

## Brief Report

# Hoarseness after pulmonary arterial stenting and occlusion of the arterial duct

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**Abstract** We report a 12-year-old girl who had multiple congenital cardiac lesions, specifically an arterial duct, left pulmonary arterial stenosis, an atrial septal defect in the oval fossa, and mild Ebstein's malformation of the tricuspid valve. Therapeutic transcatheter intervention was performed to stent the left pulmonary artery, occlude the arterial duct with a coil, and place a device to close the atrial septal defect. Subsequent to the catheterization, she complained of hoarseness, which was shown to be due to entrapment of the left recurrent laryngeal nerve between the coil used to close the arterial duct and the stent placed in the left pulmonary artery. Laryngoscopy confirmed paralysis of the recurrent laryngeal nerve.

**Keywords:** Therapeutic catheterization; patent arterial duct; coil occlusion; complication

PATENCY OF THE ARTERIAL DUCT IS COMMONLY associated with stenosis of the left pulmonary artery.<sup>1</sup> With the advances made in interventional catheterization, transcatheter therapy has now become the treatment of choice for this combination of lesions, using a variety of implantable devices.<sup>2–5</sup> Development of stenosis of the left pulmonary artery, however, is also a known complication of using a device to close a patent arterial duct.<sup>6</sup> This may require stenting of the left pulmonary artery at a separate catheterization.<sup>7</sup> The left recurrent laryngeal nerve has a long course, and is known to bear a close relationship to the arterial duct and the left pulmonary artery.

In this report, we describe acute paralysis of the left recurrent laryngeal nerve, and paresis of the vocal cords, following transcatheter closure of the arterial duct combined with stenting of the left pulmonary artery. Injury to the recurrent laryngeal nerve is well-recognized following surgical ligation of the arterial duct.<sup>8</sup> Our complication, in contrast, is due to entrapment of the nerve between the stent in the left

pulmonary artery and the coil used to close the arterial duct.

To our knowledge, this complication has not previously been reported following such interventions.

## Case report

A 9-year-old girl presented with a few months history of exertional dyspnoea and cyanosis. On examination, she had no respiratory distress, but demonstrated central and peripheral cyanosis, along with clubbing of the fingers and toes. Her saturation of oxygen in room air was 89%. She weighed 22 kg, and her remaining vital signs were normal. Cardiac auscultation revealed a normal first sound, loud and fixed splitting of the second heart sound, and an ejection systolic murmur, grade II/VI, best heard at the left upper sternal border, with a continuous murmur of grade II/VI audible at the left sub-clavicular area.

The cross-sectional echocardiogram showed dilation of the inferior caval vein, along with a dilated right atrium and right ventricle. There was a defect within the oval fossa of 14 mm diameter permitting bi-directional shunting, mild Ebstein's malformation of the tricuspid valve, moderate tricuspid regurgitation with peak Doppler velocity of 4.4 m/s, and trivial mitral regurgitation. The left ventricle was mildly dilated, but showed normal function. No obstruction was found in either the right or left

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ventricular outflow tract, but the pulmonary trunk and right pulmonary artery were dilated, the right pulmonary artery measuring 15 mm in diameter, with severe stenosis of the left pulmonary artery, which measured only 3 mm in diameter at its origin. The aortic arch was left-sided without aortic coarctation. The arterial duct was restrictive, measuring 3–4 mm in diameter.

### Cardiac catheterization

Our patient underwent interventional cardiac catheterization for stenting of the left pulmonary artery, placement of a coil in the arterial duct, and placement of a device in the atrial septal defect (Fig. 1). General anesthesia was achieved with endotracheal intubation, using a size 5.5 mm non-cuffed endotracheal tube. Intubation, and post procedural extubation, proceeded smoothly, with no difficulties encountered. The procedure lasted for 172 min. Pulse oxymetry was 89% on room air, and increased to 97% subsequent to the catheterization.

Prior to stenting, the gradient from the left pulmonary artery to the pulmonary trunk was measured at 50 mmHg. The aortogram showed a patent arterial duct of moderate size permitting a left-to-right shunt. Pulmonary arteriography confirmed the presence of a severe and long stenosis of the left pulmonary artery, with the narrowest diameter measured at 3 mm. Test occlusion of the atrial septal defect after stenting the left pulmonary artery and occluding the arterial duct showed an increase in aortic saturation

from 89% to 100%, and a decrease in pulmonary arterial to half systemic values, at 50/17 mmHg. Aortic pressure was 105/74 mmHg.

The left pulmonary artery was stented with Johnson and Johnson Interventional Systems Co size P188- and size P308-Palmaz stents. After deployment, an aortogram showed good positioning of the stent, with improvement in the diameter of the left pulmonary artery.

The arterial duct was then occluded using a 5 mm diameter detachable Gianturco coil (Cook, Inc., Bloomington, IN, USA) inserted retrogradely. The atrial septal defect was then closed under transesophageal echocardiographic guidance using a 16 mm diameter Amplatzer device (Amplatzer septal occluder, AGA Medical Corporation, Golden Valley, MN, USA). There was no residual leak.

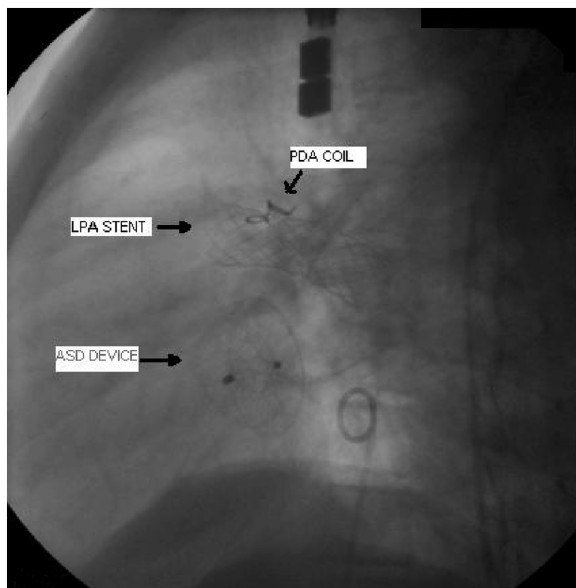
On the second day after catheterization, the patient was complained of mild hoarseness, which was initially attributed to intubation. On follow up at 3-months, the patient was asymptomatic except for the hoarseness, that persisted over 3 to 6 months following the procedure. Indirect laryngoscopy showed paralysis of the left vocal cord. Repeated laryngoscopy six months later confirmed persistent paralysis of the left vocal cord, with a mobile right cord.

### Discussion

It is now standard therapy to use coils to close the patent arterial duct and stenting to relieve pulmonary arterial stenosis.<sup>4,5</sup> This combination of lesions is well-recognized,<sup>1</sup> and stenosis of the left pulmonary artery is also a known complication of closure of the arterial duct that may require stenting.<sup>6</sup> Paralysis of the left recurrent laryngeal nerve is also well documented following surgical ligation of a patent arterial duct.<sup>8</sup>

The left recurrent laryngeal nerve, which innervates the larynx and the vocal cords, has a close anatomical relationship to the arterial duct and left pulmonary artery as it hooks around the arterial duct or ligament before ascending back to the larynx. Surgical ligation is well-established as resulting sometimes in its injury and secondary hoarseness.<sup>9</sup> In our patient, transcatheter intervention produced the same result, presumably by directly injuring or entrapping the recurrent laryngeal nerve between the devices placed in the left pulmonary artery and the arterial duct.

We have now stented the left pulmonary artery and occluded the arterial duct with coils in 5 patients, concomitantly in 2 or to relieve stenosis following initial ductal occlusion in 3. In the latter cases, we used a Rashkind 17 mm device in one and inserted multiple coils in the other two. None of them developed paresis or paralysis of the recurrent laryngeal nerve.



**Figure 1.** Posterolateral cineangiography shows the devices in place, demonstrating the close proximity of the coil in the arterial duct and the stent in the left pulmonary artery.

The hoarseness could have been related to intubation;<sup>9</sup> but the endotracheal intubation in our patient was smooth, and the non-cuffed endotracheal tube was of appropriated size for her age. Furthermore, the laterality of the injury, and its persistence beyond six months after the procedure, speaks for genuine injury to the recurrent laryngeal nerve. By our experience, we hope to raise the awareness of paediatric cardiologists for this possible complication secondary to interventional transcatheterization. It is somewhat reassuring to know that improvement of hoarseness usually occurs in the long-term following injury to the recurrent laryngeal nerve.<sup>10</sup>

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