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# **Brief Report**

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# Giant pulmonary pseudoaneurysm following balloon dilatation of the pulmonary artery to relieve pulmonary band

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# Abstract

Pulmonary artery pseudoaneurysms are uncommon. They may occur secondary to trauma, infectious diseases, vasculitis syndromes, neoplasms, congenital diseases, and pulmonary hypertension. Due to increasing number of cardiac interventions, iatrogenic complications are among the major causes of pulmonary artery pseudoaneurysms.

In this report, we present a 6-month-old patient with pulmonary pseudoaneurysm that occurred following pulmonary balloon angioplasty for the relief of a pulmonary band.

Pseudoaneurysms of the pulmonary artery are very rare. Etiologies include infections, trauma, vasculitis syndromes, neoplasms, and/or iatrogenic causes. They are defined as focal dilatations that arise among the pulmonary artery. Histologically pseudoaneurysms involve only the external layer of the arterial wall; thus, pulmonary artery pseudoaneurysms are associated with a high risk of rupture owing to the relatively low resistance of the surrounding tissue.<sup>1</sup>

Hemoptysis is the most frequent presenting symptom of such patients; however, symptoms range from acute to severe haemorrhage to incidental findings on radiographs. Treatment of pulmonary artery pseudoaneurysms include surgical ligation, wedge resection, lobectomy, angiographic embolisation, endovascular stent graft placement, and close follow-up.<sup>2</sup>

In this report, we present a 6-month-old patient with a pulmonary pseudoaneurysm.

## **Case report**

A 6-month-old female patient with a borderline left ventricle (aortic annulus size:7 mm, Z score of aortic valve: -1.72, mitral annulus size: 8 mm, Z score of mitral valve: -2.05, and Z score of left ventricle: -2.88), multiple ventricular septal defects, and sinus venosus type atrial septal defect whom underwent aortic type B interruption repair with an autologous pericardial roll anastomosed between the ascending aorta and the descending aorta as well as a pulmonary band operation with a 3-mm width PTFE tape material during the early neonatal period presented to the clinic with increasing dyspnoea, lethargy, easy fatigability, and poor feeding. The patient was intubated due to respiratory distress and a chest X-ray was performed which showed signs of pneumonia. The echocardiography indicated biventricular outflow tract stenosis with increased transaortic and transpulmonic gradients along with depressed myocardial function. Cardiac catheterisation and if necessary, balloon angioplasty were planned. During the intervention the systolic ascending aorta, descending aorta and transaortic pressures were 72, 52, and 20 mmHg, which altered to 65, 62, and 3 mmHg, respectively. The invasive pulmonary gradient was measured as 45 mmHg. A decision to mildly dilate the pulmonary band to aid biventricular outflow obstruction and myocardial contractions without leading to pulmonary over circulation was made. The pulmonary band was minimally loosened revealing a 35-40 mmHg transpulmonary systolic gradient. The procedure was finalised uneventfully.

The patient was followed at the ICU; however, the patient could not be weaned off the ventilator, and the ICU stay was complicated with sepsis attacks as well as four episodes of sudden cardiac arrest which were reversed with medical and mechanical resuscitation. Serial echocardiography controls indicated successful angioplasty of the aorta, the pulmonary artery with markedly reduced gradients, and a minimally dilated pulmonary artery. A decision was made to perform a control cardiac catheterisation to rule out any cardiac abnormalities which may have caused cardiac arrest and the inability to wean off the ventilator. The angiography indicated severely enlarged pulmonary artery, presumable a pseudoaneurysm (Fig 1, Suppl. 1). A review



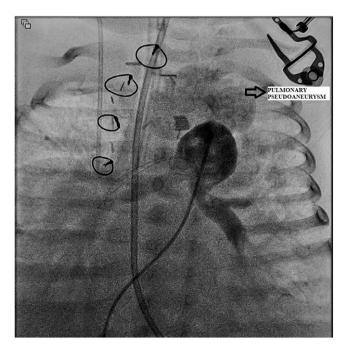


Figure 1. Cardiac catheterisation indicating giant pulmonary pseudoaneurysm.

of the echocardiography is performed before the angiography through the hospital database; however, there was no indication within the records regarding pulmonary enlargement, therefore indicating a relatively new event. Control contrasted CT of the thorax indicated a giant pulmonary pseudoaneurysm just beneath the sternum leading to bronchial obstruction and compression of major cardiovascular structures (Fig 2a, 2b, Suppl. 2).

After explaining the pathology and the risks, a consent was obtained from the patient's parents for urgent surgical treatment as well as possible use of the recorded materials in the hospital's database for academic purposes. Due to vicinity of the pseudoaneurysm to the sternum and high risk of injury, median sternotomy was performed on-pump following the right carotid artery and jugular vein cannulations. Extensive fibrous tissue formation was observed following sternotomy of the patient (Fig 3). The patient was cooled down to 14°C, and total circulatory arrest was instituted. We incised the pulmonary artery and faced with detachment of the main pulmonary artery from the pulmonary bifurcation and severely destructed main pulmonary artery. The pulmonary artery was reconstructed with a xenograft (Edwards Lifesciences Corp. One Edwards Way Irvine, CA 92614) pericardial roll. The peripheric cannulae was transferred to the ascending aorta and the right atrium afterwards the patient was re-warmed. Pulmonary banding was performed to the neopulmonary artery. Patient was weaned off cardiopulmonary bypass with inotropic support and transferred to the ICU with an open sternum due to severe oedema most probably secondary to septic cardiogenic shock. Unfortunately, the patient was lost due to deteriorating general condition and multiorgan failure in the post-operative period.

## Discussion

Pulmonary artery aneurysms and pseudoaneurysms are very rare. Pathophysiologiclly, aneurysms comprise all three layers of the arterial wall. Unlike aneurysms, pseudoaneurysms are devoid of a true three-layer wall and are usually composed of a local haematoma contained by the surrounding tissues around the artery.<sup>3</sup> The most serious complication of an aneurysm or a pseudoaneurysm is rupture. The risk of rupture is higher in cases of pseudoaneurysms when compared with a true aneurysm. In case of a rupture, it is reported that mortality rates may reach up to 50%.<sup>4</sup>

Traumatic events, congenital cardiovascular lesions, pneumonia, systemic infections (e.g., tuberculosis, syphilis, and HIV), vasculitis syndromes (Behcet's disease, Ehler–Danlos syndrome, and Hughes–Stovin syndrome) and malignancy are the main aetiologies for pulmonary artery pseudoaneurysms.<sup>5</sup> Sparse reports of pulmonary artery pseudoaneurysms following percutaneous interventions such as insertion of a Swan-Ganz catheter<sup>6</sup> or percutaneous transluminal angioplasty for pulmonary artery stenosis are also observed.<sup>7</sup>

Patients with pulmonary pseudoaneurysms may present to the clinic completely asymptomatic and be diagnosed incidentally, with compression symptoms to the adjacent structures, chest pain, hypoxaemia, or haemoptysis.<sup>4</sup> The diagnosis relies on radiographic findings. Plain chest X-rays may rarely be suggestive. Echocardiography may be helpful for intramediastinal lesions. Contrasted CT is the main diagnostic tool in the current era. Conventional angiography may be preferred in selective cases and especially when percutaneous treatment measures are planned.<sup>8</sup>

Since the pseudoaneurysms are devoid of true arterial layers, rupture may be fatal. Hence, when diagnosed, prompt action is necessary. The treatment does not only depend on the aetiology but also the location of the lesion. In symptomatic patients, endovascular treatment options such as coil embolisation, thrombin injection, and/or stent/stent graft implantation are rapid and life-saving options. On the other hand, in patients with hemothorax, uncontrolled haemoptysis, or irresponsive infections, surgery is the primary treatment option which may range between simple pseudoaneurysm sac evacuation with arterial reconstruction to resection of the effected pulmonary lobe or even pneumonectomy.<sup>4</sup>

We presume that the aetiology for pulmonary pseudoaneurysm in our case had been due to the pulmonary balloon angioplasty for the relief of the pulmonary band. In addition, the infectious condition of the patient may have complicated the aetiology as well. The pseudoaneurysm resulted in a sudden cardiac arrest most probably due to compression of adjacent structures. The diagnosis was made incidentally during cardiac catheterisation while investigating unexplained cardiac arrests and confirmed with contrasted CT of the thorax. Although the patient was a re-do case, waiting until the infection had passed was not an option because of rapid onset, huge size, and complications arising due to the pseudoaneurysm. It was not amenable to percutaneous treatment, and surgical resection of the pseudoaneurysm was conducted along with pulmonary artery reconstruction with a xenograft pericardial roll.

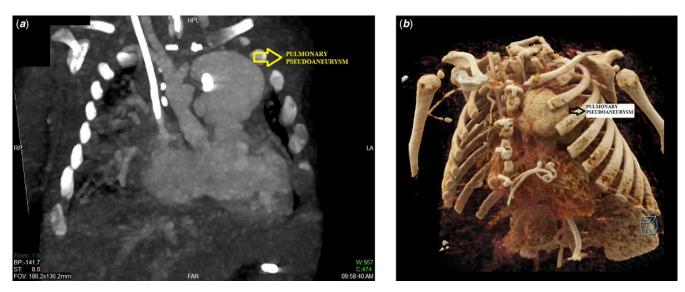


Figure 2. (a) Contrasted CT indicating giant pulmonary pseudoaneurysm and vicinity of it to the sternum. (b) 3D reconstruction of the tomography.

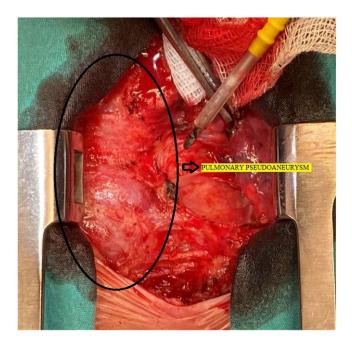


Figure 3. Perioperative image of the pulmonary pseudoaneurysm.

Supplementary material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951122002980

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Conflict of interest. None.

IRB approval. N/A (single case report)

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