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Images in Congenital Heart Disease

Embolization of a pulmonary arterial pseudoaneurysm with endovascular coils

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Abstract Unrecognized pseudoaneurysm or rupture of a pulmonary artery is a rare but potentially catastrophic complication of pulmonary arterial catheterization. Herein, we describe a teenage patient with a pulmonary arterial pseudoaneurysm, probably iatrogenic, who presented with haemoptysis following catheterization of the right heart. The pseudoaneurysm was successfully embolized using coils inserted by catheter. Increased awareness of this lesion, its rapid recognition, and prompt therapy are the keys to a successful outcome.

Keywords: Interventional catheterization; haemoptysis; congenital cardiac disease

Catheter-induced pulmonary arterial pseudoaneurysm is a rare, but serious, complication of catheterization of the right heart.[1] The estimated frequency of haemorrhage, rupture, or pseudoaneurysm resulting from catheterization of the pulmonary arteries is from 0.04% to 0.2%, but has been associated with death in half of the reported cases.[2] Risk factors that have been identified include advanced age, over 60 years, rigid catheters, anticoagulation, chronic use of steroids, pulmonary arterial hypertension, in the case of Swan-Ganz catheters, prolonged inflation of the balloon, multiple manipulations, or peripheral placement of the catheters.[1,3,4] Such a complication in a young patient undergoing diagnostic catheterization of the right heart for congenital cardiac malformations, however, is exceedingly rare.

Case report

A 17-year-old female presented for investigation of a one-year history of intermittent haemoptysis. Most episodes produced a small amount of clot. After one large haemoptysis, there was sufficient bleeding to reduce her haemoglobin by 2 milligrams per decilitre, and she required a blood transfusion. At the age of 4 weeks, she had previously undergone repair of coarctation of the aorta, along with banding of the pulmonary trunk, for a ventricular septal defect. Subsequently, uneventful closure of the defect, and removal of the band was performed at the age of 2 years. She was well until the onset of dyspnoea at the approximate age of 16 years. At that time, echocardiography at an outside institution demonstrated severe pulmonary arterial hypertension. A diagnostic cardiac catheterization, along with a vascular reactivity study, was performed. This confirmed a mean pressure of 50 millimetres of mercury in the pulmonary arteries. Her baseline pulmonary arteriolar resistance was severely elevated, at 19 Wood units per metre squared. No significant decrease in pulmonary arteriolar resistance occurred with 100% oxygen or 80 parts per million of nitric oxide. A balloon catheter was used to measure the pulmonary arterial wedge pressure. No complications were noted during the procedure, and treatment with sildenafil citrate and bosentan was initiated. Shortly thereafter, she began to experience recurrent episodes of haemoptysis.

Physical examination at our institution demonstrated a prominent right ventricular impulse, an ejection systolic murmur graded at 3 from 6 at the left upper sternal border, radiating to the carotid

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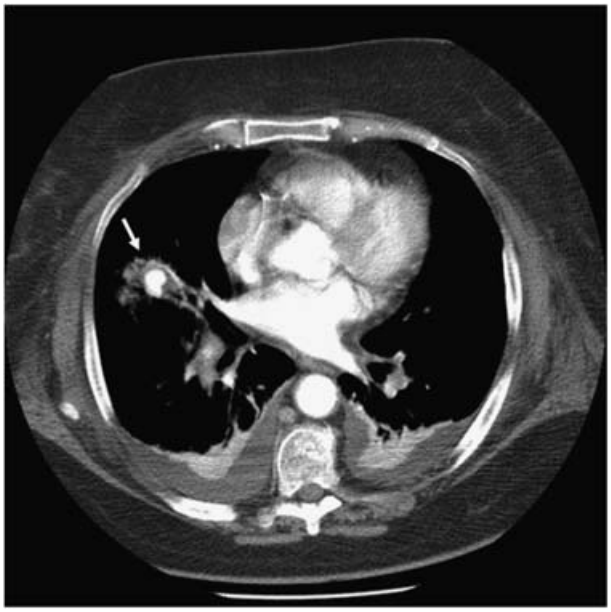

Figure 1.
Multislice contrast computed tomography of the chest showing the pulmonary arterial pseudoaneurysm (arrow) surrounded by pulmonary haemorrhage.

arteries, and a loud pulmonary component of second heart sound. Our investigations included a chest radiograph, which demonstrated a prominent pulmonary trunk. During a flexible fibreoptic bronchoscopy, thrombus was found occluding the lateral basal segment of the right lower lobe. A bronchoalveolar lavage was performed and the results excluded tuberculosis, as well as bacterial or fungal infections. A multislice contrast computed tomography scan of the chest (Fig. 1), revealed a saccular pseudoaneurysm 2 centimetres in diameter in the right lower thorax, with patchy alveolar infiltrates in the right lower lobe. This was interpreted as consistent with alveolar haemorrhage, and pulmonary arterial angiography demonstrated a pseudoaneurysm in a subsegmental branch of the right middle lobe branch pulmonary artery. A "Tracher catheter" was used to access the two arteries feeding into the false aneurysm, and multiple coils (Cook Inc.®) were used to occlude these branches (Fig. 2). Following embolization, the haemoptysis resolved completely, and she was discharged from hospital. During six months of follow-up, she has continued to remain well without any further episode of haemoptysis.

Discussion

The pulmonary arterial pseudo, or false, aneurysm develops after rupture of the vessel wall. The resultant bleeding is contained by surrounding

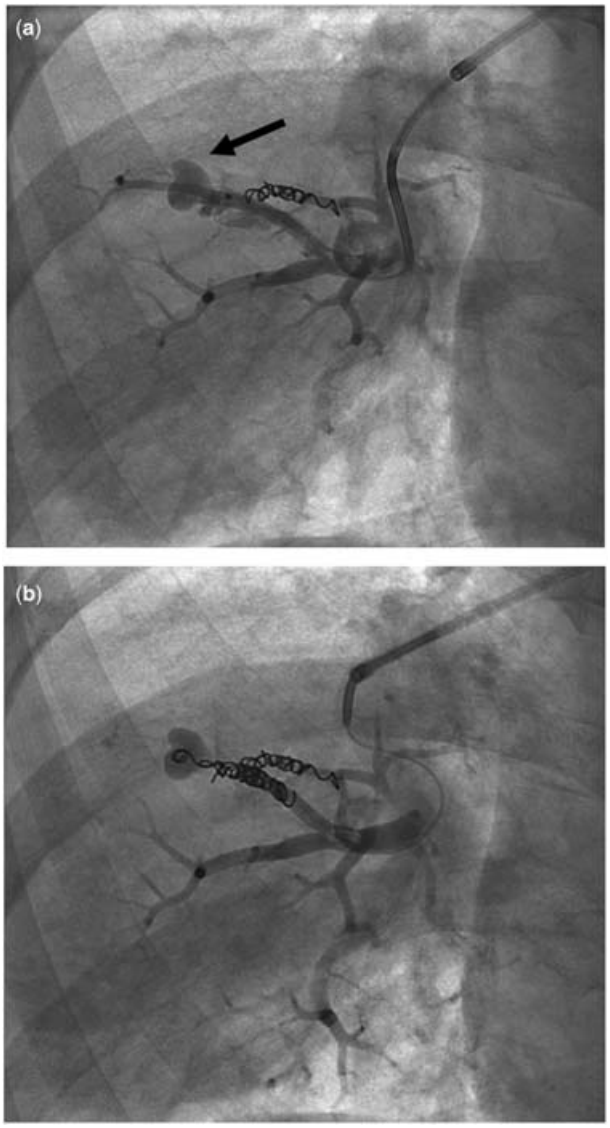

Figure 2.
*The pseudoaneurysm (arrow) with two feeding subsegmental pulmonary arteries and coil embolization (**a**) of one feeding vessel followed by (**b**) embolization of both feeding vessels.*

pulmonary parenchyma, and the vessel thus lacks normal vascular endothelium. End-stage necrotizing arteritis in pulmonary hypertension can rarely cause formation of such pseudoaneurysms in small pulmonary arteries through fullthickness obliteration of the muscular wall.[5] Vascular trauma during pulmonary arterial catheterization, usually with a Swan-Ganz or balloon tipped catheter, can also cause pseudoaneurysm, or even rupture of the pulmonary artery.[2,6] Overinflation of the balloon may result in vessel injury.

The clinical presentation of pulmonary arterial pseudoaneurysm ranges from the incidental finding of a new pulmonary mass or infiltrate on the chest radiograph in an asymptomatic patient, to

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dyspneoa, chest pain, recurrent haemoptysis, and life threatening haemorrhage.[4,7,8] Pulmonary haemorrhage is the most common mode of presentation, and may occur during the procedure, or present even months later with recurrent haemoptysis.[7] Spontaneous regression of asymptomatic lesions has been observed.[8] Pulmonary arterial rupture in the setting of pseudoaneurysm is prudently regarded as a serious and potentially life-threatening complication of catheterization of the right heart, and has proved fatal in about half of reported cases.[3] A high index of suspicion is the key to prompt diagnosis of this complication, for which appropriate intervention can be life-saving.[1,9] In subacute or chronic cases, contrast enhanced multislice chest computed tomography offers an excellent, less invasive modality for detection of these aneurysms.[1,10] Pulmonary arterial angiography allows direct visualization, and is necessary for transcatheter management.

The preferred treatment is transcatheter insertion of coils.[1,7] This may involve occlusion of the feeding vessels, as illustrated by our patient, or obliteration of the cavity of the pseudoaneurysm itself.[6,9] Conservative management, and surgical treatment including aneurysmectomy, wedge resection, or lobectomy, are associated with high mortality.[3,10]

The exact aetiology in our patient is unclear. Although extremely rare cases of spontaneous formation of pseudoaneurysms have been reported in patients with primary pulmonary hypertension,[5] our patient had neither the degree of elevated pulmonary artery pressure nor the classically described plexiform lesions. The temporal relation of her haemoptysis to pulmonary arterial catheterization supports an assumption that the vessel was injured during the procedure, but virtually all such reported cases have been in older patients. This case emphases the importance of proper technique when inflating the balloon for measurement of the pulmonary capillary wedge pressure, close observation of the pressure waveform, and the sensation of resistance as a guide to the pressure of inflation to be used to prevent this complication.[7]

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