An unusual cause of pulmonary artery pseudoaneurysm: acrylate embolism

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Abstract

Sclerotherapy is commonly used to manage bleeding from oesophageal varices. In a patient with cirrhosis of the liver, sclerotherapy with bucrylate was followed by a pulmonary embolism and then by a decline in general health. A chest radiograph taken 5 months later disclosed a left perihilar opacity, surrounding and invading the pulmonary artery. Despite moderate fixation by positron emission tomography and inconclusive bronchoscopy findings, an upper left lobectomy was deemed in order. A left pulmonary artery pseudoaneurysm was found during the surgery. The pseudoaneurysm ruptured during dissection, requiring a left pneumonectomy. The pathological examination showed shredding of the left pulmonary artery, which contained foreign material. At points of contact with this material, destruction and severe polymorphic inflammation of the pulmonary parenchyma were noted. There was no evidence of tumour or infection. These findings strongly suggested an iatrogenic pulmonary artery pseudoaneurysm related to a bucrylate embolism through porto-systemic vascular shunts. We are not aware of previously reported cases.

Keywords: Sclerotherapy • Oesophageal varices • Aneurysm • Pulmonary artery • Iatrogenic disease

INTRODUCTION

Sclerotherapy is widely used to treat bleeding from oesophageal varices \cite{1}. The most common adverse effect of sclerotherapy is a pulmonary embolism (PE) due to the migration of the sclerosing agent into the pulmonary vessels \cite{2, 3}. Here, we report a case of pulmonary artery pseudoaneurysm probably due to sclerosing agent migration through porto-systemic shunts.

CASE REPORT

A 69-year old man was referred to the thoracic surgery department for the management of a pulmonary opacity with a decline in general health and prolonged fever. His medical history was chiefly remarkable for cirrhosis of the liver due to non-alcoholic steato-hepatitis. He had no history of smoking or alcohol abuse. His first episode of haematemesis 1 year earlier led to the diagnosis of oesophageal varices. Endoscopic ligature and sclerotherapy with bucrylate (isobutyl-2-cyanoacrylate) were performed. In the first days after the sclerosis, the patient had fever (40°C) and the C reactive protein was increased from 24 to 50 mg/l. The inflammatory markers quickly normalized. One month after being discharged home, he started experiencing chest pain and dyspnoea with no fever. A PE was suspected. Computed tomography (CT) revealed a margined defect in the left lower pulmonary artery. The cardiac ultrasound was normal with a normal systolic function, and no dilation of the right cavities; systolic pulmonary arterial pressure was estimated to be 35 mmHg. Anticoagulant treatment was given for 3 months. The patient presented two episodes of small amounts of haemoptysis 3 months after the varical sclerosis. Over the next 8 months, he had chills and coughing with sputum production, which were diagnosed as recurrent pulmonary infections. Fever (38–38.4°C) remained even during periods without pulmonary infections. Several courses of antibiotics induced transient improvements, but he lost a considerable amount of weight and reported severe fatigue. CT showed a left perihilar mass located chiefly around the left apical pulmonary artery, thickening the vessel and occluding it (Fig. 1). Bronchoscopy was normal and routine biopsies were inconclusive. Positron emission tomography showed moderate uptake by the mass (maximal SUV 2.7). Surgery was first recommended for suspected lung cancer because transbronchial or CT-guided biopsy was associated with a high risk of major and fatal bleeding, especially with coagulation disorders related to liver cirrhosis (platelet level 100 G/l).

During surgery, the left pulmonary artery was seen to be surrounded by inflammatory tissue. Dissection within the lung fissure disclosed a pseudoaneurysm with inflammation adjacent to the sclerotherapy material located within the fissure. The pseudoaneurysm ruptured during lymphadenectomy, requiring a left pneumonectomy.
By gross examination, the left pulmonary artery was dilated and ruptured and contained an extensive adherent thrombus filling the lower intrapulmonary artery branches. Distal emboli of foreign material were seen (Fig. 2). Microscopic examination revealed serpiginous foreign material in the left pulmonary artery and its branches. The artery walls were dilated and eroded, with multiple rupture points and exudation of the material into the lung parenchyma. A dense inflammatory infiltrate containing a few giant cells was visible around the foreign material and there were extensive ulcerations of the bronchial mucosa and cartilage at points of contact with the material. The reactive fibrous tissue surrounding the lesions produced a tumour-like appearance. Distal areas of infarction and scarring were seen in the lung.

The patient experienced several postoperative complications including recurrent pulmonary infections, a bronchial fistula, recurrent ascites and gastric bleeding. He died of septic shock 3 months after the surgery.

**DISCUSSION**

Porto-systemic vascular shunts are common in patients with cirrhosis of the liver and portal hypertension. In particular, oesophageal varices conduct blood directly from the portal vein to the inferior vena cava. Sclerotherapy agents injected into the varices may therefore migrate to the right cardiac chambers and pulmonary circulation, causing PE. A similar mechanism has been demonstrated with foam sclerotherapy of varicose veins [4]. In our patient, multiple pulmonary emboli of sclerotherapy material led to the formation of pseudoaneurysms with multiple breaks in the arterial wall. Migration of the material through
these breaks caused perivascular pulmonary and bronchial inflammation with a fibroid reaction mimicking a tumour.

Cyanoacrylates are rapidly polymerizing adhesives that have been used for embolization therapy of complex cerebral and extra-cerebral vascular abnormalities. Most of the studies investigating the histopathological changes associated with cyanoacrylate deposition in humans focused on cerebral vascular malformations [5]. Inflammation with mural angionecrosis adjacent to bucrylate fragments, bucrylate deposits within vessel walls and the presence of bucrylate at extravascular sites have been reported. To the best of our knowledge, our case is the first report of a pulmonary artery aneurysm associated with bucrylate emboli documented by histopathological examination. The severity of the manifestations in our patient was related to the concomitant presence of distal bucrylate emboli in both lobes, with small infarcted foci, and to the local deleterious effect of bucrylate on the artery walls and surrounding parenchyma. No other foreign material was injected in this patient, and the symptoms, especially fever, started early after sclerotherapy. This diagnosis had not been suggested even if several differential hypotheses had been eliminated like a recurrent PE (no filling defects during the follow-up), endocarditis (normal cardiac ultrasonics and negative blood cultures) or infective process due to Aspergillus (negative Aspergillus research on repeated sputum, bronchial aspiration and BAL).

In conclusion, bucrylate used to treat oesophageal varices caused a pulmonary artery aneurysm with severe vessel wall damage. We are not aware of previously reported cases, suggesting that this complication may be extremely rare. However, its incidence may be underestimated given the non-specific nature of the symptoms. In a similar case, a pulmonary angiography should be performed before surgery to evaluate the possibility of pulmonary pseudoaneurysm embolization to minimize the intraoperative risk of rupture.

Conflict of interest: none declared.

REFERENCES