Strangulation of aberrant artery in extralobar pulmonary sequestration on video imaging

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Abstract

We report a case of an 18-year old female patient with symptomatic extralobar pulmonary sequestration. The initial symptom was sudden-onset right lateral abdominal pain. Enhanced computed tomography showed a 5 cm in diameter, spindle-shaped mass located in the costophrenic sinus with no aberrant artery. Exploratory thoracoscopy showed a haemorrhagic mass caused by strangulation of an aberrant vessel originating from the intercostal artery. Pathological findings revealed pulmonary sequestration with haemorrhagic infarction. The strangulated aberrant artery was clearly demonstrated by video imaging.

Keywords: Extralobar pulmonary sequestration • Strangulation • Thoracoscopy

INTRODUCTION

Extralobar pulmonary sequestration (EPS) is usually asymptomatic and incidentally detected. EPS is rarely associated with symptoms such as dyspnoea, chest pain, abdominal pain or fever, which are speculated to occur secondary to infarction and/or infection. Sudden-onset circulatory disorders of EPS may be related to torsion, but no reports have revealed such an image. Here, we demonstrate the identification of a strangulated aberrant artery by thoracoscopy in a patient with symptomatic EPS.

CASE REPORT

An 18-year old female patient presented to a nearby hospital with fever and sudden-onset abdominal pain. She had no history of trauma or illness. Blood examination revealed a white blood cell count of 13 000/mm³, haemoglobin level of 13.5 g/dl and serum C-reactive protein concentration of 7.4 mg/dl. Contrast-enhanced chest CT revealed predominantly right lateral abdominal pain. Enhanced computed tomography showed a 5 cm in diameter, spindle-shaped mass located in the costophrenic sinus with no aberrant artery. Exploratory thoracoscopy showed a haemorrhagic mass caused by strangulation of an aberrant vessel originating from the intercostal artery. Pathological findings revealed pulmonary sequestration with haemorrhagic infarction. The strangulated aberrant artery was clearly demonstrated by video imaging.

Thoracoscopy revealed bloody pleural effusion and a haemorrhagic mass adjacent to the inferior ligament. The mass was adherent to the right lower lung lobe, diaphragm and chest wall. A strangulated aberrant artery from the intercostal artery was identified during synechiotomy. Complete resection was uneventful with ligation of the aberrant vessel (Video 1). The mass was encapsulated by its own visceral pleura and region of lung parenchyma that was disconnected from the tracheobronchial tree and pulmonary artery. Histopathological examination showed an organized pulmonary parenchyma with diffusely haemorrhagic necrosis. The pathological diagnosis was an infarcted EPS. The elastic arteries from the intercostal artery and veins to the azygous vein were occluded by an organized clot (Fig. 2).

DISCUSSION

Lung malformations have an estimated incidence rate of 2.20–6.60%, and pulmonary sequestrations are rare, with an estimated...
incidence rate of 0.15–1.80% [1]. An abnormal lung segment enclosed within the pleural membrane, completely separated from the tracheobronchial tree, accounts for 25% of all pulmonary sequestrations. The preferential site is reportedly the left inferior ligament, which occurs with a frequency of 80%. The aberrant artery originates from the thoracic or abdominal aorta in more than 80% of cases of EPS. The blood supply is provided by another systemic artery (intercostal, subclavian, brachiocephalic or splenic in 15% of cases, and gastric or pulmonary arteries in 5%) [2, 3].

Diagnosis of EPS is difficult when the patient is an adult because EPS is uncommon, usually asymptomatic and generally discovered on routine chest X-ray films or during incidental examinations for other diseases. Seven case reports can be found in PubMed using a combination of the keywords ‘extralobar pulmonary sequestration’ and ‘torsion’. The symptoms in these cases were sudden-onset ipsilateral chest and abdominal pain and subsequent respiratory failure caused by haemothorax and infarction. Chen et al. [4] reported that all cases of EPS torsion noted in the literature were located in the inferior portion of the left thoracic cavity, and a pathological study showed diffusely haemorrhagic pulmonary parenchyma consistent with infarction. These previous reports suggested that symptomatic EPS may be related to infarction caused by incidental torsion during drastic respiratory movement. Torsion of a sequestration has rarely been demonstrated at the time of surgery because the aberrant artery gradually shrunk and was absent after strangulation. Tetsuka et al. [5] reported 2 patients with symptomatic EPS with no aberrant vessels and speculated that these aberrant vessels were tiny enough to easily strangulate and shrink. Thoracoscopy can demonstrate such torsion in case it is performed shortly after the initial onset.

In conclusion, we confirmed that symptomatic infarcted EPS was caused by a strangulated aberrant artery of the EPS. Urgent thoracoscopy should be recommended even when an aberrant artery belonging to a mass beside the inferior ligament is not detected in a patient with ipsilateral abdominal pain.

Conflict of interest: none declared.

REFERENCES