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Extralobar pulmonary sequestration manifesting as hemorrhagic infarction in a five-year-old male

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ABSTRACT

Extralobar pulmonary sequestration (EPS) is frequently asymptomatic and detected incidentally. We herein report a pediatric case of EPS that manifested as hemorrhagic infarction. A five-year-old male with previously normal X-ray films was referred to our hospital due to acute onset of right flank pain and a non-productive cough. The initial diagnosis was infection of an EPS; however, the symptoms persisted despite the administration of antibiotics. A CT-angiogram revealed interruption of the blood flow to the sequestrum, and a definitive diagnosis as hemorrhagic infarction of a pulmonary sequestration was made. During surgery, an aberrant artery was revealed to derive from the intercostal artery and the sequestrum was resected safely. In cases of infarction of EPS, early surgical resection should be considered, because progressed adhesion can complicate dissection from surrounding organs and the detection of the feeding vessels.

A pulmonary sequestration is defined as a mass of abnormal lung tissue which has no bronchial communication with the normal tracheobronchial tree. Pulmonary sequestrations are divided into two groups; extralobar and intralobar sequestrations, based on whether they have their own visceral pleura. Extralobar pulmonary sequestration (EPS) is usually asymptomatic and detected incidentally during examinations for other clinical conditions [1]. We herein report a case of symptomatic EPS with massive bloody pleural effusion, and the initial symptom was a sudden onset of right flank pain. The pathological findings were compatible with hemorrhagic infarction of an EPS. We also review other previously reported cases of this condition.

1. Case report

A five-year-old male was referred to our hospital due to a sudden onset of right flank pain and a non-productive cough. The patient had a past history of acute bronchitis when he was two-years-old and had undergone adenoidectomy when he was four-years-old. The patient received X-ray examinations each time, in which pulmonary sequestration was not found. Radiologic abnormalities were not identified even though our radiologists reviewed these X-ray films. At this presentation, the patient's temperature was 38.4 °C, his pulse was 140 bpm and his respirations were 60/min. The breath sounds were diminished over the right lower lobe. He had remarkable right flank pain, but there were no signs of peritoneal irritation or ascitic fluid. A peripheral blood examination showed elevation of the white blood cell count to 13,000 and a serum C-reactive protein concentration of 8.03 mg/dl. An X-ray film of the chest showed opacification of the right hemithorax. A contrast-enhanced computed tomography (CECT) scan revealed the presence of a solitary triangle mass without enhancement, adjacent to the posterior portion of the right lower lobe (Fig. 1). In addition, massive pleural effusion was demonstrated, which occupied almost all of the area of the right pleural cavity. The initial diagnosis was right EPS with acute infection, and antibiotic chemotherapy was initiated.

The patient underwent thoracentesis due to an increase in the pleural effusion on the second hospital day. The thoracentesis yielded bloody fluid, the bacterial examination of which was negative. A low-grade fever and right pleuritic pain persisted despite the intravenous administration of antibiotics and drainage

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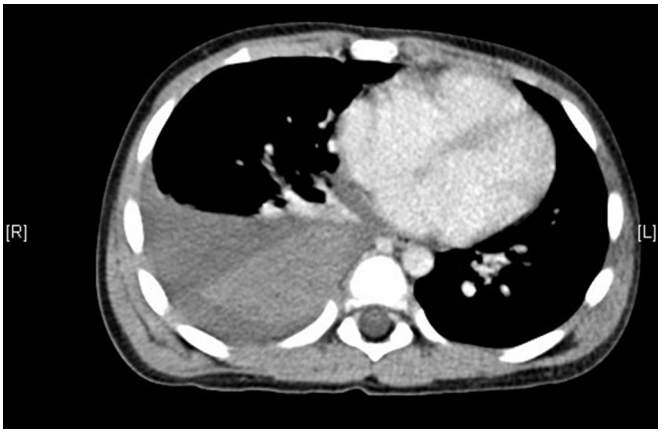

Fig. 1. A contrast-enhanced computed tomography scan showed a solitary triangular mass without enhancement, and massive pleural effusion in the right pleural cavity.

of the pleural effusion. This clinical course was considered toatypical as respiratory infection disease. Considering the absence of enhancement of the mass on the initial CECT and the bloody pleural effusion, the possibility of necrosis of the sequestrum was raised. On the fifth hospital day, CT angiography was performed to evaluate the vascular flow of the sequestrum and to detect the origin of the feeding artery. However, the vascular flow was deficient, and the feeding artery and drainage vein were not demonstrated. A definitive diagnosis of hemorrhagic infarction of an EPS was made from these findings, and the patient underwent exploratory thoracotomy on the seventh hospital day.

The patient was placed in the left lateral decubitus position under general anesthesia with unilateral ventilation. Thoracoscopy revealed bloody effusion and infarcted mass, however the vascular pedicle was not identified due to dense adhesion. A postero-lateral muscle-sparing thoracotomy via the sixth intercostal space was performed, and the dark-red sequestrum was identified at the latero-caudal part of the right pleural cavity (Fig. 2). It had its own visceral pleura, and adhesion to the right lower lobe, diaphragm and chest wall was identified. After sharp and blunt dissection of these adhesions, the sequestrum was proven to be fixed with only a fibrous structure with a short neck that derived from the posterior thoracic wall. We speculated that it was a remnant of an aberrant artery originated from the intercostal artery. The fibrous structure was ligated and divided, and the sequestrum was resected completely with no complications. The exact sequestrum size was 90 × 65 × 30 mm and the pathological findings were compatible with hemorrhagic infarction of an EPS (Fig. 3).

Almost all fields of the lung parenchyma developed hemorrhagic infarction, and the elastic lamina of the visceral pleura was entirely obscured. The corresponding site to the fibrous structure which was considered as a remnant of the feeding artery was revealed to be a necrotic feeding artery. In addition, a partially organized thrombus was revealed. Small vessels in the sequestrum were also entirely occluded with thrombi. Although the mechanism underlying the hemorrhagic infarction was not revealed during surgery, the pathological findings suggested that the hemorrhagic infarction resulted from torsion of the vascular pedicle.

The patient recovered uneventfully during the perioperative period and was discharged on the seventh postoperative day.

2. Discussion

EPS are usually asymptomatic and discovered incidentally. Hemorrhagic infarction of an EPS is rare, and only 10 cases have been reported in the literature [2–10] (Table 1). The previously reported cases included six females and four males, ranging from three to 38 years old. The affected side was the left in seven patients and the right in the remaining three patients. None of the previous reports mentioned the findings of past chest X-ray films. In the present case, we speculate that the sequestrum was located dorsal to the liver. Therefore, no abnormal findings were identified in the previous X-ray films, because it was behind the liver.

The initial clinical symptoms were sudden back pain in one patient, chest pain in four patients, abdominal pain in three patients and both of these in two patients. Many of the symptoms showed a sudden onset presentation. In the present case, a non-productive cough was seen in addition to right flank pain, and was considered to be caused by stimulation of the pleura by the bloody

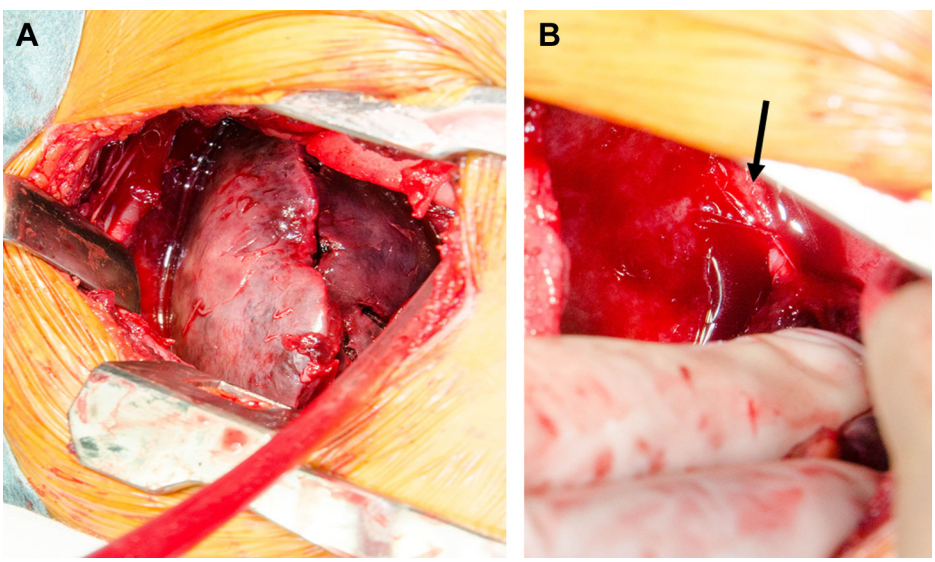

Fig. 2. (A) A dark-red colored sequestrum was identified at the latero-caudal part of the right pleural cavity. Dense adhesion between the sequestrum and surrounding tissues was found. (B) A fibrous structure which originated from the thoracic wall was detected. It was considered to be a remnant of the feeding artery (arrow).