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Case Report

Torsion of right lung sequestration mimicking a posterior mediastinal mass presenting as acute abdomen: Usefulness of MR imaging

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ABSTRACT

A 15-year-old boy with extralobar sequestration torsion is presented, who presented as an acute abdomen. Chest X-ray and computed tomography on admission revealed an apparent posterior mediastinal mass on the right side at the lower thoracic vertebral level. MR imaging, however, clearly showed scanty fluid around the mass and the subpleural fat layer between the vertebral body and the mass, suggesting its extrapulmonary and intrapleural cavity location. Its hemorrhagic nature was also suggested by the reduced signal on the in-phase as compared to out-of-phase chemical shift images, which helped make correct preoperative diagnosis.

Case report

A 15-year-old boy, who had been healthy and uneventful, was referred to our hospital for persistent severe coughing, and upper abdominal pain, which occurred during playing tennis. Laboratory test data on admission showed marked leukocytosis ($12.5 \times 10^3/\mu L$: normal range $3.3\text{-}8.6 \times 10^3/\mu L$) and slightly elevated lactate dehydrogenase (261 U/L: 124-222 U/L) and creatine kinase level (253 U/L: 59-248 U/L).

Gastroenteritis or other abdominal inflammatory processes were suspected clinically, however, chest and abdominal X-ray (Fig. 1) showed an apparent right paraspinal or posterior mediastinal mass at the lower thoracic vertebral level. Subsequently obtained contrast enhanced computed tomography (CT) (Fig. 2) revealed a soft tissue density mass adjacent to the vertebral body at the posterior mediastinum, showing no definite enhancement. There was no definite pleural effusion or abdominal abnormality, either. To further characterize the lesion, magnetic resonance imaging (MRI) was performed

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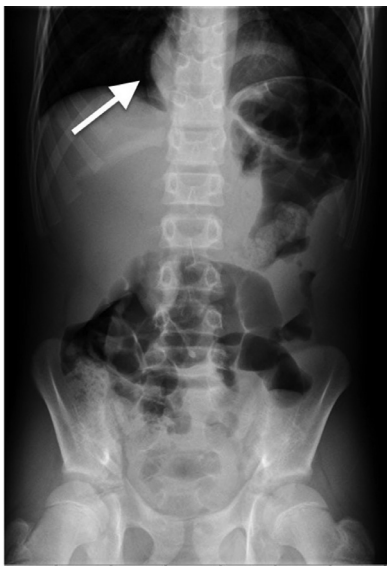

Fig. 1 – Abdominal X-ray. A smoothly marginated paraspinal mass is shown (arrow).

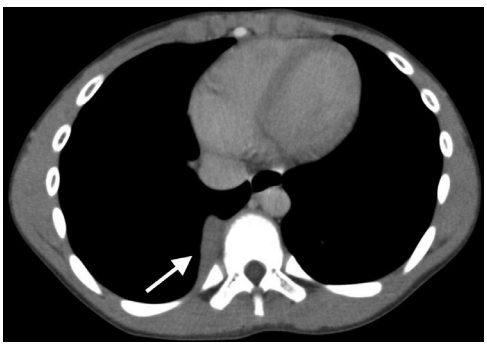

Fig. 2 – Delayed phase image of triple-phase contrast enhanced CT.
A well-defined 2 cm mass is noted on the right side of the vertebral body, mimicking a posterior mediastinal mass (arrow). There is no significant enhancement effect for the mass.

with a 1.5 T clinical unit (Ingenia, Philips medical systems, Netherland), revealing a well-defined mass of 3 cm in diameter at the right paraspinal region, showing low signal intensity both on T2- and T1-weghted images (Fig. 3A and B). Furthermore, on T2-weighted images, very high signal was observed surrounding the mass, suggesting the presence of fluid, probably representing small amount of pleural effusion (Fig. 3A); on the subtraction chemical shift images (CSI), on the other hand, the presence of subpleural fat layer was evident between the mass and the vertebral body (Fig. 3C). These 2 findings were considered to suggest the extrapulmonary and intrapleural location of the lesion. In addition, on subtraction CSI (Fig. 3C), the mass exhibited lower signal than the lung parenchyma, suggesting the presence of intralesional substance with strong susceptibility effect, namely hemorrhage.

Although there was no visualization of the aberrant vascular supply to the lesion on CT or MRI, preoperative diagnosis of extralobar sequestration (ELS) with torsion was made, and thoracoscopic resection of the lesion was performed. At surgery, a dark brownish mass was found, being attached to the mediastinal side via twisted vascular pedicle within the pleural cavity (Fig. 4). Unfortunately, origin of the aberrant vessels within the vascular pedicle could not be confirmed during the surgery. The vascular pedicle was ligated and resected, and the lesion was successfully removed. Microscopic survey of the resected specimen revealed hemorrhagic and necrotic pulmonary tissue, along with relatively thick-walled vessels, which suggested systemic circulation, rather than pulmonary vessels. Final diagnosis of infarcted ELS due to torsion was thus confirmed.

Discussion

Lung sequestration is a relatively rare congenital lung abnormality, which is divided into intralobar and extralobar subtypes, depending on the anatomical location of the lesions [1-5]. ELS is characterized by a separate pleural investment, whereas intralobar subtype is by an inclusion within the lung. ELS accounts for 25% of all lung sequestration, and occurs more frequently in male (male:female = 4:1). The majority occurs in the posterior costodiaphragmatic sulcus between the lower lobe and left hemidiaphragm [1,4]. ELS is often associated with other anomalies such as diaphragmatic herniation, bronchogenic cyst, or pericardial defect [2], and therefore is usually found in the neonatal period or early childhood. Without these anomalies, most of the patients can be completely asymptomatic, however, torsion of ELS is the one most important complication for these patients that requires prompt surgical treatment, because torsed ELS would result in infarcted, hemorrhagic, and necrotic lung tissue [1-12].

Because ELS is typically attached to the mediastinal side of the thoracic wall via only one stalk, which is the inferior pulmonary ligament containing aberrant systemic vessels (both artery and vein), it can be twisted within the pleural cavity typically during heavy physical exercise [6-12]. Symptoms are usually chest pain, coughing, and fever, but not infrequently include abdominal pain or shoulder to back pains [6-12]. In our case, the patient was playing tennis at the time of onset of symptoms, including upper abdominal pain, which is concordant to the previous descriptions.

For radiological diagnosis of sequestration, regardless of the subtypes, identification of the systemic supplying vessels is the key [1-5]. Systemic artery supplying ELS typically arises directly either from thoracic or abdominal aorta, and its draining vein is usually contiguous to the inferior vena cava or azygos system [1-5]. Rarely, cases with systemic arterial supply from small branches, including the left gastric, splenic, and intercostal arteries, or even those supplied by multiple branches of them, have been reported [2-4]. In case of ELS torsion, however, these vessels cannot be visualized due to cessation of the blood flow caused by the twist of the vascular pedicle [6-12]. Similarly in our

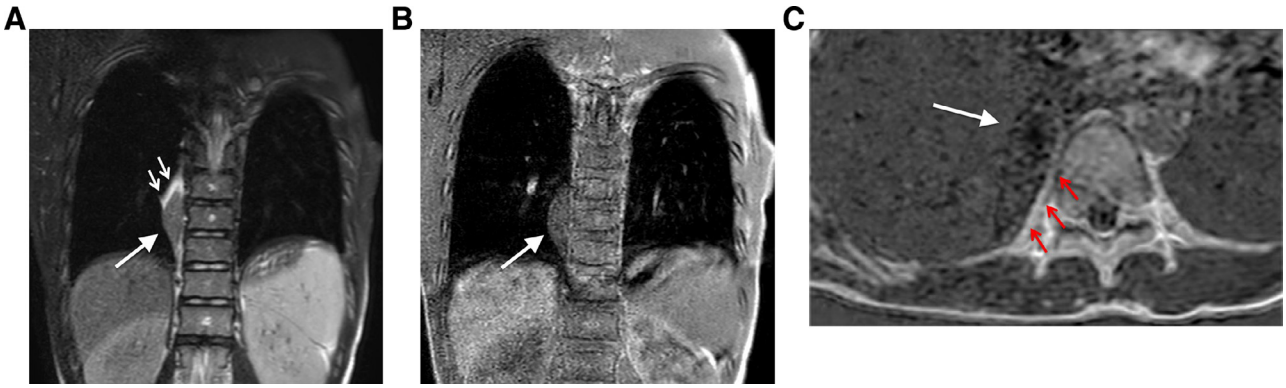

Fig. 3 – MR images obtained at a 1.5 T clinical unit.
(A) Coronal T2-weighetd fast spin-echo image with fat suppression (TR/TE = 1200/80 ms). The lesion shows hypointensity (arrow), surrounded by very high signal, indicative of small amount of pleural effusion (small arrows).
(B) Coronal T1-weighed gradient-echo image with fat suppression (TR/TE = 107.9/4.6 ms). The lesion shows hypointensity (arrow).
(C) Axial subtraction image of gradient-echo chemical shift images (TR/TE = 150.8/4.6 and 2.3 ms). High signal line is evident between the lesion and vertebral body, representing the presence of intact subpleural fat (small arrows), which suggest the lesion is located within the pleural cavity. Also note that the lower signal of the lesion as compared to the adjacent intact lung parenchyma (arrow), which suggests the presence of intralesional hemorrhage.

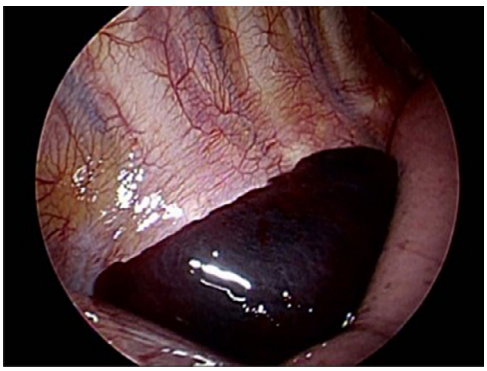

Fig. 4 – Laparoscopic view. Note dark brownish appearance of the lesion (arrow). 360 degree torsion of the vascular pedicle was confirmed during the surgery (not shown).

case, these aberrant vessels could not be confirmed on the preoperative imaging including contrast enhanced CT and MRI.

In the majority of the reported cases of ELS torsion, infarcted lesions are typically found within considerable amount of pleural effusion, and therefore one can readily diagnose the intrapleural cavity location of the lesion without difficulty, for example, on CT [8,12]. In our case, however, the amount of pleural effusion was so scanty that the lesion appeared as a posterior mediastinal mass on CT or on X-ray (Figs. 1 and 2). The reason why the pleural fluid is so little in this particular case, despite of the presence of leukocytosis, is unknown. On MR images, this scanty pleural effusion was clearly depicted on T2-weighted images surrounding the lesion, and subtraction images of the out-of-phase (OP) from in-phase images (IP) of CSI successfully visualized the subpleural fat layer between the lesion and the vertebral body (Fig. 3), indicating that the lesion location is "extrapulmonary and intrapleural."

In CSI, 2 echo times are set at 2.3 and 4.6 ms for OP and IP at 1.5 T unit, respectively, and signal intensity of OP should be smaller than that of IP when fat and water coexist within the same voxel due to cancelling effect of signals on OP [13]. Therefore, when the signal intensity of a lesion on the subtraction CSI is higher than that of the normal lung parenchyma containing air, the presence of small amount of fat is indicated, as shown for the subpleural fat layer in our case. Furthermore, the subtraction images of CSI works in another way. That is, when water and iron coexist within the voxel, signal intensity of IP should be smaller than that of OP due to the susceptibility effect of iron on IP. Therefore, when the signal on the subtraction image is lower than that of the normal lung tissue, the presence of iron or hemorrhage is strongly suggested [13,14], as confirmed for the torsed ELS (infarcted lung tissue) in our case.

Radiological differential diagnoses in this particular case include several posterior mediastinal masses, such as neurogenic tumors, duplicated cyst, or extramedullary hematopoiesis, which could be excluded based on the "extrapulmonary intrapleural" location of the lesion, as mentioned above. As for extrapulmonary intrapleural lesion, solitary fibrous tumor or other mesenchymal tumors may be considered [15], but little or no enhancement along with hemorrhagic nature as seen in the current case may be against these conditions. As an etiologically related disorder to ELS, congenital pulmonary airway malformation, or CPAM, may also be included in the differential diagnoses, which actually can coexist with lung sequestration [1-5]. CPAM may appear to be a solid mass, however, it usually is associated with aerated cystic component, at least in part of it [1-5], which was not seen in the current case.

In conclusion, radiologists should include ELS with torsion as a differential diagnosis when a young patient presents as an acute abdomen along with radiological findings suggesting a posterior mediastinal mass. Meticulous analysis of MR imaging may reveal characteristic features and precise location of ELS even in an atypical case as presented here, which may facilitate correct diagnosis and proper management.

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Declaration

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This case report has never been published or is not under consideration for publication elsewhere in English or other language. Informed consent was waived by our institutional review board in this case report. All the authors declare that there are no conflicts of interest. This study was not funded by any company.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2019.02.008.

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