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CASE REPORT

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A seven-month-old baby presenting excessive crying for pulmonary sequestration with torsion: A case report and literature review

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Abstract

Pulmonary sequestration with torsion is a rare condition. We describe a seven-month-old baby presenting excessive crying for pulmonary sequestration with torsion. Contrast-enhanced chest computed tomography demonstrated an oval-shaped mass in the posteromedial right lower chest, no systemic arterial supply was evident. The edge of the mass showed slight linear reinforcement, and its interior had no reinforcement. Thoracoscopic segmentectomy was carried out and histology confirmed pulmonary sequestration with torsion.

KEYWORDS
child, pulmonary sequestration, torsion

INTRODUCTION

Pulmonary sequestration (PS) is an uncommon congenital pulmonary malformation, it has been described as a cystic mass of non-functioning lung parenchyma that lacks a demonstrable connection to the tracheobronchial tree and receives its blood supply anomalously from the systemic circulation. It can be manifested as recurrent pneumonia, hemoptysis, dyspnea, or asymptomatic, occasionally found in prenatal or physical examination.[1] The two main variants are extralobar sequestrations (ELS) and intralobar sequestrations (ILS). ELSs are isolated from the remaining lung tissue, having their own pleural investment. ELS concurrent pedicle torsion is rare in clinic, and the symptoms are nonspecific, early identification difficulties, and easily misdiagnosed. Herein we report a 7-month old baby presenting excessive crying for ELS with torsion.

CASE REPORT

A 7-month-old previously healthy infant was brought to our hospital with the chief complaint of excessive crying for the last 2 days. The parents reported that about 2 days prior the infant suddenly appeared crying and restless, no vomiting and diarrhoea, occasional cough, no sputum, no nasal congestion and runny nose. Fever occurred 1 day ago, with the highest temperature of 38.1°C, without convulsions and chills. The chest radiograph of the external hospital showed pneumonia in the lower right lung, and amoxicillin and clavulanate potassium were given. But the crying became persistent, and all efforts to console the infant were futile. His vital signs were: pulse rate of 148 beats/min, respiratory rate of 36 breaths/min, temperature 38°C, and pulse oximetry of 96% in room air. The head, eye, ear, nose, and throat examination was normal. There were no skin rashes or ulcers, breath sounds were equal bilaterally and normal, and

abdominal examination was unremarkable with no organomegaly. There were no signs of meningitis, and the remaining neurological and musculoskeletal examination was normal. Cardiovascular examination was mild sinus tachycardia. The abdominal ultrasound was normal, and there was no evidence of urinary system infection. The patient's and his family histories were unremarkable, except that the right pulmonary sequestration was found during the prenatal examination.

A targeted right lower chest ultrasound was performed. The ultrasound confirmed a predominantly homogeneously echogenic solid mass in the right lower chest, which did not move with the lung on respiration. No vascular supply or internal vascularity within the mass could be demonstrated on Doppler evaluation (Figure 1). Adjacent consolidated lung and a small right pleural effusion were also identified.

To help further delineate the origin of the mass and determine whether a vascular supply was present, the contrast-enhanced chest computed tomography (CT) was performed. CT demonstrated an oval-shaped mass with average density of 68HU in the posteromedial right lower chest measuring 1.3 cm transverse 2.5 cm anteroposterior 2.2 cm craniocaudal. No systemic arterial supply was evident, the edge of the mass showed slight linear

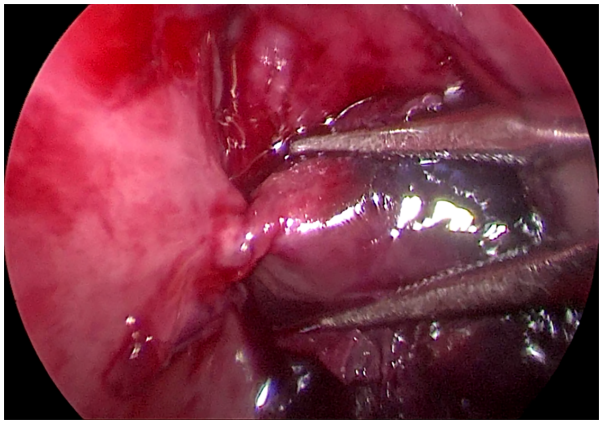

FIGURE 3 Intraoperative photo showing infarcted extralobar sequestration (S) and twisted feeding vessel stalk (arrow).

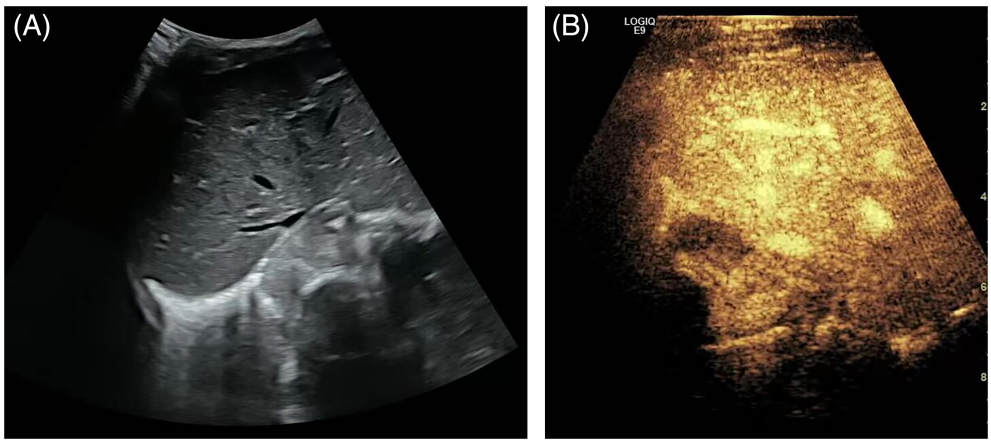

FIGURE 1 (A) Chest ultrasound demonstrates a predominantly homogeneously echogenic solid mass (arrow). (B) No vascularity or feeding vessel was discernible.

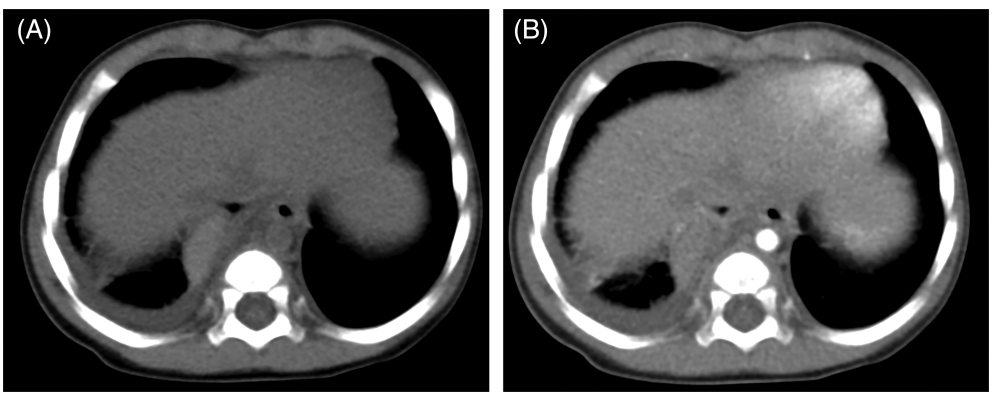

FIGURE 2 (A) Chest computed tomography showed a homogeneous solid mass in the right lower thorax (arrow), pleural effusion was also identified (E). (B) Contrast medium-enhanced chest computed tomography showed the edge of the mass with slight linear reinforcement and no reinforcement in its interior (arrow).

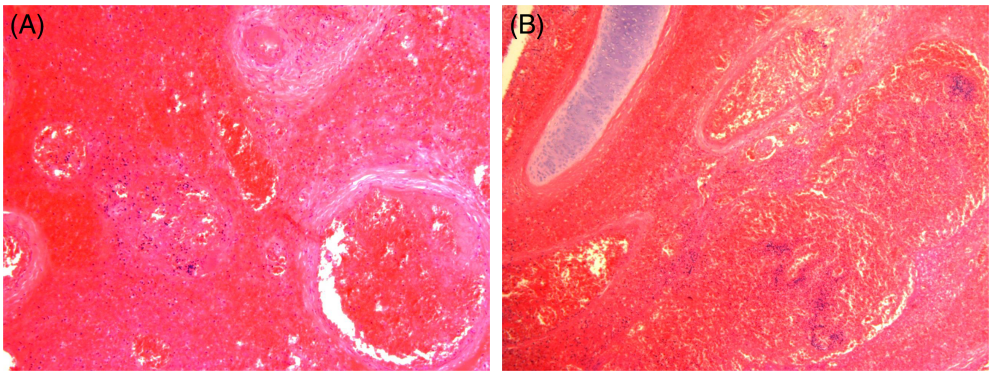

FIGURE 4 (A, B) Photomicrograph of the excised mass with recognizable lung structures showing hemorrhagic coagulative necrosis, consistent with extralobar sequestration.

TABLE 1 Summary of torsion extralobar pulmonary sequestration reported in literature.

No.	Authors	Sex	Age (y)	Chief complaint	Location	Main CT imaging	Feeding artery on image	Hydrothorax	Preoperative diagnosis
1	Chen et al.[9]	M	13	Abdominal pain	Left	Soft tissue mass	−	+	Neurogenic tumour
2	Lima et al.[10]	F	11	Abdominal pain	Left	A paraspinal mass	−	+	Pulmonary malformation
3	Choe and Goo[11]	M	10	Abdominal pain	Left	Hypodense soft tissue lesion	+	+	Torsion ELS
4	Kirkendall and Guiot[12]	M	13	Abdominal pain and Chest pain	Left	Nodule	+	+	−
5	Shah et al.[13]	F	11	Abdominal pain and Chest pain	Left	Soft-tissue density mass	−	+	−
6	Gawlitza et al.[7]	M	11	Abdominal pain	Left	Soft-tissue density mass	−	+	ELS
7	Huang et al.[14]	F	13	Chest pain	Left	Solid mass	−	+	−
8	Zucker et.al.[15]	M	6	Abdominal pain	Left	Soft-tissue density mass	−	+	−
9	Uchida et al.[16]	M	4	Abdominal pain	Left	Teardrop-shaped mass	−	+	ELS
10	Yang and Yang[3]	M	10	Chest pain	Left	Soft-tissue density mass	+	+	
11	Son et al.[2]	F	13	Abdominal pain and Chest pain	Left	A mass-like lesion	−	+	ELS
12	Yunxing Ti et al.[17]	M	7	Vomiting and abdominal distension	Left	Soft-tissue density mass	−	+	Neurogenic tumour/ELS
13	Yunxing Ti et al.[17]	M	3	Abdominal pain	Left	Soft-tissue density Mass	−	+	Lung consolidation
14	Yunxing Ti et al.[17]	M	5	Chest pain	Right	Soft-tissue density mass	−	+	Neurogenic tumour/ELS
15	Yunxing Ti et al.[17]	F	6	Abdominal pain and Chest pain	Right	Soft-tissue density mass	+	+	Torsion ELS
16	Yunxing Ti et al.[17]	F	10	Abdominal pain and Chest pain	Right	Soft-tissue density mass	−	+	ELS
17	Yunxing Ti et al.[17]	F	12	Abdominal pain and Chest pain	Right	Soft-tissue density mass	−	+	Torsion ELS
18	Walcutt, J et al.[18]	M	13	Abdominal pain	Left	A hyperdense mass	−	+	Torsion ELS

Note: "+" means presence and "−" means absence or not described or unclear diagnosis.
Abbreviations: CT, computed tomography; ELS, extralobar pulmonary sequestration; F, female; M, male.

reinforcement, and its interior had no reinforcement (Figure 2). The CT examination also demonstrated a small right pleural effusion, moderate right lower lobar consolidation. Video-assisted thoracoscopic surgery (VATS) was performed. The mass with intact capsule, congestion and necrosis was seen, accompanied by bloody pleural effusion and had an appearance suggestive of sequestration with torsed blood supply (Figure 3) and was resected. Pathological examination confirmed an ELS with haemorrhagic infarction (Figure 4). The patient recovered well postoperatively without complication and was discharged 3 days later.

DISCUSSION

ELS comprises 25% of all PS, the visceral pleura is not continuous with that of the regular lung, and the blood supply of an ELS is systemic and in approximately 80% of cases is from direct branches of the thoracic or abdominal aorta.[2] ELS is usually asymptomatic and found prenatally or incidentally in young children, torsion and infarction of ELS is extremely rare. To date, only 18 paediatric cases have been reported in the English literature (Table 1), the youngest is 3 years old. The chest pain or discomfort was the main symptom among adult patients of ELS with torsion,[3] whereas abdominal pain was the primary clinical manifestation in children (15/18), followed by chest pain (3/18). The location of PS is close to the diaphragm, when torsion necrosis occurs, the necrosis tissue and pleural effusion stimulate the diaphragm and cause abdominal pain. Our case was a 7-month-old baby, he could not express his pain in words, crying was his only way. Children presenting with excess or persistent crying with no physical examination clues, pose a diagnostic challenge to paediatrician. There are different reasons for persistent crying in children, including foreign body ingestion, urinary tract infections, intussusception et al.[4] We examined the patient carefully, cardiovascular examination and abdominal ultrasound were performed, but no positive results. The most important personal history is his prenatal discovery of the PS, then contrast-enhanced chest CT was performed, ELS with torsion was suspected.

Imaging examination is the main means to diagnose PS, multi-planar CT with 3-dimensional reconstruction has taken over as a better noninvasive diagnostic tool.[5] Definitive diagnosis requires the establishment of a systemic arterial supply and venous drainage of the sequestered lung tissue. But torsion of the feeding and draining vessels prevented visualization of the vascular pedicle resulting in atypical CT findings. The enhancement pattern of ELS with torsion is also variable. Our patient's torsed ELS presented mild peripheral enhancement, like the case reported by Takeuchi et al.,[6] the case reported by Gawlitza et al.[7] did not enhance following contrast administration. In summary, if there are school-age children with abdominal pain as the first or main manifestation, with or without chest pain, or a baby present excessive crying with personal history of PS, chest CT suggesting the following signs should be considered PS with torsion: (1) A soft-tissue density mass in the thorax paravertebral region; (2) The mass enhancing only on the periphery or not on CT; (3) Reactive pleural effusion or hemothorax; (4) No feeding artery identified. Surgical resection is main treatment for PS with torsion. VATS lobectomy is now accepted as an alternative surgical treatment to thoracotomy.[8] All the 19 cases underwent thoracoscopic surgery, the prognosis was well.

AUTHOR CONTRIBUTIONS

Li Li was involved in the investigation, methodology, writing of the original draft, and review and editing of the final manuscript. Yuanxiang Wang, Longwei Sun were involved in the conceptualization, formal analysis, and investigation. Wenjian Wang was involved in the conceptualization, investigation, and the methodology. All authors reviewed and approved the final manuscript.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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