



# Intradiaphragmatic pulmonary sequestration: advantages of the thoracoscopic approach

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**Abstract** Pulmonary sequestrations are accessory foregut lesions that are most commonly located within the thorax and occasionally in the abdominal cavity. Sequestrations arising within the diaphragm are exceedingly rare. We describe 2 patients found to have left peridiaphragmatic lesions on prenatal ultrasound and postnatal computed tomography. In the first patient, an initial laparoscopic approach was abandoned in favor of a thoracoscopic approach after no intraabdominal mass was found. The second patient had an uncomplicated thoracoscopic resection of a similar lesion. To our knowledge, these represent the first intradiaphragmatic pulmonary sequestrations to be resected via a minimally invasive approach. The rarity of these lesions makes definitive diagnosis without operative intervention challenging. Thoracoscopy appears to be a reasonable approach for resection of such intradiaphragmatic lesions.

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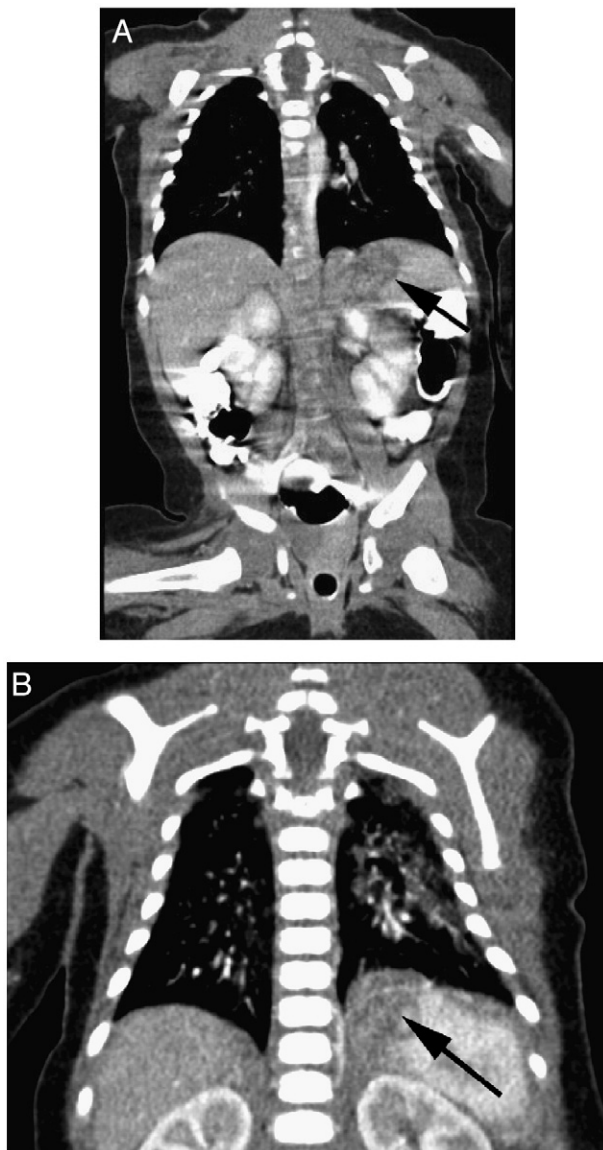
Pulmonary sequestration is a congenital anomaly that arises as a result of an accessory tracheobronchial bud originating from the primitive foregut. Intralobar sequestrations (ILSs) share the visceral pleura with the normal lung as compared with extralobar sequestrations (ELSs), which do not [1]. Extralobar sequestrations may be located inside (intrathoracic) or outside the thorax (ectopic). Although subdiaphragmatic ectopic ELSs are reported with some frequency, intradiaphragmatic ELSs have only been described twice before in the literature [2,3]. We present 2 cases of intradiaphragmatic ELS resected via thoracoscopy. The possible embryologic mechanisms for such entities are

considered as well as difficulties with radiologic localization to guide the operative approach.

## 1. Case 1

A 7-week-old female infant was referred for evaluation of a left-sided peridiaphragmatic lesion detected on prenatal ultrasound. Pregnancy and delivery were otherwise unremarkable. Postnatal ultrasounds could not definitively localize the mass relative to the diaphragm. A computed tomographic (CT) scan showed a  $2.0 \times 1.1 \times 1.8$  cm soft tissue mass separate from the paraspinal region and adrenal gland (Fig. 1A). No feeding or draining vessels could be identified. A review of all the diagnostic imaging was still

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**Fig. 1** Abdominal CT images showing peridiaphragmatic masses in left suprarenal area (black arrows) in case 1 (A) and case 2 (B).

unable to localize the mass, but discussion with radiologists concluded that the lesion was most likely located within the abdomen based on its proximity to the left adrenal gland. Urinary vanillylmandelic acid (VMA) and homovanillic acid (HVA) were within reference range.

The patient was brought for exploratory laparoscopy and resection. The stomach was mobilized, and the hiatus was exposed. Dissection proceeded to expose the spleen, pancreas, and left adrenal gland, all of which were normal. No mass was visualized in the abdomen, but a rounded density was noted upon inspection of the left hemidiaphragm, originating either within the diaphragm itself or above it. A decision was made to approach the lesion from the chest, and the abdominal ports were thus closed, and the patient turned onto her right side. Three 5-mm ports were placed in the left chest (fourth interspace in the midclavicular line, sixth interspace in the anterior axillary line, and eighth

interspace in the anterior axillary line) for a thoracoscopic approach. Pneumothorax at 5 mm Hg was used to allow identification of a bulge in the diaphragm. A combination of hook cautery and sharp dissection was used to dissect the lesion free from between the 2 diaphragm leaflets (Fig. 2). A small feeding vessel was noted and clipped. The residual diaphragmatic defect was closed with interrupted 3-0 silk sutures with intracorporeal ties and a chest tube placed. The chest tube was removed on the first postoperative day, and the patient was discharged the following day. Pathology was consistent with extralobar pulmonary sequestration.

## 2. Case 2

A 4-week-old male infant was referred for evaluation of a left lower lobe cystic pulmonary mass detected on prenatal ultrasound. Pregnancy and delivery were unremarkable. A CT scan showed a soft tissue mass in the left posterior costophrenic sulcus measuring  $2.3 \times 2.0 \times 1.7$  cm (Fig. 1B). No feeding or draining vessels were identified, and the location of the lesion relative to the diaphragm could not be determined. A subsequent ultrasound showed the mass to be separated from the adrenal gland by a well defined tissue plane, suggesting an intrathoracic lesion. Urinary VMA and HVA were within reference range.

The patient was taken to the operating room for thoracoscopic exploration. Right mainstem intubation and pneumothorax at 5 mm Hg were used to collapse the left lung. Four 3-mm ports were placed in the left chest (3 in the anterior axillary line approximately 1, 3, and 5 interspaces below the tip of the scapula, and 1 in the midaxillary line at level of the tip of the scapula), and a bulge was visualized in the diaphragm consistent with the location of the lesion noted on CT. The diaphragm around the mass was opened with electrocautery, and the mass was noted to be intradiaphragmatic and densely adhered to the crural fibers of the diaphragm. A plane was dissected on the retroperitoneal side of the mass, and it was noted to be separate from the kidney and adrenal gland. The mass was removed, the diaphragmatic defect was closed with interrupted 2-0 pledgeted polyester sutures, and a chest tube was placed. The chest tube was removed on the second postoperative day, and the patient was discharged the following day. Pathology was consistent with an intradiaphragmatic extralobar pulmonary sequestration.

## 3. Discussion

Pulmonary sequestration is a focus of nonfunctioning lung parenchyma that develops apart from the normal tracheobronchial tree. These anomalies account for up to 6% of congenital pulmonary malformations. Sequestrations are classified as ILS or ELS depending upon whether they

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