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Laparoscopic resection of an intradiaphragmatic pulmonary sequestration: A case report and review of the literature

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Abstract Extralobar pulmonary sequestrations have been occasionally described in the abdomen but rarely in the diaphragm. We present the case of a 10 month old girl with an intradiaphragmatic pulmonary sequestration. The minimally invasive operative technique is outlined in detail, including the combine use of laparoscopy and thoracoscopy. The case is discussed and the literature is reviewed.

A pulmonary sequestration is a mass of nonfunctioning lung tissue that is supplied by an anomalous systemic artery and only rarely has a bronchial connection to the native tracheobronchial tree. Approximately 25% of sequestrations are extralobar, and they may be found in the chest, within the diaphragm, or in the abdomen. Arterial blood supply may arise above or below the diaphragm, and the venous drainage is into the systemic or pulmonary venous circulation [1]. Although intradiaphragmatic pulmonary sequestration has been reported, we present a successful combined thoracoscopic and laparoscopic resection with detailed techniques and illustrations [2–4].

1. Case report

The patient is a full-term female who initially presented to the Pediatric Surgery clinic at 7 months of age with a left lower lobe lung lesion first noted prenatally. The prenatal diagnosis for the lesion was a congenital cystic adenomatoid malformation (CCAM). The mass was further evaluated with chest computed tomography (CT) that revealed a focal soft tissue density in the posterior medial left lower lobe which measured 12×28×36 mm (Fig. 1). There were two vessels extending from the upper abdomen to the mass. The diagnosis of an extralobar sequestration was made, but an intralobar lesion could not be excluded.

Immediately following presentation, the patient developed a respiratory syncytial virus (RSV) infection complicated by pneumonia. Her operation was postponed until 10 months of age, when her pulmonary issues had resolved. She underwent a left video-assisted thoracoscopy based on the presumed location of the lesion. She was prepped and positioned to facilitate exposure to both the chest and the abdomen in case rapid control of the aberrant artery was needed.

Intraoperatively, no mass was found in the left hemithorax (Fig. 2). Furthermore, no evidence of an aberrant arterial vessel was found. The patient did appear to have an eventration of the diaphragm, but there was no appreciable

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Fig. 1 Computed tomography revealing the extralobar sequestration.

mass effect to suggest opening the diaphragm would expose the mass. At this point, it was suspected the sequestration was located intraabdominally, so the decision was made to explore the patient laparoscopically. The patient underwent diagnostic laparoscopy and again no mass was found. The left hemidiaphragm did appear to have a mass located within it from this view, and a large aberrant vessel was visualized across the abdominal esophagus that entered the diaphragm through the left crus (Fig. 3).

The vessel was dissected and divided laparoscopically (Fig. 4). The vessel was then carefully followed distally to the level of the hemidiaphragm. The decision was made to explore the diaphragm laparoscopically, and the diagnosis of an intradiaphragmatic extralobar pulmonary sequestration was confirmed. The dissection proceeded meticulously, and two additional smaller vessels were clipped and divided. An area of dense adherence to the greater curvature of the stomach was divided laparoscopically with a stapler. The consistency was that of a bronchus. Upon successful resection of the mass (Fig. 5), a large diaphragmatic defect was noted associated with the eventration (Fig. 6), and this was repaired laparoscopically with silk sutures (Fig. 7A). Care was taken not to suture the lung by switching the camera back and forth between the abdominal and thoracic ports (Fig. 7B).

Pathology revealed an incomplete lobular architecture dominated by a confusion of variably dilated and collapsed distal airways lined by ciliated columnar respiratory epithelium. An intact bronchus coursed through the middle of the specimen and was composed of ciliated columnar respiratory epithelium, lamina propria with a few unremarkable submucosa glandular elements, and hyaline cartilage all showing proper anatomic relationships. Thick-walled muscular vessels accompany the bronchus and were present in the surrounding interstitium.

The patient was admitted postoperatively to the pediatric intensive care unit, immediately extubated, and discharged home tolerating a regular diet on the first postoperative day. She remains asymptomatic in good health six months postoperatively.

2. Discussion

The management of sequestrations is controversial in the asymptomatic patient. Some authors have reported successful observation of these lesions without resection [1]. Others, however, have advocated extirpation due to the risk of infection, malignant degeneration, and pathologically defining the lesion, especially if the lesion is extrathoracic [3]. Additionally, mixed CCAM and sequestration-hybrid lesions have been reported, as well as mixed bronchogenic cyst and sequestration hybrid lesions [3]. There is even a report of a lesion with mixed intralobar and extralobar components [3]. The long-term sequelae of such entities are unknown.

The indications for operation in this case were multiple. The patient's CT delineated a presumed extralobar sequestration, but it was felt to be intrathoracic upon review. An intralobar

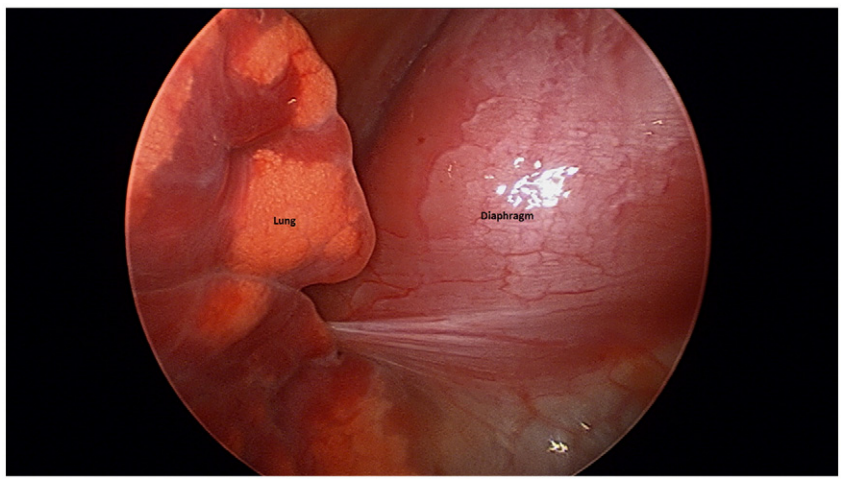

Fig. 2 Intrathoracic view revealing no evidence of a congenital anomaly.