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Ectopic Lung Tissue: An Unusual Extra-Lobar Pulmonary Sequestration

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ABSRACT

Pulmonary sequestration is a relatively rare pulmonary congenital malformation. It consists of an isolated lung segment that lacks communication with the tracheobronchial tree and receives its own blood supply through the systemic circulation, most commonly from the descending thoracic aorta. We report an unusual case of extra-lobar pulmonary sequestration presenting with an associated large mucocele and supplied by aberrant branches of the intercostal vessels. It was successfully resected through 2 cm incision uniportal video assisted thoracoscopic surgery.

KEYWORDS

Pulmonary sequestration; Extra-lobar; Intercostal vessels

INTRODUCTION

Pulmonary sequestration is a rare clinical entity, and represents less than 2% of congenital anomalies. It is defined as a malfunctioning lung tissue that has no connection with the lung tracheobronchial elements. Thoracic and abdominal aortas are the main source of its arterial blood supply. The sequestration could be intra-lobar (ILS), the most common type, if it lies within pleural layer surrounding the lobar lung, or extra-lobar (ELS) if it has its own pleura completely separated from adjacent normal lung. We present a case of ELS that was diagnosed intra-operatively, pedicled to aberrant branches of the intercostal vessels and secreting viscous mucous collected in a large dual-contour cyst.

CASE REPORT

A 28-years old man previously healthy was referred to our department after an incidental finding of a posterior mediastinal mass that appeared first on chest X-Ray. The patient was asymptomatic, the physical exam was unremarkable and the laboratory work-up was in the normal range. The patient also had a Magnetic Resonance Imaging (MRI) of the chest showing a 63 mm × 36 mm × 44 mm right sided sub-pleural paravertebral mass at T9-10 level, abutting the costovertebral angle, exerting mild mass effect by displacing the lung parenchyma. The mass is well encapsulated, septate with a dual-contour shape, of decreased signal on T1 sequence and increased signal on T2 sequence images, with no enhancement after contrast (Figure 1).

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No sub-pleural pulmonary tissue was described on the imaging. Considering the location and characteristics of the mass, cystic Schwannoma was the most probable diagnosis.

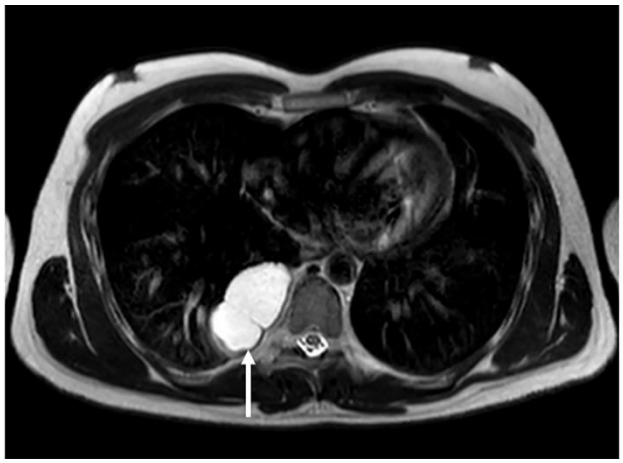

Figure 1: MRI showing the right paravertebral dual-contour cystic mass with increased intensity on T2-weighted sequence.

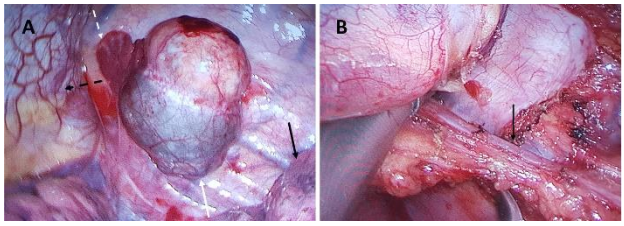

Figure 2: (A) Endoscopic view showing the ELSwith its two components: the cyst (white arrow) and the parenchymal tissue (dashed white arrow) located between the right lower lobe (black arrow) and the diaphragm (dashed black arrow). (B) Endoscopic view showing the irrigating vessels arising from the intercostal pedicle.

The patient was referred to us for surgical resection. The mass was approached through uniportal video-assisted thoracoscopic surgery (VATS); a 2 cm incision in the middle of the right fifth intercostal space was performed. The tumor was found at the postero-basal costo-vertebral groove. It had a dual-contour cystic shape in contact with extra-pulmonary lung parenchyma (Figure 2A). The surrounding pleura was opened using hook cautery, and its chest wall bed was released. It remained pedicled to small artery and vein that took origin from the intercostal vessels (Figure 2B). The pedicle was cauterized and cut. During retrieval of the mass in an endoscopic bag, the cyst ruptured; it contained viscous mucous (Figure 3). The patient recovered well and was discharged home on the second post-operative day. The pathology result was consistent with lung parenchymal tissue and a ruptured mucocele.

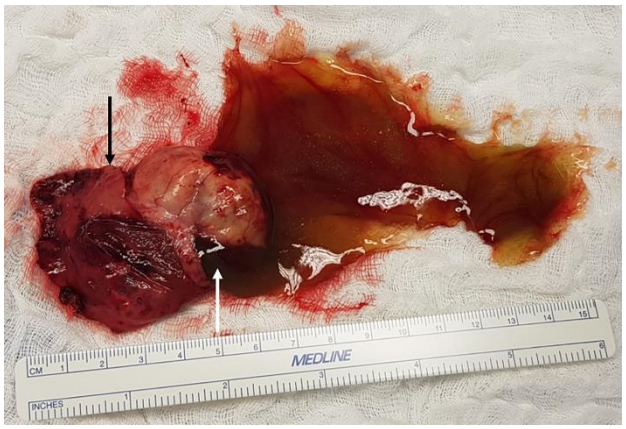

Figure 3: Parenchymal lung tissue (black arrow); ruptured mucocele with viscous mucous (white arrow).

DISCUSSION

Pulmonary sequestration (PS) is a rare congenital malformation where a non-functional lung tissue develops without any communication with the tracheobronchial tree and receiving its blood supply from aberrant systemic vessels. PSs are classified into intra-lobar (75%) or extra-lobar (25%) according to their pleural investment [1]. Extra-lobar pulmonary sequestration (ELS) has its own visceral pleura and it is separated from the normal lung tissue, whereas intra-lobar pulmonary sequestration (ILS) shares the same visceral pleura with the adjacent lung parenchyma [2]. Patients with ELS are often asymptomatic. The gold standard imaging tool for diagnosis is the computed tomographic angiography that can clearly show the origin of the aberrant feeding vessel from the systemic circulation. This is also true for the MRI or the Magnetic Resonance Angiography [3]. In majority of cases, the arterial supply comes from the descending thoracic aorta followed by the abdominal aorta [2]. In our patient, ELS was misdiagnosed as a posterior mediastinal tumor. The MRI showed a cystic mass due to the long-term mucous secretions in a blinded bronchial lumen; Also, it couldn't identify the feeding artery that wasintra-operatively found to be a branch of the intercostal artery which seldom happens [4].

Regardless of the presence of symptoms, surgical resection of the PS remains the preferred contemporary management to prevent possible future complications including recurrent infection and hemorrhage [5]. It may be managed through uniportal VATS in selected cases such our patient.

Although ELS is rarely reported in the right thoracic cavity, its' association with mucocele and aberrant vascularization from intercostal bundle makes it noteworthy.

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