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## Intra-abdominal pulmonary sequestration as an exceptional cause of abdominal mass in the adult<sup>☆</sup>



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## ABSTRACT

**INTRODUCTION:** Pulmonary sequestration (PS) is an extremely rare malformation defined as a portion of lung tissue isolated from the pulmonary system. PSs are classified into intralobar type and intra-abdominal PS that represents only 2.5% of cases. There are 20 cases of PS reported in adults and only two were managed by laparoscopic approach. We report a case of intra-abdominal PS mimicking a gastroesophageal duplication cyst in an adult. Besides its rarity, this is the first intra-abdominal PS in an adult managed by an anterior laparoscopic approach.

**PRESENTATION OF CASE:** A 60-year-old female patient had had epigastric and left upper quadrant pain for several days. Physical examination was normal. Image test were consistent with a gastroesophageal duplication. The patient was taken to the operating room for laparoscopic exploration and resection. The pathological diagnosis was extralobar pulmonary sequestration.

**DISCUSSION:** Less than 20 cases of PS have been reported in adults and only two cases were managed by a lateral laparoscopic approach. In contrast to these reports, we used an anterior approach due to the GEJ suspected origin of the mass.

**CONCLUSION:** Extralobar intra-abdominal PS is an extremely rare condition during adulthood but this diagnosis should be included in the differential diagnosis of a left-sided suprarenal mass. Due to the difficulty in achieving a definitive preoperative diagnosis, surgery is recommended. Laparoscopic resection is safe and effective but careful preoperative imaging studies are recommended in order to plan the most suitable approach.

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A 60-year-old female patient had had epigastric and left upper quadrant pain for several days. Her medical history was irrelevant. Physical examination was normal. Abdominal ultrasound revealed a left suprarenal mass. Computed tomography showed a 7 × 4 cm cystic soft tissue mass with internal calcification just above the left adrenal gland and beneath the diaphragm (Fig. 1). Biochemical testing was performed and showed normal plasma levels of adrenocortical hormones and catecholamines. Abdominal magnetic resonance imaging showed a 7.5 × 6 × 4 cm cystic mass possibly arising from the gastroesophageal junction (GEJ) and adjacent to but independent from the adrenal gland, spleen, pancreas and stomach. Vascularization was not detected (Fig. 2). Although no other diagnosis could be excluded, gastroesophageal duplication was suggested. This diagnosis was also suggested by an upper

endoscopic ultrasound that revealed a 7 cm cystic mass arising from the posterior wall of the GEJ.

After discussing the diagnosis, benefits, risk and alternatives with the patient, she was taken to the operating room for laparoscopic exploration and resection.

The laparoscopic procedure was performed in the supine decubitus position. A Hasson trocar was placed in the umbilicus. Under direct vision, a 12 mm trocar was placed in the left upper quadrant and 5 mm trocars were placed in the right quadrant, left subcostal margin and epigastrium. Upon inspection of the abdomen no obvious lesions were found. After gastrosplenic and short vessels freed, the mass was evident. Using the ultrasonic coagulating shears the lesion was dissected from the spleen, pancreas, left diaphragm, left crus, abdominal aorta and gastric posterior wall. The GEJ was dissected and isolated completely. The mass was attached to the posterior aspect of the GEJ and the resection was completed by stapling this tissue.

The pathological diagnosis was extralobar pulmonary sequestration.

### 1. Discussion

Pulmonary sequestration (PS) is an extremely rare malformation defined as a portion of lung tissue isolated from the pulmonary

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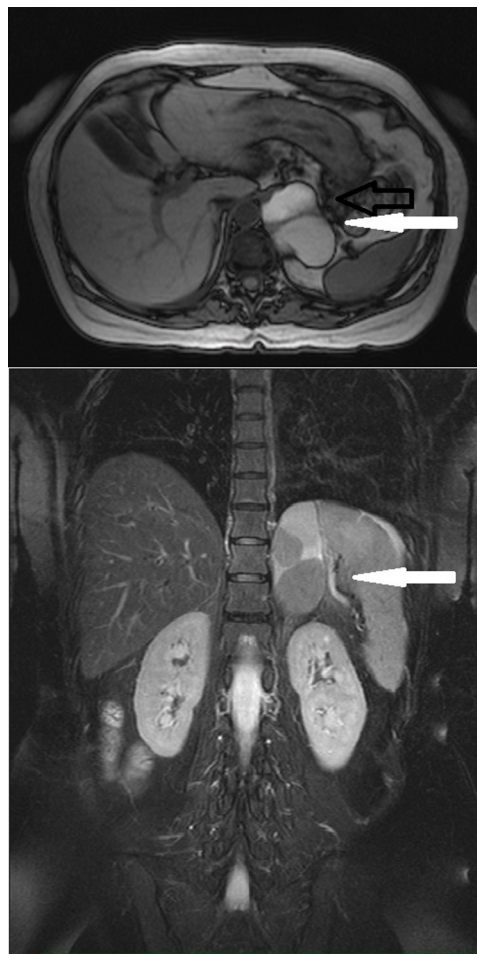
**Fig. 1.** Computed tomography showed a 7 × 4 cm left subdiaphragmatic hyperdense mass with internal calcification which was isolated from left adrenal gland.

system.<sup>1</sup> This condition, which arises from the primitive foregut, has no bronchial connections and its vascular supply can originate in the aorta, abdominal visceral vessels or coronary system.<sup>2</sup> PSs are classified into two types. The intralobar type has a segment of normal lung, while the extralobar type has a completely separated pleura covering of lung tissue and can be supra- or subdiaphragmatic. The latter is called intra-abdominal PS and is the least common subtype (2.5%).<sup>3</sup>

As a congenital malformation, PSs are usually diagnosed in the pediatric age.<sup>4</sup> They may remain asymptomatic initially and are usually discovered accidentally in imaging studies during adulthood, when adrenal disease is often suspected.<sup>2–4</sup>

Although fewer than 20 cases of PS have been reported in adults, this condition should be included in the differential diagnosis of a left-sided suprarenal mass.<sup>2</sup> As in our case, the evaluation should include imaging studies and biochemical testing to exclude adrenal functional tumors. The radiological features usually include a left-sided infradiaphragmatic cystic mass with calcified areas and its vascularization can be demonstrated,<sup>3</sup> which could not be proven in our case. However, although computed tomography and magnetic resonance imaging can suggest the diagnosis of PS, these techniques do not provide a conclusive diagnosis of extralobar PS.<sup>5</sup> A more accurate diagnosis can be made by percutaneous tissue biopsy.<sup>6</sup> However, the mass is usually located in an area not easily amenable to percutaneous approach.<sup>7</sup>

There are no cases of malignant transformation reported in adults but surgery is usually required to obtain a definitive diagnosis.<sup>2</sup> Only two cases of laparoscopic resection of a PS in an adult have been reported previously and both were



**Fig. 2.** A magnetic resonance imaging showed a 7.5 × 6 × 4 cm cystic mass, hyperintense on T1 and hypointense on T2.

accomplished via the lateral approach commonly used to perform a laparoscopic left adrenalectomy.<sup>2,4</sup> In contrast to these reports, we used an anterior approach due to the GEJ suspected origin of the mass and it was not necessary to remove the adrenal gland.

In summary, extralobar intra-abdominal PS is an extremely rare condition during adulthood but this diagnosis should be included in the differential diagnosis of a left-sided suprarenal mass. Due to the difficulty in achieving a definitive preoperative diagnosis, surgery is recommended. Laparoscopic resection is safe and effective but careful preoperative imaging studies are recommended in order to plan the most suitable approach.

#### Conflict of interest

None of the authors have any conflict of interest.

#### Funding

None.

#### Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of this written consent is available for review by the Editor-in-Chief of this journal on request.

### Author contributions

C. Moreno-Sanz, M.L. Herrero-Bogajo and J. Picazo-Yeste performed the surgery.

C. Moreno-Sanz wrote the manuscript.

A. Morandeira-Rivas, M. Manzanera-Díaz and C. Sedano-Vizcaino were involved in the management and follow up of the patient. The later were also involved in data collection and edition.

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