



CASE REPORT

A CASE OF EXTRALOBAR PULMONARY SEQUESTRATION: POSTERIOR MEDIASTINAL HYPERVASCULAR CYSTIC LESION MIMICKING VASCULAR TUMOR

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ABSTRACT

Hypervascular lesions in the posterior mediastinum can pose diagnostic challenges as they resemble various entities, including vascular tumors and neurogenic tumors. One frequently overlooked possibility is extralobar pulmonary sequestration (ELS), a congenital abnormality of non-functional lung tissue with blood supply from systemic arteries and venous drainage to the systemic venous system. This study aims to report a case of ELS in an adult patient that radiologically resembled a vascular mediastinal tumor. The research done by using case study with clinical evaluation, imaging using contrast-enhanced MSCT thorax, and histopathological confirmation. The outcomes of this study are that an asymptomatic adult patient was found to have a hypervascular cystic mass in the posteroinferior region of the left lower lobe with blood supply from the left posterior intercostal artery and drainage to the azygos and hemiazygos venous systems. No connection with bronchial branches was observed. Histopathological imaging reinforced the ELS diagnosis. From the findings it can be concluded that ELS diagnosis should be considered in cases of hypervascular posterior mediastinal masses, especially those with vascular supply from systemic arteries and without bronchial communication. Contrast-enhanced thoracic MDCT examination is crucial for establishing diagnosis, preventing unnecessary invasive procedures, and supporting appropriate management.

Keywords: Extralobar Pulmonary Sequestration, Mediastinum Posterior, Lesi Hipervaskular, MSCT Toraks, Diagnosis Banding Tumor Vaskular

INTRODUCTION

Vascular lesions in the posterior mediastinum are relatively rare findings that can pose diagnostic challenges as they resemble various entities such as hemangiomas, arteriovenous malformations (AVM), and vascular neurogenic tumors.¹⁻³ Hypervascular posterior mediastinal masses can also manifest from congenital abnormalities rarely considered in adults, one of which is extralobar pulmonary sequestration (ELS).^{4,5}

ELS is a congenital abnormality consisting of non-functional lung tissue that lacks communication with the normal tracheobronchial system and receives blood supply from systemic arteries with drainage to the systemic venous system, such as the azygos or hemiazygos veins.⁵ Although more commonly

diagnosed in infancy or childhood, ELS can also appear incidentally in adults as a hypervascular posterior mediastinal mass, which may be misinterpreted as a neoplastic or other vascular process.^{4,6}

Imaging plays a crucial role in establishing diagnosis, particularly with contrast-enhanced multidetector computed tomography (MDCT) of the thorax, which allows clear visualization of unusual feeding arteries and draining veins.⁷ Accurate identification through imaging is important to avoid unnecessary invasive procedures and to plan appropriate interventions, such as embolization or surgical resection.

PULMONARY SEQUESTRATION

Pulmonary sequestration is a congenital

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malformation consisting of non-functional lung tissue that lacks connection to the bronchial system and receives supply from systemic arteries.^{4,5} PS is divided into two types: intralobar sequestration (ILS), located within normal lung tissue without its own pleural layer, and extralobar sequestration (ELS), which exists outside the main lung and has separate pleura.^{8,9} The extralobar type (ELS) is usually separated from normal lung by its own pleura, with a characteristic vascular pattern consisting of supply from the aorta or its branches and drainage to the systemic venous system.^{10,11}

EPIDEMIOLOGY

ELS is a rare form of pulmonary sequestration rarely found in adults and often detected incidentally.^{12,13} The most common location is the posteroinferior region of the left lower lobe. With increasing use of imaging such as CT angiography, incidental detection of ELS is increasing despite patients showing no respiratory symptoms.^{8,14-16}

In the past decade, prenatal diagnostic techniques for ELS have rapidly developed, particularly through fetal ultrasound and fetal MRI. Ultrasonographic diagnosis can be performed from the second trimester of pregnancy (around 16-22 weeks), showing a sharply defined homogeneous echogenic mass, usually supradiaphragmatic, with visible systemic veins or arteries leading to the lesion (especially from the aorta) when using Color Doppler.^{17,18} Meanwhile, fetal MRI (including MRA) becomes an important complement¹⁹, especially when ultrasound is unclear or complex abnormalities are present. On T2-weighted images, ELS appears as a homogeneous hyperintense mass with recognizable systemic vascular supply without ionizing radiation use.¹⁷ MRI efficacy in differentiating BPS from other congenital lesions such as CCAM is also high - one study reported prenatal MRI accuracy of 15/16 cases, including several ELS with vascular supply visualization as an important diagnostic feature.²⁰

DIAGNOSIS IN ADULTS

Accurate diagnosis of ELS in adult patients, as in this case, is a crucial clinical aspect considering ELS is very rare (about 15-25% of all pulmonary sequestration cases) and often asymptomatic in adulthood.²¹ Clinical challenges arise because adult ELS manifestations can resemble mediastinal neoplasms, AVMs, or cysts, as reported in a case where unusually located ELS lesions led to premature surgical exploration due to suspected malignancy.²²

Modern imaging modalities, particularly multiphasic CT angiography, are important diagnostic tools as they enable identification of systemic arterial supply

and venous drainage - characteristic features of ELS that distinguish it from other thoracic lesions.²³ CT angiography has proven to provide up to 90% accuracy in establishing PS diagnosis, and visualization of abnormal vasculature can avoid needle biopsy procedures, which are highly risky in hypervascular lesions like ELS.^{8,21} MRI and MR-angiography become useful additions when CT is unclear or in patients requiring radiation protection, although CT remains superior in displaying cystic and vascular details.²¹

The misdiagnosis rate is quite high: in one adult case series, presurgical ELS diagnosis was achieved in only 9% of cases through imaging, with the remainder detected only during surgery.^{22,24} Additionally, in another series of 25 adult patients, several ELS cases were initially misdiagnosed as fibrous pleural tumors or bronchogenic cysts until postoperative histological confirmation.²⁵ Such diagnostic errors can potentially lead patients to unnecessary invasive procedures, high bleeding risks, or even inappropriate therapy.

Accurate diagnosis enables safer and more appropriate management decisions: for example, conservative evaluation in asymptomatic adult patients, early feeding artery embolization as a minimally invasive alternative before surgery, or structured resection planning through VATS or thoracotomy with consideration of already mapped vascular supply.²⁵ Without proper diagnosis, there are risks of serious complications such as massive hemoptysis, spontaneous hemothorax, or severe infections that are difficult to manage.^{22,25}

ANTENATAL DIAGNOSIS

Extralobar sequestration (ELS) is non-functional lung tissue that does not communicate with the bronchial tree and receives blood supply from abnormal systemic arteries, usually originating from the thoracic aorta or its branches. ELS is wrapped in its own pleura, distinguishing it from the intralobar form both anatomically and clinically.^{11,26} In rare cases, ELS can be found in the posterior mediastinum and display hypervascular characteristics with cystic components. On postnatal imaging such as CT or MRI, this condition often causes diagnostic difficulties as it resembles vascular mediastinal masses such as hemangiomas or neurogenic tumors.^{11,27}

Postnatal CT angiography, particularly with multidetector technique (MDCT), is the primary imaging modality for detailed identification of feeding arteries and draining veins. The characteristic feature of ELS is the presence of one or several feeding arteries originating from the descending aorta or posterior intercostal arteries with venous drainage system toward systemic veins such

as azygos or hemiazygos. This pattern is very helpful in differentiating it from mediastinal vascular tumors that generally show diffuse vascularization patterns with low-resistance flow without clearly defined single feeding arteries.^{17,28}

Specifically, the presence of cystic components can cause diagnostic confusion with CPAM (Congenital Pulmonary Airway Malformation) or bronchogenic cysts, while its location in the posterior mediastinum increases the risk of misdiagnosis as a neurogenic tumor. Several case reports show that adult ELS can even remain asymptomatic and be misdiagnosed as mediastinal tumors for years before finally being diagnosed through CT angiograph.^{27,29}

THORACIC MSCT EXAMINATION

Multislice Computed Tomography (MSCT) of the thorax is an advanced imaging method that utilizes multidetector technology to produce thin slice images with high acquisition speed. This technique can provide detailed three-dimensional (3D) reconstruction, making it very useful in evaluating lung, mediastinal, pleural, and thoracic wall structures.^{30,31}

In cases of extralobar pulmonary sequestration (ELS), the role of thoracic MSCT is vital as it can detect feeding arteries originating from systemic sources (such as posterior intercostal arteries) and venous drainage to the azygos/hemiazygos system - which becomes the diagnostic key to differentiate it from vascular or neurogenic tumors.

The advantages of MSCT compared to conventional CT lie in the ability to acquire multi-slice simultaneously, enabling multiplanar reconstruction (MPR) and three-dimensional techniques such as volume rendering and virtual bronchoscopy. This technology can also scan the entire thoracic cavity in seconds, reducing artifacts due to respiratory or cardiac motion, making it very beneficial for critical patients or those who have difficulty holding their breath.^{32,33}

Thoracic MSCT examination protocols typically use slice thickness of 3-5 mm with sub-millimeter reconstruction capabilities (≤ 1 mm) to detect small abnormalities in lung parenchyma. Research by Bisra et al. showed that 2 mm slice thickness provides the best image quality for detecting lung lesions, with statistically significant results ($p < 0.05$).³⁴ Intravenous contrast media administration, generally 80-100 ml iodine with injection speed of 2-4 ml/second, is performed in the vascular phase with approximately 35-45 second delay for vessel and vascular lesion evaluation.³⁵

In clinical practice, thoracic MSCT has an important role in early detection of lung tumors, small nodules, and evaluation of interstitial disease, pulmonary embolism, aortic aneurysms, and other vascular

diseases. The main advantages of MSCT are speed and high resolution, but the challenge faced is relatively higher radiation dose, requiring parameter optimization and use of iterative reconstruction algorithms to reduce exposure without reducing image quality.³⁶

CASE DESCRIPTION

A 35-year-old female was hospitalized for dengue hemorrhagic fever (DHF), without accompanying respiratory complaints. During routine thoracic imaging evaluation as part of supportive examination during treatment, an incidental discovery was made of a previously asymptomatic thoracic mass. Contrast-enhanced and non-contrast thoracic MSCT examination performed on April 23, 2025, showed a large well-defined cystic lesion ($\pm 10.0 \times 9.6 \times 10.1$ cm) located in the posteroinferior region of the left lower lobe. The lesion appeared hypervascular with necrotic components (22 HU), hemorrhage (78 HU), and significant contrast enhancement (126 HU) in the vascular portion.

Vascularly, the lesion received supply from the left posterior intercostal artery and drainage to the azygos and left hemiazygos venous systems. No connection was found between the lesion and bronchial branches, and the lesion appeared attached to surrounding structures such as the descending aorta, left visceral pleura, left hemidiaphragm, and posterolateral hemithorax wall. Additionally, mild bilateral pleural effusion and compression atelectasis in the right lower lobe were found, along with other lung abnormalities including fibrosis and mediastinal lymphadenopathy.

These radiological findings are very characteristic of extralobar pulmonary sequestration (ELS), particularly with identification of systemic arterial supply and drainage to the systemic venous system, without bronchial communication. ELS diagnosis was established based on a combination of clinical, radiological data, and post-resection histopathological confirmation. This finding emphasizes the importance of systematic advanced imaging examination, even in patients without respiratory symptoms, to detect rare congenital abnormalities such as ELS that can resemble neoplastic masses in the posterior mediastinum.

INVESTIGATION

The patient in this case was diagnosed with extralobar pulmonary sequestration (ELS), one form of PS that has separate pleura and systemic vascular supply, commonly found incidentally in adults.

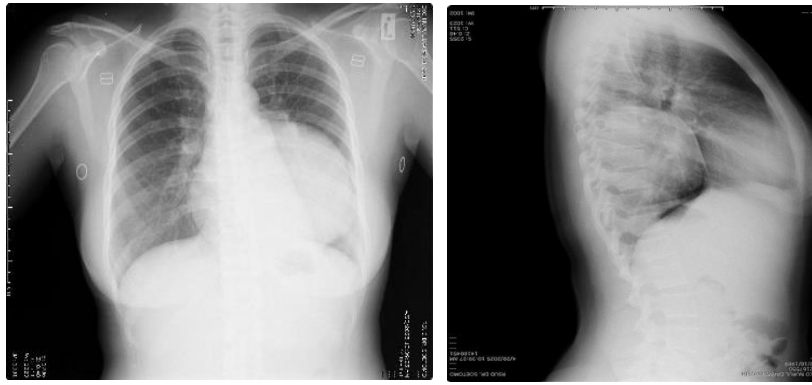


Figure 1. Chest X-ray

Embryologically, ELS is thought to originate from an accessory lung bud that separates from the primary airway during the pseudoglandular phase, between the 5th and 17th weeks of gestation. This bud develops independently outside the main lung, forming non-functional lung tissue with separate pleura and blood supply from systemic arteries.³⁷⁻³⁹

The most common ELS location is the left lower lobe (56-77%). Most feeding arteries originate from the thoracic aorta (54-87%).^{12,40} Venous drainage can go to pulmonary veins or the systemic venous system (azygos/hemiazygos).³⁷ Clinical symptoms in adults are generally mild or absent - only cough, hemoptysis, or chest pain are found in intralobar PS. ELS wrapped by its own pleura rarely becomes infected, so the frequency of asymptomatic presentation is higher.⁸

Chest X-ray examination shows the presence of a faint homogeneous opacity shadow in the lower left hemithorax that obscures part of the left diaphragmatic silhouette. This finding does not show specific characteristic signs of specific lung abnormalities but may raise suspicion of mediastinal mass or congenital lung lesions such as pulmonary sequestration. In ELS, X-rays often do not provide definitive diagnosis because the lesion does not communicate with the bronchial tree and only appears as a non-specific mass or shadow in the

posterior or basal lung area. Therefore, this X-ray examination becomes the basis for further imaging with thoracic MSCT/CTA, which is the gold standard method for visualizing systemic arterial supply and venous drainage in PS.^{41,42}

In this case, the adult patient was found to have a well-defined, hypervascular posterior mediastinal lesion with cystic, hemorrhagic, and necrotic components, along with arterial supply from the left posterior intercostal artery and drainage to the azygos and hemiazygos venous systems. These characteristics are very typical of ELS. The lesion's location attached to the descending aorta, visceral pleura, and hemidiaphragm without connection to bronchial branches reinforces this suspicion.

In ELS, non-functional lung tissue has its own pleura (separate from normal lung), does not communicate with the bronchial tree, receives supply from systemic circulation, and generally drains to systemic veins such as azygos or hemiazygos veins; this drainage pattern actually becomes a marker that distinguishes it from ILS which commonly drains to pulmonary veins. This finding showing feeding arteries originating from posterior intercostal rather than directly from aorta has also been described in cases of posterior mediastinal ELS in adults, thus reinforcing its pathoanatomical coherence.²⁷

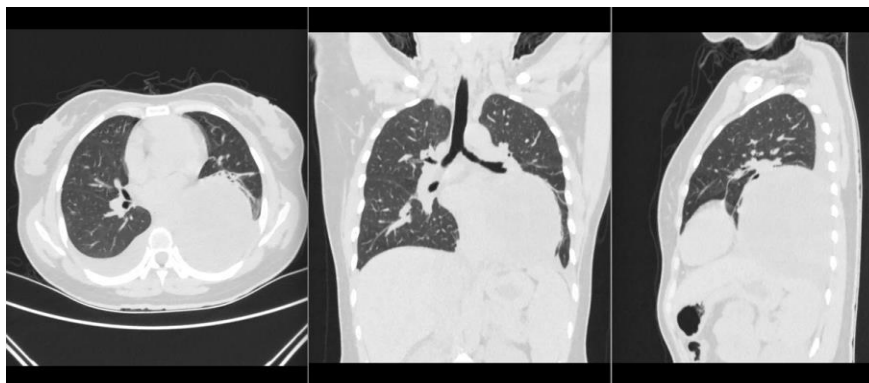


Figure 2. Thoracic MSCT results in three reconstruction views (axial, coronal, and sagittal)

Based on the multi-planar chest CT images (axial, coronal, and sagittal) displayed, there is a large cystic

mass in the posteroinferior region of the left lower lobe, with clear boundaries, heterogeneous

consistency, and mass effect on surrounding structures. The mass appears hypodense with varying density, reflecting necrotic components (low HU values) and hemorrhagic areas (higher HU values), consistent with the previous description from thoracic MSCT results. On axial sections, the lesion appears to occupy almost the entire left lower lobe and pushes the mediastinum and heart toward the right. Coronal sections show dislocation of the descending aorta and elevation of the left hemidiaphragm, while on sagittal sections, the lesion appears attached to visceral pleura and possibly in direct contact with diaphragmatic structures. No direct connection between the lesion and bronchial branches is visible, which reinforces the ELS diagnosis, a congenital abnormality where non-functional lung tissue does not communicate with the tracheobronchial tree and receives systemic vascular supply, usually from the thoracic aorta or its branches.

ELS diagnosis is supported by findings of vascularization from the left posterior intercostal artery and drainage to the azygos and hemiazygos systems, clearly visible from dilated vessel images

around the lesion. Literature shows that ELS is a rare form of bronchopulmonary foregut malformation and accounts for about 15-25% of all pulmonary sequestration cases. Characteristic radiological features of ELS include solid cystic lesions in the lower lung lobe (more often left side), with systemic arterial supply from aorta and venous drainage to systemic veins, and without bronchial connection.^{42,43} Contrast-enhanced MSCT is very important for detailed identification of feeding and draining vessels, and in this case, this has been well confirmed.

Regarding prenatal diagnosis, extralobar pulmonary sequestration can often be detected from the second trimester through antenatal ultrasound. Characteristic features on prenatal examination include a homogeneous, hyperechoic, non-cystic mass in the lower fetal thorax, usually left side, and often associated with pleural effusion or fetal hydrops. Further examination with fetal MRI can also help differentiate ELS from congenital pulmonary airway malformation (CPAM) and other congenital abnormalities.^{44,45}

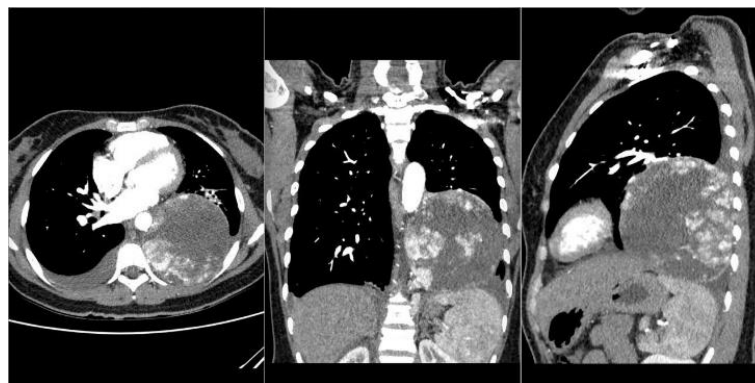


Figure 3. Thoracic CT scan with multiplanar reconstruction (MPR): axial (left), coronal (center), and sagittal (right)

Based on multi-planar chest CT images (axial, coronal, and sagittal), there is a large mass in the posterior mediastinum or left paravertebral area with very characteristic features requiring systematic diagnostic approach. Initially, the posterior location (behind the heart, anterolateral to vertebrae) indicates that neurogenic tumors such as schwannoma or neurofibroma are the main differential diagnosis, considering about 90-95% of posterior mediastinal tumors originate from nerve tissue.⁴⁶ This type of tumor usually appears as a soft mass, can be homogeneous or heterogeneous, and sometimes shows bone compression (scalloping) or extension to intervertebral foramen (“dumbbell tumors”).

However, the mass has heterogeneous density, prominent due to clear areas of necrosis or internal hemorrhage with varying HU values. This is

consistent with characteristics of large neurogenic tumors, which can show cystic or hemorrhagic degeneration.⁴⁷ Additionally, the mass appears not to destroy bone - no vertebral destruction or foramen cavities at the posterior mediastinal border.

Nevertheless, other possibilities must also be considered: lymphoma or lymph node metastases, as they can appear as heterogeneous masses providing mediastinal compression effects without bone erosion, and are often initially asymptomatic.⁴⁷ Foregut duplication cysts, such as bronchogenic or esophageal duplication cysts, can also be found behind the esophagus as masses with thin walls and fluid content; however, they usually appear homogeneous and without solid or hemorrhagic components.^{48,49}

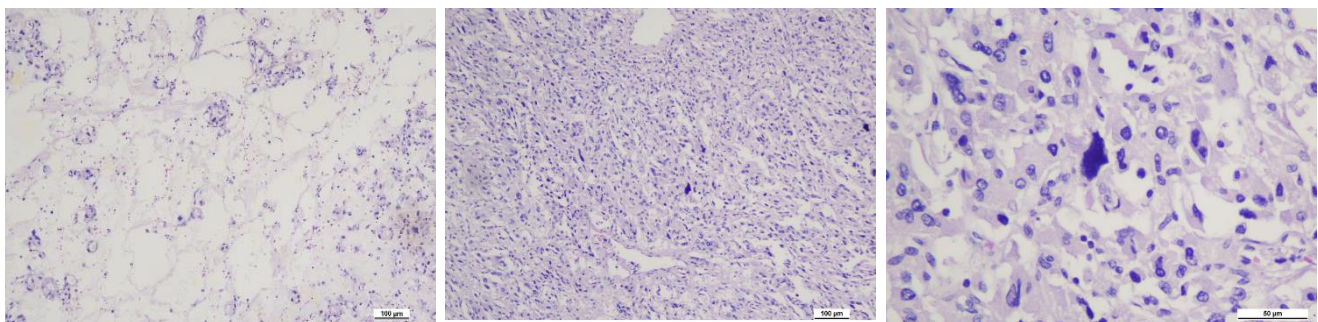
Furthermore, heterogeneous appearance and posterior location also suggest possibility of vascular

tumors such as paraganglioma, though less common.⁵⁰ Contrast angiography or MRI imaging is needed to evaluate vascular involvement and enhancement patterns. Contrast CT can also confirm vascular structures and assess whether the mass compresses large vessels such as aorta or azygos vein.

In conclusion, posterior location, clear boundaries, heterogeneous density, and absence of bone invasion make neurogenic tumor (schwannoma/neurofibroma) the most likely diagnosis - most common in posterior mediastinum.⁴⁶ However, lymphoma, nodal metastases, duplication cysts, or vascular neoplasms cannot be ruled out without further evaluation, such as CT-guided biopsy or advanced tissue characterization MRI. Such evaluation is also important for mapping management plans including surgical resection options (ideally VATS) or non-surgical therapy based on tumor etiology and condition. In Gabelloni et al. report⁵, MDCT imaging

with multiplanar reconstruction is the primary method for identifying PS, particularly in detecting arterial supply and characteristic venous drainage patterns. ELS often shows arterial supply from thoracic aorta or intercostal arteries, and drainage to systemic veins such as azygos/hemiazygos, as seen in this case.

PS management depends on symptoms, size, and risk of complications. In large asymptomatic lesions compressing mediastinal structures (as in this case), surgical resection becomes a primary consideration. Another alternative is endovascular embolization of feeding arteries, especially in high-risk surgical patients. However, embolization has risks of recanalization and devascularized tissue infection. Additional findings of bilateral pleural effusion and compression atelectasis in this patient are likely mass effects from this large lesion on surrounding lung tissue. The presence of fibrosis and lymphadenopathy could be residual from chronic inflammatory or reactive processes.



The low-magnification, 100 µm left image shows lung tissue with a loose structure and many empty spaces resembling alveoli.

The central image at medium magnification (100 µm) shows a significant increase in cell density, with a pattern of cellular proliferation and densely packed nuclei.

The right image at high magnification (50 µm) clearly shows nuclear pleomorphism, with variations in nuclear size and shape, as well as the presence of mitosis and hyperchromatic nuclei.

Figure 4. Histopathological appearance of Extralobar Pulmonary Sequestration (ELS)

The histopathological image shows a spectrum of tissue changes consistent with extralobar pulmonary sequestration (ELS) diagnosis in the patient. The left image shows normal lung tissue with intact alveolar structure, while the center image shows dense cellular proliferation without alveolar structure, reflecting non-functional sequestered tissue with abnormal vascularization. The right image shows nuclear pleomorphism and high cell density likely related to chronic inflammatory response, hemorrhage, or necrosis consistent with MSCT findings showing necrotic and hemorrhagic components in the lesion. These three findings support ELS diagnosis histopathologically.

Differential diagnosis through imaging has narrowed possibilities, but definitive proof can only be obtained through histopathological examination.

Anatomical pathology results become a key element confirming that this hypervascular lesion in the posterior mediastinum is not a vascular neoplasm, infectious process, or malignancy. Microscopic findings show non-functional lung tissue, with imperfect alveolar structure, dense cellular proliferation, and nuclear pleomorphism with high density - characteristic findings of extralobar pulmonary sequestration (ELS). No neoplastic cell invasion, pathological mitoses, or purulent inflammation leading to malignancy or infection were found.

Additionally, identification of separate pleura and vascular supply from systemic arteries reinforces the characteristic anatomical features of ELS. These findings are consistent with literature stating that definitive ELS confirmation can only be achieved

through histopathological evaluation, particularly to differentiate from similar radiological entities such as hemangioma, bronchogenic cysts, or neurogenic tumors.^{5,51}

CONCLUSION

This case demonstrates the importance of considering extralobar pulmonary sequestration (ELS) as a differential diagnosis for hypervascular posterior mediastinal masses, especially when there is systemic feeding artery and drainage to azygos/hemiazygos veins. Contrast-enhanced thoracic MDCT examination is crucial in establishing diagnosis as it can clearly identify the lesion's vascular anatomy.

Although ELS is more commonly found in infants, incidental diagnosis in adults remains possible. Good understanding of ELS radiological features can prevent diagnostic errors and unnecessary interventions, and help plan appropriate management approaches, both surgically and endovascularly.

DECLARATIONS

Ethics approval and consent to participate

Not applicable

Consent for publication

Not applicable.

Competing interests

The authors declare no conflict of interest.

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