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Case Study

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INCIDENTAL FINDING OF INTRALOBAR PULMONARY SEQUESTRATION OF THE LUNG IN 41-YEAR-OLD ADULT MALE

[1]*Dr. Hemanth Gowda M. C. (MBBS) and [2]Dr. Parul Dutta (MD, DMRD)

[1]Post Graduate Resident, Department of Radiology Gauhati Medical College and Hospital, Guwahati- 32.
[2]Professor and Head of the Department Department of Radiology Gauhati Medical College and Hospital, Guwahati - 32.

***Corresponding Author: Dr. Hemanth Gowda M. C.**

Post Graduate Resident, Department of Radiology Gauhati Medical College and Hospital, Guwahati- 32.

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ABSTRACT

Pulmonary sequestration is defined as a nonfunctioning mass composed of dysplastic lung parenchyma, embryologically detached from the tracheobronchial tree and receiving its own blood supply from a systemic artery, usually the thoracic or abdominal aorta. It may be intralobar or extralobar depending on the presence of an independent pleural envelope. Here, we present a rare case of incidental intralobar pulmonary sequestration in an adult.

KEYWORDS: Congenital lung conditions, intralobar sequestration, asymptomatic adult, close follow-up.

INTRODUCTION

Pulmonary sequestration is an uncommon congenital anomaly of the primitive foregut. The sequestered segment of the lung is completely separated from trachea-bronchial tree. The definite diagnosis of pulmonary sequestration depends on the demonstration of the systemic arterial supply and venous drainage. Here, we present a rare case of incidental intralobar pulmonary sequestration in an adult.

CASE REPORT

A 41- year old man referred from department of urology with history of left flank pain and hematuria for 15 days. Past medical and surgical history was insignificant. NCCT KUB revealed left renal calculus. Incidentally, a triangular mass of non-aerated segment measuring 5.2x3.2x3.9 cm is noted occupying the postero-basal segment of left lung. Patient underwent lithotripsy for renal calculus and referred to pulmonary medicine for further evaluation of incidental finding.

A detailed retrospective history did not reveal neonatal respiratory distress, recurrent respiratory tract infections, chest pain or any other respiratory symptoms. Prior chest radiograph done 2 months back revealed triangular opacity in left lower zone overlapping cardiac shadow.

A CT thoracic angiography is done to rule out any vascular etiology, which revealed aberrant arterial supply to the non-aerated segment by the direct branches from the abdominal aorta originating postero-laterally 2.9 cm above the origin of celiac trunk and venous drainage through tributaries into inferior pulmonary veins. No communication is noted between the lesion and the remainder of the trachea-bronchial tree. With these findings, a diagnosis of intralobar sequestration (ILS) of the left lung was made. To confirm the diagnosis, biopsy of the lesion was done under CT guidance, which on microscopy revealed dysplastic pulmonary tissue with no malignant cells suggesting pulmonary sequestration.

Given that the patient was currently asymptomatic, after consulting with the department of cardiothoracic and vascular surgery, patient was offered with two options: immediate preventive resection versus close follow-up with surgical resection should he become symptomatic. The patient opted for the later and patient is currently under close follow-up and is asymptomatic till date.

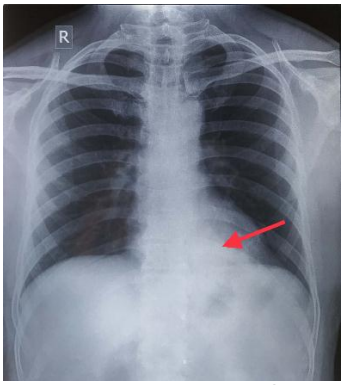

Figure 1: Chest radiograph showing a triangular opacity in left lower zone overlapping the cardiac shadow.

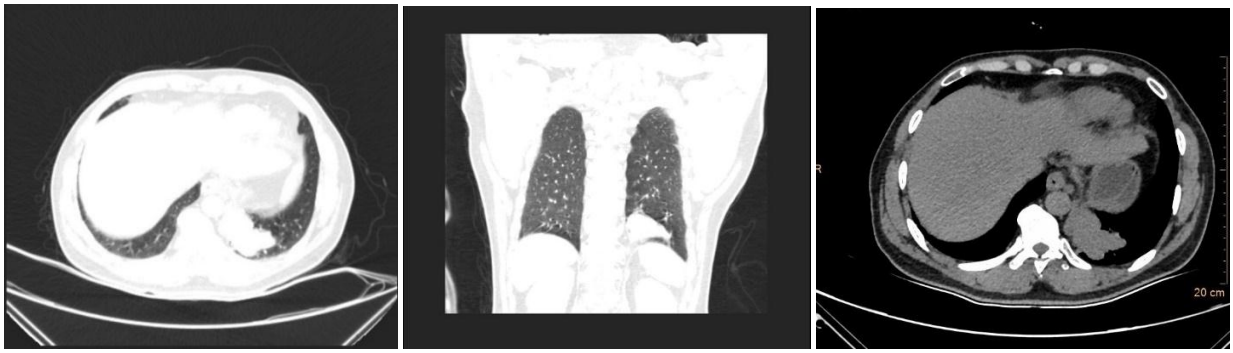

Figure 2: HRCT chest axial and coronal sections in lung and mediastinal window showing triangular mass of non-aerated lung occupying postero-basal segment of left lower lobe.

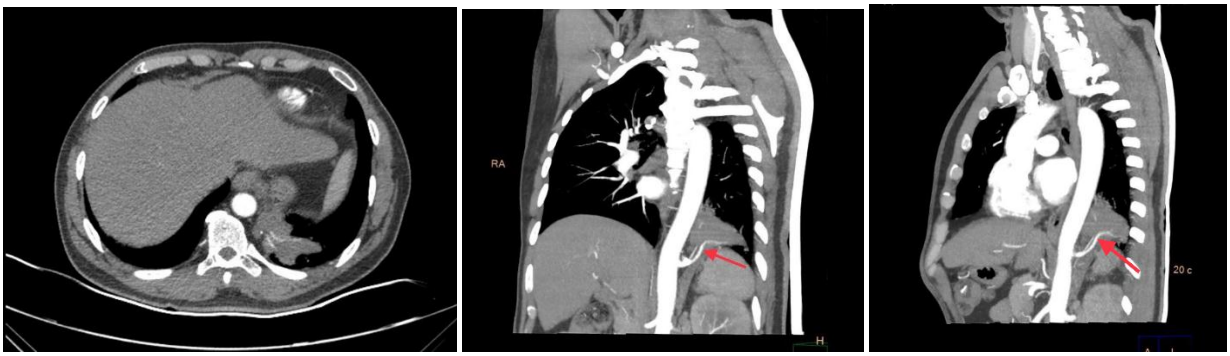

Figure 3: CT angiography thorax(axial, oblique coronal and sagittal with mip) showing aberrant arterial supply from direct branches from abdominal aorta(red arrow)

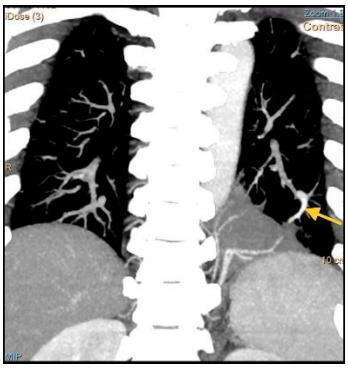

Figure 4: CT angiography thorax(delayed phase with mip) showing venous drainage into the pulmonary veins(yellow arrow)

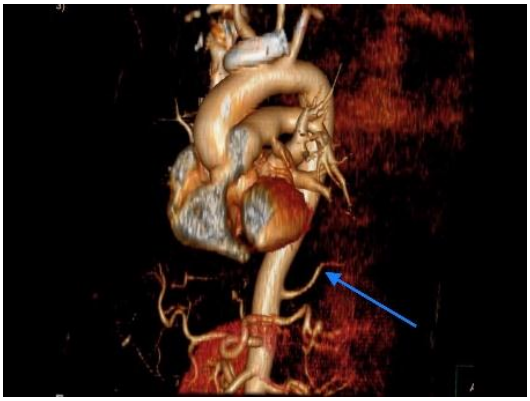

Figure 5: Multiplanar reconstructive image showing aberrant artery (blue arrow) arising postero-laterally from abdominal aorta.

DISCUSSION

Pulmonary sequestration was first described by Huber in 1777. It is characterized as a dysplastic mass of lung tissue that lacks communication with the tracheobronchial tree and receives systemic rather than pulmonary arterial blood supply.[1] Pulmonary sequestration can be further classified into two distinct types: intralobar and extralobar. These differ based on clinical features, location, pleural covering and venous drainage.[Table 1] Pulmonary sequestration is a rare condition and is encountered at a rate of less than one case per year at tertiary referral centres.[2] The etiology of ILS is unclear, with evidence equally supporting both acquired and congenital hypotheses. The latter is well described by Pryce,[1] who proposed that ILS occurs as a result of either the formation of an accessory lung bud or due to the capture and subsequent traction of a tip of the developing lung by a systemic artery. An alternative explanation was later offered by Smith,[3] who proposed that the initial defect was in the primitive pulmonary arterial supply. According to Smith,[3] a lack of blood flow to the developing lung leads to the retention of vessels from the primitive dorsal aorta, and exposure to systemic arterial pressure after birth leads to the changes observed. Recently, the observation that ILS is more commonly encountered in adults and that coexisting congenital anomalies in this condition are rare has led to the proposal that ILS is an acquired phenomenon. Although the details are yet to be established, it has been suggested that an unknown trigger serves as a stimulus for the development of an aberrant systemic arterial supply in late childhood or early adulthood.[4] The key to establishing the diagnosis of ILS lies in identifying the aberrant arterial supply. Both CT and magnetic resonance imaging have proven to be effective in this regard. However, multidetector CT angiography is emerging as the diagnostic test of choice because it is better able to simultaneously visualize and provide details of the arterial supply, lung parenchyma and venous drainage.[5-7] Classically, ILS in adults has been identified in the work-up for recurrent lower respiratory tract infections.[2] In our patient diagnosis of ILS was made incidentally, during imaging for a completely unrelated condition. We found that current guidelines did not offer recommendations for management in this case. Traditionally, as a preventive measure or as a treatment option for recurrent pneumonia, most centres recommend early surgical resection.[8] Potentially adding further support for early resection are recent case reports that describe more severe complications such as life-threatening hemorrhage and malignant transformation.[9,10] However, it is important to note that in the largest case series on ILS published to date, a large percentage of patients were asymptomatic.[2] Also, although it would be difficult to study, it is possible that the number of patients with undiagnosed ILS is greater than those who develop symptoms. Finally, although severe complications, such as malignant transformation, have been documented in case reports, we should remain cognizant of the fact that these are extremely rare occurrences. And adult patients with asymptomatic pulmonary sequestration may be observed without surgical resection.[12] Given that the patient was currently asymptomatic, after consulting with the department of cardiothoracic and vascular surgery, patient was offered with two options: immediate preventive resection versus close follow-up with surgical resection should he become symptomatic.

Table 1[11]

	Intralobar sequestration	Extralobar sequestration
Patient age	Adult or older child	Infant or child
Symptoms	Infection common	Infection rare
Morphology	Within a lobe	Within its own pleural envelope
Location	65% at left base	90% at lung base
Arterial supply	Thoracic or abdominal aorta	Usually abdominal aorta
Venous drainage	Usually pulmonary veins	Usually systemic veins
Appearance	Commonly contains air	Rarely contains air

CONCLUSION

ILS is a rare congenital anomaly that can present in adulthood. As advanced imaging modalities become more routine and available, the diagnosis of ILS as an incidental finding will likely become more common. Although most centres recommend early resection, given that many patients remain asymptomatic and that severe complications are rare, close outpatient follow-up may be a reasonable management option for patients with ILS discovered incidentally. In the absence of established guidelines, the decision to proceed with resection should be made with the patient on a case-by-case basis when ILS is discovered incidentally.

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