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Case Report

Radiology Section

Multimodality Imaging and Endovascular Management of Intralobar Pulmonary Sequestration in a Young Female: A Case Report

ARNAV VIVEK RAMTEKE[1], KAJAL MITRA[2], PRASHANT ONKAR[3], SURESH PHATAK[4], ASHISH AMBHORE[5]

ABSTRACT

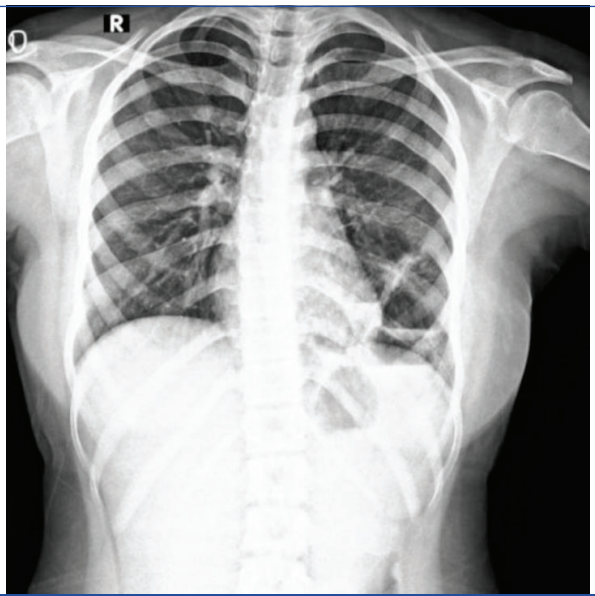
Pulmonary sequestration is an uncommon congenital lung anomaly in which non-functioning lung tissue lacks communication with the tracheobronchial tree and receives blood supply from an abnormal systemic artery. Intralobar sequestration, though less frequent, often presents with recurrent respiratory infections in children and adolescents. This case report describes a young female with a long history of recurrent fever and pneumonia who was evaluated using multimodality imaging. Imaging revealed cavitary lesions in the left lower lung. CT angiography demonstrated an aberrant artery arising from the abdominal aorta, findings consistent with intralobar pulmonary sequestration The patient was initially managed with drainage and antibiotics, followed by endovascular coil embolisation of the anomalous artery. This reduced the risk of bleeding and facilitated a safe, planned surgical removal of the sequestered lung tissue. This case demonstrates the value of combining modern imaging and minimally invasive embolisation with surgery to achieve safe and effective management of pulmonary sequestration.

Keywords: Abdominal aorta, Cavitary lesions, Embolisation, Lung anomaly, Lung zone

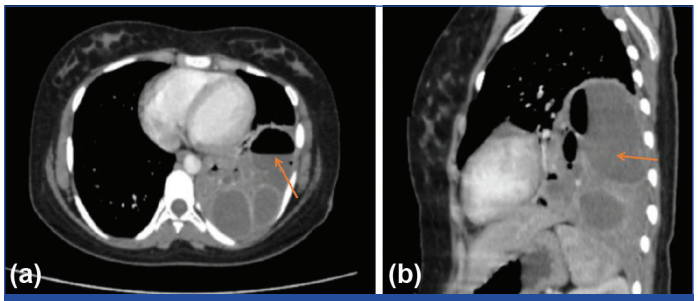
CASE REPORT

A 14-year-old female presented with complaints of recurrent cough, fever, and mild dyspnoea for the last 15 days. There was no history of haemoptysis, chest pain, tuberculosis or previous thoracic surgery. No previous diagnosis of congenital anomalies was present. No significant family history was noted. Her vital signs were within normal limits. On auscultation, there was reduced air entry on the left side, infra-scapular, infra-mammary and infra-axillary area. A pulmonary function test was also which showed mild restrictive pattern and suboptimal effort.

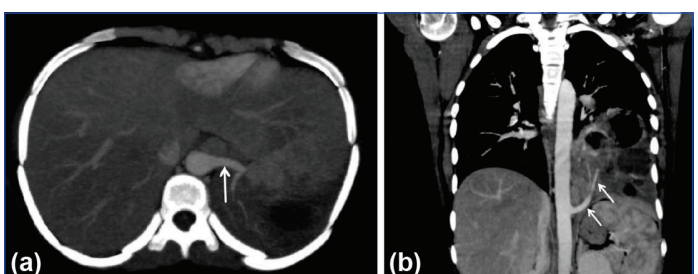
A chest radiograph was done on presentation, which showed cystic airspace lucencies with an air-fluid level in the left lower lung zone, suggestive of loculated collections [Table/Fig-1]. A consequent CECT thorax was performed, which showed multiple interconnecting thick-walled peripherally enhancing fluid-filled cavities within the left lower lung lobe [Table/Fig-2a,b]. Differential diagnoses such as congenital pulmonary airway malformation, bronchogenic cyst, and lung abscess were initially considered. However, the demonstration of a systemic arterial supply arising from the left lateral wall of the abdominal aorta on CT angiography was diagnostic, allowing these possibilities to be ruled out and confirming intralobar pulmonary sequestration [Table/Fig-3a,b]. Intercostal Drainage (ICD) of collections and antibiotics were given to treat the infection. She was planned for digital subtraction angiography followed by



[Table/Fig-1]: Chest radiograph (PA view) demonstrating multiple cavitary air-space lesions with air-fluid levels in the left lower lung zone, suggestive of infected cystic lesions/loculated collections.

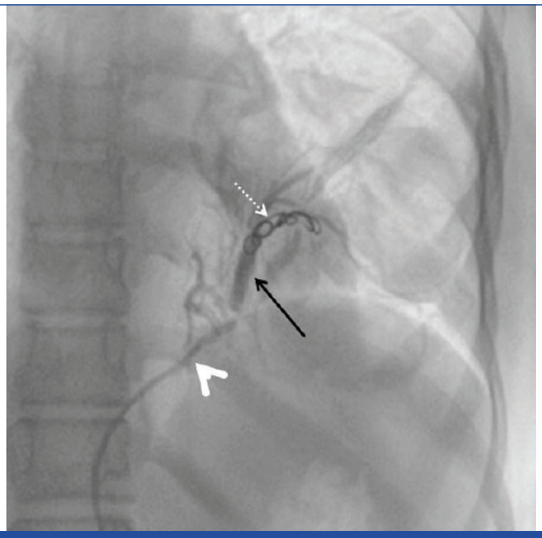


[Table/Fig-2a,b]: Axial and Sagittal reformatted images showing multiple inter-communicating, thick-walled, peripherally enhancing cavitary lesions with air-fluid levels involving the left lower lobe, consistent with infected sequestrated lung tissue (Marked by arrows).

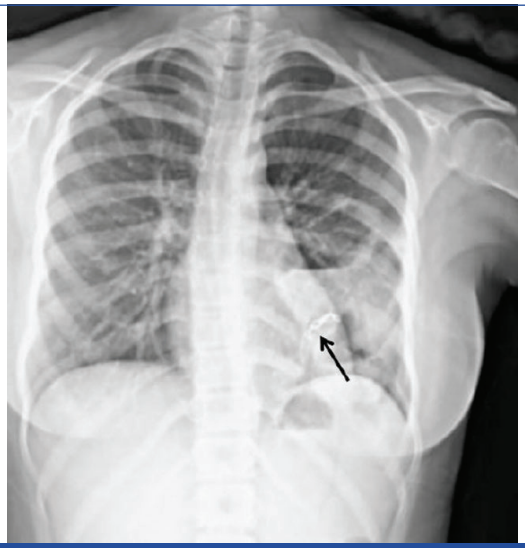


[Table/Fig-3a,b]: Axial and coronal post-contrast images demonstrating an aberrant systemic arterial branch arising from the abdominal aorta supplying the left lower lobe, confirming the diagnosis of intralobar pulmonary sequestration. (Marked by white arrows).

pulmonary artery embolisation using endovascular coiling in the same setting. A catheter was passed through the descending aorta into the anomalous artery, and endovascular coiling was done under Digital Subtraction Angiography (DSA)/fluoroscopic guidance [Table/Fig-4]. A post-embolisation X-ray was taken to confirm the coil position [Table/Fig-5]. Following successful embolisation, the patient underwent elective sequestrectomy approximately three weeks later via posterolateral thoracotomy. The postoperative period was marked by transient respiratory issues, for which the patient was closely monitored and managed with supportive care. Bronchoscopy performed during this period revealed blood clots in the left main bronchus, which were successfully removed, following which her respiratory status gradually improved. She was clinically stable at discharge and was advised to have regular follow-up. Postoperative cross-sectional imaging could not be performed, as the patient declined further imaging.



[Table/Fig-4]: DSA image during endovascular coil placement demonstrating the contrast-opacified anomalous artery (black arrow), deployed coil (dotted white arrow), and guidewire in situ (white arrowhead).



[Table/Fig-5]: Post-embolisation chest radiograph demonstrating radio-opaque embolisation coils in situ, confirming appropriate coil positioning with no evidence of migration. Black arrow showing normal coil position.

DISCUSSION

Pulmonary sequestration is a congenital lung anomaly in which dysplastic lung tissue lacks communication with the tracheobronchial tree and receives an abnormal systemic arterial supply [1]. It accounts for approximately 6% of congenital lung malformations and may be diagnosed prenatally or present later in life with recurrent pneumonia, chest pain, cough, dyspnea, or haemoptysis [2,3]. Pulmonary sequestration is considered a congenital primitive foregut anomaly. The exact mechanism is not known, but the most accepted theory is that there is development of an accessory lung bud below or in proximity to the normal lung bud. In intralobar sequestration, abnormal lung tissue develops within the normal lung parenchyma before pleural formation and shares the visceral pleura, with systemic arterial supply [4].

There are two main types of sequestration: Intralobar and extralobar. A third type has been described if there is a communicating bronchopulmonary foregut malformation [5,6].

Extralobar sequestration accounts for 25% of total cases of sequestration that are diagnosed postnatally. Here, the extra lobe is invested by its own pleura and gets its blood supply from a systemic artery [7]. The venous drainage can be into the azygous vein, hemiazygos vein, and/or into the vena-caval system. However, in almost 25% cases, venous drainage is through pulmonary veins. The most common location (almost 80% cases) for extralobar sequestration is in between the left lower lobe and the underlying diaphragm, usually in the posterior costo-diaphragmatic sulcus. It can also occur in the sub-diaphragmatic zone, usually in the suprarenal location [8,9].

In intralobar sequestration, the abnormal lung tissue is within the normal lung. It is covered by the visceral pleura and drains into the pulmonary veins. Few cases have shown drainage into the vena cava, azygous vein, or drainage directly into the right atrium has also been reported. Involvement of the lower lobes more often on the left-side is the most common presentation. Intralobar sequestration comprises 75% of cases diagnosed postnatally [10].

Sequestration can be detected on antenatal ultrasound by almost 16 weeks of gestation. It appears as a well-defined, homogeneous, echogenic wedge-shaped lesion in the left lower lobe adjacent to the underlying diaphragm. To differentiate between other congenital lung masses, one must look for a systemic artery feeding the mass using colour doppler [2,11].

MRI can help confirm the diagnosis and location of the mass. The abnormal lobe tends to have high T1 and T2 signals as compared to normal lung tissue. MR Angiography can be used to identify the feeding vessel from the aorta to the mass. Some instances of resolution of masses have been reported, which may be due to torsion of the vascular pedicle supplying the lobe [12,13].

Primary treatment for both intra- and extralobar sequestration has been surgical resection of the abnormal/supernumerary lobe, as they help reduce the incidence of recurrent pulmonary infections, haemoptysis or other symptoms. Segmental resection, lobectomy or open thoracotomy is some of the approaches. However, these procedures have complications like haemothorax, atelectasis, ARDS, pulmonary embolism, etc., [14].

Coil embolisation of the aberrant systemic branch has recently been established as a treatment approach. As the anomalous blood supply is cut-off, there is resorption of the affected lobe due to processes like infarction, necrosis, and fibrosis [5]. It is beneficial because it reduces the incidence of intraoperative bleeds and hospital stays. However, the recurrence rate after embolisation is high (3-5%). It is also associated with complications like broncho-pulmonary fistula formation, thrombosis or infection at the access site, embolisation of the wrong artery etc. [14]. Thus, while endovascular embolisation shows promising results, further studies are needed to better define its long-term outcomes, associated complications, and its role as an adjunct or alternative to surgical management.

In keeping with the evolving role of endovascular management, recent reports in the literature describe cases of intralobar pulmonary sequestration managed with endovascular embolisation, either as a standalone treatment or as a preparatory step before surgery. Borzelli A et al., reported successful coil embolisation with good clinical outcomes, demonstrating embolisation as a minimally invasive option in carefully selected patients [3]. Similarly, Healy J et al., described embolisation in symptomatic intralobar pulmonary

sequestration, noting its usefulness in simplifying subsequent surgery and reducing perioperative bleeding [14]. Together with the present case, these observations highlight the increasing role of multimodality imaging and endovascular techniques in the management of intralobar pulmonary sequestration.

CONCLUSION(S)

Sequestration is a congenital anomaly that may be incidentally diagnosed or present with recurrent pneumonia. A CT/MR angiogram can be used to identify the aberrant artery supplying the abnormal lung lobe. Segmental resection/lobectomy is the classical treatment approach. Arterial embolisation is a comparatively newer treatment modality with its pros and cons. Thus, further studies and research are necessary to determine the long-term outcomes and benefits of this modality.

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PARTICULARS OF CONTRIBUTORS:
1. Junior Resident, Department of Radiodiagnosis, Lata Mangeshkar Hospital, NKPSIMS, Hingna, Nagpur, Maharashtra, India.
2. Professor, Department of Radiodiagnosis, Lata Mangeshkar Hospital, NKPSIMS, Hingna, Nagpur, Maharashtra, India.
3. Professor, Department of Radiodiagnosis, Lata Mangeshkar Hospital, NKPSIMS, Hingna, Nagpur, Maharashtra, India.
4. Professor, Department of Radiodiagnosis, Lata Mangeshkar Hospital, NKPSIMS, Hingna, Nagpur, Maharashtra, India.
5. Lecturer, Department of Radiodiagnosis, AIIMS, Nagpur, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Arnav Vivek Ramteke,
NKP Salve Institute of Medical Sciences, Vindhyachal Boys Hostel, Room No. 6, Ngapur-440019, Maharashtra, India.
E-mail: arnavramteke798@gmail.com

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