

CASE REPORT

Intralobar pulmonary sequestration with an aneurysmal celiac arterial feeder coexisting with superior mesenteric artery syndrome: A case report and literature review

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Pulmonary sequestration is a rare bronchopulmonary foregut malformation characterized by the abnormal development of non-functional lung tissue enclosed by visceral pulmonary pleura, supplied by a systemic arterial blood supply, and lacking communication with the bronchial tree. The blood supply commonly arises from the thoracic or abdominal aorta. We reported a 44-year-old female who presented with vomiting and recent weight loss of about 20 kilograms in the last 6 months. The physical and biochemical examination was unremarkable. A contrast-enhanced abdominal CT scan reveals signs of superior mesenteric artery syndrome (SMAS), including a decreased aortomesenteric angle and aortomesenteric distance. The lower chest sections show an irregular consolidative lesion in the posterior segment of the left lower lobe supplied by an aberrant aneurysmal vessel from the celiac artery, consistent with intralobar pulmonary sequestration (ILS). To our knowledge, co-occurrence of SMAS and ILS with a celiac aneurysmal feeder has not been previously reported, as both conditions affect organs of different locations and embryological origins. A literature review was conducted to examine the presentation and vascular supply of ILS.

Keywords: pulmonary sequestration, aberrant vessel, superior mesenteric artery syndrome

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Introduction

Pulmonary sequestration is a congenital pulmonary malformation of the lower respiratory tract, accounting for 0.15–6.4% of all pulmonary anomalies, making it a relatively uncommon condition. This anomaly consists of an anomalous lung segment with a systemic arterial supply and different types of venous drainage [1]. The affected lung segment is not functioning and is not connected to the tracheobronchial tree [2]. The two main anatomical types of sequestrations are intralobar and extra lobar; the former is the most common, accounting for about 75 to 90% of sequestrations, while 10 to 25% are extralobar [2]. The intralobar type differs from the extralobar type by sharing the same native pleural lining. In contrast, the extralobar type exhibits pleural investment of its own [3].

Two-thirds of cases of ILS are located in the costophrenic sulci of the posterior basal segment of the left lower lobe, making it more prevalent in the left lung [4]. The blood supply of the sequestered segment typically originates from the thoracic or abdominal aorta in 75% of cases, although other arteries, such as the splenic, left gastric, coronary, or renal arteries, may also supply it [5]. ILS typically presents with symptoms such as recurrent cough, fever, hemoptysis, and chest pain, often due to recurrent pulmonary infections later in childhood or adulthood. However,

9.7% of case reports were detected as incidental findings [6]. The first line of investigation for such symptoms is a chest X-ray, which shows homogeneous or heterogeneous opacity, mass lesion, or even a cavitary lesion. These findings have a wide differential diagnosis, including infection, mass, or even congenital anomalies, which warrant further evaluation by contrast-enhanced CT scan and CT angiography. This reveals aberrant arterial supply to the sequestration as well as abnormal lung parenchyma [7].

Many congenital malformations of the pulmonary airways may be associated with sequestration, such as congenital heart disease, gastric duplication, bronchogenic cysts, and diaphragmatic hernias [5]. Although surgical resection was the standard procedure, as this abnormal lung tissue is still a source of infection even in asymptomatic cases, endovascular intervention is now being used to treat an increasing number of cases [8].

We report a case of ILS incidentally detected during the workup of signs and symptoms of SMAS. The case is unique because each condition is uncommon on its own, and its coexistence is even rarer. In addition, symptoms of both conditions may overlap or mask each other, like abdominal pain from the SMAS or from the lower pulmonary sequestration. To our knowledge, this is the first reported case of such co-occurrence of two conditions of different organs and different embryology based on our search in databases like Google Scholar, Web of Science, and Scopus, using various keywords and titles.

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Case presentation

A 44-year-old female patient, married G4P4A0, with unremarkable past medical and surgical history, presents with a history of recurrent vomiting and recent weight loss (20 kilograms) within 6 months. She frequently experiences emesis after eating, mostly undigested food, not projectile, and no hematemesis. Her physical examination and the biochemical tests showed no significant abnormalities. Therefore, she was sent for a contrast-enhanced CT scan of the abdomen to exclude any hidden pathology. CT scan (Figure 1) shows a decreased aorto-mesenteric angle of 23° and an aorto-mesenteric distance of 5.1 mm, compressing the 3rd part of the duodenum with a distended contrast-filled stomach. These findings are consistent with superior mesenteric artery syndrome. The visualized sections through the lower chest show an incidental pulmonary lesion in the form of an irregular area of consolidation involving the posterior segment of the left lower lobe abutting the medial pleura. There is a large aneurysmal aberrant vessel arising from

the celiac artery, measuring 10 mm in maximal diameter, that supplies this pulmonary lesion. It ascends through the esophageal hiatus, reaches, and branches within the lesion (Figure 1). The feeding vessels exhibit normal wall thickness, no mural calcification, and no mural hematoma. The pulmonary drainage of this pulmonary lesion is ascending to the left inferior pulmonary vein (Figure 2).

Regarding SMAS: The aortomesenteric distance in our case was 5.1 mm, and the aortomesenteric angle was 23° . These values mostly match the distance-based CT criteria for the diagnosis of superior mesenteric artery syndrome that are frequently mentioned in the literatures. However, it is crucial to mention that imaging thresholds are helpful but not definitive; the patient's response to nutritional therapy and clinical assessment is still crucial for diagnosis.

The patient managed conservatively by advising the patient to eat frequent small meals with a high-calorie diet and practice posture treatment, including lying in the left lateral decubitus position instead of recumbent, in addi-

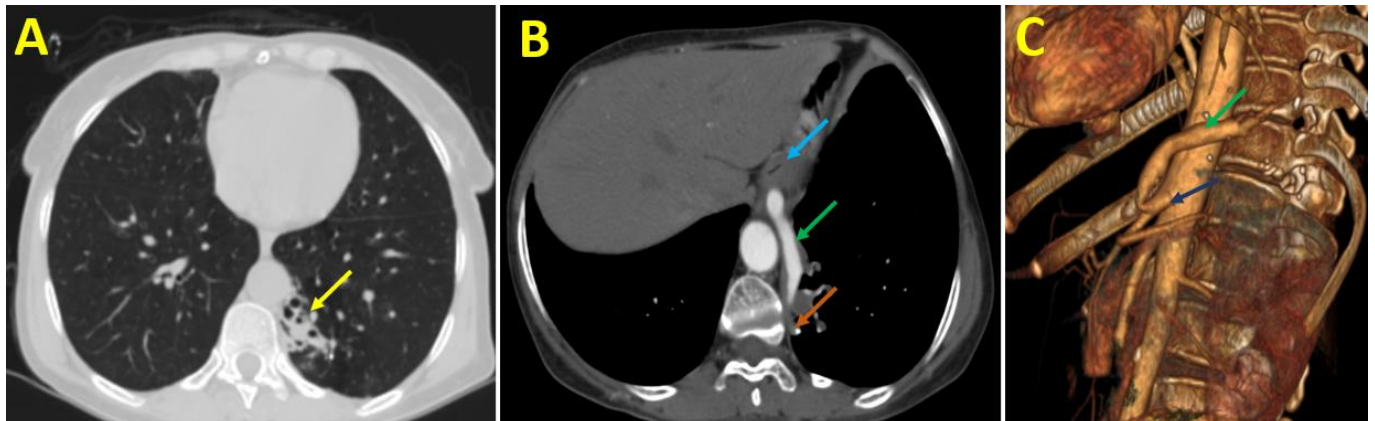


Fig. 1. Contrast-enhanced CT scan of the lower chest. A: Lung window; B: mediastinal window shows heterogeneous patchy area of consolidation involving the medial aspect of the posterior segment of the left lower lobe abutting the medial pleura with internal bronchiectatic changes (yellow arrow) showing coarse calcification (orange arrow) and there is aberrant vessel (green arrow) supplying this pulmonary lesion ascending from the abdominal cavity through the esophageal hiatus posterior to the gastro-esophageal junction (blue arrow); C: volume rendered image showing the aberrant vessels arising from the celiac trunk (blue arrow).

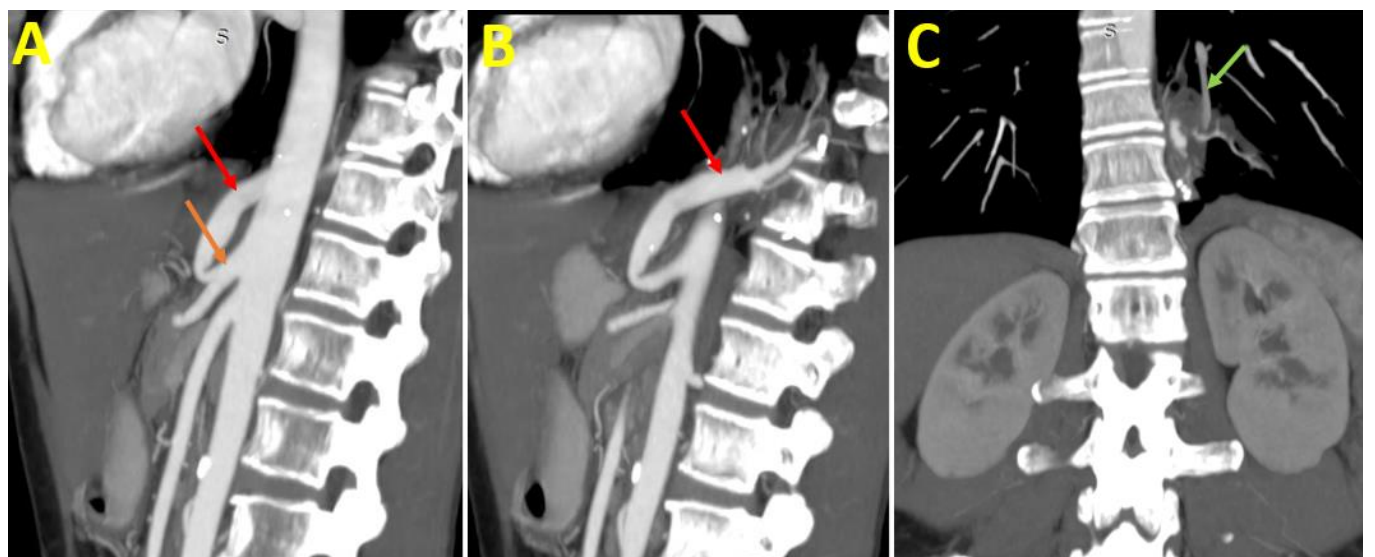


Fig. 2. Contrast-enhanced CT scan arterial phase MIP images. A and B: Sagittal reformatted images (arterial phase) show an apparent artery (red arrow) supplying the pulmonary lesion arising from the celiac artery (orange arrow). C: Coronal reformatted MIP image (venous phase) shows the venous drainage (green Arrow) of this pulmonary lesion to the left inferior pulmonary vein.

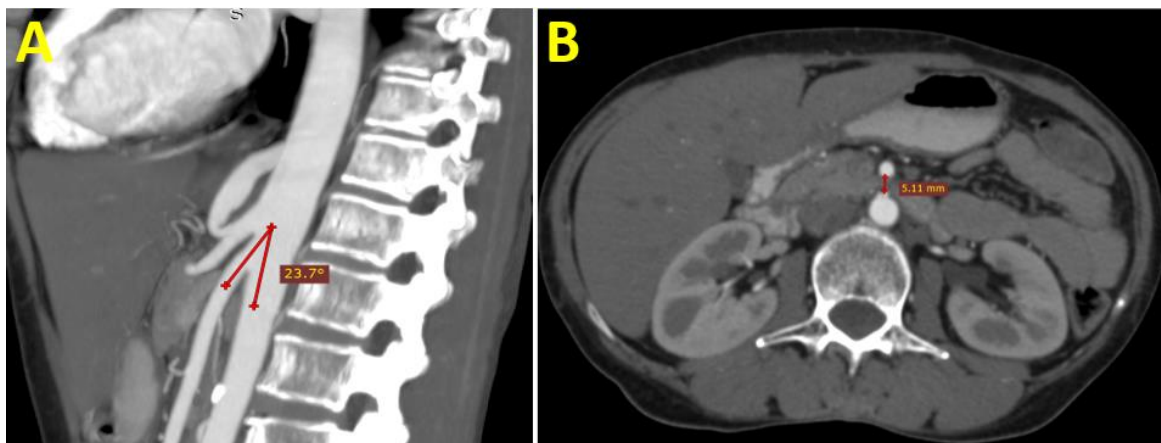


Fig. 3. Contrast-enhanced CT scan of the upper abdomen (MIP) images. A: Sagittal reformatted image shows that the aortomesenteric angle measured 23°. B: Axial section shows the aortomesenteric distance = 5.1 mm.

tion to hyper-alimentation, is crucial in an effort to enhance the mesenteric fat pad, which will raise the aortomesenteric angle, alleviating her symptoms.

Regarding the pulmonary sequestration, the patient was referred to the thoracic surgeon, who advised her to undergo surgical resection to prevent potential complications. However, given the incidental and asymptomatic nature of the ILS, she declined any intervention, even after being informed of the potential risk of bleeding, infection, pulmonary hypertension, and, rarely, malignant transformation. The patient was followed clinically for 6 months after diagnosis without repeating the contrast-enhanced CT scan. The vomiting decreased in frequency, and the patient started to gain weight by following prescribed conservative measures. She was advised to promptly report to her physician any signs of complications from the pulmonary sequestration, which had already been explained to her.

Methodology

This is a literature review that was conducted to describe the reported data on ILS, such as clinical presentation, presenting age, feeding vessels, diagnostic imaging modalities, and coexisting conditions. The electronic search was done between the period of inception and [23 November 2025] in Google Scholar, Web of Science, and Scopus. Search terms were used that consisted of: (pulmonary sequestration) OR (aberrant vessel), AND (aberrant) OR a(anomalous plus feeding vessel) OR (superior mesenteric artery syndrome).

Titles/abstracts were screened by two independent reviewers, after which the full-text review was performed. Records identified were (n=93) after duplicate removal, screening for eligibility, and full text assessment, we finally included (n=71) in the final analysis, see below (Figure 4).

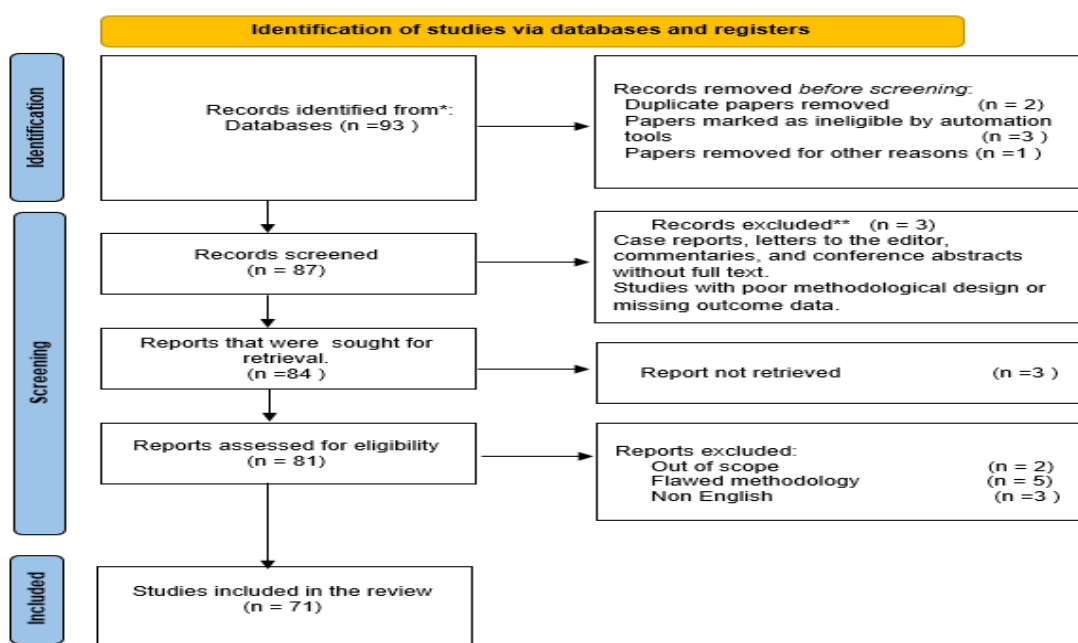


Fig. 4. Prisma flow chart for study methodology (inclusion, exclusion, and analysis).

Inclusion criteria: case reports, case series, review articles, and cohort studies (retrospective/prospective) that report ILS cases in English. The exclusion criteria: missing articles, non-English articles. Two reviewers extracted the data, and disagreements were solved by discussion. The extraction variables were: patient demographics, vascular anatomy, clinical presentation, complications, and management. Synthesis of extracted data was carried out by tabulating (Tables 1-3).

Discussion

The embryological theory about the origin of pulmonary sequestration remains unclear, but the most likely theory is that its development occurred before the separation of the aortic and pulmonary circulations [9]. Other theories suggest that the lung bud emerges before the pleura's development, and the anomalous arterial vessel proliferation is caused by acquired origin from chronic pulmonary infection [10]. The clinical features of pulmonary sequestration can vary widely, from recurrent chest infection, hemoptysis, to rarely heart failure, due to volume overload and left-to-right shunt [11] although some literature suggests that most of the cases show no symptoms [2] but the recurrent respiratory infections remain the commonest reported presentation [12]. It may present as acute coronary syndrome, as a case presented by Rahim et al [13].

Different imaging modalities are used for diagnosis depending on the clinical features, the age at presentation, and the suspicion. MRI and ultrasound are the two main radiologic modalities that can identify bronchopulmonary sequestration in utero. The prenatal obstetrical ultrasound is crucial for early diagnosis as part of an anomaly scan, but the MRI is done for further evaluation and to demonstrate the anomalous vessel [14]. While diagnosis in adults and children is best done with a computed tomography (CT) scan with intravenous contrast and, ideally, CT angiography (CTA)[15,16]. The treatment of asymptomatic lung sequestration is a matter of debate. However, it is recommended to resect these lesions due to the risk of bleeding, the necessity for a larger resection if the sequestration becomes chronically infected, and the likelihood of recurrent infection [17].

In the current case, the aberrant vessel is aneurysmal (10 mm in diameter), arising from the celiac artery, then curving superiorly, passing through the esophageal hiatus, and reaching the Left lower lobe. The aneurysmal feeding artery is rarely reported in the literature [18]. Actually, Savic et al. reported only one case of 540 pulmonary sequestration cases supplied by an aneurysmal artery [19]. The presence of aneurysmal dilatation of the feeding vessel increased the risk of hemorrhage and intraoperative bleeding control, especially if associated with atherosclerosis and hypertension [20].

Table 1. The variable feeding vessel origin

	Name of vessel	No of cases	references
1	Descending thoracic aorta	236	1, 2, 6, 8, 12, 13, 17, 18, 21, 27, 29, 30-48
2	Abdominal aorta	247	5, 33, 34, 49-55
3	Celiac artery	1	34
4	Superior mesenteric artery	1	56
5	Hepatic artery	1	57
6	Inferior Phrenic artery, diaphragmatic artery	33	33, 34, 58
7	Intercostal arteries	37	33, 34
8	Aortic arch	8	34
9	Subclavian artery	6	34
10	Pulmonary artery	5	34
11	Left gastric artery	4	34
12	Coronary arteries, Left circumflex artery	9	22-24, 34, 59
13	Renal artery	4	60-63
14	Renal and internal thoracic*	1	60
15	Aortic and celiac*	1	3
16	11th left intercostal artery and the left bronchial artery*	1	64
17	Internal thoracic artery	3	60,65,66
18	Inferior vena cava	1	4

*Multiple feeders from different origins.

Table 2. Pulmonary sequestration involving the upper and middle lobes

	Lobe	Feeding vessel	Draining vein	Gender	Age at presentation	Symptoms	References
1	RML	Internal mammary artery	Not reported	Male	67 years	Hemoptysis	66
2	RML	Left circumflex coronary artery	Right lower pulmonary vein.	Female	64 years	Recurrent hemoptysis and angina	69
3	RML	Descending thoracic aorta	Pulmonary veins	Male	46 years	Hemoptysis	70
4	RUL	Aorta	Azygos vein	Female	30 years	Weight loss	66,68
5	LUL	Aorta	Left superior pulmonary vein	Female	antenatal	Antenatal	71
6	lingula	Left gastric artery	Left Superior Pulmonary Vein	Female	51 years	Hemoptysis	10
7	lingula	Pericardiophrenic artery and a branch from the aorta	Left superior pulmonary vein	Female	Cadaver 62 years	-	72
8	lingula	Celiac artery	Left superior pulmonary vein	Male	24 years	Repeated infection	73

Table 3. Cases of pulmonary sequestration that have an apparent vessel from the celiac artery

Reference	Year	Age at presentation / years	Sex	Side	Clinical feature	Treatment
1 Jung et al [74]	2010	29	Female	LLL	Hemoptysis	Endovascular embolization
2 Ayloo et al [75]	2011	50	Female	RLL	Hemoptysis	RT lobectomy
3 Erden et al [3]	2012	27	Female	RLL	Pneumonia	Lobectomy
4 Litt et al [76]	2013	25	Male	RLL	Pneumonia	Resection
5 Majumdar et al [77]	2018	40	Male	LLL	Left flank pain	Resection
6 Wang et al [7]	2020	61	Female	RLL	Recurrent chest infection and hemoptysis	Endovascular embolization
7 Dehbi et al [78]	2020	2	Female	RLL	Respiratory distress and fever	RLL lobectomy
8 Mashkov et al [79]	2021	10	Female	LLL	Recurrent pneumonia	Resection
9 Shafiq M et al [53]	2021	32	Female	LLL	Hemoptysis	Surgical resection
10 Boyle et al [80]	2023	25	Female		Hemoptysis	Endovascular embolization
11 Marwah V et al [73]	2023	24	Male	Lingula	Recurrent pneumonia	Left segmentectomy
12 Monfregola et al [81]	2024	69	Male	LLL	Hemoptysis	Resection with endovascular embolization
13 Chataut et al [82]	2024	30	Male	LLL	Hemoptysis	Endovascular embolization

Finding the systemic arterial supply is crucial for pulmonary sequestration diagnosis, as this is due to variable presentations and radiological patterns of the sequestered lung segment [21]. Variable feeding systemic vessels from the thoracic and abdominal aorta and their branches have been reported as shown in (Table 1), but most commonly arising from the descending thoracic aorta, followed by the upper abdominal aorta, and other feeders from the superior mesenteric artery, internal thoracic artery, renal, and celiac arteries have been reported. The question is what happens if the feeding vessel arises from the arteries of vital organs, such as the coronary circulation? Theoretically speaking, a pulmonary sequestration supplied by the coronary circulation can cause symptoms of ischemic heart disease through a mechanism of blood steal [22]. Although due to the rare incidence, the natural history of sequestration provided by a coronary artery is still unknown, and the presentation of ischemic heart disease, especially in old age, is due to atherosclerotic stenosis or the steal effect [23]. The case reported by Ioannis Tsitouridis et al showed that the myocardial ischemic changes occurred due to the steal phenomenon involving the left circumflex artery, which supplied the RT middle lobe sequestration, confirmed by thallium scintigraphy before and after exercise [22], while the case reported by D Sheshagiri Rao states that the stealing effect of the RT coronary artery causes ischemic ventricular tachycardia [23]. So Suspicions of coronary supply are crucial in sequestration patients, necessitating preoperative angiography before surgical ligation of the feeding artery, as the ligation or injury to the coronary artery may result in Infarction, myocardial ischemia, or even death, and it is important to mention that the feeding coronary artery may not be detected on aortogram, and selective coronary angiography is required [24].

The majority of ILS empties into the pulmonary veins, as in the current case [15]. However, it can be drained

through the azygos vein/hemiazygos system, portal vein, right atrium, or IVC [15].

The sequestered lung is usually supplied by a solitary feeding artery, although multiple feeders of single origin or from different origins have been reported in adults, like cases mentioned in table 1 [25-28] and in a 10-month-old infant case reported by Theodoropoulos et al [29].

The most common site of sequestered pulmonary segment documented in literature was the left lower lobe (74%) as seen in the current case, followed by the right lower lobe (25%), and extremely rarely in the upper lobe, accounting for less than 0.5% of all pulmonary sequestrations [10,67], as demonstrated in Table 2

The atypical position of these non-functional lung segments and variable radiological patterns, which sometimes resemble a mass, leads to misdiagnosis as malignancy, so identification of the feeding vessels is mandatory [68].

Table 3 shows the reported sequestration cases supplied by an aberrant artery from the celiac artery. It was found that most cases arise from the lower (Right or Left) lobes and presentation variable between hemoptysis (about 50%) and recurrent chest infection (about 50%), and one case presented with Left flank pain, in contrast to our case with is completely asymptomatic and incidental.

It has been stated that's ILS is uncommonly associated with other congenital anomalies [17]. Although other authors documented that about 5% of cases are associated with congenital anomalies [83]. The current case is associated with signs of symptoms of superior mesenteric artery syndrome, which is confirmed by CT scan. Table 4 shows the reported cases associated with congenital anomalies or other non-congenital pathologies. The acquired cases show aortitis and RA [50] and vasculitis (medium and large vessels)[84] raising the possibility of associated vascular anomalies or inflammatory conditions.

Table 4. The cases of intrapulmonary sequestration associated with other pathologies.

Reference	Year	Age at presentation	Sex	Side	Associated pathology	Presentation
1 E. HoRCHEr et al [85]	1987	6 months	Male	RLL	Scimitar syndrome	Repeated chest infection
2 Sueishi et al [50]	1989	48 years	Female	LLL	Aortitis and RA	Polyarthralgia of the extremities
3 Grewal et al [36]	1994	36 years	Female	LLL	Bronchogenic cyst	History of LLL pneumonia
4 MacKenzie et al [86]	2001	neonate	Female	LUL	Bronchogenic cyst and congenital cystic adenomatoid malformation	Antenatal diagnosis
5 Fujisawa et al [37]	2007	13 years	Female	RLL	Lung herniation	Fever and cough
6 Barbut J et al [71]	2010	3 weeks	Female	LUL	Bronchogenic cyst	Antenatal diagnosis
7 Nowak K et al [55]	2013	41years	Male	LLL	Carcinoid within	Hemoptysis
8 Gładki et al [87]	2023	5months	Female	RLL	Scimitar, ASD	RDS and tachypnea
9 Malik et al [84]	2014	25 years	Female	LLL	Medium and large vessel vasculitis	Abdominal pain, malaise, and weight loss
10 Lukman Lawal et al [88]	2015	67 years	Male	LLL	Adenocarcinoma within	Hemoptysis and recurrent pneumonia
11 Daniel Lim [63]	2018	48 years	Male	RLL	Bochdalek hernia	Right hydropneumothorax
12 Toumi et al [9]	2025	One month	Male	RLL	Extralobar sequestration	Intrauterine diagnosis

Malignant transformation within pulmonary sequestration has been documented in a few isolated case reports; however, being uncommon. The overall risk is thought to be quite low, and this possibility is still debatable. Although there were no malignant characteristics in our case, it is crucial to be aware of this uncommon relationship[89].

Case strengths

The current case is a rare coexistence of ILS with an aneurysmal feeding of the celiac and SMA. It provides a novel insight into uncommon conditions with a rigorous diagnostic approach using contrast-enhanced CT Scans. The latter efficiently reveals the reduced aortomesenteric angle and the aberrant vascular anatomy of SMAS and ILS, respectively. The associated review highlights the case finding in the current knowledge, underscoring the significance of recognizing atypical thoraco-abdominal symptoms. Such a case will improve our understanding of rare congenital and vascular anomalies, emphasizing the need for further investigation to unveil their pathogenesis for optimizing medical care.

Case limitation

Being a single case has its inherent limitations that restrict the generalization of its findings. The potential genetic and embryological link between ILS and SMAS remains unexplored. The diagnostic accuracy of the case is limited by the absence of histopathological confirmation. The follow-up period was not long, which constricted management and therapeutic insight. The lack of long-term possible complications or implications for this abnormality will limit the case's clinical applicability

Conclusion

The unusual coexistence of pulmonary sequestration and SMAS reflects a rather uncommon clinical situation. This example underlines the necessity of maintaining a high index of suspicion while investigating atypical thoracoabdominal symptoms, especially in young patients with unexplained gastrointestinal and respiratory issues.

This case added value to the limited body of knowledge on such uncommon vascular and congenital abnormalities,

underlining the need for further research and documentation to better understand their pathogenesis, clinical implications, and therapeutic outcomes. Early detection and focused treatments remain crucial to improving prognosis and preventing potential consequences.

Abbreviations

CT: Computed tomography.

LLL: Left lower lobe.

RLL: Right lower lobe.

LUL: Left upper lobe.

RDS: Respiratory distress syndrome.

ASD: Arterial septal defect.

RA: Rheumatoid arthritis.

Authors' contribution

SKA (Conceptualization; Methodology; Writing – original draft; Writing – review and editing); WN (Conceptualization; Methodology; Supervision; Writing – review and editing); QAH (Conceptualization; Validation; Supervision; Writing – review and editing); NNA (Supervision; Writing – review and editing).

Ethical statement

Written approval was gained from the Ethical Approval Committee of Al-Kindy College of Medicine. The patient gave informed consent.

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