

## Pediatric Hybrid Lung Lesion: A Case Series and Literature Review with a Unique Case of Congenital Pulmonary Airway Malformation (CPAM) and Bilateral Pulmonary Sequestration (PS)

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### Abstract

**Background:** Hybrid lung lesion combining PS and CPAM represent a complex subset of these anomalies. Bilateral PS is an exceptionally uncommon variant, characterized by non-functioning lung tissue supplied by systemic arteries on both sides.

**Objective:** To present a case series of hybrid lung lesion in children, including a rare case combining CPAM with bilateral PS, and to provide a literature review.

**Method:** A retrospective review of four patients with hybrid CPAM and PS was conducted using electronic medical records.

**Result:** Four patients with hybrid CPAM and PS were identified. One case demonstrated a rare presentation of CPAM with bilateral PS. All cases presented with respiratory symptoms, with variable age of presentation and imaging findings. All cases underwent surgical intervention with a favorable outcome.

**Conclusion:** Understanding these complex congenital anomalies and recognizing unusual presentations is crucial for appropriate patient care. Considering the variety of presenting symptoms is important. Management approaches range from conservative to surgical interventions, emphasizing the need for a multidisciplinary approach in diagnosis and treatment.

**Keywords:** Congenital Pulmonary Airway Malformation (CPAM); Pulmonary Sequestration (PS); Hybrid Lung Lesions

### Abbreviations

AA: Abdominal Aorta; AbPV: Aberrant Pulmonary Vein; Ao: Aorta; AoA: Aortic Arch; BC: Bronchogenic Cyst; Bil: Bilateral; BIL: Bilateral; CBCD: Complete Blood Cell Counts and Differential; CA: Celiac Artery; Cao: Coronary Artery; Cons: Conservative; CPAM: Congenital Pulmonary Airway Malformation; CT: Celiac Trunk; DAE: Diminished Air Entry; DA: Descending Aorta; DBI: Died Before Intervention; DiphA: Diaphragmatic Artery; ELS: Extra-Lobar Sequestration; EX-PT: Extreme Preterm; FBV: Feeding Blood Vessel; G6PD: Glucose-6-

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Phosphate Dehydrogenase Deficiency; HLL: Hybrid Lung Lesion; Hybrid lesion: Cystic Pulmonary Airway Malformation and Pulmonary Sequestration; ICA: Intercostal Artery; IDA: Infradiaphragmatic Aorta; ILS: Intra-Lobar Sequestration; IMV: Invasive Mechanical Ventilation; IR: Intervention Radiology; LAB: Large Aortic Branch; LBW: Low Birthweight; LCCA: Left Common Carotid Artery; LGA: Left Gastric Artery; LH: Left Hemi-Thorax; LL: Lower Lobe; LLL: Left Lower Lobe; MHA: Microcytic Hypochromic Anemia; MVS: Mitral Valve Stenosis; MVR: Mitral Valve Regurgitation; N: Normal; ND: Not Done; NF: Not Found; NI: No Information; PA: Pulmonary Artery; PBWT: Peri-Bronchial Wall Thickening; Post: Posterior; PS: Pulmonary Sequestration; PV: Pulmonary Vein; RD: Respiratory Distress; RLL: Right Lower Lobe; RUL: Right Upper Lobe; SA: Systemic Artery; SCA: Subclavian Artery; SS: Scimitar Syndrome; Surg: Surgical; TA: Thoracic Aorta; TDSA: Transdiaphragmatic Systemic Artery; UNC: Others Not Confirmed; VD: Venous Drainage; WBC: White Blood Cells; WES: Whole-Exome Sequencing

## **Introduction**

Congenital lung anomalies include hybrid lung lesions, which is combining features of congenital pulmonary airway malformation (CPAM) and pulmonary sequestration (PS). These conditions represent a rare and complex subset of congenital lung anomalies, and these lesions pose significant diagnostic challenges, requiring careful radiological and pathological evaluation and the importance of accurate classification [1]. Hybrid lung lesions with bilateral pulmonary sequestration are carry unusual presentation and there is limited scientific literature specifically addressing this condition.

Pulmonary sequestration is a rare congenital anomaly characterized by a mass of non-functioning lung tissue that is not connected to the normal bronchopulmonary tree and receives its blood supply from systemic circulation [2].

Sequestrations are typically classified as either intralobar or extralobar. While bilateral pulmonary sequestration is rare, it is important to note that pulmonary sequestrations can occur in either lung. The majority of extralobar sequestrations are found in the left lower lobe [3]. It's worth mentioning that the term "hybrid lesion" was first introduced by clinicians at the Children's Hospital of Philadelphia in 1998 to describe lesions that combine features of CPAM and bronchopulmonary sequestration, and these lesions are more common in males [4,5].

## **Methods**

A retrospective review of pediatric patients diagnosed with CPAM-PS lesions at King Faisal Specialist Hospital and Research Center and one case from King Soud Medical City.

Data were obtained from electronic medical records and imaging systems.

The requirement for informed consent was waived by the institutional review board of King Faisal Specialist Hospital and Research Center because the study involved a retrospective review of de-identified patients' records and did not include any direct patient contact or intervention.

Ethical approval was obtained from the institutional review board of King Faisal Specialist Hospital and Research Center, Riyadh, Saudi Arabia (RAC #2251364).

This retrospective study has been reported in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines.

## Case Presentations

### Case 1-AA (Table 1 and 2)

A 3-year-old boy with no significant past medical history was admitted to a local hospital due to persistent fever unresponsive to oral antibiotics. Chest radiography revealed necrotizing pneumonia, for which he received dual antimicrobial therapy. He was subsequently referred to our center for further evaluation. His antenatal, neonatal, medical, and surgical histories were unremarkable, and he was up to date with vaccinations and demonstrated age-appropriate development.

On examination, the patient was hemodynamically stable with normal vital signs and growth parameters within the normal percentile for age and sex according to WHO standards. He exhibited good bilateral air entry without respiratory distress but appeared pale; the remainder of the physical examination was unremarkable.

Laboratory investigations showed a white blood cell count of  $5.1 \times 10^9/L$  (70% neutrophils, 16% lymphocytes, 11% monocytes, 2% eosinophils), hemoglobin of 104 g/L with microcytic indices suggestive of chronic anemia, and normal platelet count. Immunoglobulin levels and vaccine's antibody responses were within normal limits, as were lymphocyte markers and histocompatibility class II expression. Arterial blood gas analysis indicated normal gas exchange. Notably, G6PD activity was reduced ( $< 1.0$  U/g Hgb).

Initial chest X-ray showed demonstrated multicystic changes predominantly in the left lower lobe with peribronchial infiltrates, mild perihilar interstitial thickening, and a left lower lobe airspace opacity containing cystic lucencies and an air-fluid level (Figure 1A), Contrast-enhanced CT of the chest (Figure 1B) revealed a prominent bilateral arterial branch originating from the descending aorta, supplying the posterior segments of both lower lobes. The left lower lobe contained multilocular, thin-walled cystic structures, the largest measuring  $2.6 \times 2.0 \times 3.5$  cm, as well as a thick-walled, pleural-based cyst with an internal air-fluid level and surrounding atelectasis and consolidation. There was evidence of air trapping adjacent to right lower lobe sequestration. Pulmonary venous drainage of the lower lobes was preserved (Figure 1B).

A multidisciplinary team approach was employed, and in collaboration with pediatric surgery, the patient underwent left lower lobectomy with surgical ligation of the aberrant feeding vessel.

Histopathological examination of the resected specimen (Figure 3A-3D) revealed a congested lung with dilated central and peripheral airways. The parenchyma displayed features of intralobar sequestration and lymphoid hyperplasia with foamy macrophage-rich mixed inflammation. Additionally, peripheral dilated bronchiolar structures, separated by alveolar tissue and reminiscent of CPAM, were noted—consistent with a hybrid lesion comprising intralobar sequestration and CPAM-like changes, likely related to intrauterine bronchial obstruction or pulmonary sequestration.

### Case 2- RH (Table 1 and 2)

A 14-month-old boy, with a known history of mitral valve stenosis and regurgitation, presented to the emergency department with fever, cough, and vomiting. He was part of a twin pregnancy, born preterm at 26 weeks of gestation with a low birth weight of 1.2 kg.

On admission, his vital signs were within normal limits. Anthropometric measurements revealed a weight at the 5<sup>th</sup> percentile and height at the 10<sup>th</sup> percentile for age and sex. There was no finger clubbing. Chest examination demonstrated equal bilateral air entry without added sounds.

Initial chest radiography revealed a cystic lung lesion in the right upper lobe, which had not been previously detected. The patient was admitted for intravenous antibiotic therapy and further evaluation.

Contrast-enhanced computed tomography (CT) of the chest identified a thin-walled, air-filled cystic lesion in the right upper lobe. Additionally, a large systemic artery arising from the descending aorta was noted, supplying a sequestered segment in the left lower lobe.

Based on these findings, a diagnosis of a hybrid lung lesion was established, specifically congenital pulmonary adenomatoid malformation (CPAM) associated with sequestration of the left lower lung lobe.

At the age of two years, the patient underwent surgical resection of the left lower lobe sequestration. Intraoperatively, coiling and closure device placement were performed to occlude the aberrant systemic artery supplying the sequestered lung tissue. The procedure was successfully completed without complications.

### **Case 3-AH (Table 1 and 2)**

A 14-month-old boy who presented to the emergency department with fever and cough. His past medical history was unremarkable.

On physical examination, his vital signs were within normal limits. His weight was at the 5<sup>th</sup> percentile and height at the 10<sup>th</sup> percentile for age and sex. There was no finger clubbing. Chest examination revealed equal bilateral air entry without any added sounds.

Initial chest radiography showed bilateral perihilar bronchial wall thickening. Moderate air space opacity was observed within the left lower lobe, accompanied by adjacent atelectasis and focal areas of increased lung lucency in the left lower lobe. These radiological findings were consistent with the clinical suspicion of congenital pulmonary airway malformation (CPAM) (Figure 2A and 2B).

Based on clinical and radiological assessment, a diagnosis of hybrid lung lesion was established, specifically, congenital pulmonary adenomatoid malformation (CPAM) associated with sequestration of the left lower lung lobe.

A multidisciplinary team reviewed the case, and, at 18 months of age, the patient underwent left thoracoscopic surgery, which was converted to thoracotomy. Left lower lobe resection and surgical ligation of the aberrant feeding vessel in the left lower chest were performed. The procedure was completed without complications.

### **Case 4-NA (Table 1 and 2)**

A newborn female who presented on the first day of life with significant respiratory distress, necessitating mechanical ventilation. On initial assessment, she was noted to be tachypneic and tachycardic. Physical examination revealed diminished air entry in the left lower lung field.

Laboratory evaluation, including a complete blood count, was within normal limits for age. Chest radiography demonstrated multiple cystic lesions in the left lung, initially raising suspicion for congenital diaphragmatic hernia. However, subsequent contrast-enhanced computed tomography (CT) of the chest identified a feeding systemic vessel in association with multiple cystic lesions in the left lower lobe.

Based on these findings, a diagnosis of a hybrid lung lesion-combining features of congenital pulmonary airway malformation (CPAM) and pulmonary sequestration (PS) in the left lower lobe-was established.

At 18 days of age, the patient underwent surgical resection of the left lower lobe along with ligation of the aberrant feeding vessel. The procedure was completed successfully and without complications.

Cases	Case 1-AA	Case 2-RH	Case 3-AH	Case 4-NA
Final Diagnosis	HLL: CPAM, Bil PS: ILS in the LLL and RLL	HLL: CPAM, ILS of the LLL	HLL: CPAM, ILS of the LLL	HLL: CPAM, PS of the LLL
Age of presentation	3 years	13 months	14 months	First day of life
Presenting symptoms	Persistent fever, cough	Fever, cough, vomiting	Fever, Cough	RD required IMV
Medical hx:	G6PD deficiency	Twin pregnancy, EX-PT 26 weeks, LBW of 1.2 kg, MVS, MVR, G6PD deficiency	Unremarkable	Unremarkable
<b>Physical examination</b>				
Vital signs	N	N	N	Tachypnea, Tachycardia
Growth Parameters	N	N	N	N
Chest exam	N	N	N	DAE in LLL.
<b>Intervention</b>				
Age	At 3 years	At 2 years	At 18 months	At 18 days
Procedure	Left thoracotomy LLL resection Surgical ligation of feeding FBV	Left Thoracotomy LLL resection Surgical Coiling and closure device placement of FBV	Left thoracoscopic converted thoracotomy LLL resection Surgical ligation of FBV	Left Thoracotomy LLL resection
Complications	None	None	None	None
Histopathology	Figure 3A and 3B: Patchy areas of fibrosis, lymphoid hyperplasia and accumulation of foamy macrophages suggestive of ILS Figure 3C and 3D: Cystic spaces resembling dilated bronchioles separated from each other by alveolar structures picture of CPAM	NF	Dilated airways with congested lung consist with intra-lobar sequestration. Rare space with no definitive cyst lining.	NF

**Table 1:** Clinical presentation of cases and histopathology examination.

*Legend table 1: HLL: Hybrid Lung Lesion; CPAM: Congenital Pulmonary Airway Malformation; Bil: Bilateral; PS: Pulmonary Sequestration; ILS: Intra-Lobar Sequestration; LLL: Left Lower Lobe; RLL: Right Lower Lobe; RD: Respiratory Distress; IMV: Invasive Mechanical Ventilation; G6PD: Glucose-6-Phosphate Dehydrogenase Deficiency; EX-PT: Extreme Preterm; LBW: Low Birthweight; MVS: Mitral Valve Stenosis; MVR: Mitral Valve Regurgitation; N: Normal; DAE: Demised Air Entry; RUL: Right Upper Lobe; FBV: Feeding Blood Vessel; NF: Not Found.*

Cases	Case 1-AA	Case 2-RH	Case 3-AH	Case 4-NA
<b>Lab workup</b>				
CBCD	WBC: N, MHA (G6PD).	WBC: N, MHA (G6PD).	WBC: N, Neutrophilia 75%, lymphopenia 22%, MHA	N
Immuno-globulins	N	ND	ND	ND
WES	ND	ND	Negative result	ND
<b>Radiology</b>				
Chest X-Ray	BIL PBWT LLL multi-cysts, air space opacity, increased lung lucency and air-fluid level. (Figure 1A)	BIL PBWT LLL Multi-cysts	BIL PBWT LLL air space opacity, atelectasis and increased lung lucency. (Figure 2A)	Diffuse opacity in LH
CT chest Angiography	LLL multiple cystic lesions, surrounding area of atelectasis and consolidation. Bil supplying systemic artery arising from the DA supply both LL. (Figure 1B)	RUL Multiple cystic lesion LLL heterogeneous density with multiple cysts Supplying systemic artery arising from the DA to LLL.	LLL multiple cystic lesions, surrounding area of atelectasis and consolidation. Supplying systemic artery arising from the TA to LLL. (Figure 2B)	LLL multiple cystic lesion

**Table 2:** Lab workup and radiology of hybrid lung lesion.

*Legend table 2: CBCD: Complete Blood Cell Counts and Differential; WBC: While Blood Cells; N: Normal; MHA: Microcytic Hypochromic Anemia; G6PD: Glucose 6-Phosphate Dehydrogenase Deficiency; WES: Whole-Exome Sequencing; ND: Not Done; BIL: Bilateral; PBWT: Peri-Bronchial Wall Thickening; LLL: Left Lower Lobe; LH: Left Hemi-Thorax; DA: Descending Aorta; LL: Lower Lobe; RUL: Right Upper Lobe; TA: Thoracic Aorta.*



**Figure 1A**



**Figure 1B**

**Figure 1:** Radiological images of case 1-AA. 1A: Chest x-ray showed multicystic changes, in the left lower lobe with peri-bronchial infiltrates, left lower lobe air space opacity, with a few air-cystic lucencies within and an air-fluid Level. 1B: Chest computed tomography angiography (CTA) of case 1, multiple cystic lesions on the left side with variable densities, blood supply coming from the lower descending aorta, and another blood supply going from the descending thoracic to the right side.

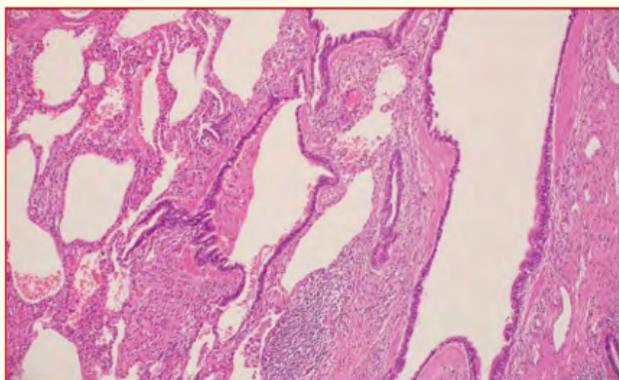


**Figure 2A**

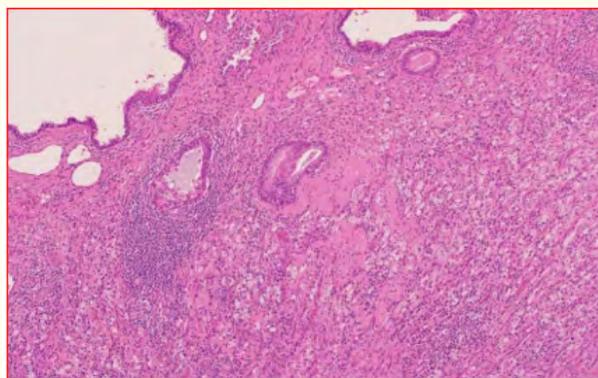


**Figure 2B**

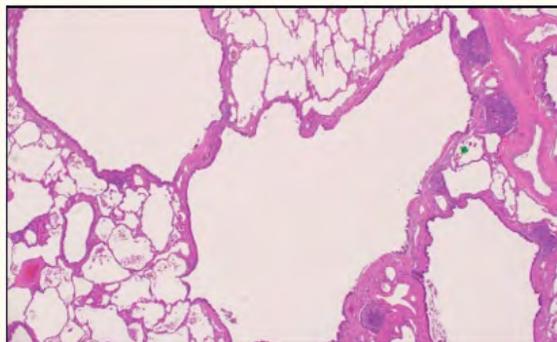
**Figure 2:** Radiology of case 3-AH. 2A: Chest x-ray, large cystic lesion in the left lower lung zone. 2B: Chest computed tomography angiography (CTA) of Case 3, multiple large cystic lesions filled with air, in the left lower Lobe.



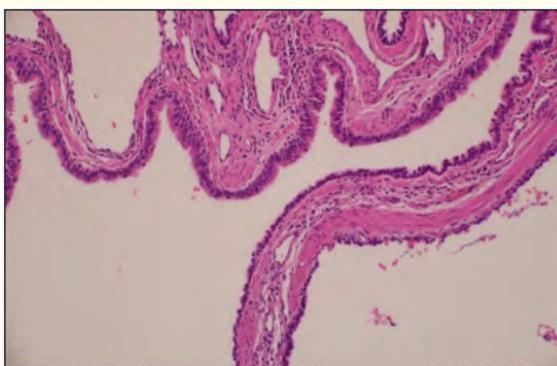
**Figure 3A**



**Figure 3B**



**Figure 3C**



**Figure 3D**

**Figure 3A-3D: Histopathology.**

*Figure 3A and 3B histopathology examination of case 1: Patchy areas of fibrosis, lymphoid hyperplasia, and accumulation of foamy macrophages suggestive of Intra-lobar sequestration.*

*Figure 3C and 3D histopathology examination of case 1: C: Cystic spaces resembling dilated bronchioles separated from each other by alveolar structures suggestive of CPAM. D: High power shows dilated airspaces lined by ciliated columnar epithelium.*

## Result

Four patients were identified, aged 1 month to 3 years. All presented with respiratory symptoms. A CT scan revealed cystic lesions with systemic feeding vessels. All patients underwent surgical intervention with favorable postoperative outcomes

## Discussion

Hybrid lung lesions, combining features of congenital pulmonary airway malformation (CPAM) and pulmonary sequestration, represent a rare and complex form of congenital lung anomaly. These lesions are characterized by the presence of both cystic adenomatoid tissue and an aberrant systemic arterial supply [1].

The coexistence of these two distinct pathologies in a single lesion highlights the intricate nature of embryonic lung development and the potential for multiple malformations to occur simultaneously [6].

Based on the referenced literature (Table 3), hybrid lung lesion represent a complex congenital anomaly combining pulmonary sequestration and congenital pulmonary airway malformation (CPAM). Conran, *et al.*'s and Cass DL, *et al.*'s reported published cases highlight the rarity and diagnostic challenges of these conditions [7,8]. Stocker and Husain's pathological perspectives emphasize the critical importance of precise classification of cystic lung lesions in children [9].

	Author	Year	Number of Cases	Lesion Type	Affected Site of lung	Surgery	Blood supply
1	Khushdil, <i>et al.</i> [1]	2018	1	Hybrid	LLL	Yes	AA VD from PV
2	Cass, <i>et al.</i> [8]	1997	6	Hybrid	LLL: 2 RLL: 3 Right chest region: 1	Yes	3AoV: 1 IDA: 1 TDSA: 3 LAB: 1
3	Conran and Stocker [7]	1998	50	Hybrid PS without CPAM PS Not classified with/out cpam	<b>Hybrid lesion:</b> Lt side: 17 Rt side: 4 Post mediastinum: 2 <b>BPS without CPAM:</b> Left: 11 Right: 6 Ant mediastinum: 4 Post mediastinum: 1 Chest wall, left diaphragm: 1 <b>BPS Not classified with/out CPAM:</b> Left: 3 Right: 1	NI	Aortic arch: 1 TA: 5 AA: 1 DA: 2 AO: 10 DiphA: 1 SA: 3 IDA: 3 TDSA: 2 CA: 2 PA: 3 LCCA: 1 IMA: 1 UNC: 6
4	A. Albassam, <i>et al.</i> [18]	1998	57	Cystic lung lesion	CLE: 37 CPAM: 7 BC: 8 PS: 5	Surg: 53 cons: 7	

**Table 3:** Summary of literature review included of hybrid lung lesion.

*Legend table 3:* VD: Venous Drainage; PV: Pulmonary Vein; TA: Thoracic Aorta; AA: Abdominal Aorta; DA: Descending Aorta; Ao: Aorta; DiphA: Diaphragmatic Artery; SA: Systemic Artery; IDA: Infradiaphragmatic Aorta; TDSA: Transdiaphragmatic Systemic Artery; CA: Celiac Artery; CT: Celiac Trunk; PA: Pulmonary Artery; LCCA: Left Common Carotid Artery; IMA: Internal Mammary Artery; LAB: Large Aortic Branch; AbPV: Aberrant Pulmonary Vein; 3AoV: Three Large Aortic Vessels; UNC: Others Not Confirmed; ICA: Intercostal Artery; AoA: Aortic Arch; SCA: Subclavian Artery; LGA: Left Gastric Artery; Cao: Coronary Artery; NI: No Information; Post: Posterior; Cons: Conservative; Surg: Surgical.

Bilateral pulmonary sequestration which is an exceptionally rare condition (Table 4), with only a handful of cases reported in the literature.

	Author	Year	Number of Cases	Lesion Type	Intervention	Blood supply
1	Roshan, <i>et al.</i> [15]	2023	1	BIL PS	Cons	TA
2	DePaRedes, <i>et al.</i> [16]	1970	12	ELS: 7 ILS: 5	Surg	TA: 9 IDA: 3
3	Joseph, <i>et al.</i> [11]	2024	1	BIL PS	Surg planned	TA
4	Savic, <i>et al.</i> [10]	1979	540	PS One whole lung: 5 Bil ILS: 2 ILS: 400 ELS: 133	Surg: 365 Cons: 1 NI 26	TA: 322 AA: 99 UNC: 43 <b>Bil BPS:</b> ICA: 1 AA: 2 TA: 1 UNC: 2
5	Stern, <i>et al.</i> [19]	N/A	17	Bil PS: 15 NI: 2	Surg: 15 DBI: 2	TA: 22 AA: 2 CA: 8 PA: 1 UNC: 1
6	Ou, <i>et al.</i> [17]	2014	48	PS	Surg: 24 IR: 2 Cons: 4	TA: 22 AA: 5 ICA: 1 UNC: 2
7	Zhu Y, <i>et al.</i> [3]	2011	2625	ILS: 1873 ELS: 358 Bil PS: 3	Surg	TA: 1384 AA: 334 ICA: 36 DA: 28 AoA: 8 SCA: 6 PA: 5 LGA: 4 CAo: 2 CT: 1
8	Van Raemdonck, <i>et al.</i> [2]	2001	28	ELS: 7 ILS: 21	Surg	TA: 17 AA: 9 ICA: 1
9	Bratu, <i>et al.</i> [6]	2001	39	ELS: 14 ILS: 8 Bil: 2  Communicating with esophageous: 2 Hybride lesion: 7 BC with ELS: 1 Multiple anomalies: 1 SS: 4	Surg: 36 Cons: 3 IR: 1	DA:1 Ao: 1 PA: 1
10	Von Scheidt F, <i>et al.</i> [22]	2018	1	Bil PS	AE	AA

11	Tekinhatun M., <i>et al.</i> [23]	2023	1	Bil PS	NI	CA
12	Oliveri Aruete F., <i>et al.</i> [34]	2017	1	Bil PS	Surg	AA TA
13	Nagendran S., <i>et al.</i> [24]	2009	1	Bil PS	Surg	TA
14	lv l., <i>et al.</i> [25]	2022	1	Double ELS	Surg	AA
15	Gudbjartsson T., <i>et al.</i> [26]	2007	1	Bil ELS	Surg	AA
16	Poh ME., <i>et al.</i> [27]	2018	1	Bil ILS	Surg	TA
17	Fontalba Navas M., <i>et al.</i> [28]	2013	1	Bil ILS	NI	TA
18	Murugesan N., <i>et al.</i> [29]	2024	1	Bil PS	NI	CA
19	Ren G., <i>et al.</i> [30]	2023	1	Bil PS	NI	TA
20	Joy MG., <i>et al.</i> [31]	2005	1	Bil ILS	Surg	CT
21	Gülsen., <i>et al.</i> [32]	2019	1	Bil ILS	Cons	CT
22	Woerner A., <i>et al.</i> [33]	2008	1	Bil ILS	Surg	CA

**Table 4:** Summary of literature review included of bronchopulmonary sequestration.

*Legend table 4:* Bil: Bilateral; PS: Pulmonary Sequestration; Cons: Conservative; TA: Thoracic Aorta; ELS: Extra Lobar Sequestration; ILS: Intra-Lobar Sequestration; Surg: Surgical; IDA: Infradiaphragmatic Aorta; NI: No Information; AA: Abdominal Aorta; UNC: Others Not Confirmed; ICA: Intercostal Artery; DBI: Died Before Intervention; PA: Pulmonary Artery; IR: Intervention Radiology; DA: Descending Aorta; AoA: Aortic Arch; SCA: Subclavian Artery; LGA: Left Gastric Artery; Cao: Coronary Artery; CT: Celiac Trunk; Hybrid Lesion: Cystic Pulmonary Airway Malformation and Pulmonary Sequestration; BC: Bronchogenic Cyst; SS: Scimitar Syndrome; Ao: Aorta; AE: Arterial Embolization; CA: Celiac Artery.

Basd on Zhu., *et al.*'s retrospective analysis of 2,625 cases in China and Savic., *et al.*'s comprehensive review of 540, the rarity of bilateral sequestration can be attributed to the complex embryological processes involved in lung development. Most cases of pulmonary sequestration are unilateral, making bilateral occurrence a unique and challenging presentation for both diagnosis and management [3,10].

The presentation and management of pulmonary sequestration vary significantly based on its type. Intralobar sequestration (ILS) often remains asymptomatic for years and is frequently discovered incidentally during imaging studies performed for unrelated conditions. When symptomatic, the most common clinical presentations include recurrent pneumonia, cough, and hemoptysis. On the other hand, extralobar sequestration (ELS) typically manifests in early infancy, often presenting with respiratory distress. ELS is also commonly associated with congenital anomalies such as congenital diaphragmatic hernia, cystic pulmonary adenomatoid malformation, congenital heart disease, and tracheoesophageal fistula [11].

The long-term presence of these lesions, even if asymptomatic, can lead to complications such as infection, bleeding, heart failure, or malignant transformation, emphasizing the importance of early detection and intervention [6].

The management of hybrid lung lesions and bilateral sequestration requires a multidisciplinary approach. surgical intervention is generally recommended for symptomatic patients or those at risk of complications [6].

The work of Ren., *et al.* specifically demonstrates the potential for hybrid lesions in adult patients, particularly in the context of complex clinical scenarios like COVID-19. Ziegler., *et al.*'s underscores the necessity of a comprehensive approach to managing these rare developmental lung anomalies [12].

Surgical treatment is critical in managing all cystic lung lesions, either isolated cyst or associated with (PS) as a hybrid lesion, to prevent complications [6].

Options include open thoracotomy, which involves a larger incision and may result in more postoperative pain, versus thoracoscopic surgery, a minimally invasive approach linked to reduced postoperative discomfort. With advancements in video-assisted thoracic surgery (VATS) instrumentation and visualization systems in the era of minimally invasive thoracic surgery, VATS is recommended as the primary approach. Open surgery should be reserved for more complex cases [13].

Early surgical intervention offers benefits by preventing respiratory symptoms and chronic inflammation from recurring infections. Even asymptomatic cases of intralobar sequestration (ILS) may require treatment to minimize the risks of recurrent pneumonia, lung abscesses, pneumothorax, or rare malignancies [13].

Hybrid lung lesions, combining features of (CPAM) and (PS), present a significant diagnostic challenge in both pediatric and adult populations. This case underscores the critical importance of maintaining a high index of suspicion for these rare congenital malformations. Accurate diagnosis hinges on a comprehensive approach, including a detailed medical history with particular attention to episodes of recurrent or treatment-resistant pneumonia, coupled with a thorough radiological assessment of chest pathology. The potential for misdiagnosis is considerable, emphasizing the need for heightened clinical vigilance, as undetected hybrid lesions can lead to serious complications and substantially impact patient quality of life.

Continued research and long-term follow-up studies are essential to further our understanding of these rare conditions and optimize management strategies.

This report highlights the importance of considering unusual presentations and the possibility of hybrid lesions with bilateral involvement lung sequestration components. A multidisciplinary approach, incorporating expertise from radiology, pulmonology, pediatric surgery, and histopathologist is crucial for timely identification and appropriate management of these complex congenital lung malformations.

## **Conclusion**

In conclusion, hybrid lung lesions and bilateral pulmonary sequestration represent rare and complex congenital anomalies that pose unique challenges in diagnosis and management. The rarity of these conditions, particularly bilateral sequestration, underscores the importance of thorough evaluation and individualized treatment planning. Surgical intervention remains the mainstay of treatment, and this approach should be tailored to the patient's age, symptoms, and overall health status.

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