Case Report

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Bilateral Intralobar Pulmonary Sequestration in an Elderly Patient

Internal Medicine Section

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ABSTRACT

Pulmonary Sequestrations (PS) are congenital anomalies resulting from defective pulmonary development. PS consist of abnormal lung tissue that is not associated with the normal airway. They have their own abnormal vascular system consisting of arterial connections (usually from the aorta and its branches) and venous drainage through pulmonary venules. These lung lesions do not contribute to the exchange of oxygen and carbon dioxide. However, intralobar PS is usually seen in childhood, unilaterally in the left lower lobe of the lung. PS is rarely encountered bilaterally or in older patients. Herein, we describe a case of bilateral PS detected by computed tomography angiography in an elderly patient. The patient refused additional interventional examination and operation, so clinical follow-up was continued.

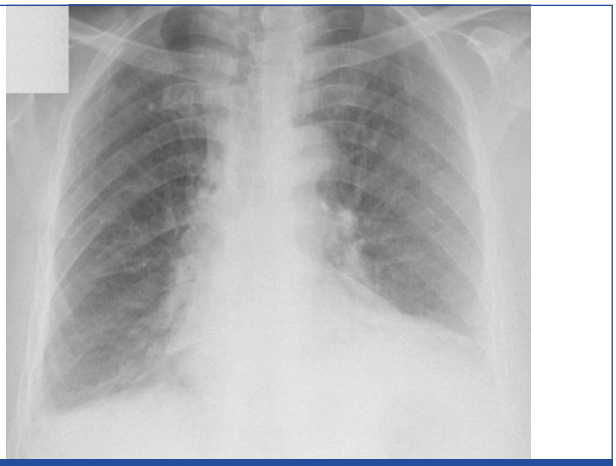
Keywords: Asymptomatic, Drycough, Lung

CASE REPORT

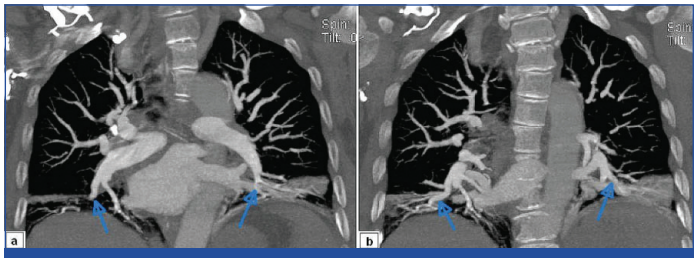
A 64-year-old female patient was admitted to the university hospital chest disease clinic due to chest pain, following one month history of persistent dry cough and a flu infection. The patient had no chronic complaints other than a dry cough. Otherwise, the patient's history was not noteworthy. She had no known lung disease, no complaints of chronic cough or sputum production, and no recurrent pulmonary infections.

A physical examination revealed a blood pressure of 130/90 mm Hg; pulse rate: 68 beats/min; respiratory rate: 15 breaths/min; and body temperature: 38°C. On respiratory system examination, the breath sounds were reduced in the lungs in bilateral lower lobes. Complete blood count showed a white blood cell count of 12.500 10^3 cells/mm^3, a C-reactive protein level of 5.4 mg/dL, and D-dimer level of 550 µg/L.

Chest radiograph showed right sinus blunting, increased cardiothoracic ratio, and possible left lower lobe consolidation [Table/Fig-1]. Chest Computed Tomography Angiography (CTA) scan was performed to rule out pulmonary embolism. CTA revealed a bilateral aberrant lung tissue with left and right abnormal systemic arterial supply separated from celiac truncus, and a venous vessels flowing into both lower pulmonary veins [Table/Fig-2a,b]. There was no sign of pulmonary embolism. The lesions in the CTA scan were compatible with bilateral PS. Echocardiographic evaluation showed no abnormal findings.



[Table/Fig-1]: Chest radiograph showing right sinus blunting, increased cardiothoracic ratio, and possible left lower lobe consolidation.



[Table/Fig-2]: CT-Angiography showing bilateral aberrant lung tissue with left and right abnormal systemic arterial supply separated from celiac truncus, and a venous vessels flowing into both lower pulmonary veins.

Antibiotic treatment was started because of increased inflammation parameters and mild fever. Cough and chest pain regressed slowly within four weeks. Surgical treatment for bilateral intralobar PS was recommended, but she refused additional interventional examination and operation, so clinical and radiological follow-up was continued.

DISCUSSION

Patients with Pulmonary Sequestration (PS) may present to the clinics with no symptoms, unknown fever, haemoptysis, pulmonary infections, dry cough, dyspnea or progressive respiratory distress [1]. Along with these symptoms, chest radiographic pattern of the patients are usually unilateral nonhomogeneous parenchymal consolidation in the lower lobe [2]. This congenital anomaly results in a non-functional area of the lung following a deterioration in embryogenesis. They have their own abnormal vascular systems (often, arterial feeding through aorta, and venous drainage through pulmonary venules) and are not connected to the bronchial system [Table/Fig-3] [3]. These lung lesions do not contribute to the exchange of oxygen and carbon dioxide [4]. In 1946, Pryce and colleagues used the terms "extralobar" and "intralobar" PS for the first time [5]. PS is more commonly intralobar (75%) than extralobar (25%) [6]. Intralobar PS is usually seen in childhood, unilaterally in the left lower lobe of the lung [7].

PS is a rare congenital anomaly that accounts for 0.1% to 6.4% of all pulmonary malformations, and rarely encountered bilaterally or in older patients [8]. Wei Y et al., completed the most extensive work on this subject in 2011 [9]. As a result of the analysis of 2625 PS patients, it was reported that only three of them had bilateral PS and two had intralobar PS. In a review of 540 patients by Savic B et al., only two of the patients had bilateral, intralobar PS [8]. Therefore, the index patient is very rare because of the presence of bilateral

	Extralobar	Intralobar
Anatomy	Within lung parenchyma	Separate from lung
Localisation	Within basal segments	Below lower lobe
Predominant side	Left	Left
Age	60% <1 yr of age	50% adults; Rare <2 yr of age
Male /Female	4:1	1:1
Artery	Systemic	Systemic
Vein	Systemic (azygous)	Central (pulmonary vein)
Associated anomalies	Frequent (diaphragmatic hernia)	Rare
Origin	Congenital	Congenital or acquired

[Table/Fig-3]: Characteristic features of pulmonary sequestration.
Quoted from Nicolette LA, et al., [3] with permission

PS and asymptomatic clinically and he was over 50 years of age. Approximately, half of PS patients are diagnosed before the age of 20 with the first symptom appearing before the age of 10 years in 37.2% of cases [8,10]. PS may lead to various chronic symptoms or may be asymptomatic. It causes complaints such as cough, fever, frequent sputum production, and very rarely chest pain [9]. It is often an incidental finding during chest radiography in patients with frequent, recurrent, or incurable lung infections. Recurrent respiratory tract infections are the most common reason for referral to clinics. In addition, 13.3-15.5% of the cases are reported to be asymptomatic and incidentally diagnosed. This patient was referred because of a persistent dry cough and a new onset chest pain that did not resolve after the flu-infection.

In a recent study, the most common symptoms in patients was cough and sputum (82.6%) [11]. In addition, intermittent fever, chest pain and haemoptysis have also been reported, 17.4%, 13.0% and 4.0%, respectively. The mean age of the patients was 48.3 years, 17.4% were asymptomatic and the diagnosis was made incidentally. CT findings were reported as solid (69.5%), mixed cystic-solid (30.4%) and cystic (4.3%) [11].

Chest CTA examination often reveal mass lesions, cystic lesions, cavitary and pneumonic lesions (49.0%, 28.5%, 11.5% and 7.9%, respectively) [9]. The differential diagnosis of PS are: lung cancer, lung abscess, empyema, congenital cystic malformation, diaphragmatic hernia, bronchiectasis and pneumonia [12]. Multidetector-row CTA is the most commonly used diagnostic test, and surgical procedures including video-assisted thoracoscopic surgery are recommended in symptomatic cases.

However, the surgeries can be performed by thoracotomy or thoracoscopic approach. Lobectomy can generally be performed as sub-lobar resection, segmentectomy or atypical resections [13]. Evaluation of the artery, feeding the PS segment is very important in patients planned for resection. In addition, pre-operative embolization may be required in patients with hemoptysis and artery fed from the abdominal aorta [1]. Coil embolization may be used as an alternative to surgery in patients with intralobar PS [14]. [Table/Fig-4] shows an overview of previous studies [1,8,9,11,15].

Author	Year	n	IL %	EL %	IL+EL %	Bilateral %	Age	Asymp. %
Schlöricke E et al., [1]	2016	14	79.0	21.0	-	-	mean 50 y	21.4
Savic B et al., [8]	1979	540	74.0	24.6	1.1.	0.3	<20-%50	15.5
Wei Y et al., [9]	2011	2625	83.9	16.0	-	0.1	-	13.3
Dong Q et al., [11]	2019	23	100	-	-	-	mean 48.3 y	17.3
Alsunraim M et al., [15]	2018	32	81.0	19.0	-	-	mean 42 y	40.0

[Table/Fig-4]: Overview of previous published studies [1,8,9,11,15].
n: Patients; y: Years; IL: Intralobar; EL: Extralobar; Asypm: Asymptomatic

CONCLUSION

PS is rarely seen in a patient over 60 years of age, but is diagnosed incidentally in symptomatic cases. Present patient is one of the very few cases in the literature due to bilateral, asymptomatic and elderly presentation. We would like to emphasise the need to consider PS as well as other possible causes, in the light of differential diagnoses such as pneumonia, pleural effusion and embolism.

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