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**A Huge Pulmonary Sequestration in An Elderly Patient; A Case Report**  
*Yaşlı Hastada Çok Büyük Boyutlu Pulmoner Sekestrasyon Olgusu*

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**Abstract**

Pulmonary sequestration (PS) is a rare congenital pulmonary malformation with unknown origin. It may be clinically asymptomatic, or may cause recurrent infections, chronic cough, and hemoptysis. PS has two types as intralobar (ILS) and extralobar sequestration (ELS). The mainstay of diagnosis is the identification of the aberrant systemic arterial supply. We report a case of an elderly patient who has 17x14x15 cm sized mass of intralobar pulmonary sequestration supplied via the coeliac trunk. To the best of our knowledge, this case represents the oldest patient with a large sized PS in the literature with aberrant arterial supply from the coeliac trunk. The reported case is presented along with current literature review.

**Keywords:** Pulmonary Sequestration, Coeliac Artery, Computed Tomography

**Özet**

Pulmoner Sekestrasyon (PS), akciğer parankiminin oldukça nadir ve etyolojisi bilinmeyen konjenital malformasyonudur. Klinik olarak asemptomatik olabileceği gibi tekrarlayan hemoptiziler, kronik öksürük ve akciğer enfeksiyonları ile de başvurulabilirler. İntralobar ve ekstralobar olmak üzere iki tipi vardır. PS sistemik arterden kanlanır ve bunun gösterilmesi ile tanı konulur. Çölyak trunkustan aberran sistemik arteri gösterilerek tanı konulan 17x14x15 cm boyutlarında intrapulmoner sekestrasyon olgumuzu, literatürdeki en ileri yaştaki hasta olması, yetişkinde en büyük boyutlardaki olgu olması ve çölyak trunkustan arteryal kanlanmanın pulmoner sekestrasyon olguları arasında oldukça nadir bulunması nedeniyle literatür eşliğinde sunmayı uygun bulduk. **Anahtar Kelimeler:** Pulmonary Sekestrasyon, Çölyak Arter, Bilgisayarlı Tomografi

## Introduction

**P**ulmonary Sequestration (PS) is a rare congenital malformation of the lung parenchyma of unknown etiology. It may be asymptomatic or be the cause of recurrent infections, chronic cough and hemoptysis. It may mimic the lung cancer on chest radiograms. There are two types of PS: Intralobar (ILS) and extralobar (ELS). The mainstay of diagnosis is the identification of the aberrant systemic arterial supply.

PS was first described by Rochitansky and Rectorzik in 1861, as an accessory pulmonary lobe. The term, PS, was first used by Pryce in 1946. PS occurs in two forms: Intralobar Sequestration (ILS) and Extralobar Sequestration (ELS). Both of them have a common embryopathogenic basis. The majority of the PS consists of ILS (75% of all cases). ILS approximately equally distributed between sex while ELS is found more commonly in men (80% of cases) (1,2,5,8).

Here, we present a 77 year old patient who had been healthy with an ILS sized 17x14x15 cm with a systemic arterial supply from the coeliac trunk. To the best of our knowledge, this case represents the oldest patient with large sized of PS among the cases in the literature with aberrant arterial supply from the coeliac trunk. The reported case is presented along with current literature review.

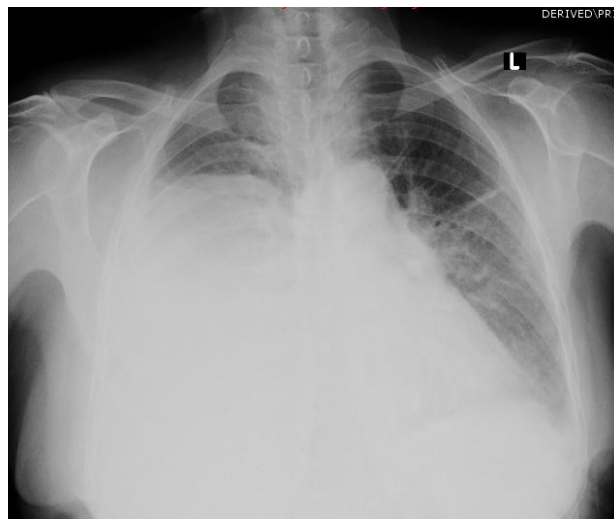
## Case presentation

A 77-year-old female patient presented to our pulmonary diseases outpatient clinic with a complaint of exertional dyspnea. Her symptom was not associated with hemoptysis, chronic cough, sputum production, fever, and chest pain. Her medical history was unremarkable; there was no history of recurrent pulmonary infections.

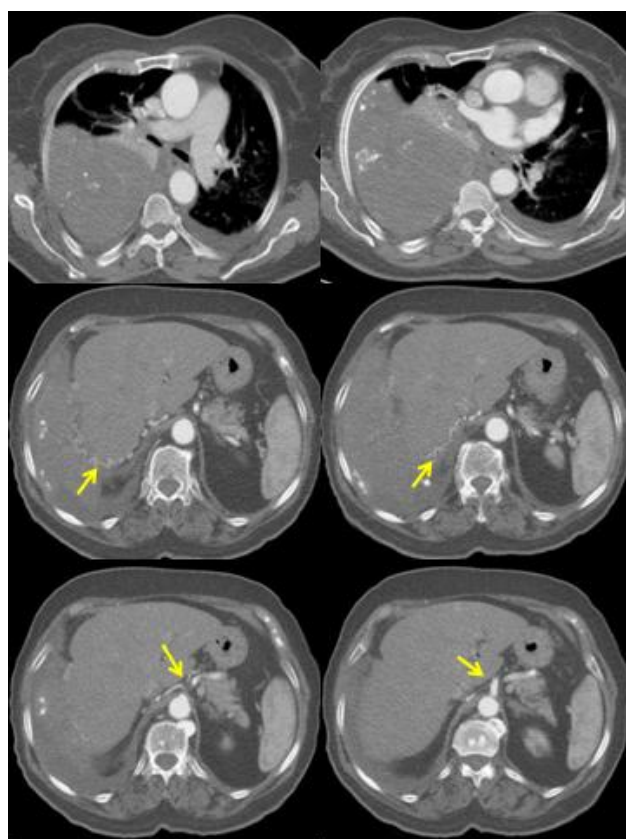
The physical examination revealed decreased breath sounds in right lower lobe and in the area below the right scapula. Vibration thoracic was lost and there was the sign of dullness on the same area. Other physical examinations and laboratory findings were normal.

The posterior-anterior chest radiograph showed the deletion of the cardiac silhouette by a homogenous lesion (Figure 1). Computed Tomography (CT) of thorax with intravenous contrast injection was performed for further evaluation. The CT revealed a PS with an aberrant systemic arterial supply to the lower right lobe. The origin of the arterial supply was coeliac trunk. The size of the PS was 17x14x15 cm (Figure 2).

According to the these findings the diagnosis of ILS originated from celiac trunc was made. Surgery was considered but the patient refused any operation. The patient is currently alive and followed by medical treatment.



**Figure 1.** Chest X Ray. Deletion of the cardiac silhouette by a homogenous lesion in right hemithorax and shift of mediastinum to the left.



**Figure 2.** Computed Tomography (CT) of thorax with intravenous contrast. The arterial supply of pulmonary sequestration originating from coeliac trunk was shown by yellow arrow.

## Discussion

Pulmonary sequestration (PS) is an uncommon congenital malformation of the lung, consisting of about

0.15 to 6.45 % of all congenital pulmonary anomalies. It is a non-functional mass which is not affiliated with the trachea – bronchial system. PS occurs in two forms: Intralobar Sequestration (ILS) and Extralobar Sequestration (ELS). Both of them have a common embryo-pathogenic basis. The majority of the PS consists of ILS (75% of all cases). ILS approximately equally distributed between sex while ELS is found more commonly in men (80% of cases) (1,2,5).

The ELS is surrounded by visceral pleura whereas ILS is encapsulated within a distinct pleural covering separate from the normal lung tissue. Therefore, ELS appears as a different pulmonary lobe (1,2). Sixty to seventy percent of ILS and the 90 % of the ELSs are located in the left hemithorax. ILS and ELS are usually observed in the posterobasal segment of the lower lobes which preferring the paravertebral areas (1-3). The localization of the lobes except for the lower lobes is uncommon (4). The study of Savic et al showed that only 9 out of 540 PS cases were located in upper or middle lung lobes (5). Contrary to the general opinion, in our case, PS was located in the right lower lobe.

Associated congenital abnormalities are uncommon in ILS whereas 50 % of ELS cases are associated with diaphragmatic and cardiac congenital abnormalities (2). In our case, there was not any associated congenital anomaly.

Patients with PS are usually symptomatic. Asymptomatic lesions may be latent until infection. Degeneration of the normal lung tissue and trauma lead to symptoms. The most common symptoms are fever, cough, purulent sputum, hemoptysis and chest pain. The presentation of ILS is usually as a recurrent pneumonitis (1,2,5). Medical history in our case was unremarkable; there was no history of recurrent pulmonary infections. Her single complaint was exertional dyspnea.

The clinical presentation of PS may appear at any age. However, many of the PSs are asymptomatic and therefore are not discerned until adulthood and are discovered during routine tests. The sixty percent of patients with ELS are diagnosed during childhood, whereas most of patients with ILS are diagnosed during adolescence or early adulthood. Asymptomatic patients with ILS until advanced age, as in our case, is found to be rare in the literature (1,5-8).

The radiographic findings of PS are infiltration of pneumonitis, cavity containing an air-fluid level suggesting complicated PS by infection, and rarely a dense opacity or mass (9,10). In our case, the chest radiography revealed a diffuse homogeneous lesion deleting the cardiac silhouette in the right hemithorax suggesting pleural effusion. The CT of thorax showed large mass (17x14x15 cm) with localized calcification areas and slight pleural effusion.

The sequestered pulmonary tissue is vascularized by an aberrant artery which derives from systemic arteries. This aberrant artery is originated in 75

% of all cases from the thoracic aorta, and in 25 % from the abdominal aorta. PS has no direct connection to the tracheobronchial tree or to the pulmonary arteries. Meanwhile, different origins and numbers of aberrant artery are reported in literature (1,2). In our case, CT with contrast revealed a PS with an aberrant systemic arterial supply to the lower right lobe. The origin of the arterial supply was coeliac trunk (Figure-2). This type of arterial supply of a PS by coeliac artery is seen very rare in the literature (1,2,5,8).

The mainstay of diagnosis of PS is the identification of the systemic aberrant arterial supply by angiography. Computed tomography angiography (CTA) may suffice in many cases as it successfully delineates the origin and course of the anomalous systemic artery. The development of CTA imaging reduced the need for angiography which is an invasive technique. Magnetic resonance angiography is also helpful for showing the aberrant arterial supply and especially for planning of the surgery. The venous return of the ILS is via the pulmonary veins, whereas the ELS drains into systemic veins via azygos and hemi-azygos veins. Rarely, there are cases reported in which the PS drains into various veins and atrium (8-10). In our case, venous drainage could not be showed because of both tomography shooting technique and patient's refusal to the operation.

The case reported here was referred to our clinic for the suspicion of lung malignancy due to her advanced age. The diagnosis of PS was made by showing the aberrant arterial supply originating from the coeliac trunk, which is rare in the current literature. Since the patient refused surgery, the diagnosis could not be verified pathologically.

The recommended treatment of PS is surgical resection of the involved lung in order to avoid infection, massive hemoptysis and the destruction of the normal pulmonary tissue, even in asymptomatic patients with PS (2,5). In some PS cases with infection and pulmonary damage, lobectomy or pneumonectomy should be the choice of treatment (11). The long-term outcome is highly favorable.

Limitations of our present case report is the lack of histopathological diagnosis of the sequestration because of the absence of patient's informed consent for operation. In conclusion, we reported a huge sized intralobar pulmonary sequestration in the oldest patient with its arterial blood supply from celiac trunk diagnosed by CT angiography first in the literature.

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