

A Rare Case of Pulmonary Sequestration in a Young Male Patient

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SUMMARY

We present a case of a young male administered to our pulmonary clinic with symptoms that included high fever, pain into the chest and dyspnea. On auscultation there were bilateral crackle sounds, while on chest X-ray there were signs of pneumonia. There were three previous hospitalizations due to necrotizing interstitial pneumonias. Our patient underwent bronchoscopy and microbiological tests were sampled. He stayed into the hospital for 10 days. The investigation included a CT scan plus angiography that confirmed the diagnosis of pulmonary sequestration.

Background

Pulmonary sequestration belongs to rare congenital lung malformations with a population impact around 6% of all inherited lung disorders. There is an autonomic dysplastic lung segment or lobe without communication with the trachea or bronchi and with abnormal separate blood supply. It is divided into three main categories. Pulmonary sequestration can be into the lung and it is called intralobar sequestration (ILS), it can be outside of the lung like an abnormal "third lung" into the thoracic cavity and it is called extralobar sequestration (ELS) or it can be subdiaphragmatically.

Case Presentation

A 26-year-old male patient visited the emergency department of our pulmonary clinic with severe chest pain, dyspnea (breathing difficulty), dry cough, shortness of breath, and fever between 37,8C-38,8C that started 6 days before his hospitalization. He was a smoker, but he quit a year ago. He consumed alcohol occasionally. He had a dry cough that was not connected to allergies and did not have diurnal variations. He experienced chest pain which affected his sleeping and

daily activities. Dyspnea was persistent and it was not connected to any heart complications as it was confirmed on multiple heart examinations including ECG and echocardiography. In his medical history he had three previous hospitalizations due to necrotizing interstitial pneumonias with similar symptoms including mainly dyspnea, cough, chest pain and high fever. Previous chest X-ray examinations showed an opacity in the right lower zone of the lung. On auscultation bilateral crackles were audible in the lung bases. X-ray showed pneumonia of the right lung so an intravenous (iv) antibiotic treatment with piperacillin plus tazobactam was administered. He also received short acting bronchodilator. We collected blood samples in order to estimate inflammatory markers such as white blood cells WBCs, c-reactive protein (CRP), state of renal function (urea and creatinine) as well as electrolytes. He underwent a Broncho-alveolar lavage (BAL). Ebus- bronchoscopy was also performed and we collected samples for biopsy from the lungs that were tested for tuberculosis. A Mantoux test was done. His blood oxygen saturation was 98%, heart rate 86 beats/minute, he had a normal- ECG and he was three-time vaccinated against Covid-19.

Immediately after his hospitalization and initiation of antibiotic treatment, there was a significant remission of fever from 38C to 37C. However, on the fourth day the patient showed an increase of his fever to 39C persistently so we changed our antibiotic regimen from piperacillin/tazobactam to linezolid and meropenem for 10 days. In addition, we repeated bronchoscopy, blood and bronchoalveolar lavage cultures. Blood gas analysis showed a pH-7,473 with pO₂-80mmHg and pCO₂-36mmHg. Although our patient improved after the initiation of the second line antibiotic treatment resulting in improvement of his physical status, auscultation findings were still abnormal exhibiting

bilateral crackle sounds. The blood cultures and the bronchoalveolar lavage cultures were tested negative for a specific infectious cause. We decided to perform a CT scan examination plus angiography. CT angiography scan proved the presence of a lung sequestration with different blood supply than the rest of the lungs originating from the descending thoracic aorta which is extremely susceptible into lung infections and inflammation [1] due to its abnormal blood supply as well as due to the presence of dysfunctional lung tissue. Our patient had a right extralobar sequestration located at the base of the right lung. The size of sequestration affected a whole lung lobe (Figure 1).

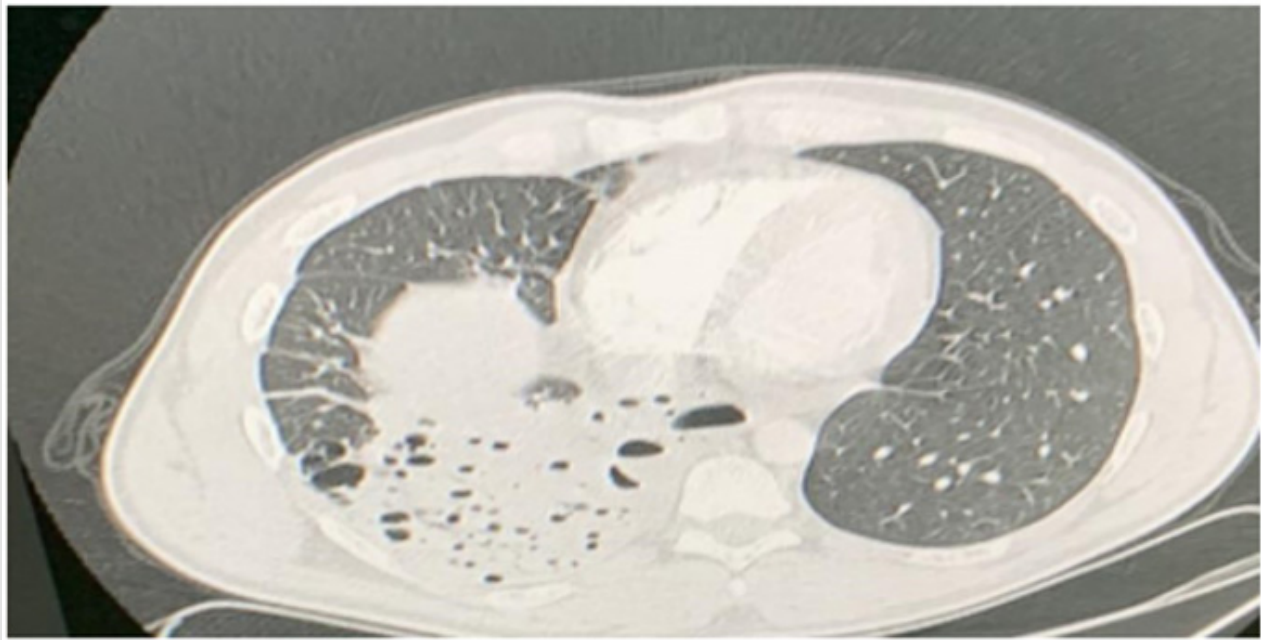


Figure 1: Pulmonary sequestration imaging of our patient.

Investigations

Described in the course of the case report.

Differential Diagnosis

Differential diagnosis in our patient included bacterial or viral pneumonia, tuberculosis, inherited abnormal airway malformations, lung abscess or cysts, and lung cancer. Lab tests and blood and bronchoalveolar lavage cultures excluded the case of bacterial pneumonia as well as lung cancer through pathoanatomical examination of lung tissue. Mantoux test was negative.

Treatment

Treatment included linezolid, berovent, meropenem and Nexium for 10 days and we decided that in case of new relapsing a thoracic surgery can be done in order to remove the sequestrant avoiding further complications.

Outcome and Follow Up

The last 2 days of hospitalization (9th and 10th day), our patient had no fever, he had clear auscultation without cough and dyspnea and we released him from the hospital. Our patient was finally evaluated by thoracic surgeon and we advised him to consider a future surgical resection of the lung sequestration to prevent possible relapses and complications.

Discussion

Pulmonary sequestration is defined as an abnormal lung tissue with independent blood supply. [1-4] It accounts for around 6 percent of all inherited lung malformations. [4] It is divided as intralobar sequestration, extralobar sequestration and extralobar subdiaphragmatic sequestration which is extremely rare. There is an abnormal lung tissue segment, lobe or even bigger that is separated from the rest tracheobronchial tree and with independent blood supply and

venous drainage. It causes several episodes of pneumonia, hemoptysis, cough, chest pain, dyspnea and fever. There are several difficulties for diagnosis due to the location of it but CT scan and CT scan angiography are the methods of choice for confirmation. [1,5] In some cases MRI may also be performed. [4] Lung cancer, pleural or mediastinal tumors and lung cysts are several cases of misinterpretation of lung sequestration. [6] In addition, in some cases there are multiple fibrotic areas into the sequestration, around of it while there is possible bronchiectasis into the rest normal lung. [1] It is mainly a childhood disease because there are multiple and severe attacks of recurrent pneumonias so surgical resection is the treatment of choice. [1] The interesting fact of our case is that we have an adult patient without so severe cases of pneumonia during childhood but with several recent attacks during adulthood. Blood supply of sequestration may come directly from thoracic aorta, bronchial arteries, abdominal aorta or may derive also from other vessels such as intercostal arteries, diaphragmatic artery, aortic arch, subclavian artery, brachiocephalic artery, pulmonary artery, left gastric artery, coronary artery, arteria lienalis, celiac trunk and renal artery.

[7] Lung sequestration in extremely rare cases is related with lung malignancies or with presence of two sequestrations in both lungs simultaneously [8,9] and in these cases surgical resection is done immediately. However, in cases without severe symptoms, the surgical resection is controversial because there is no clear evidence that the patient has better prognosis. [9,10] The interesting fact of our case is that we have an adult patient without so severe cases of pneumonia during childhood, even absent but with several recent attacks during adulthood. Because of that, there was a debate between us if this patient needs to proceed directly to surgery or it is better to proceed in case of the next relapsing. Based on the fact that surgery

is treatment of choice and there is no sufficient scientific data to support it we decided to avoid it for now and we advised our patient to see a pulmonologist on regular basis.

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