RESPIRATORY CASE REPORTS

Maybe it is More than Pneumonia: Case Report of an Intralobar Sequestration in a 20-Year-Old Male

Pnömoni Pnömoniden de Fazlası Olabilir: Yirmi Yaşında Erkek İntralober Sekestrasyon Olgusu Sunumu

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Abstract

Pulmonary sequestration (PS) is rare congenital lung malformation typically diagnosed with fetal ultrasound or computed tomography scans. Potential complications of PS include recurrent respiratory infections, hemorrhage, heart failure, and respiratory distress. Recommended treatment is surgical resection. Presently described is case of a 20-year-old male diagnosed with intralobar PS.

Key words: Congenital Malformations, Infections, Pneumonia, Epidemiology, Pulmonary Sequestration.

Özet

Pulmoner sekestrasyonlar (PS) nadir görülen konjenital malformasyonlar olup çocuklarda fetal ultrason ve erişkinlerde bilgisayarlı tomografi ile tipik olarak tanı almaktadırlar. PS'nin potansiyel komplikasyonları tekrarlayan solunum yolu infeksiyonları, hemoraji, kalp yetmezliği ve solunumsal yetmezliklerdir. Önerilen tedavi cerrahi rezeksiyondur. Bu makalede intralober PS tanısı konmuş 20 yaşında erkek olgu tartışıldı.

Anahtar Sözcükler: Konjenital malformasyonlar, enfeksiyon, pnömoni, epidemiyoloji, pulmoner sekestrasyonterapi.

Pulmonary sequestration (PS) is rare, amounting to 0.15% to 6.4% of all congenital pulmonary malformations (1). Pulmonary sequestration involves nonfunctional, not fully developed lung tissue. Sequestration typically receives blood supply from systemic circulation, rather than the pulmonary arteries, and specifically from the thoracic aorta (74%) (1,2). Venous blood return from PS is usually from the pulmonary veins, although occasionally venous return is through systemic circulation (1,2). In addition, PS is separated from the bronchial system (3). There are 2 types of sequestration: intralobar and extralobar. Intralobar sequestration is contained within same pleural lining as the lobe in which it occurs (2). Extralobar sequestrations are contained within their own pleura (2). Intralobar sequestrations make up 75% to 85% of all PSs (4). PS is typically diagnosed with fetal ultrasound or computed tomography (CT) scans. Most common symptoms of sequestration are cough, recurrent pneumonia, and expectoration (1). Potential complications of PS include recurrent pulmonary infections, hemorrhage, respiratory distress, and heart failure (3,4). Recommended treatment option is usually surgical resection to remove the sequestration (5).

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CASE

A 20-year-old male went to his primary care physician for physical examination and complained of chest pressure and discomfort on left side. He stated that sometimes the pressure radiated down his left arm and had been ongoing for about a month. He described pressure as being constant throughout the day, with no changes due to exertion, lying down, or exercise. The patient had no history of cough or shortness of breath. He had previously been in good health and had no history of smoking. Electrocardiogram (ECG) was performed and the results were normal. His blood pressure was 110/80 mm Hg, and his pulse was 80 bpm and regular. Laboratory values were normal except for positive mycoplasma immunoglobulin M (IgM) and equivocal mycoplasma immunoglobulin G (IgG). Purified protein derivative skin test was performed and results were negative. Chest X-ray was performed. Left lung was clear; however, right lung indicated reticulonodular haziness at the base on frontal view. Impression was suspicious of developing right lower lobe infiltrate. Initial diagnosis was mycoplasma pneumonia. The patient had mycoplasma pneumonia twice previously, at 8 years of age and again at 9 years of age. He was prescribed clarithromycin 500mg twice daily with blood work to be repeated in 2 weeks. Laboratory values showed mycoplasma IgM was still positive and mycoplasma IgG was still equivocal. Symptoms were still present. Clarithromycin treatment was extended for 2 more weeks. The patient was seen for 2 follow-up appointments within the next 4 weeks with symptoms continuing and positive mycoplasma IgG and mycoplasma IgM values. Second chest X-ray was performed. Left lung remained clear. Right lung showed persistent haziness in the right medial and posterior lung base and was suspicious for pneumonia. There was also mild pulmonary hyperinflation and mild peribronchial thickening. Chest CT scan with contrast was performed. CT scan revealed superimposed infection. It also indicated right lower lobe opacity medially with apparent dilated bronchus which appeared separated from main segment bronchi and was suspicious of congenital abnormality of either lung sequestration or congenital pulmonary airway malformation. Consultation appointment was made with a pulmonologist. Pulmonologist performed physical exam and found normal lung sounds. The pulmonologist interpreted CT scan to be PS. CT angiography chest scan was performed and revealed dilated bronchus separated from the main segment bronchi (Figure 1). Findings were most consistent with intralobar PS. Arterial supply of sequestration was aberrant branch of the descending thoracic aorta (Figure 2). Venous return was from branch of the lower lobe pulmonary vein. There were small areas of glass opacity in the right lower lobe suspicious of infection. After follow-up with the pulmonologist, surgical removal of sequestration was planned. The patient was scheduled for video-assisted thoracic surgery (VATS) 4 months later. Right lower lobe wedge resection via VATS to remove PS was performed without complication. The patient was discharged from hospital the next morning and no complications or pain were reported in follow-up with the surgeon 2 weeks later. The patient no longer experienced chest pain or pressure on his left side. Follow-up with pulmonologist 6 months later revealed no complications and no chest pain or pressure since the surgery.



Figure 1: Computed tomography (CT) angiography of the chest with the arrow pointing to the intralobar sequestration



Figure 2: Computed tomography (CT) angiography of the chest with the arrow pointing to the feeding vessel of the sequestration stemming of the descending thoracic aorta

DISCUSSION

This patient had classic symptoms of PS. He had recurrent pneumonia, having mycoplasma pneumonia 3 times in his life, with latest occurrence not responding to antibiotics. PS was discovered due to constant non-identifiable chest pain and pressure that radiated down his left arm. There is case report of an 18-year-old male who had intralobar sequestration of the lower left lobe, yet experienced chest pain on his right side radiating down his right arm (2). There may be a previously unrecognized correlation between PS and chest pain. Based on location and type of pain described, it is consistent with neuropathic pain. The present patient reported not experiencing pain or pressure after surgery to remove sequestration. Therefore, it is plausible that in both of these cases, PS was affecting phrenic nerve, which led to continuous radiating pain. PS in currently described patient had typical features: It received blood supply via systemic circulation from the thoracic aorta and venous return was through the pulmonary veins. This patient's PS was intralobar, which is consistent with most PS cases (75% to 85%) (4). Intralobar sequestration is contained within the same visceral pleura of the adjacent lobe. Extralobar sequestration is contained within its own visceral pleura and is separate from the lung (2,6). Intralobar sequestration is most commonly in lower lobe of the lung, and nearly 67% are in the left lung (4). Surgical resection is recommended treatment for PS. Most common surgical techniques are either thoracotomy or VATS (7). Wedge resection may be sufficient to remove PS, but if severely infected or difficult to resect, lobectomy may be required (7,8). In this patient, VATS was recommended to minimize complications to surgery and shorten recovery time. PS in this patient was removed via wedge resection and lobectomy was not necessary.

CONCLUSION

Pulmonary sequestration is typically diagnosed with fetal ultrasound or chest CT scan. Early diagnosis is key to prevent potentially life-threatening complications. Surgical resection is currently primary treatment option to prevent complications.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - E.P.B.; Planning and Design - E.P.B.; Supervision - E.P.B.; Funding - E.P.B.; Materials - E.P.B.; Data Collection and/or Processing - E.P.B.; Analysis and/or Interpretation - E.P.B.; Literature Review - E.P.B.; Writing -E.P.B.; Critical Review - E.P.B.

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