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Intralobar Bronchopulmonary Sequestration: A Case Report and Review of the Literature

Intralober Bronkopulmoner Sekestrasyon: Bir Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT

Pulmonary sequestration is a rare malformation characterized with abnormal lung tissue with an anomalous systemic blood supply, which has no/or partial connection with the tracheobronchial tree. Here, we report the radiological diagnosis of an adult patient with intralobar type of bronchopulmonary sequestration.

Keywords: *Sequestration, Computed tomography, Lung*

ÖZET

Pulmoner sekestrasyon sistemik olarak kanlanan, trakeobronşial ağaç ile ilişkisiz veya sadece parsiyel olarak ilişkili olabilen anormal akciğer dokusunu tanımlar. Burada yetişkin hastada intralober tipte bronkopulmoner sekesterasyonun radyolojik tanısı sunulmaktadır.

Anahtar Kelimeler: *Sekestrasyon, Bilgisayarlı tomografi, Akciğer*

INTRODUCTION

Intralobar bronchopulmonary sequestration is a relatively uncommon abnormality of unknown etiology. This pathology is characterized by the presence of non-functioning lung tissue that is devoid of normal connection with the bronchopulmonary tree which is supplied by an aberrant systemic artery rather than a pulmonary artery branch (1, 2).

Two types of bronchopulmonary sequestration are usually recognized, these are; intralobar (75% of cases) and extralobar (25 %), depending on whether or not the abnormal lung tissue posses its own pleural covering. In intralobar bronchopulmonary sequestration, the tissue mass is incorporated in the otherwise normal parenchyma of the lobe and the venous drainage is usually

through the pulmonary vein. Extralobar bronchopulmonary sequestration, on the other hand, has its own pleural envelope, corresponding to a true accessory lung. Patients with this form of bronchopulmonary sequestration are most often diagnosed as newborns or in early childhood, compared to late adulthood for intralobar bronchopulmonary sequestration (2, 3). Computed tomography (CT) and magnetic resonance imaging (MRI) can provide the necessary information required for the diagnosis and pre-operative planning. Here, we report the computed tomography diagnosis of an adult patient with intralobar bronchopulmonary sequestration and briefly review the imaging findings of this entity.

CASE REPORT

A previously healthy 44 year old man who had a heavy smoking history (60 pack per year) presented for routine medical examination. On physical examination, heart sounds, rhythm and vital signs were in normal limits. Auscultation of the lung revealed vesicular respiratory sounds. Frontal chest radiograph revealed ill-defined soft tissue densities at the left basal lung area. For further evaluation, contrast enhanced chest CT scan was performed. The examination was carried out with a 40-slice multidetector computed tomography (MDCT) scanner (Brilliance 40, Philips Medical Systems, NL). The examination was performed 16 seconds after the administration of 100 ml of iodinated non-ionic contrast material at a flow rate of 3.5 ml/s, using the following scan parameters: collimation, 32 X 1.25 mm, gantry rotation time 0.4 sec, slice thickness, 4 mm, slice increment, 2 mm, with 120 Kv and 190 mAs. Data on the acquired volume were transferred to a dedicated workstation where, beside the axial images, multiplanar (MPR) reconstructions were visualized.

Axial and coronal images that were evaluated in both mediastinal and parenchymal window settings showed; bilateral emphysematous changes mostly evident on the upper lobes. At the mediobasal segment of the left lower lobe, there were nodular structures, some of them containing dilated bronchi with air-fluid levels (Figure 1). On the coronal image these structures had finger-like projections towards the sorrounding normal lung parenchyma (Figure 2). There were lucent areas corresponding to air trapping sorrounding these nodular lesions (Figure 3).

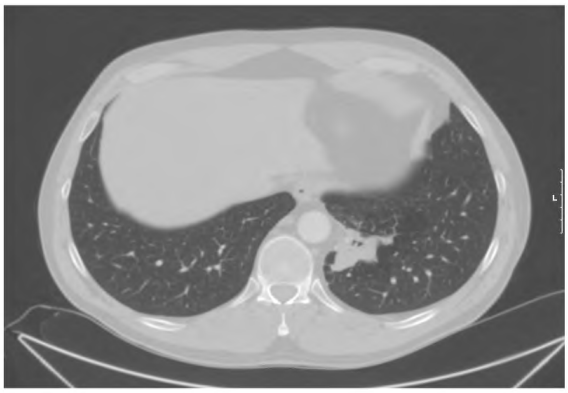

Figure 1: *Axial CT image shows nodular lesions and accompanying dilated bronchi with air-fluid levels at the mediobasal segment of the left lobe.*

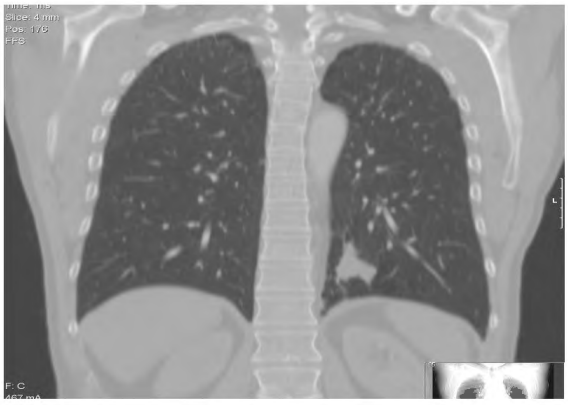

Figure 2: *Coronal MPR image shows that these nodular structures have finger-like projections towards the lung parenchyma.*

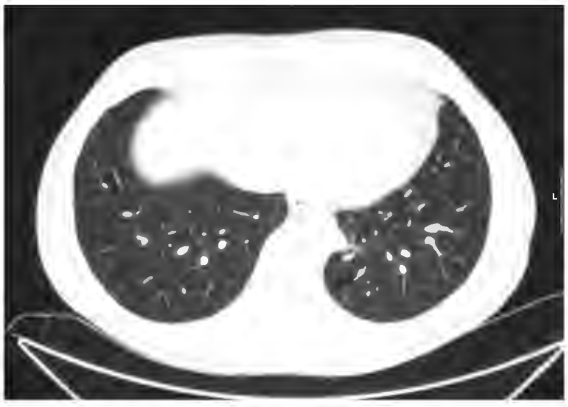

Figure 3: *On the axial CT image lucent areas corresponding to air trap are noted around the abnormal lung.*

At the level of the distal thoracic aorta on the left side, three anomalous arteries were seen emanating from the aorta supplying the abnormal lung (Figure 4). The presumptive diagnosis was intralobar bronchopulmonary sequestration.

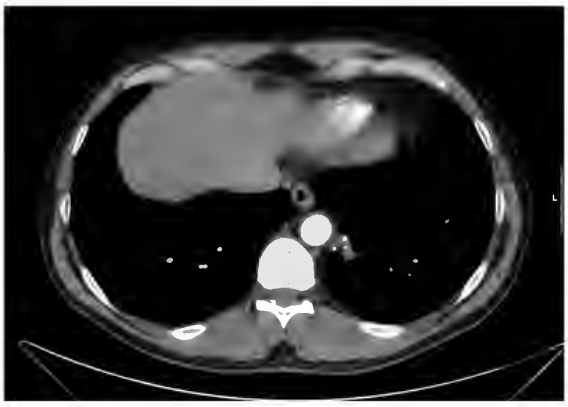

Figure 4: *At the level of the distal thoracic aorta on the left side, three feeding arteries can be seen originating from the aorta supplying the abnormal lung tissue.*

DISCUSSION

Pulmonary sequestration (PS) is a rare anomaly characterized with the presence of abnormal lung tissue supplied by systemic circulation, lacking the normal connection with the tracheobronchial tree. This anomaly was first described by Rokitansky and Rektorzik in 1861 and it accounts for approximately 0.15- 6.4 % of all congenital malformations of the lung (1, 2, 3).

Regardless of the type, the bronchopulmonary sequesterations are supplied from the systemic circulation, via one or more aberrant arteries. The most frequent site of origin of these aberrant arteries is thoracic aorta and aortic arch. Rarely, they may arise from the abdominal aorta, renal arteries or celiac trunk (3, 4).

Most of the patients with PS remain asymptomatic and hence the diagnosis is frequently incidental, as in our patient. This is especially true for extralobar sequesterations. In intralobar type, signs and symptoms can appear at any time of life, most commonly in the second to third decades. Chronic cough, infection and hemoptysis are the most common symptoms, but hemothorax and congestive heart failure due to shunting may also occur (2-4).

In addition to the differences in the age of presentation and symptomatology, location is another criterion to differentiate between intra and extralobar PS. Intralobar sequestrations almost always occur within the lower lobes and are seen slightly more often in the left than in the right lung (5, 6). On the other hand extralobar type tends to be located above, below or within the diaphragm (3, 5, 6).

Extralobar bronchopulmonary sequesteration is believed to be a congenital anomaly and it is often associated with other congenital pathologies such as, gastrointestinal tract anomalies, congenital cardiac disesases and adenomatoid malformation. On the other hand the etiology and pathogenesis of the intralobar type is not clear. Although some authors suggest this type being congenital, it is more likely to be acquired as a result of postinflammatory degenerative changes (4 - 6).

At radiography, intralobar sequestrations can manifest as an area of increased

opacity simulating pneumonia, as a mass with or without air fluid levels, or as cysts. CT can demonstrate a parenchymal lesion, which manifests as a homogeneous mass of soft-tissue attenuation, emphysematous change, or a cystic area (7, 8). The blinded bronchi at the area of sequestration may become distended and trapped with mucous which can give rise to mucous plugs and mucocele formation. CT reveals parenchymal lesions and demonstrates the anomalous systemic artery in up to 80 % cases after contrast material administration. The anomalous systemic artery is sometimes not visualized on spiral CT scans typically when the feeding vessel is single and has a small diameter. The diagnosis can be confirmed by means of CT angiography, which shows the systemic arterial supply. An important key to the diagnosis of bronchopulmonary sequestration as in our case is the finding of an anomalous systemic arterial supply. It should also be kept in mind that although intralobar sequestrations lack normal communications with the tracheobronchial tree, they can be ventilated by collateral air drift or through fistulous bronchial communications that may develop after an episode of infection (5-8).

MRI is a good diagnostic alternative when CT angiography is contraindicated in case of known allergy to iodinated contrast agents or renal insufficiency. In MRI, the abnormal lung is seen hyperintense on both T1 and T2 weighted sequences. Postgadolinium images can demonstrate the systemic arterial supply and also the venous drainage (2, 6, 8).

Diagnosis of intralobar bronchopulmonary sequestration should be kept in mind in a young adult with recurrent lower lobe infections and persistent lower lobe abnormalities on chest radiographs. CT angiography is the diagnostic method of choice not only for the diagnosis, but also for preoperative planning.

REFERENCES

1)Non-invasive diagnosis in a case of bronchopulmonary sequestration and proposal of diagnostic algorithm. Caradonna P, Bellia M, Cannizzaro F, Regio S, Midiri M, Bellia V. Monaldi Arch Chest Dis. 2008; 69: 137-41.

2)Pulmonary sequestration--a review of 8 cases treated with lobectomy.Pikwer A, Gyllstedt E, Lillo-Gil R, Jönsson P, Gudbjartsson T.Scand J Surg. 2006; 95: 190-4.

3)Intralobar pulmonary sequestration associated with an aneurysmal aberrant aortic branch. Schena S, Crabtree TD, Zoole JB, Patterson GA. J Thorac Cardiovasc Surg. 2007; 134: 535-6. No abstract.

4)Intralobar bronchopulmonary sequestration. Momeni A, Hood MN, Carter WR, Ho VB. Mil Med. 2007; 172: viii-ix. No abstract.

5)Bronchopulmonary sequestration: CT assessment. Ikezoe J, Murayama S, Godwin JD, Done SL, Verschakelen JA.Radiology. 1990; 176: 375-9.

6)Intralobar sequestration: a rare cause of severe hemothorax.Wandschneider W, Illiasch H. J Thorac Cardiovasc Surg. 2003; 126: 872-3. No abstract.

7)Systemic arterial supply to the lungs in adults: spiral CT findings.Do KH, Goo JM, Im JG, Kim KW, Chung JW, Park JH.Radiographics. 2001; 21: 387-402.

8)Developmental lung anomalies in the adult: radiologic-pathologic correlation. Zylak CJ, Eyler WR, Spizarny DL, Stone CH.Radiographics. 2002; 22 Spec No: S25-43.