Olgu Sunumu / *Case Report*

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A Case of Intrapulmonary Sequestration Seeming as An Intrathoracic Mass

İntratorasik Kitle Görüntüsü Veren İntrapulmoner Sekestrasyon Olgusu

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Abstract

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As a rare congenital thoracic malformation, pulmonary sequestration is composed of nonfunctioning primitive lung tissue. It has anomalous systemic supply rather than the pulmonary circulation and no connection with the tracheobronchial tree. It may present as respiratory infection or asymptomatic mass on chest imaging studies. Surgical removal is the treatment of choice. This paper presents a case of intralobar pulmonary sequestration that presented during childhood.

***Keywords*:** sequestration, pulmonary, lung

Özet

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Nadir bir konjenital toraks malformasyonu olan pulmoner sekestrasyon, işlevsiz pirimitif akciğer dokusudur. Pulmoner dolaşımdan farklı sistemik dolaşımdan beslenmesi olup trakeobronşial sistemle ilişkisi yoktur. Solunum enfeksiyonları olarak görülebilir veya radyolojik çalışmalarında asemptomatik kitle olarak rastlanabilir. Tedavisi cerrahi rezeksiyondur. Bu yazıda çocukluk döneminde görülen intra lober pulmoner sekestrasyon olgusunu sunuyoruz.

Anahtar Kelimeler: *sekestrasyon, pulmoner, akciğer*

Intorduction:

Bronchopulmonary sequestrations are cystic or solid masses and characterized by the abscence of communication with the tracheobronchial system and supplied by an anomalous systemic artery[1]. Pulmonary sequestration represents approximately 6% of all congenital pulmonary malformations. One part of the lung develops abnormally during embryogenesis. Two types of sequestration are associated with different clinical features regarding to whether it has independent pleura[2]. The intralobar sequestration is located within the visceral pleura of the adjacent lung, while the extralobar type is contained within its own visceral pleura, separate from the remaining lung. The common aspect is discontinuity with the bronchial tree and pulmonary arterial vasculature. Medical treatment is limited by the present infection. Surgical resection provides definitive management[3].

Case Report

A 10-year-old boy presented to the our pediatric clinic complaining of one and half months of respiratory distress and intermittent abdominal and chest pain. Physical examination revealed a healthy-appearing however auscultation revealed no breath sonuds over the left basal area. A chest radiograph revealed an opacity in his left lower lung field (Figure 1) and ultrasonography confirmed this lesion as heterogen structure. The septated mass of 137 mm x 92 mm involving left lower lobe was seen on his chest computerised tomography. CT (computed tomography) also showed the lesion with its artery from aorta in detail (Figure 2,3). His blood tests were within normal ranges except leucocytosis (WBC 17700) . The patient was evaluated and considered for surgery. At thoracotomy, the left lower lobe was seen to be supplied by abdominal aorta and fullfilled with loculated infected material. Its arterial branch from the aorta was carefully isolated and ligated and the lesion was removed without complication. Left lower lobectomy was performed (Figure 4). Pathology of the resected specimen showed inflammatory lung parenchyma, with cystic spaces of up to 4 cm. The cysts had smooth walls and contained a viscous, brown, opaque, mucus-like material. A small artery was noted along the pleural surface. The patient made an uneventful recovery and he is at the follows.

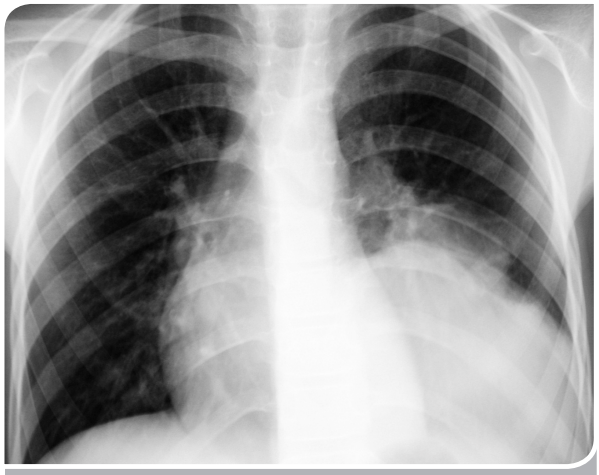

Figure 1: Chest x-ray of the patient

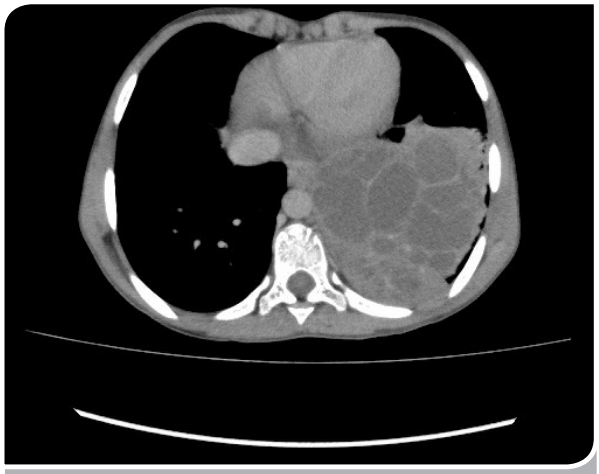

Figure 2: A CT scan of the sequestration

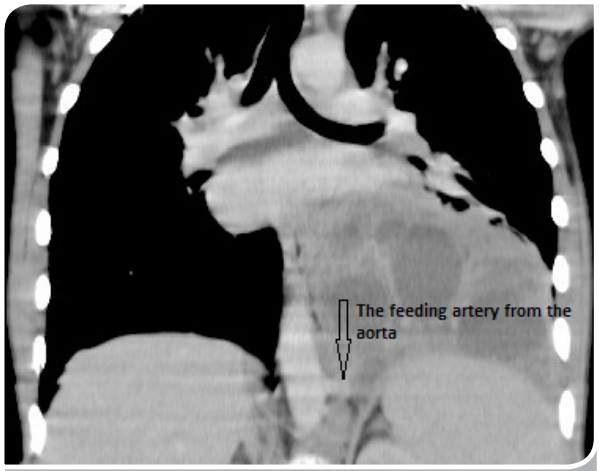

Figure 3: Coronal view of the patient showing the aorta and the feeding artery

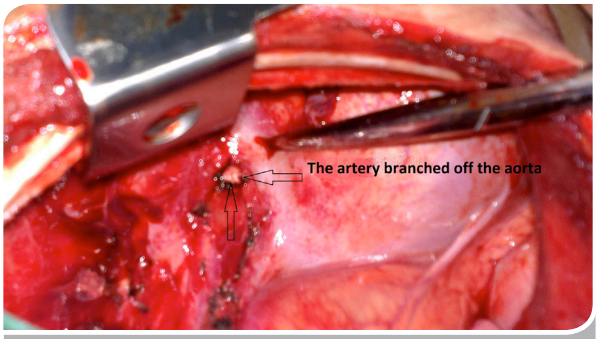

Figure 4: Peropertative Picture showing the feding artery of the sequestration

Discussion

Bronchopulmonary sequestration means a part of lung parenchyma that is separated from the tracheobronchial tree of the lung tissue.1 Its blood supply is usually from an aberrant artery arising from the aorta or one of its branches. sequestration is believed to result from abnormal diverticulation of foregut and aberrant lung buds. It is characterised by an aberrant vessel. This rare congenital abnormality represents less than 2 % of in general population[4]. It is classified into 2 different forms regarding early or later maldevelopment of the bud: intralobar, as in our patient, and extralobar, according to whether it has independent pleura. Intralobar forms comprise 75 % of sequestrations while the extralobar type is found in the remaining 25 % of cases[5,6] The most common location is in the posterior basal segment, and nearly two thirds of pulmonary sequestrations appear in the left lung[7].

Clinical symptoms vary from breathing difficulties to an asymptomatic mass found incidentally on chest x- rays. They are associated with the sequesration type. Intralobar sequestration usually presents during childhood in contrast the extralobar form is a disease confined to neonates because of the associated congenital abnormalities. The majority of intralobar sequestrations are asymptomatic and carry the abnormality for years, only to be diagnosed during a routine chest radiograph for unrelated symptoms. Symptoms of sequestration include include chest pain, pleuritic pain, shortness of breath, and wheezing; however, signs of recurrent infection, such as fever and productive cough, are common. Sequestrations are not completely isolated from the native lung despite no communication with bronchial tree. Therefore, an infection in the sequestration can invade normal bronchial system. It can even present with frank hemoptysis. To ensure a correct diagnosis, the radiographic imaging of sequestration has to characterize the lesion in detail for excluding alternative pathology, and to clarify the aberrant arterial supply of the sequestration to facilitate operative management (8). For this purpose, plain chest radiograph is usually nonspecific and incompetent. Chest CT usually shows a discrete mass in the lower lobe, with, as in our case, or without cystic changes. Lesions present in lobes other than the lower lobe should remind investigation of differential diagnoses, sequestration may cause emphysematous changes in the adjacent lung tissue due to air trapping. Pre-operative identification of the aberrant arterial supply is important in preventing operative morbidity. Spiral CT and magnetic resonance angiography (MRA) are the imaging method of choice for identifying the aberrant artery and the venous return[8,9]. The arterial supply of intralobar sequestration is a branch off of the thoracic aorta in up to 75% of patients. Rarely, however, the arterial supply can come from branches of the aorta, such as the subclavian artery or intercostal arteries. Venous drainage is commonly provided to the pulmonary veins[10]. In our case located in the left lung basal area, the feding artery was branched off the thoracic aorta and venous return into the pulmonary vein. The condition was consistent with the intrapulmonary type of sequestration. The risk of vascular complications is greatly reduced with this procedure before surgery.

Surgical removal is the treatment of choice for intrapulmonary sequestration, but the timing of surgery is in question[3]. Careful preoperative evaluation can prevent surgical complications[11]. Patients with symptoms should be referred to surgery for definitive treatment. Ongoing infections can trouble operative success. Surgery usually involves lobar resection via standard thoracotomy because periphery of the sequestration can not be clearly defined. Thorascopic surgery has been described for some cases. Close attention to division and ligation of the artery prevents bleeding problems. Close monitoring for symptoms should be essential if the surgery is cancelled. Complications of watchful waiting include recurrent pneumonia, hemoptysis, and chest pain. As an alternative treatment to surgery, angiographic embolization of the aberrant feeding artery can be used in patients with high operative risk.

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