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A case of lobar agenesis recognized following tracheobronchial foreign body aspiration

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Özet

Trakeobronşial yabancı cisim aspirasyonu sonrası tanı konan bir lober agenezi olgusu

Sol üst lober agenezi saptadığımız üç yaşında bir kız olgu sunulmaktadır. Lober agenezi olgularında eşlik eden anomali olasılığı yüksekse de, bizim olgumuzda hiçbir ek anomali saptanmamıştır. Bu olguda akciğer perfüzyon sintigrafisi ve göğüs bilgisayarlı tomografi incelemeleri, ayırıcı tanı için yeterli görülmüştür.

Anahtar kelimeler: *Akciğer, anomaliler*

Summary

A case of lobar agenesis of the left upper lobe of the lung in a 3 year-old girl is presented. Although, this anomaly is usually associated with other congenital anomalies, our patient had no associated anomaly. Pulmonary perfusion scintiscan and chest computed tomography examinations were found to be useful diagnostic modalities in our case.

Key words: *Lung, abnormalities*

Introduction

Developmental anomalies of the lung are seldom reported. Unilateral pulmonary agenesis is seen once in 10.000 to 15.000 autopsies and lobar agenesis is more rare. Agenesis and aplasia of the lung are usually detected after birth due to early onset of symptoms. Conversely, lung hypoplasia and lobar agenesis may remain clinically silent for a long time [4].

Tracheabronchial foreign body aspiration is one of the most important emergency procedure with its risks and high complication rate displaying its highest incidence in children younger than 3 years [7].

Here, we present a case with agenesis of the left upper lobe of the lung without any associated congenital anomaly, diagnosed accidentally in early childhood following tracheabronchial foreign body aspiration. We would like to emphasize how the symptoms and radiological findings of these two conditions can be confusing and also, the importance of selecting the proper diagnostic techniques which will help to determine the route of therapy in small children.

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Case Report

A 3 year-old girl was admitted to the hospital with the history of cherry aspiration. The patient was born by vaginal delivery following an uncomplicated term gestation without history of polyhidramniosis. No remarkable symptom of pulmonary disease was noted before this aspiration. At admission, the patient's general condition was not good with the moderate cyanotic appearance, dyspnea and tachypnea. In physical examination, hyperressonance was percutated on the right hemithorax.

There was a localized wheezing on the left hemithorax where the breath sounds were absent. Routine laboratory studies were normal. Chest x-ray did not reveal any radiolucent foreign body but revealed density on the upper left hemithorax and a mediastinal shift away from right hemithorax to the left. The ribs were closed at the upper left he-

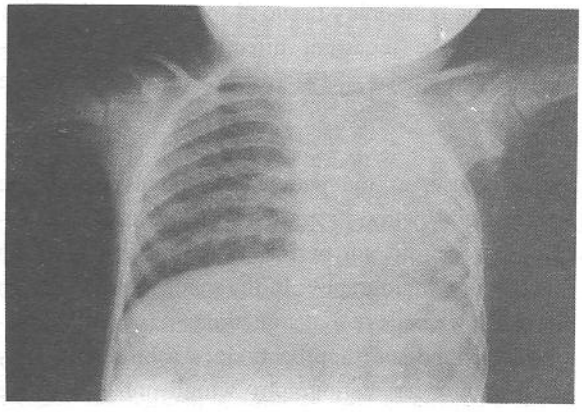

Figure 1. The chest x-ray of the patient showing the shift of the heart and mediastinum to the left. The left hemithorax is smaller than the right with the crowding of the upper chest.

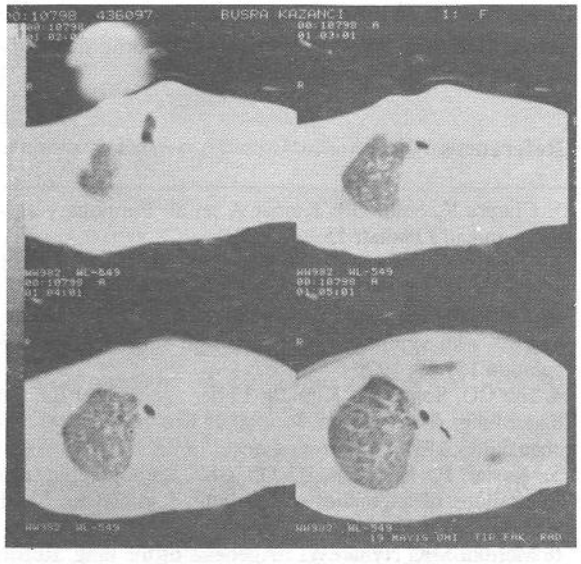

Figure 2. Chest computed tomography examination of the patient after bronchoscopy showing no pulmonary tissue in the left upper hemithorax.

mithorax in addition to a decrease of hemithoracal volume of the same side (Figure 1).

With these findings, patient was taken to the emergency operation room for rigid bronchoscopy under general anesthesia. Some of the cherry pieces were removed from trachea. In contrast with radiological and clinical findings, no foreign body was found in the lower bronchial tree. Although, the pulmonary symptoms of the patient improved after bronchoscopy, there was no difference in the control chest x-ray film compared with the initial x-ray. With the computed tomography (CT) examination of the thorax, left upper lobar agenesis was suspected (Figure

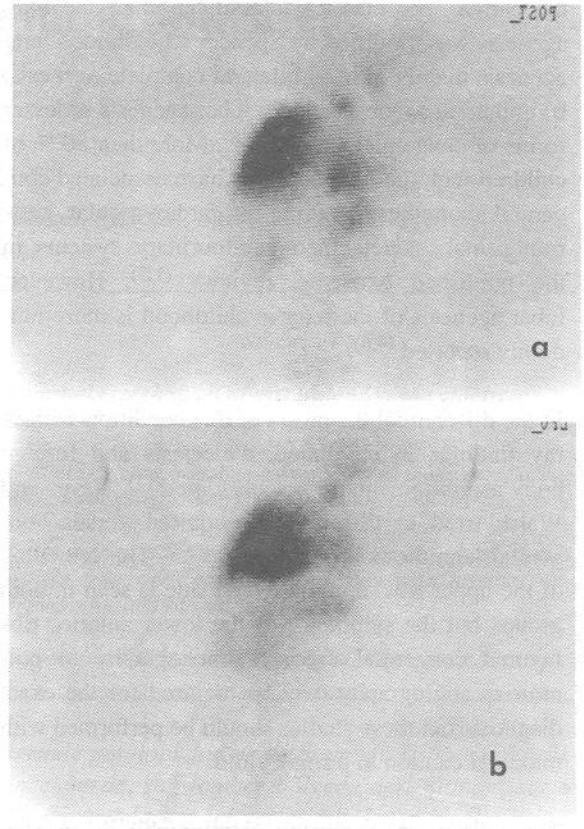

Figure 3. Pulmonary perfusion scintigraphy of the patient after bronchoscopy showing no perfusion in the left upper hemithorax. a) oblique view, b) posterior view.

2). The pulmonary perfusion scintigraphy performed later, using 3 m Ci Tc-99m macroagregated albumin revealed no perfusion in the left upper lobe (Figure 3a,b). When all these findings were taken into consideration, the diagnosis of lobar agenesis was confirmed.

Other diagnostic studies which were performed in order to evaluate the organ systems did not reveal any other associated congenital malformation in this child. By the end of second week, the patient was discharged without any symptom following a regimen of antibiotherapy and expectorant. With the follow up of 18 months, the child is asymptomatic without any medication.

Discussion

Agenesis of the lung refers to the total absence of the pulmonary parenchyma as well as the vasculature and bronchial components of the lung distal to

the carina. The classical classification of the lung agenesis was modified by Spencer who divided lung agenesis mainly into: **a)** bilateral complete agenesis, **b)** unilateral agenesis and, **c)** lobar agenesis or lesser forms of congenital anomaly [8]. More than 50 % of children with the lung agenesis have associated congenital anomalies involving the cardiovascular, gastrointestinal, skeletal and genitourinary systems in the published extensive reviews [1,5]. However, lobar agenesis of the lung in childhood is more randomly reported [4,6].

In the differential diagnosis of this condition with x-ray findings in childhood, atelectasis and foreign body aspiration should be considered [5]. Daves and Walsh tried to differentiate acquired versus congenital hemithoracic volume loss [2]. The crowding of the upper ribs on the affected side is seen in both groups but the symmetry of the lower anterior ribs favored congenital cause. Bronchography or pulmonary angiography may be required for the exact diagnosis but these studies should be performed with maximal caution in a small child.

The pulmonary perfusion scintigraphy is an available method for the diagnosis and limits the differential diagnosis in patients with the similar findings of chest x-ray to three disorders: pulmonary artery agenesis, pulmonary artery hypoplasia or pulmonary embolism [3]. With the addition of a CT examination or a pulmonary ventilation study, the pulmonary embolism can be excluded, as there would be a lung tissue on the affected side.

Pulmonary agenesis and aplasia are very similar disorders for practical and developmental purposes, as there is no pulmonary tissue or ipsilateral pulmonary artery. If the recurrent infections are noted particularly in aplasia, the source of infection should be removed.

In this case, as the patient was an asymptomatic small child without any history of recurrent infections, we did not perform a bronchography or a pulmonary angiography. In the management of such patients, we believe that sometimes it is a better choice "wait and see" approach since a bronchography or a pulmonary angiography would not change the route of therapy. On the other hand, performing a pulmonary perfusion scintigraphy which is a completely noninvasive method available everytime will lead us to differentiate the radiological x-ray findings of foreign body aspiration and some developmental anomalies of the lung.

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