Congenital Bronchial Atresia Presenting as a Cavitary Lesion—A Case Report

—Case Report—

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Abstract

Bronchial atresia is a rare congenital anomaly usually identified as an incidental finding during routine examinations in adulthood. We report a case in which routine chest radiograph revealed bronchial atresia with a cavitary lesion surrounded by hyperinflated lung tissue and few vascular markings. Chest computed tomography (CT) revealed a cavitary lesion with air-fluid level and segmental emphysematous changes. Bronchoscopy revealed a very small orifice at the beginning of the bronchus.

The initial chest radiograph findings of bronchial atresia can mimic those of many pulmonary diseases. The diagnosis of congenital bronchial atresia can be confirmed on the basis of chest CT findings. Bronchoscopy can be helpful in excluding the possibility of other pulmonary diseases in doubtful cases.

Key words: Bronchial atresia, cavitary lesion, computed tomography

Introduction

Bronchial atresia was first reported by Ramsay and Byron in 1953 [1]. It is an uncommon anomaly caused by focal obliteration of the bronchial lumen and the absence of communication between a lobar, segmental or subsegmental bronchus and the central airway [2-4]. Stenosis results in the collection of mucus in the bronchi and formation of a bronchocele. In most patients with bronchial atresia, the anomaly is detected as an incidental finding during routine examinations. Bronchial atresia is associated with various abnormalities, including pulmonary sequestration, congenital adenomatoid malformation, congenital lobar emphysema, bronchogenic cyst, pericardial defects, and aplastic lungs [2-4]. Herein, we report a case of bronchial atresia with a tubular mass, which was diagnosed by chest radiography. The chest computed tomography (CT) and bronchoscopy findings confirmed this diagnosis.

Case report

A 17-year-old non-smoking woman underwent routine chest radiography during a health check-up. Her chest radiograph revealed a cavitary-like lesion surrounded by hyperinflated lung tissue with a few vascular markings on the left upper lobe (Figure 1). She was symptom-free. The results of all laboratory tests and physical examinations were normal.

A chest CT scan revealed a thin-walled cavitary lesion with air-fluid level and emphysematous changes in the anterior segment of the left upper lobe. These findings were consistent with those for mucocele. No other pulmonary masses were visible on the CT image (Figure 2). Bronchoscopy revealed a very small orifice at the beginning of the anterior segmental bronchus of the left upper lobe, which possibly corresponded to the “atretic” anterior segment (Figure 3). The results of microbiologic and cytologic examinations of the washed specimens from the cavitary lesion were negative.

The patient was diagnosed with congenital bronchial atresia and received no further intervention. Follow-up examination 1 year later revealed that she was asymptomatic, and no changes were observed in the chest CT scan.

Discussion

Bronchial atresia is a pulmonary anomaly of unknown cause in which a segmental bronchus does not communicate with the central airways. Although a pathological report is not available for our patient, the combination of mucocele, bronchial occlusion and local emphysematous changes supports our diagnosis of bronchial atresia, on the basis of Matsushima's criteria [5].

There are 2 possible theories about the pathogenesis of bronchial atresia. First, the separated primitive bronchial bud may have continued to develop, but lost connection with the central airway. The second proposal has attributed its cause to a local vascular insult which resulted in failure of canalization of the bronchial buds [9].

Bronchial atresia is characterized by a branching mass or mucocele formed by the mucus-filled dilated bronchi distal to the atretic segment. The lung hyperinflation is caused by collateral ventilation from the adjacent normal lung via a check valve mechanism; this ventilation occurs through the pores of Kohn and the channels of Lambert [3, 9]. Bronchial atresia usually involves a segmental bronchus and is most commonly found in the posterior apical segment of the left upper lobe [6-9]. This condition is usually diagnosed in the second or third decade of life and exhibits male predominance, with an estimated prevalence of 1.2 cases per 100,000 males [10]. About half to two-thirds of the reported patients were asymptomatic before diagnosis.

Recurrent pneumonia, dyspnea, cough, or hemoptysis has been reported in some cases [10].

Congenital bronchial atresia is a rare and benign condition. Initial radiographic examination of patients with this condition may occasionally reveal findings that resemble those of serious underlying pulmonary diseases. The criteria proposed by Matsushima et al. (mucocele, bronchial occlusion and local emphysema) may serve as an objective basis for its diagnosis. [5]

以開洞性病灶來表現的支氣管閉鎖症-病例報告

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支氣管閉鎖症通常是意外發現的肺部結構異常, 它在主要氣道和細支氣管間失去了交通性。影像學上的表現常可和其他疾病混淆。我們提出一位年輕女性, 在常規的胸部放射線檢查中發現管狀的腫塊, 電腦斷層表現為開洞性的病灶伴隨周邊的肺氣腫變化。支氣管鏡檢查顯示在左上葉的前分支開口處狹窄。細胞學和細菌培養皆為陰性。電腦斷層是最佳的檢查利器, 而支氣管鏡檢查可協助排除其他疾病。

關鍵字: 支氣管閉鎖症

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Figure legends

Figure 1. Chest radiograph revealed a tubular mass surrounded by hyperinflated lung tissue with a few vascular markings on the left upper lung field.

Figure 2. Chest computed tomography scan showed a cavitary lesion with an air-fluid level and emphysematous changes in the anterior segment of the left upper lobe.

Figure 3. Bronchoscopy revealed a very small orifice at the beginning of the anterior segmental bronchus of the left upper lobe.

References

1. Ramsey BH, Byron FX. Mycocele, congenital bronchiectasis and bronchogenic cyst. J Thorac Surg 1953; 26: 21-9.
2. Jederlinic PJ, Sicilian LS, Baigelman W, *et al.* Congenital bronchial atresia. A report of 4 cases and a review of the literature. Medicine (Baltimore) 1987; 66: 73-83.
3. Bailey PV, Tracy T Jr, Connors RH, *et al*. Congenital bronchopulmonary malformations. Diagnostic and therapeutic considerations. J Thorac Cardiovasc Surg 1990; 99: 597-602.
4. Kuhn C, Kuhn JP. Coexistence of bronchial atresia and bronchogenic cyst: Diagnostic criteria and embryologic considerations. Pediatric Radiology 1992; 22: 568-70.
5. Matsushima H, Takayanagi N, Satoh M, *et al.* Congenital bronchial atresia: Radiologic findings in nine patients. J Comput Assist Tomogr 2002; 26: 860-4
6. Cohen AM, Solomon EH, Alfidi RJ. Computed tomography in bronchial atresia. AJR Am J Roentgenol 1980; 135: 1097-9.
7. Rappaport DC, Herman SJ, Weisbrod GL. Congenital bronchopulmonary diseases in adults: CT findings. AJR Am J Roentgenol 1994; 162: 1295-9.
8. Kinsella D, Sissons G, Williams MP. The radiological imaging of bronchial

atresia. Br J Radiol 1992; 65: 681-5.

9. Shady K, Siegel MJ, Glazer HS. CT of focal pulmonary masses in childhood. Radiographics 1992; 12:505-14.

10. Psathakis K, Lachanis S, Kotoulas C, *et al*. The prevalence of congenital bronchial atresia in males. Monaldi Arch Chest Dis 2004; 61: 28-34.