Case Report

Asymptomatic Atresia of the Lobar Bronchus of the Lung: A Case Report

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A 64-year old woman presented with an asymptomatic occlusion of the intermediate bronchus associated with a peripheral mass occupying the entire middle and lower lobes. As malignancy was suspected, inferior bilobectomy was done. There was a complete atelectasis of both lobes, with massive parenchymal necrosis. Pathological examinations suggested a tuberculous granuloma in the bronchus and parenchyma although tuberculous bacilli were not found. This case was unusual as congenital anomaly, and was suspected as bronchial tuberculosis. (Ann Thorac Cardiovasc Surg 2001; 7: 301–3)

Key words: bronchial atresia, tuberculosis, aberrant vessel

Introduction

Congenital bronchial atresia is a rare disease first described in 1953 by Ramsay and Byron.[1)] The etiology is unknown. The segmental or lobar bronchus fails to construct normally, hence this malformation leads to accumulation of mucus within the distal bronchi and lung, or hyperinflation of the obstructed segment. This entity is usually asymptomatic but symptoms such as cough, mucinous sputum, recurrent chest infections, and dyspnea may be present. Secondary bronchial atresia can occur with the presence of a mucosal flap, hypertrophy of the bronchial mucosa, bronchial kinking secondary to herniation, or external compression of bronchi by vascular formation.[2)]

We treated a 64-year old woman with lober bronchial atresia of the right intermediate bronchus and a peripheral mass which was increasing in size with unknown etiology.

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Case Report

In a 64-year-old Japanese woman weighting 43.5 kg and 1.5 m tall a chest roentgenogram taken during a health examination in May 2000, revealed an abnormal shadow in the right lower lung field (Fig. 1). At the time she presented at Kyushu University Hospital in Fukuoka, respiratory symptoms were absent and pulmonary function was normal. Serum chemistry and blood counts were normal and the woman had no loss of weight, general fatigue or a childhood history of respiratory symptoms or disease, including tuberculosis. A repeat chest roentgenogram showed a large shadow in the right lower lung field, and computed tomography (CT) revealed a cystic mass, including necrotic material which occupied the entire middle and lower lobes (Fig. 2). Segmental pulmonary arteries and veins seemed to normally branch. On bronchoscopy the intermediate bronchus appeared blind. Biopsy of the blind bronchus wall revealed normal epithelial tissue and a mild chronic inflammation.

When compared with a chest roentgenogram taken at another hospital in 1996, the cystic mass had increased in size and malignancy was suspected. Surgical excision of the mass was done on July 3, 2000. At thoracotomy, the right middle and lower lobes were in a state of complete atelectasis and we suspected collection of mucus in the lungs. On dissecting the hilum and interlober aspect, we found no aberrant vessels

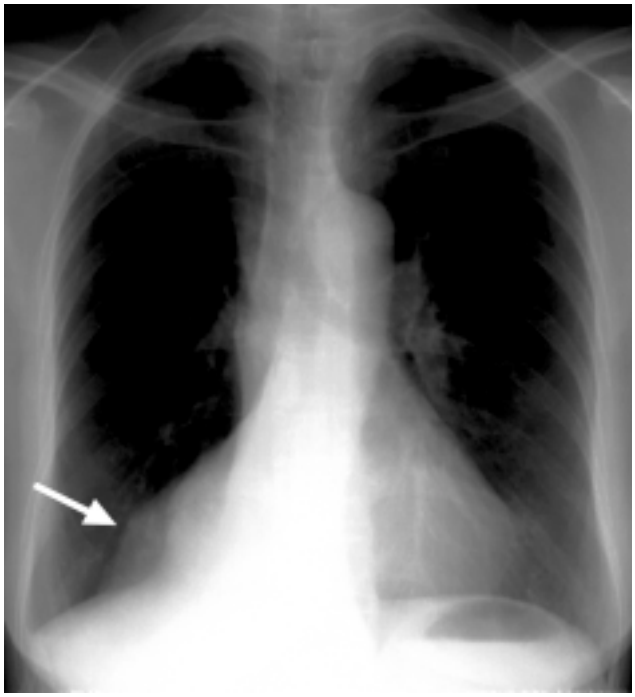

Fig. 1 Chest roentgenogram. Chest roentgenogram on May 2, 2000, showed an abnormal shadow in the right lower lung field (arrow).

between the lung and mediastinum, and the branches of the segmental pulmonary arteries were normal (A4, 5, 6, 7+8, and 9+10) as were pulmonary veins (V4+5 and the inferior pulmonary vein). The bronchial wall outside the atresia had no cartilage (Fig. 3). A middle and lower bilobectomy was done, while, bronchial resection was done at a distal site of the atresia since a sleeve resection would be required to resect the atresic portion and fibrous adhesions between the pulmonary artery and bronchus were present.

The cystic mass in the middle and lower lobes measured 5×4×4 cm and 11×10×7 cm in diameter, respectively. Odorless, viscous, and gray-yellow necrotic material filled the mass. Bacterial cultivation of the mass revealed no bacilli or fungus. Histologically, there was a massive necrosis of the lung parenchyma with epithelial granuloma of the bronchial wall. Tuberculosis was suspected, however acidphilic bacilli were absent with Ziehl Neelsen or PAS staining of the tissues. There were no malignant cells (Fig. 4). DNA of mycobacterium tuberculosis was not evident, as determined by polymerase chain reaction of paraffin fixed sections.

The patient made an uneventful recovery and was discharged on the 18th postoperative day.

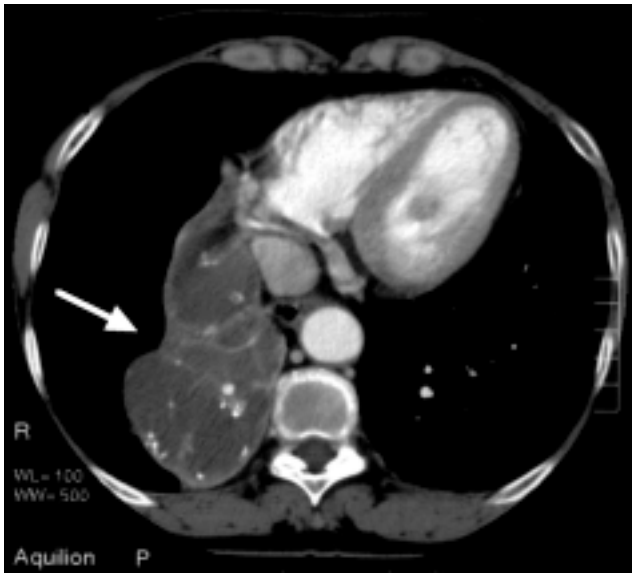

Fig. 2 Chest CT. Chest CT scan revealed a mass shadow with fluid impaction in the right middle and lower lobe (arrow).

Discussion

Bronchial atresia is a rare congenital disease usually characterized by a segmental or lobar emphysema associated with or without mucoid impaction.[1,3] The etiology is estimated that at the time that bronchi are formed in the 5th from 16th week of gestation, a vascular insult may occur leading to necrosis of bronchial tissues.[4] Raynor and colleagues reported cases of secondary bronchial atresia caused by the presence of a mucosal flap, hypertrophy of bronchial mucosa, bronchial kinking secondary to herniation, or external compression of bronchi by anomaly vascular formation such as patent ductus arteriosus and aneurysm of a pulmonary vein.[2] Congenital bronchial atresia is usually discovered in neonates or in middle aged persons, but usually in young patients whereas secondary bronchial atresia seems to be frequent in middle or older age persons. The typical chest roentgenographic shows hyperlucency and a hilar or extrahilar mass, in case of both congenital and acquired diseases. Sometimes, an air fluid level is recognized in the bronchocoele.[5] Computed tomography (CT) shows branching mucus plugs or a mucoid impacted mass and a hyperinflated lobe.

The present case was unusual in terms of the central location of the atresia (intermediate bronchus), and massive necrotic change in the entire middle and lower lobes. At the first presentation, roentogenological and CT

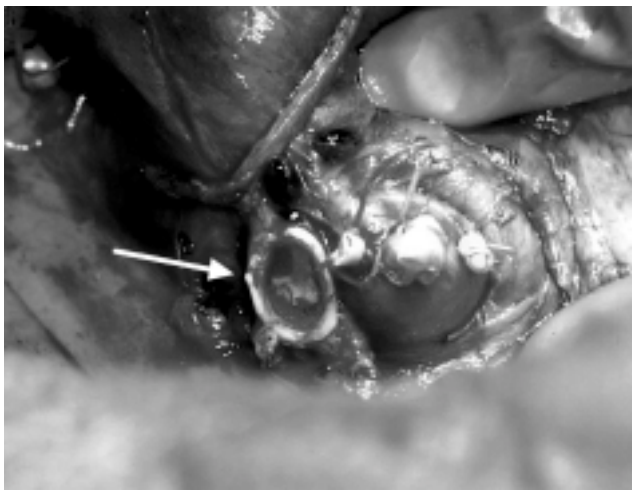

Fig. 3 Operative finding. Macroscopic findings of bronchial atresia in the intermediate bronchus (arrow).

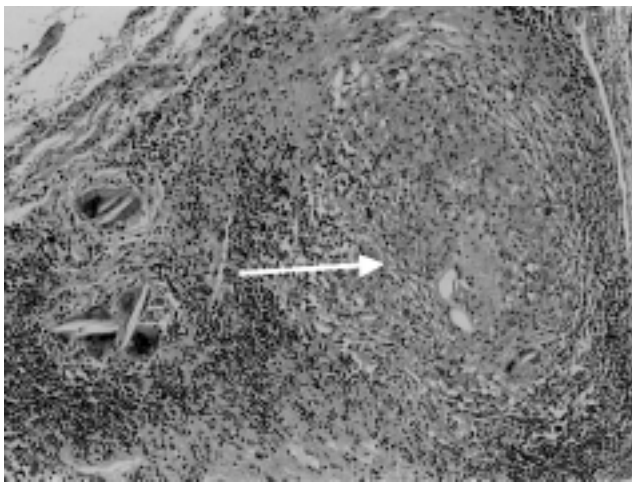

Fig. 4. Microscopic finding. Microscopic finding of the resected mass in the lung. Section showed many foci of epithelial granuloma (arrow) with massive necrosis, partly involving the bronchial wall. Tuberculosis was considered, but mycobacterium tuberculosis was not detected using Ziehl-Neelsen stains and there was no evidence of malignant cells.

imagings of the mass in the right thorax suggested extralobar pulmonary sequestration, however, magnetic resonance angiography revealed no aberrant artery or vein around the mass. Histological examination of the resected specimen suggested a tuberculous infectious feature such as a number of foci of epithelioid granuloma with massive necrosis in the parenchyma and bronchial. However, tuberculous germs were not detected by Ziehl-Neelsen stains, or DNA of mycobacterium tuberculosis was not evident in the polymerase chain reaction. Our patient had no complaints and had been healthy for over 6 decades of life, however, her mother and elder sister had had pulmonary tuberculosis when the patient was a child. We speculated that the cause of the bronchial atresia and the peripheral lung lesion were related to tuberculosis.

Treatment of asymptomatic bronchial atresia is usually conservative with regular chest roentgenogram during follow up. When the patient has definite or recurrent symptoms with the possibility of severe infection, surgical treatment is indicated. Our patient had a large mass without aeration, unusual findings as a morphological change in bronchial atresia. Therefore, surgical resection was deemed necessary as an associated malignancy had to be considered.

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