Case Report

Bronchopulmonary Sequestration Associated with Aspergilloma, with Special Reference to the Finding of Radionuclide Venoangiography

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A case of intralobar bronchopulmonary sequestration associated with aspergilloma was reported. It was suggested by radionuclide venoangiography and aortic cineangiography. The diagnosis was confirmed by subsequent pathologic examination of the lobectomized lung tissue. The pulmonary perfusion scan with ^{99m}Tc -MAA showed a perfusion defect in the right lower lung field. Perfusion defect could also be traced in the same anatomical location in the pulmonary phase of radionuclide venoangiography with ^{99m}Tc -pertechnetate. However, the area of perfusion defect was visualized as hot area, when the injected radionuclide was pumped into the aorta after its first pass through the lung. The radionuclide venoangiography can efficiently detect abnormal systemic blood supply of the lung tissue. It can be an useful method for suggestive diagnosis of bronchopulmonary sequestration.

Key Words: Bronchopulmonary sequestration, Aspergilloma, Radionuclide venoangiography

INTRODUCTION

Bronchopulmonary sequestration is a congenital anomaly of the lung, a portion of which is supplied by the systemic artery without normal connection with the tracheobronchial trees^{1, 2}. It is rare in association with aspergilloma^{1, 3}. It is also a rare cause for focal perfusion defect on the pulmonary perfusion scan⁴. We present this case of bronchopulmonary sequestration associated with aspergilloma with special reference to the significance of radionuclide venoangiography to detect this anomaly non-invasively.

CASE REPORT

The patient, a 28 years old male was admitted to the medical ward of Kaohsiung Medical College Hospital on Nov. 1982 with the chief complaint

of intermittent cough and hemoptysis for around one year. Allergic rhinitis could be traced since his teenager. Physical examination was unremarkable, and no murmur detected on auscultation. The admission routine, including blood, urine and stool routine, liver function tests, renal function tests, and electrolytes were within normal limits. Electrocardiogram revealed normal sinus rhythm with incomplete right bundle branch block. No pathogen was found in the smear and culture of the sputum. Neither obstructive nor restrictive defect was defined in the pulmonary function tests. Chest roentgenographs (PA and Lat. view) and tomographs showed a thin-walled cystic lesion with air-fluid level in the right lower lung field (Fig. 1). Bronchogram revealed displacement and crowding of the segmental bronchi of

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Fig. 1. Chest roentgenographs shows a thin walled cystic lesion with air-fluid level in the right lower lobe.

the right lower lobe and no contrast medium entering the cystic lumen. No communication between the cystic lesion and the esophagus could be visualized in the esophagogram. There was no specific finding in bronchofiberscopic examination.

The pulmonary perfusion scan using ^{99m}Tc -macroaggregate albumin showed a perfusion defect in the right lower lung field corresponding to the cystic lesion in the chest roentgenograph (Fig. 2A). Perfusion defect could also be traced in the same anatomical location in the pulmonary phase of radionuclide venoangiography using ^{99m}Tc -Pertechnetate. However, the area of perfusion defect become visualized in perfusion in the systemic phase in the same study, when the injected radionuclide was pumped into the aorta after its first pass through the lung (Fig. 2B).

Aortic cineangiography showed the cystic lesion deriving its arterial supply from the descending aorta with orifice located in between those of the 10th and the 11th right intercostal artery (Fig. 3). The venous drainage was via the pulmonary vein.

Surgical intervention with right lower lobe lobectomy was performed. Grossly, the thin walled sequestration lay in the central-medial portion of the right lower lobe of the lung without separating pleura from the normal tissue (Fig. 4). It was measured 5.1×4.3 cm in the greatest diameter, and a brownish ball-like material was found in the cystic lumen. In the microscopic examination, the cystic wall was lined with ciliated columnar epithelium. The aberrant systemic arterioles showed constituents of elastic fiber under Orcein's stain (Fig. 5A). Hyphae of the *Aspergillus* species was found in the brownish ball-like material within the cystic lumen (Fig. 5B).

DISCUSSION

Bronchopulmonary sequestration comprises 0.15–6.4% of all congenital pulmonary malformation, and usually the posterior basal segment is affected³⁾. Two types of sequestration are recognized: the intralobar and extralobar type^{1, 2)}. The former is enclosed within the visceral pleura of the affected lobe and the latter is separated

Fig. 2. Pulmonary perfusion scan shows perfusion defect in the right lower lung field (A). Perfusion defect can be defined in the same anatomical location in the pulmonary phase of the radionuclide venoangiography (B, at the 23 and 25 sec frame).

The area of perfusion defect was contrasted out as a hot area in perfusion in the early systemic phase, while the lung is to complete its first pass by the radionuclide (B, at the 29 and 31 sec frame).

from the normal lung tissue and enclosed in its own visceral pleura. However, both derive their systemic arterial supply (aberrant artery) from the aorta or one of its branches.

As detailed previously, this patient is a typical case of intralobar bronchopulmonary sequestration. Unexpectedly a fungus ball due to *Aspergillus* species was detected in the cystic lumen within the sequestered lung. In review of 540

Fig. 3. Aortic cineangiography shows the cystic lesion deriving its arterial supply from the descending aorta (arrows) located in between those of the 10th and 11th right intercostal artery.

published cases of lung sequestration, Savic et al. reported six occasions of tuberculous disease and a case of nocardial infection within the sequestration. However, there was no case of associated infection with *aspergillus* species³⁾. The case of intralobar sequestration associated with aspergilloma was much less reported¹⁾.

Bronchopulmonary sequestration is a rare cause for focal perfusion defect in the pulmonary perfusion scan⁴⁾. The perfusion defect shown by the sequestration can be explained by its blood supply originated from the aorta instead of the pulmonary artery. In the radionuclide venoangiography, the same rationale can be applied to the perfusion defect shown by the sequestration as shown in pulmonary phase. The sequestration is flooded with the radionuclide from the aorta in the early systemic phase of the venoangiography, when the lung with decreasing radioactivity is to complete its first pass by the radioactive agent. Thus, the sequestration contrasts out as a "hot area" in the early systemic phase of the venoangiography. Although it had been applied for this purpose only in a few limited institutions^{5,6)}, radionuclide venoangiography can be a valuable method for suggestive diagnosis of the bronchopulmonary sequestration.

Fig. 4. The specimen from right lower lobe lobectomy. The sequestration lies in the central medial portion of the right lower lobe, with a brownish ball-like material in the cystic lumen (fixed in formalin).

Fig. 5. In the microscopic examination, the cystic wall is lined by ciliated columnar epithelium (A, arrows), and the surrounding arterioles shows constituents of elastic fiber (A, arrow heads, Orcein's stain 25 $\times$). Hyphae of the *Aspergillus* species (B) was found in the brownish ball-like material within the cystic lumen (H.E. stain, 400 $\times$).

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REFERENCES

- 1) Fraser RG, Pare JAP: *Diagnosis of Diseases of the Chest*. 2nd ed, WB Saunder, Philadelphia, p 606-609, 1977.
 - 2) Spencer H: *Pathology of the lung*. 3rd ed, Pergamon Press, Oxford, p 75-87, 1977.
 - 3) Savic B, Birtel FJ, Tholen W, et al: Lung sequestration: report of seven cases and review of 540 published cases. *Thorax* 34: 96-101, 1979.
 - 4) Aman MEV, Lloyd TV and Johnson JC: Focal perfusion defect caused by sequestration in two cases. *Am J Roentgenol* 131: 904-905, 1978.
 - 5) Kawakami K, Tada S, Katsuyama N, et al: Radionuclide study in pulmonary sequestration. *J Nucl Med* 19: 287-289, 1978.
 - 6) Ohkawa H, Uchiyama G, Takahashi H, et al: A case of intralobar pulmonary sequestration in childhood, examined by RI scanning methods. *Jpn J Clin Radiol* 23: 289-293, 1978.
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