Pulmonary Hamartoma : Report of Two Cases

Hajime Goto, M.D. and Hideo Kato, M.D., F.C.C.P.

Department of Internal Medicine, The Institute of Medical Science Hospital,
University of Tokyo, Tokyo

Yoju Miyamoto, M.D.

Department of Surgery, The Institute of Medical Science Hospital,
University of Tokyo, Tokyo

Jun Yamauchi, M.D.

Department of Replacement Surgery, The Institute of Medical Science Hospital,
University of Tokyo, Tokyo

Tomo Wakabayashi, M.D.

Department of Clinical Pathology, The Institute of Medical Science Hospital,
University of Tokyo, Tokyo

We recently encountered two cases of pulmonary hamartoma. One (Case 1) was an endobronchial, and the other (Case 2) was an intrapulmonary hamartoma. Case 1 involved a 50-year-old man who had experienced recurrent bloody sputa. The sagittal tomographic study showed a round mass in the right upper bronchus. Under fiberoptic bronchoscopy, a smooth orange-colored and relatively fragile tumor was found. A right upper lobectomy was performed and the tumor was diagnosed histopathologically as an endobronchial hamartoma. Case 2 involved a 44-year-old man who had a coin lesion in his mid-left lung. A segmentectomy of the left S5 was performed. The histopathological diagnosis of this case was intrapulmonary hamartoma. Endobronchial hamartoma is fairly rare and this case is thought to be the eighth reported case in Japan.

Key words : endobronchial hamartoma, intrapulmonary hamartoma

Introduction

An increased number of lung cancer and benign lung tumor cases has been observed. We encountered two cases of benign lung tumors, one of which was an endobronchial hamartoma and the other an intrapulmonary hamartoma. As noted below, the former disease is fairly rare. Since these two types of hamartoma have considerably different features in their clinical manifestations, we would like to report these cases while also referring to other published materials.

Case Reports

Case 1 involved a 50-year-old man, a heavy smoker, who had for the past five months experienced recurrent bloody sputa, an irritating cough and wheezing in his right retrosternal region. He was hospitalized on April 4, 1978. The physical examination including the PA and lateral roentgenograms revealed no significant findings. The repeated bacteriological examination of sputa was negative. However, a sagittal tomographic study showed a distinct

Fig. 1

Fig. 2

Fig. 3

Fig. 4

Table 1 Results of Pulmonary Function and Blood Gas Analysis of Case 1.

Pulmonary Function		Blood Gas Analysis	
VC (%)	114.7	pH	7.406
FEV (%)	70.2	PaO ₂	95.4 mmHg
RV/TLC (%)	16.0	PaCO ₂	34.8 mmHg
DLCO (%)	82.3	SaO ₂	97.3%
		BE	-2.5

Fig. 5

Fig. 6

round mass lesion in the right upper bronchus, with a small section of radio-opacity (Fig. 1). The results of pulmonary function tests and blood gas analysis were almost normal, as summarized in Table 1. The bronchographic study of the right upper bronchi showed a tumor-like lesion which was located at the bifurcation of right apical (B1) and subapical (B2a) bronchus. The contrast medium hardly filled the periphery of this lesion (Fig. 2). A widened bifurcation between A1 and A2 was observed through angiography of the pulmonary artery. A smooth, orange-colored and relatively fragile protrusion was found attached to the posterior wall of the right upper bronchus, so as to almost obstruct the lumen. This discovery was made by flexible fiberoptic bronchoscopy (Fig. 3). Although transfiberscopic cytological examination by the brushing method did not reveal the characteristics of malignancy, a right upper lobectomy was performed on April 9, 1978 for fear of malignant disease. The induration was found to be palpable at the bifurcation of right upper bronchi. There was no pleural fluid, adhesion or lymph node involvement. The patient subsequently recovered completely. Macroscopic studies showed a tumor located at the bifurcation of right B1 and B2a, extending about 1 cm to the periphery of B2a with a size of 10 mm × 5 mm × 5 mm (Fig. 4). This tumor consisted of both a cartilaginous and a soft papillary portion. Microscopic studies of the tumor showed irregular cartilage formations covered with bronchial epithelium, accompanied by partial papillary proliferations. Squamous metaplasia and erosion were also found. In the submucosa, chronic inflammatory cell infiltration, edema and fibrosis were found (Fig. 5). Apart from these lesions, a caseous necrosis with the diameter of about 2 mm was found at the periphery of B2a. From these findings, this lesion was finally diagnosed as an endobronchial cartilaginous hamartoma with old tuberculous scarring.

Case 2 involved a 40-year-old man, a heavy smoker, who was discovered in August, 1970, to have a homogeneous coin lesion with a clear border and a size of 18 mm × 15 mm × 15 mm located in his lingula by PA and lateral radiograms. However, no clinical symptoms accompanied this lesion. He was hospitalized on July 9, 1974 for further examination to determine exact nature of this shadow. Over the preceding four years, the size of the lesion had remained unchanged (Fig 6). No definite findings were obtained by bronchography, fiberoptic bronchoscopy, bacteriological and other examinations. However, since the patient's previous history, his family history and habits would not allow us to rule out the possibility

Fig. 7

of malignancy, partial resection of the lingula was performed on September 20, 1974, according to the histopathological results from the freezing method. The findings in the chest were as follows. The induration of the tumor, with a smooth surface and elastic-soft hardness, was palpable in the left S5. There was no pleural fluid, adhesion, or lymph node involvement. After several days of postoperative subcutaneous emphysema, the patient recovered completely. The resected tumor had a multilobular conformation with white elastic-soft cartilaginous tissue in the center, surrounded by gelatinous substances. The border of the tumor was clear. Histopathological examination of the tumor showed multilobular myxomatous tissue and cartilaginous substances with clefts lined by cuboidal and columnar epithelium (Fig. 7). The final diagnosis was an intrapulmonary cartilaginous hamartoma.

Discussion

The first report of hamartoma and coinage was made by Albrecht¹⁾ and the first report of pulmonary hamartoma was by Hart²⁾. In Japan, Ozawa's report in 1942³⁾ is believed to be the first.

These pulmonary hamartomas are generally classified into two categories, based on location: intrapulmonary and endobronchial. The clinical manifestations differ between two types considerably. The former has very few subjective symptoms and signs other than lung shadow on chest roentgenograms. The latter, however, is not always discovered during routine roentgen examination of the chest, except where there is pneumonic infiltration or emphysema due to bronchial obstruction, but it shows clinical manifestations from the early stage of growth, including coughing, sputa, hemoptysis, dyspnea, atelectasis and pneumonia. Bronchographic or bronchoscopic studies would also likely to yield considerable information in the case of endobronchial hamartoma, because of its location but not in the case of intrapulmonary hamartoma. However, there are some problems in the diagnosis and therapy of this disease. The possibility of malignant change of hamartoma is generally denied, except in the report of Simon and Ballon⁴⁾, therefore, when a lesion is definitely diagnosed as hamartoma, it is thought that the resection of the tumor itself is sufficient. As for endobronchial hamartoma, the indication would be for resection through a bronchoscope; there are, in fact, some reports on this maneuver^{5) 6) 7)}. However, in practice, there are many cases which have problems in diagnosis. For example, Shiozawa's report⁸⁾ showed that, in patients aged 40 to 50, 41.2%

to 52.3% of the cases of hamartoma were diagnosed as lung cancer. Furthermore, the case report by Kuroda, et al.⁹⁾ showed that the results of cytology from a specimen obtained through bronchial brushing were Papanicolaou class V, and this case was tentatively diagnosed as squamous cell carcinoma. As to our two cases, these features were also found in clinical manifestations, diagnosis and therapy. The difficulty of making a correct preoperative diagnosis was especially recognized. The final diagnoses were made after the histopathological examinations of the resected lung.

The incidence of this disease differs considerably between intrapulmonary and endobronchial hamartoma. According to foreign case studies, the latter is rarer than the former; 3.3% of 215¹⁰⁾, 12.0% of 200⁵⁾, and 16.5% of 200¹¹⁾ cases of pulmonary hamartomas. This trend is clearly found in Japanese cases. That is, so far as we know, the reported cases of the endobronchial hamartoma in Japan were as follows. In 1964, a case was reported by Sato, et al. and then, further, six cases were documented up to this point⁹⁾¹²⁾¹³⁾¹⁴⁾¹⁵⁾. In this connection, the first case of endobronchial chondroma in Japan was reported by Maeda, et al. in 1973. Furthermore, each case was documented by Nagase, et al.¹⁶⁾, by Nishiuchi, et al.¹⁷⁾, and by Iwase, et al.¹⁸⁾ Thus, endobronchial hamartoma is fairly rare in Japan, as well as is endobronchial chondroma. Moreover, the incidence of this lesion within all pulmonary hamartoma cases seems to be relatively low when compared with reports from abroad, although the reasons are not yet clear⁵⁾⁸⁾¹¹⁾.

A part of this work was presented at the 46th Kanto Annual Scientific Meeting, Japan Society of Chest Diseases, Tokyo, May 10, 1980.

Legends

Fig. 1 Sagittal tomography of Case 1. A round mass lesion is seen in the right upper bronchus.

Fig. 2 Bronchography of Case 1. A tumor-like lesion is seen at the bifurcation of right B1 and B2a. Contrast medium hardly fills the periphery of this lesion.

Fig. 3 Case 1. Smooth orange-colored protrusion is found to be attached to the posterior wall of right upper bronchus through the flexible fiberoptic bronchoscope.

Fig. 4 Macroscopic finding of the endobronchial hamartoma of Case 1. This lesion is located at the bifurcation of right B1 and B2a extending about 1 cm to the periphery of B2a.

Fig. 5 Microscopic finding of the endobronchial hamartoma of Case 1. Irregular cartilage formation covered with bronchial epithelium accompanied by partial papillary proliferation is found.

Fig. 6 Chest roentgenogram of Case 2. A coin lesion is seen in the lingula with the size of 18 mm × 15 mm.

Fig. 7 Microscopic finding of Case 2. Multilobular myxomatous tissues and cartilaginous substances are found.

References

- 1) Albrecht: Ueber Hamartome. Verhandl. d. deutsch. path. Gesellsch., 7: 153-157, 1904.
- 2) Hart, C.: Ueber die primaeren Enchondrome der Lunge. Ztschr. Krebsforsch., 4: 578-585, 1906.
- 3) Ozawa, Y.: Resection of the lung. J. Jpn. Surg. Soc., 42: 1863-1944, 1942.
- 4) Simon, M. A. and Ballon, H. C.: An unusual hamartoma (so-called chondroma of the lung). J. Thorac. Surg., 16: 379-391, 1947.
- 5) Young, J.M., Jones, E., Hughes, F.A., Foley, F.E. and Fox, J.R., Jr.: Endobronchial hamartoma. Report of two cases. J. Thorac. Surg., 27: 300-305, 1954.
- 6) Davidson, M.: A case of pulmonary chondroma of the bronchus. Br. J. Surg., 28: 571-574, 1941.

- 7) Kallqvist, I. and Carlens, E.: Endobronchial chondroma as a trigger mechanism in asthma. Report of a case with endobronchial extirpation of chondroma. Dis. Chest, 41: 46-48, 1962.
- 8) Shiozawa, M.: Hamartoma of the lung. J. Jpn. Surg. Soc., 21: 85-100, 1973.
- 9) Kuroda, T., Morimoto, M., Shida, H., Noguchi, O., Ozawa, K. and Maruyama, Y.: Report of two cases of resected endobronchial hamartomas. Jpn. J. Thorac. Dis., 17: 409-413, 1979.
- 10) Bateson, E.M.: Relationship between intrapulmonary and endobronchial cartilage-containing tumor (so-called hamartoma). Thorax, 20: 447-461, 1965.
- 11) Hodges, F.V.: Hamartoma of the lung. Dis. Chest, 33: 43-51, 1958.
- 12) Terashima, M., Yoshizumi, A., Endo, S., Eguchi, S. and Sakashita, I.: A case of endobronchial cartilaginous hamartoma. Jpn. J. Chest Dis., 29: 396-399, 1970.
- 13) Kitahara, M., Tajima, G., Nonaka, H., Tsuzuki, N. and Horie, K.: A case of endobronchial hamartoma successfully treated by surgery. Jpn. J. Chest Dis., 35: 702-707, 1976.
- 14) Okazaki, N., Izumi, S., Arai, T., Kira, S., Hasegawa, T. and Saito, K.: A case of endobronchial hamartoma which showed characteristic symptom and recurrent pneumonia. Intern. Med., 40: 465-468, 1977.
- 15) Kimura, S., Morita, T., Inagaki, K., Arai, T., Hirata, M., Nakano, A., Suzuki, T., Otani, T., Tajima, H., Kadosawa, K., Saito, A., Oshima, T. and Ino, K.: A case of endobronchial hamartoma. J. Jap. Bronchol., 3: 207-212, 1981.
- 16) Nagase, C., Yamamoto, T., Ito, M., Oshima, S. and Takada, Y.: A case of endobronchial chondroma. Jpn. J. Chest Dis., 33: 216-220, 1974.
- 17) Nishiuchi, M. and Hirota, N.: A case of endobronchial chondroma. Jpn. J. Chest Dis., 36: 129-134, 1977.
- 18) Iwase, T., Watanabe, H., Hashiba, A., Iwa, T., Kitagawa, M. and Yokohama, S.: A case of endobronchial chondroma. Jpn. J. Chest Dis., 37: 137-141, 1978.

別刷請求：〒329-04 栃木県河内郡南河内町
大字薬師寺3311-1
自治医科大学ウイルス学教室
後藤 元

(投稿受付 1981年10月13日)