

## An Intrapulmonary Chondromatous Hamartoma Penetrating the Visceral Pleura: Report of a Case

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**A 65-year-old woman had been followed up for a hamartoma-like mass in the right upper lobe of the lung since 1995. A follow-up CT scan showed an increase in the size of the lesion and a new lesion next to the old mass, which was highly suspected to be lung cancer. A right upper lobectomy was thus performed in May 2000. The hamartoma penetrated the visceral pleura, and adhered to the mediastinal pleura. Such a growing hamartoma with pleural invasion has so far rarely been previously reported. (Ann Thorac Cardiovasc Surg 2002; 8: 42–44)**

**Key words:** hamartoma, pleural invasion

### Introduction

A hamartoma is a benign tumor of the lung, accounting for the about 8% of all pulmonary tumors.<sup>1)</sup> Among all benign tumors, it is the most frequent. Most hamartomas are located in the lung parenchyma, and only a few are found in the bronchi. Pulmonary hamartomas, which used to be regarded as developmental abnormalities, are now accepted as genuine benign neoplasms, probably originating from mesenchymal cells of the bronchial wall.<sup>2)</sup> To our knowledge, however, a growing hamartoma penetrating the pleura has rarely been previously reported.

### Case Report

A 65-year-old female patient had been followed for a right pulmonary abnormal shadow since 1995. In 1998, since the shadow had increased in size, a needle biopsy assisted by chest computed tomography (CT) was performed at Kyushu University Hospital in Fukuoka, Japan. The diagnosis was a hamartoma consisting of mature

cartilagenous tissue. Thereafter she had been followed up by periodical chest CT scans (Fig. 1a). On April 21, 2000, her chest CT scan revealed another abnormal mass in the right upper lobe (Fig. 1b). The irregular mass was highly suspected of being adenocarcinoma. No particular symptoms were observed and her pulmonary function, serum chemistry and blood counts were all normal. In addition, no abnormalities were observed on bronchoscopy.

A right upper lobectomy was performed on May 29, 2000. During the thoracotomy, neither pleural effusion nor pleural dissemination were recognized. The hamartoma of the S1 penetrated the visceral pleura and was exposed in the thoracic cavity (Fig. 2). The mass at the S2 that was diagnosed as well differentiated adenocarcinoma was accompanied with pleural retraction but no pleural invasion.

The masses in S1 (hamartoma) and S2 (adenocarcinoma) measured  $3.6 \times 3.0 \times 3.0$  cm and  $2.7 \times 2.4 \times 1.5$  cm in diameter, respectively. Histologically, the hamartoma mainly consisted of mature cartilagenous tissue. Bronchial epithelium, adipose and connective tissue were rimming. There were no malignant cells. The figure clearly shows that a part of chondromatous hamartoma was exposed without any pleura (Fig. 3). The adenocarcinoma had no pleural invasion, vascular or lymphatic permeation of cancer cells. The hilar and mediastinal lymph nodes were dissected and found to be free from any metastasis.

The patient made an uneventful recovery and was discharged on the 11th postoperative day.

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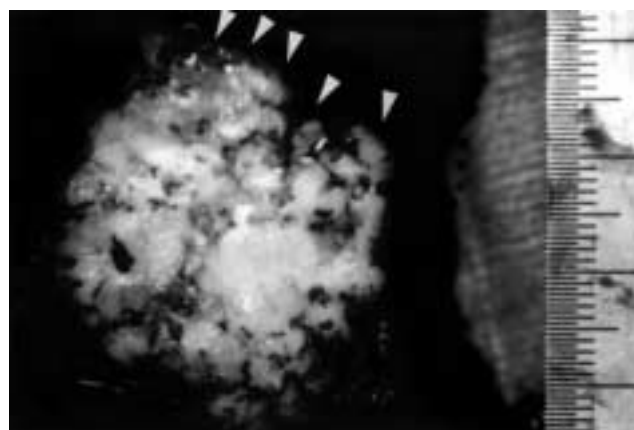


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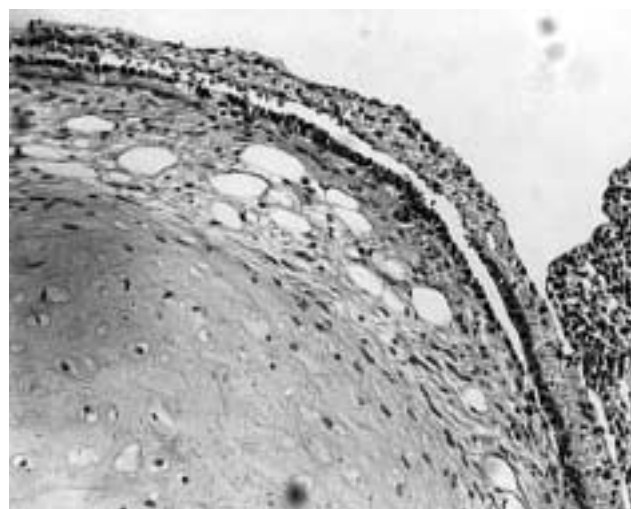


b

**Fig. 1.** a: A chest CT scan revealed a well bordered mass shadow in the right upper lobe.  
b: Follow-up CT scan revealed a new mass lesion.



**Fig. 2.** Macroscopic findings of the hamartoma penetrating visceral pleura.



**Fig. 3.** Histological findings of the resected specimen.

The hamartoma mainly consisted of mature cartilaginous tissue. Bronchial epithelium, adipose and connective tissue were rimming. This figure clearly shows that a part of chondromatous hamartoma was exposed without any pleura. (×200 hpf)

## Discussion

Hamartomas are usually peripherally located, grow slowly and possess an invariably benign nature. They are usually solitary, and well-demarcated nodules generally measuring less than 4 cm in diameter, and approximately 15% of them are calcified. Perihilar localization is rare and an endobronchial location is reported in 3-19% of all patients.<sup>3,4)</sup> According to a report of the Mayo Clinic, a male preponderance is found to be approximately 2:1 and the vast majority of hamartomas were asymptomatic. In this series of 215 cases, sixty-three patients (29.3%) had concurrent neoplasms most commonly, lung carcinoma.<sup>5)</sup> No

malignant changes in leiomyomatous or chondromatous types of hamartomas have been reported however, a few such cases have been mentioned.<sup>4)</sup> A cytogenetic analysis of the pulmonary hamartomas showed an abnormal karyotype and revealed recombinations between 6p21 and 14q24.<sup>6)</sup> These recombinations are common and probably pathogenetically important, thus supporting the view that a hamartoma of the lung is a true neoplasm.

In this case, the hamartoma penetrated the visceral pleura and adhered to the mediastinal pleura, and such findings are quite unusual. In addition, the tumor did not invade the parietal pleura, this to our knowledge, is the first report of a hamartoma showing such a growth tendency. No malignant changes have been reported and this case also had no malignant cells histologically.

This case also presented with lung carcinoma in the same lobe which appeared whilst following the hamartoma for 16 months. There might be some factors promoting tumorigenicity in the local sites.

The diagnosis of a hamartoma can be made with a fine needle biopsy in several cases. Treatment is usually conservative with regular chest roentgenograms during the follow-up. However, when malignancy cannot be ruled out by this procedure or when the nodules are rapidly growing, then an open lung biopsy must be performed. Our patient had another tumor (adenocarcinoma) in the same lobe of the right lung. Regardless of the second growth, a surgical resection was indicated due to the growing tendency of the hamartoma.

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