Multiple Pulmonary Hamartomas Masquerading as Metastatic Lung Tumors: A Case Report

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ABSTRACT —— ***Background.*** The majority of pulmonary hamartomas are recognized as solitary tumors. It is difficult to differentiate multiple pulmonary hamartomas from pulmonary malignant nodules in a patient who has undergone previous surgery for metastatic lung tumor. We report an extremely rare surgical case of multiple pulmonary hamartomas. ***Case.*** A 57-year-old woman with an abnormal chest radiograph was referred to our hospital in August 2004 for further evaluation. Chest radiography and computed tomography showed multiple nodular lesions in the posterior basal segment of the right lung. She had undergone surgery for rectal cancer in September 2002 and had undiagnosed bilateral pulmonary nodules identified at that time. Because the pulmonary nodules in the left lung increased in size, partial resection of the left upper lobe was performed in February 2003. Postoperative histopathologic diagnosis of the lung tumor was metastatic adenocarcinoma originating from the rectal cancer. She underwent chemotherapy starting in March 2003, but an increase in number and size of the pulmonary nodules in the right lower lobe was demonstrated on chest films. She underwent partial resection of the posterior basal segment of the right lung by video-assisted thoracoscopic surgery in September 2004 to remove all nodules. Postoperative histopathologic examination of the resected specimens revealed multiple pulmonary hamartomas consisting of various-sized bronchioles without a cartilageous component. She recovered uneventfully and there have been no signs of recurrence for 40 months since the pulmonary resection in February 2003. ***Conclusion.*** We encountered an extremely rare surgical case of multiple pulmonary hamartomas. If it is difficult to differentiate it from metastatic lung tumors in this case, minimally invasive surgery such as video-assisted thoracoscopic surgery should be performed to establish a definitive diagnosis. (*JJLC.* 2007; 47:37-40)

KEY WORDS —— Multiple pulmonary hamartomas, Metastatic lung tumor, Video-assisted thoracoscopic surgery (VATS), Chemotherapy

INTRODUCTION

Pulmonary hamartoma is usually discovered by a screening chest radiograph or by a post-mortem examination in adults. The majority of pulmonary hamartomas involve a solitary tumor, and microscopically usually contain islands of cartilage and cleft-like spaces lined in part by ciliated epithelium. We report an extremely rare surgical case of multiple pulmonary hamartomas consisting only of various sized bronchioles without a cartilageous component.

CASE REPORT

A 57-year-old woman with an abnormal chest radiograph was referred to our hospital in August 2004 for further examination. Chest radiography and computed tomography (CT) showed plural nodular lesions surrounded by micronodular shadows in the posterior basal segment (S^{10}) of the right lung. She had undergone surgery for rectal cancer in September 2002. Preoperative chest CT in 2002 had revealed a round 8-mm nodular lesion (Figure 1A) in the left upper lobe and plural nodular lesions in the right S^{10} (Figure 2A, 2B, 2C). During routine postoperative outpatient evaluation, chest radiogra-

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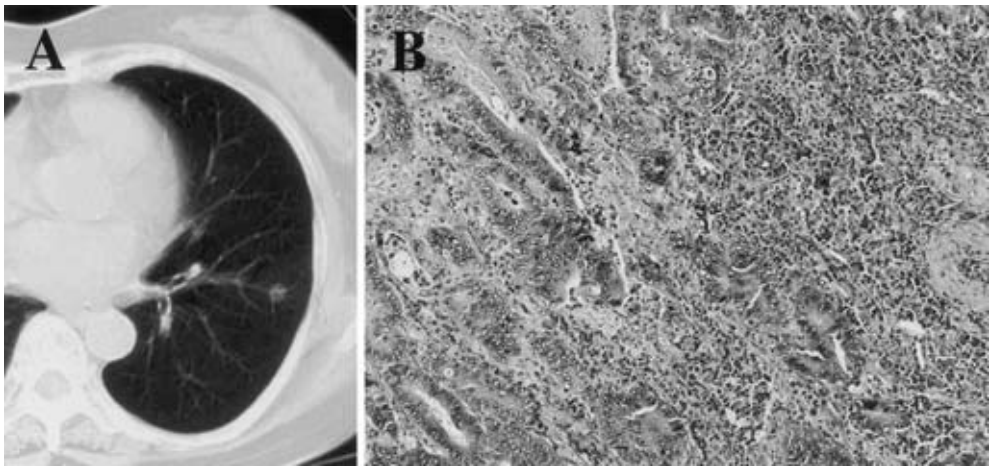

Figure 1. Computed tomography of the left lung in September 2002, showing a round 8-mm nodule (**A**). Histopathologic examination of the nodule revealed metastatic adenocarcinoma from rectal cancer (**B**). **B**, hematoxylin and eosin stain.

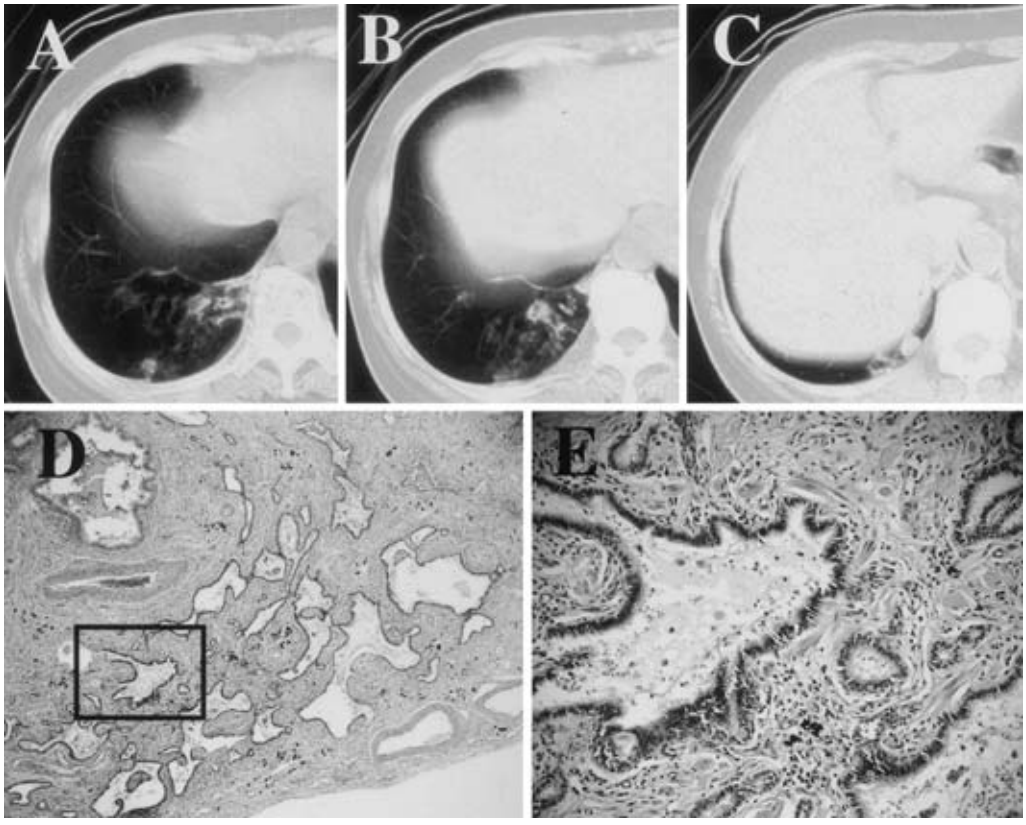

Figure 2. Computed tomography of the right lung in September 2002, showing plural nodular lesions (**A**, **B**, **C**). Histopathologic examination of the nodular lesions revealed multiple pulmonary hamartomas (**D**). Higher magnification of the rectangular portion in **D**, showing various sized bronchioles (**E**). **D**, **E**, hematoxylin and eosin stain.

phy demonstrated enlargement and spicula formation in the left pulmonary nodule. She underwent partial resection of the left upper lobe in February 2003 on a provisional diagnosis of metastatic lung tumor. Histopathologic examination of the lung tumor revealed metastatic adenocarcinoma originating from the rectal cancer

(Figure 1B).

Subsequently the patient received oral uracil-tegafur (UFT) chemotherapy between March 2003 and September 2003. However, the pulmonary nodules in the right S^{10} increased in number and size, and intravenous treatment with 5-fluorouracil (5-FU) plus leucovorin (LV) under a presumptive diagnosis of metastatic lung tumor was administered from January 2004 until March 2004. This was discontinued when she complained of dyspnea on effort and diarrhea due to right pleural effusion and enterocolitis. Modified combination chemotherapy with a novel oral fluoropyrimidine with two modulators (TS-1) plus cisplatin (CDDP) was started in May 2004, but the pulmonary nodules in S^{10} of the right lung increased in number and size. The patient was referred to our hospital in August 2004. Physical examination, blood tests, and serum tumor marker revealed no abnormalities. She underwent partial resection of the S^{10} in the right lung by video-assisted thoracoscopic surgery (VATS) in September 2004 to remove all nodules. Postoperative histopathologic examination of the resected specimens revealed multiple pulmonary hamartomas (Figure 2D, 2E). The patient recovered uneventfully and has had no signs of recurrence for 40 months since resection of the left pulmonary metastatic lesion.

DISCUSSION

The term "hamartoma" was proposed by Albrecht in 1904 to describe certain tumors that result from a localized error in development of a normal constituent or constituents. A hamartoma develops as the result of unbalanced, uncoordinated, and expansive but noninvasive growth in a limited anatomic field. The component tissues follow the general course of development and may reach varying degrees of maturity and functional ability. Further growth of a hamartoma continues after similar normal tissues have reached maturity. [1]

The majority of pulmonary hamartomas are recognized as solitary tumors and usually contain islands of cartilage and cleft-like spaces lined in part by ciliated epithelium. Multiple pulmonary hamartomas are rare. Furthermore, the majority of multiple pulmonary hamartomas consist of either leiomyomatous or chondromatous component. [2-9] To the best of our knowledge multiple pulmonary hamartomas consisting only of various sized bronchioles without cartilage have not been reported previously.

For metastatic lung tumors from colorectal carcinoma, multiple pulmonary metastasectomy is the standard therapeutic procedure when pulmonary functional reserve is sufficient. Complete surgical excision of all pulmonary metastases is often technically feasible with low rates of morbidity and mortality. [10-12] In our case, the plural nodules in the right lung were evaluated preoperatively as metastatic tumors from the rectal cancer, and chemotherapy seemed to have failed to control their progression. Therefore, we performed surgery and a diagnosis of multiple pulmonary hamartomas was obtained. When it is difficult to differentiate multiple pulmonary hamartomas from metastatic lung tumors, as in this case, minimally invasive surgery such as VATS should be performed to make a definitive diagnosis.

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