A Case of Sclerosing Hemangioma Forming a Pedunculated Mass

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We report our experience with an unusual case of sclerosing hemangioma (SH) that formed a pedunculated mass protruding into the thoracic cavity. A pulmonary tumor was found in a 60-year-old female during the medical examination. Computed tomography showed a 19 × 17-mm nodule with a clear border and smooth margin contiguous with the diaphragm in the right S8 segment. Uneven enhancement following contrast medium administration was observed. We performed a 3-port thoracoscopic wedge resection of the right lower lobe. We observed a yellow pedunculated tumor protruding from the diaphragmatic surface of the right lower lobe. The surface of the tumor was smooth and encapsulated. Microscopically, we diagnosed it as a SH. SHs usually exist adjacent to the visceral pleura, but rarely form pedunculated tumors protruding into the cavity as seen in this case. By thoracoscopic surgery, we successfully diagnosed and treated the patient in a minimally invasive manner. Since there have been reported cases of recurrence, we anticipate that periodic follow-up observations will be required.

Key words: sclerosing hemangioma, VATS, lung tumor

Introduction

Pulmonary sclerosing hemangioma (SH) is a relatively uncommon tumor, accounting for approximately 0.2% to 1% of all primary lung tumors. Its preoperative diagnosis is difficult even with a biopsy because differentiation from other diseases such as lung cancer and carcinoid is often difficult.1 In this report, we report our experience with an unusual case of SH that formed a pedunculated tumor protruding into the thoracic cavity. On the basis of intraoperative findings, the mass was suspected to be a pleural tumor such as solitary fibrous tumor.

Case Report

A pulmonary tumor was found in a 60-year-old female during the medical examination. She had no symptoms. She was a nonsmoker and had a past history of bronchiectasis of the right middle lobe. Computed tomography (CT) showed a 19 × 17-mm nodule with a clear border and smooth margin that was contiguous with the diaphragm in the right S8 segment (Fig. 1). Uneven enhancement was observed following contrast medium administration. The tumor showed no internal calcification. There were no signs of involvement of pulmonary arteries and veins or lymph node enlargement. Magnetic resonance imaging revealed equal signal intensity on T1-weighted images and high and low signal intensity areas on T2-weighted images. Although imaging initially led us to suspect a benign tumor such as SH, we could not rule out the possibility of a malignancy. Therefore, surgery was performed for definitive diagnosis as well as treatment.

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tumor protruding from the diaphragmatic surface of the right lower lobe. The surface of the tumor was smooth and encapsulated (Fig. 2). There was no pleural effusion or any dissemination in the thoracic cavity, and the contiguous diaphragm was uninvolved. Although the middle and lower lobes adhered to one another extensively due to the presence of bronchiectasis, they were not adherent to the tumor. Judging from the intraoperative findings, we concluded a high possibility for the presence of a pleural tumor such as solitary fibrous tumor. We performed wedge resection using an endoscopic stapler after securing a sufficient margin. The duration of surgery was 40 min and the blood loss was minimal. She was discharged from the hospital 2 days after the surgery without complications.

Microscopic examination revealed that the resected tumor had resulted from the hyperplasia of moderately large type II alveolar epithelial cells and small round cells with bright vesicles. Solid, papillary, and sclerotic patterns were observed (Fig. 3). Immunostaining revealed that the tumor was positive for thyroid transcription factor-1 and epithelial membrane antigen, and we diagnosed it as a SH.

Discussion

SH was originally regarded as a pathology originating from blood vessels, and it was named so by Liebow et al. in 1956. The subsequent progress in immunohistological and electron microscopic research has led to its current classification as a benign tumor or one with an extremely low malignant potential originating from alveolar epithelial cells. It accounts for 0.2% to 1% of primary lung tumors, is frequent among middle-aged women, has a smooth margin, and is encapsulated by a desmoplastic membrane. Its surface consists of a mixture of ash gray, yellow, and brown areas.

It usually exists adjacent to the visceral pleura, but rarely does it form a pedunculated tumor protruding into the cavity as seen in this case. Kim et al. examined 16 cases of pulmonary SH and reported one unusual case of a pedunculated mass that presented clinically as a mediastinal mass. Im et al. have reported one case of a SH with a tumor on the interlobar surface of the lung, and Sakamoto et al. reported a case of SH isolated in the mediastinum.
On CT, a SH appears to be a rounded tumor with a clear border and smooth margin, and often shows uneven enhancement after contrast administration. The CT findings of the present case fell into this typical pattern of the disease. Under histopathological examination, its typical pattern can be summarized into four patterns: papillary areas, solid areas, sclerotic areas, and hemorrhagic areas. In most cases, at least three of these components can be observed. Using needle biopsy, it may be difficult to differentiate SH from bronchioloalveolar carcinoma or metastatic papillary adenocarcinoma, particularly in SH cases exhibiting a predominant papillary pattern or multiple nodules.8–10)

There have been reports of cases that develop lymph node metastasis,4, 11) local recurrence after enucleation,12) and local recurrence after 10 years.13) Although most cases are benign, there remains some risk for the low malignant type. Iyoda et al.14) reported one case with recurrent tumor. They performed immunohistochemical examination for Ki-67 and p53 expression in order to assess the biological activity of SH. The Ki-67 labeling index and p53 expression of the recurrent tumor was low. They concluded that this indicated a low proliferative activity in the recurrent case and, therefore, suggested that this recurrence might have been secondary to incomplete resection of the tumor. We anticipate that regular follow-up observations will be required for this case in the future.

References