

Solitary Capillary Hemangioma of the Lung: A Report of Two Cases

It has long been recognized that tumors of capillary vessels in the lung are extremely rare. While pulmonary capillary hemangiomatosis, which is a rare cause of pulmonary hypertension, is relatively well known, there have been only a few reports of solitary capillary hemangioma (SCH) of the lung. Herein we report two cases of SCH. Both cases were first detected as a solitary nodule of the lung in chest computed tomography images. Both lesions were recognized as early lung cancer and surgical resections were performed. At low power view, one was not encapsulated but was well delineated from the non-neoplastic lung and the other was pseudo-capsulated. Both tumors consisted of uniform capillaries with cuboidal or flattened endothelial cells. There was no cytologic atypia. Endothelial cells were highlighted by CD31 stain. Awareness of this entity is important for pathologists for differential diagnosis of a solitary pulmonary nodule. (*J Lung Cancer* 2012;11(2):102–104)

Key Words: Lung, Hemangioma, Capillary, Solitary pulmonary nodule

Hyun-Woo Lee, M.D.¹
Soomin Ahn, M.D.¹
Young Mog Shim, M.D., Ph.D.²
Yong Soo Choi, M.D., Ph.D.²
Kyung-Soo Lee, M.D., Ph.D.³, and
Joungho Han, M.D., Ph.D.¹

Departments of ¹Pathology, ²Thoracic Surgery, and ³Radiology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, Korea

Received: October 16, 2012
Accepted: November 18, 2012

Address for correspondence
Joungho Han, M.D., Ph.D.
Department of Pathology, Samsung Medical Center, Sungkyunkwan University School of Medicine, 50, Irwon-dong, Gangnam-gu, Seoul 135-710, Korea
Tel: 82-2-3410-2765
Fax: 82-2-3410-0025
E-mail: Joungho.han@samsung.net

It has long been recognized that tumors of capillary vessels in the lung are extremely rare. Among this group, pulmonary capillary hemangiomatosis is relatively well known, characterized by a proliferation of pulmonary capillaries in the alveolar septa, perivascular connective tissue, bronchial wall, and pleura (1). It is also known to be a rare cause of pulmonary hypertension and obstruction of pulmonary veins and venules (2).

However, there have been only a few reports of solitary capillary hemangioma (SCH) of the lung (3-6). Although most have reported cases occurring in neonates or children, Fugo et al. reported two cases of adult SCH (5). Awareness of this clinical entity is important because the lesions are difficult to differentiate radiologically from early lung cancer. In the cases in Fugo et al. (5), the lesions were incidentally detected as small nodules with ground glass opacity. The radiologist diagnosed both lesions as early lung cancer, however histologic

and immunohistochemical findings were consistent with the diagnosis of SCH of the lung (5).

Herein, we report two further cases of SCH of the lung. Both cases were first detected as a solitary nodule of the lung in chest computed tomography (CT) images. Both lesions were recognized as early lung cancer and surgical resections were performed.

CASE REPORT

Case 1

A 55-year-old man visited the thoracic surgery department of the Samsung Medical Center, Seoul, for further evaluation of an abnormal finding on lung examination that had been incidentally detected by routine medical check-up. His physical condition was good, and there were no significant signs or

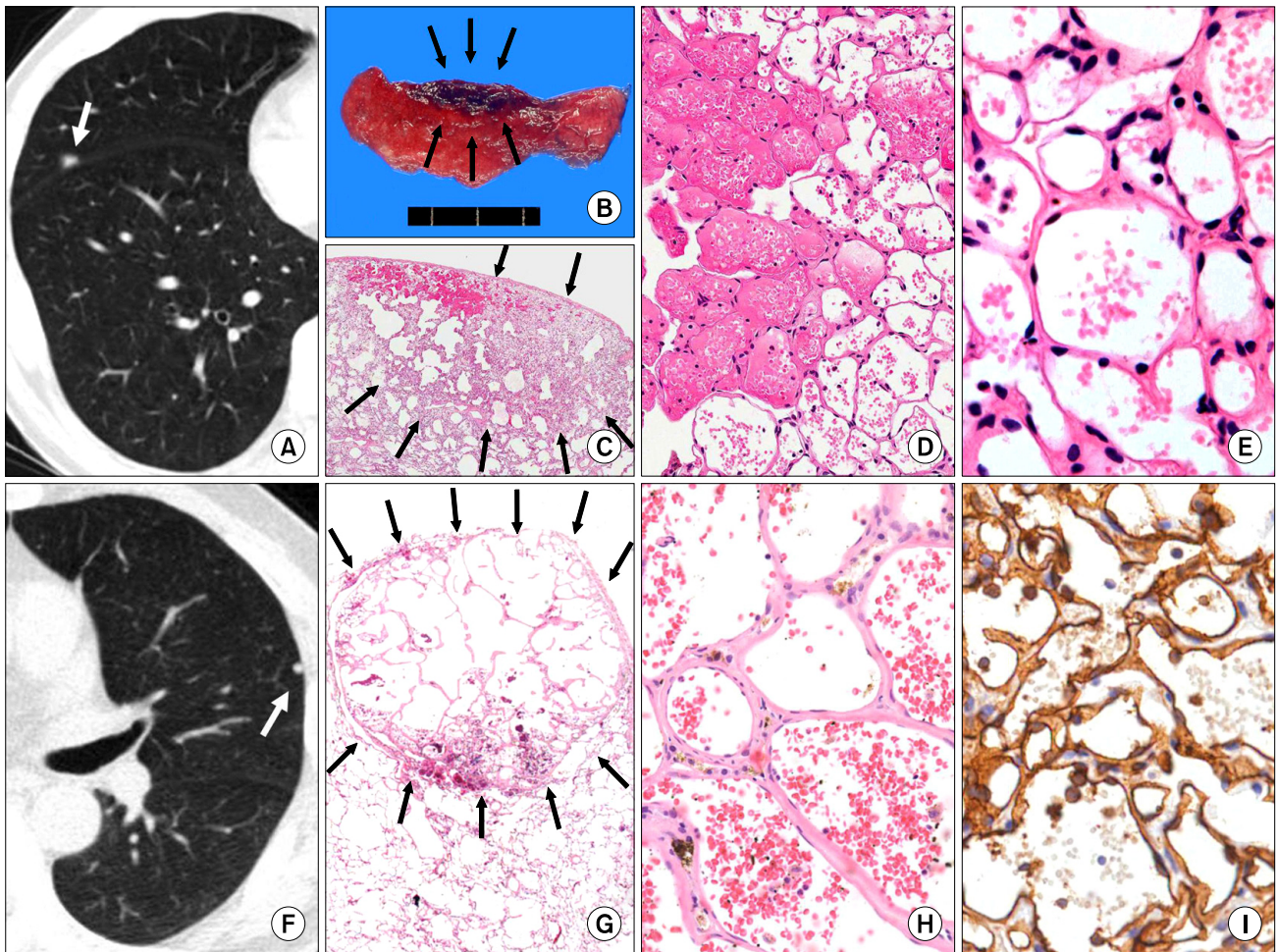


Fig. 1. (A) In the first case, chest computed tomography (CT) shows a small single nodular mass (arrow). (B) Wedge resected lung shows an ill-defined soft hemorrhagic mass (arrows). (C) The tumor is not encapsulated but well delineated from the non-neoplastic lung (arrows). (D, E) The tumor consists of uniform capillaries, with cuboidal or flattened endothelial cells. There is no cellular atypia or mitosis. (F) In the second case, chest CT shows a single nodular mass (arrow). (G) The tumor is round in shape and is pseudo-encapsulated (arrows). (H) The tumor consists of dilated capillaries. There is no cellular atypia or mitosis. (I) CD31 immunohistochemical stain highlights endothelial tumor cells (1 : 80, Dako, Glostrup, Denmark) (C and G, H&E, $\times 10$; D, H&E, $\times 100$; E and H, H&E, $\times 200$; I, CD31, $\times 200$).

symptoms that were consistent with pulmonary hypertension or veno-occlusive disease, such as hemoptysis or dyspnea on exercise. Other cutaneous or internal organ lesions were not found. Chest contrast computed tomography (CT) showed a growing 9 mm-sized solid nodule in the bottom portion of the right middle lobe (Fig. 1A). The radiologist reported this lesion as representing an area of minimally invasive adenocarcinoma or invasive adenocarcinoma with lepidic tumor growth. No preoperative histological confirmation was performed.

The patient underwent a diagnostic wedge resection of the right middle lobe. The wedge resected lung showed an ill-defined soft hemorrhagic mass. The tumor was not encap-

sulated but was well delineated from the non-neoplastic lung (Fig. 1B, C). The tumor consisted of uniform capillaries with cuboidal or flattened endothelial cells (Fig. 1D, E). The endothelial cells were highlighted by CD31 stain (1 : 80, Dako, Glostrup, Denmark). There was no other abnormality in the non-neoplastic lung. No significant cytologic atypia was identified. The Ki-67 index was positive in 5% of tumor cells. There was no evidence of recurrence during 12 months of follow-up.

Case 2

A 50-year-old man visited the thoracic surgery department of

the Samsung Medical Center, Seoul for detailed evaluation of abnormal findings that had been discovered incidentally by low-dose screening lung CT scan. He had no significant signs or symptoms. There were no other lesions of skin or internal organs. Non-contrast helical chest CT revealed a 10 mm-sized ground-glass opacity nodule in the left upper lobe, and another subpleural small nodule measuring 5 mm in the left upper lobe below the main lesion (Fig. 1F). During follow up, the main lesion slightly increased in size, therefore histologic confirmation was recommended for this lesion. The subpleural small nodule showed no change in size during follow-up. Wedge resections for both nodules were performed for histologic diagnosis.

The 10 mm-sized nodule was histologically confirmed to be adenocarcinoma. The other subpleural mass was well-defined from the adjacent lung parenchyma and pseudo-encapsulated (Fig. 1G). This was a 5 mm-sized round hemangioma composed of dilated capillaries, lined by single layered flattened endothelial cells (Fig. 1H). Endothelial cells were highlighted by CD31 stain (Fig. 1I). There has been no recurrence during six months of follow-up.

DISCUSSION

Vascular tumors of the lung are extremely rare. There have been a limited number of reports of pulmonary capillary hemangiomatosis (1,2), which has been described as comprising multiple nodules in the lung parenchyma or bronchovascular walls, composed of infiltrating thin-walled capillary vessels (1,2). It can be a rare cause of pulmonary hypertension and secondary pulmonary veno-occlusive disease. SCH is even more rare and hemangioma has been seen more commonly in children (3,6). There have been only a few case reports of SCH of the lung, most of which have been in very young patients (4-6). Fugo et al. (5) reported two cases of SCH affecting middle-aged adults. Herein, we report two further cases of adult SCH. These are worth reporting because SCH can be a rare cause of a solitary pulmonary nodule. Localized capillary hemangioma manifests as a localized cystic lesion or focal ground-glass opacity nodule in a CT scan (3,5). Therefore it is easily misinterpreted by radiology as cancerous lesion. It is important for pathologists to be aware of this disease entity for accurate diagnosis.

Histologically, capillary hemangioma consists of dilated cap-

illaries, lined by single layered flattened endothelial cells. The border of this type of tumor can be either well-circumscribed or relatively ill-defined, as was reported in several previous studies (4,5). The differential diagnosis includes other vascular tumors such as cavernous hemangioma, sclerosing hemangioma or angiosarcoma. Angiosarcoma is easily distinguished from hemangioma due to its cellular atypia. In addition, capillary hemangioma is composed of well-formed capillaries. Cavernous hemangioma is composed of large, dilated vessels filled with blood (6). Galliani et al. (6) reported a case of pulmonary cavernous hemangioma causing massive hemoptysis. Sclerosing hemangiomatosis of the lung can be confused with capillary hemangiomatosis due to their prominent angiomatoid features. However, sclerosing hemangiomatosis is, in fact, a misnomer and current consensus favors these being a benign or very low-grade neoplasm arising from primitive respiratory epithelium. This tumor consists of superficial cuboidal epithelium and round stromal cells with hemorrhagic areas. Tumor cells do not express vascular markers and show epithelial differentiation (7).

In conclusion, SCH of the lung is a rare vascular tumor that can be clinically confused with lung cancer. It is composed of dilated capillaries, lined by single layered flattened endothelial cells. It is important for pathologists to be aware of this entity for differential diagnosis of a solitary pulmonary nodule.

REFERENCES

1. Umezu H, Naito M, Yagisawa K, Hattori A, Aizawa Y. An autopsy case of pulmonary capillary hemangiomatosis without evidence of pulmonary hypertension. *Virchows Arch* 2001; 439:586-592.
2. Wagenvoort CA, Beetsla A, Spijker J. Capillary haemangiomatosis of the lungs. *Histopathology* 1978;2:401-406.
3. Abrahams NA, Colby TV, Pearl RH, Chipps BE, Juris AL, Leslie KO. Pulmonary hemangiomatosis of infancy and childhood: report of two cases and review of the literature. *Pediatr Dev Pathol* 2002;5:283-292.
4. Bowyer JJ, Sheppard M. Capillary haemangioma presenting as a lung pseudocyst. *Arch Dis Child* 1990;65:1162-1164.
5. Fugo K, Matsuno Y, Okamoto K, et al. Solitary capillary hemangioma of the lung: report of 2 resected cases detected by high-resolution CT. *Am J Surg Pathol* 2006;30:750-753.
6. Galliani CA, Beatty JF, Grosfeld JL. Cavernous hemangioma of the lung in an infant. *Pediatr Pathol* 1992;12:105-111.
7. Kim GY, Kim J, Choi YS, Kim HJ, Ahn G, Han J. Sixteen cases of sclerosing hemangioma of the lung including unusual presentations. *J Korean Med Sci* 2004;19:352-358.