A 44-year-old woman underwent surgery for lung cancer. Although preoperative computed tomography did not reveal a tiny nodule, pathological examination of the background lung showed that type II pneumocyte-like tumor cells grew papillary in an area of approximately 2.3 × 1.2 mm. This lesion exhibited hemorrhage, hemosiderosis, calcification, and varying degrees of fibrosis, leading to the diagnosis of sclerosing hemangioma. This is the first reported case of microscopic sclerosing hemangioma undetectable by chest computed tomography.

Keywords: sclerosing hemangioma, lung cancer, histology

A 44-year-old woman was noted to have an abnormal shadow in the right upper lung field on her chest X-ray during a routine medical checkup (Fig. 1A), and was referred to our department. She had a history of smoking 10 cigarettes per day for a year, but stopped smoking at the age of 21 years.

Blood chemistry showed no abnormalities, and the tumor markers NSE, CEA, SLX, and SCC were within normal limits. Chest computed tomography (CT) demonstrated an ill-defined, irregularly bordered tumor of 4.8 cm in diameter, in the right S1 (Fig. 1B).

The tumor was diagnosed as a well-differentiated adenocarcinoma by CT-guided needle biopsy. With the diagnosis of lung cancer (cT2N0M0, stage IB), the patient underwent a right upper lobectomy with a hilar and mediastinal lymph node dissection. Postoperative histopathological examination showed that hobnail-shaped tumor cells grew along the alveolar septa, forming an elastotic scar in the center of the tumor, which led to the diagnosis of bronchioloalveolar carcinoma, pT2N0M0, stage IB (Fig. 1C). On the other hand, in the background lung at a distance from the tumor, type II pneumocyte-like tumor cells, which have cuboidal to low columnar cytoplasm and small round nuclei with minimal nuclear atypia, grew papillary with fibrotic sclerosis in an area of...
approximately 2.3 × 1.2 mm. Two tiny lesions faced each other, with one being characterized by marked and the other mild fibrosis (Fig. 2A). They exhibited diverse histological features mainly consisting of sclerosis and vascular proliferation and including hemorrhage, hemosiderosis, and calcification, leading to the diagnosis of sclerosing hemangioma (Fig. 2B–2D). The tumor histology featured two neoplastic cell types, and cuboidal tumor cells lined papillae and formed tubules, whereas pale cells filled the papillary cores and formed sheets in solid areas (Fig. 2B–2D).

The patient’s postoperative course was uneventful, and she has remained in good health since discharge. At present, 2 years after surgery, chest CT shows no abnormal intrapulmonary shadows. The patient has remained free from lung cancer recurrence, and will continue to be followed-up on an outpatient basis.

Discussion

Sclerosing hemangioma was first described by Liebow and Hubbell in 1956.5) According to the 1999 histological classification of lung tumors by the World Health Organization, sclerosing hemangioma is defined as a miscellaneous tumor. Although never considered a vascular lesion, it was named such on account of the frequent hemorrhage and hemosiderin deposition within the tumor. The name sclerosing hemangioma reflects two common histological features, sclerosis and vascular proliferation, that are now thought to represent secondary changes in an essentially epithelial neoplasm.4) Ultrastructural and immunohistochemical studies showed that pulmonary sclerosing hemangioma is likely to be the manifestation of the primary proliferation of epithelial cells, probably type II pneumocytes.4, 6, 7) The term sclerosing pneumocytoma is, therefore, more appropriate than sclerosing hemangioma, although the latter is still preferred by the World Health Organization for historical reasons.

Sclerosing hemangioma is quite rare in Western countries, but has an incidence that is comparable to carcinoid tumor in Japan.8) It typically occurs in middle-aged females, and most patients are asymptomatic.1, 4) Most lesions are solitary, but 4% are multifocal.9) Although this tumor has generally been thought to have a benign nature, a few cases with associated lymph node metastasis have been previously reported in the literature.4, 7, 9) In our case, no lymph node metastases were noted.

Sclerosing hemangiomas have been reported to range from 1 to 11 cm in diameter, with a mean of approximately 3 cm.1–4) In contrast to previously reported sclerosing hemangiomas, the present case showed a tiny lesion
Microscopic Sclerosing Hemangioma

Ann Thorac Cardiovasc Surg Vol. 17, No. 5 (2011) 509

as small as 2.3 mm in diameter. A search of the literature revealed reports of sclerosing hemangiomas of approximately 10 mm, but not 2.3 mm, in diameter. In the present case, we reviewed the patient’s CT images after surgery, but could not identify tiny nodular shadows suggestive of sclerosing hemangioma. Since there have been no reports of sclerosing hemangiomas detected incidentally on the close histopathological examination of lung sections, the present case is valuable in understanding the natural course of sclerosing hemangioma.

Sclerosing hemangioma is composed of two types of cell: cuboidal cells covering papillary structures and solid-growing polygonal round cells with pale cytoplasm. The majority of tumors demonstrate papillary, sclerotic, solid, and hemorrhagic growth patterns. One of these patterns may predominate, but they are more commonly seen in combination, as in our case. The papillary pattern is the most common but may merge with sclerotic areas. The sclerotic pattern shows hyalinized collagen in papillae or solid areas. Sheets of round cells bordered by surface cells form the solid pattern, and the hemorrhagic areas feature blood-filled spaces lined by tumor cells. In the papillary pattern, complex papillae are comprised of surface cells covering a stroma of round cells, as shown in the present case. Associated histological findings include hemorrhage, hemosiderosis, foamy macrophage accumulation, focal dystrophic calcification, cystic degeneration, and granuloma, and the microscopic lesion in the present patient also exhibited histological features including hemorrhage, hemosiderosis, and calcification. The sclerosing hemangioma in this patient consisted of two tiny lesions with different degrees of fibrosis. The tendency of tiny fibrosing hemangiomas to differentiate in two different directions suggests the neoplastic aspect of the tumor.

Sclerosing hemangioma is a clinically benign tumor which grows very slowly, with a reported doubling time of 660 to 1250 days. No tumor-related deaths have been reported, even in the rare instances of multifocal tumors or intrathoracic lymph node metastases. Surgical excision alone is curative, and this patient required no further treatment.
References