A case of pulmonary sclerosing hemangioma with low ^{18}FDG uptake in PET

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Abstract. Pulmonary sclerosing hemangioma (PSH) is a relatively rare benign neoplasm, often asymptomatic and presenting as a solitary pulmonary nodule on radiological imaging studies. In the present case report, we examined a case of PSH in a young adult female, and reviewed the literature pertaining to PSH with an emphasis on ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}FDG PET/CT) and pathology. Immunohistochemical staining was also performed to confirm the diagnosis of sclerosing hemangioma. The results revealed that the tumor cells were immunopositive for epithelial membrane antigen, thyroid transcription factor-1 and vimentin and cytoskeleton 7. The patient recovered and was discharged. Thus, ^{18}FDG PET/CT may be used in the diagnosis of a solitary benign pulmonary nodule.

Introduction

Pulmonary sclerosing hemangioma (PSH) is a relatively rare neoplasm first described by Liebow and Hubbell in 1956 (1). In most patients, it is often asymptomatic, the clinical course is benign and the prognosis following resection is excellent (2). Biopsy is regarded as an important diagnostic tool in PSH. Moreover, immunohistochemical staining markedly suggests that sclerosing hemangioma originates from primitive respiratory epithelium.

Radiological diagnosis is usually based on computed tomography (CT) findings, typically presenting as a smooth or slightly lobular, peripheral and solitary pulmonary nodule. To the best of our knowledge, little is known regarding the appearance of pulmonary sclerosing hemangioma, making it difficult to identify this neoplasm on a ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}FDG PET/CT) scan. An example of this rare benign neoplasm occurring in a young adult female of Asian ethnicity is described in this study.

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Case report

A 32-year-old woman was incidentally admitted to the hospital for an abnormal nodule in the right lower lung field on chest X-ray following an annual medical checkup. The patient was asymptomatic and a non-smoker. The medical history, physical examination and laboratory fndings were all normal. Serum levels of carcinoembryonic antigen, squamous cell carcinoma antigen, cancer antigen 125, cancer antigen 19-9 and neuron-specific antigen were all normal. Sputum cytology was negative for malignancy.

To differentiate malignant from benign pulmonary nodule, a whole-body ^{18}FDG PET/CT scan was performed on November 19, 2007. Chest CT scan revealed a 1.1 cm, solitary lesion located in the right lower lobe. The margin appeared irregular and slightly lobular (Fig. 1). A clearly lobulated nodule with heterogeneous moderate enhancement and possible involvement of the vessel upon contrast-enhanced CT examination of the thorax was revealed (Fig. 2). It demonstrated low FDG uptake in the right lower lobe of lung [standardized uptake value (SUV) not shown]. No enlarged mediastinal lymph nodes, hepatic metastases or mediastinal invasion were evident upon ^{18}FDG PET scan (Fig. 3).

Since the nodule is peripheral and small, the bronchoscopic biopsy and fine needle aspiration cytology under CT guidance was unable to be performed. Consequently, the patient underwent an excisional lung biopsy for a definite diagnosis.

The resected specimens from the nodule were diagnosed as sclerosing hemangioma (SH) following an intraoperative examination of the frozen sections. Macroscopically, the tumor was well circumscribed without invasion of the surrounding tissue. Microscopically, the tumor exhibited the typical histological features of round stromal cells and cuboid surface cells in papillary, sclerotic, solid and hemorrhagic patterns. The stromal cells were small with well-defined borders, centrally located round to oval bland nuclei and eosinophilic cytoplasm (Fig. 4). Immunohistochemical staining revealed that the tumor cells were immunopositive for epithelial membrane antigen, thyroid transcription factor-1 and vimentin and cytoskeleton 7.

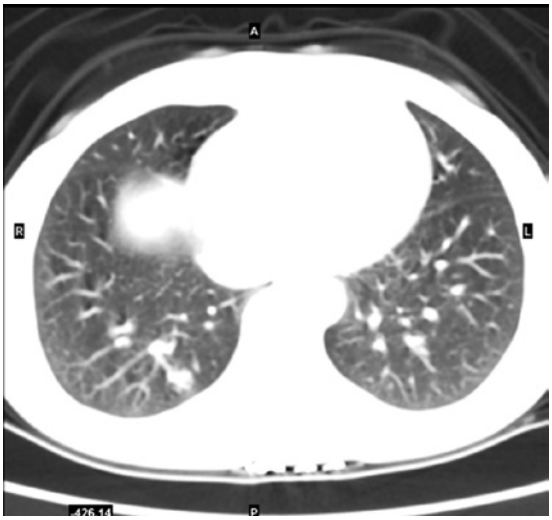

Figure 1. Chest computed tomography (CT) on admission shows a mass with an irregular margin, and which is slightly lobulated in the right lower lobe.

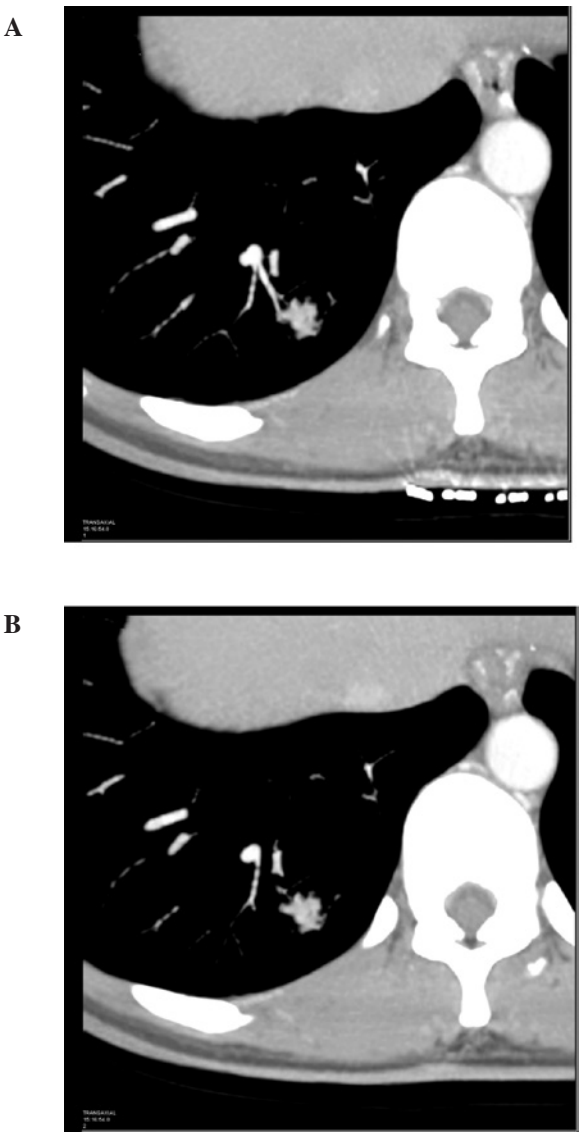

Figure 2. Contrast-enhanced CT image of the thorax with mediastinal windows showing the clearly lobulated mass as a hypervascular lesion. CT, computed tomography.

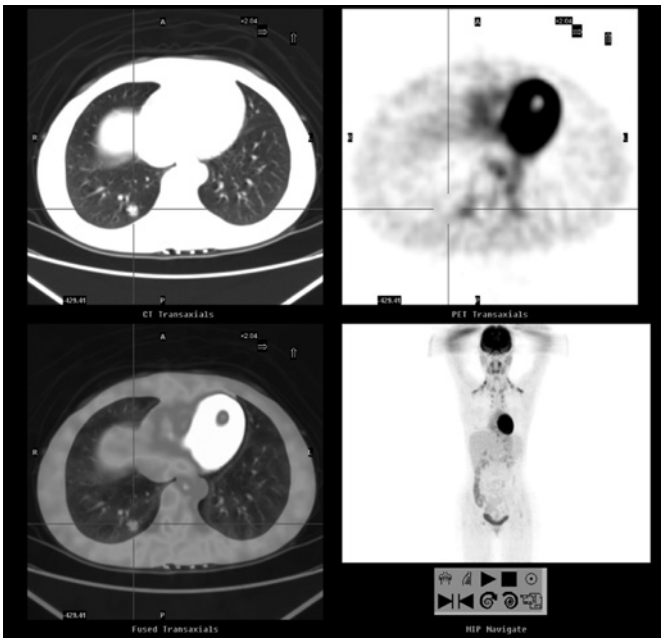

Figure 3. FDG-PET/CT imaging shows a well-defined nodule with low FDG uptake. FDG-PET/CT, F-fluorodeoxyglucose positron emission tomography/computed tomography.

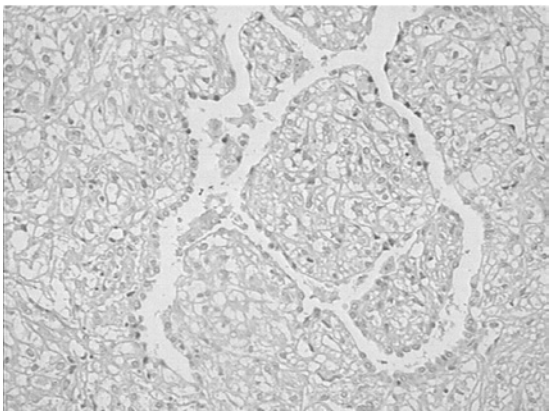

Figure 4. Microscopic histopathological image of the sclerosing hemangioma shows typical histological patterns that characterize sclerosing hemangioma of the lung.

The patient recovered well and was discharged following an uncomplicated postoperative course.

The study was approved by the local ethics committee and the patient provided informed consent.

Discussion

PSH was first reported by Liebow and Hubbell in 1956 (1). PSH is predominantly identified in middle-aged women and accounts for approximately 1% of all benign pulmonary tumors. Most patients are asymptomatic, with the lesion being found incidentally upon chest radiographs performed for alternative reasons. Additionally, only a few patients experience hemoptysis, dyspnea, cough or chest pain due to an enlargement of the tumor and compression of surrounding tissue (2-4).

The characteristic radiological and CT imaging features of the PSH are reported as a solitary, well-defined, round or oval nodule of less than 3 cm in diameter, and occasionally exhibiting marked heterogeneous contrast enhancement and calcification in a minority of tumors (4). However, cases involving large mass, multiple lesions or nodal metastasis have also been reported (5-7). In the present case, the patient was free of symptoms and the tumor was found incidentally upon X-ray examination. The chest CT examination revealed a solitary heterogeneous lesion located in the right lower lobe. The margin appeared irregular and slightly lobular. Contrast-enhanced CT examination of the thorax revealed a clearly lobulated nodule with heterogeneous moderate enhancement, with possible involvement of the vessel. Furthermore, the mean size of PSH is 1.1 cm with no calcification or fat in CT. We suspected that the lesion was benign, but the diagnosis could not be confirmed preoperatively. Thus, ^{18}FDG PET/CT studies may be useful when neither calcification nor fat are revealed on CT studies.

^{18}FDG PET has been shown to be more accurate than contrast-enhanced CT in differentiating malignant from benign pulmonary nodules. Most malignant lesions have increased uptake due to higher glycolytic activity, whereas benign lesions are typically associated with lower uptake. However, certain carcinoid tumor or inflammatory conditions may demonstrate a false positive result to limit the accuracy of ^{18}FDG PET. The experience using ^{18}FDG PET scans in sclerosing hemangioma is limited. To the best of our knowledge, only a few cases are available on ^{18}FDG PET reported in PSH. In the patient reported by Hara *et al*, only slightly elevated uptake with an SUV of 1.8 was noted (8). In the patient reported by de Koning *et al*, there was a slight amount of ^{18}F-FDG uptake (SUV of 1.6) (9). By contrast, the two patients reported by Hsu *et al* demonstrated SUV of 2.72 and 3.93, which were well above the cut-off value (SUV of 2.5), suggestive of malignancy. The high FDG uptake may be attributed to the larger tumor size and potentially low-grade malignant nature of sclerosing hemangioma (10). The patient in the present study also exhibited low uptake, which was preoperatively believed to most likely represent a carcinoid on the ^{18}FDT-PET/CT scan. The ^{18}FDG-PET scan features of our patient were compatible with the pathological evaluation of PSH.

Cell proliferation is rarely identified in PSH, suggesting a benign behavior. However, certain authors regard sclerosing hemangioma as a potentially low-grade malignancy since few cases of lymph node metastases and postoperative local recurrence of PSH have been reported. The prognosis following surgical resection is excellent, even when lymph node metastasis or multiple lesions are present. The patient described in the present study was also asymptomatic and had a favorable prognosis. Although PSH is a benign condition, further careful follow-up is required (6,11).

The pathogenesis of PSH has yet to be fully elucidated. Previous studies have proposed various possibilities, including mesothelial, mesenchymal, neuroendocrine and epithelial (12). Based on immunohistochemical and electron microscopic techniques, PSH reportedly arises from type II pneumocyte and multipotential primitive respiratory epithelium (13). Macroscopically, PSH is typically well-defined, well circumscribed, encapsulated and often hemorrhagic. Microscopically, sclerosing hemangiomas are composed of round stromal cells and cuboid surface cells with papillary, sclerotic, solid and hemorrhagic patterns (2). The use of immunohistochemical staining is crucial in the diagnosis of PSH (3). It is reported that concomitant positivity of round stromal cells and cuboidal surface cells for epithelial membrane antigen and thyroid transcription factor-1, and negativity of round stromal cells for cytokeratin confirm the diagnosis of sclerosing hemangioma (2).

In conclusion, PSH is a relatively rare, benign pulmonary neoplasm. The radiology findings are relatively characteristic, particularly those of ^{18}FDG PET scan. When a benign tumor is highly suspected, observation is reasonable. However, when the lesion has grown more rapidly than is expected and is large and symptomatic, patients should undergo resection.

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