Preoperative Diagnosis in 46 Cases of Pulmonary Sclerosing Hemangioma

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Key words: Computed Tomography; Diagnosis; Pulmonary Sclerosing Hemangioma

Pulmonary sclerosing hemangioma (PSH), a thoracic tumor, was first described by Liebow and Hubbell[1] in 1956. Recently, immunohistochemical evidence and characterization of antibodies have clarified that PSHs are true tumors. They are mainly composed of epithelial cells (Type II pneumocytes) aligned in different directions and accompanied by proliferation and reaction of other components. Because these tumors, especially when multiple, are rare, there is little published information about their clinical manifestations and imaging data. Thus, they can easily be misdiagnosed as malignant disease of the lungs. The presence of a solitary, well-defined, round or oval mass on computed tomography (CT) scanning suggests, however, that pathological examination is necessary to make a definite diagnosis.

We enrolled 46 cases of PSH attending our hospital from March 1994 to March 2013 in this retrospective study. We analyzed their clinical characteristics, imaging and pathological findings, diagnosis, treatment, and prognoses. We, here, summarized this clinical experience with special reference to the diagnosis, growth, and treatment of these tumors.

Five of the 46 patients were men. Their ages ranged from 20 to 76 years (mean 51.4 years) and 17 patients were asymptomatic, whose tumors having been discovered during routine check-ups. Eleven of the symptomatic patients presented with hemoptysis, 22 with cough, six with chest pain, nine with expectoration of sputum, and two with fever. Three of the five patients with multiple lesions were asymptomatic, one presented with hemoptysis and chest pain, and one with expectoration of sputum.

Access this article online

Quick Response Code:

Website: www.cmj.org

DOI: 10.4103/0366-6999.182839

CT scans showed solitary, well-defined, round or oval masses, with lobulation (n = 11), calcification (n = 7), or aerial semilunar sign (n = 1) [Figure 1]. The diameters of the lesions varied from 0.9 cm to 6.9 cm (mean 2.6 cm). Five patients had one major lesion of diameter 1.2 cm to 5.1cm and multiple smaller lesions scattered throughout both lungs and pleura.

In 34 patients who underwent enhanced CT, the mean CT attenuation was 86.8 HU (range, 35.5–140.3 HU) in the enhanced phase. Four patients showed enhancement on delayed scans with CT attenuation ranging from 82.5 to 114.6 HU. The lesions were of homogeneous density in 25 patients and heterogeneous density in nine.

A diagnosis of PSH was made on CT images in only seven patients. The most frequent radiologic misdiagnoses were primary lung cancer (n = 6), metastasis (n = 3), followed by inflammatory pseudotumor (n = 4) and hamartoma (n = 2).

Macroscopically, these tumors had four major patterns such as solid, sclerotic, papillary, and hemorrhagic; most contained a mixture of these patterns. Of 34 cases who underwent enhanced CT, 25 had lesions of homogeneous density composed of a uniform proportion of all four patterns (n = 13) or a mixture of hemorrhagic and papillary

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Received: 19-12-2015 **Edited by:** Peng Lyu
How to cite this article: Hu AM, Zhao D, Zheng H, Wang QH, Lyu Y, Li BL. Preoperative Diagnosis in 46 Cases of Pulmonary Sclerosing Hemangioma. Chin Med J 2016;129:1377-8.

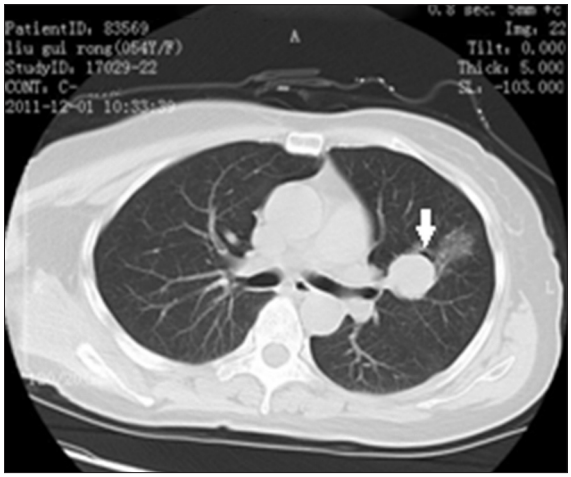

Figure 1: Computed tomogram image showing a solitary lesion (white arrow) with aerial semilunar sign in the left chest.

patterns (n = 12). The remaining nine lesions were of heterogeneous density with an uneven proportion of all four patterns, sclerotic and solid being the most frequent.

Surgery was performed in 45/46 patients; 21 patients underwent enucleation (the most common procedure), nine patients were wedge resection, and 15 patients were lobectomy. Lymph node samplings were performed in four patients; there were no lymph node metastases.

In five patients with multiple lesions, enucleation was performed in two patients and wedge resection in two patients to remove large lesions. All resected lesions were diagnosed as sclerosing hemangioma. One patient refused surgery after being diagnosed by lung biopsy.

No recurrence occurred in any patient during follow-up by telephone, chest X-ray, or CT review ranging from 4.2 to 13.5 years. In particular, one patient who had multiple lesions and refused surgery was followed for 4.2 years, during which all nodules remained unchanged.

PSH occurs mostly in East Asia. Devouassoux-Shisheboran *et al*.[2] summarized 100 patients who were mostly women (83/100, 83%) with a mean age of 46.2 years. In the current series, 89.1% (41/46) of the patients were female and the mean age was 51.4 years. The minor discrepancies between these data and those of the present case series might be related to racial differences or our smaller sample size.

PSH of the lung is typically asymptomatic. However, in the current series, 63.0% (29/46) of subjects had symptoms, including hemoptysis, cough, chest pain, expectoration, and fever. Some of the symptoms, such as expectoration and fever, have rarely previously been reported. The greater proportion of symptomatic subjects in our series might reflect a lack of regular health checks.

On chest radiographs, PSHs usually appear as peripheral, solitary nodules or masses with smooth margins. In our group, the mean CT attenuation was 86.8 HU (range, 35.5–140.3 HU) in the enhanced phase and ranged from 82.5 to 114.6 HU in four delayed enhanced scans. Macroscopically, the lesions mainly contained hemorrhagic and papillary patterns and were rich in blood vessels. Thus, obvious enhancement is a major characteristic of PSHs.

PSHs are easily misdiagnosed on CT scans as lung cancer, inflammatory pseudotumor, and hamartoma. Maximum attenuation of 20–60 HU is reportedly a good predictor of malignancy;[3] Whereas, PSH shows much stronger and rapid homogeneous enhancement with maximum CT values ranging from 90 to 110 HU.[4] On CT images, hamartomas have smooth borders and focal collections of fat or fat alternating with areas of calcification.[5] Inflammatory pseudotumors are associated with respiratory symptoms and CT scans mostly show single or multiple small lesions or a honeycomb-like low-density shadow.

PSHs seldom recur or metastasize, therefore, which are thought to be benign. However, Devouassoux-Shisheboran *et al*.[2] reported four multifocal PSHs, four pleural metastases, and one hilar lymph node metastasis in 100 cases of PSH. Four of our patients underwent lung resection and mediastinal lymph nodes sampling, no lymph nodes showed metastasis. In the current series, chest CT showed three patients had bilateral multiple tumors and two multiple pleural metastases. None of the five patients with multiple PSHs developed recurrence or metastasis during follow-up ranging from 4.2 to 13.5 years.

Our data suggest that PSH occurs mostly in middle-aged women. CT images show solitary nodules or masses with smooth margins, and obvious enhancement is characteristic. Multiple lesions could appear in PSH, therefore, it could not exclude the possibility of PSH.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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