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Diagnosis and Video-Assisted Thoracic Surgery of Pulmonary Sclerosing Pneumocytoma

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Abstract: **This work was to explore the clinical characteristics and the efficacy of thoracoscopy surgery of pulmonary sclerosing pneumocytoma (PSP). The clinical features, imaging, pathology and surgical methods of 35 patients with PSP treated by video-assisted thoracic surgery (VATS) in the affiliated Hospital of Qingdao University from January 2014 to January 2019. All cases were screened and collected for retrospective analysis. 35 patients, 32 were female and 3 were male, average age of 52.69 ± 10.23. The cases in different methods VATS wedge resection, segmental resection, lobectomy, compound lobectomy, lymph node dissection were 17, 11, 5, 2, 4 respectively, and all lymph nodes were negative. The postoperative complication rate was 8.57%. The average follow-up time was 35.6 months. 2 cases were lost to follow-up. The other 33 patients survived well without recurrence, metastasis, or death. VATS can confirm the histopathology and remove the lesion. At present, it is the most effective method for the treatment of PSP, and the patient has a good prognosis.**

Keywords: Pulmonary sclerosing pneumocytoma; Video-assisted thoracic surgery; Diagnosis; Treatment

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1. Introduction

Pulmonary sclerosing pneumocytoma (PSP) is called pulmonary sclerosing hemangioma (PSH), which is rare in the clinic. It is a benign tumor that occurs in the lungs. In 2015, the WHO lung tumor classification officially named this tumor as PSP, which belongs to adenoma in pulmonary neuroendocrine tumors [1]. The clinical symptoms and signs of patients with this disease have no obvious specificity. Most patients were found during physical examination that the imaging characteristics are similar to other benign tumors and even some malignant tumors, which is difficult to distinguish clinically. Therefore, we collected the clinical data of 35 patients with PSP treated in our hospital and retrospectively analyzed their clinical characteristics, imaging, pathology, and prognosis to improve the diagnosis and treatment of the disease.

2. Materials and Methods

2.1.General information

A retrospective analysis was performed on 35 patients with PSP in affiliated hospital of Qingdao University from January 2014 to January 2019. The basic information is in Table 1. All patients in this group were isolated lesions: 2 cases were near the hilar or lobar bronchus, and the rest were located in the parenchyma around the lung.

Table 1. Basic data of all patients

Variable	Number	Proportion
Main symptoms		
No symptoms	27	77.14%
Chest tightness and suffocation	1	2.86%
Cough and sputum	5	14.29%
Chest pain	2	5.71%
Location		
Upper lobe of right lung	1	2.86%
Middle lobe of right lung	6	17.14%
Lower lobe of right lung	6	17.14%

Middle and inferior lobe of right lung	2	5.71%
Upper lobe of left lung	7	20.00%
Left lower lobe of lung	13	37.14%
Preoperative diagnosis		
PSP	9	25.71%
Benign tumor	7	20.00%
Lung nodules	7	20.00%
Malignant tumor	6	17.14%
Pulmonary space-occupying lesion	5	14.29%
Pleural mesothelioma	1	2.86%

Table 2. Surgical methods and intraoperative histopathology results

Variable	Number	Proportion
Surgical methods		
VATS Wedge pneumonectomy	17	48.57%
VATS segmentectomy	11	31.43%
VATS lobectomy	5	14.29%
VATS composite lobectomy	2	5.71%
Lymph nodes dissection	4	11.43%
Intraoperative histopathology		
PSP	31	88.57%
Malignant tumors cannot be ruled out	2	5.71%
PSP in the middle lobe with adenocarcinoma in the lower lobe of the right lung	1	2.86%
PSP in the middle lobe and lower lobe of the right lung	1	2.86%

2.2. Operation method

All patients were provided video-assisted thoracic surgery(VATS), double-lumen endotracheal intubation, general anesthesia, and single-lung ventilation. The surgical approach was single or double hole. The choice of surgical methods were VATS wedge resection with the lesion located in the peripheral tissue and a smaller diameter, segmental resection while the wedge resection is difficult, and if the lesion is extensive, invades the leaf bronchus, close to the hilum, choose lobectomy or composite lobectomy.

2.3. Follow-up method

All patients were followed up by telephone or outpatient review. Follow-up was performed at 1 and 3 months after operation, and every 6 months thereafter. The follow-up items were mainly chest CT. The follow-up deadline was December 2019.

3. Results

3.1. Surgical results

All patients were successfully completed the operation and no adverse or malignant events occurred during the operation. The operation method was in Table 2. The average operation time was 111.77 ± 39.94 min, intraoperative bleeding was 59.14 ± 65.36 ml, postoperative tube days were 3.37 ± 1.57 days, and postoperative hospitalization days were 5.49 ± 1.95 days. Two patients were converted to open surgery due to extensive chest adhesions. Lymph node dissection was performed in 4 patients due to intraoperative histopathology that could not rule out malignant lung tumors and all lymph nodes were negative. Postoperative complications: There were 2 patients with pneumonia and 1 patient with air leaks. The complication rate was 8.57%.

3.2. Pathological and immunohistochemical results

The intraoperative histopathology results were shown in Table 2. Postoperative histopathology showed that all 34 patients were PSP, and one patient had PSP in the right middle lobe combined with lower adenocarcinoma. The pathological results of lymph node tissues were all negative. Immunohistochemistry: All patients' pathology includes epithelial cells and round cells. Both cells express the two genes EMA and TTF-1. The other expressed genes are shown in Figure 1.

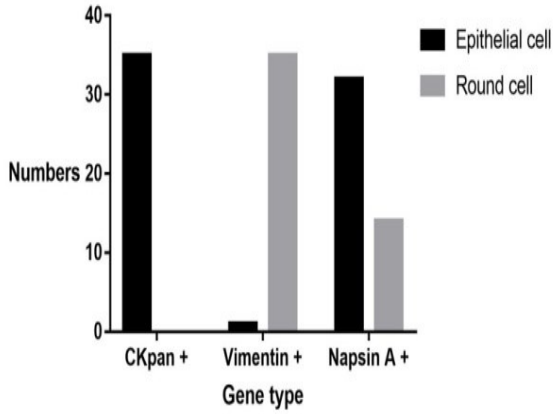

Figure 1. Other genes expressed by epithelial cells and round cells.

3.3. Follow-up results

The average follow-up time was 35.6 months and 2 patients were lost to follow-up. All other patients survived well during the follow-up period, and no recurrence, metastasis, or death occurred.

4. Discussion

PSP is a rare benign tumor, first was reported by Liebow et al. under the name PSH in 1956 [2]. In 2015, WHO reclassified it as a lung adenoma, suggesting that it is a benign tumor. At present, it is believed that PSP tumor cells are mainly derived from original respiratory epithelial cells [3], because both PSP and original respiratory epithelial cells express genes such as TTF-1 and EMA and Napsin A [4,5].

The disease is more common in Asia. The incidence of female patients is significantly higher than that of males, which may be related to the expression of estrogen receptors, progesterone and other sex hormone receptors in women [4]. Most patients do not have any discomfort, and a few patients have symptoms such as cough and sputum. The ratio of women to men in this group was 32:3, and 27 (77.14%) asymptomatic patients found abnormalities due to physical examination. The other 8(22.86%) patients had cough, expectoration, chest pain, breath holding and other symptoms.

The lesions are mostly isolated in the surrounding lung parenchyma. The lesions adjacent to the hilum or main bronchi, subpleural, superior diaphragm, multifocal and bilateral lesions [6] are rare. Some scholars studied 89 patients with PSP, of which 69 (77.53%) were peripheral isolated lesions. 13 (14.6%) were central lesions, 3 (3.4%) were multiple lesions, and 4 (4.5%) were combined with Lung cancer. Maximum lesion diameter is 0.3 cm to 6.0 cm [7]. In this study, 31 cases were located in the surrounding lung parenchyma, 2 cases had lesions adjacent to the hilum and main bronchus, 1 case was multiple, and 1 case had lung adenocarcinoma. Lesion diameter range: 0.7 to 5 cm, with an average of 3.25 ± 5.98 cm, mostly in the left lower lobe (13/35, 37.14%).

Common CT signs are well-defined solitary nodules or tissue masses. There are burrs, lobes, or speckle-like ground glass shadows in some lesions. Other uncommon CT manifestations include: air crescent sign, halo sign, calcification, and clinging vascular sign [8]. The CT enhancement of most benign tumors is often not obvious, and the CT enhancement value is <20HU. The PSP is the opposite. Due to the microvessel density (MVD) is high, the CT value is more than 20HU, because the degree of enhancement is positively correlated with MVD [9,10]. The enhanced features of the dynamic enhanced CT scan can be distinguished from other benign tumors and some malignant tumors, and the diagnosis rate can be improved.

Pathology is the gold standard for the diagnosis of PSP. Pathological specimens were white-yellow or red-brown in section, tough or soft, with or without capsules, and generally did not invade surrounding tissues. "Four growth patterns and two types of cells" can be seen under the microscope: four growth patterns including papillary, sclerotic, solid and haemorrhagic; two types of cells, epithelial cells and round cells [11]. Four growth patterns and two kinds of cells are the key points in the diagnosis of PSP [12]. Percutaneous puncture or bronchial biopsy has some limitations. Insufficient material can cause misdiagnosis or difficult diagnosis. Traditional thoracotomy surgery is more traumatic and patients recover slowly, and VATS can confirm the pathological diagnosis and is currently the most effective treatment [13]. Its advantages are small incision, light interference of cardiopulmonary function, less postoperative complications, short hospital stay, and the surgical method can be adjusted according to pathological results, and the patient has a good prognosis. Except for 2 patients who were lost to follow-up in this group, the rest were cured, and no recurrence, metastasis, or death occurred during follow-up. The incidence of postoperative complications was 8.57%.

Although PSP is a benign tumor [14], some cases show multiple tumors [15], accompanied by malignant tumors [16], mediastinal lymph node metastasis [17,18], pleural spread [19], stomach [20], liver [16], bone [21] metastasis, etc. It is potentially malignant or low-grade malignant. At present, no death has been reported so far. Patients with mediastinal lymph node metastasis and distant metastasis should be completely reselected after pathology is confirmed and closely followed up.

5. Conclusion

Most patients with PSP are asymptomatic. Preoperative diagnosis is mainly based on chest CT, but it lacks specificity and is easily misdiagnosed. So histopathology is the gold standard for diagnosis. VATS has little effect on cardiopulmonary function, minor trauma, fewer postoperative complications, and rapid recovery of the patient. It can confirm the histopathology and remove the lesion. At present, it is the most effective method for the treatment of PSP, and the patient has a good prognosis.

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