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# Pulmonary sclerosing pneumocytoma with lymph node metastasis and high 18FDG uptake in PET/CT: A rare case report and literature review

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Pulmonary sclerosing pneumocytoma (PSP) was first reported by Liewbow and Hubbellin (1956). It was initially called pulmonary sclerosing hemangioma (PSH) because it was thought to be derived from endothelial cells with rich vascularity. However, the 2015 World Health Organization Classification of Lung Tumors removed the term “sclerosing hemangioma”, since these tumors lack vascular origin (Travis et al., 2015), and the renamed tumor was considered a rare and benign lung tumor. However, several reports have described PSP with lymph node metastasis (Soo et al., 2017). Thus, PSP is not always benign and has a potential to metastasize. PSP shows varying levels of 18F-fluorodeoxy-glucose (FDG) accumulation, with the maximum standardized uptake value (SUVmax) ranging from the background value to 6.4 (Chen et al., 2012; Jiang et al., 2018). Therefore, it is easily misdiagnosed as other malignant tumors and is often removed surgically or treated improperly. Here, we present a rare case of PSP in a young woman with lymph node metastasis, a higher 18FDG uptake in the positron emission tomography-computed tomography (PET/CT) scan, and a SUVmax of 6.1.

A 23-year-old woman was incidentally admitted to the First Affiliated Hospital, Zhejiang University School of Medicine in Hangzhou, China with a diagnosis of an abnormal nodule in the right upper lung field on chest X-ray following her annual medical checkup.. The asymptomatic nonsmoker patient had unremarkable medical history. The serum levels of carcinoembryonic antigen, squamous cell carcinoma antigen, cancer antigen 125, cancer antigen 199, and neuron-specific antigen were normal. Sputum cytology showed a negative result for malignancy. The family and medical histories were negative for relevant diseases.

A chest CT scan upon admission revealed a 9.1 × 5.5 × 5.2 cm mass in the right upper lobe. Tumor localization was visually identified in CT images (Figs. 1a, 1b). The tumor was a well-defined nodule with strong and homogeneous enhancement. The CT values of the lesion were 45 and 97 Hounsfield units (HU) in the plain scan and contrast-enhanced scan, respectively. Lymph node metastasis was also observed in the right hilar, with a size of 1.2 × 1.0 × 0.7 cm. No associated mediastinal lymph node enlargement was detected.

We then conducted a PET/CT scan and found a high FDG accumulation within the mass, with a SUVmax of 6.1. The lymph node in the right lung hilar showed an SUVmax of 5.1 (Figs. 1c, 1d).

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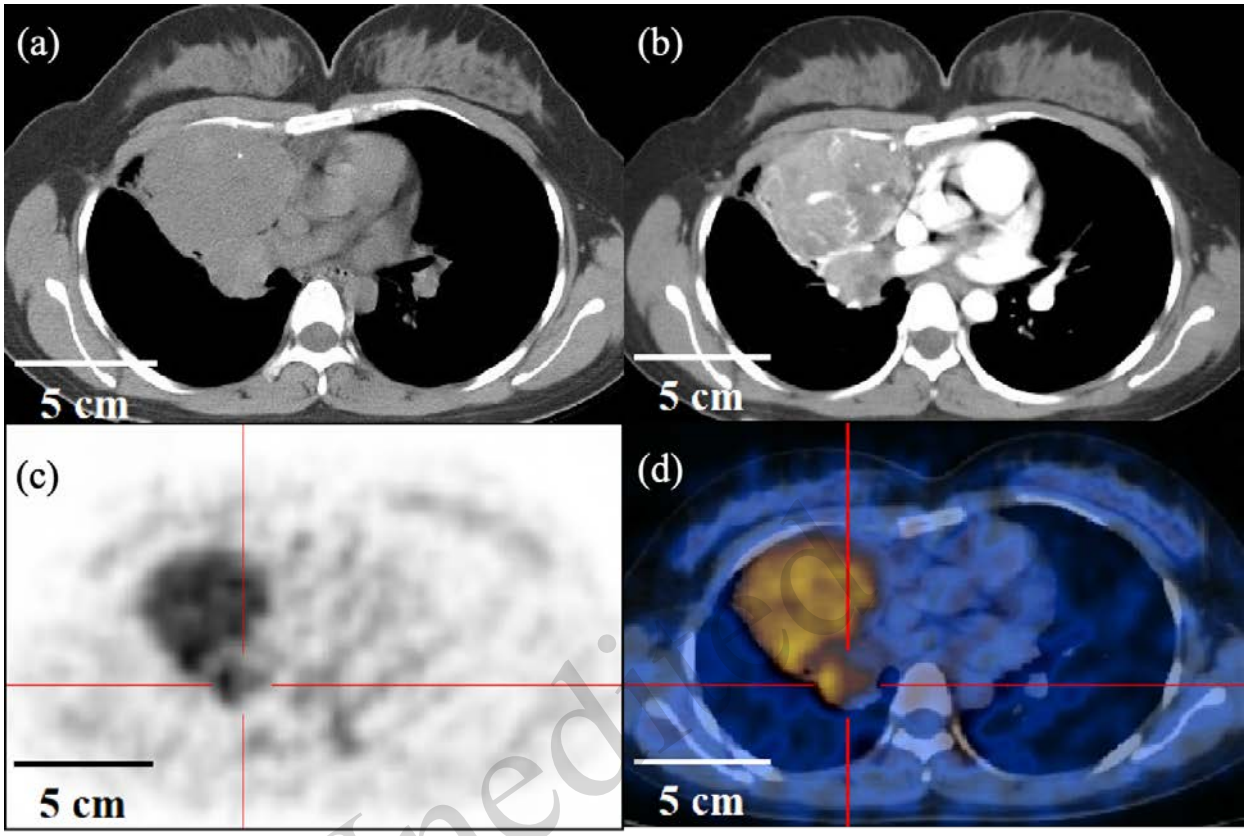


**Fig. 1 Chest CT axial scan: plain (a) and enhanced (b), showing the right upper lobe tumor. The corresponding positron emission tomography scan (c) and fused positron emission tomography-computed tomography scan (d) images show a relatively high uptake of 18F-fluorodeoxy-glucose in the tumor (the maximum standardized uptake value (SUVmax)= 6.1) and lymph node (SUVmax = 5.1).**

To establish a precise pathological diagnosis of PSP, percutaneous CT-guided core needle biopsy was performed. Because the tumor size was substantial, the patient underwent right upper lobectomy. The tumor mass and right hilar lymph node were removed. 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 and had centrally located round to oval-shaped bland nuclei and eosinophilic cytoplasm (Figs. 2a, 2b). Immunohistochemical staining revealed that the tumor and right hilar lymph node were positive for thyroid transcription factor (TTF-1) (Fig. 2c) and cytokeratin 7 (CK7) (Fig. 2d) and negative for CD31 and CD34. The Ki-67 positivity was 15%.

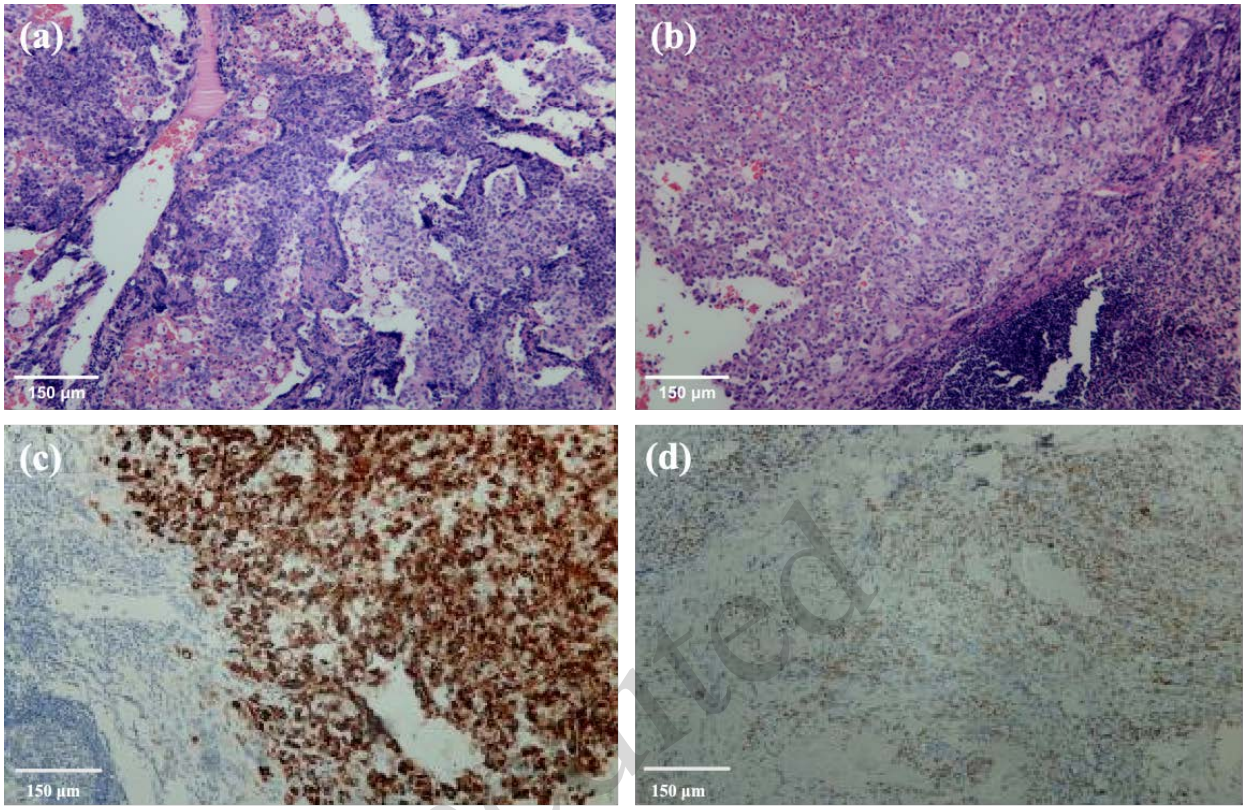


**Figure 2. Histopathological examination with hematoxylin and eosin staining of the right upper lobe lesion (a) and right hilar lymph node metastasis (b); original magnification, ×100. Immunohistochemical staining revealed that the right hilar lymph node metastasis was positive for Cytokeratin 7 (c) and Thyroid Transcription Factor-1, (TTF-1) (d).**

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PSP typically occurs in middle-aged adults, with a female-to-male ratio of 7.5:1. It has a higher incidence in Asia and accounts for 5.8% of surgically resected benign tumors in Japan (Masuda et al., 2018). The most common form is an asymptomatic solitary pulmonary mass. PSP-related symptoms include hemoptysis, chronic cough, chest pain, etc. In the present case, although the lesion was large, the patient did not present any clinical symptoms. This may be attributed to the sufficient growth space of the lung and the fact that the lesion did not involve the trachea or bronchi.

In the chest CT scan, PSP typically presents as a peripheral, solitary, round, well-defined mass with homogeneous opacity. Most patients with PSP show no calcification within the lesions, and apparent enhancement is a major characteristic of PSP. Since its first report, this type of tumor has been considered a rare and benign form of lung tumor; PSP with lymph node metastasis is an extremely rare case. Since the first report of PSP with lymph node metastasis by Tanaka et al (1986), several studies have described PSP with metastasis in the lymph nodes (Kim et al., 2003; Pokharel et al., 2016; Chien et al., 2009; Pokharel et al., 2016; Katakura et al., 2005; Miyagawa-Hayashino et al., 2003; Yano et al., 2002), bone, mediastinum, and pleura (Kim et al., 2015; Arumugam et al., 2017; Pokharel et al., 2016). Thus, PSP is not always benign and has a potential to metastasize. Although the mechanism of PSP metastasis remains unclear, it may be related to age, gender, primary tumor location, and tumor size. Based on previous reports, PSP metastasis may be more likely to occur in young patients, males, and individuals with large tumors (Pokharel et al., 2016; Sun et al., 2012). It was considered that the epithelial-mesenchymal transition (EMT) may play an important role in PSP metastasis; this is because the round cells of PSP may be derived from the EMT of the surface cells.

The patient of the current case report was diagnosed to have a malignant tumor on admission by CT and PET/CT scans based on the presence of a large mass in the lung and hilum with a high FDG uptake. Because the tumor mass had abundant blood supply, the large blood vessels were wrapped and traversed. Therefore,

chemotherapy was recommended as the main treatment approach considering that the surgical approach might not lead to successful recovery. Accordingly, a biopsy was performed, and the results suggested the possible diagnosis of PSP. Considering the prognosis of PSP and the young age of the patient, the lung mass and hilar lymph node were completely removed during the surgery. The lesions and hilar lymph nodes were finally diagnosed as PSP based on immunohistochemical analysis. The patient has been followed up for 8 years, with no tumor recurrence detected. Moreover, the patient gave birth to two healthy children during this period.

Tanaka et al (1986) reported the first case of PSP with lymph node metastasis. To date, only approximately 20 cases of PSP with lymph node involvement have been reported, and all these cases were from Asian populations (Soo et al., 2017). Devouassoux-Shisheboran et al. (2000) reported a 1% incidence of hilar lymph node metastasis in all PSPs. In most cases, the tumor size does not appear to correlate with the extent of lymph node involvement (Soo et al., 2017). Yano et al. (2002) reported that larger tumors had greater potential for lymph node metastasis. Our observation was consistent with this study; however, it was also observed that young male patients with PSP tend to have lymph node metastasis, and some patients even showed no tumor progression or recurrence during regional lymph node metastasis (Miyagawa-Hayashino et al., 2003).

PSP patients show varying levels of FDG accumulation, with the SUVmax ranging from the background value to 6.4 (Jiang et al., 2018; Satoh et al., 1989). Furthermore, the tumor size of PSPs was shown to be positively correlated with the SUVmax of these tumors (Xu et al., 2019). We searched and reviewed all articles published in English in the PubMed database on PSPs with pathological confirmation and 18F-FDG PET/CT findings. Nineteen PSP tumors were described in 14 reports published in the last 10 years. The relationship between tumor size and FDG uptake is summarized in Table 1 and Figure 3. In previous reports, the FDG uptake of the tumor was positively correlated with tumor size. However, some authors (Travis et al., 2015) indicated that tumor size does not appear to correlate with the extent of lymph node involvement, suggesting that the malignant potential of PSP is independent of tumor size (Chen et al., 2012). The Ki-67 proliferation index, an indicator of cell proliferation fraction in the tumor tissue, shows a correlation with increased FDG uptake in a variety of malignant cancers (Nguyen et al., 2007). In the present case, the Ki-67 index value was 15%; this value was slightly higher than previously reported (Cho et al., 2017; Schiergens et al., 2011; Morikawa et al., 2019). This finding could be attributed to the larger tumor size, lymph node metastasis, and increased FDG uptake. Our patient showed substantial uptake of FDG by the PSP and metastatic lymph node, which resembled malignancy, and this finding was similar to the results of a previous report (De et al., 2015). Therefore, the lymph node metastasis of PSP could be easily misdiagnosed as a malignant lesion, leading to mistreatment.

**Table 1. Review of tumor size and FDG uptake of the reported PSP cases**

| Author (year) | Age | Gender | Tumor size (cm) | SUVmax |
|---|---|---|---|---|
| Yuki Shiinal et al.(2014) | 34 | F | 5.0 | 5.5 |
| Hideyuki Maeda et al. (2015) | 26 | M | 4.0 | 4.7 |
| G. De Luca et al. (2015) | 55 | F | 2.2 | 3.7 |
| Jung-Hwan et al.(2016) | 36 | F | 3.0 | 3.9 |
| Alex Cheen Hoe Khoo et al.(2017) | 60 | F | 5.5 | 4.0 |
| Ing Xiang Soo et al. (2017) | 40 | F | 2.5 | 4.2 |
| Sujith V. Cherian et al.(2017) | 33 | F | 5.3 | 6.4 |
| Kyungsoo Bae et al. (2019) | 56 | F | 1.6 | 2.0 |
| Akriti Khanna et al.(2019) | 43 | F | 2.4 | 3.5 |

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| Takashi Sakaiet al.(2019) | 64 | F | 15.0 | 6.2 |
| Ko Leea et al. (2019) | 61 | F | 4.0 | 5.2 |
| Morikawa K et al. (2019) | 25 | F | 3.0 | 2.9 |
| Nishok David et al. (2022) | 25 | M | 2.0 | 4.5 |
| Nishok David et al. (2022) | 43 | F | 2.6 | 2.0 |
| Kocaman G et al. (2022) | 25 | F | 3.5 | 4.4 |
| Kocaman G et al. (2022) | 52 | F | 2.0 | 3.4 |
| Kocaman G et al. (2022) | 59 | M | 3.2 | 4.4 |
| Kocaman G et al. (2022) | 41 | F | 1.7 | 1.3 |
| Kocaman G et al. (2022) | 53 | F | 2.5 | 3.5 |
| Present case | 23 | F | 9.1 | 6.1 |

FDG:18F-fluorodeoxy-glucose; PSP:Pulmonary sclerosing pneumocytoma; SUVmax:the maximum standardized uptake value; M:male; F:femal.

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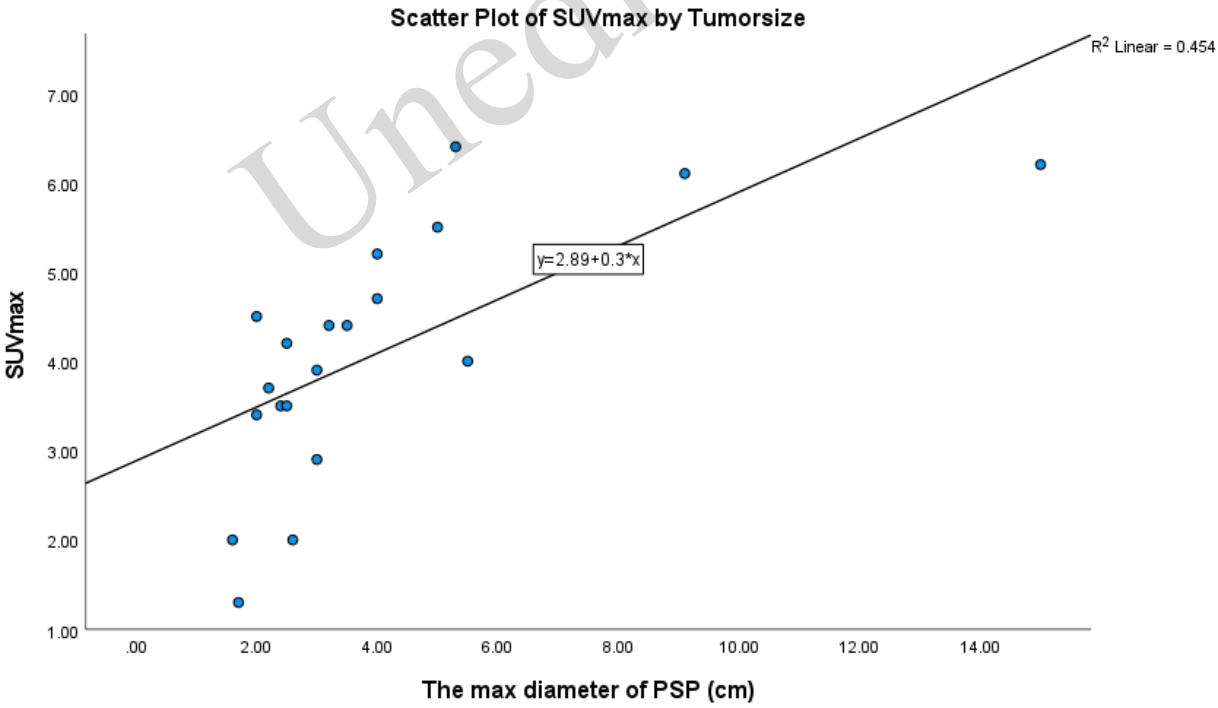


**Figure 3. Linear regression analysis showing the correlation between the maximum diameter of Pulmonary sclerosing pneumocytoma (PSP) lesions and the maximum standardized uptake value (SUVmax) among all 20 PSPs. R = 0.642, R2 = 0.454, P = 0.000.**

Lymph node metastasis and high FDG uptake were rarely reported in patients with PSP. Based on literature review and analysis, lymph node metastasis and FDG uptake were found to be correlated with tumor size in PSP patients. However, it remains unclear whether lymph node metastasis and FDG uptake are related to the age and/or gender of patients, necessitating further studies involving more PSP patients. In young patients where the lung CT scan shows an enlarged well-defined mass with no apparent necrosis and high FDG uptake in

the PET/CT scan, the possibility of PSP should be considered, and further examinations, such as fine needle aspiration and immunohistochemistry, should be performed. The prognosis of PSP patients is favorable after tumor resection, even though it may present with large lesions, lymph node metastasis, and high FDG uptake.

**Data availability statement**

Data sharing is not applicable to this article as all the data are already listed in this paper.

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**Author contributions**

Jingfeng XU, Qiuli WU provided clinical care for the patient and collected the clinical data. Jingfeng XU ,Feng CHEN and Yilei ZHAO made the diagnosis. Jingfeng XU wrote the original draft of the manuscript. Feng CHEN and Yilei ZHAO edited the final manuscript. All authors have read and agreed to the published version of the manuscript.

**Compliance with ethics guidelines**

Jingfeng XU, Yilei ZHAO, Qiuli WU and Feng CHEN declare that they have no conflict of interest.

All procedures followed were in accordance with the ethical standards of the Ethics Committee of the First Affiliated Hospital, Zhejiang University School of Medicine (IIT20250730B) and with the Helsinki Declaration of 1975, as revised in 2013. Informed consent was obtained from the subject for being included in the report.

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