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*Case Report*

# Alternative treatment modalities for pulmonary sclerosing pneumocytoma: a case report and review of the literature

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**Abstract:** Pulmonary Sclerosing pneumocytoma (PSP) is a rare lung tumour with non-specific clinical manifestations. Chest computed tomography (CT) often reveals round nodules or masses in the lung. It is often mistaken for a malignant tumor. Here a case of PSP is reported that was initially suspected to be malignant because of the imaging characteristics of the patient. It was situated at the hilum of the lung, and enhancing CT showed signs of enhancement. The patient was treated with bronchial artery embolization (BAE), which is not previously reported. Chest computed tomography showed tumor shrinkage after BAE. Surgical resection is often used to treat this disease, but this case also demonstrated the effectiveness of bronchial artery embolization.

**Keywords:** Pulmonary sclerosing pneumocytoma (PSP), bronchial artery embolization (BAE)

## Introduction

Pulmonary sclerosing pneumocytoma (PSP) is a rare pulmonary tumor, often discovered by chance and it is initially thought to be malignant [1]. It is most common among non-smoking middle-aged women in Asia. PSP usually has no clinical symptoms or specific symptoms compared to other respiratory diseases. The imaging features are isolated nodules, or multiple nodules, and there are also reports of large masses on CT [2]. It is defined histologically by two cell types: cuboidal surface cells and round stromal cells, arranged into four distinct patterns: papillary, solid, sclerosing and haemorrhagic/angiomatoid. This current patient was asymptomatic and lung nodules were only found during physical examination with chest CT. Histopathology is the only method to diagnose it. Surgical resection is the recommended definitive management. Both limited resection and lobectomy are commonly used. Bronchotomy and bronchoplasty have also been reported [3-5]. In this case, surgery was impossible for the patient because of the location of the tumor. Bronchial artery embolization was found to be effective in this case report.

## Case report

A 45-year-old woman complained of a twenty-one day history of abnormal chest computed tomography scans. Enhanced computed tomography (CT) of the chest showed a 3.8 cm × 2.4 cm mass occupying the left hilum of the lung, with a rich blood supply. It also found some small lung nodules (**Figure 1**). Positron emission tomography (PET)-CT showed a slight increase in glucose metabolism of the hilar mass of the lung, and some solid nodules in the middle lobe of the right lung and the upper lobe of the left (**Figure 2**).

Because of a nondiagnostic fiber bronchoscopy, the patient underwent a CT-guided percutaneous lung puncture biopsy and pathology examination led to a diagnosis of pulmonary sclerosing pneumocytoma (PSP) (**Figure 3**). We performed a multidisciplinary consultation with thoracic surgery and interventional radiology.

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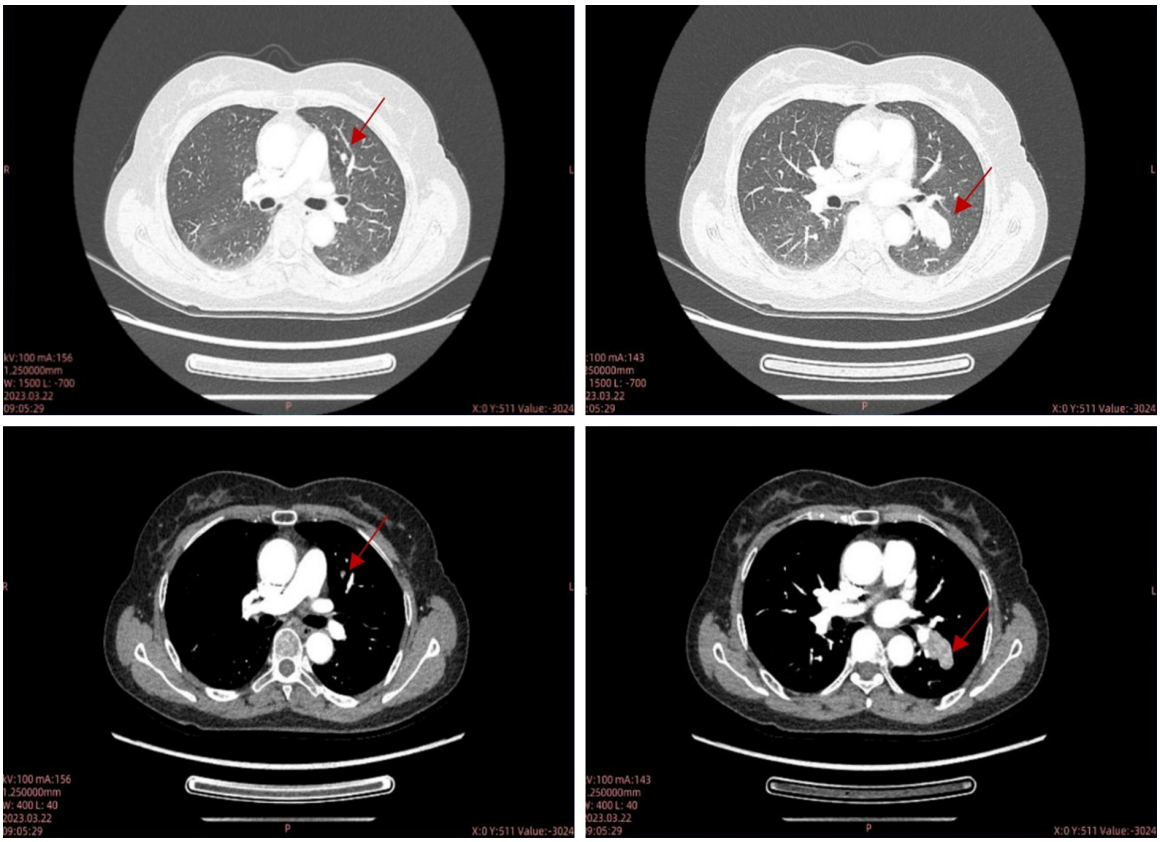

**Figure 1.** Initial chest enhanced CT scan of the lung window (top row) and mediastinal window (bottom row) revealed an occupied left hilum with smooth edges and enhancement, and a few small nodules in both lungs.

Finally, the patient was treated with bronchial artery embolization (BAE) (**Figure 4**). BAE is the injection of an embolic agent into a specific bronchial artery through a catheter to block the blood vessel for hemostasis or other therapeutic purposes. It is mainly used in the treatment of massive hemoptysis or lung tumor. Chest CT at 3 months after the therapy of BAE showed marked shrinkage (2.1 cm × 1.4 cm) (**Figure 5**).

## Discussion

PSP was first described over 60 years ago. It was originally thought to originate in the endothelial cells of blood vessels [6]. It is considered to be benign or a low-grade malignant tumor, but some of them have the ability for metastasis. Only one death has been reported so far, due to respiratory failure caused by tumor size [7].

PSP usually has no specific clinical symptoms compared to other respiratory diseases. The imaging features are isolated nodules, or multiple nodules, and there are also reports of large masses on CT [2]. The current patient was asymptomatic and only CT found lung nodules. This finding was followed up by chest enhanced CT, bronchoscopy, PET-CT, and percutaneous pulmonary biopsy in our hospital. The diagnostic procedure was similar to that for lung cancer.

Surgical resection is the main treatment for PSP. There are few reports of recurrence after surgery [8]. There are different reports of the surgical method and whether lymph node dissection is performed. Park et al. [3] found limited resection could shorten the length of hospital stay. Zheng et al. [4] found sublobectomy was superior to lobectomy. There is no clear evidence on the significance of bronchotomy and angioplasty for endobronchial tumors [5]. Radiation therapy and radiofrequency ablation are also used as alternative treatments for inoperable patients [9].

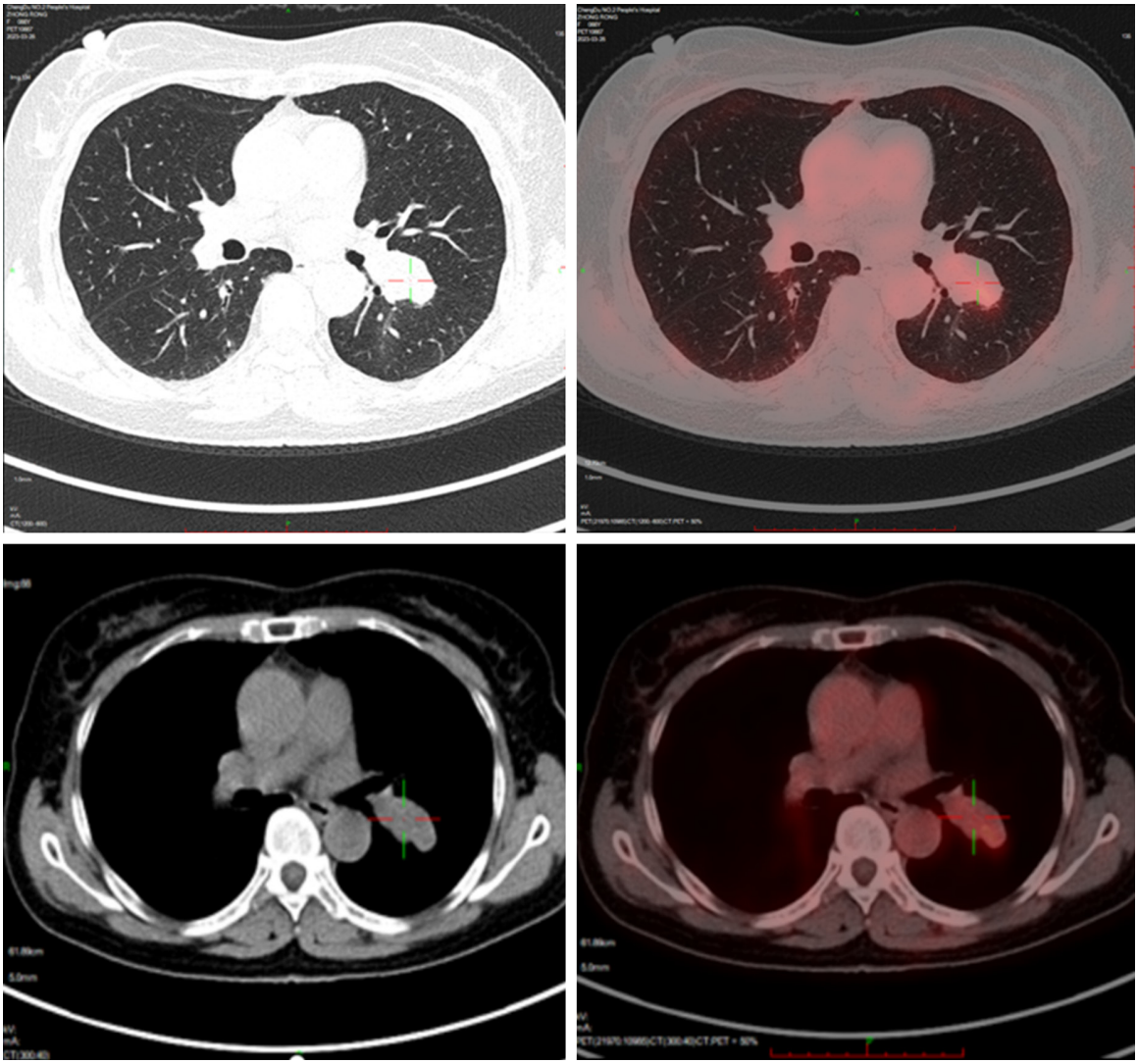

**Figure 2.** There were mildly increased mass of hilar glucose metabolism in the left lung, solid nodules in the middle lobe of the right lung and the upper lobe of the left lung, increased glucose metabolism in the mediastinum and lymph nodes in both hilar lungs.

The specific location of the tumor in this patient was not suitable for surgical resection. But bronchial artery embolization (BAE) for PSP has not been reported previously. The nodules in this patient were located in the hilum of the lung, closely related to the large blood vessels, and had the characteristics of a malignant tumor. Finally, the patient was treated with bronchial artery embolization (BAE). Chest CT at 3 months after BAE showed tumor shrinkage, but longer follow-up is needed to verify effectiveness of the treatment.

The rarity and non-specific manifestations of PSP are why it is often misdiagnosed. We report this case with PSP that had no manifestations, and lung nodules were the major imaging finding. Histopathology made a definitive diagnosis of PSP.

This report may reduce misdiagnosis of PSP. It also provides a new method to treat the disease, which has not been reported to our knowledge. But as with other treatments, long-term follow-up is needed to verify its effectiveness.

## Acknowledgements

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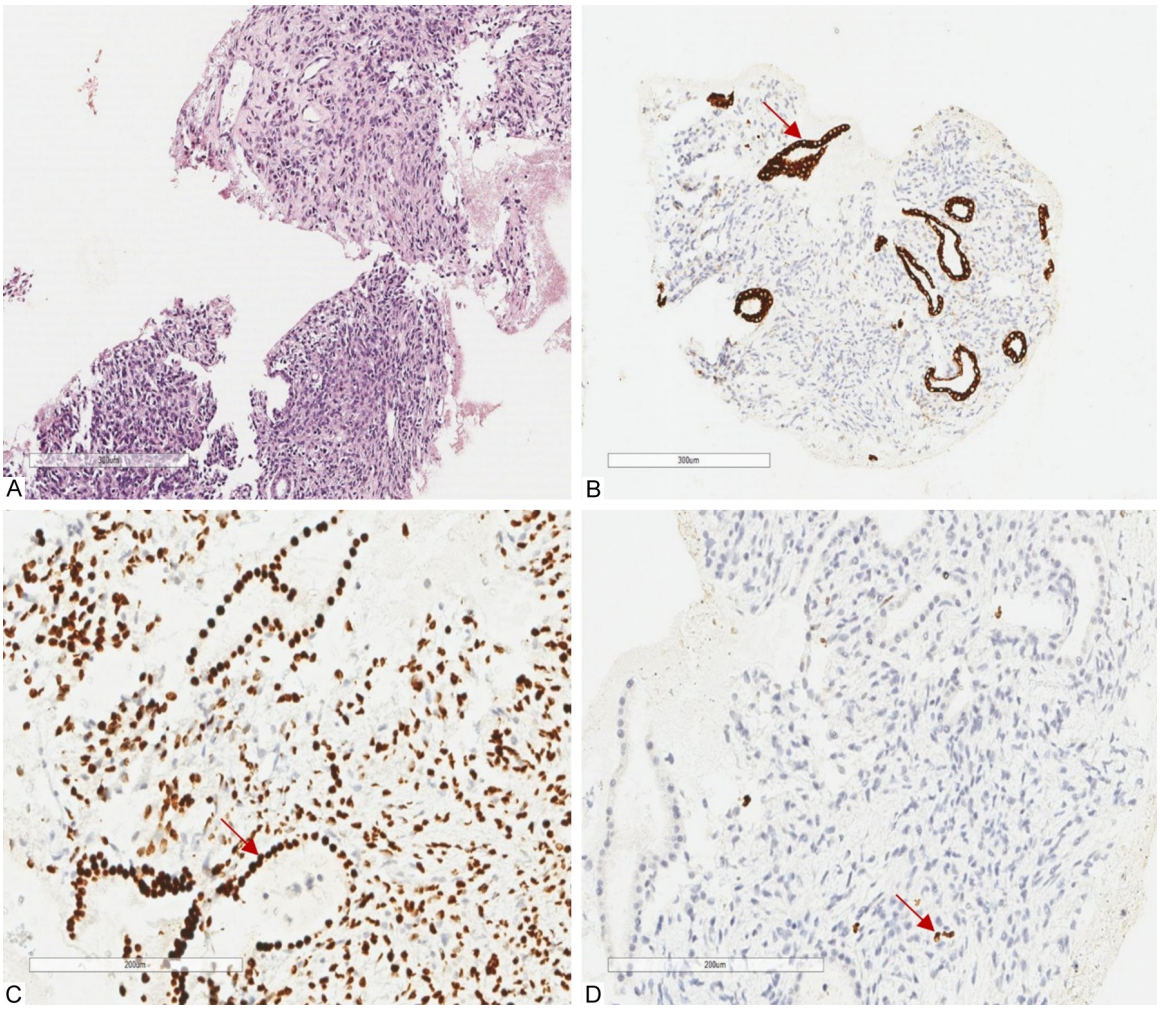


**Figure 3.** The tumor cells were distributed in a short, lamellar pattern with a focal glandular tubule structure. No mitotic figures were seen (A) ×100. The stroma had scattered mast cells and eosinophils. Immunohistochemistry showed that the tumor cells to be PCK (+) (B) ×100, TTF-1 (+) (C) ×200, Ki-67 (+) (D) ×200.

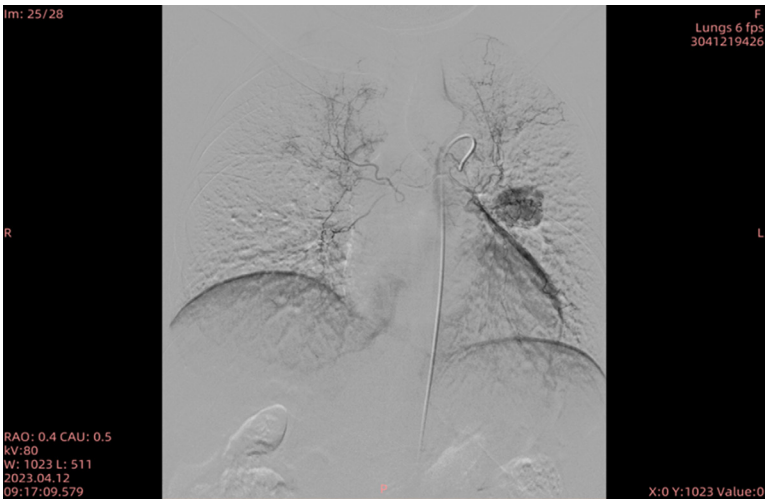


**Figure 4.** The patient underwent bronchoangiography and bronchial artery embolization (BAE).

Written informed consent was obtained from all subjects and/or their legal guardian(s).

## Disclosure of conflict of interest

We declare that we have no financial and personal relationships with other people or organizations that can inappropriately influence our work, there is no professional or other personal interest of any nature or kind in any product, service and/or company that could be construed as influencing the position presented in.

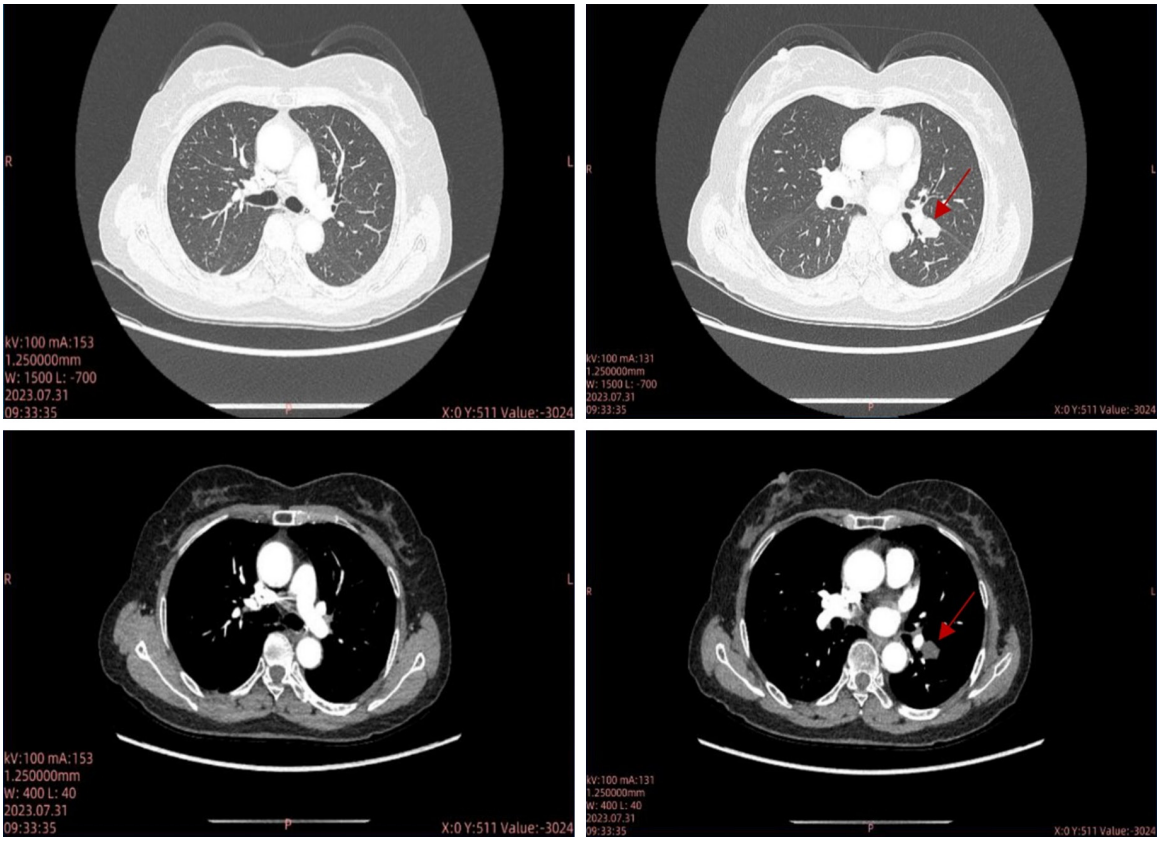


**Figure 5.** Contrast-enhanced CT scan 3 months after BAE showed that the size of the left lung nodules decreased significantly.

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## References

[1] Zhang W, Cui D, Liu Y, Shi K, Gao X and Qian R. Clinical characteristics of malignant pulmonary sclerosing pneumocytoma based on a study of 46 cases worldwide. Cancer Manag Res 2022; 14: 2459-2467.

[2] Shin SY, Kim MY, Oh SY, Lee HJ, Hong SA, Jang SJ and Kim SS. Pulmonary sclerosing pneumocytoma of the lung: CT characteristics in a large series of a tertiary referral center. Medicine (Baltimore) 2015; 94: e498.

[3] Park JS, Kim K, Shin S, Shim H and Kim HK. Surgery for pulmonary sclerosing hemangioma: lobectomy versus limited resection. Korean J Thorac Cardiovasc Surg 2011; 44: 39-43.

[4] Zheng Q, Zhou J, Li G, Man S, Lin Z, Wang T, Chen B and Lin F. Pulmonary sclerosing pneumocytoma: clinical features and prognosis. World J Surg Oncol 2022; 20: 140.

[5] Wani Y, Notohara K, Tsukayama C and Okumura N. Sclerosing hemangioma with florid endobronchial and endobronchiolar growth. Virchows Arch 2007; 450: 221-3.

[6] Liebow AA and Hubbell DS. Sclerosing hemangioma (histiocytoma, xanthoma) of the lung. Cancer 1956; 9: 53-75.

[7] Zhang W, Liu Y, Chai Y, Shi K, Chen J, Shi D and Wu X. Case report: rare pulmonary sclerosing pneumocytoma: large, multiple, metastatic, and fatal. Front Med (Lausanne) 2021; 8: 661032.

[8] Wang Q, Lu C, Jiang M, Li M, Yang X, Zhang L, He Y, Mao C, Fu P, Yang Y and Xiao H. Case report and literature review: pulmonary sclerosing pneumocytoma with multiple metastases harboring AKT1 E17K somatic mutation and TP53 C176Y germline mutation. Front Med (Lausanne) 2021; 8: 655574.

[9] Fayers RW, Lim TS and Havlat MF. Pulmonary sclerosing pneumocytoma (sclerosing haemangioma): radical radiation therapy. J Med Imaging Radiat Oncol 2016; 60: 693-695.