Journal of Zhejiang University-SCIENCE B 2026 27(2):202-206
www.jzus.zju.edu.cn; www.springer.com/journal/11585
E-mail: jzus_b@zju.edu.cn

Correspondence
https://doi.org/10.1631/jzus.B2500159

Check for updates

Pulmonary sclerosing pneumocytoma with lymph node metastasis and high ^{18}FDG uptake in PET/CT: a rare case report and literature review

Jingfeng XU[1], Yilei ZHAO[1], Qiuli WU[2], Feng CHEN[1]✉

[1]*Department of Radiology, The First Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310003, China*
[2]*Department of Pharmacy, The Second Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310009, China*

Pulmonary sclerosing pneumocytoma (PSP) was first reported by Liebow and Hubbell (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 the potential to metastasize. PSP shows varying levels of ^{18}F-fluorodeoxy-glucose (^{18}FDG) accumulation, with the maximum standardized uptake value (SUV_{max}) 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 high ^{18}FDG uptake on positron emission tomography/computed tomography (PET/CT) scan, and an SUV_{max} 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 an unremarkable medical history. The serum levels of carcinoembryonic antigen, squamous cell carcinoma antigen, cancer antigen 125 (CA125), CA199, 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 cm×5.5 cm×5.2 cm mass in the right upper lobe. Tumor localization was visually identified in CT images (Figs. 1a and 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 cm×1.0 cm×0.7 cm. No associated mediastinal lymph node enlargement was detected.

We then conducted a PET/CT scan and detected a high ^{18}FDG accumulation within the mass, with an SUV_{max} of 6.1. The lymph node in the right lung hilar showed an SUV_{max} of 5.1 (Figs. 1c and 1d).

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

✉ Feng CHEN, chenfenghz@zju.edu.cn
Feng CHEN, https://orcid.org/0000-0003-4402-4955
Jingfeng XU, https://orcid.org/0000-0003-4080-3433

Received Mar. 30, 2025; Revision accepted May 19, 2025; Crosschecked Jan. 4, 2026

bland nuclei and eosinophilic cytoplasm (Figs. 2a and 2b). Immunohistochemical staining revealed that the tumor and right hilar lymph node were positive for thyroid transcription factor-1 (TTF-1) (Fig. 2c) and cytokeratin 7 (CK7) (Fig. 2d) and negative for cluster of differentiation 31 (CD31) and CD34. The Ki-67 positivity rate was 15%.

PSP typically occurs in middle-aged adults, with a female-to-male ratio of 7.5:1. It has a relatively high incidence in Asia and accounts for 5.8% of surgically resected benign tumors in Japan (Masuda et al., 2018).

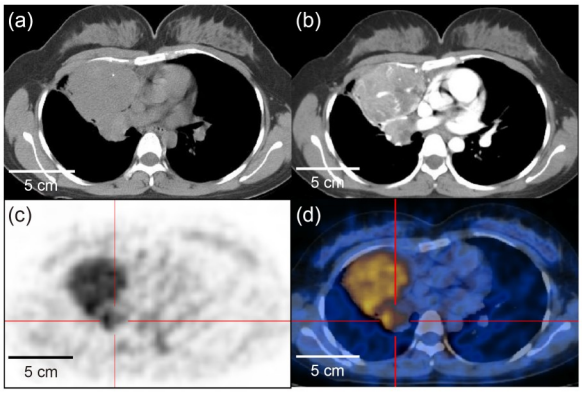

Fig. 1 Chest CT and PET/CT images of the patient. (a, b) Chest CT axial scans: plain (a) and enhanced (b), showing the right upper lobe tumor. (c, d) The corresponding PET scan (c) and fused PET/CT scan (d) images show a relatively high uptake of ^{18}FDG in the tumor (SUV_{max}=6.1) and lymph node (SUV_{max}=5.1). CT: computed tomography; PET: positron emission tomography; ^{18}FDG: ^{18}F-fluorodeoxyglucose; SUV_{max}: the maximum standardized uptake value.

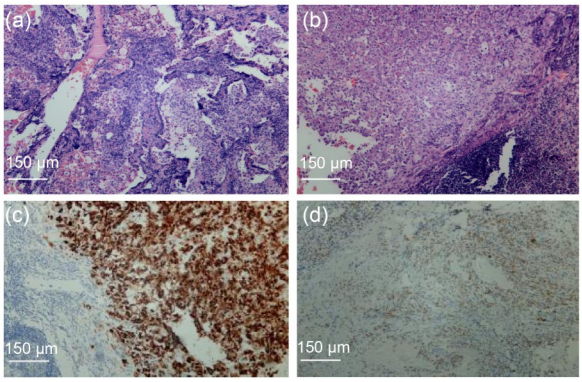

Fig. 2 Pathological results with evidence of PSP. (a, b) Histopathological examination with hematoxylin and eosin (H&E) staining of the right upper lobe lesion (a) and right hilar lymph node metastasis (b). (c, d) Immunohistochemical staining revealed that the right hilar lymph node metastasis was positive for thyroid transcription factor-1 (TTF-1) (c) and cytokeratin 7 (CK7) (d). PSP: pulmonary sclerosing pneumocytoma.

The most common form is an asymptomatic solitary pulmonary mass. PSP-related symptoms include hemoptysis, chronic cough, and chest pain. 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, and 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 as a rare and benign form of lung tumor; PSP with lymph node metastasis is extremely rare. 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 (Yano et al., 2002; Kim et al., 2003; Miyagawa-Hayashino et al., 2003; Katakura et al., 2005; Chien et al., 2009; Pokharel et al., 2016), bone, mediastinum, and pleura (Kim et al., 2015; Pokharel et al., 2016; Arumugam et al., 2017). Thus, PSP is not always benign and has the 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 (Sun et al., 2012; Pokharel et al., 2016). 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 in the current case report was diagnosed with 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 high ^{18}FDG uptake. Because the tumor mass had an 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 a 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 surgery. The lesions and

hilar lymph nodes were finally diagnosed as PSP based on immunohistochemical analysis. The patient was followed for eight years, and no tumor was 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 PSP patients. In most cases, 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 ^{18}FDG accumulation, with the SUV_{max} ranging from the background value to 6.4 (Satoh et al., 1989; Jiang et al., 2018). Furthermore, the tumor size of PSP was shown to be positively correlated with the SUV_{max} (Xu et al., 2019). We searched and reviewed all articles published in English in the PubMed database on PSPs with pathological confirmation and ^{18}FDG PET/CT findings. Nineteen PSP tumors were described in 14 reports published in the last ten years. The relationship between tumor size and ^{18}FDG uptake is summarized in Table 1 and Fig. 3. In previous reports, ^{18}FDG uptake of the tumor was positively correlated with tumor size. However, some authors (Travis et al., 2015) have 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 tumor tissue, shows a correlation with increased ^{18}FDG uptake in a variety of malignant cancers (Nguyen et al., 2007). In the present case, the Ki-67 index value was 15%, which was slightly higher than that previously reported (Schiergens et al., 2011; Cho et al., 2017; Morikawa et al., 2019). This finding could be attributed to the larger tumor size, greater degree of lymph node metastasis, and increased ^{18}FDG uptake. Our patient showed substantial uptake of ^{18}FDG by the PSP and metastatic lymph node, which resembled malignancy, and this finding was similar to the results of a previous report (de Luca 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 ^{18}F-fluorodeoxy-glucose (^{18}FDG) uptake of the reported pulmonary sclerosing pneumocytoma (PSP) cases

Reported case	Age (years)	Gender	Tumor size (cm)	SUV_{max}
Shiina et al. (2014)	34	F	5.0	5.5
Maeda et al. (2015)	26	M	4.0	4.7
de Luca et al. (2015)	55	F	2.2	3.7
Lim et al. (2016)	36	F	3.0	3.9
Khoo et al. (2017)	60	F	5.5	4.0
Soo et al. (2017)	40	F	2.5	4.2
Cherian et al. (2017)	33	F	5.3	6.4
Bae et al. (2019)	56	F	1.6	2.0
Khanna et al. (2019)	43	F	2.4	3.5
Sakai et al. (2019)	64	F	15.0	6.2
Lee et al. (2019)	61	F	4.0	5.2
Morikawa et al. (2019)	25	F	3.0	2.9
David et al. (2022)	25	M	2.0	4.5
	43	F	2.6	2.0
Kocaman et al. (2021)	25	F	3.5	4.4
	52	F	2.0	3.4
	59	M	3.2	4.4
	41	F	1.7	1.3
	53	F	2.5	3.5
Present case	23	F	9.1	6.1

SUV_{max}: the maximum standardized uptake value; M: male; F: female.

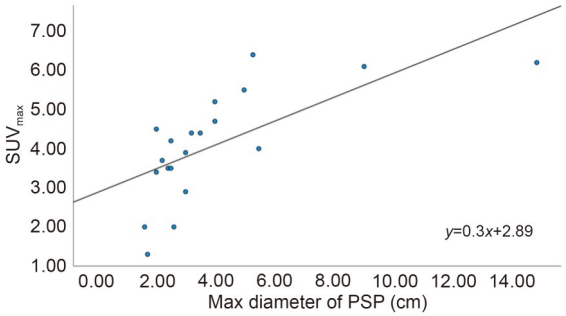

Fig. 3 Linear regression analysis showing the correlation between the maximum diameter of pulmonary sclerosing pneumocytoma (PSP) lesions and the maximum standardized uptake value (SUV_{max}) among all 20 PSP lesions. R=0.642, R^2=0.454, P<0.001.

Lymph node metastasis and high ^{18}FDG uptake were rarely reported in patients with PSP. Based on literature review and analysis, lymph node metastasis and ^{18}FDG uptake were found to be correlated with tumor size in PSP patients. However, it remains unclear whether lymph node metastasis and ^{18}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 or high ^{18}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 these patients may present with large lesions, lymph node metastasis, and high ^{18}FDG uptake.

Data availability statement

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

Acknowledgments

This study was supported by the Zhejiang Province Public Welfare Technology Application Research Project (No. LGF19H220003), China.

Author contributions

Jingfeng XU and 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 approved the final manuscript, and therefore, have full access to all the data in the study and take responsibility for the integrity and security of the data.

Compliance with ethics guidelines

Jingfeng XU, Yilei ZHAO, Qiuli WU, and Feng CHEN declare that they have no conflicts 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 (Approval No. IIT20250730B) and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Informed consent was obtained from the patient for being included in the study.

Declaration on the use of generative AI tools

No generative AI tools were used in the preparation of this manuscript.

References

Arumugam VG, Joseph LD, Thangavel P, et al., 2017. Sclerosing pneumocytoma of the lung: a case report. *J Clin Diagn Res*, 11(2):ED12-ED14. https://doi.org/10.7860/JCDR/2017/22279.9271

Bae K, Song DH, Jeon KN, et al., 2019. Pulmonary sclerosing pneumocytoma presenting a peritumoral halo and an intervening lucent zone on computed tomography: radiology–pathology correlation. *Thorac Cancer*, 10(5): 1295-1296. https://doi.org/10.1111/1759-7714.13069

Chen QY, Wu LJ, Hu HZ, et al., 2012. A case of pulmonary sclerosing hemangioma with low ^{18}FDG uptake in PET. *Oncol Lett*, 3(3):646-648. https://doi.org/10.3892/ol.2011.5248492317727668

Cherian SV, Estrada-Y-Martin RM, Irani AD, et al., 2017. A 33-year-old woman with a fluorodeoxyglucose-avid left lower lobe mass. *Chest*, 151(5):e119-e122. https://doi.org/10.1016/j.chest.2016.10.049

Chien NC, Lin CW, Tzeng JE, 2009. Sclerosing haemangioma with lymph node metastasis. *Respirology*, 14(4):614-616. https://doi.org/10.1111/j.1440-1843.2009.01523.x

Cho HJ, Lee JH, Lee GK, et al., 2017. Case of sclerosing pneumocytoma combined with a typical carcinoid and pulmonary adenocarcinoma in different lobes. *Thorac Cancer*, 8(4):372-375. https://doi.org/10.1111/1759-7714.12449

David N, Chandy ST, Benjamin SR, et al., 2022. Pulmonary sclerosing pneumocytoma—a case series. *Indian J Thorac Cardiovasc Surg*, 38(2):167-172. https://doi.org/10.1007/s12055-021-01269-z

de Luca G, Martucci N, Setola S, et al., 2015. Sclerosing hemangioma of the lung mimicking pulmonary metastasis. *Lung*, 193(3):447-448. https://doi.org/10.1007/s00408-015-9711-0

Devouassoux-Shisheboran M, Hayashi T, Linnoila RI, et al., 2000. A clinicopathologic study of 100 cases of pulmonary sclerosing hemangioma with immunohistochemical studies: TTF-1 is expressed in both round and surface cells, suggesting an origin from primitive respiratory epithelium. *Am J Surg Pathol*, 24(7):906-916. https://doi.org/10.1097/00000478-200007000-00002

Jiang L, Huang Y, Tang QY, et al., 2018. ^{18}F-FDG PET/CT characteristics of pulmonary sclerosing hemangioma vs. pulmonary hamartoma. *Oncol Lett*, 16(1):660-665. https://doi.org/10.3892/ol.2018.8660

Katakura H, Sato M, Tanaka F, et al., 2005. Pulmonary sclerosing hemangioma with metastasis to the mediastinal lymph node. *Ann Thorac Surg*, 80(6):2351-2353. https://doi.org/10.1016/j.athoracsur.2004.06.099

Khanna A, Alshabani K, Mukhopadhyay S, et al., 2019. Sclerosing pneumocytoma: case report of a rare endobronchial presentation. *Medicine*, 98(15):e15038. https://doi.org/10.1097/MD.0000000000015038

Khoo ACH, Hamzah F, Ong CK, 2017. Incidental sclerosing pneumocytoma detected on bone scintigraphy. *Clin Nucl*

Med, 42(1):e77-e79. https://doi.org/10.1097/RLU.0000000000001371

Kim KH, Sul HJ, Kang DY, 2003. Sclerosing hemangioma with lymph node metastasis. *Yonsei Med J*, 44(1):150-154. https://doi.org/10.3349/ymj.2003.44.1.150

Kim MK, Jang SJ, Kim YH, et al., 2015. Bone metastasis in pulmonary sclerosing hemangioma. *Korean J Intern Med*, 30(6):928-930. https://doi.org/10.3904/kjim.2015.30.6.928

Kocaman G, Yenigün MB, Ersöz CC, et al., 2021. Pulmonary sclerosing pneumocytoma with mediastinal lymph node metastasis: a case report. *Gen Thorac Cardiovasc Surg*, 69(1):142-146. https://doi.org/10.1007/s11748-020-01431-1

Lee K, Sasaki M, Kakimoto T, et al., 2019. Pulmonary sclerosing pneumocytoma demonstrating intratumoral hematoma. *Respir Med Case Rep*, 28:100870. https://doi.org/10.1016/j.rmcr.2019.100870

Liebow AA, Hubbell DS, 1956. Sclerosing hemangioma (histiocytoma, xanthoma) of the lung. *Cancer*, 9(1):53-75. https://doi.org/10.1002/1097-0142(195601/02)9:1<53::aid-cncr2820090104>3.0.co;2-u

Lim JH, Lee N, Choi DW, et al., 2016. Pulmonary sclerosing pneumocytoma mimicking lung cancer: case report and review of the literature. *Thorac Cancer*, 7(4):508-511. https://doi.org/10.1111/1759-7714.12341

Maeda H, Kanzaki M, Isaka T, et al., 2015. Video-assisted thoracoscopic enucleation after congenital cardiac surgery. *J Surg Case Rep*, 2015(9):rjv123. https://doi.org/10.1093/jscr/rjv123

Masuda M, Endo S, Natsugoe S, et al., 2018. Thoracic and cardiovascular surgery in Japan during 2015. *Gen Thorac Cardiovasc Surg*, 66(10):581-615. https://doi.org/10.1007/s11748-018-0968-0

Miyagawa-Hayashino A, Tazelaar HD, Langel DJ, et al., 2003. Pulmonary sclerosing hemangioma with lymph node metastases: report of 4 cases. *Arch Pathol Lab Med*, 127(3):321-325. https://doi.org/10.5858/2003-127-0321-pshwln

Morikawa K, Misumi S, Fukuda T, et al., 2019. Pulmonary sclerosing pneumocytoma presenting as slow-growing multiple nodules over a long period. *Radiol Case Rep*, 14(5):602-607. https://doi.org/10.1016/j.radcr.2019.02.024

Nguyen XC, Lee WW, Chung JH, et al., 2007. FDG uptake, glucose transporter type 1, and Ki-67 expressions in non-small-cell lung cancer: correlations and prognostic values. *Eur J Radiol*, 62(2):214-219. https://doi.org/10.1016/j.ejrad.2006.12.008

Pokharel S, Dhillon SS, Ylagan L, et al., 2016. Sclerosing pneumocytoma with lymph node metastasis. *J Thorac Oncol*, 11(10):1802-1804. https://doi.org/10.1016/j.jtho.2016.06.005

Satoh Y, Tsuchiya E, Weng SY, et al., 1989. Pulmonary sclerosing hemangioma of the lung. A type II pneumocytoma by immunohistochemical and immunoelectron microscopic studies. *Cancer*, 64(6):1310-1317. https://doi.org/10.1002/1097-0142(19890915)64:6<1310::aid-cncr2820640624>3.0.co;2-s

Sakai T, Miyoshi T, Umemura S, et al., 2019. Large pulmonary sclerosing pneumocytoma with massive necrosis and vascular invasion: a case report. *Oxf Med Case Reports*, 2019(7):omz066. https://doi.org/10.1093/omcr/omz066

Schiergens TS, Khalil PN, Mayr D, et al., 2011. Pulmonary sclerosing hemangioma in a 21-year-old male with metastatic hereditary non-polyposis colorectal cancer: report of a case. *World J Surg Oncol*, 9:62. https://doi.org/10.1186/1477-7819-9-62

Shiina Y, Sakairi Y, Wada H, et al., 2018. Sclerosing pneumocytoma diagnosed by preoperative endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA). *Surg Case Rep*, 4:20. https://doi.org/10.1186/s40792-018-0429-0

Soo IX, Sittampalam K, Lim CH, 2017. Pulmonary sclerosing pneumocytoma with mediastinal lymph node metastasis. *Asian Cardiovasc Thorac Ann*, 25(7-8):547-549. https://doi.org/10.1177/021

Sun Y, Zhou LX, Zhao M, et al., 2012. Histogenesis of pulmonary sclerosing hemangioma. *Chin J Pathol*, 41(4):239-242 (in Chinese). https://doi.org/10.3760/cma.j.issn.0529-5807.2012.04.006

Tanaka I, Inoue M, Matsui Y, et al., 1986. A case of pneumocytoma (so-called sclerosing hemangioma) with lymph node metastasis. *Jpn J Clin Oncol*, 16(1):77-86. https://doi.org/10.1093/oxfordjournals.jjco.a039119

Travis WD, Brambilla E, Nicholson AG, et al., 2015. The 2015 World Health Organization classification of lung tumors: impact of genetic, clinical and radiologic advances since the 2004 classification. *J Thorac Oncol*, 10(9):1243-1260. https://doi.org/10.1097/JTO.0000000000000630

Xu J, Dong YW, Yin GT, et al., 2019. ^{18}F-FDG PET/CT imaging: a supplementary understanding of pulmonary sclerosing pneumocytoma. *Thorac Cancer*, 10(7):1552-1560. https://doi.org/10.1111/1759-7714.13100

Yano M, Yamakawa Y, Kiriyama M, et al., 2002. Sclerosing hemangioma with metastases to multiple nodal stations. *Ann Thorac Surg*, 73(3):981-983. https://doi.org/10.1016/s0003-4975(01)03122-8