

Multiple pulmonary sclerosing hemangiomas (pneumocytoma) mimicking lung metastasis detected in fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography

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ABSTRACT

Pulmonary sclerosing hemangioma (PSH), or the alternative name of “sclerosing pneumocytoma,” is a rare benign neoplasm. PSH is often asymptomatic and presents as a solitary or multiple pulmonary nodules on radiologic imaging studies. Few articles have been reported to describe the fluorodeoxyglucose positron emission tomography (FDG PET) findings about PSH. The authors describe an interesting but uncommonly encountered cause of false positive FDG PET scan in the thorax in a 25-year-old woman, a known case of arteriovenous malformation of oral cavity who underwent embolization and presented with incidental detection of bilateral lung nodules. She is asymptomatic and is on follow-up.

Keywords: 18-F fluorodeoxyglucose uptake, bilateral pulmonary nodules, metastasis, pulmonary sclerosing hemangioma

INTRODUCTION

Pulmonary sclerosing hemangioma (PSH) is a relatively rare benign tumor, which was first described by Liebow and Hubbell in 1956.^[1] Most patients are asymptomatic, with less than one-third experiencing hemoptysis, coughs, or chest pain.^[2] Most cases of PSH appear on computed tomography (CT) scans of the chest as a solitary round or oval well-defined nodule. Multiple nodules reportedly account for only 4% of all cases.^[3] Although the CT scan features of these nodules were compatible with the differential diagnosis of sclerosing hemangioma, multiple nodules and increased uptake on fluorodeoxyglucose positron emission tomography (FDG-PET) scan raised the suspicion of malignancy.

CASE REPORT

The present case report is about a 25-year-old female patient, already a known case of Arteriovenous malformation of oral cavity and she underwent embolization of the lesion, presented with incidental detection of bilateral multiple pulmonary nodules on CT scan. CT showed round or oval pulmonary nodules, with smooth margin associated with marked enhancement. She was referred for whole body positron emission tomography/computed tomography (PET/CT) for detection of primary site and to differentiate between benign and malignant nodules. PET/CT showed an intense uptake in the bilateral pulmonary nodules [Figure 1] with largest nodule located in right lower lobe with a standardized uptake value (SUV_{max}) of 10.5. Some of the lesions did not show FDG uptake patient underwent minithoracotomy and wedge biopsy of the lesion [Figure 2] and biopsy confirmed PSH [Figure 3]. A diagnosis of bilateral multiple PSH was made and she was advised follow-up.

DISCUSSION

PSH is a rare benign neoplasm, first described in 1956.^[1] Recent

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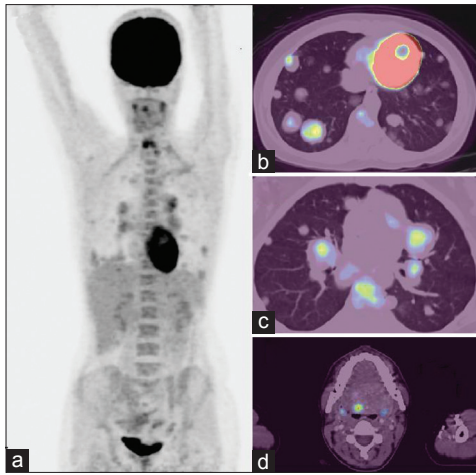


Figure 1: Whole body fluorodeoxyglucose-positron emission tomography/computed tomography (PET/CT) maximum intensity projection image (a), axial fused PET/CT (b and c) showed an intense uptake in bilateral round enhancing nodular lesions in the lungs and (d) showing uptake in the sclerosing hemangioma in the sublingual region

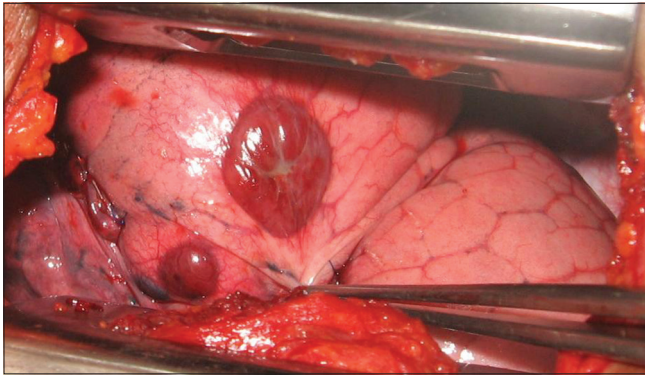


Figure 2: Intra-operative wedge biopsy picture showing multiple sclerosing hemangioma in the lungs

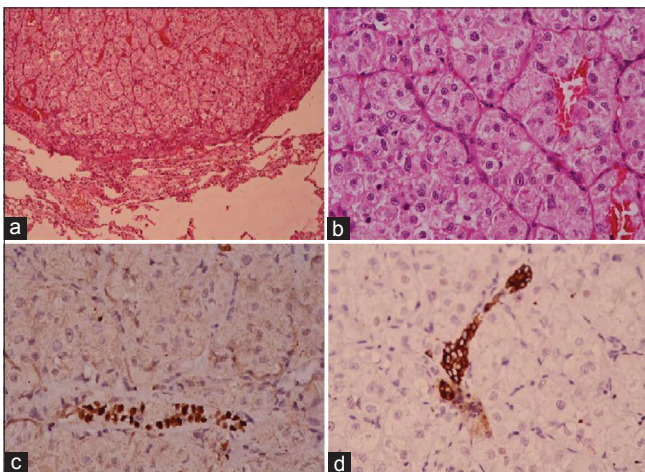


Figure 3: (a) Histology showing lung parenchyma with well-circumscribed solid nodules (X and E, ×10). (b) Microscopically, solid nodules with nests and trabeculae of large polygonal cells with vesiculae nuclei and eosinophilic granular chromatin (X and E, ×40). Immunohistochemistry showing (c) thyroid transcription factor 1st (d) Pan-cytokeratin negative in tumor cells and positive in entrapped alveoli

studies suggested that PSH arises from type II pneumocyte and multipotential primitive respiratory epithelium.^[2] Thus, the alternative name of “sclerosing” has been proposed. This tumor affects more frequently middle-aged women in the Far Eastern individuals.^[3] Most cases are discovered incidentally with a solitary lesion. However, cases with multiple lesions or nodal metastases have also been reported. The radiologic and CT imaging features of the PSH are reported as a solitary, well-defined, round or oval mass, and often showing good contrast enhancement.^[4] Because of its rarity, the natural course of multiple PSHs has not been well-understood. FDG PET has been shown to be more accurate than contrast enhanced CT in differentiating malignant from benign pulmonary nodule. However, false-positive results occur. To the best of our knowledge, only few case reports had reported F18-FDG PET of PSH.^[5,6] The gross and histopathological findings of SH are well-described in the literature. The co-existence of chronic inflammation along with other common microscopic findings could be a possible factor causing SH to be FDG avid on PET scans.^[7] Benign and slow-growing tumors usually showed low glucose metabolism. However, some PSH may exceed greater than 5 cm, even though mitotic figures are rarely seen. These larger PSH probably have more active cell proliferation which may lead to higher FDG uptake; or may have more cell components, which are responsible for higher FDG uptake than smaller tumors. Therefore, larger PSH will be misreading as a malignant neoplasm.^[8] Previous reports have indicated that, although rapid progression might occur in a solitary PSH, multiple PSHs tend to be slow-growing, another case study described multiple PSHs that were stable over 10 years.^[9] We also followed-up our case as she is asymptomatic for lung lesions. Our case report demonstrates 18F-FDG PET/CT uptake in bilateral multiple PSH. The possibility of false positive result caused by PSH should be made aware of when using FDG PET for diagnosis of malignant pulmonary neoplasm.

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