Multiple FDG-avid sclerosing hemangiomas mimicking pulmonary metastases in a case of soft tissue sarcoma

Nilendu C. Purandare a, Sumeet G. Du a, Sneha Shah a, Anshu R. Sharma a, Pallavi V. Suryawanshi b and Venkatesh Rangarajan a

Bioimaging Unit, Department of Pathology, Tata Memorial Hospital, E. Borges Road, Parel, Mumbai, Maharashtra 400012, India

Corresponding address: Venkatesh Rangarajan, Bioimaging Unit, E. Borges Road, Parel, Mumbai, Maharashtra 400012, India. Email: drvrangara@gmail.com

Date accepted for publication 9 July 2010

Abstract

Non-neoplastic lesions have been known to mimic malignancies and metastases on positron emission tomography/computed tomography. We report the rare occurrence of multiple fluorodeoxyglucose-concentrating sclerosing hemangiomas in a patient with soft tissue sarcoma mimicking lung metastases.

Keywords: Sclerosing hemangioma; PET/CT; soft tissue sarcoma metastasis.

Introduction

Morphologically sclerosing hemangiomas can mimic metastases in a patient with a known primary neoplasm. Lack of sufficient data on the fluorodeoxyglucose (FDG) uptake of sclerosing hemangioma confounds the problem even further. We report the occurrence of multiple FDG-avid sclerosing hemangiomas, which by virtue of their multiplicity and avidity, were misinterpreted as pulmonary metastases in a patient with a soft tissue sarcoma of the hand.

Case report

A 37-year-old female was referred with complaints of swelling on the dorsal aspect of the right hand for 2 months. A biopsy report of a synovial sarcoma (type of soft tissue sarcoma) was followed by a staging positron emission tomography (PET)/computed tomography (CT) scan as per institution protocol for soft tissue malignancies. In addition to hypermetabolism (maximum standardized uptake value (SUV max) 8.1) at the site of the primary tumor in the right hand (arrows in Fig. 1a,b), a few FDG concentrating foci were noted in the chest (arrows in Fig. 2a,d,g). These foci localized to soft tissue nodules of varying sizes in the lungs on the CT component of the PET/CT study (arrows in Fig. 2b,e,h) which was confirmed on the fusion images (arrows in Fig. 2c,f,i). The SUV max of the nodules ranged from 4.1 to 4.4. In view of the primary diagnosis of a soft tissue sarcoma, the lung nodules were considered to be metastatic in nature. A CT-guided biopsy was performed from 2 nodules, one in each lung to confirm the etiology. Histologic examination of the biopsy specimens revealed cuboidal cells arranged in papillary fronds with focal lymphocytic infiltrates (Fig. 3). On immunohistochemistry the cuboidal cells expressed CK 7. In accordance with the histopathology report the lung nodules were diagnosed as sclerosing hemangiomas and the patient was treated with curative intent for the primary tumor. She is being followed up, and the lung lesions have remained unchanged for 1 year.

Discussion

Combined FDG-PET/CT is being used increasingly in staging and assessing treatment response in patients with soft tissue sarcoma[1]. As many as 20% of adult
Figure 1  Coronal PET (a) and coronal fusion PET/CT (b) images show increased FDG uptake at the site of the primary soft tissue sarcoma in the right hand (arrows).

Figure 2  Axial PET (a,g) and coronal PET (d) images show FDG-avid foci in the lungs (arrows). Axial CT (b,h) and coronal reformatted (e) images in lung window show bilateral lung nodules (arrows) corresponding to the FDG-avid foci. Fusion PET/CT images (c,f,i) confirm the findings seen in the PET and CT images.
patients with an extremity soft tissue sarcoma can have lung metastasis during the course of the disease\textsuperscript{[2]}. Most lung metastases appear as well-demarcated nodules surrounded by the lung parenchyma on the CT component of the study, and often show FDG uptake in the fusion and PET images. Such a finding is considered a surrogate marker for metastatic dissemination of the disease to the lungs. However, certain benign and inflammatory conditions can occasionally mimic this appearance and thereby produce a false-positive result on FDG-PET/CT altering patient management.

Sclerosing hemangioma (SH) is an uncommon pulmonary neoplasm. Since the first report of SH in 1956 by Leibow and Hubbell, its origin has been variously ascribed to vascular, mesothelial, mesenchymal, epithelial and neuroendocrine precursors. In the largest series of immunohistochemical analysis of SHs, the authors attributed its origin to primitive respiratory epithelium by virtue of TIFF-1 expression\textsuperscript{[3]}. Most SHs are asymptomatic and are incidentally detected in middle-aged females (M/F ratio 5:1) on chest radiographs as a solitary lung nodule\textsuperscript{[4]}. They can be multiple in 3–4% of cases\textsuperscript{[3]}. On CT scans, an SH appears as a well-marginated nodule that enhances on contrast administration\textsuperscript{[5]}. Morphological features such as shape, margin, attenuation and calcification seen on unenhanced CT scans can help narrow the differential diagnosis of lung nodules. Spiculated and lobulated margins usually favour malignancy, whereas nodules with smooth margins, calcification and a homogenous attenuation are benign. Enhancement of benign nodules such as SH on contrast-enhanced CT is often similar to that of vascular lung metastases and can add to the diagnostic dilemma. Dynamic CT characteristics coupled with morphological features have been used to differentiate benign lesions such as SH from malignant lung nodules\textsuperscript{[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\textsuperscript{[7]} could be a possible factor causing SH to be FDG avid on PET scans. There are just a handful of reports so far describing PET findings in an SH of the lung\textsuperscript{[8–14]} showing moderate FDG uptake. The higher metabolic uptake in the nodules in our case (SUV\textsubscript{max} ranging from 4.1 to 4.4), which also showed the morphologic characteristics of metastases on CT images, raised a very strong suspicion of lung metastases. Other than a few cases showing nodal metastases\textsuperscript{[15]}, SHs are largely considered benign with an excellent prognosis following surgical resection\textsuperscript{[7]}

The combination of high FDG uptake and a CT morphology suggestive of metastases can occasionally be misleading. The rare occurrence of multiple SHs in a patient with a known primary tumor can cause false-positive results by virtue of avid FDG uptake, as depicted in our case. This is of particular relevance in malignancies such as bone and soft tissue sarcomas, which have a high propensity for lung metastases; pulmonary nodules detected on imaging are presumed to be metastatic often without histological confirmation. All the cases reported so far in the literature have shown FDG avidity in an SH presenting as a solitary pulmonary nodule, whereas our report describes a patient with primary soft tissue sarcoma with multiple SHs presenting as FDG-avid lung nodules mimicking pulmonary metastases. It remains to be seen whether the varying intensity of FDG uptake can be correlated with histology of SHs, an apparently benign tumor but with the potential to cause false-positive findings on a FDG-PET/CT examination.

References

\textsuperscript{[1]} Piperkova E, Mikhail M, Mousavi A, \textit{et al.} Impact of PET and CT in PET/CT studies for staging and evaluating treatment response in bone and soft tissue sarcomas. Clin Nucl Med 2009; 34: 146–50. doi:10.1097/RLU.0b013e318196609d. PMid:19352275.

\textsuperscript{[2]} Gadd MA, Casper ES, Woodruff JM, McCormack PM, Brennan MF. Development and treatment of pulmonary metastases in adult patients with extremity soft tissue sarcoma. Ann Surg Oncol 1993; 218: 705–12. doi:10.1097/00000658-199312000-00002. PMid:8257219.

\textsuperscript{[3]} Devoissouxs-Shisheboran M, Hayashi T, Linoila RI, Koss MN, Travis WD. 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 2000; 24: 906–16. doi:10.1097/00000478-200007000-00002. PMid:10895813.

\textsuperscript{[4]} Haaga J, Lanzier CF, Gilkeson RC. CT and MR imaging of the whole body; Vol 1, 4th ed. St. Louis, MO: Mosby; 2003.

\textsuperscript{[5]} Chung MJ, Lee KS, Han J, Sung YM, Chong S, Kwon OJ. Pulmonary sclerosing hemangioma presenting as solitary pulmonary nodule: dynamic CT findings and histopathologic comparisons. Am J Roentgenol 2006; 187: 430–7. doi:10.2214/AJR.05.0460. PMid:16861548.

\textsuperscript{[6]} Yi CA, Lee KS, Kim EA, \textit{et al.} Solitary pulmonary nodules: dynamic enhanced multidetector row CT study and comparison with vascular endothelial growth factor and microvessel density. Radiology 2004; 233: 191–9. doi:10.1148/radiol.2331031535. PMid:15304661.
Keylock JB, Galvin JR, Franks TJ. Sclerosing hemangioma of the lung. Arch Pathol Lab Med 2009; 133: 820–5.

Chang JM, Lee HJ, Goo JM, et al. False positive and false negative FDG-PET scans in various thoracic diseases. Korean J Radiol 2006; 7: 57–69. doi:10.3348/kjr.2006.7.1.57. PMid: 16549957.

Hara M, Iida A, Tohyama J, et al. FDG-PET findings in sclerosing hemangioma of the lung: a case report. Radiat Med 2001; 19: 215–8.

van Kouwen MC, Drenth JP, van Krieken JH. Ability of FDG-PET to detect all cancers in patients with familial adenomatous polyposis, and impact on clinical management. Eur J Nucl Med Mol Imaging 2006; 33: 270–4.

Neuman J, Rosiorceanu A, Schuss A, et al. Radiology-pathology conference: sclerosing hemangioma of the lung. Clin Imaging 2006; 30: 409–12. doi:10.1016/j.clinimag.2006.05.030. PMid: 17101410.

de Koning DB, Drenth JP, Oyen WJ, Wagenaar M, Aliredjo RP, Nagengast FM. Pulmonary sclerosing hemangioma detected by fluorodeoxyglucose positron emission tomography in familial adenomatous polyposis: report of a case. Dis Colon Rectum 2007; 50: 1987–91.

Hsu PK, Cheng HF, Yeh YC, Wu YC, Hsu WH. Pulmonary sclerosing haemangioma mimicking lung cancer on PET scan. Respirology 2009; 14: 903–6. doi:10.1111/j.1440-1843.2009.01559.x. PMid:19659837.

Miura N, Shoji F, Kawano D, et al. A pulmonary sclerosing hemangioma with an increasing uptake on PET. Thorac Cardiovasc Surg 2009; 57: 498–9. doi:10.1055/s-2008-1039219. PMid:20013629.

Miyagawa-Hayashino A, Tazelaar HD, Langel DJ, Colby TV. Pulmonary sclerosing hemangioma with lymph node metastases: report of 4 cases. Arch Pathol Lab Med 2003; 127: 321–25.