

First Case Report of F-18 FDG PET/CT in a Rare Case of Bilateral Adrenal and Lung Metastasis from Primary Hepatic Angiosarcoma

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ABSTRACT

Hepatic angiosarcoma is a very rare disease, accounting for only 2% of primary liver malignancy. They are rare tumours with poor prognosis, with patients usually dying within 6 months. Metastases mainly occur in lymph nodes, spleen, lungs, bones and adrenals. There are some studies that reported the use of Fluorine -18 Fluorodeoxyglucose (FDG)-positron emission tomography (PET)/ computer tomography (CT)(F 18 FDG PET/CT) in angiosarcomas (hepatic, cardiac, venous and cutaneous angiosarcomas) in the literature. Bilateral adrenal metastasis from angiosarcoma of liver is a rare phenomenon. We report the first case of 36 year old patient, angiosarcoma of liver, post right lobe hepatectomy status, presented with bilateral adrenal and lung metastases imaged using F-18 FDG PET/CT.

Key words: Hepatic angiosarcoma, Bilateral adrenal metastasis, Lung metastasis, F -18 FDG PET/CT.

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History

- Submission Date: 27-05-2017;
- Review completed: 18-06-2017;
- Accepted Date: 28-07-2017;

DOI : 10.5530/ogh.2018.7.1.16

Article Available online

<http://www.oghreports.org>

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INTRODUCTION

Angiosarcomas account for ~1-2% of all soft tissue sarcomas, which are aggressive and malignant neoplasms of the endothelial-type cells that line vessel walls. They may occur in any region of the body, although the primary sites are usually the skin, the breast, soft tissues and bone. Primary hepatic angiosarcomas are rare and represent ~5% of all angiosarcomas.^[1] There are some studies that reported the use of Fluorine -18 Fluorodeoxyglucose (FDG)-positron emission tomography (PET)/ computer tomography (CT)(F-18 FDG PET/CT) in angiosarcomas (hepatic, cardiac, venous and cutaneous angiosarcomas) in the literature.^[2] Metastatic hepatic angiosarcomas to adrenals are particularly rare, and, to the best of our knowledge, no case of a bilateral adrenal and lung metastasis from a primary hepatic angiosarcoma has been reported to date in F-18 FDG PET/CT.

CASE REPORT

A 36 year-old male, known case of angiosarcoma of liver, post right lobe hepatectomy done 2 years before presented with complaint of abdominal pain. Ultrasound abdomen revealed large mass lesion in left adrenal gland, suspicious for metastasis. He was referred for whole body F-18 FDG PET/CT (figure 1a) for restaging which showed a large necrotic enhancing lesion in left adrenal (with an SUVmax of 20, figure 1b), also right adrenal showed a smaller lesion with intense FDG uptake (SUVmax of 8, figure 1c) and bilateral pulmonary metastasis (figure 1d, arrows). He underwent chemotherapy and on follow up.

DISCUSSION

Angiosarcoma is an uncommon mesenchymal malignant neoplasm of the vascular or lymphatic endothelium, accounting for 2% of all soft-tissue sarcomas, that can affect any organ but seems to have a predilection for the skin in the head and neck regions. Although primary hepatic angiosarcoma is rare and accounts for only 2% of primary hepatic tumor, it is the most common malignant mesenchymal tumor of the liver.^[3] Hepatic angiosarcoma is ranked as fifth in the list of most common sites of angiosarcoma. Hepatic angiosarcoma occurs with more frequency during the sixth decade of life, with a men-to-women ratio of 1.9 :1.

Risk factors include exposure to vinyl chloride monomer, ingestion of thorium dioxide (thorotrast), or ingestion of arsenic. Although the symptoms are usually nonspecific, abdominal distension and discomfort, weight loss, and fatigue are commonly found. Others like jaundice, ascites, or hepatomegaly are usually associated with advanced liver angiosarcoma.^[4] Angiosarcomas are aggressive tumors with poor prognosis, therefore, it is quite important to determine disease extension and detect local recurrence and/or distant metastases for appropriate therapy management. Hepatic angiosarcoma metastasis to adrenal gland is a rare phenomenon. Twenty-two cases of primary adrenal angiosarcomas in the medical literature (English language alone) have been reported.^[5] There are no case reports of metastasis to bilateral adrenal glands.

Cite this article: Kamaleshwaran KK, Radhakrishnan EK, Jose RP, Shinto AS. First case report of F-18 FDG PET/CT in a Rare case of Bilateral adrenal and lung metastasis from primary hepatic angiosarcoma. OGH Reports. 2018;7(1):75-6.

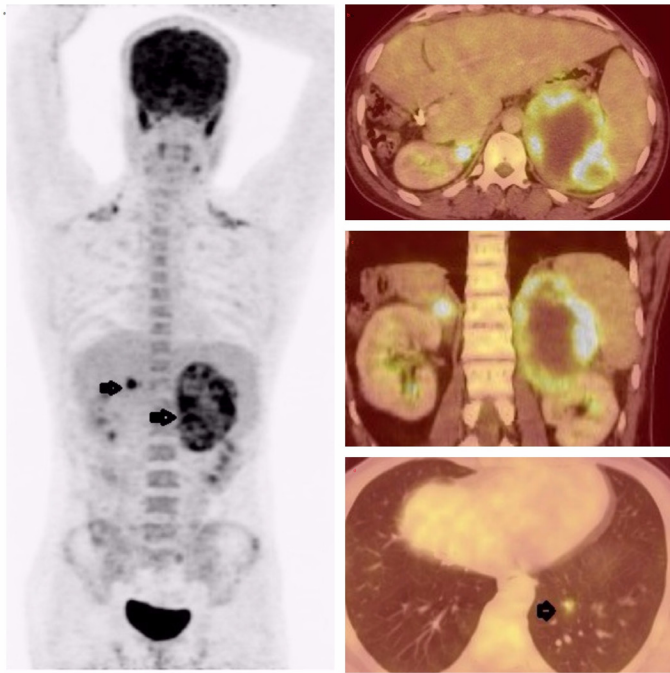


Figure 1: Whole body F-18 FDG PET/CT maximum projection image (MIP) (a), axial (b) and coronal (c) PET/CT showing enhancing necrotic lesion in both adrenal glands, also post right lobe hepatectomy status noted and axial PET/CT (d) showing pulmonary nodules.

To our knowledge there are only a few reports about angiosarcomas at uncommon localizations (cardiac, hepatic and venous regions) detected by FDG PET and PET/CT and the value of FDG PET in diagnosis and staging was well-defined in these studies. However, early detection of local and/or distant metastases is a challenging problem in high-risk patients and has an important role to predict the patients' outcomes. For the primary hepatic angiosarcoma and metastatic lesion, PET/CT was useful for the detection of the metastasis in the current case. An angiosarcoma does not have a consistent appearance in imaging, due to its varied histological composition. Thus, it is challenging to differentiate an angiosarcoma from other types of vascular tumors preoperatively.^[6] The reason FDG accumulates in adrenal angiosarcoma is not known.

However, in general, the degree of FDG accumulation in various tumors is considered to depend on the activity of glucose transporters, especially GLUT-1, and of hexokinases in the cells based on the findings of experimental and clinical studies. In our patient, overexpression of GLUT-1 was seen. Thus, it can be the reason for the marked FDG accumulations in the adrenal tumors.

Oe *et al* reported the first case of FDG PET/CT in hepatic angiosarcoma.^[7] Maeda *et al*^[8] originally detailed the utility of PET/CT for the detection of primary hepatic angiosarcomas, and suggested that PET/CT may contribute to confirming the presence or absence of distant metastases in other organs, and then assist in the selection of an appropriate treatment regimen. Hur *et al*^[9] reported the clinical course of hepatic angiosarcoma, in which metastases were noted in the spleen, lung, pericardium, and bone.^[8] Our patient presented with bilateral adrenal and pulmonary metastasis which have not been reported yet in literature.

FDG PET/CT can be a promising imaging tool in angiosarcomas. It enables to detect disease extension and multiple metastasis at a single session and is cost-effective by providing morphologic and functional images concurrently. Our case is the first to report FDG PET/CT findings in hepatic angiosarcoma, post-surgery presented with bilateral adrenal and pulmonary metastasis.

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