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Case Report

Aneurysmal angiosarcoma associated with vascular graft revealed by ^{18}F-FDG-PET imaging

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ABSTRACT

We report a rare case of vascular graft-associated aneurysmal angiosarcoma by ^{18}F-Fluorodeoxyglucose (FDG) positron emission tomography (PET). An 81-year-old male patient, with a prior history of graft interposition 1 year previously, was referred to ^{18}F-FDG-PET because of an inflammatory syndrome of unknown origin. FDG-PET images revealed a particular pattern of intense circular uptake within the arterial wall (SUVmax = 10) in a popliteal aneurysm and, additionally, a large hypermetabolic mass centered by the graft. Remote hypermetabolisms in lung nodules and pleural thickenings were also detected. The diagnosis of angiosarcoma was ascertained through histopathological analysis of surgical samples. Development of an aneurysmal angiosarcoma at the site of a vascular graft is a rare entity, often misdiagnosed. ^{18}F-FDG-PET appears to be useful in its detection with a PET pattern of intense circular uptake within the arterial wall. Such finding should lead to the search for distant metastasis.

Introduction

Intimal angiosarcoma, a rare entity of soft tissue sarcoma, has been described in association with arterial thrombosis in the particular setting of vascular grafts or aneurysm [1–3]. Preoperative diagnosis is difficult due to the presence of intimal angiosarcomas with diverse and nonspecific symptoms, and imaging studies often fail to differentiate between atheroma, mural thrombus, aneurysm, and tumors [2]. ^{18}F-Fluorodeoxyglucose (FDG) positron emission tomography (PET) computed tomography (CT) imaging has already been shown to be useful for the diagnosis of angiosarcoma [4], especially in instances of rare localization [5,6].

We report herein a case in which an aneurysmal angiosarcoma associated with a vascular graft was revealed at a multimetastatic stage with a particular pattern of intense circular uptake within the arterial wall on ^{18}F-FDG-PET imaging.

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Case report

An 81-year-old male patient, with a prior history of femoral aneurysm treated surgically with graft interposition 1 year previously, was referred to ^{18}F-FDG-PET because of an inflammatory syndrome of unknown origin. ^{18}F-FDG-PET was performed on a Biograph hybrid system (Siemens, Knoxville, TN) with an intravenous injection of 4 MBq/kg of ^{18}F-FDG after an overnight fast. The CT was first performed 60 minutes after the injection for providing the attenuation-correction map and was immediately followed by the 3D PET recording. A CT angiography was performed thereafter using a 320-detector CT scan system (Aquilion ONE, Toshiba, Japan) after intravenous administration of 150 mL of contrast medium (OMNIPAQUE 350, GE Healthcare, Chalfont St. Giles, UK). The final histologic diagnosis was obtained after surgery. Informed consent was obtained from this patient.

As illustrated in Figure 1, the FDG-PET images revealed a particular pattern of intense circular uptake within the arterial wall (SUVmax = 10) in a right popliteal aneurysm in addition to a large hypermetabolic mass in the right leg muscles centered by the graft. CT angiography revealed a total thrombosis of the right popliteal aneurysm (Fig. 2). Finally, remote hypermetabolisms were also detected, corresponding to the lung nodules and pleural thickenings shown in Figure 3.

The diagnosis of intimal angiosarcoma was ascertained through the histopathological analysis of surgical samples from both the femoral aneurysm and the leg mass. Of note, the surgical specimen was reviewed by pathology at the time of the graft, and no sarcoma was detected, thereby excluding a neoplastic lesion prior to the graft.

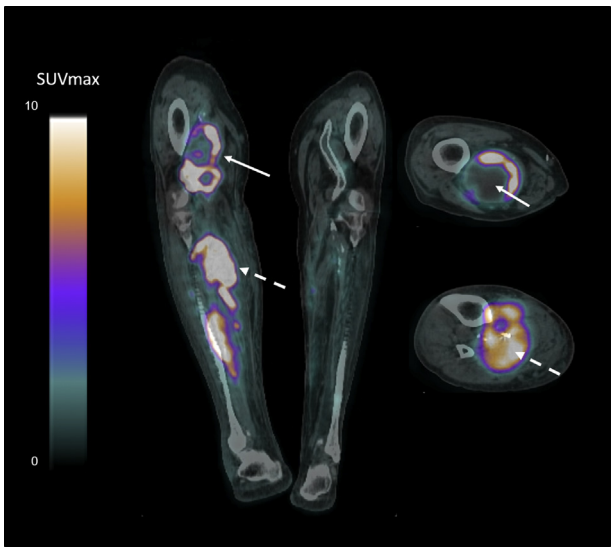

Fig. 1 – Coronal and axial slices of 18F-FDG-PET imaging showing a particular pattern of intense circular uptake within the arterial wall in a right popliteal aneurysm (white solid arrow) and, additionally, a large hypermetabolic mass in the right leg muscles centered by the graft (white dotted arrow). 18F-FDG, ^{18}F-Fluorodeoxyglucose; PET, positron emission tomography.

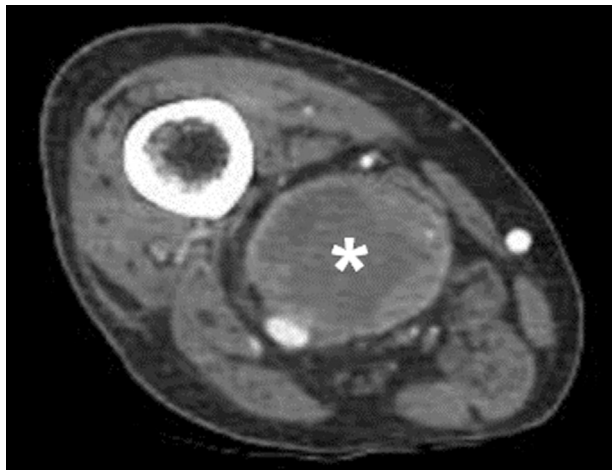

Fig. 2 – Axial slice of a CT angiography revealing a total thrombosis of the right popliteal aneurysm (asterisk). CT, computed tomography.

Discussion

The development of angiosarcomas at the site of vascular prosthesis or arising from an arteriovenous graft site has been described, leading to hypothesize that chronic repair processes and alterations in blood flow with thrombosis could constitute predisposing factors [1,3]. Moreover, the graft material may in itself directly provoke angiogenesis and subsequent tumorigenesis and could represent a risk factor in our patient previously treated for graft interposition. Indeed, newer-generation "biopolymers" seek to incorporate bioactive molecules that directly stimulate vascular regeneration [3]. In addition, cases of angiosarcoma have been described in association with an arterial aneurysm [2], such as that documented in the present case report, although no causal relationship could be conclusively established.

In this rare case of vascular graft-associated aneurysmal angiosarcoma, ^{18}F-FDG-PET imaging revealed a particular pattern of circular aneurysmal uptake, which was substantially

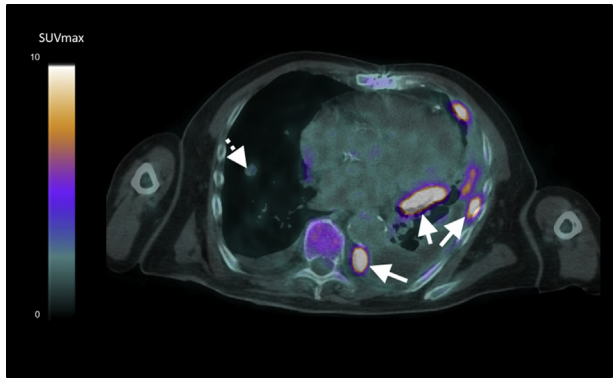

Fig. 3 – Axial slice of 18F-FDG-PET imaging revealing distant hypermetabolisms in a lung nodule (white dotted arrow) and pleural thickenings (white solid arrows) related to metastasis. 18F-FDG, ^{18}F-Fluorodeoxyglucose; PET, positron emission tomography.

more intense than that described for common atherosclerotic aneurysms [7]. Therefore, it is likely that SUVmax values may help in differentiating aneurysmal sarcoma from the common forms of aneurysmal atherosclerosis in a manner similar to that already documented for differentiating pulmonary artery sarcoma from common forms of pulmonary embolism [8]. Alternatively, the particular pattern of circular FDG uptake allows distinguishing aneurysmal angiosarcoma with infectious diseases such as mycotic aneurysms, in which FDG uptake foci are described in a setting of infective endocarditis [9].

Moreover, the detection of distant metastasis on ^{18}F-FDG-PET images herein provided an opportunity for whole-body imaging, yielding considerable additional information for the diagnosis of angiosarcoma. Indeed, distant pulmonary metastasis has already been reported for angiosarcoma [10]. However, it should be emphasized that increased FDG uptake is not specific and can be observed in a variety of conditions including infection [9]. While the present FDG-PET findings are suggestive, they are nonetheless not diagnostic of sarcoma. Diagnosis is made by pathologic analysis.

In summary, we report herein a case of aneurysmal angiosarcoma development at the site of a vascular graft, which is a rare entity and often misdiagnosed. ^{18}F-FDG-PET appears to be useful in its detection with a PET pattern of intense circular uptake within the arterial wall. Such uptake appears of greater intensity than in atherosclerotic aneurysm, commonly associated with severe thrombosis, and should lead to the search for distant metastasis.

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