

CASE REPORT

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Pigmented villous nodular synovitis mimicking metastases on ^{18}F -FDG PET/CT in a patient with rectal mucosal melanoma: a case report

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Abstract

Background: Mucosal melanomas are rare and have a high potential for metastasizing. Surgical resection is the treatment of choice for single distant metastases. Malignant melanoma usually shows the highest uptake of fluorine-18 fluorodeoxyglucose (^{18}F -FDG). ^{18}F -FDG positron emission tomography /computed tomography (PET/CT) is usually used for melanoma staging. An extensive literature review revealed only 4 published case reports and an original paper involving 8 cases (12 cases in total) of patients with skin melanomas in whom pigmented villous nodular synovitis (PVNS) mimicked metastatic melanoma, however, none of the melanomas reported were of rectal mucosal origin.

Case presentation: A 60-year-old woman presented with recent diagnosis of rectal mucosal melanoma, two additional ^{18}F -FDG-avid lesions in the left ankle and left foot were detected on ^{18}F -FDG PET/CT. Metastases were initially suspected; however, the final diagnosis was PVNS.

Conclusions: This is the first report of PVNS mimicking metastases on ^{18}F -FDG PET/CT in a patient with rectal mucosal melanoma. Although high ^{18}F -FDG-avid lesions in patients with rectal mucosal melanoma are highly suspected to be metastasis and warrant an meticulous examination, the present case is a reminder that in such patients, not all lesions with high ^{18}F -FDG uptake, especially those near a joint, are metastases and that more extensive resection is unnecessary.

Keywords: Melanoma, Pigmented villous nodular synovitis (PVNS), Positron emission tomography (PET), Rectum

Background

Only 1% of all melanomas arise from mucosa; most melanomas arise from skin [1]. Mucosal melanomas arise primarily in the head and neck, anorectal, and vulvovaginal regions. Of all colorectal malignancies, anorectal mucosal melanomas are rare (0.05%) [2], and they have a high potential for metastasis [1, 3, 4]. Surgical resection of the primary tumor, performed by wide local excision, is the mainstay of treatment.

Of all cancers, malignant melanoma usually shows the highest uptake of fluorine-18 fluorodeoxyglucose (^{18}F -FDG) [5]. Positron emission tomography/computed tomography

(PET/CT) with ^{18}F -FDG is a highly effective way to screen for metastases of malignant melanoma throughout the body [6–8]. ^{18}F -FDG PET/CT can reveal unexpected metastases, often outside the field of view of the other imaging modalities; such findings necessitate a change in patient management [9]. Surgical excision of metastases is recommended if only one or a few sites of disease are apparent [10].

However, not all highly ^{18}F -FDG-avid lesions are malignant. Benign conditions and lesions can have high ^{18}F -FDG uptake, including hyperplasia, benign tumors, and any inflammation or infection [11, 12]. Therefore, lesions should be histologically confirmed, particularly when PET/CT findings might prompt a change of treatment.

Pigmented villous nodular synovitis (PVNS) represents an uncommon benign proliferative process characterized by focal or diffuse hyperplasia of synovial villi that affects

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the synovial joints, tendon sheaths, and bursa membranes. In asymptomatic cases, no additional treatment is required [13–16]. The knee, followed by the hip, is the most common location of PVNS. Other large joints affected include the ankle, the shoulder, and the elbow, in decreasing order of frequency. PVNS lesions have high ^{18}F -FDG uptake and are known to have a false-positive appearance on ^{18}F -FDG PET/CT [17, 18].

We describe the case of a patient with rectal mucosal melanoma and two additional ^{18}F -FDG-avid lesions, one in the left ankle and one in the left foot. These lesions were detected on ^{18}F -FDG PET/CT and initially suspected to be metastases, but the final diagnosis was PVNS. To our knowledge, this is the first report of PVNS that mimics metastases on ^{18}F -FDG PET/CT in a patient with rectal mucosal melanoma.

Case presentation

A 60-year-old woman presented to her primary care physician with bloody stool for 2 months. Laboratory examination revealed Hb and Hct levels were 11.8 g/dL and 34.2%, (reference range: 11.6–14.8 g/dL and 34–44%), respectively. Stool occult blood was <7 ng/mL (reference range: <12 ng/mL). Physiological examination did not reveal any other skin lesion that could be suspected for melanoma. Colonoscopy and biopsy performed at another hospital revealed a malignant melanoma at the anorectal site; thereafter, the patient was transferred to our hospital only with pathology report for further management. Routine ^{18}F -FDG PET/CT examination performed after the biopsy for melanoma staging revealed a highly ^{18}F -FDG-avid lesion in the rectum (Fig. 1). The maximum standardized uptake value (SUV_{max}) was 15.3. Two additional high ^{18}F -FDG-avid lesions were found in her left ankle and left foot (SUV_{max} 8.9; Fig. 2).

The lesion with the highest ^{18}F -FDG avidity noted was diagnosed as rectal melanoma; moreover, the lesions in the left ankle and the left foot showed high ^{18}F -FDG avidity. For these reasons, and because melanoma has a high metastatic potential, these lesions were first thought to be distant metastases.

The patient was referred to a surgical oncologist to undergo additional workup. Magnetic resonance images (MRI), sagittal T1-weighted turbo spin echo (TSE) image (Figs. 3 and 4a), sagittal T1-weighted TSE fat-suppressed with Ga-DTPA contrast -enhanced image with Ga-DTPA fat suppression (Figs. 3 and 4b), coronal proton density (PD)-weighted TSE fat -suppressed image (Figs. 3 and 4c), and transverse T1-weighted TSE fat-suppressed with contrast-enhanced image with Ga-DTPA fat suppression (Figs. 3 and 4d) of the left foot, obtained to further characterize the lesions, revealed the two masses beside the flexor hallucis longus muscle and tendon, which were



Fig. 1 Anterior (left) and left lateral (right) whole-body Fluorine-18 fluorodeoxyglucose (^{18}F -FDG) maximum-intensity projections in a patient with rectal melanoma. An intensely ^{18}F -FDG-avid lesion was present in the rectum (arrow) and two additional intensely ^{18}F -FDG-avid lesions were present in the left ankle and in the left foot (arrowheads)

locations suggestive of metastases from melanoma; however, the differential diagnosis included PVNS. Out of concern that they represented metastatic melanoma, she was taken to the operating room. Surgery on the left ankle revealed firm, yellowish subcutaneous tumors beneath the deep fascia and the Achilles tendon. Intraoperative pathological study revealed that the lesion in the left foot was benign; therefore, only the mass in the left ankle was resected (Fig. 5), and special pathological staining revealed that the correct diagnosis was PVNS (Fig. 6). Thus, this patient underwent abdominoperineal resection. However, a follow-up abdominal CT conducted 3 months later

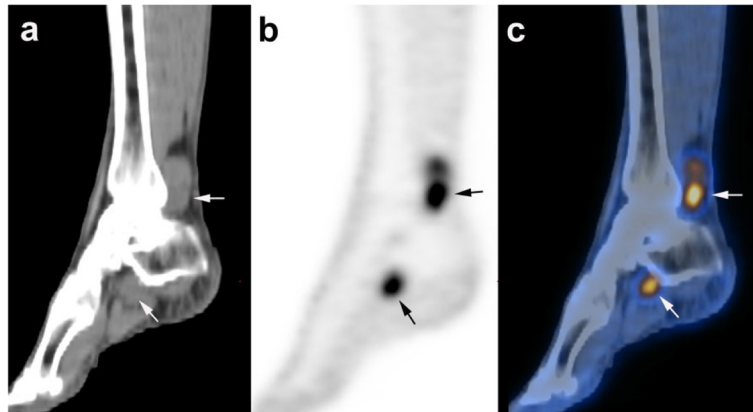


Fig. 2 Sagittal positron emission tomography (PET; **a**), computed tomography (CT; **b**), and fused Fluorine-18 fluorodeoxyglucose (¹⁸F-FDG) PET/CT (**c**) of the patient’s left ankle and left foot. Two ¹⁸F-FDG-avid lesions were present in these locations beside the flexor hallucis longus muscle and tendon (arrows)



Fig. 3 Magnetic resonance images of the patient’s left ankle. **a** Sagittal T1-weighted turbo spin echo (TSE) image. **b** Sagittal T1-weighted TSE contrast-enhanced image with Ga-DTPA fat-suppressed. **c** Coronal proton density (PD)-weighted TSE fat-suppressed image. **d** Transverse T1-weighted TSE contrast-enhanced image with Ga-DTPA fat-suppressed. A well-enhanced mass was identified beside the flexor hallucis longus muscle at the level of the ankle joint (arrows). Considering that the patient was diagnosed with melanoma, these appearances were in favor of metastases from melanoma according to a radiologist. However, the differential diagnosis was pigmented villous nodular synovitis

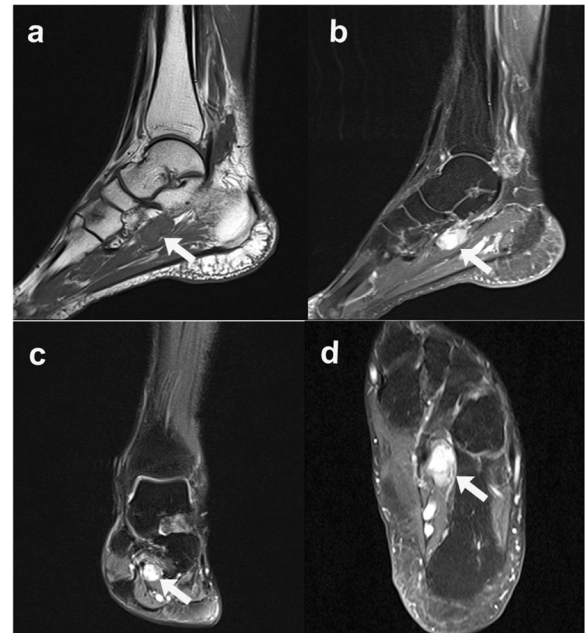


Fig. 4 Magnetic resonance images of the patient’s left foot. **a** Sagittal T1-weighted turbo spin echo (TSE) image. **b** Sagittal T1-weighted TSE contrast-enhanced image with Ga-DTPA fat-suppressed. **c** Coronal proton density (PD)-weighted TSE fat-suppressed image. **d** Transverse T1-weighted TSE contrast-enhanced image with Ga-DTPA fat-suppression. Another well-enhanced mass was identified beside the flexor hallucis longus tendon at the level of the talonavicular joint (arrows). Considering that the patient was diagnosed with melanoma, these appearances were in favor of metastases from melanoma according to a radiologist. However, the differential diagnosis was pigmented villous nodular synovitis

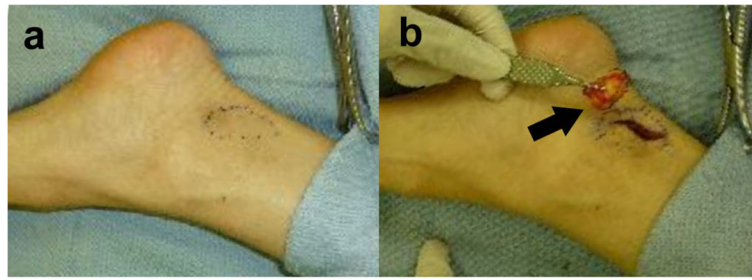


Fig. 5 Excision of the tumor in the left ankle. **a** The location was marked (arrow) before surgery. **b** The mass (arrow) was removed

revealed new multiple liver metastases, and chemotherapy treatment was initiated. Fortunately, a follow-up abdominal CT conducted 2 years later revealed that the liver lesions were nearly in complete regression. The patient is alive, and after another 2 years after complete regression, the patient is stable at present.

Discussion and conclusion

Because anorectal melanomas have a high metastatic potential and high ^{18}F -FDG uptake, any distant lesion that is highly ^{18}F -FDG-avid is suspected as being a possible metastasis, and for any resectable ^{18}F -FDG-avid lesions that are identified, aggressive surgical management is presumably necessary. However, in the present case, intraoperative pathological tests revealed benign tumor. Therefore, only lesion excision was performed.

An extensive literature review revealed only four published case reports and an original paper about 8 cases (12 cases in total) of patients with skin melanomas in whom PVNS mimicked metastatic melanoma, but none of the melanomas were mucosal [18–22]. To our knowledge, our case is the first report of PVNS mimicking metastases on ^{18}F -FDG PET/CT in a patient with rectal mucosal melanoma.

In retrospect, there can be two indications suggesting that these lesions are not metastases from melanoma. First, we noted that although the lesions in the left ankle and left foot showed high ^{18}F -FDG uptake (SUV_{max} , 8.9), the uptake was moderately lower than that of the rectal melanoma (SUV_{max} , 15.3). This suggests that these lesions were not the highest ^{18}F -FDG-avid lesions. Second, the lesions were located on the ankle and foot,

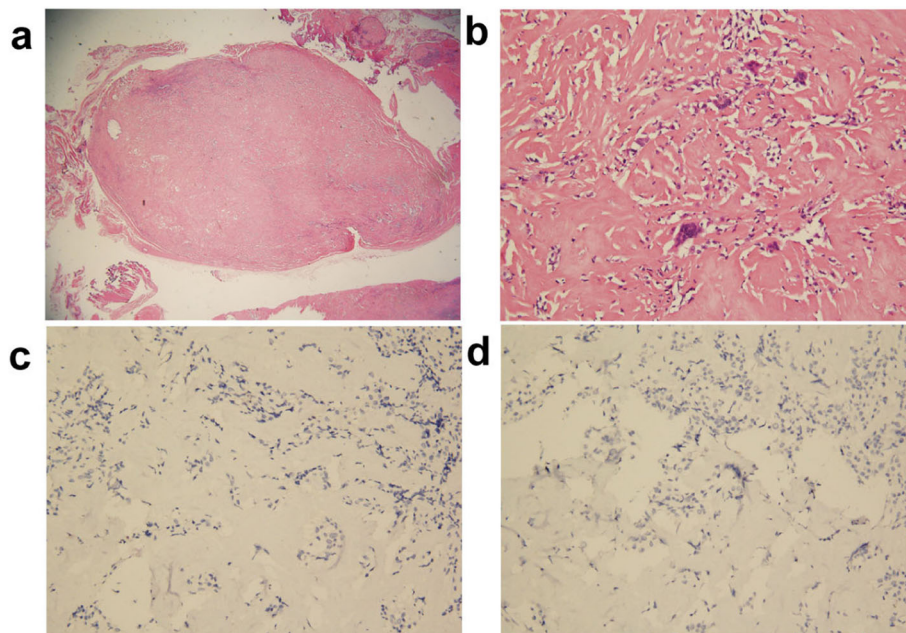


Fig. 6 Pathological appearances of the mass from the left ankle. **a** Low-power view of the hypocellular, ovoid shaped tumor. **b** Focal areas had increased cellular infiltrates consisting of lymphocytes, macrophages, and multinucleated giant cells, surrounded by highly collagenized stroma. Special staining with Melan-A (**c**) and HMB45 (**d**), both of which are markers of melanoma, yielded negative results. The pathologically proved diagnosis was pigmented villous nodular synovitis (PVNS)

which are sites preceded by the knee and hip as the most common sites; therefore, the locations of the lesions were not at the most common sites of PVNS.

Because the treatment of PVNS differs significantly from that of metastatic melanoma, PVNS should be included in the differential diagnosis of melanoma, especially in cases when ^{18}F -FDG PET/CT identifies possible metastatic spread near the joints, as in this case (in the left ankle and the left foot).

Although any highly ^{18}F -FDG-avid lesion in patients with rectal mucosal melanoma is high suspect for being a metastasis and warrants an aggressive workup, some benign conditions and lesions can have high ^{18}F -FDG uptake. This case is a reminder that not all lesions with high ^{18}F -FDG uptake, especially not extremely high ^{18}F -FDG uptake and near the joint, are metastases and that more extensive resection may not be necessary.

Abbreviations

^{18}F : Fluorine-18; FDG: Fluorodeoxyglucose; PET/CT: Positron emission tomography /computed tomography; PVNS: Pigmented villous nodular synovitis; SUV_{max} : The maximum standardized uptake value

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Not applicable.

Authors' contributions

Study concept and design: CHL. Acquisition of data: LCW, NML, and CHL. Analysis and interpretation of data: YAY and CHL. Drafting of the manuscript: LCW and CHL. Critical revision of the manuscript for important intellectual content: CHL. Administrative, technical, and material support: CHL. Study supervision: CHL. Provision of pathological images: NML. All authors reviewed and provided feedback and insights for further research. All authors read and approved the final manuscript.

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Availability of data and materials

Not applicable.

Ethics approval and consent to participate

All procedures performed in this case report was in accordance with the ethical standards of the institutional review board of Chi Mei Medical Center (assurance number: 10806-E01) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Consent for publication

The case report has been granted exemption from review by the Institutional Review Board of Chi Mei Medical Center. (Assurance number:10806-E01). Because the patients were receiving scheduled and routine PET/CT for disease evaluation or surveillance, and not for research study, no any extra protocol was performed, therefore, there was no extra informed consent given. We just retrospective reviewed data, No identifying information of patients, including names, initials, or hospital numbers was involved in this manuscript.

Competing interests

The authors declare that they have no competing interests.

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