

FDG-PET/CT findings of atypical (bizarre/symplastic) uterine leiomyoma in a patient with abdominal leiomyosarcoma

Fabrice Hubelé, MD; Gerlinde Averous, MD; Edmond Rust, MD; Alessio Imperiale, MD; and Izzie Jacques Namer, MD

Atypical (bizarre) leiomyoma is a benign uterine smooth-muscle tumor characterized by a) a significant number of cells with dense eosinophilic cytoplasm and enlarged, bizarre single/multiple hyperchromatic or multiple nuclei without tumor necrosis and b) poor mitotic activity. We report the case of an atypical (bizarre) leiomyoma revealed by focal fluoro-deoxyglucose (FDG) uptake during a PET-CT in a patient with relapsing abdominal and retroperitoneal leiomyosarcoma.

Case report

Twenty years ago, a 50-year-old woman with multiple cancers suffered a multifocal ductal carcinoma in situ with microinvasion that was treated by mastectomy with axillary lymph node dissection, followed by radiotherapy and hormone therapy. One year later, she developed a well-differentiated leiomyosarcoma on her left arm measuring 2.5 cm, which was treated by complete surgical excision. Three years later, she again developed multifocal ductal carcinoma in situ with microinvasion of the left breast, this time treated by mastectomy and axillary lymph-node dissection. She was free of tumor for sixteen years until 2009, when she was diagnosed with an abdominal and retroperitoneal leiomyosarcoma extending from the inner side of the iliac wing to the left oblique muscle of the abdominal wall. The tumor measured 5x5x4 cm and had a proliferation

index (Ki67) of 40%. The treatment consisted in a wide surgical excision followed by chemotherapy. Only one year later, she relapsed with a bifocal tumor: one part on the left abdominal wall measuring 3x3.3x3.3 cm, and the other part below the first on the inner side of the left iliac wing measuring 6x5.5x6 cm.

Preoperative staging included a F18-deoxyglucose (FDG) PET-CT to assess tumor extension and metastasis. The PET-CT acquisition was in 3D-mode, 60 minutes after intravenous injection of 4.5 MBq/kg 18F-FDG. A dual-phase study was also performed on the pelvis 150 min after the injection.

The whole-body PET-CT scan found intense uptake of the bifocal known leiomyosarcoma of the left abdominal wall. Fortunately, another focal uptake on the uterus was found (Figs. 1A, D). The dual time-point imaging showed an increased FDG uptake; the maximum Standard Uptake Value was 8.5 kBq/ml one hour after injection and 12.3 kBq/ml 150 min later. PET-CT scan was completed by a 3T-MRI scan with T1-FAT-SAT and T2 sequences after gadolinium injection. The MRI could not exclude malignancy of the uterine mass.

The multidisciplinary cancer committee decided to perform a large excision of the abdominal tumor with hysterectomy during the same surgery.

Histology confirmed the bifocal, FNCLCC grade-3 leiomyosarcoma with a mitotic activity of 40 mitoses/10 HPF and a high Ki67 index of about 50-60%. The leiomyosarcoma was estrogen-receptor-negative (Figs. 1B, C).

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Drs. Hubelé, Imperiale, Rust, and Namer are associated with the Service de Biophysique et Médecine Nucléaire, and Dr. Averous is associated with the Service d'Anatomie Pathologique, all at Hôpital de Haute-pierre, CH Strasbourg, France. Contact Dr. Hubelé at fabrice.hubele@chru-strasbourg.fr.

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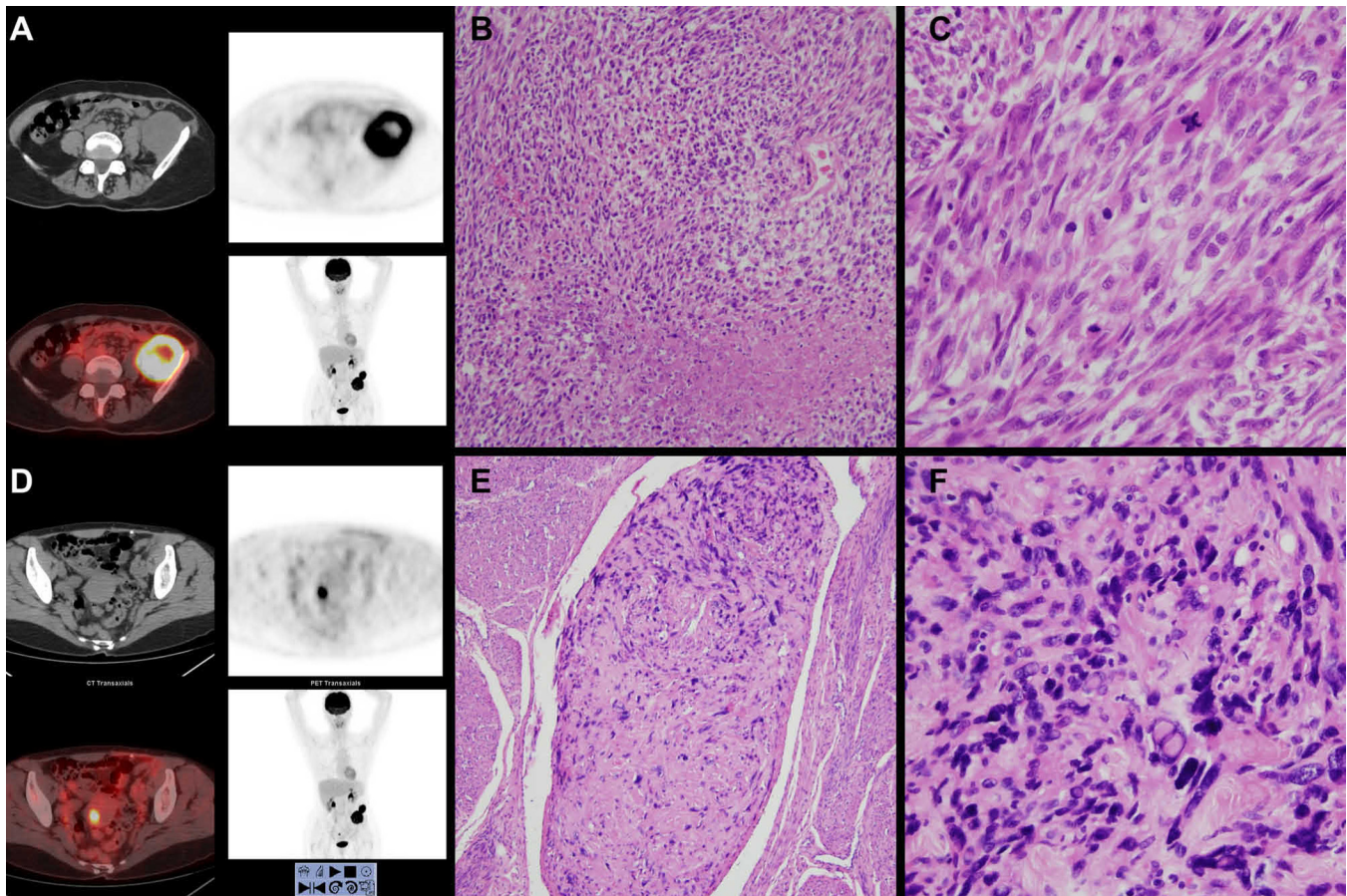


Figure 1. 50-year-old woman with carcinoma in situ (20 and 16 years ago) and leiomyosarcoma on the left arm (19 years ago). She recently developed another leiomyosarcoma of the left abdominal wall. FDG-PET-CT shows intense uptake of the recurrent tumor on the abdominal wall muscle and inner side of the iliac wing (A: axial CT, PET on the first line, fused and maximum-intensity projection [MIP] on the second line). Histology confirmed a leiomyosarcoma with coagulative necrosis; H&E staining x200 (B) and x400 (C). The same FDG-PET-CT showed abnormal focal uptake in the uterine body (D: axial CT, PET on the first line, fused and MIP on the second line). Histology revealed a uterine atypical (bizarre) leiomyoma with bizarre single or multiple hyperchromatic nuclei; H&E x100 (E) and x400 (F).

The histological examination of the uterine nodule revealed a 1.5-cm, well-circumscribed tumor composed of spindle cells with dense eosinophilic cytoplasm and enlarged, often bizarre single or multiple hyperchromatic nuclei without necrosis or myometrial invasion. Tumor cells were caldesmon, desmine, and smooth-muscle-actine positive, corresponding to the immunohistochemical phenotype of smooth muscle. The Ki67 level was low, with a maximum value of 15%. The mitotic activity was 1 mitosis/10 HPF. This tumor expressed estrogen receptors, whereas the leiomyosarcoma did not. Diagnosis was atypical (bizarre/symphastic) leiomyoma of the uterus, a benign variant of uterine leiomyoma (Figs. 1E, F).

Discussion

This case shows that atypical (bizarre) uterine leiomyoma can be suspected on FDG PET-CT due to glucose hyper-

metabolism. We think that despite the benign behavior of this kind of tumor, the focal uptake may be explained by the relatively high cellularity of the tissue and can be seen as a sign of increased proliferative and metabolic activity—as shown with the Ki67 level, which focally achieved 15% in the presence of very rare mitotic figures.

In view of the clinical history of the patient, who has a tendency to develop multiple malignant tumors, we should also discuss the possibility of a precancerous small tumor in *statu nascendi*, with a particular metabolic signature on FDG PET-CT.

Further comparative investigations involving histopathology and imaging should try to find out whether FDG PET-CT could help in assessing the category of uterine nodules, particularly those with increased metabolic and proliferative activity, in order to understand the gray-zone tumors better, as there are atypical leiomyomas and smooth muscle tumors of uncertain malignant potential (STUMP).

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