

Placental-site trophoblastic tumor and fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography

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Abstract

Objective: Pre-operative imaging characteristics of placental site trophoblastic tumor (PSTT) are variable and non-specific. Although magnetic resonance imaging (MRI), ultrasonography, chest CT/X-rays findings have been studied, the fluorine-18 fluorodeoxyglucose positron emission tomography/ computed tomography (¹⁸F-FDG PET/CT) findings of PSTT have not been previously documented. We present the findings of a first case of PSTT evaluated by pre-operative ¹⁸F-FDG PET/CT. A suspicious mass was biopsied and revealed PSTT in post-operative pathological examination. She was referred to the gynecology - oncology department. The ¹⁸F-FDG PET/CT scan revealed a 27x20mm laterally expanded lesion that showed increased ¹⁸F-FDG uptake (SUVmax :5.20) on the right isthmus of the uterus. The ¹⁸F-FDG PET/CT findings were in accordance with those from chest X-ray/s, CT and pelvic ultrasonography. A systematic, nerve sparing, paraaortic and pelvic lymph node dissection along with total hysterectomy and salpingoophorectomy was performed. The patient was discharged uneventfully. **Conclusion:** ¹⁸F-FDG PET/CT scan was able to identify the mass in the uterus which was shown by pathology to be PSTT. This finding of PET/CT was in accordance with other imaging techniques. Lymphatic mapping of ¹⁸F-FDG PET/CT in this case of was also in accordance with surgery and pathology findings.

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Introduction

Placental site trophoblastic tumor (PSTT) is the rarest gestational trophoblastic disease, and accounts for 0.23-1% of all gestational trophoblastic neoplasias [1]. Due to the rarity of placental site trophoblastic tumor, there is little information about its epidemiology and about 300 cases have been reported in the literature [2, 3]. Placental STT presents as a slow growing tumor which arises from intermediate trophoblasts and can occur following a normal or complicated pregnancy. Placental STT is characterized by low beta-hCG levels, a greater tendency for lymphatic spread and chemoresistance, unlike other gestational trophoblastic neoplasias (GTN). Lymphatic spread is encountered for about 5.9% of the cases at the time of diagnosis or recurrence. Most of PSTT at first diagnosis are confined to the uterus; however 14%-30% of them may have metastases to the lungs, liver, vagina, gastrointestinal tract, brain, lymph nodes, bladder, ovary, omentum, thoracic diaphragm, pancreas, spleen, kidney, bone marrow and scalp [4]. Pre-operative imaging characteristics of PSTT are variable and non-specific. Although MRI, and ultrasonographic, chest CT/X-ray findings are reported, ¹⁸F-FDG PET/CT findings of PSTT have not been previously described. We here present the ¹⁸F-FDG PET/CT preoperative findings of a PSTT case.

Case Report

A female 37 years old, gravida 2, parity 2, delivered a term healthy baby by Cesarean section. During Cesarean section, a suspicious mass was seen next to the hysterotomy site and a biopsy was performed which revealed PSTT. The patient was referred to the gynecology-oncology department. She was breastfeeding her baby. Her body mass index was 27, and Eastern Cooperative Oncology Group score was 0. Her physical examination was unremarkable and her laboratory values were within normal limits, hu-

man chorionic gonadotropin (hCG) level was 2.7 IU/mL. A preoperative chest X-ray, chest and a computerized tomography (CT) of the abdomen were normal. Abdomino pelvic ultrasonography revealed an echogenic nodular mass of 2x3 cm in the uterine corpus, without distinct borders.

The ^{18}F -FDG-PET/CT imaging performed 60 minutes after the intravenous injection of 270 MBq of ^{18}F -FDG by a scanner from (Philips Gemini TF) from the skull to the proximal thighs revealed a 27x20mm laterally expanded lesion on the right isthmus of the uterus that showed increased ^{18}F -FDG uptake. Maximum standardized uptake value (SUVmax) of the lesion was 5.20 (Figure 1A-B).

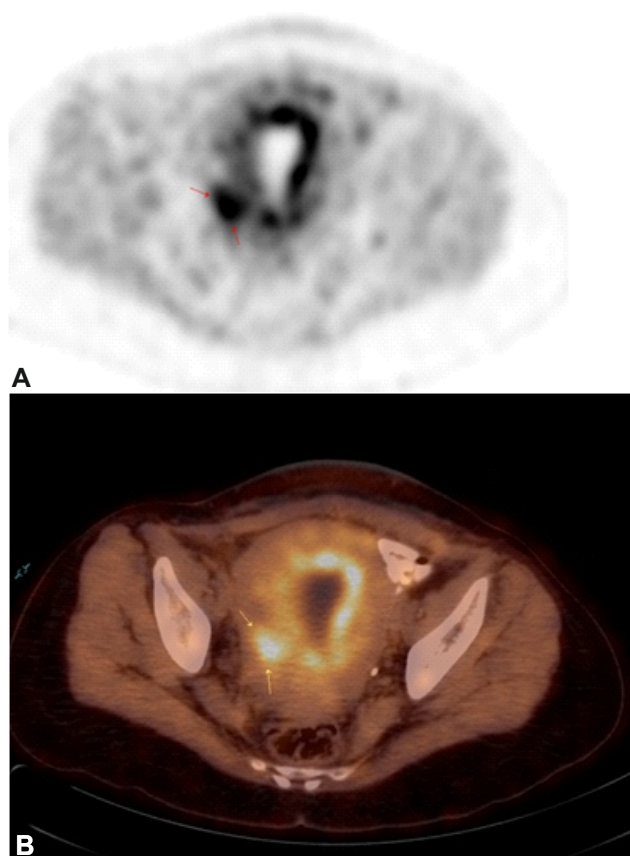


Figure 1. A-B: Axial PET and PET/CT fusion images demonstrated increased ^{18}F -FDG uptake on the tumor at the right isthmus of the uterus (arrow) (A, B) and the endometrium.

Wide spread increased ^{18}F -FDG uptake was detected on endometrial tissue secondary to postpartum hazards (SUVmax: 5.39). Increased breast uptake in the maximum intensity projection (MIP) image due to lactation was also observed (Figure 2).

The patient was then operated. Abdominal exploration was normal except for two paraaortic lymph nodes, in size 1x1cm. There was a mass of about 3x3cm in the anterior surface of the uterus. It was taken care of a systematic nerve sparing, during paraaortic and pelvic lymph node dissection, total hysterectomy and salpingoopherectomy. A 1000 mL of ascitic fluid was drained. Ascitic fluid was eventually decreased by diet. Patient's course was unremarkable and she was discharged on the postoperative day.



Figure 2: Maximum intensity projection image (MIP) illustrated increased and spotted ^{18}F -FDG uptake on both breasts and on the tumor at the right isthmus of the uterus (arrow).

Her final pathology showed: A tumor sized 3.5x2.5x2cm, infiltrating more than half myometrium close to the serosa by 0.1cm. There was venous invasion of the tumor (Figure 3A-B).

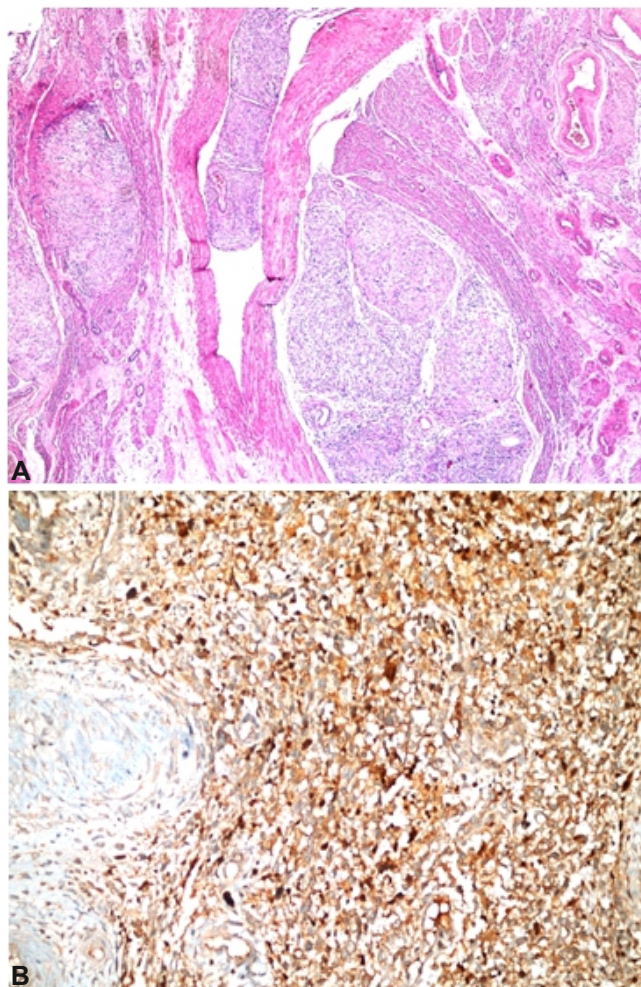


Figure 3. A-B: A) Myometrial invasion and adjacent blood vessel invasion of the placental site by a trophoblastic tumor (H&E x100) B) Immunohistochemical human placental lactogen (hPL) expression in neoplastic cells (hPLx200).

Adenomyosis and leiomyoma were observed in the non-neoplastic myometrium. There were no lymph node metastases (pelvic 0/28, and paraaortic 0/14).

Discussion

The PSTT are rare tumors that may have metastases to extrauterine genital organs or to more distant sites at the time of diagnosis. The FIGO stage which is used in PSTT (Table 1) does not include lymphatic spread.

Table 1. FIGO staging of GTN

Stage I	Limited to the uterus
II	Extended outside the uterus but limited to genital structure
III	Extended to lungs with or without known genital tract reached
IV	Any other site of metastasis

FIGO: International Federation of Gynecology and Obstetrics; GTN: Gestational Trophoblastic Neoplasia

The value of the FIGO/WHO risk scores (Table 2) is controversial. Survival rate 48 months after diagnosis was reported

to be 91% for stage I and 0% for stage III-IV [5]. Poor prognostic factors in PSTT are: An interval of more than 24 months since the last pregnancy, a level of mitosis higher than 5/10, age older than 34 years, a term birth for the last pregnancy, a myometrium invasion of more than 50%, presence of cells with clear cytoplasm and an extensive coagulation necrosis. Deep myometrial invasion in clinically apparent stage I PSTT patients is a risk factor for lymphatic spread.

There are limited data on the role of PET-CT in gestational trophoblastic neoplasia (GTN). No data are available for PSTT staging by ^{18}F -FDG PET/CT. Mapelli et al (2013) [6] studied the value of ^{18}F -FDG PET/CT in primary staging and monitoring GTN. In the staging procedure, ^{18}F -FDG PET/CT was in 91%, 84% and 81% in accordance with ultrasonography, chest radiography and CT, respectively. They have concluded that ^{18}F -FDG PET/CT was not superior to conventional imaging methods for staging GTN, but ^{18}F -FDG PET/CT might be useful in high risk GTN patients. In small cohort studies ^{18}F -FDG PET-CT was found to be an important imaging modality to detect metastases and exclude false positive lesions in GTN [7, 8]. In our case, ^{18}F -FDG PET/CT revealed an uterine mass with SUVmax 5.20 and PET/CT was in accordance with ultrasonography and chest X-ray.

The PSTT is usually located in the uterine corpus as in our case. PSTT is a rare form of trophoblastic diseases. A solid or cystic mass may be seen in ultrasonography. In patients where tumor cannot be demonstrated by ultrasonography, MRI may be useful to locate PSTT [9]. Hysterectomy is the main treatment modality in PSTT, where multi-agent etoposide, methotrexate, actinomycin D, cyclophosphamide, vincristine or etoposide, methotrexate, actinomycin D,

Table 2. FIGO/WHO risk classification. If the risk score was greater than 7, the patient was regarded as high risk. The distinction between low and high risk patients was practically applied for stages II and III.

Risk score	0	1	2	4
Age	<40	40	-	-
Previous pregnancy	Mole	Abortion	Term	-
Plasmatic HCG before treatment (IU/l)	<1000	<10.000	<100.000	>100.000
Interval months from index pregnancy	<4 months	4-7 months	7-13 months	>13 months
Tumor Size (cm)	-	3-5	>5	-
Number of metastasis	0	1-4	5-8	>8
Metastatic sites	Lung	Kidney, Spleen	Bowel	Brain, Liver
Failure of prior chemotherapy	-	-	Single agent	Two or more

FIGO: International Federation of Gynecology and Obstetrics; WHO: World Health Organisation

etoposide, cisplatin (EMA-CO or EMA-EP) chemotherapy is used in high risk or in metastatic patients. Lymphatic spread of PSTT, particularly to paraaortic lymph nodes, is reported in about 5.9% [10]. Of 11 patients with lymph node metastases, 5 had paraaortic 5 pelvic metastases and the remaining 6 metastases to other organs [10]. Lan et al (2010) suggested performing retroperitoneal lymphadenectomy in high risk stage I and stage II patients. We agree that every effort including lymphatic dissection should be undertaken in surgical treatment of this curable disease. The ^{18}F -FDG PET/CT scan may have a role in diagnosing PSTT, better or differently from other common forms of GTN. However, more data should be available in the literature to correctly identify the role of ^{18}F -FDG PET/CT in PSTT.

The authors declare that they have no conflicts of interest

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