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Placental site trophoblastic tumor in early stage: a case report

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Summary

Here, the authors present a case of a placental site trophoblastic tumor (PSTT) in a 28-year-old gravida 1 para 1 living 1 woman three months after vaginal delivery of a female infant at diagnosis in 2014. The patient was FIGO Stage I and finally underwent a total laparoscopic hysterectomy with ovarian conservation. Subsequently, the patient received two cycles of EMA/CO chemotherapy. Patient is on regular follow-up (clinical exam, β-hCG tests, pelvic and abdominal sonography) and has shown no signs of local or systemic recurrence for 24 months.

Key words: Placental site trophoblastic tumor (PSTT); Diagnosis; Treatment.

Introduction

Placental site trophoblastic tumors (PSTTs) are the rarest subtype of gestational trophoblastic neoplasm (GTN), typically occurred in reproductive-aged females. Surgery is the mainstay of treatment. Adjuvant chemotherapy was recommended in addition to surgery, considered for patients with Stage I disease who also have risk factors for recurrence [1, 2]. Here, the authors report a case which was confirmed to be PSTT (FIGO Stage I) on biopsy.

Case Report

A 28-year-old woman G1P1A0L1 (gravida, para, abortion, and living). She visited a local hospital with irregular vaginal bleeding after three months of vaginal delivery of a female infant in 2014. The serum β-hCG level at diagnosis was 80.05 mIU/ml. Ultrasonography found an irregular echogenic mass (32×31 mm) in the rear wall of the uterus with increased vascularity. A dilatation and curettage procedure was performed, which revealed PSTT with blood clots and extensive necrosis, and the hCG level decreased to 23.72 mIU/ml.

The patient was referred to the present center. The laboratory examination was unremarkable except that hCG turned to 42.5 mIU/ml. Physical examination revealed no abnormal finding. Transvaginal ultrasonography (TUS) and positron emission tomography/computed tomography (PET/CT) scan showed that the legion was confined to the uterus (Figure 1). After seven months of delivery, the patient received a total laparoscopic hysterectomy. Intraoperative appearance of the uterus and bilateral tubes and ovaries were normal. Grossly, the tumor was 4.5×2.5 cm in size located in the right and posterior wall of the uterus with poorly defined border, and showed a yellow, soft polypoidal mass, which infiltrated the whole myometrium and nearly to the serosal surface. Microscopically (Figure 2), the tumor was comprised of large and polygonal tumor cells, hyperchromatic with vesicular nuclei, and abundant eosinophilic cytoplasm. Obvious vascular invasion and severe nuclear atypia were also observed. The mean mitotic count was one mitoses per 10 high-power fields (HPFs). Immunohistochemically, the tumor cells showed positivity for AE1/AE3 (pan-cytokeratin antibody), inhibin-α, human placental lactogen (HPL), and p53, slight positivity for hCG, and the Ki-67 proliferative index was about 1%, but negative for placental alkaline phosphatase (PLAP) and smooth muscle actin (SMA). Subsequently, the patient was administered two cycles of EMA/CO chemotherapy, because of postoperative slight positivity for HCG. EMA: dactinomycin 0.5 mg, intravenously, days 1 and 2; etoposide 100 mg/m², intravenously, days 1 and 2; methotrexate 300 mg/m², intravenously, day 1; folinic acid 15 mg, orally, twice a day, days 2 and 3; CO: vincristine 1.4 mg/m², intravenously, day 8; cyclophosphamide 600 mg/m², intravenously, day 8 (treatments were given in two-week cycles). Patient was on regular follow-up (clinical exam, β-hCG tests, and pelvic and abdominal sonography) and has shown no signs of local or systemic recurrence for 24 months.

Discussion

PSTT is the rarest variant of gestational trophoblastic diseases and originates from the implantation site intermediate trophoblast. Due to its rarity and varied biological behavior, it presents a diagnostic challenge and treatment dilemma to both the pathologist and clinicians. It can develop following any type of pregnancy and mostly occurs after full-term normal delivery. The most common symptom is irregular vaginal bleeding [3-5]. The prognostic factors are age, interval from antecedent pregnancy (> 4 years indicating poor prognosis), FIGO staging, mitotic count (> 5/10 HPF high power field), and metastasis [6, 7]. The appropriate treatment of a patient with Stage I disease is hysterectomy and ovarian conservation, unless the patient has a family history of ovarian cancer or is postmenopausal [8].

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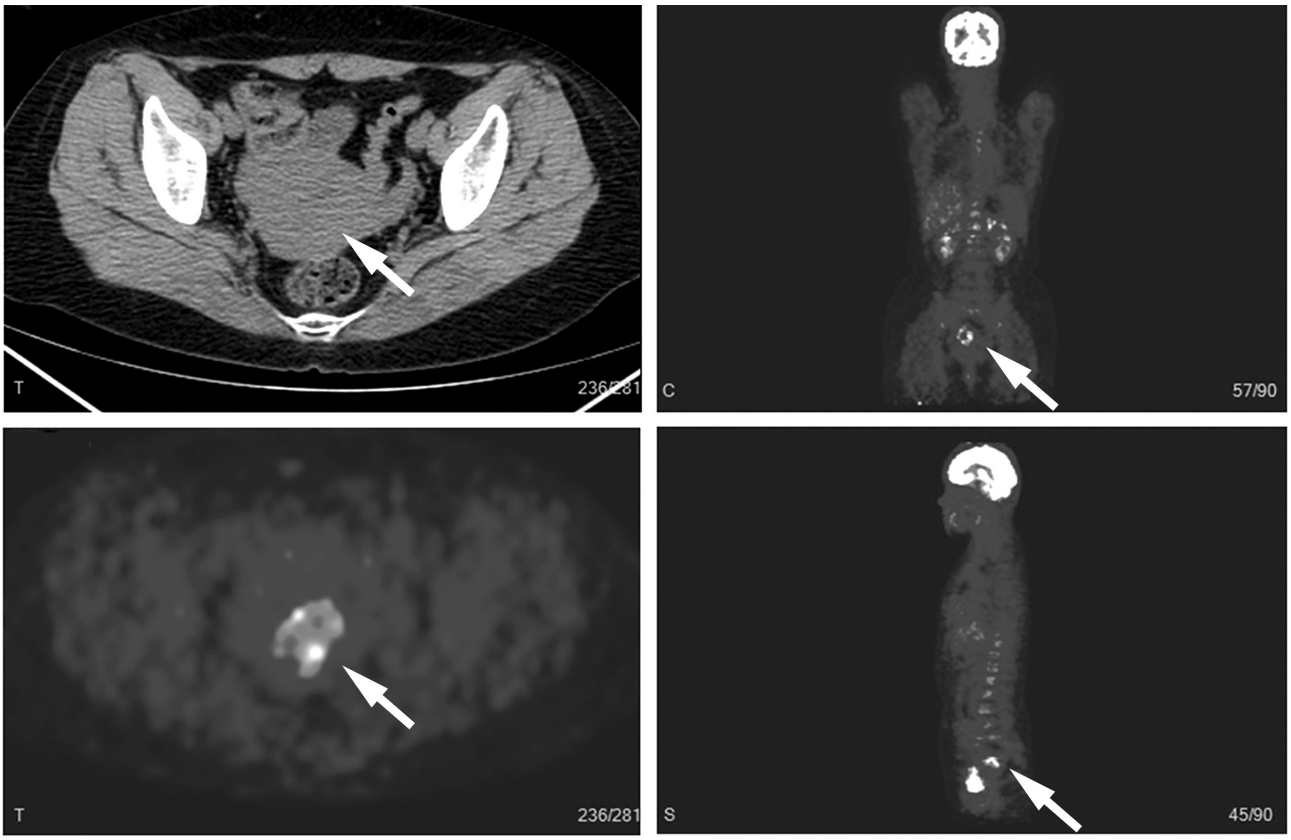

Figure 1. — PET/CT showed that the legion was confine to uterus (arrows) with no evidence of metastasis.

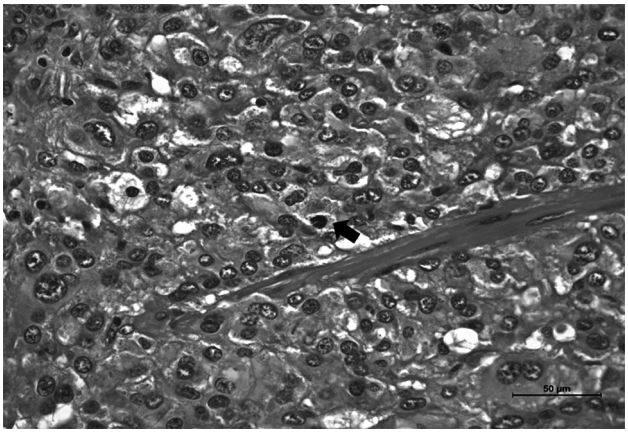

Figure 2. — PSTT area with severe nuclear atypia and mitotic (arrows). (HE stain ×40)

However, postoperative chemotherapy for early stage is controversial.

In the present case, a young patient with PSTT (Stage I, FIGO Stage, International Federation of Gynecology and Obstetrics) with irregular vaginal bleeding after three months of vaginal delivery at diagnosis underwent a total laparoscopic hysterectomy with ovarian conservation, as the legion was confined to the uterus and whole body PET/CT noted no involvement of pelvic lymph nodes and distant metastasis. Soon, the patient received two cycles of EMA/CO chemotherapy, because of persistent slight positivity for hCG. Now, the patient is alive with no evidence of disease on follow-up of 24 months.

Chemotherapy was recommended in addition to surgery, considered for patients with Stage I disease who also have risk factors for recurrence, such as long interval from AP, vascular invasion, deep myometrial invasion, serosal involvement, lymphatic spread, high mitotic index, or persistently raised postoperative hCG [1, 2]. Multi-agent regimen like EMA-CO (etoposide, methotrexate, actinomycin D, cyclophosphamide, vincristine) and EMA-EP (P-platinum) were the most available adjuvant chemotherapy, and BEP (bleomycin, etoposide, cisplatin) or MTX (methotrexate) was also successful [4]. Otherwise, PSTT were known to be chemoresistant, and the toxicity of chemotherapy would counteract the benefit for patient. It was indicated that scrupulousness is needed before adding postoperative chemotherapy on PSTT with Stage I disease.

Conclusion

PSTT is a rare subset of the GTNs. As it is relatively chemoresistant and has a varied prognosis, the benefit from postoperative chemotherapy on early stage PSTT is still equivocal. More high quality studies are required. There is a need for scrupulousness before adding postoperative chemotherapy or more powerful multi-agent regimen on Stage I PSTT.

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