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Case report

A case of placental site trophoblastic tumor complicating nephrotic syndrome in which hysteroscopic biopsy did not yield a definitive diagnosis

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ABSTRACT

Placental site trophoblastic tumor (PSTT) is the rarest subtype of gestational trophoblastic neoplasm. We present a case of PSTT complicating nephrotic syndrome. A 32-year-old woman experienced irregular menstrual bleeding and lower extremity edema 18 months after delivery. She was diagnosed with nephrotic syndrome and exaggerated placental site based on the hysteroscopic biopsy results. During follow-up, transvaginal color Doppler ultrasound showed an enlarged uterus filled with a hypervascular mass. Positron emission tomography–computed tomography showed diffuse accumulation in the entire uterus. The patient was diagnosed with PSTT only after total hysterectomy. Postoperatively, serum β-human chorionic gonadotropin decreased to within the normal range and her nephrotic syndrome resolved. She has remained without evidence of recurrence for 15 months. It is difficult to diagnose PSTT definitively. Most patients with PSTT are of reproductive age, therefore, to maintain fecundity, therapy development is expected.

Introduction

Placental site trophoblastic tumor (PSTT) is the rarest subtype of gestational trophoblastic neoplasm (GTN), which is characterized by intermediate trophoblasts at the site of placental implantation.[1] In 1976, Kurman et al described this entity for the first time using the name trophoblastic pseudotumor.[2] In 1981, Scully and Young coined the term PSTT to describe the malignant potential of this tumor.[3] PSTT is a rare subtype of GTNs, with almost 300 cases reported worldwide.[4] GTNs are rarely associated with nephrotic syndrome, and only a few cases have been documented.[5] It is difficult to diagnose PSTT definitively. Most cases are often diagnosed definitively after total hysterectomy. Most patients with PSTT are of reproductive age, therefore, to maintain fecundity, therapy development is expected.

Case report

A 32-year-old woman, gravida 1 para 1, gave birth to a healthy full-term baby through normal vaginal delivery. For 17 months after delivery, the patient had amenorrhea, and urinary human chorionic gonadotropin (hCG) was positive. Because irregular bleeding was detected, the patient consulted a medical practitioner. The diagnosis was chemical abortion, because laboratory investigations showed that urinary hCG was positive. At 18 months after delivery, she was admitted to hospital with irregular menstrual bleeding and sudden onset of lower extremity edema. Laboratory investigations showed severe proteinuria, urinary hCG positivity, low serum protein (5.0 g/dL), and low serum albumin (2.7 g/dL). A renal biopsy showed thrombotic microangiopathy. She was diagnosed with nephrotic syndrome, and she was treated with steroid. Computed tomography (CT) for etiological determination revealed an enlarged uterus, and she was referred to our department. Transvaginal ultrasound and magnetic resonance imaging (MRI) showed an enlarged uterus, but the tumor was not found clearly in the uterus (Figure 1B). The serum β-hCG level was 289 mIU/mL. During hysteroscopy, villus-like pathological changes were observed in the

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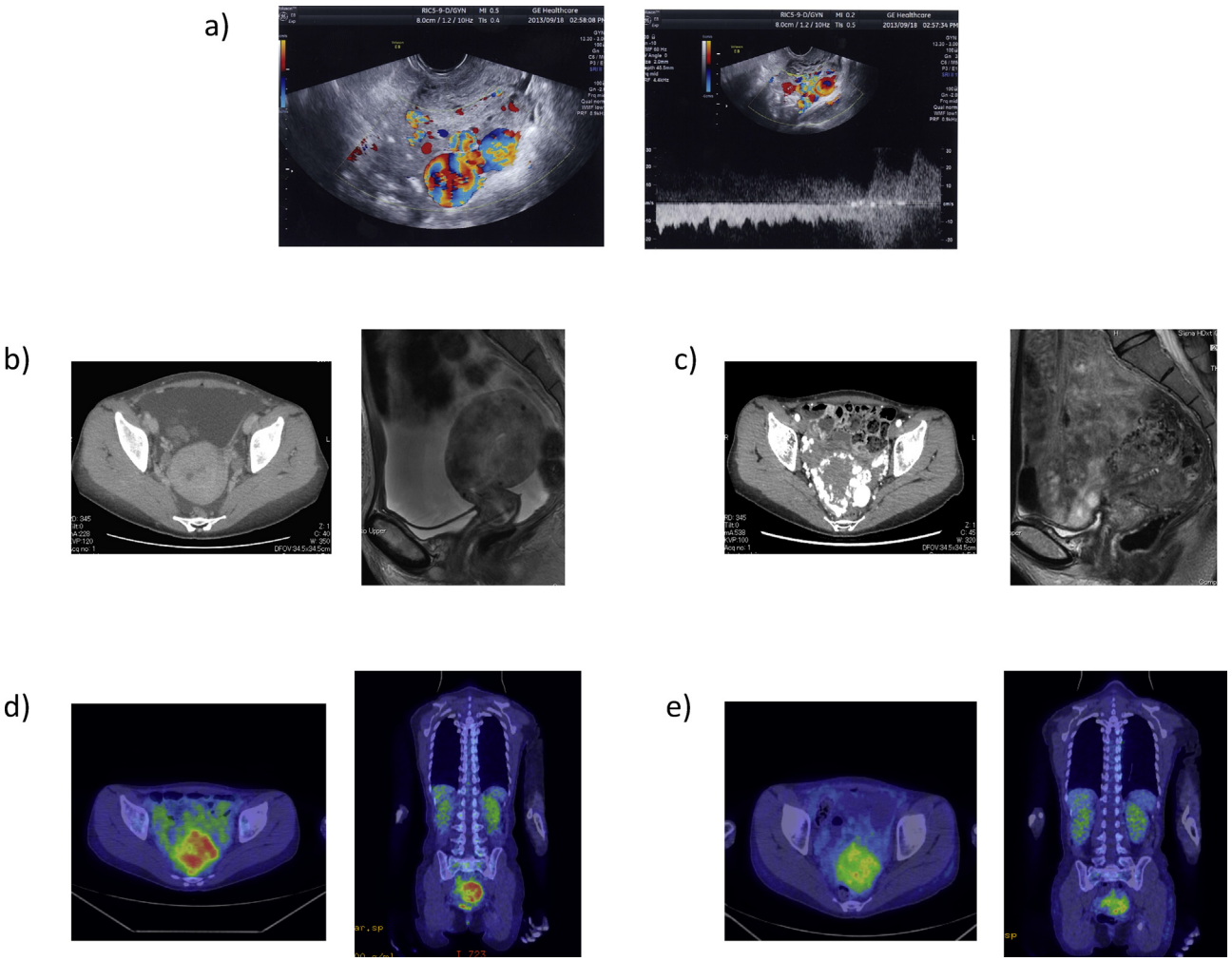

Figure 1. (A) Before total hysterectomy, transvaginal color Doppler ultrasound revealed an enlarged uterus filled with a hypervascular mass in the endometrium and myometrium. (B) Before MTX therapy, CT and MRI showed an enlarged uterus, but the tumor was not found clearly in the uterus. (C) Before total hysterectomy, CT and MRI revealed an enlarged uterus filled with a hypervascular mass in the endometrium and myometrium, enlarged vessels in the myometrium, and enlarged gonadal vessels. After MTX treatment, repeat PET-CT showed uptake in the accumulation images, but there was a clear decrease in maximum standardized uptake value. (D) Before MTX treatment; (E) after MTX treatment. CT = computed tomography; MRI = magnetic resonance imaging; MTX = methotrexate; PET = positron emission tomography.

uterine cavity, so hysteroscopic biopsy was performed to make a definitive diagnosis. Microscopically, there were increased numbers of intermediate trophoblasts, and tumor cells were arranged in sheets and cords throughout the smooth muscle fibers of the myometrium, without invading blood vessel walls in the myometrium. Immunohistochemically, the tumor cells were positive for hPL and hCG, and the Ki-67 labeling index was ~20%. She was diagnosed with an exaggerated placental site (EPS), because the Ki-67 labeling index was high, but a neoplastic lesion was not detected clinically and mitotic figures were not found by histopathology. In addition, the entire uterus showed uptake on 18F-fluorodeoxyglucose (18F-FDG) positron emission tomography (PET)-CT (Figure 1D). Methotrexate (MTX) therapy was commenced after diagnosis of EPS. MTX therapy was given as an intramuscular injection of 20 mg/day for 5 days. Serum β-hCG values decreased from 292 mIU/mL to 88 mIU/mL after MTX therapy. On PET-CT, the accumulation image accepted it, but showed a clear decrease in the maximum standardized uptake value (Figure 1E). We were going to continue MTX therapy subsequently. However, MTX therapy had to be discontinued because of severe side effects such as stomatitis. Therefore, she was followed up in the outpatient department. In the meantime, serum β-hCG values decreased gradually from 88 mIU/mL to 22 mIU/mL, but they did not decrease to within the normal range (< 0.5 mIU/mL). Seven months after MTX therapy, transvaginal color Doppler ultrasound showed an enlarged uterus filled with a hypervascular mass in the endometrium and myometrium, which was confirmed by MRI and CT (Figures 1A and 1C). She was diagnosed with uterine arteriovenous malformation after EPS. Based on this diagnosis, we performed total hysterectomy. Microscopically, the tumor cells consisted of intermediate trophoblasts arranged in sheets and cords throughout the smooth muscle fibers of the myometrium, with invasion of the blood vessel walls in the myometrium, nuclear atypia, and an increased number of mitotic figures (4–10/10 high-power fields). Immunohistochemically, the tumor cells were positive for hPL and hCG, and the Ki-67 labeling index was > 13% (Figure 2). She was diagnosed with PSTT, International Federation of Gynecology and Obstetrics (FIGO) Stage I disease. Serum β-hCG values decreased steadily from 22 mIU/mL prior to total hysterectomy to within normal ranges 1 month postoperatively. Her nephrotic syndrome resolved after hysterectomy. Monthly serum β-hCG values have been negative for 15 months, and she has remained without evidence of recurrence.

We present a case of PSTT diagnosed only after total hysterectomy. Although the application of hysteroscopy is useful in

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