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Case Report

Diagnosis and management of a rare case with placental site trophoblastic tumor

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ABSTRACT

Objective: Placental site trophoblastic tumor (PSTT) is rare and is characterized by a slow growth. The objective of this report is to present a case of PSTT associated with irregular vaginal spotting that occurred 1 year after normal vaginal delivery.
Case Report: This report provides interesting ultrasound, hysteroscopy, and histology findings of PSTT. It is difficult to make a clinical diagnosis of PSTT at an early stage. Without the use of immunohistochemical analysis, PSTT may evade histological detection. An operative hysteroscopy using electrocauterization reduces active bleeding during the removal of PSTT with markedly engorged tumor vessels.
Conclusion: Transvaginal sonography using color Doppler imaging plays a vital role in identifying residual PSTT with microscopic infiltration to the myometrium and a negative serum β-human chorionic gonadotropin level.

Introduction

Placental site trophoblastic tumor (PSTT) derives from intermediate trophoblastic cells. The tumor infiltrates the smooth muscle cells of decidual spiral arterioles during early normal gestation. PSTT has a malignant potential to invade the myometrium or metastasize. PSTT accounts for 0.23–3% of gestational trophoblastic disease and may arise several months or years after a normal or abnormal pregnancy [1]. Most patients present with nonspecific symptoms and signs at diagnosis, such as abnormal vaginal bleeding, amenorrhea, mild elevated serum β-human chorionic gonadotropin (β-HCG) levels (< 1000 mIU/mL in 79% of cases) [2], and an endometrial or myometrial nodule on transvaginal sonography [3]. The clinical course of PSTT is unpredictable. A high β-HCG level and a deep myometrial infiltration are likely to be associated with metastases to the lung, liver, vagina, intestine, brain, lymph nodes [1,4,5], or even through the placenta to the fetus [6].

Case Presentation

A 30-year-old patient, gravid 1 para 1, presented with irregular vaginal spotting for several days. She previously gave birth to a normal baby, weighing 3100 g, through the vagina. The patient was still breastfeeding her 1-year-old female baby. Her serum β-HCG level was 15 mIU/mL. Transvaginal sonography revealed an endometrial nodule measuring about 1.3 cm × 1.2 cm × 1.3 cm, which contained low-resistance blood flow as seen from color Doppler imaging. Under a suspicion of residual placental tissues, an operative hysteroscopy was arranged. Active bleeding occurred during cervical dilation and impeded further hysteroscopic surgery. After endometrial curettage, tumor bleeding was controlled by intravenous administration of tranexamic acid and external compression using SURGICEL FIBRILLAR (Ethicon, Somerville, NJ, USA; oxidized regenerated cellulose) and an inflated Foley balloon inserted into the uterine cavity.

Histology of the specimens demonstrated products of conception without evidence of trophoblastic disease. Follow-up serum β-HCG levels decreased to 5 mIU/mL, but sonography revealed that the endometrial lesion with low-resistance blood flow still persisted. Under the impression of focal placenta accreta, 50 mg/m^2 of methotrexate was injected through the uterine

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cervix. One week later, another follow-up serum β-HCG level measurement showed an increase to 9 mIU/mL. Follow-up sonography revealed an increase in tumor size and vascularity (Figures 1A and 1B). Diagnostic hysteroscopy clearly showed a 1.5-cm tumor with engorged vessels protruding into the uterine cavity (Figure 1C). After careful dilation of the cervical canal, the tumor was removed as thoroughly as possible under an operative hysteroscopy using electrocauterization (Figure 1D). The final histologic report indicated PSTT with up to four mitotic figures per high-power field (Figure 2A). Immunohistochemical analysis showed positive staining for human placental lactogen and Ki-67 (Figure 2B). Another follow-up serum β-HCG level showed a decrease to 1.2 mIU/mL. Positron emission tomography (PET) revealed neither a residual uterine lesion nor metastatic sites. A repeated gray-scale transvaginal sonography did not show any irregularities, but color Doppler imaging identified low-resistance trophoblastic flows in the myometrium (Figures 2C and 2D), consistent with a residual PSTT. After discussion with the couple, a laparoscopic-assisted vaginal hysterectomy was performed. A 2.5-mm residual PSTT infiltrating to the myometrium was confirmed by microscopic examinations and immunohistochemical analysis (Figure 3).

Discussion

It is difficult to make a clinical diagnosis of PSTT at an early stage because of mild symptoms, a slight elevation of β-HCG level, a small lesion in the uterus, and the rarity of PSTT. The suspicion of a retained placenta or focal placenta accreta tends to be aroused when clinicians first encounter this entity. Invasive mole or choriocarcinoma is usually associated with a high and progressively growing serum β-HCG level. Of note, the presence of prominent vessels and low-resistance trophoblastic flows noted using color Doppler imaging could not distinguish PSTT from placenta accreta or residual placental tissues [3,7]. Nevertheless, placenta accreta is known to be responsive to methotrexate therapy [8].

Without the use of immunohistochemical analysis, PSTT may also evade histological detection [1]. PSTT is characterized by positive staining for inhibin, human placental lactogen, cytokeratin, epidermal growth factor receptor, vascular endothelial growth factor, and pregnancy-associated major basic protein. Pregnancy-associated major basic protein is a marker for intermediate trophoblasts and is useful to differentiate PSTT from epithelioid trophoblastic tumors.

PSTT associated with engorged tumor vessels, partly due to arteriovenous shunt, is prone to contact bleeding during the process of cervical dilation and tumor excision. The active bleeding may obscure the visual field of hysteroscopy and make further procedures of the operative hysteroscopy difficult. After delicate dilation of the cervix, careful hysteroscopic electrocauterization can prevent tumor bleeding and achieve the removal of the visible PSTT bulging into the uterine cavity. After tumor removal by operative hysteroscopy, the residual PSTT with microscopic invasion to the myometrium may not be detectable using serum β-HCG, PET, or a gray-scale transvaginal sonography. In this case, transvaginal sonography using color Doppler imaging seemed to be the most sensitive examination to detect the presence of residual PSTT.

PSTT is characterized by a slow growth, 61% resistance or incomplete response to chemotherapy [9], and a tendency to metastasize through lymphatic pathways. During the early stages, hysterectomy is the best way to cure PSTT. Although partial resection of the uterus has been reported in patients with the desire to preserve fertility [10], it is difficult to guarantee complete excision

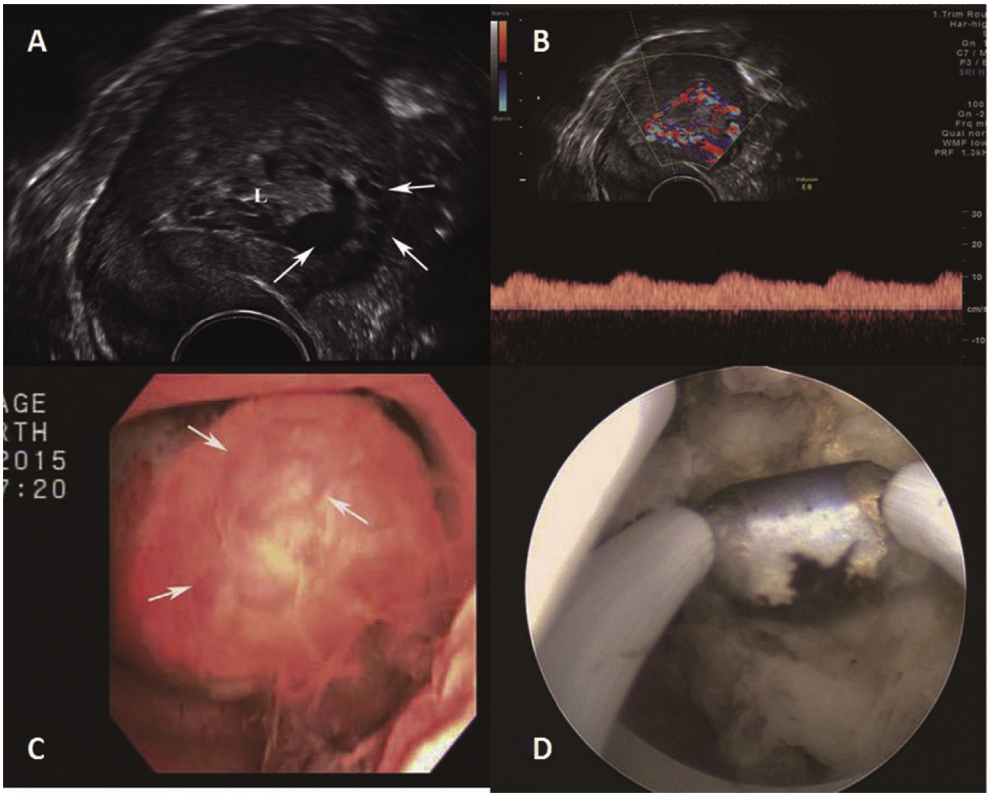

Figure 1. (A) Gray-scale transvaginal sonography displayed an endometrial lesion (L) with multiple sonolucent areas (arrows), (B) which were found to be composed of low-resistance tumor vessels (resistance index of 0.32) in color Doppler imaging. (C) Hysteroscopy displayed a mass with obvious tumor vessels (arrows). (D) Electrocauterization was performed to prevent bleeding during removal of the tumor.