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Case report

Postmolar choriocarcinoma after an interval of 7 years: Case report and literature review

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ABSTRACT

Choriocarcinoma is a rare pregnancy-related malignancy. The majority is arising from non-molar pregnancy. Here we report a patient who was diagnosed with postmolar choriocarcinoma after an interval of 7 years. Before surgery, we suspected the diagnosis of the patient was intramural pregnancy or choriocarcinoma. Laparoscopy was performed and hysterectomy was carried out. Postoperative pathological evaluation of the surgical specimen confirmed the case was choriocarcinoma. Hysterectomy through laparoscopy was feasible and safe for selected patients.

Introduction

Gestational trophoblastic disease (GTD) encompasses a unique group of uncommon but interrelated conditions that derived from the anomalous growth of placental trophoblastic tissue, which differ in propensity for regression, invasion, metastasis, and recurrence. The World Health Organization (WHO) divides GTD into the following groups: pre-malignant disease represented by complete hydatidiform moles (CHM) and partial hydatidiform moles (PHM), and malignant forms represented by invasive moles, choriocarcinoma, placental site trophoblastic tumor (PSTT), and epithelioid trophoblastic tumor (ETT).[1,2] This disease is very uncommon and is often highly complicated. In addition, the incidence of the disease is declining, presumably due to improved medical care and to social, economic and educational changes.[3] Hydatidiform mole accounts for about 80% of all cases of GTD, and occurs in 0.6–1.1/1000 pregnancies in North America.[4] Invasive mole develops when molar villi invade the myometrium through venous channels. The development of local invasion after CHM occurs in about 3%–5% of patients, and metastatic disease is rare.[5] While choriocarcinoma is rarer than hydatidiform mole, occurring in 1–9.2/20,222–40,000 pregnancies.[6] It is usually arises in the uterine cavity. And an other type of this tumor is classified in the germ cell tumors of the ovary that arises from this organ. Extrauterine choriocarcinoma is rarely seen in clinical practice.[7] Gestational choriocarcinoma may follow any type of pregnancy and mostly is associated with coincident or antecedent pregnancy. These pregnancy may be a miscarriage, a term pregnancy, or a molar pregnancy. The majority arise following a non molar pregnancy. Here we report a case that is diagnosed to be postmolar choriocarcinoma after a long interval of 7-year, which is difficult to distinguish it from intramural pregnancy pre-operatively.

Case report

A 47-year-old woman (Gravida 2, para 1) was admitted to our clinic with the complaint of vaginal bleeding for one month after 3-month menopausal. Before this, she had an irregular menstrual cycle with 3–5 days of menstrual period and 30–60 days of menstrual cycle. She used a condom or rhythm method for contraception. In this month, she had vaginal blotting accompanied with lower abdomen dull pain that can be endured. No frequent urination, odynuria, anus belly, fever and others discomfort was accompanied. So she did not pay attention to it and did not to see doctor. After 20 days blotting, the vaginal bleeding increased, she went to a community hospital. Ultrasonography has detected an echogenic focus about 6.4 × 4.6 × 5.1 cm in posterior myometrium of the uterus. No physical or other auxiliary examination was

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suggested. So she came to our hospital. Ultrasonography showed a strong echo mass of 6.4 × 5.1 × 7.1 cm at the intramural layer in left fundus of the uterus which slightly convex to the serous layer. The serum level of β-HCG was 65,576.8 mIU/ml. There was no positive foundation of her lung CT. Intramural pregnancy was suspected and hospitalization was suggested. In the course of the disease, the patient didn't accompanied with cough, phlegm, hemoptysis and weight lose. She had a history of hydatidiform mole 7-year-ago, and had received two times of evacuation. She received follow-up till the serum β-HCG decreased to be normal. She did not have a routine physical examination.

After admission, we performed physical examination and ultrasonic acoustic imaging. Bimanual examination has demonstrated an enlarged uterus as 10-gestation-week in the lower part of abdomen without tenderness. Doppler analysis revealed an heterogenic mass measuring 7.1 × 5.4 × 5.6 cm with high blood flow and low resistance vascularity, and area of 4.9 × 4.9 × 5.9 cm lack of contrast agent was found in the mass (see in Fig. 1). The serum level of β-HCG was 74,145.8 mIU/ml, which was higher than that of the first result (4-say interval). Because of the nonspecific characteristics of ultrasonography, we arranged pelvic CT the next day after admission, the results showed a low density lesion on the upper posterior uterine wall that was no clear boundary with uterine myometrium. It was heterogenic after T2-weight imaging (see in Fig. 2). So uterine choriocarcinoma was suspected, and intramural pregnancy could not excluded. After a sufficient preoperative preparation and content with patient, laparoscopy was performed under systematic anesthesia. An enlarged uterus as 10-week gestational size was found. A maroon and crispy lesion was on the left posterior myometrium converting to serous layer and was infiltrating anterior serous muscular layer of the rectum. Bilateral ovaries and tubes were normal. We removed the lesion firstly. The frozen section result showed scattered low-differentiated carcinoma cells in the extensive hemorrhage and necrosis tissue. Because the patient had no desire for fertility, we performed hysterectomy. The final pathological findings (see in Fig. 3) were uterine choriocarcinoma accompanied with extensive hemorrhage and necrosis, infiltrating the whole muscular layer. Results of IHC were P63++, CD146+++, CK-P+++, PLAP-, HCG+++, 90% positive rate of Ki 67. According to the stage and scoring system (see in Tables 1 and 2), the diagnosis was postmolar choriocarcinoma, stage 1, score 9. Treatment is based on classification into risk groups defines by stage and scoring system.[8] For patients with the high-risk disease (FIGO stage IV and stages II–III score ≥7) should be treated initially with multi agent chemotherapy with or without adjuvant surgery or radiotherapy.[9] So when we confirmed the diagnose after surgery, EMA-CO regimen was given in order to prevent recurrence and metastasis. Now the patient was in following-up with a fine condition.

Discussion

As a kind of highly malignant disease, choriocarcinoma is characterized by hyperplastic and anaplastic syncytioblasts and cytotrophoblasts, hemorrhage, tissue necrosis, and absence of chorionic villi. It spreads by directly invading the myometrium and vascular channels, resulting in involvement at distant sites, most commonly the lungs (80%), vagina (30%), brain (10%), liver (10%).[10,11] The clinical symptoms and signs may be nonspecific, making the diagnosis difficult. When patients present with vaginal bleeding after term pregnancies or miscarriages, abnormal serum level of β-HCG, choriocarcinoma should be considered. The Cancer Committee of the International Federation of Gynecologists and Obstetricians (FIGO) has established the guidelines for the diagnosis of postmolar GTN. The components include at least 1 of the following: ① HCG plateau for 4 consecutive values over 3 weeks; ② HCG rise of ≥10% for 3 values over 2 weeks; ③ HCG persistence 6 months after molar evacuation; ④ histopathologic diagnosis of choriocarcinoma; or ⑤ presence of metastatic disease.[12] The following two points are important for judging invasive mole or choriocarcinoma: ① The interval of pre-molar evacuation: invasive mole is less than one year, while choriocarcinoma is more than one year; ② Histological examination: for invasive mole, villi can be found microscopically, while for choriocarcinoma, only trophoblastic cells can be found.[13] For our patient, she had a history of complete mole 7-year-ago and had received evacuation. The level of serum β-HCG decreased to normal in the follow-up. After that, she did not conceive. The final pathological diagnose confirmed postmolar choriocarcinoma. It is a rare case for its long interval from her mole history. It is suggested that when patients were older than 40-year-old, the risk of developing postmolar GTN will increase 37%, while it will increase 56% when older than 50-year-old.[14] Pathological characteristically, choriocarcinoma is associated with proliferating sertoli cells that losing the normal histological manifestation and infiltrating uterine muscular layer extensively, which causes necrosis, hemorrhage and distant metastasis.[15] In gross appearance, the uterus increases irregularly in size and is soft. The surface shows blue purple nodules sometimes. When viewed sectionally, the focus is maroon, accompanied with hemorrhage, necrosis and infection. Actively growth of sertoli cells can be seen around the foci, which can infiltrate vascular.

GTN is the only malignancies that can be diagnosed basing on clinical symptoms and signs without pathological evidence, which agreed by FIGO and ISGC. Because of the particularity, GTN is

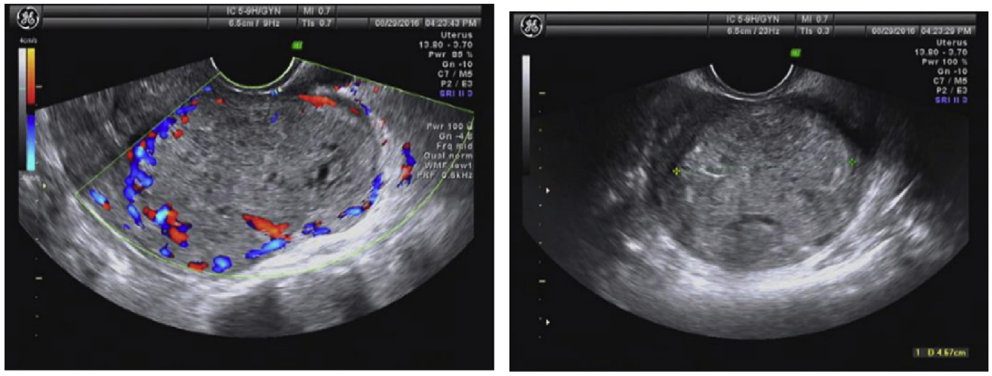

Fig. 1. The ultrasonography showed an heterogenic mass with high blood flow and low resistance vascularity.