Case report

Post-partum choriocarcinoma mimicking retained adherent placental remnants: A rare case report

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ABSTRACT

Patients with post-partum choriocarcinoma have poorer prognosis than those with hydatidiform mole due to metastasis and delayed diagnosis. We report a rare case of post-partum choriocarcinoma manifesting as retained placental remnants caused by placental adhesion, wherein a delay in diagnosis occurred due to failure of serum human chorionic gonadotrophin (hCG) surveillance and pathological examination of the placenta or curettage tissues. In summary, it is important to pay more attention to retained adherent placental remnants by pathological examination for abnormal placenta and β-hCG surveillance for retained placenta to shorten the interval between antecedent pregnancy and diagnosis.

1. Introduction

Choriocarcinoma, a malignant type of gestational trophoblastic neoplasia, most commonly occurs after an abnormal pregnancy and may occur after any type of pregnancy, including abortion, term pregnancy, premature delivery, and stillbirth [1]. The incidence of choriocarcinoma following term delivery is approximately 1 in 50,000 births [2, 3]. It has been reported that choriocarcinoma following a full-term birth accounts for 16.2–22.5% of choriocarcinomas [2]. Post-partum choriocarcinoma is divided into two subgroups: short (<4 months) and long (>4 months) interval groups, according to the time interval between disease onset and previous pregnancy [2, 4]. The most common symptom of patients with post-partum choriocarcinoma is vaginal bleeding, especially in patients belonging to the short interval group [2, 4]. Vaginal bleeding can mimic diseases manifested as post-partum haemorrhage, of which the retained placenta is a major cause. It is easy to miss diagnosis of post-partum choriocarcinoma in this condition. We present a rare case of post-partum choriocarcinoma manifesting as retained adherent placental remnants with vaginal bleeding after delivery.

2. Case report

A 32-year-old woman (gravida 4, para 3) complained of uterus placenta remnants. Her previous two deliveries were normal vaginal deliveries, with no history of gestation trophoblastic disease. Her family history, drug history, and allergy history were unremarkable. The patient underwent a vaginal delivery of a healthy female infant in a local hospital four months ago. Her antenatal ultrasound in this pregnancy demonstrated normal placenta without evidence of previa or placental invasion. The third stage of labour was complicated by retained adherent placenta, which was removed manually. After delivery of the placenta, ultrasound detected 'placental remnants' about 3 cm in diameter in the uterine cavity. Dilatation and curettage under ultrasound guidance was performed, but was immediately stopped due to heavy vaginal bleeding. After treatment with intravenous oxytocin and prophylactic antibiotics, the patient was discharged on post-partum day 3 in a stable condition, with instructions for serum β-hCG surveillance. However, the patient did not follow this instruction because she did not complain of any discomfort or vaginal bleeding. Two months after delivery, ultrasound still detected 'placental remnants' 0.6*0.5cm in diameter in the uterine cavity. The patient refused treatment because she had no complaints or symptoms. Three months after delivery, the patient was referred to a local hospital because of light vaginal bleeding for 2 weeks. Her serum β-hCG level was 38371 mIU/ml (Normal serum β-hCG level is <10 mIU/ml). Ultrasonographic examination detected an intrauterine mass (diameter, 2.1 × 1.8 cm) in the right uterine horn. Chest radiography revealed no abnormalities. A diagnosis of late secondary haemorrhage due to retained placenta was made. And then hysteroscopic curettage was performed to...
remove this detected ‘placental remnants’. Blood loss during the operation was 950 ml. Histopathological examination of intraterine objects indicated trophoblasts and decidual tissues in this local hospital. On postoperative day 3, the serum β-hCG level declined to 8,008 mIU/ml, and the patient was discharged with regular outpatient β-hCG monitoring. One week later, serum β-hCG level declined to 4,653 mIU/ml, and 2 weeks later, it rebounded to 29,566 mIU/ml. Accordingly, diagnose of gestational trophoblastic tumours were suspected then. The patient was referred to our hospital for further examination and treatment. Approximately 6 scattered tiny nodular shadows of 0.1–0.6 cm in diameter were observed in the right middle lobe and inferior lobe on thoracic computed tomography (CT) (Figure 1A, C). Magnetic resonance imaging (MRI) showed abnormal mixed signals in the right corner of the uterus, 3.2 × 3.8 cm in diameter; the edge of the lesion was clearly enhanced in the early stage, and the boundary with the adjacent muscle layer was unclear (Figure 2A and B). Brain CT findings were unremarkable. Tissues obtained from hysteroscopic curettage were re-examined by pathologist in our institution, and histopathologic diagnosis of ‘choriocarcinoma’ was made (Figure 3A&B). Serum β-hCG level was 84409.8 mIU/ml. According to the modified Bagshawe’s scoring system by the World Health Organization, the patient scored 7, placing her in the high-risk group (>7). The tumour was classified as FIGO stage III. EMA-CO systematic chemotherapy was initiated. After four cycles of chemotherapy, β-hCG levels declined to normal and thoracic computed tomography scan showed scattered metastatic lesions in the bilateral lungs regressed (Figure 1B and D) however, localised disease in the uterus remained. The patient did not wish to preserve fertility since she already had three children, then laparoscopic total hysterectomy and bilateral salpingectomy were performed. The gross appearance of the uterus after total hysterectomy indicated a lesion located at the uterine fundus (Figure 3C). The patient received two cycles of postoperative systemic therapy with EMA-CO. She is currently in follow-up for more than 12 months with a serum β-hCG level of <2 mIU/ml.

Informed consent was obtained and the study has been approved by the ethics committee of West China Second University Hospital of Sichuan University (2020076).

3. Discussion

Patients with post-partum choriocarcinoma have poorer prognosis than gestational trophoblastic neoplasm occurred after hydatidiform mole or abortion due to widespread metastatic disease and delays in diagnosis [4, 5]. An increased time interval between disease onset and previous pregnancy is associated with worse outcomes [6]. Early diagnosis is critical for directing the clinical management of post-partum choriocarcinoma. In this case, incomplete placenta was delivered after manual extraction due to placentation adhesion, followed by evacuation and curettage which was soon stopped due to heavy vaginal bleeding. Neither the placenta nor evacuation tissue was sent for further pathological examination. There is a possibility that this case may have arisen from intraplacental choriocarcinoma, which usually occurs in the third trimester and is often asymptomatic with no macroscopic placental abnormalities [7]. The presentation of intraplacental choriocarcinoma ranges from clinically silent lesions to symptoms of metastatic disease. Since pathological examination of the placenta is not routinely performed and half of the patients are asymptomatic, it is likely that there are missed cases and the true incidence may be higher than data reported [7].

Retained placenta is a common cause of postpartum hemorrhage with an incidence of 3–5% after routine vaginal delivery [8]. According to previous studies, surgery may be avoided in cases of abnormal placental adherence by leaving the placenta in situ [9]. Differential diagnoses between retained placenta and choriocarcinoma is necessary, since postpartum abnormal vaginal bleeding is the same initial presentation for both of them. Choriocarcinoma is characterized by myometrial and vascular invasion with high incidence of pulmonary metastasis in the form of nodules with surrounding ground glass opacities [10]. Combined gray-scale and color Doppler US allow real-time assessment of the uterine cavity and blood flow, vascularity of the uterine mass in addition to the myometrial invasion suggested a malignant process. CT or MRI can serve as diagnostic adjuncts in complicated cases [11]. Evacuation is often used to stop vaginal bleeding and acquire tissue for pathologic examination for differential diagnoses [2].

Figure 1. Thoracic computed tomography scan showing scattered metastatic lesions in the bilateral lungs (A, C arrows) and regressed lesions after systematic chemotherapy (B, D arrows).
Besides, in such cases, hCG surveillance is critical for early diagnosis. It is postulated that the serum β-hCG level depends on the amount of active trophoblasts that are still attached to the uterine wall. Serum hCG levels will decline due to spontaneous degeneration of the retained placenta. The half-life of serum hCG elimination is 5.2–6.1 days during conservative management of retained adherent placenta [12, 13]. It is reported that serum hCG level drops to negative 21–35 days after delivery when leaving adherent placenta remnants in situ [14], though it extends to 10 weeks after delivery in a few cases [15]. While the half-life of serum β-hCG was 32.3h after normal delivery without placenta remnants. Thus, it is important to be alert about the possibility of choriocarcinoma in β-hCG positive cases diagnosed as retained placenta 6 weeks after delivery. In this case, the patient did not follow the instructions to monitor serum β-hCG level after discharge. Two months after delivery, ultrasound still detected ‘placental remnants’ in the uterine cavity. The patient refused treatment because she did not experience any discomfort or symptoms. Unfortunately, serum β-hCG levels were not examined at that time and may miss the opportunity for early diagnosis. In our own opinion, surveillance of serum β-hCG will assist to evaluate the activity of trophoblasts and absorption of placenta, more over, in rare conditions, it will help for early diagnosis of post-partum choriocarcinoma when β-hCG level increases sharply or decreased abnormally.

This is a rare case of post-partum choriocarcinoma misdiagnosed as a retained placenta. Histological study of placental tissue and surveillance of serum β-hCG may lead to early diagnosis and initiation of treatment. Obstetricians should have increased awareness of the symptoms and the importance of early diagnosis of post-partum choriocarcinoma. Early recognition of post-partum choriocarcinoma may reduce mortality by limiting the extent of distant metastasis.
In summary, it is important to pay more attention to retained adherent placental remnants by pathological examination for abnormal placenta and β-hCG surveillance for retained placenta to shorten the interval between antecedent pregnancy and diagnosis.

Declarations

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

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Data availability statement

Data will be made available on request.

Declaration of interests statement

The authors declare no conflict of interest.

Additional information

No additional information is available for this paper.

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