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Case Report

Retroperitoneal Hematoma as an Atypical Presentation of Choriocarcinoma: A Case Report

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Abstract
A 38-year-old female with an etonogestrel implant in place and history of previous ectopic pregnancy presented with acute abdominal pain and vaginal bleeding. She was found to have a beta-hCG of >12,000 mIU/mL and free fluid noted on a focused assessment with sonography in trauma exam. She underwent an emergent diagnostic laparoscopy due to the suspicion of a ruptured ectopic pregnancy. Findings at the time of surgery included a normal-appearing uterus and left fallopian tube, a surgically absent right fallopian tube and large volume hemoperitoneum with a rapidly expanding left retroperitoneal hematoma. A postoperative computerized tomography (CT) angiogram suggested active bleeding from a pseudoaneurysm of the left renal artery which was successfully embolized by interventional radiology. Biopsy confirmed gestational trophoblastic neoplasia (GTN) after metastases to the brain. In this report, we describe the details of this case of GTN with an atypical presentation.

This case was presented at the 2023 ACOG Annual Clinical and Scientific Meeting, Baltimore, Maryland 5/2023.

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Introduction

Gestational trophoblastic neoplasia (GTN) encompasses a spectrum of disorders from abnormal proliferation of the placental trophoblast and includes invasive mole, choriocarcinoma, placental site trophoblastic tumor among others [1]. The typical initial presentations include vaginal bleeding, irregular sized uterus, beta-hCG >100,000 mIU/mL, and multiple echoes or focal cystic spaces noted on ultrasound [1]. The most common sites of metastases in GTN include the lungs, brain, and vagina [2]. GTN can be fatal due to distant and rapidly growing metastases to the brain with intracranial hemorrhage or abdominal bleeding from uterine rupture [1]. There are only a few rare case reports describing metastases to the spleen or to the kidneys [2–5]. In this report, we highlight the unusual case of a woman who presented with spontaneous massive retroperitoneal hematoma caused by GTN.

Case Presentation

A 38-year-old gravida 6, para 2 woman with an etonogestrel implant in place was presented to the emergency center with sudden onset abdominal pain and vaginal bleeding. She had a history of previous ectopic pregnancy requiring salpingectomy and documented negative pregnancy test at the time of contraceptive implant placement 1 year before presentation. She was found to have a beta-hCG of 12,712 mUI/mL with an acute abdomen and vital signs suggestive of hypovolemia. A focused assessment with sonography in trauma was performed and a large amount of free fluid was seen in the abdominal-pelvic cavity. No intrauterine pregnancy was noted on transvaginal ultrasound. Laboratories revealed a hemoglobin of 10.9 g/dL and creatinine of 0.9 mg/dL. After initial resuscitation, she was taken emergently to the operating room for a diagnostic laparoscopy due to concern for a ruptured ectopic pregnancy with activation of the massive transfusion protocol. On laparoscopy, she was found to have 1.5 L of hemoperitoneum along with a normal-appearing uterus and left fallopian tube and a surgically absent right fallopian tube. The abdomen was thoroughly inspected with no identification of an ectopic pregnancy. A large expanding left retroperitoneal hematoma was visualized and noted to be pushing the sigmoid colon to the right pelvis (Fig. 1). The hematoma extended from below the left round ligament to above the pelvic brim. Blood was noted to be draining from the retroperitoneum into the pelvic cavity from pinpoint peritoneal openings, resulting in large volume hemoperitoneum. Intraoperative hemoglobin returned at 5.4 g/dL.

Due to the expanding hematoma, a laparotomy was performed, and the retroperitoneum was opened without a clear source of the bleeding identified. At this point, the patient was suspected to be coagulopathic given the extensive hemorrhage, and decision was taken to pack the abdomen with laparotomy sponges and the abdomen was closed with an active temporary abdominal closure device. She was taken emergently for a computerized tomography (CT) angiogram and found to have active bleeding from a pseudoaneurysm of the left renal artery. She underwent emergent interventional radiology embolization with good clinical response (Fig. 2). On postoperative day one, the laparotomy sponges were removed and the abdomen was closed. The evaluation after surgery included an endometrial biopsy that showed fragments of inactive endometrium, slightly elevated CA 125 (71 U/mL), elevated LDH (560 U/L), normal CEA (<1 ng/mL), and normal AFP (<2.5 ng/mL). A CT scan of the chest revealed small non-specific lesions which were biopsied with insufficient cellularity noted. She was discharged after stabilization with outpatient plans for serial beta-hCG trending given the clinical suspicion of GTN. The etonogestrel implant remained in place.

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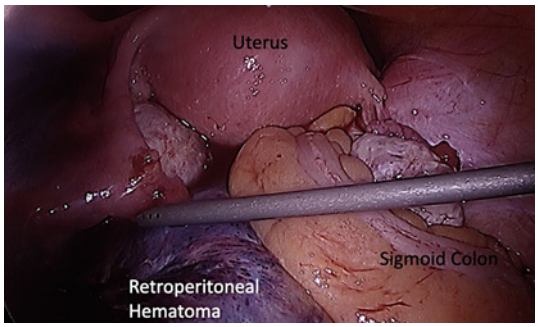

Fig. 1. Image of expanding left retroperitoneal hematoma on initial diagnostic laparoscopy.

After several weeks of follow-up, she remained asymptomatic and her beta-hCG outpatient had decreased to 6,230 mUI/mL. However, she was readmitted to the hospital due to new symptoms of headache, dizziness, and imbalance with imaging concerning for 6 mm right cerebellar infarct and small left occipital lesions (Fig. 3). Beta-hCG had risen to 100,811 mUI/mL. During her hospital stay, the patient developed new onset sudden right flank pain and became hemodynamically unstable. Imaging revealed a 16-cm right perinephric hematoma, and the patient underwent a successful emergent embolization of the right renal artery by interventional radiology (Fig. 3). CT scan of the chest revealed larger lesions suggestive of metastasis. She was diagnosed with GTN with a World Health Organization (WHO) of 14. She therefore received inpatient chemotherapy with cisplatin/etoposide induction for 3 cycles followed by etoposide, methotrexate, actinomycin, cyclophosphamide, and oncovorin (EMA-CO). After the first cycle of EMA-CO, she developed a hemothorax secondary to bleeding from pulmonary metastasis and a chest tube was placed. The patient also underwent gamma knife surgery to treat her brain metastases. She then received seven cycles of EMA-CO with good response. Her beta-hCG level decreased to <5 mIU/mL and she was placed on surveillance with plans to repeat imaging in 3 months. However, she presented with symptoms of headache, dizziness, and blurry vision and imaging revealed acute right occipital hemorrhage concerning for new metastasis. She underwent craniotomy with pathology, resulting in metastatic choriocarcinoma. Her b-hCG increased to 20,832. The patient continues to recover and continues to undergo chemotherapy for the brain metastases. Her most recent b-hCG is 9, a year after initial presentation.

Discussion

GTN can be difficult to diagnose due to diverse initial presentations. Our case has multiple unusual characteristics that led to delays in diagnosis: (1) initial clinical presentation was consistent with a ruptured ectopic pregnancy; (2) an etonogestrel implant in place for more than 1 year prior to her presentation with a documented negative pregnancy test before its placement; (3) initial abdominal survey with only a retroperitoneal hematoma identified; and (4) no definitive tissue diagnosis. Early diagnosis is critical because GTN is responsive to chemotherapy with >90% of all cases being treatable despite metastatic involvement [1]. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000534036).

Because prompt recognition is important to prevent delays in treatment, there must be a high level of clinical suspicion. Abnormal vaginal bleeding is the most commonly recognized symptom of GTN [1]. Diagnosis can be made clinically using imaging and trending of beta-hCG levels with initiation of treatment, but tissue pathology is definitive. In this case, there was no

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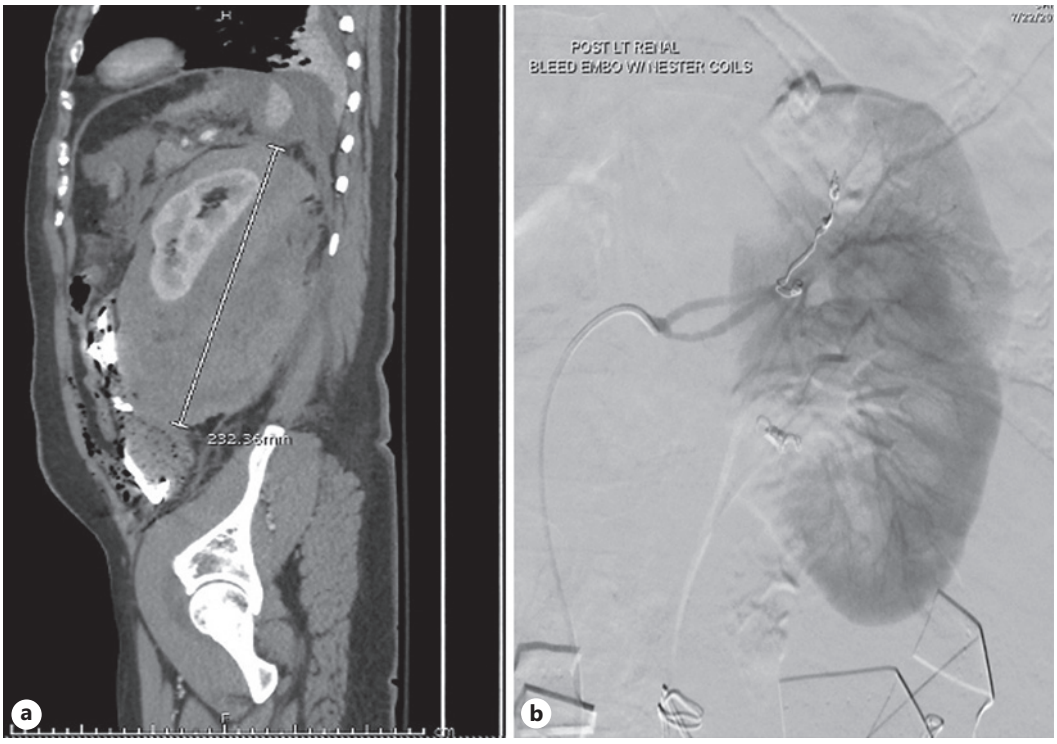

Fig. 2. a Retroperitoneal hematoma measuring 232 mm on postoperative computerized tomography. **b** Post-embolization of the left renal artery by interventional radiology.

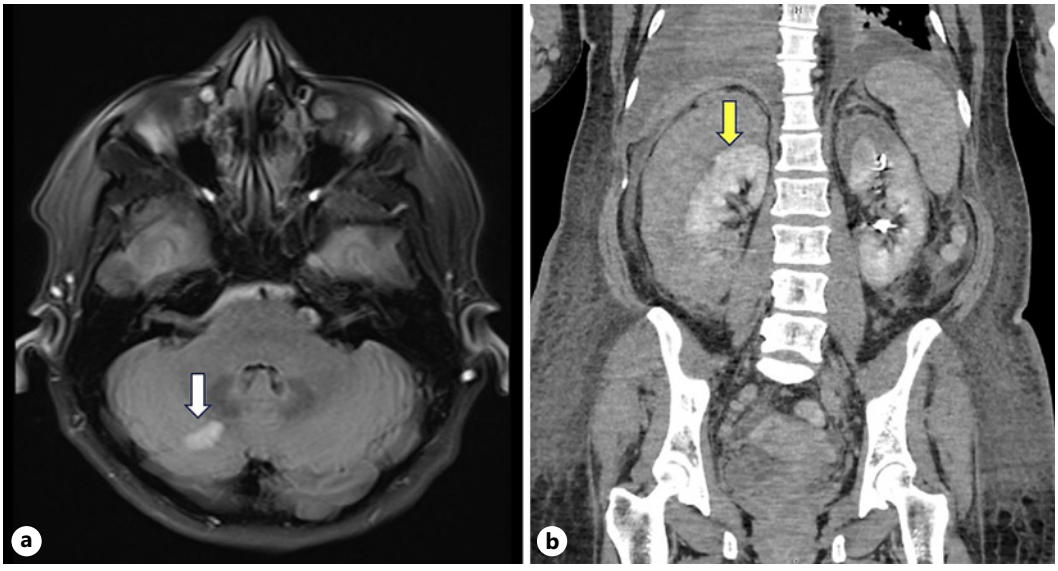

Fig. 3. a Head magnetic resonance imaging with right cerebellar ischemic infarct T2 hyper intensity (white arrow). **b** Right perinephric hematoma on computerized tomography (yellow arrow).

tissue diagnosis until months after treatment; however, clinical suspicion was high enough to warrant treatment. Our patient had biopsies of a lung nodule and an endometrial biopsy during her first admission without a diagnosis of GTN. No intrauterine involvement at the time of diagnosis may be due to autoimmune clearance of the intrauterine disease after distant metastases [6]. Craniotomy due to hemorrhagic brain metastases resulted in tissue diagnoses of GTN months after initial treatment.

Our patient developed retroperitoneal involvement with pseudoaneurysm of the left renal artery with active extravasation with a second episode affecting her right kidney. Several case reports have characterized extrauterine involvement into the retroperitoneal space [3–5]. In a previously reported case, a patient developed diffuse abdominal pain after initiation of chemotherapy with concern for intra-abdominal bleeding [3]. She underwent exploratory laparotomy with profuse bleeding found in the retroperitoneal space. Similarly, a patient with rising beta-hCG levels, vaginal bleeding, and abdominal pain was found to have a ruptured left kidney that eventually required nephrectomy [4]. A subsequent case report described a patient with abdominal pain and abdominal masses on imaging [5]. She underwent a diagnostic laparoscopy with findings of a small hematoma in the retroperitoneal space. Our patient underwent chemotherapy but was then readmitted with another retroperitoneal hematoma that was managed expectantly. Although rare, retroperitoneal disease must be on the differential for complications of GTN metastases due to possible massive hemorrhage that may seriously compromise renal function and be life-threatening. Exploratory laparotomy and activation of a massive transfusion protocol may be necessary initial steps after identification of unstable and rapid bleeding. In cases of unstable retroperitoneal bleeding, consultation of other services such as interventional radiology and urology may be required. The presentation of patients with GTN is highly variable and includes the possibility of back pain with retroperitoneal bleeding. A high clinical suspicion is required in these patients in order to diagnose early and expedite treatment.

Statement of Ethics

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Ethical approval is not required for this study in accordance with local or national guidelines.

Conflict of Interest Statement

The authors do not have any conflicts of interest to report.

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Author Contributions

Truce Pham contributed for the manuscript draft. Mateo G. Leon and Danica Lapid contributed with manuscript edits and figure editing. Kathleen M. Schmeler and J. Alejandro Rauh-Hain performed the manuscript review.

Data Availability Statement

All data generated during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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