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CASE REPORT

A Rare Case of Spontaneous Uterine Perforation with Hemoperitoneum after Normal Pregnancy due to Choriocarcinoma

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ABSTRACT

A patient with one previous term delivery presented with spontaneous uterine perforation due to choriocarcinoma (CC), requiring emergency hysterectomy due to hemoperitoneum. This tumor appears to follow a more aggressive course with extensive metastatic spread. The CC presenting as a spontaneous uterine perforation with intraabdominal hemorrhage is an extremely rare and life-threatening condition. It should be considered in the differential diagnoses of patients in the reproductive phase with a history of recent pregnancy having acute abdomen with hemoperitoneum. Early diagnosis and aggressive chemotherapy is important for patient survival. The patient should be closely followed up for a better outcome.

Keywords: Choriocarcinoma, Term pregnancy, Uterine perforation.

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INTRODUCTION

Trophoblastic diseases comprise a variety of biologically interrelated conditions, which form a clinical spectrum consisting of four distinct clinical pathological entities like (1) molar pregnancy, (2) invasive mole, (3) placental site trophoblastic tumors, and (4) choriocarcinoma (CC). Even though it is rare, acute abdomen due to hemoperitoneum as a result of uterine perforation may be the first symptom of CC. So, it should be considered in women of childbearing age with acute intraabdominal hemorrhage. Presentation with history of molar pregnancy is seen only in about 50% cases; 25% cases have a history of ectopic pregnancy and another 25% develop after normal pregnancy. Previous normal delivery increases the diagnostic challenge as it may occur months to years after a normal pregnancy and may often be mistaken for an ectopic pregnancy. In this study, we report a case of hemoperitoneum with uterine perforation due to CC, which was successfully managed by surgery and chemotherapy.

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CASE REPORT

A patient aged 22 years, P_1L_1, who had normal term pregnancy 6 months earlier, presented in the emergency unit with complaints of amenorrhea of 2 months, continuous pain in lower abdomen, giddiness, and bleeding per vaginum since 4 to 5 days. Patient was afebrile with severe pallor, pulse was 120/min, and blood pressure was 90/60 mm Hg. On per abdomen examination, guarding and tenderness were present. Per speculum examination revealed minimal bleeding through os. Vaginal examination showed uterus enlarged to about 10 weeks and cervical movements were tender. Bilateral fornices were boggy. Urine pregnancy test was found positive. Ultrasound (USG) showed a large mixed echogenic mass on the left uterine fundus with gross amount of free fluid with internal echoes in pouch of Douglas and peritoneal cavity, suggestive of hemoperitoneum.

Emergency laparotomy with a provisional diagnosis of ruptured ectopic was taken up with two units of blood. On opening the abdomen, about 1 L of hemoperitoneum was evacuated. The bilateral tubes and ovaries were normal. The uterus was enlarged and showed a black-colored, highly vascular mass, of size 5 × 4 cm, which completely filled the uterine cavity and perforated the left fundic region and the cervix (Fig. 1). Decision of emergency hysterectomy was taken because of torrential hemorrhage. Histopathological examination revealed a biphasic trophoblast (cytotrophoblast and syncytiotrophoblast) infiltrating the myometrium with characteristic absence of villous pattern with hemorrhage and large areas of necrosis suggestive of gestational CC (Fig. 2).

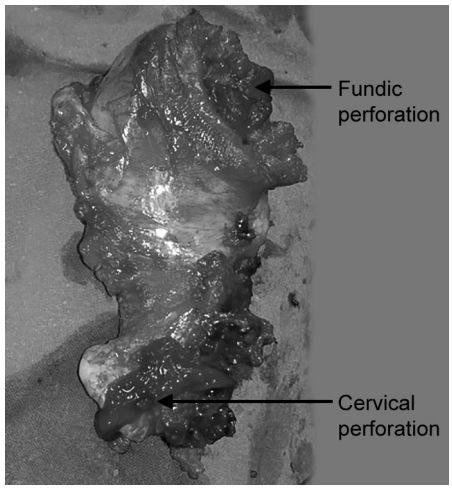

Fig. 1: Hysterectomy specimen showing cervical perforation below and fundic perforation above

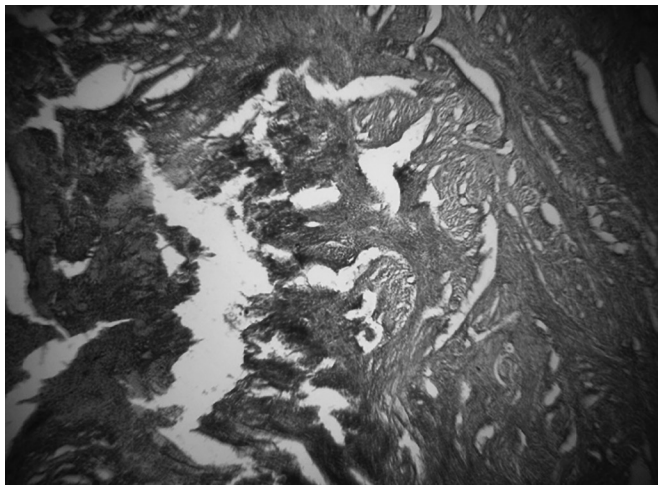

Fig. 2: Histopathological picture (with hematoxylin and eosin stain: 100×) showing intermediate trophoblast in clusters, invading the myometrium, with necrosis. The cells are large with abundant eosinophilic cytoplasm with large irregular nuclei and prominent nucleoli

Postoperatively, chest X-ray (CXR), USG abdomen-pelvis, and computed tomography of brain, chest, and abdomen were done. No signs of metastasis were seen. Serial beta-human chorionic gonadotrophin (β-hCG) values on postoperative days 1 and 7 were 222,400 and 14,548 mIU/L respectively. Confirmation of normal blood tests and USG of abdomen and pelvis was done prior to initiation of chemotherapy.

Etoposide, methotrexate, actinomycin-D, cyclophosphamide, vincristin (EMA–CO) regimen was initiated with EMA on days 1 and 2 and CO on day 8. This cycle was repeated every 2 weeks until β-hCG was normal (5 mIU/L), following which two additional cycles were given. Patient was referred to a specialized oncology center for further management and followed up every month in the outpatient department clinically and screened by β-hCG, USG, and CXR for evidence of metastasis. Till date, no evidence of the same has been found.

DISCUSSION

The incidence of gestational trophoblastic neoplasia (GTN) after a live birth is estimated at 1 in 50,000.[1] Bleeding per vaginum is found to be the most common presenting symptom. Any patient with persistent vaginal bleeding and positive urine pregnancy test should be suspected for GTN.[2] Thus, a high index of suspicion is required for accurate and early diagnosis, which is associated with better prognosis. Previous normal delivery is a poor prognostic factor due to delay in diagnosis and may present with metastasis in liver or central nervous system.

The tumors metastasize by early vascular invasion and they frequently present with acute hemorrhage. Acute hemoperitoneum, as seen in our case, was due to perforation of uterus by the tumor. Differential diagnosis of hemoperitoneum in patients of CC includes ruptured liver metastasis, ovarian metastasis, and ruptured theca lutein cysts. Metastatic CC often presents with symptoms entirely unrelated to the genital tract like stroke, intracerebral hemorrhage, spinal cord tumor, hematuria, gastrointestinal bleeding, pulmonary disease, or malignancy of uncertain origin. Patients may also have signs and symptoms of eclampsia, threatened or missed abortion, ectopic pregnancy, or delayed postpartum hemorrhage. The patients with intrauterine CC presenting with symptoms of threatened or missed abortion have typical history of amenorrhea followed by uterine enlargement, vaginal spotting, and positive pregnancy test. Frequently, these patients undergo emergency laparotomy with a presumptive diagnosis of ruptured ectopic pregnancy. There are only 11 reports on uterine perforation owing to CC in the literature (April 2014, Mesh database). Although the exact cause of uterine perforation in CC is not known, the following theories have been proposed to explain this condition: (1) Trophoblasts can invade the uterine veins, and blood vessel may be damaged. As a result of such damage, multiple infarctions occur due to thrombosis, vascular aneurysms, and intratumoral bleeding; (2) in line with its hypervascular nature, myometrial invasion; (3) depending on the chemotherapy regimen used, rapid trophoblast death can lead to necrosis and eventually uterine perforation; and (4) the presence of a 90% necrotic CC mass led us to speculate that necrosis, in addition to tumoral invasion, may be the underlying cause of perforation in our case.

Treatment is based on classification into risk groups defined by the stage and scoring system. Patients with nonmetastatic (stage I) and low-risk metastatic (stages II and III, score <7) GTN can be treated with single-agent chemotherapy, with resulting survival rates approaching 100%. Patients classified as having high-risk metastatic disease (stage IV and stages II–III, score ≥7) should be

treated in a more aggressive manner with multiagent chemotherapy and/or adjuvant radiation or surgery to achieve cure rates of 80 to 90%. Use of the Fédération Internationale de Gynécologie et d'Obstétrique (FIGO) staging system is essential for determining initial therapy for patients with GTN to assure the best possible outcomes with the least morbidity.[3]

Our patient had a FIGO prognostic score of 7, classified as high risk (age < 39 = 0, antecedent term pregnancy = 2, pregnancy interval 6 months = 1, pretreatment β-hCG 224,000 = 2, largest tumor >5 cm = 2, no metastasis, no prior chemotherapy). So, we initiated treatment with combination chemotherapy and β-hCG progressively decreased during chemotherapy administration and follow-up.

Abdominal hysterectomy may have to be undertaken in emergency situation. The tumor is highly responsive to chemotherapy. Low-risk and nonmetastatic tumors can be treated with methotrexate monotherapy. The EMA-CO protocol is currently the initial treatment of choice for high-risk metastatic GTN because of low toxicity allowing adherence to treatment schedules, high complete response rates, and overall high resultant survival.[4] Chemotherapy for high-risk disease is continued for at least two to three courses after the first normal hCG.

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