



Case report

Fertility-sparing uterine lesion resection in a woman with hemoperitoneum due to invasive mole: A rare case report

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ABSTRACT

Introduction: Gestational trophoblastic neoplasia comprises a unique group of human neoplastic diseases that derive from fetal trophoblastic tissues. The hydatidiform mole is the most common form of GTD, representing 80 % of cases. An invasive mole is a hydatidiform mole characterized by the enlarged hydropic villi invading into the myometrium, into vascular spaces, or into extrauterine sites.

Case presentation: Here is a case with invasive mole after the evacuation of complete molar pregnancy, presented with an acute abdomen. We desired to preserve the uterine because our 21 years old patient doesn't have a child.

Clinical discussion: An emergency abdominal ultrasound scan showed a 47 * 34 * 55 mm ill-defined hyperechoic heterogeneous mass with anechoic cystic vascular spaces within it, in the posterior wall of the uterus away from the endometrium that extended to the serous layer of the uterus. Laparotomy was done. After the evacuation of 2 L of hemoperitoneum, an approximately 5 × 4 metastatic, vesicular mass was seen in the posterior wall of the uterus, which was resected and uterine preservation was successful.

Conclusion: This case report describes the clinical, imaging, surgical and histopathological findings of Invasive mole after a hydatidiform molar pregnancy. Our case highlights the feasibility of fertility-preserving surgery in the case who experienced life-threatening hemorrhage due to a ruptured uterus.

1. Introduction

Gestational trophoblastic neoplasia comprises a unique group of human neoplastic diseases that derive from fetal trophoblastic tissues. This group is composed of choriocarcinoma, placental-site trophoblastic tumor, and epithelioid trophoblastic tumor, and many forms are derived from the precursor lesions, hydatidiform moles [1]. Approximately 50% of GTN are diagnosed after a hydatidiform molar pregnancy, with a diagnosis being made by the clinical and/or histopathologic criteria based on the following Federation of Gynecology and Obstetrics (FIGO) recommendations (1) an hCG plateau for at least four values over 3 weeks; (2) an hCG increase of 10% or greater for at least three values over 2 weeks; (3) hCG persistence 6 months after molar pregnancy evacuation, (4) histopathologic diagnosis of choriocarcinoma; or (5) presence of metastatic disease [2]. The hydatidiform mole is the most common form of GTD, representing 80% of cases. An invasive mole is a hydatidiform mole characterized by the enlarged hydropic villi invading into the myometrium, into vascular spaces, or into extrauterine sites.

The abnormal villi penetrate deeply into the myometrium. These lesions may be differentiated from choriocarcinoma in that they contain hydropic villi along with the marked trophoblastic proliferation. Approximately 15% of invasive moles remain localized to the uterus, and 5% are metastatic. Deep myometrial invasion may occur, and molar villi may be observed on the uterine serosa. Uterine rupture and severe intraperitoneal hemorrhage may occur if a deep myometrial invasion is left untreated [3]. Here is a case with invasive mole after the evacuation of complete molar pregnancy, presented with acute abdomen. We desired to preserve the uterine because our 21 years old patient doesn't have a child. Management and fertility-preserving surgery, in this case, was fortunately successful despite uterine rupture due to metastasis and immense hemoperitoneum. This case report has been reported in line with the improved SCARE checklist. The SCARE Guidelines were published in 2020.

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2. Case description

A 21-year-old Gravida 2 Abb 1 woman with no allergy history and drug was referred to the emergency ward of Obstetrics and Gynecology of a university-based hospital with a history of abdominal pain, nausea, and spotting. The patient was a known case of complete molar pregnancy that had been evacuated on 21/2/2021 (2 months earlier). The pre-evacuation hCG titer was 269,000 IU/L. She was followed by weekly hCG. During follow up the amount of hCG decrease to 700 but the last hCG was reported 106,907 IU/L on the day of admission (Table 1).

We admitted the patient to the emergency ward. On arrival, her initial vital signs were detected as Bp 100/70, PR: 120, RR: 16, T: 37, O2sat: 97%. Physical exam was remarkable for abdominal distension and diffuse tenderness with a rebound. On speculum examination, the vagina examined was normal. On detailed examination, slight vaginal bleeding was noted. A general physical examination with a focus on the likely site of metastases performed (i.e., lungs, liver, central nervous system [CNS]). Labs revealed anemia (Hb 8.6). An emergency abdominal ultrasound scan showed a 47 * 34 * 55 mm ill-defined hyperechoic heterogeneous mass with anechoic cystic vascular spaces within it, in the posterior wall of the uterus away from the endometrium that extended to the serous layer of the uterus. A large hematoma adjacent to the posterior wall of the uterus was noted (Fig. 1). Color Doppler revealed prominent blood flow signals within the mass and spectral Doppler showed high velocity and low impedance arterial blood flow and non-pulsatile venous flow. Heterogeneous endometrium suggestive of hemorrhage within the endometrial cavity was noted (Fig. 2). Both ovaries were unremarkable. A large amount of free intraperitoneal fluid with internal echo in favor of hemoperitoneum was seen. Based on the previous history of molar pregnancy, an ultrasound diagnosis of GTN (invasive mole) of the myometrium with uterine rupture was suggested (Fig. 2). The level of hCG (106,907 IU/L) strongly suggested GTN and due to acute abdomen, the uterine rupture was inferred. CXR, TFT, and LFT were normal. The patient immediately underwent exploratory laparotomy. Residents of gynecology did surgery under the direct supervision of their attending surgeon. Laparotomy was done under general anesthesia by a Pfannenstiel incision. Since she was very young and had no child and wished to preserve her fertility, we attempted for resection of the tumor with uterine reconstruction. After the evacuation of 2 L of hemoperitoneum, an approximately 5 × 4 metastatic, vesicular mass was seen in the posterior wall of the uterus (Fig. 3). We resected the metastatic mass by wedge resection from the posterior wall of the uterus (similar to the Jones technique), which was the origin of bleeding, thereafter; the uterus was reconstructed by suturing in a continuous locked manner (Fig. 4). Bilateral uterine artery ligation was done. A total of 4 units of packed cells and 4 units of FFP were transfused. Her recovery from the surgery was uneventful. Metastatic workup was done and was found to be negative. Pathologic examination revealed rare molar villi and atypical trophoblastic cells, consistent with invasive mole. The patient had stage 1 GTN and candidate for chemotherapy. She was discharged 2 days after surgery by advising her to check hCG weekly, assumption of DMPA for contraception, and be referred for chemotherapy. The patient chemotherapy with Methotrexate (MTX) started. HCG wasn't decreased properly so the regime been changed to Actinomycin. Fortunately HCG decreased to 94 on 29/5/2021 after two cycle chemotherapy with Actinomycin every two weeks. The patient responded to the treatment well.

3. Discussion

Invasive mole develops after a molar pregnancy and is characterized by the presence of edematous chorionic villi with trophoblastic proliferation invading the myometrium. Postmolar GTN develops in approximately 15 to 20% of patients following complete hydatidiform mole [3]. Uterine rupture in the setting of high-risk GTN is a rare and potentially catastrophic event [11]. This patient had a history of complete hydatidiform mole near 2 months ago and the high-risk feature for developing GTN was hCG levels >100,000. (Patient hCG before succession curettage was 265,000 IU/L.) GTN usually presents with AUB due to invasion of uterine tumor or bleeding from a metastatic site. Bleeding from uterine perforation may result in abdominal pain. For women with a prior molar pregnancy, serial measurement of hCG is part of post-treatment surveillance, and an elevation, plateau, or persistence of hCG suggests the development of GTN. The use of transvaginal ultrasound and color flow Doppler is particularly useful when the uterus is enlarged to identify patients at risk of perforation or who might be candidates for hysterectomy to reduce tumor burden [4]. In this case report, we described a young woman with a history of complete molar pregnancy. The patient was evaluated for spotting and mense retardation while the gestational age was 9 weeks. Transvaginal sonography revealed complete molar pregnancy which was confirmed by pathology assessment after succession curettage. The sequence of hCG titer after suction curettage is shown in Table 1. The downward trend of hCG was stopped for 3 weeks and unfortunately, the patient didn't return to the clinic because of the fear of Corona Virus, according to the fourth wave of covid-19 in Iran. She postponed attending the clinic because the hCG wasn't rose extremely during 2 weeks ago (Table 1).

On 14/4/2021she referred to the emergency ward by the acute abdomen. Sonography revealed a large hemoperitoneum and a heterogeneous hypervascular mass in the posterior wall of the uterus. She was just 21 years old and strongly maintained a desire to preserve her fertility; therefore a conservative management approach was employed.

Informed consent was taken and all the probabilities in the operation room (hysterectomy, massive transfusion) were discussed in detail with the patient and her husband.

Fertility sparing surgery in gestational trophoblastic neoplasia wasn't popular. The majority of them was done in elective condition for persistent GTN which didn't respond to chemotherapy [5]. In most instances, GTN is cured by surgical evacuation of the uterus, with the persistent disease being very sensitive to chemotherapy. Hysterectomy is recommended for persistent chemotherapy-resistant uterine disease [6]. Myometrial invasion in GTN may be followed by uterine perforation and intraperitoneal bleeding. Uterine rupture can be potentially lethal and often entails hysterectomy. In patients who desire to preserve fertility, hysterectomy is usually unacceptable. Uterine resection of localized disease with uterine reconstruction can be an alternative, but in certain cases, as shown in Table 2, primary repair, as conservative management, should be considered as an option [8].

Among 78 patients with GTN in the study by Xiaoyu Wang who achieved fertility-preserving surgery, only 5 patients underwent surgery due to suspected uterine rupture [7]. There have been several previous reports of cases of uterine perforation by an invasive mole, all of which were managed with abdominal hysterectomy [13]. To our knowledge, invasive mole perforation with active bleeding management by fertility-preserving surgery without hysterectomy is rare. She underwent an exploratory laparotomy where 2 L of hemoperitoneum was evacuated, and a 5 cm bulging mass was noted on the posterior aspect of the uterus

Table 1
BhCG titration.

Date	21/2/2021 (before succession curettage)	First week	Second week	Third week	Fourth week	Fifth week	sixth week	14/04/2021 (admission time)
hCG level	269,000 IU/L	10,672	897	700	- ^a	-	-	106,907 IU/L

^a Patients hadn't performed BhCG test for 3 weeks.

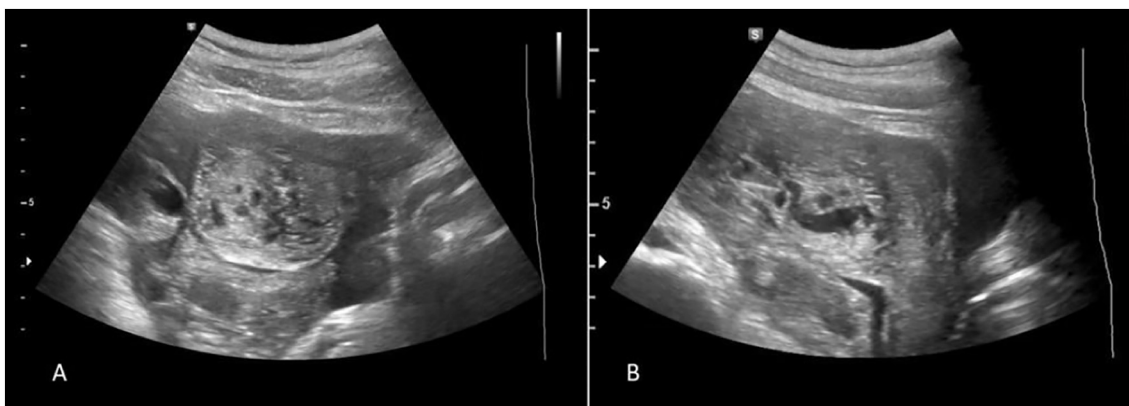


Fig. 1. A) Axial and B) sagittal images from transabdominal gray scale ultrasound show hyperechoic heterogeneous mass with anechoic cystic spaces within it, in the posterior wall of the uterus.

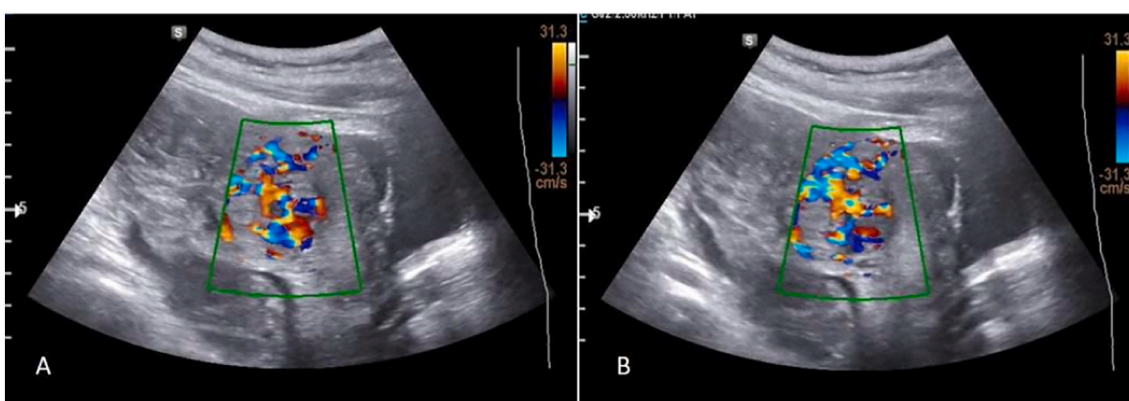


Fig. 2. A) and B) Transabdominal color Doppler image shows heterogeneous hypervascular mass in the posterior wall of the uterus.

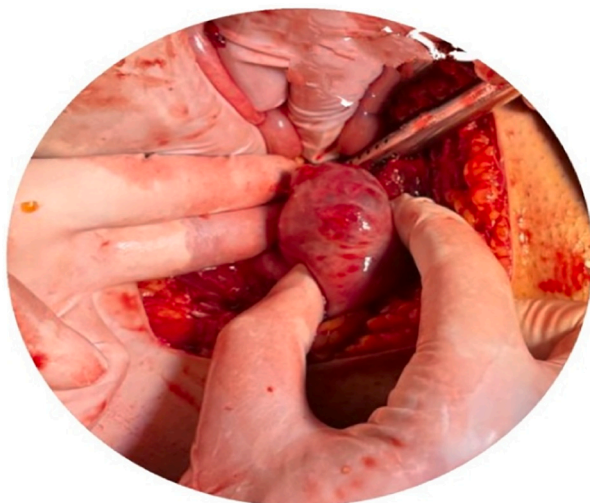


Fig. 3. Metastatic, vesicular mass was seen in the posterior wall of the uterus.

at the level of the serosa. Within the mass, a 1 cm serosal rupture was noted with emerging tumor and active bleeding. We resected the metastatic mass by wedge resection of the posterior wall of the uterus. Bilateral uterine artery ligation was done thereafter. Inspection of the abdomen and pelvis was performed, revealing no other intraabdominal or pelvic masses. The site of bleeding was inspected again and excellent hemostasis was noted. At the time of abdominal wall closure, a Jackson-



Fig. 4. The uterus was reconstructed by suturing in a continuous locked manner.

Pratt (JP), drain was placed in the pelvis to monitor any post-operative bleeding.

Recently David-West G described the patient similar to our case but bilateral uterine artery embolization using Gelfoam slurry to temporize bleeding done before surgery by the interventional radiology team [12].

Table 2

Review of GTN cases (acute abdomen & uterine rupture) with fertility sparing surgery.

Publication	Case	Mode of surgery	Chemotherapy after surgery	Etiology of surgery
Behtash et al. [9]	- 18yr-G1P0 - 19yr-G1P0	Uterus localized resection	No chemotherapy	Acute abdomen & shock
Estrella et al. [10]	- 23yr-G1P0	Mass resection & uterine repair	MTX change to Actinomycin-D (5 cycles)	Tumor rupture & hemoperitoneum in ultra sonography
Hasanzadeh et al. [8]	- 19yr-G1P0 - 27yr-G3AB2P0	Uterine defect primary repair Molar tissue resection	11 cycles of MTX 9 cycles of MTX (50 mg/m ² weekly)	Abdominal pain & shock under chemotherapy Shock (class 3)

For the present case, we ligated uterine arteries during surgery while evacuating 2 lit blood from the abdomen. Furthermore, David-West G coated the site of bleeding thrombin spray and packed it with Surgiflo and Gelfoam soaked in thrombin, but we repaired the uterine wall carefully like myomectomy by continuously locked sutures. Our case highlights the feasibility of fertility-preserving surgery in the case who experienced life-threatening hemorrhage due to a ruptured uterus, a very high-risk clinical scenario. This case report has been reported in line with the improved SCARE checklist. The SCARE Guidelines were published in 2020 [14].

Consent informed

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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CRedit authorship contribution statement

Alizadeh SH: Investigation, Writing - Original draft, Writing - Review & editing; critical revision

Mollabashi M: study concept or design, data collection, Investigation, Writing - Original draft

Zamani M: performed the surgery, Investigation.

Declaration of competing interest

The authors report no declarations of interest.

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