



## Case report

## Vaginal metastasis of gestational trophoblastic neoplasia resulting in hemorrhage: A case report

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## ABSTRACT

Complete molar pregnancies complicate approximately 1 in 1500 pregnancies in the United States and result in gestational trophoblastic neoplasia in about 15–20% of these cases. Vaginal metastasis is the second most common site of metastasis and may present with vaginal bleeding and hemorrhage. This report describes a case of a 19-year-old Hispanic primigravida who presented with hemorrhage from an anterior vaginal wall metastasis two weeks after dilation and curettage for complete molar pregnancy. Hemorrhage resolved after extrusion of the lesion from the anterior vaginal wall. Pathology showed markedly atypical trophoblastic tissue from the lesion. Vaginal involvement of gestational trophoblastic neoplasia can present with life-threatening hemorrhage.

## 1. Introduction

Hydatidiform mole falls within the spectrum of gestational trophoblastic neoplasia (GTN), cellular proliferations arising from placental villous trophoblast (Lurain, 2010). Hydatidiform moles are abnormal pregnancy events, occurring in 1 out of 1500 pregnancies in the United States. They are defined as complete or partial based on distinct chromosomal and cytologic appearance. Complete moles have a karyotype of 46, XX or 46, XY, whereas partial moles have a triploid karyotype, most commonly 69, XXY. Abnormal trophoblastic tissue of moles have dysfunctional regulatory mechanisms, which can result in aggressive, vascularized invasion (Seckl et al., 2010).

Complete moles can initially be managed with dilation and curettage (D&C), particularly in patients desiring fertility (Ngan et al., 2018). However, they do have the potential to invade the uterus and metastasize hematogenously, most commonly to the lungs, pelvis, and brain, developing into gestational trophoblastic neoplasia (Lurain, 2010). Persistent molar pregnancy, invasive mole or choriocarcinoma, is estimated to occur in approximately 15–20% of complete moles (Ngan et al., 2018). The presence of metastases contributes to the stratification of disease into low- and high-risk, thereby guiding further management (Ngan et al., 2003). While metastatic disease may be occult, presenting as a plateauing or rising beta-hCG level on serial evaluation, it may also present with hemorrhage due to the hypervascularity of the

trophoblastic metastatic tissue.

## 2. Case presentation

A 19 year old Hispanic primigravida presented to the Emergency Department with sudden-onset vaginal bleeding, hypotension, and syncope. The patient had recently undergone an uncomplicated suction D&C for a suspected molar pregnancy fifteen days prior, at which point her beta-hCG was >700,000 mIU/mL. Pathologic review of the specimen obtained showed hydropic villi and mild trophoblastic proliferation, confirming the diagnosis of complete molar pregnancy. Weekly beta-hCG was trended with decline to 12,718 mIU/mL.

Upon presentation to the ED, the patient was initially evaluated in the trauma bay where she was noted to be normotensive and non-tachycardic. Her beta-hCG returned 14,253 mIU/mL and her hemoglobin was 11.8 g/dl. A rapid bedside sonogram revealed a small uterus measuring 9.7 × 6.0 × 4.0 cm with a relatively thin endometrial stripe (0.87 cm). Multiple bilateral adnexal cysts, the two largest measuring 7.0 × 5.4 cm and 6.0 × 4.9 cm, were noted. During evaluation, the patient's vaginal bleeding continued and large clots were evacuated from the vagina. The patient soon became hypotensive despite an IV fluid bolus and her estimated blood loss in the ED was 500 cc. Transfusion of packed red blood cells was started and the patient was consented for emergent pelvic examination under anesthesia, D&C under

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ultrasound guidance, and possible exploratory laparotomy and hysterectomy.

Once in the operating room, bedside ultrasound again revealed a thin endometrial stripe. Her exam under anesthesia showed no bleeding coming from the cervix but new blood in the vaginal vault. Removal of the speculum and inspection of the vaginal walls revealed a 1.5 × 1.5 cm friable mass in the anterior vaginal wall, 3 cm proximal to the urethral orifice and directly midline (Fig. 1). The mass was actively bleeding and appeared to be markedly different from the surrounding vaginal epithelium. Prior to proceeding, the lesion's superficial location and size were considered, with the potential for increased bleeding with surgical intervention weighed against the possibility that bleeding could be controlled. Gentle pressure was applied adjacent to the mass, completely extruding it from the vaginal wall (Fig. 2). The bleeding immediately ceased. On gross inspection, the mass appeared to be placental-like. Palpation of the vaginal defect revealed only a thin layer of tissue intervening between the base of the mass and the urethra. Urethroscopy was performed which showed a normal bladder survey, trigone, bilateral ureteral orifices with jets, and urethra without defects or discoloration. No saline extravasation was noted into the anterior vaginal wall defect. A Foley catheter was placed and the defect was closed with 3–0 Vicryl suture in a layered closure.

The patient was discharged home on postoperative day one. She was evaluated at a follow-up appointment and was doing very well with no further vaginal bleeding. Pathologic review of the specimen showed marked atypical trophoblast. She was subsequently referred to Gynecologic Oncology and obtained CT of the chest, abdomen, and pelvis, as well as MRI of the brain. She was diagnosed with Stage III, low-risk GTN and started on medroxyprogesterone for birth control, single-agent chemotherapy with methotrexate, and serial beta-hCG monitoring (Fig. 3). Her prognosis after treatment is excellent, with almost all low-risk cases being cured (Ngan et al., 2018).

### 3. Discussion

While GTN is a curable disease, it may be acutely life-threatening when presenting as sudden-onset hemorrhage. In the literature, cases of bleeding vaginal metastases of gestational trophoblastic disease have been managed with plication of the lesion, vaginal packing with subsequent chemotherapy, or selective angiographic embolization to control bleeding from ruptured metastatic lesions (Yingna et al., 2002; Bloch and Atad, 1983). However, no cases to our knowledge describe

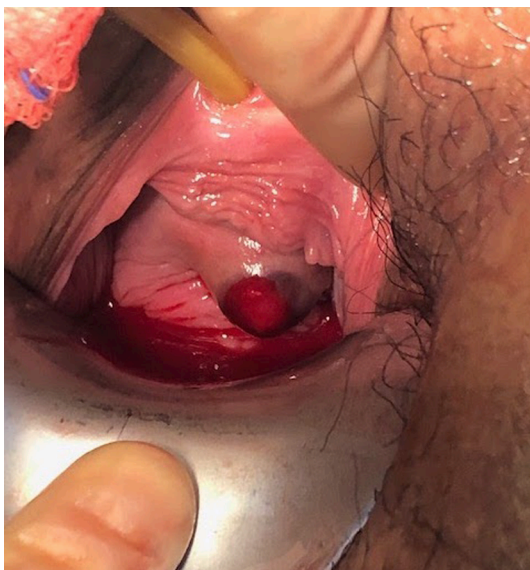


Fig. 1. Anterior vaginal wall mass as the source of the hemorrhage.

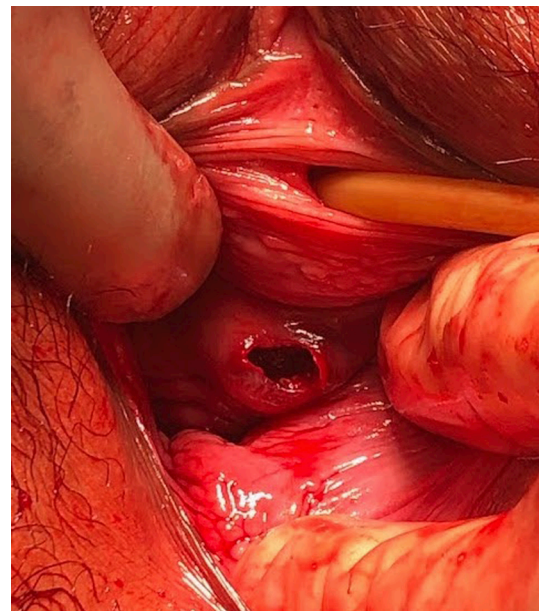


Fig. 2. Vaginal wall defect after extrusion of the anterior vaginal wall mass.

extrusion of a lesion from the wall with cessation of hemorrhage, without the need for further surgical intervention, as described in this report. In fact, experts recommend avoiding biopsy or excision of such lesions to limit catastrophic hemorrhage (Wong et al., 1990). Surgical intervention is only recommended to control bleeding, and can still be a hazardous endeavor in these scenarios.

We suspect that in this case, the superficial nature of the lesion resulted in its early rupture, and the relatively small size of the lesion lent itself to resolution of bleeding with removal of the mass. In addition, it is possible that the atypical trophoblast pathology contributed to the ability to resect the mass, compared to more malignant or aggressive pathologies. However, it is important to emphasize that even small lesions have the potential to hemorrhage, which may not resolve with excision (Method et al., 1996). The hypervascularity of molar tissue in the setting of the vaginal venous plexus increases the likelihood for bleeding. If removal of the mass resulted in continued hemorrhage, the vagina would have to be packed and angiographic embolization sought.

There are no guidelines or recommendations for predicting which lesions can be removed successfully, and therefore, it is reasonable to assume that these lesions will bleed significantly if disrupted. Maintaining a high clinical suspicion for ruptured vaginal metastasis in patients with vaginal bleeding after molar pregnancy can allow the practitioner to promptly manage bleeding and limit mortality in these patients. Despite the management and outcome described in this case, removal of such lesions should only be attempted with great caution and avoided altogether in the hemodynamically stable patient.

### 4. Patient consent

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.

### CRedit authorship contribution statement

**Emily Du:** Writing - original draft. **Melissa Perez:** Writing - original draft. **Tabetha Harken:** Conceptualization, Writing - review & editing, Supervision. **Jill H. Tseng:** Conceptualization, Writing - review & editing, Supervision.

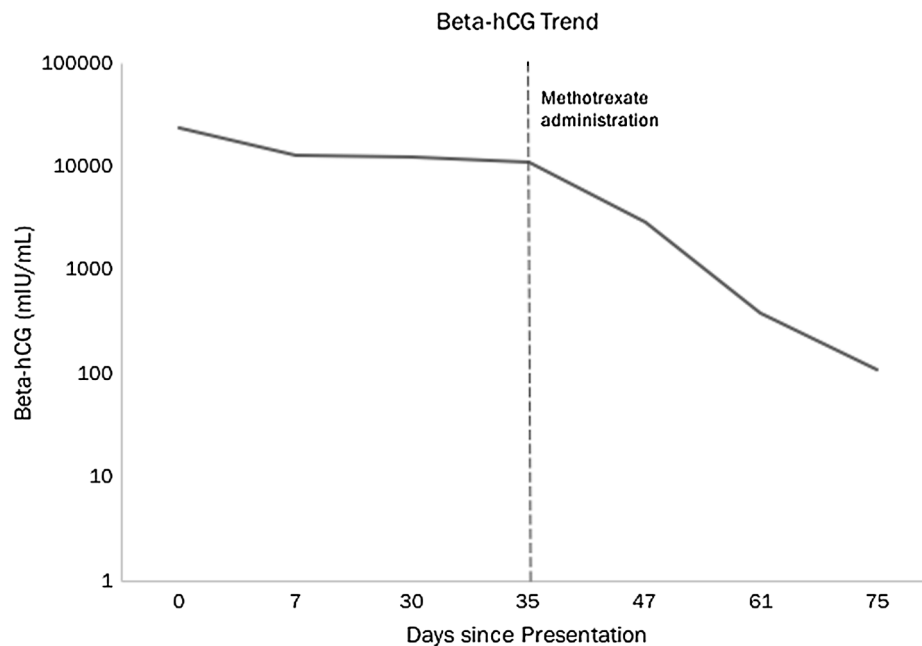


Fig. 3. Beta-hCG trend after removal of vaginal metastasis and treatment.

#### Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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