



Perforating Mole in a Nullipara Leading to Massive Hemoperitoneum: Fertility Preservation, a Challenge

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Abstract

Invasive mole, also known as chorioadenoma detrusens, is a neoplastic entity involving the trophoblastic tissues and penetrating deep into the myometrium. It may present as massive intraperitoneal hemorrhage, following uterine perforation, in which uterine conservation becomes difficult. We report a rare case of chorioadenoma detrusens, presenting as massive hemoperitoneum in a nulliparous woman, in which, a challenging decision of conserving the uterus was taken in low resource settings. Operative findings showed hemoperitoneum of 1000 cc, an enlarged uterus with a rent on fundus with torrential bleeding through it and grape-like vesicles popping from it. Two more thin uterine blebs representing thinned out myometrium, were seen on the anterior and right lateral surface of uterus on the verge of rupture. Hemostatic sutures were applied on the rent and bleeding was controlled.

Keywords: Invasive mole; Hemoperitoneum; Conservative management

Introduction

Gestational Trophoblastic Neoplasia (GTN) encompasses pathologic entities that are characterized by aggressive invasion of endometrium and myometrium by trophoblastic cells. Invasive mole, also known as chorioadenoma detrusens, is one such entity which involves trophoblastic tissues, penetrating deep into the myometrium, and at times, involving peritoneum, adjacent parametrium or vaginal vault. They are locally invasive but generally lack tendency to develop wide spread metastasis. Classic presentation of invasive mole is history of amenorrhea, bleeding per vaginum and pain abdomen. However, it may present as massive intraperitoneal hemorrhage, following uterine perforation. We report a rare case of chorioadenoma detrusens, presenting as massive hemoperitoneum in a nulliparous woman, in which, a challenging decision of conserving the uterus was taken in low resource settings.

Case Presentation

A 25-years-old nulliparous female was referred from a small public hospital to the casualty department of a tertiary care hospital, as a case of ruptured ectopic pregnancy. The patient had history of dilatation and curettage done four months back in view of missed abortion at eight weeks gestation, in a private hospital. No histopathology had been sent. Patient remained asymptomatic for one month and developed bleeding per vaginum thereafter. She assumed it as menstruation, though pattern of bleeding was different from normal. After five days of bleeding, she developed acute pain in abdomen for which she consulted the public hospital. There, her ultrasound showed normal size uterus with endometrial thickness of 12 mm. Bilateral ovaries were normal, however, increased vascularity in bilateral adnexa and mild amount of heterogenous collection in pouch of Douglas was noted. The β hCG levels came out to be 2,97,198 IU. With these reports, patient was referred to our hospital.

On admission, patient was conscious but had signs of hypovolumic shock (blood pressure of 80/60 mmHg, pulse rate of 130 bpm to 140 bpm). She had severe pallor. Abdominal examination revealed a distended and tense abdomen with tenderness in lower abdomen. Vaginal examination showed minimal bleeding, closed external OS and fullness in fornices. Uterine size and bilateral adnexa could not be assessed due to tense abdomen. Paracentesis showed non-clotting blood. Patient was taken for laparotomy immediately. Operative findings showed hemoperitoneum of 1000 cc, uterus enlarged to 10-12 weeks size with a rent of 2 cm \times 1 cm on fundus with torrential bleeding through it and grape-like vesicles popping from it. Two more thin uterine blebs (2 cm \times 1 and 4 cm

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Figure 1: Uterus enlarged to 10 to 12 week's size with a rent of 2 cm x 1 cm on fundus with torrential bleeding through it, held by Green Armytage Forceps.



Figure 2: Grape-like vesicles popping from the rent in uterus.

× 4 cm), bluish in color, representing thinned out myometrium, were seen on the anterior and right lateral surface of uterus on the verge of rupture (Figure 1-3). Bilateral fallopian tubes were normal but bilateral ovaries enlarged with theca-lutein cysts. A clinical diagnosis of invasive mole or choriocarcinoma was made. Keeping in mind, the nulliparous status of patient, uterine conservation seemed to be important. However, risk of rupture of the blebs was a prime concern. Moreover, there was a possibility of inability to achieve hemostasis. The patient's husband was explained about the gravity of situation and requirement of relaparotomy if she bleeds again. Hemostatic sutures were applied on the rent and bleeding was controlled. The protruding grape like tissues was sent for histopathologic examination. Intraperitoneal drain was inserted due to risk of subsequent rupture of the blebs leading to hemoperitoneum. As ultrasound revealed no mass in uterine cavity, no evacuation was attempted.

Patient remained stable in postoperative period with her stitches and drain removed on 8th postoperative day. Her histopathology showed hyperplastic trophoblasts, generalized cystic degeneration of chorionic villi and presence of molar villi within myometrium. Her β hCG had fallen to 56,781 IU/ml by 8th post operative day. No metastasis found in her chest X-ray and USG whole abdomen. Oncology consultation was done. After reviewing all reports and prognostic scoring (=7) patient was put on multiple agent chemotherapy (EMACO) on outpatient basis by oncology department; patient discharged with proper contraceptive and follow up advice. She received eight cycles of EMACO. Weekly β hCG follow up was done and it was found to be 1.5 IU/ml after 20 weeks follow up.

Discussion

Invasive moles comprise of 15% cases of gestational trophoblastic



Figure 3: Two more thin uterine blebs (2 cm x 1 cm and 4 cm x 4 cm), bluish in colour, representing thinned out myometrium, were seen on the anterior and right lateral surface of uterus on the verge of rupture (Shown by arrow).

neoplasia. They almost exclusively originate from partial or complete mole. Indicative signs of disease are persistent unexplained metrorrhagia or abnormal ultrasound appearance (large size uterus for gestational age and theca-lutein cyst >6 ml), non-normalization of serum β hCG at 6 months after evacuation of hydatiform mole or massive intraperitoneal hemorrhage [1]. These are definitely diagnosed on the basis of myometrial invasion on histology.

Management of invasive mole basically depends on two factors- prognostic scoring of GTN and parity of patient. Low risk GTN includes both non-metastatic and metastatic GTN with prognostic score <7. In patients with stage one disease, the selection of treatment primarily depends on whether patient desires to retain fertility. If patient does not want to preserve fertility hysterectomy with adjuvant single agent chemotherapy is performed as primary treatment. But in patients wanting to preserve fertility, management solely rests on chemotherapy.

Mitani et al. [2] proposed partial resection of uterus for young women if invasive mole presents with hemoperitoneum. Five women have been reportedly treated this way and four of them delivered healthy babies with cesarean section. Local uterine resection together with bilateral internal iliac artery ligation to combine hemostasis with preservation of fertility has been reported by Goldstein et al. [3]. Atala et al. [4] reported a case of chorioadenoma detrusens with uterine rupture where trophoblastic tissue invaded broad ligament and right ureter [4]. Hysterectomy followed by chemotherapy was done although it could not save the patient. In our case, the nulliparous status of patient along with point source of bleeding which could be easily controlled by hemostatic sutures deferred us from hysterectomy. Nonetheless there was further risk of uterine perforation through those thinned out blebs but we undertook that risk and left an intraperitoneal drain to watch for any further episode of hemoperitoneum. Uterine Artery Embolization (UAE) is an alternative and has been reported by Shen et al. [5] to successfully control internal hemorrhage caused by invasive trophoblastic disease. But UAE was not an option for us, as the facility is not available in our setting. If available, preoperative or immediate postoperative UAE could have been an option in our patient, as it would have alleviated the risk of further hemorrhage to an extent. In patients with stage one disease who desire to retain fertility chemotherapy is preferred option with excellent remission rates. Though a case has been reported by Sunesh et al. [6] where local uterine wedge resection following uterine evacuation without any chemotherapy cured the patient.

Conclusion

Invasive mole though not very common can present in a

nulliparous patient and hysterectomy should only be opted as last resort. Conservative approach should be considered.

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