Review of a case of choriocarcinoma with vaginal metastases

Hoo P.S., Nik Rafiza Afendi, Mas Irfan Jaya Mahamooth, Ahmad Amir Ismail, Rahimah Abdul Rahim, Ahmad Akram Omar

ABSTRACT
Introduction: Choriocarcinoma is a rare highly malignant tumor that arises from trophoblastic tissue. It metastasizes rapidly to vital organs including lungs and brain. It shows very good response to appropriately chosen chemotherapeutic regimen. Case Report: We illustrate a case of gestational choriocarcinoma with vaginal metastasis. This lady initially presented to us with heavy menstrual bleeding approximately nine months after giving birth to her first child. On vaginal examination revealed the presence of a anterior vaginal wall growth measuring about 2x3 cm which was biopsied. While waiting for the HPE results, patient developed another two episodes of hypovolemia due to profuse blood loss, warranting uterine artery embolization. Formal histopathology report (HPE) revealed the presence of gestational trophoblastic disease consistent with metastatic choriocarcinoma. Patient is currently undergoing chemotherapy with EMACO (etoposide, methotrexate, actinomycin D, cyclophosphamide and vincristine) regime and is responding well. Conclusion: Metastatic choriocarcinoma may present solely as a vaginal growth. It may pose initial diagnostic dilemmas and thus treatment delay.

Keywords: Choriocarcinoma, Uterine artery embolization, Vaginal metastases

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INTRODUCTION
Gestational trophoblastic disease (GTD) encompasses a diverse group of lesions that originated from fetal trophoblast whose behavior may be benign or malignant. Fortunately, it is highly curable even when the tumor has spread to distant organs. Gestational trophoblastic neoplasia (GTN) is subgroup of GTD. It includes choriocarcinoma, invasive mole and placental site trophoblastic tumor. The lack of histological confirmation in its malignant form often poses much difficulty and controversy in its diagnosis, investigation and management.
Choriocarcinoma is a malignant tumor with absence of chorionic villi, abnormal syncytiotrophoblast and cytotrophoblast, necrosis, and hemorrhage [1]. There is ethnic variation in the incidence of choriocarcinoma, with women from Asia having a higher incidence compared with non-Asian women, studies have quoted the incidence at 1 in 2000 [1, 2].

We report a case of choriocarcinoma presenting initially as a vaginal nodule with heavy menstrual bleeding. Primarily a disease of the uterine corpus, it may invade surrounding organs such as fallopian tube, cervix, ovary and vagina thus making diagnosis and subsequent management difficult as it may be misdiagnosed as a threatened miscarriage, ectopic pregnancy, benign or malignant lesions of the cervix or vagina [3].

CASE REPORT

A 27-year-old para 1 female presented to our hospital in late June 2015 with complaint of heavy menstrual bleeding, nine months after normal vaginal delivery. She had been bleeding vaginally for about two weeks prior to admission with usage of at least 10 pads per-day with blood clots. This was also associated with anemic symptoms like giddiness and palpitations. On admission she was pale, with Blood pressure 114/75 mmHg and pulse rate 112 bpm. She had no goitre or any palpable lymph nodes.

Cardiorespiratory examination was unremarkable. Her abdomen was soft, with a palpable 12 week size uterus. A speculum examination was performed with consent. On examination a smooth growth was seen at the anterior vaginal wall, 2 cm from the hymen ring, 5 cm from the cervix, measured 2x3 cm. It had smooth surface with regular margin, bluish discoloration, blood clots were seen through an opening on the nodule. The vaginal nodule was biopsied. Her cervix was normal, measured about 2x2 cm. Bimanual examination revealed an anteverted uterus with size of 12 weeks.

Transvaginal ultrasound was done showed an anteverted uterus with measurement of 12x6 cm, with the presence of which was thought as two submucosal fibroid measuring 3.2x3.4 cm and 4.3x4.4 cm respectively. Endometrium was regular homogenous 10 mm thick. Bilateral ovaries were normal, with no other adnexal masses seen. Urine pregnancy test done was negative at this stage. Hemoglobin on admission was 6.5 g/dl, hence patient was transfused with four pints packed cells. Provisional diagnosis of abnormal uterine bleeding secondary to submucosal fibroid was given while waiting for the full HPE report. She was subsequently discharged home with oral tranexamic acid and progesterone.

Unfortunately two weeks later, the patient presented again with heavy peri-vaginal bleeding, with anemic symptoms. As hemoglobin was 6.0 g/dl, she was transfused another 3 pints pack cell and discharge home after four days. Two days later, patient developed active, profuse per vaginal bleeding with total blood loss of 1200 cc. Resuscitation done with transfusion of 4 pints packed cells and 2 cycles of DIVC regime. Vaginal packing was done to stop the bleeding, and patient was subsequently undergone uterine artery embolization which performed by interventional radiologist. The procedure was successful.

On that admission a urine pregnancy test was done, and it was positive. Urgent serum beta hCG was 664,456 IU/L. Biopsy of the vaginal nodule revealed the presence of gestational trophoblastic neoplasm, consistent with metastatic choriocarcinoma. Computed tomography scan of thorax, abdomen and pelvis showed the presence of choriocarcinoma of the uterus with local infiltration into the vagina and lung metastases.

The patient subsequently underwent nine cycles of chemotherapy (EMACO) and responded well. Per vaginal bleeding was stopped, and vaginal wall nodule was disappeared. Her latest β-hCG level was 0.8 IU/L. She is currently under follow-up of our GTD clinic.

DISCUSSION

The frequency of vaginal metastases in choriocarcinoma varies between 16% and 36% [4, 5]. Gestational trophoblastic neoplasia usually disseminates widely by the hematogenous route and the resultant metastatic foci are often nourished by an extensive network of fragile and tortuous vessels. These trophoblastic implants are often friable and vascular and frequently induce severe hemorrhagic complications [5].

Often situated suburethrally or in the fornices, vaginal metastases usually appear stuffed with blood, and have a blue, purplish or red hue. The sites of the vaginal metastases indicate an almost certain hematogenous spread of the disease, with lymphatic involvement being extremely rare [6].

The friable and hypervascular nature of a metastatic lesion places the patient at risk for significant hemorrhage. Traditionally, surgery, such as abdominal hysterectomy or uterine artery ligation, has been the treatment of choice for controlling intractable hemorrhage but recently other less invasive methods have been used like in this case, with use of uterine artery embolization to control acute blood loss [6].

Gauze packing might able to control the bleeding of most ruptured nodules, but it has some drawbacks. For example, prolonged tamponade could cause vaginal infection. Changing the gauze frequently will lead to vaginal mucosa abrasion, affecting its healing [7]. In addition, wrong packing method could be more dangerous because it may cause more damage and bleeding.

Recently, angiographic embolization has showed to be a safe and highly effective alternative procedure for massive genital bleeding [8]. This technique offers several advantages, including the avoidance of major surgery and general anesthesia and the preservation of fertility.
As in our case, her uterus was preserved by performing the angiographic embolization. There are several studies who report successful pregnancies after uterine artery embolization [7, 8].

**CONCLUSION**

This case illustrates the importance of having a high index of suspicion for gestational trophoblastic neoplasia, especially in patients presenting with recurrent per-vaginal bleeding which does not respond to medication after a recent normal pregnancy. An earlier diagnosis obtained would enable clinicians to treat the condition thus reducing morbidity and mortality. The usage of uterine artery embolization also illustrates the importance of alternative treatment modalities. Angiographic embolization may decrease the time required for gauze packing, reduce the amount of blood loss, and prevent multiple transfusions and infections.

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**Author Contributions**

Hoo P.S. – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Nik Rafiza Afendi – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Mas Irfan Jaya Mahamooth – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Ahmad Amir Ismail – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Rahimah Abdul Rahim – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Ahmad Akram Omar – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

**Guarantor**
The corresponding author is the guarantor of submission.

**Conflict of Interest**

Authors declare no conflict of interest.

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