

# A case report: Co-existence of uterine arteriovenous malformation with persistent trophoblastic disease – The diagnostic and management dilemmas

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### SUMMARY

Uterine arteriovenous malformation (UAVM) is a rare vascular anomaly characterised by abnormal connections between the uterine arteries and veins. It can be either congenital or acquired, often presenting with abnormal uterine bleeding. Persistent Trophoblastic Disease (PTD) is a form of gestational trophoblastic tumor, which is an uncommon pregnancy-related condition that can arise following a molar pregnancy or a normal pregnancy, due to retained trophoblastic tissue. Both conditions are rare and can potentially cause life-threatening hemorrhage independently. It is rare for both to coexist, and when they do, the diagnosis and management can be challenging, as exemplified in our patient.

### INTRODUCTION

Uterine Arteriovenous Malformation (UAVM) is a rare condition that can occur either congenitally or be acquired. It is described as abnormal connections that occur between the uterine arteries and veins with no intervening capillary.

Gestational Trophoblastic Disease (GTD) is a group of tumors that arise with either normal or abnormal pregnancies. It can be malignant or benign. Persistent Trophoblastic Disease (PTD) is when there is a persistent elevation of beta-human chorionic gonadotrophin ( $\beta$ -hCG) hormone after GTD, and it forms part of the condition referred to as Gestational Trophoblastic Neoplasia (GTN).<sup>1</sup> This rare condition arises after a molar pregnancy or after the loss of a pregnancy.

However, co-existence of UAVM with PTD is very rare but can be potentially life-threatening due to uncontrollable uterine bleeding that can harm the patient. This case report highlights a 35-year-old woman who was managed at Hospital Tuanku Ja'afar (HTJ) with both these conditions co-existing. The diagnosis and management of the woman were challenging but gratifying once the exact diagnosis was made. She had a spectrum of modalities for diagnosis and management that resulted in a good management outcome for her.

### CASE PRESENTATION

A 35-year-old woman, Para 0 + 1, was seen in the Obstetrics and Gynecology Clinic of HTJ for persistent vaginal bleeding

for about 1 month in May 2024. She is not known to have any medical problems.

She had a history of Evacuation of Retained Product of Conception (ERPOC) for incomplete miscarriage in October 2019. She had retained products of conception (POC) complicated with endometritis at that time. It was a 13-week spontaneous pregnancy loss with excessive vaginal bleeding. A transvaginal ultrasound had revealed POC within the endometrial cavity measuring 6 cm by 2.5 cm. She was pyretic at that time and was given intravenous broad-spectrum antibiotics. During the ERPOC procedure, she had bled excessively, and she was hypotensive intraoperatively. She had 4 units of packed red blood cells (RBCs) transfused during this admission and was discharged home well on the 4th day after the procedure when she was hemodynamically stable.

She did not conceive after that until May 2024, when she noted that her urine pregnancy test was positive. However, she was having lower abdominal pricking pain with irregular vaginal bleeding at that time. The General Practitioner who saw her did a transvaginal scan that revealed an intrauterine gestational sac that was irregular with no embryo seen. She was referred to HTJ for a missed miscarriage for further management.

The transvaginal scan showed an irregular intrauterine gestational sac with a  $\beta$ -hCG level of 398,322 mIU/ml. A diagnosis of partial mole was made. She had ERPOC with excessive blood loss. No vesicles to suggest molar pregnancy were noted at the procedure, and a repeat ultrasound post-evacuation showed an endometrial cavity that was thin and linear. The post-procedure hemoglobin was low, and she required 2 units of packed cell transfusions.

Post-procedure  $\beta$ -hCG had dropped to 39,549 mIU/ml. She was discharged home on the 3rd day after ERPOC with stable vitals and no vaginal bleeding. The transabdominal ultrasound before discharge showed mixed echogenicity within the uterine cavity measuring 1.3 cm in the mid-sagittal view.

During her review 3 weeks later in the gynecology outpatient clinic, she started to have excessive vaginal bleeding. Examination showed that she was bleeding and soaking

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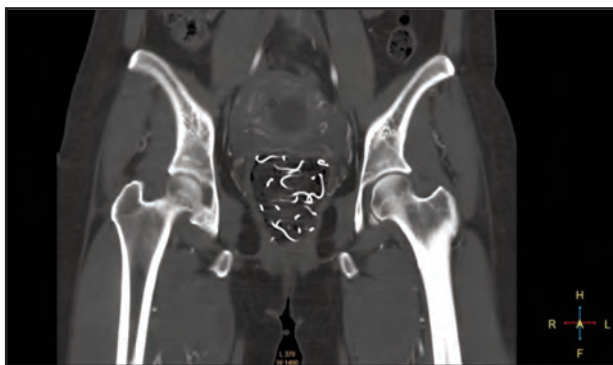


Fig. 1: CTA of the pelvis



Fig. 2: CTA of the pelvis

through her clothes and wetting the drapes. The speculum examination showed that there were about 50-100 ml of blood clots in the vaginal cavity. Transabdominal ultrasound revealed a retroverted uterus with thin endometrium. However, a homogenous mass measuring 3 cm by 1.8 cm with large tortuous vessels was noted over the anterior lower endometrial cavity extending into the myometrium. The Doppler uptake of the mass was high, with a Peak Systolic Velocity of 65 cm/s. The  $\beta$ -hCG, which had been done at the outpatient clinic two days prior, was 162,234 mIU/ml. A provisional diagnosis of Invasive Molar Pregnancy with possible UAVM was made.

She had an evacuation of the uterine cavity through a suction and curettage procedure that day under ultrasound guidance with Karmen's catheter size 6, to control her excessive vaginal bleeding. Her pre-operative hemoglobin was 8.8 gm/dL. There was minimal curetting with excessive bleeding post procedure requiring uterine massage, bimanual uterine compression, and Foley's catheter insertion with 60 ml of Normal Saline to control her uterine bleeding. She was also given intramuscular Syntometrine, intravenous Oxytocin Infusion, and intravenous Tranexamic Acid to control her bleeding. She lost about 1000ml of blood intraoperatively and required 1 unit of RBC transfusion post-procedure. The  $\beta$ -hCG post procedure was 5390 mIU/ml.

She was stable post-operatively and had an urgent Computed Tomography Angiography (CTA) of the pelvis at HTJ. The imaging revealed a rounded out-punching vascular structure at the lower anterior uterine body measuring 0.6 cm by 0.6 cm. (Figure 1 and Figure 2). This structure was most likely a UAVM. The patient was referred to Hospital Kuala Lumpur (HKL) radiology department with an available interventional radiologist for uterine artery embolization, as none were available in HTJ.

Bilateral uterine artery embolisation was done successfully at HKL. During the procedure, bilateral hypervascularity of the submucosal region of the lower uterine wall was noted. Patient recovered well post-procedure with no vaginal bleeding and was discharged home after 5 days following the procedure.

Her follow-up visits revealed that her  $\beta$ -hCG was trending downwards to 2152 mIU/ml and then to 788.5 mIU/ml. The histopathology of the endometrial curetting confirmed a

partial molar picture. However, the patient started having lower abdominal cramps with irregular vaginal bleeding, and the transvaginal ultrasound revealed a vascular lower uterine mass measuring 4.2 cm by 2.4 cm by 4.1 cm. Chest radiography was reported as unremarkable.

The International Federation of Gynecology and Obstetrics (FIGO) 2000 scoring system, was used to risk-stratify the patient. The score was 2, being positive for a history of pregnancy/miscarriage before the event, and tumor size between 3 to 5 cm.

She was started on intramuscular Methotrexate with folinic acid. She was planned for 6 chemotherapy cycles, and her  $\beta$ -hCG dropped from 214 mIU/ml to 114 mIU/ml to < 5 mIU/ml after the third cycle. The patient is still under gynecology outpatient follow-up. She is on a barrier method of contraception with no abnormal vaginal bleeding or positive transvaginal ultrasound findings. She plans for a pregnancy in 2026.

## DISCUSSION

Persistent molar tissues characterise PTD after a normal pregnancy, or molar pregnancy, or a miscarriage. It is associated with abnormal vaginal bleeding with abdominal pain, or cramps. The  $\beta$ -hCG does not return to normal levels and remains elevated. This was the scenario with the patient above, as her  $\beta$ -hCG levels were elevated even after evacuation of the uterus for partial mole.

UAVM is a rare condition, but it can be life-threatening because of uncontrollable vaginal bleeding. PTD is a more common condition found among Southeast Asians. However, for both PTD and UAVM to co-exist is even rarer. Very few case reports are available in the literature. Nandeesh and her group reported a similar case in 2022.<sup>2</sup> Their patient had uterine artery embolization after chemotherapy.

The diagnosis is complicated and challenging when there is co-existing UAVM with PTD because both share similar ultrasonic features. However, in UAVM, the PSV will be high, as seen in this patient. UAVM is a rare condition and may be congenital or acquired after uterine surgery, especially following an evacuation of the endometrial cavity. The UAVM in this patient most likely developed following her uterine evacuation in 2019. It is a potentially life-threatening

condition as patients can have profuse bleeding either spontaneously or following any uterine evacuation procedures. UAVM is best diagnosed with Color Doppler Ultrasound, where the PSV will be elevated more than 40 cm/s, as was seen in this patient. A confirmatory diagnosis can be made using CT angiography.<sup>3</sup> The diagnosis was confusing in this patient because the elevated  $\beta$ -hCG implied PTD. The patient was bleeding profusely, and to control it, evacuation of the uterus with minimal curetting was done. The intrauterine tamponade effect of the Foley's catheter controlled the bleeding.

Patients who wish to conserve the uterus can have uterine artery embolisation by an interventional radiologist. UAVM can result in heavy menstrual flow or life-threatening menstrual bleeding if untreated. Our patient had uterine artery embolisation for her UAVM. However, failures of uterine artery embolisation have been reported.<sup>4</sup>

Our patient's diagnosis was challenging as both conditions were co-existing. We proceeded with uterine artery embolization and then with chemotherapy as the  $\beta$ -hCG was persistently elevated.

#### CONCLUSION

Co-existing PTD with UAVM can be challenging to manage and diagnose. We need to have a high index of suspicion and excellent clinical acumen to diagnose the condition.<sup>5</sup> Early recognition and timely intervention were crucial in the management of this patient.

UAVM can be life-threatening due to uncontrollable bleeding.

Uterine artery embolisation is performed and recommended as a fertility-preserving measure for UAVM when expertise is available. Successful pregnancies with no increased risk for miscarriages, fetal growth restrictions or abnormal invasion of the placenta have been reported.<sup>6</sup> Single-agent chemotherapy is the accepted mode of management for low-risk PTD. A multidisciplinary approach is essential in achieving a good clinical outcome, as was seen in our patient.

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#### DECLARATION

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