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Uterine Arteriovenous Malformations After Remission of Gestational Trophoblastic Disease: A Seven-Year Case Series from a Resource-Limited Tertiary Center

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A B S T R A C T

Background:
Uterine arteriovenous malformations (UAVMs) are rare but potentially life-threatening vascular abnormalities that may occur after gestational trophoblastic disease (GTD). Their clinical presentation may mimic other causes of abnormal uterine bleeding, creating diagnostic and therapeutic challenges, particularly in resource-limited settings.

Objective: To describe the clinical presentation, diagnostic evaluation, and management of uterine arteriovenous malformations occurring after remission of gestational trophoblastic disease and to discuss management strategies in a tertiary referral center.

Methods:
we conducted a retrospective descriptive case series including patients diagnosed with UAVMs after documented remission of GTD between January 2015 and December 2022 at a tertiary referral center in Algeria. Data collected included patient characteristics, type of initial GTD, clinical presentation, imaging findings (Doppler ultrasound and pelvic angio-MRI), treatment modalities, and clinical outcomes. Therapeutic success was defined as complete cessation of uterine bleeding without additional intervention during 12 months of follow-up.

Results:
Three patients aged 33–42 years were diagnosed with UAVM after GTD remission. Initial GTD consisted of one partial hydatidiform mole, one complete hydatidiform mole, and one low-risk gestational trophoblastic neoplasia (invasive mole). Severe metrorrhagia was the presenting symptom in all cases. Management included selective uterine artery embolization (n=1), emergency hysterectomy (n=1), and bilateral hypogastric artery ligation (n=1). Complete resolution of bleeding was achieved in all patients, with no recurrence during the 12-month follow-up.

Conclusion:
UAVMs after GTD remission are rare but potentially severe complications. Diagnosis relies on β-hCG measurement and imaging techniques, including Doppler ultrasound and angio-MRI. Management should be individualized according to hemodynamic status, fertility desire, and available resources. Conservative approaches such as embolization or arterial ligation can be effective, while hysterectomy remains life-saving in emergency.

1. Introduction

Uterine arteriovenous malformations (UAVMs) are rare vascular anomalies characterized by abnormal direct communications between uterine arteries and veins, bypassing the normal capillary network (1). Although congenital forms have been described, most reported cases are acquired and occur following uterine trauma, pelvic surgery, miscarriage, childbirth, or gestational trophoblastic disease (GTD) (2,3).

The true incidence of acquired UAVMs is difficult to determine because many cases remain asymptomatic and are detected incidentally during imaging. Nevertheless, several studies suggest that these lesions represent a rare complication following GTD (4,5).

Clinically, UAVMs may present as recurrent metrorrhagia or, in severe cases, life-threatening uterine hemorrhage (6). In women with a history of GTD, this presentation may mimic recurrent trophoblastic disease, leading to diagnostic confusion and inappropriate therapy if not recognized. Therefore, a careful diagnostic approach combining serum β-hCG monitoring and imaging is essential.

Doppler ultrasonography is considered first-line due to its ability to identify abnormal hypervascularization with high-velocity, low-resistance turbulent flow. MRI, particularly angio-MRI, provides complementary anatomical information and helps confirm the diagnosis while delineating the extent of the malformation (7,8).

Although several case reports and small series have described UAVMs following GTD, data remain limited, especially in low- and middle-income countries with restricted access to interventional radiology. In such settings, treatment decisions are influenced by available technical resources.

This study aims to describe the clinical presentation, diagnostic approach, and therapeutic management of UAVMs occurring after GTD remission over a seven-year period in a tertiary referral center.

2. Methods

2.1. Study Design and Setting:

Retrospective descriptive case series (January 2015–December 2022) at the reference center for GTD in the Mitidja Plain, affiliated with University Hospital of Blida, Algeria. The center manages GTD cases from a large regional population within a resource-limited healthcare system, where interventional radiology is not always available. Ethics committee approval was obtained.

2.2. Study Population:

All consecutive patients diagnosed with UAVMs after documented remission of GTD were included.

Inclusion criteria:

- Diagnosis of UAVM following GTD
- Documented normalization of serum β-hCG prior to UAVM diagnosis

Exclusion criteria:

- Persistent trophoblastic disease (non-normalized or rising β-hCG)
- Incomplete medical records

2.3. Data Collection:

Medical records were reviewed using standardized forms. Variables included age, parity, obstetric history, type of initial GTD, interval from GTD remission to symptoms, clinical presentation, imaging findings, treatment modality, and outcomes.

2.4. Diagnostic Evaluation:

All patients underwent transvaginal or pelvic Doppler ultrasound. UAVM was suspected with abnormal hypervascularization and high-velocity, low-resistance arterial flow. Pelvic angio-MRI confirmed the diagnosis and mapped vascular anatomy.

2.5. Therapeutic Management:

Decisions were individualized based on hemodynamic status, bleeding severity, patient age, fertility desire, and availability of interventional radiology. Options included selective uterine artery embolization, uterine vascular pedicle ligation (hypogastric artery ligation), and emergency hysterectomy.

2.6. Follow-up:

All patients were followed for 12 months. Therapeutic success was defined as complete cessation of bleeding, no recurrence, and no need for additional intervention.

3. Results

Three patients with uterine arteriovenous malformations (UAVMs) occurring after remission of gestational trophoblastic disease (GTD) were identified during the study period. The patients' ages ranged from 33 to 42 years, with parity between 1 and 4.

The initial GTD diagnoses included one partial hydatidiform mole, one complete hydatidiform mole, and one low-risk gestational trophoblastic neoplasia (invasive mole). The interval between the initial GTD

event and the onset of symptoms ranged from 8 to 12 months.

Severe metrorrhagia was the presenting symptom in all cases and required urgent medical management. In the first case, a 33-year-old woman with a history of partial mole presented with heavy uterine bleeding eight months after uterine evacuation. Selective uterine artery embolization was performed, resulting in immediate and sustained hemostasis.

The second patient, a 42-year-old multiparous woman with a previous complete mole, presented with severe uterine bleeding twelve months after completion of surveillance. Due to the unavailability of interventional radiology at the time of presentation, an emergency hysterectomy was performed, which successfully controlled the hemorrhage.

The third patient, a 37-year-old woman previously treated for an invasive mole, developed severe metrorrhagia twelve months after treatment. Bilateral hypogastric artery ligation was performed, allowing effective control of bleeding while preserving the uterus.

Overall, all three therapeutic approaches resulted in complete cessation of bleeding. No recurrence of hemorrhage or UAVM-related complications was observed during a follow-up period of 12 months.

4. Discussion

Uterine arteriovenous malformations (UAVMs) are a rare but potentially life-threatening cause of abnormal uterine bleeding. While congenital forms exist, most cases are acquired and occur after uterine trauma, surgical procedures, miscarriage, childbirth, or GTD (1,2). The small number of cases in our series reflects this rarity and is consistent with the literature (3,4).

4.1. Pathophysiology:

In the context of GTD, abnormal angiogenesis and vascular remodeling induced by trophoblastic invasion may lead to arteriovenous shunts within the myometrium (3). Repeated uterine curettage during molar evacuation or post-molar surveillance can predispose to vascular injury and abnormal shunts (4,5). Persistent angiogenic signaling, such as elevated vascular endothelial growth factor activity after trophoblastic regression, may also play a role (16,17).

4.2. Clinical Presentation and Diagnosis:

Patients typically present with intermittent or severe metrorrhagia, sometimes leading to hemodynamic instability (6). In post-GTD patients, this can mimic recurrent gestational trophoblastic neoplasia. Serum β-hCG measurement is critical for differentiating UAVMs from persistent disease (6,16). In our series, all patients had normalized β-hCG.

4.3. Imaging:

Color Doppler ultrasound is first-line, showing high-velocity, low-resistance turbulent flow. MRI and angio-MRI provide detailed mapping of vascular anatomy and lesion extent, revealing serpiginous channels and early venous filling (7–9,17,18). Advanced ultrasound techniques, such as three-dimensional power Doppler, can further improve visualization (23). Differential diagnoses include retained products of conception, subinvolution of the placental bed, and GTN (21,22).

4.4. Management:

Management depends on bleeding severity, hemodynamic status, reproductive desire, and technical resources. Selective uterine artery embolization is the preferred fertility-preserving option, with success rates of 85–90% (10,11,19,24). In settings lacking interventional radiology, surgical alternatives, such as uterine artery or bilateral hypogastric artery ligation, effectively control hemorrhage while preserving the uterus (12,18,20). Emergency hysterectomy remains life-saving in uncontrolled hemorrhage or hemodynamic instability (13,16).

4.5. Reproductive Outcomes:

Conservative management generally yields favorable reproductive outcomes. Successful pregnancies post-embolization or vascular ligation have been reported, with low recurrence rates of 5–10%, emphasizing the importance of follow-up (14,19,25).

4.6. Clinical Implications

Our findings highlight the importance of considering uterine AVM in women presenting with post-GTD bleeding despite normalized β-hCG levels. Misdiagnosis may lead to unnecessary uterine curettage, which can aggravate hemorrhage and worsen the vascular lesion. Therefore, Doppler ultrasound should be systematically performed before any invasive uterine procedure in patients with suspected vascular abnormalities.

4.7. Future Perspectives

Further prospective multicenter studies with larger patient populations are needed to better define optimal management strategies and long-term reproductive outcomes. In addition, improved access to interventional radiology in resource-limited settings

may significantly enhance conservative management options for these patients.

4.8. Study Limitations

Several limitations of this study should be acknowledged. First, the study design was retrospective and descriptive, which may introduce potential biases related to data collection and limit the ability to establish causal relationships. Second, the small sample size (three cases) reflects the rarity of uterine arteriovenous malformations after gestational trophoblastic disease but restricts the generalizability of the findings and prevents meaningful statistical analysis. Third, the study was conducted in a single tertiary referral center, which may not fully represent the diversity of clinical presentations and management strategies in other healthcare settings. In addition, the availability of treatment modalities was influenced by local resources, particularly the limited access to interventional radiology, which affected therapeutic decision-making in one case and led to emergency hysterectomy. Finally, although the 12-month follow-up demonstrated no recurrence, longer follow-up would be valuable to better assess long-term outcomes, particularly regarding fertility preservation and the risk of delayed complications.

Despite these limitations, this case series provides clinically relevant insights into the diagnosis and management of UAVMs occurring after GTD remission, especially in resource-limited settings, and highlights the importance of individualized therapeutic strategies based on patient condition, fertility desires, and available medical resources.

5. Conclusion

Uterine arteriovenous malformations after GTD remission are rare but potentially severe, with significant hemorrhagic risk. Accurate diagnosis relies on serum β-hCG monitoring and imaging, particularly Doppler ultrasound and angio-MRI.

Management should be individualized based on hemodynamic status, reproductive desire, and available resources. Selective uterine artery embolization is the preferred fertility-preserving treatment when available, while hypogastric artery ligation is an effective alternative in resource-limited settings. Emergency hysterectomy remains life-saving in critical cases.

Early recognition by clinicians managing GTD is essential for timely intervention and improved patient outcomes.

6. Declarations

Ethics Approval and Consent to Participate

This study was conducted in accordance with the ethical principles of the Declaration of Helsinki. Given the retrospective nature of the study and the anonymization of patient data, formal ethical approval was waived by the institutional review board of the participating center. Written informed consent for the use of clinical data was obtained from all patients when possible.

Consent for Publication

Written informed consent was obtained from the patients for publication of their clinical data in an anonymized form.

Availability of Data and Materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Competing Interests

The authors declare that they have no competing interests.

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Authors' Contributions

All authors contributed substantially to the conception and design of the study. Data collection and clinical management were performed by the clinical team. Data analysis and manuscript drafting were carried out by the authors. All authors critically revised the manuscript and approved the final version for publication.

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