Uterine Arteriovenous Malformation Following Gestational Trophoblastic Neoplasia After Complete Hydatidiform Mole: A Case Report.

Abstract:

Background: Uterine arteriovenous malformation (UAVM) is a rare vascular abnormality that may occur following uterine instrumentation or in association with gestational trophoblastic disease (GTD). Its diagnosis is particularly challenging during post-molar follow-up, as imaging findings may mimic persistent trophoblastic neoplasia.

Case Presentation: We report the case of a 42-year-old woman diagnosed with a complete hydatidiform mole with an initial serum β-hCG level of 650,000 IU/L. Following suction evacuation, the patient developed gestational trophoblastic neoplasia and was treated with methotrexate, achieving complete normalization of β-hCG levels at the end of treatment. Four months after uterine evacuation, she presented with metrorrhagia. Pelvic Doppler ultrasonography revealed a uterine arteriovenous malformation. Given the absence of biological evidence of persistent trophoblastic disease and the patient's hemodynamic stability, a conservative approach with close clinical and ultrasonographic surveillance was adopted. The patient experienced progressive resolution of vaginal bleeding without the need for invasive intervention.

Conclusion: Uterine arteriovenous malformation should be considered in patients presenting with abnormal uterine bleeding during follow-up of gestational trophoblastic disease, even after complete β-hCG normalization. Doppler ultrasonography plays a key role in diagnosis, while conservative management may be a safe and effective option in selected clinically stable patients.

Keywords: Gestational trophoblastic neoplasia; Complete hydatidiform mole; Uterine arteriovenous malformation; Doppler ultrasonography; Methotrexate; Conservative management.

INTRODUCTION

Gestational trophoblastic disease (GTD) comprises a spectrum of disorders arising from abnormal proliferation of placental trophoblastic tissue, ranging from hydatidiform mole to gestational trophoblastic neoplasia (GTN). Owing to the routine monitoring of serum β-human chorionic gonadotropin (β-hCG) levels and the high sensitivity of GTN to chemotherapy, cure rates currently exceed 90% in most patients [1].

Uterine arteriovenous malformation (UAVM) is a rare vascular anomaly characterized by abnormal direct communications between uterine arteries and veins without an intervening

capillary network. UAVMs may be congenital or acquired, the latter most commonly occurring after uterine trauma such as dilation and curettage, suction evacuation, cesarean section, or other invasive uterine procedures [2]. Clinically, they typically present with abnormal uterine bleeding of variable severity and may occasionally lead to life-threatening hemorrhage.

The coexistence of GTD and UAVM is uncommon but represents a significant diagnostic challenge. Hypervascular uterine lesions detected on Doppler ultrasonography may mimic persistent or recurrent GTN, making differentiation between these entities difficult [3]. Accurate diagnosis is essential because management strategies differ substantially; while persistent GTN generally requires chemotherapy, inappropriate uterine instrumentation in patients with UAVM may precipitate severe hemorrhagic complications [4].

We report the case of a 42-year-old woman with gestational trophoblastic neoplasia following a complete hydatidiform mole, successfully treated with methotrexate and achieving β-hCG normalization. Four months after suction evacuation, she developed abnormal uterine bleeding, and Doppler ultrasonography revealed a uterine arteriovenous malformation. This case highlights the importance of considering UAVM in the differential diagnosis of post-molar bleeding despite complete biochemical remission and emphasizes the role of imaging in guiding appropriate management.

CASE PRESENTATION

A 42-year-old multiparous woman was referred to our department for the management of a complete hydatidiform mole. At presentation, her serum β-human chorionic gonadotropin (β-hCG) level was 650,000 IU/L. Pelvic ultrasonography revealed findings consistent with a molar pregnancy, and the patient subsequently underwent suction evacuation. Histopathological examination confirmed the diagnosis of a complete hydatidiform mole.

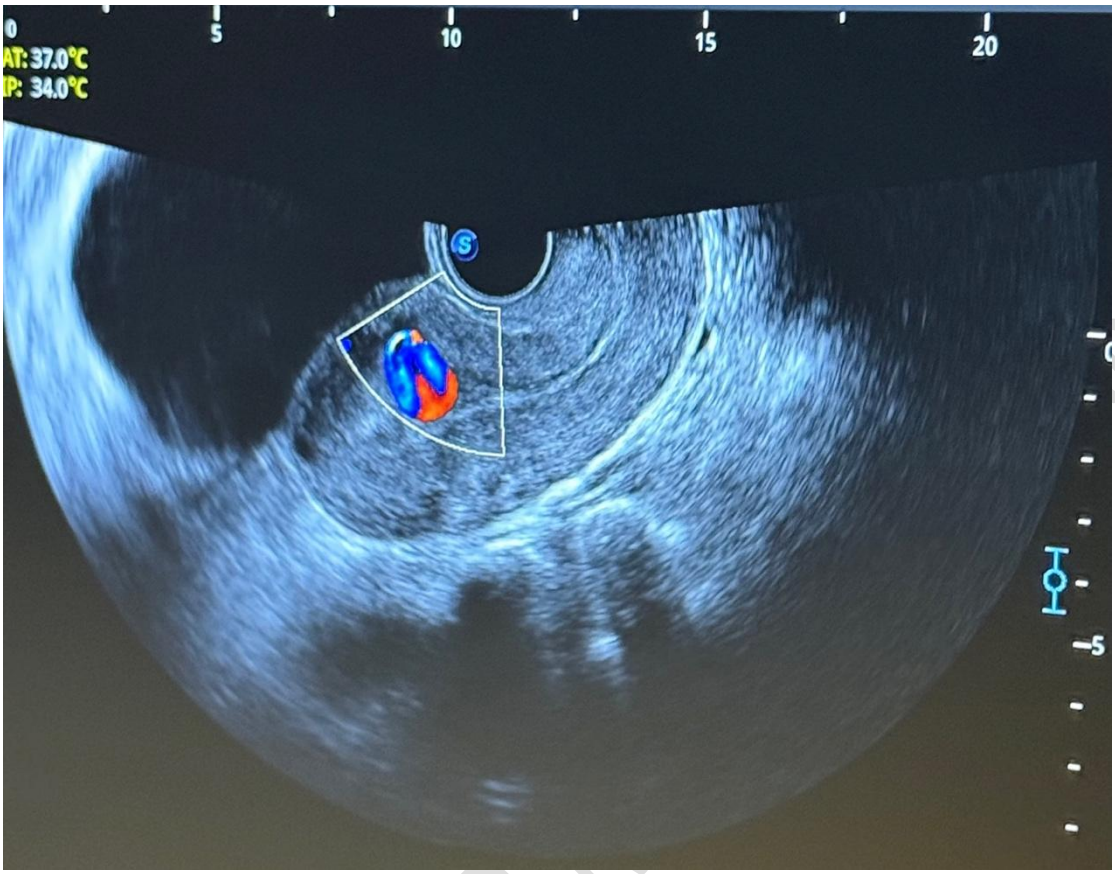
Post-evacuation surveillance was performed with serial serum β-hCG measurements. Owing to persistent elevation of β-hCG levels, the patient was diagnosed with gestational trophoblastic neoplasia and received methotrexate chemotherapy. Treatment was well tolerated and resulted in complete biochemical remission, with normalization of serum β-hCG levels at the end of therapy.

Four months after uterine evacuation, the patient presented with intermittent vaginal bleeding (metrorrhagia). She was hemodynamically stable, and physical examination revealed no signs of active severe hemorrhage. Repeat serum β-hCG testing remained negative, arguing against persistent or recurrent trophoblastic disease.

Transvaginal color Doppler ultrasonography demonstrated a hypervascular lesion within the myometrium characterized by multiple tortuous vascular channels with increased blood flow, highly suggestive of a uterine arteriovenous malformation. Given the absence of biochemical evidence of active gestational trophoblastic disease, the diagnosis of acquired uterine arteriovenous malformation was established.

Considering the patient's stable clinical condition and the self-limited nature of the bleeding, conservative management with close clinical and ultrasonographic follow-up was adopted. No embolization or surgical intervention was required. During follow-up, the metrorrhagia

progressively resolved, and the patient remained asymptomatic, with no evidence of recurrent bleeding or trophoblastic disease.



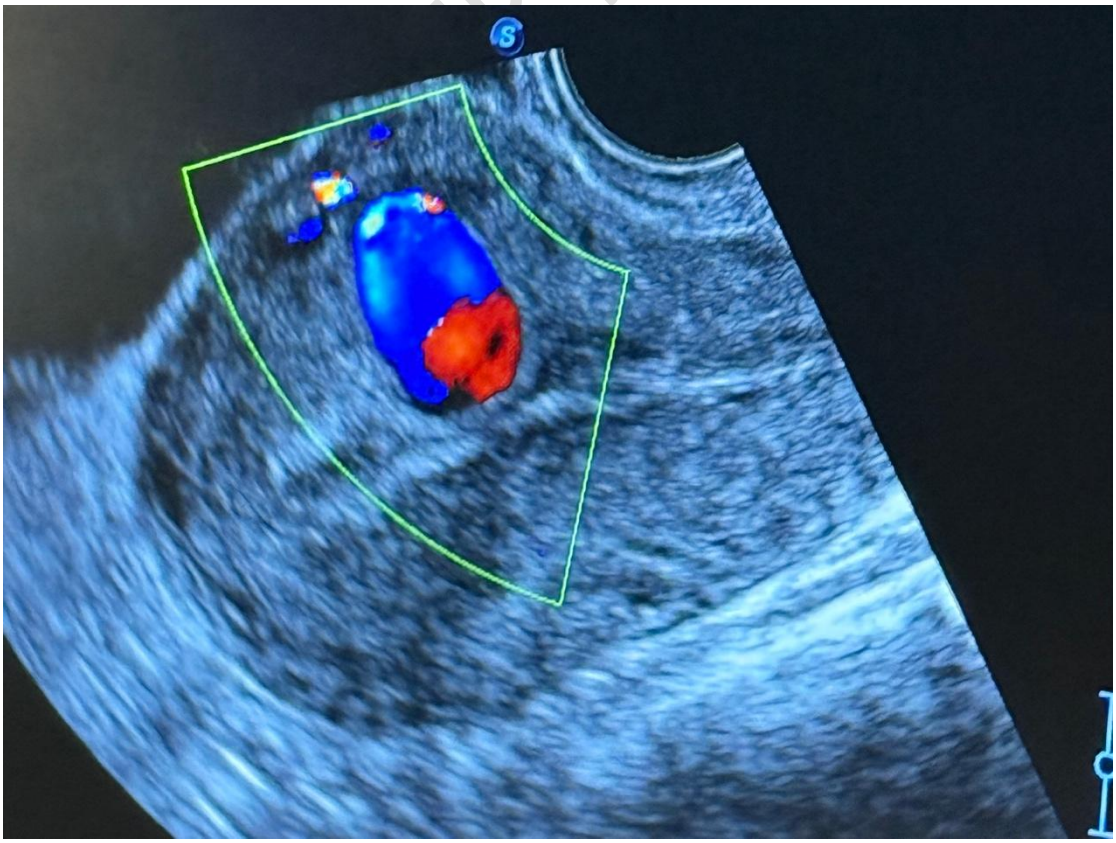


Figure 1-2 Transvaginal ultrasound demonstrating a heterogeneous hypervascular myometrial lesion following treatment of gestational trophoblastic neoplasia.

DISCUSSION

Uterine arteriovenous malformation (UAVM) is an uncommon but potentially life-threatening vascular lesion characterized by abnormal direct communications between the arterial and venous systems without an intervening capillary network. Although congenital forms have been described, most cases encountered in gynecological practice are acquired and occur following uterine trauma, including curettage, cesarean section, uterine surgery, or gestational trophoblastic disease (GTD) [1,2].

The association between GTD and UAVM remains rare. The exact pathophysiological mechanism has not been fully elucidated; however, several hypotheses have been proposed. Persistent trophoblastic invasion may promote abnormal angiogenesis and vascular remodeling, while uterine instrumentation performed during evacuation of a molar pregnancy may contribute to the development of arteriovenous fistulous communications [3]. In our patient, both suction evacuation and subsequent gestational trophoblastic neoplasia may have played a role in the formation of the vascular malformation.

The diagnosis of UAVM during follow-up of GTD represents a significant clinical challenge. Patients commonly present with abnormal uterine bleeding, ranging from mild metrorrhagia to severe hemorrhage requiring urgent intervention [4]. In the present case, the patient developed intermittent vaginal bleeding four months after evacuation. The principal diagnostic dilemma was distinguishing UAVM from persistent or recurrent gestational trophoblastic neoplasia. This distinction is crucial because management strategies differ substantially. While recurrent GTN requires further chemotherapy, invasive uterine procedures performed in patients with UAVM may precipitate catastrophic hemorrhage [3,5].

Serial β-hCG monitoring remains a key element in the differential diagnosis. In our patient, complete normalization of β-hCG levels following methotrexate treatment strongly argued against persistent trophoblastic activity. Similar observations have been reported in previous studies, highlighting the importance of correlating biological and imaging findings before establishing the diagnosis of recurrent GTN [3].

Color Doppler ultrasonography is considered the first-line imaging modality for the diagnosis of UAVM. Typical findings include a heterogeneous myometrial lesion containing multiple serpiginous vascular channels with high-velocity, low-resistance blood flow [4]. Although angiography remains the gold standard for definitive diagnosis, it is generally reserved for cases requiring therapeutic embolization [6]. In our case, Doppler ultrasonography provided sufficient evidence to establish the diagnosis and guide management.

Management of UAVM depends on the severity of symptoms, hemodynamic status, reproductive wishes, and imaging findings. Uterine artery embolization has become the preferred treatment for symptomatic patients or those experiencing significant bleeding, with reported success rates exceeding 85–90% [6,7]. Nevertheless, conservative management may be appropriate in selected clinically stable patients with limited bleeding and no evidence of

ongoing trophoblastic disease [8]. Our patient was managed expectantly because she remained hemodynamically stable and her bleeding was not severe. This approach resulted in spontaneous resolution of metrorrhagia without the need for invasive intervention.

This case emphasizes the importance of considering UAVM in the differential diagnosis of abnormal uterine bleeding occurring after treatment of gestational trophoblastic neoplasia, even in the presence of complete β-hCG remission. Early recognition of this rare complication may prevent unnecessary procedures and avoid potentially life-threatening hemorrhagic events.

CONCLUSION

Uterine arteriovenous malformation (UAVM) is a rare but clinically significant complication that may occur following gestational trophoblastic disease and uterine instrumentation [1,2]. Its diagnosis during post-molar follow-up is particularly challenging because its clinical and sonographic features may mimic persistent gestational trophoblastic neoplasia [3]. This case highlights the importance of correlating Doppler ultrasonography findings with serial β-hCG measurements. In our patient, normalization of β-hCG levels after methotrexate therapy was instrumental in excluding persistent trophoblastic activity, while Doppler ultrasound established the diagnosis of UAVM. As previously reported, conservative management may be a safe and effective option in hemodynamically stable patients with mild symptoms [2,4]. Early recognition of this uncommon condition is essential to avoid unnecessary invasive procedures and potentially life-threatening hemorrhagic complications. A multidisciplinary approach involving gynecologists, radiologists, and trophoblastic disease specialists is crucial for optimizing patient outcomes [3,5].

References

1. Ngan HYS, Seckl MJ, Berkowitz RS, Xiang Y, Golfier F, Sekharan PK, et al. Update on the diagnosis and management of gestational trophoblastic disease. *Int J Gynaecol Obstet*. 2021;155(Suppl 1):86–93.
2. Peitsidis P, Manolakos E, Tsekoura V, Kreienberg R, Schwentner L. Uterine arteriovenous malformations induced after diagnostic curettage: a systematic review. *Arch Gynecol Obstet*. 2011;284(5):1137–1151.
3. Touhami O, Grégoire J, Noël P, Trinh XB, Plante M. Uterine arteriovenous malformations following gestational trophoblastic neoplasia: A systematic review. *Eur J Obstet Gynecol Reprod Biol*. 2014;181:54–59.
4. Timmerman D, Van den Bosch T, Peeraer K, Debrouwere E, Van Schoubroeck D, Stockx L, et al. Vascular malformations in the uterus: ultrasonographic diagnosis and conservative management. *Eur J Obstet Gynecol Reprod Biol*. 2000;92(1):171–178
5. Timmerman D, Van den Bosch T, Peeraer K, Debrouwere E, Van Schoubroeck D, Stockx L, et al. Vascular malformations in the uterus: ultrasonographic diagnosis and conservative management. *Eur J Obstet Gynecol Reprod Biol*. 2000;92(1):171–178.
6. O'Brien P, Neyastani A, Buckley AR, Chang SD, Legiehn GM. Uterine arteriovenous malformations: From diagnosis to treatment. *J Ultrasound Med*. 2006;25(11):1387–1392.
7. Seckl MJ, Sebire NJ, Berkowitz RS. Gestational trophoblastic disease. *Lancet*. 2010;376(9742):717–729.

8. Yoon DJ, Jones M, Taani JA, Buhimschi C, Dowell JD. A systematic review of acquired uterine arteriovenous malformations: Pathophysiology, diagnosis, and transcatheter treatment. *AJR Am J Roentgenol*. 2016;207(4):725–732.