

# Super Selective Arterial Embolization for Hemorrhage Due To Malignant Gestational Trophoblastic Tumor: A Case Report

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**Abstract:** *Gestational trophoblastic neoplasia (GTN) is a highly vascularized gynecologic malignancy associated with heavy bleeding. Bleeding due to GTN can be very difficult to control, due to the proliferation of the tumor and its myometrial invasion. Selective arterial embolization (SAE) is widely used for the control of life-threatening bleeding. We report the case of a young patient who was hospitalized for management of a gestational trophoblastic tumor and presented with a fulminant hemorrhage that was controlled by selective arterial embolization.*

**Keywords:** gestational trophoblastic tumor.hemorrhage. transcatheter arterial embolization. superselective

## Introduction:

Gestational trophoblastic neoplasia (GTN) is the most curable gynecologic malignancy because it is chemo sensitive[1,2]. However, GTN is highly vascularized and associated with massive bleeding[3,4].

Bleeding from GTN can be very difficult to control, due to the proliferation of the tumor and its invasion into the endometrium and myometrium. Selective arterial embolization (SAE) is widely used for the control of life-threatening bleeding[5,6].

## Case Presentation:

Mrs. A. E., 18 years old, with no notable pathological history, primigravida (a miscarriage of 2 months without curettage) who consulted for metrorrhagia on an amenorrhea of 2 months. Clinical examination showed moderate metrorrhagia from the uterine endocervix, an enlarged uterus halfway to the umbilicus), HCG level at 600,000 IU/L and pelvic ultrasound showed a heterogeneous echogenic intrauterine grape image of 7cm/10cm, both ovaries normal size; the diagnosis of a molar pregnancy was suspected. An ultrasound-guided aspiration was performed until a good interface line was obtained. Pathological examination of the product came back in favor of a complete hydatidiform mole. 10 days later, the HCG level had decreased to 200,000 IU/L with a 3 cm intrauterine retention image on the follow-up ultrasound scan, the 2 ovaries still being of normal size. A second ultrasound-guided aspiration was performed until uterine vacuity was obtained. The patient was seen in a follow-up consultation 10 days after the second aspiration: the HCG level continued to fall to 120,000 IU/L, the follow-up ultrasound scan showed the reappearance of a 2cm retention image, with the visualization of large ovaries measuring 10cm each, macropolycystic, and absence of effusion in the pouch of Douglas. we continued the weekly monitoring of the HCG level and to repeat a control ultrasound in one week. The patient reconsulted in 10 days with the notion of the appearance of intense diffuse pelvic pain and the reappearance of metrorrhagia. The clinical examination showed a bleeding made of red blood of small abundance coming from the endocervix. The HCG level continued to evolve well, returning to 15,000 IU/L.

Pelvic ultrasound showed an image of retention that increased in volume compared with the previous examination, measuring 4cm/6cm (Figure 1), with a small image of the fundal uterine wall measuring 1.5cm/2cm, hypo-echogenic and heterogeneous, taking the color Doppler, making it possible to suspect invasion of the myometrium (Figure 1).

During her hospitalization, the patient presented very heavy metrorrhagia of abrupt onset, which led to the indication for emergency embolization of the uterine arteries. This stopped the haemorrhage (Figure 2).

After puncture of the right femoral artery, catheterization of the left iliac artery then left uterine arteriography revealed a hypervascularized fundic image lateralized on the left (figure 2.1). A selective embolization of the left uterine artery was performed using gelatin fragments and then an angiographic series was performed to verify the correct occlusion of the vessel. Catheterization of the right side was performed in the same way, objectifying a participation of the right uterine artery in the vascularization of the mass (figure 2.2) hence the decision to perform an embolization even on the right side.

Pelvic MRI showed an enlarged uterus measuring 170 mm/65 mm in height and 65 mm in anteroposterior diameter, with a fundal mass in the left lateral wall, intramyometrial, described in T1 and T2 iso signal, mostly liquefied and measuring 30 mm/17 mm in diameter. The 2 ovaries are increased in size and polymacrocystic. The diagnosis of gestational trophoblastic tumor was retained on the basis of the clinical data and the imaging data (ultrasound and MRI) even though the evolution of the HCG hormone was favorable. The extension work-up made of cerebro-thoraco-abdomino-pelvic CT scan reported a secondary pulmonary localization and suspicious right hilar adenopathy (figure 3, 4). A standard chest X-ray showed a right lower lobar lung parenchymal nodule. The tumor was scored as a low-risk 4. Monochemotherapy with methotrexate 1mg/kg intra-muscularly was started. Our patient received her first treatment with good clinical evolution.

**Discussion:**

The hypervascular and friable nature of the GTN puts the patient at increased risk of significant bleeding [9]. Vaginal hemorrhage caused by GTN can be a challenge in the management of these patients. Traditionally, gauze packs and surgery, such as abdominal hysterectomy or uterine artery ligation, were the treatment of choice to control refractory hemorrhage [10,11]. Recently, SAE has been shown to be a safe and highly effective alternative procedure for massive genital hemorrhage [5,7]. As with postpartum hemorrhage, interventional therapies play a key role in the management of patients with gynecologic hemorrhage. Most of the data in the literature have focused on the postpartum hemorrhage setting, but the same principles apply to the management of gynecologic malignancies, such as GTN [12,15]. This technique has several advantages, including avoiding major surgery and general anesthesia and preserving fertility in young patients [6].

Embolization may be more complicated in GTN than in postpartum hemorrhage. GTN can destroy blood vessel walls and connect arteries and veins, facilitating the formation of uterine arteriovenous malformations [9,16]. If there is a large fistula in the AVM, SAE should be performed with caution. In the case of a large fistula, coils are first used to embolize the feeding arteries, as the gelatin sponge may pass through the fistula and cause complications. The vessels that cause bleeding in the female reproductive tract are usually predictable. The uterine artery supplies the uterus and cervix and accounts for the majority of bleeding; however, the tumor often extends outside of these structures into the pelvis, and other vessels of the anterior division are recruited to supply the tumor. It seems logical that the presence of collaterals to the uterus increases the risk of rebleeding [17]. Several studies confirm the importance of non-selective pelvic arteriography to identify all the branches vascularizing the tumor.

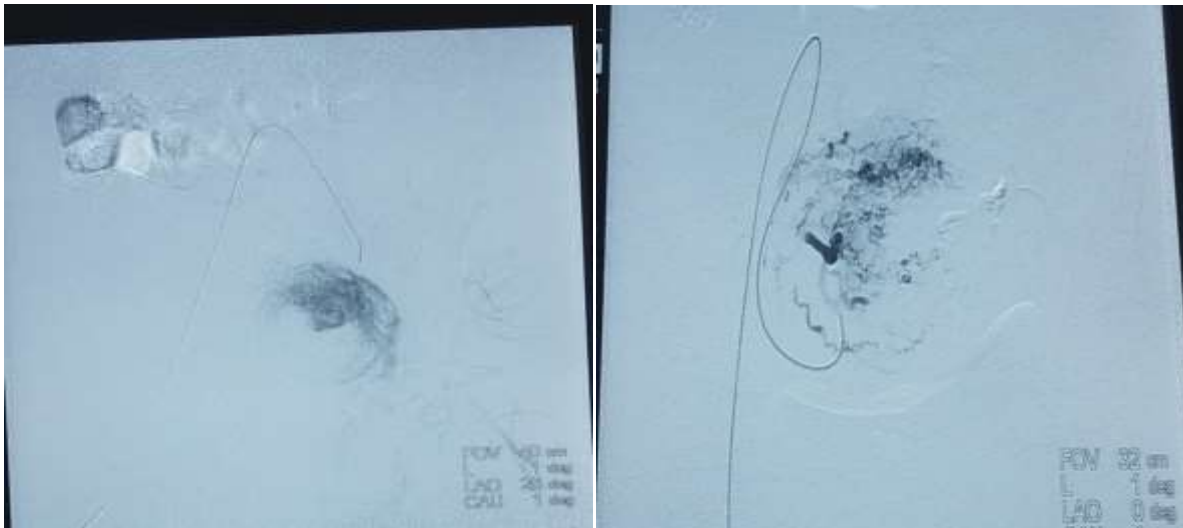
Following the introduction of chemotherapy, cure rates for choriocarcinoma and invasive mole have increased significantly. Because of their remarkable sensitivity to chemotherapy, cure rates are almost 100% in the low-risk group and almost 80% in the high-risk group with current chemotherapy regimens. [1,2,18,19]. Chemotherapy is the primary treatment modality for patients with GTN. Some physicians were unwilling to accept SAE, and only did so when the patient had life-threatening bleeding. They thought that embolization of a tumor-feeding artery could block the release of chemotherapeutic drugs into the tumor tissue. Studies done in this direction showed that SAE did not affect subsequent systemic chemotherapy, and that the prognosis of malignant GTN with massive vaginal bleeding was good when systemic chemotherapy was applied after successful SAE. Carlini et al [20] reported on a patient who did not receive chemotherapy after the diagnosis of GTN and had decreasing and then normal HCG levels after uterine AVM embolization. SAE facilitates trophoblast shrinkage by significantly reducing blood supply to uterine vessels; however, Maleux et al [16] reported that this underlying AVM malignancy was the source of early recurrent bleeding, even after technically successful embolization, due to neoangiogenesis and formation of another AVM. Although SAE itself can control tumor growth, all patients underwent chemotherapy after successful SAE, and no recurrent massive bleeding occurred during the chemotherapy period. Lim et al [14] reported the persistence of uterine AVMs in 10% to 15% of TNM patients, even after complete tumor remission after chemotherapy. [14] The current data also showed that uterine bleeding recurred in two patients due to uterine AVMs, even after complete embolization and CR of GTN. In our case, gelatin sponge was used as an embolic agent because the temporary occlusion caused by gelatin sponge seems to be sufficient to prevent rebleeding during chemotherapy, whereas AVMs may recur during long-term follow-up, possibly due to the opening of collateral vessels [21]; however, repeated embolotherapy may be successful in treating these patients. Uterine hemorrhage may recur due to uterine AVMs even after complete embolization and CR of GTN.

**Conclusion:**

In conclusion, the transcatheter superselective arterial embolization to treat hemorrhage from malignant gestational trophoblastic tumor appear to be efficacious and relatively safe. This kind of treatment option should be the first line of therapy, and the superselective arterial catheterization must be applied.



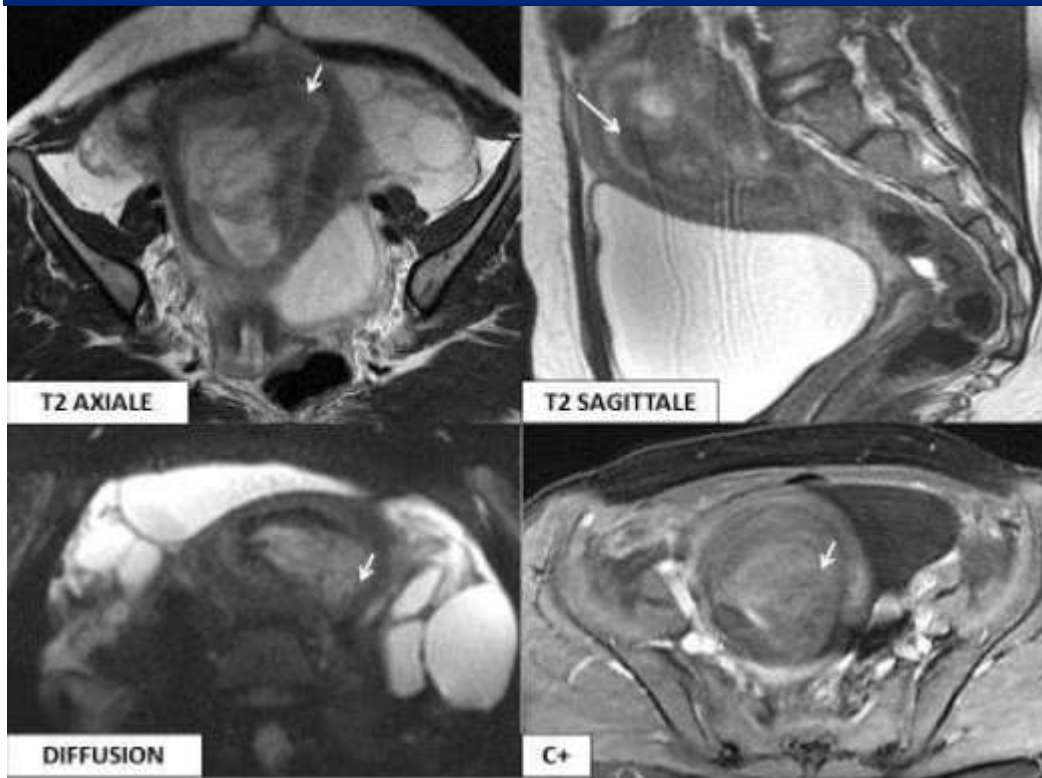
**Figure 1:** Heterogeneous echogenic intra uterine image of 4.x6cm with presence of heterogeneous hypo echogenic posterior uterine wall image taking color Doppler making suspicion of myometrial invasion.



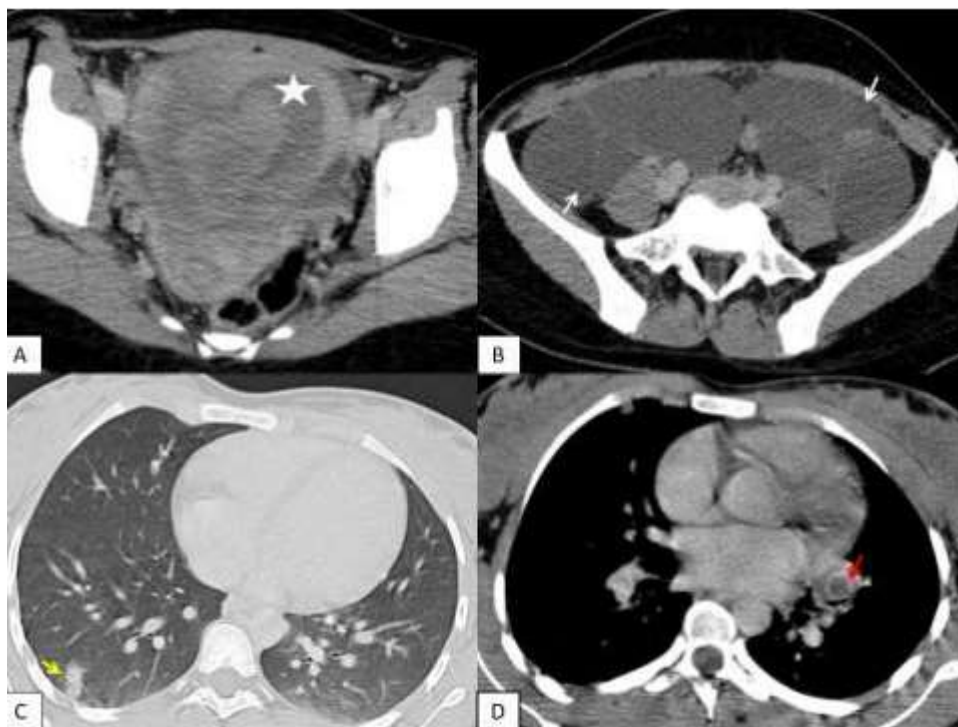
**Figure 2.1:** left uterine artery

**Figure 2.2:** right uterine artery

**Figure 2:** uterine arteriography showing the hypervascularized fundic image .



**Figure 3:** Pelvic MRI showing an enlarged uterus with a fundal mass on the left lateral wall, intra myometrial, described in T1 and T2 isosignal. It is associated with the presence of a significant hematoma, which hinders the study of the enhancement.



**Figure 4:** thoracic-abdominal-pelvic CT scan showing a heterogeneous uterus with a left lateral fundal mass (star) with hematometry and macro follicular ovaries (white arrows). Secondary localizations

pulmonary (yellow arrow) and suspicious right hilar ADP (red arrow).

#### Disclosure of interest

The authors declare that they have no competing interest.

#### Figures :

**Figure 1:** Heterogeneous echogenic intra uterine image of 4.x6cm with presence of heterogeneous hypo echogenic posterior uterine wall image taking color Doppler making suspicion of myometrial invasion.

#### Figure 2:

**Figure 3:** Pelvic MRI showing an enlarged uterus with a fundal mass on the left lateral wall, intra myometrial, described in T1 and T2 isosignal. It is associated with the presence of a significant hematoma, which hinders the study of the enhancement.

**Figure 4:** thoracic-abdominal-pelvic CT scan showing a heterogeneous uterus with a left lateral fundal mass (star) with hematometry and macro follicular ovaries (white arrows). Secondary localizations pulmonary (yellow arrow) and suspicious right hilar ADP (red arrow).

#### References:

1. Froeling FE, Seckl MJ. Gestational trophoblastic tumours: an update for 2014. *Curr Oncol Rep* 2014;16:408.
2. Mangili G, Lorusso D, Brown J, et al. Trophoblastic disease review for diagnosis and management: a joint report from the International Society for the Study of Trophoblastic Disease, European Organisation for the Treatment of Trophoblastic Disease, and the Gynecologic Cancer InterGroup. *Int J Gynecol Cancer* 2014;24:S109e16.
3. Touhami O, Gregoire J, Noel P, et al. Uterine arteriovenous malformations following gestational trophoblastic neoplasia: a systematic review. *Eur J Obstet Gynecol Reprod Biol* 2014;181:54e9.
4. Moodley M, Moodley J. Transcatheter angiographic embolization for the control of massive pelvic haemorrhage due to gestational trophoblastic disease: a case series and review of the literature. *Int J Gynecol Cancer* 2003;13:94e7.
5. Hongsakul K, Songjamrat A, Rookkapan S. Transarterial embolization for the treatment of massive bleeding in gynecologic and obstetric emergencies: a single center experience. *Emerg Radiol* 2014;21:333e9.
6. Inoue S, Masuyama H, Hiramatsu Y, Multi-Institutional Study Group of Transarterial Embolization for Massive Obstetric Haemorrhage in Chugoku & Shikoku Area Society of Obstetrics and Gynecology. Efficacy of transarterial embolisation in the management of post-partum haemorrhage and its impact on subsequent pregnancies. *Aust N Z J Obstet Gynaecol* 2014;54:541e5.
7. Salazar GM, Petrozza JC, Walker TG. Transcatheter endovascular techniques for management of obstetrical and gynecologic emergencies. *Tech Vasc Interv Radiol* 2009;12:139e47.
8. Hancock BW. Staging and classification of gestational trophoblastic disease. *Best Pract Res Clin Obstet Gynaecol* 2003;17:869e83.
9. Method MW, Hirschfield M, Everette HE. Angiographic-guided embolization of metastatic invasive mole. *Gynecol Oncol* 1996;61:442e5.
10. Paraskevaides E, Noelke L, Afrasiabi M. Internal iliac artery ligation (IIAL) in obstetrics and gynaecology. *Eur J Obstet Gynecol Reprod Biol* 1993;52:73e5.
11. Chattopadhyay SK, Deb Roy B, Edrees YB. Surgical control of obstetric haemorrhage: hypogastric artery ligation or hysterectomy? *Int J Gynaecol Obstet* 1990;32:345e51.
12. Method MW, Hirschfield M, Everette HE. Angiographic guided embolization of metastatic invasive molar pregnancy. *Gynecol Oncol* 1996;61:442e5.
13. Shen K, Yang X, Song H, et al. Selective arterial embolization in the management of internal bleeding caused by trophoblastic diseases. *Chin Med J* 1996;109:151e6.
14. Lim AK, Agarwal R, Seckl MJ, et al. Embolization of bleeding residual uterine vascular malformations in patients with treated gestational trophoblastic tumours. *Radiology* 2002;222:640e4.

15. Frati A, Ducarme G, Wernet A, et al. Uterine artery embolization as treatment for life-threatening haemorrhage from a cervical choriocarcinoma: a case report. *Eur J Obstet Gynecol Reprod Biol* 2008;141:87e8.
16. Maleux G, Timmerman D, Heye S, et al. Acquired uterine vascular malformations: radiological and clinical outcome after transcatheter embolotherapy. *Eur Radiol* 2006;16:299e306.
17. Sentilhes L, Gromez A, Clavier E, et al. Predictors of failed pelvic arterial embolization for severe postpartum haemorrhage. *Obstet Gynecol* 2009;113:992e9.
18. Maesta I, Berkowitz RS, Goldstein DP, et al. Relationship between race and clinical characteristics, extent of disease, and response to chemotherapy in patients with low-risk gestational trophoblastic neoplasia. *Gynecol Oncol* 2015;138:50e4.
19. Chu MM, Ma Y, Tse KY, et al. Cyclophosphamide, hydroxyurea, actinomycin D, methotrexate, and vincristine in the treatment of gestational trophoblastic neoplasia. *Int J Gynecol Cancer* 2015;25:498e503.
20. Carlini L, Villa A, Busci L, et al. Selective uterine artery embolization: a new therapeutic approach in a patient with low-risk gestational trophoblastic disease. *Am J Obstet Gynecol* 2006;195:314e5.
21. Vaidya S, Tozer KR, Chen J. An overview of embolic agents. *Semin Interv Radiol* 2008;25:204e15.