A Delayed Metastatic Storm after Molar Pregnancy: CT Chest and Brain Findings of Choriocarcinoma with Life-Threatening Hemoptysis

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Dr. Omima Elemam- Lab report review, study-specific physical exams, medical history and patient's treatment.
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DISCLOSURES
The authors declare no conflicts of interest related to this study.

CONSENT
Written consent from the patient

HUMAN AND ANIMAL RIGHTS
Written consent was obtained from the patient and IRB approval from King Abdullah Medical City (approval number: 25 - 1435).

ABSTRACT

Choriocarcinoma is a highly aggressive form of gestational trophoblastic neoplasia that may develop years after a molar pregnancy. We report a 22-year-old woman with severe hemoptysis and a history of molar pregnancy two years prior. Her β-hCG level was 225,000 mIU/mL. Chest CT showed a hypervascular right lung mass with arterial supply from multiple systemic vessels, suggestive of arteriovenous shunting. Brain MRI revealed hemorrhagic occipital metastasis. No uterine lesion was found. Metastatic choriocarcinoma was diagnosed. She underwent arterial embolization followed by systemic chemotherapy with etoposide–cisplatin and EMA-CO. Her β-hCG level declined to 10,090 mIU/mL. This case illustrates an atypical, delayed metastatic presentation of choriocarcinoma. In women of reproductive age with a prior gestational event, hypervascular pulmonary and cerebral lesions with elevated β-hCG should prompt consideration of metastatic choriocarcinoma.

CASE REPORT

BACKGROUND

Choriocarcinoma is an uncommon and very aggressive form of gestational trophoblastic neoplasia (GTN), likely to metastasize via the hematogenous pathway. While the majority of cases occur within a year of a molar pregnancy, this case illustrates a very delayed life-threatening metastatic presentation two years after a molar pregnancy evacuation. It highlights the diagnostic challenges and some critical clinical considerations associated with metastatic choriocarcinoma. The profound hemoptysis in our patient—along with imaging findings of hypervascular pulmonary and cerebral metastases resembling vascular malformations—demonstrate the considerable difficulties encountered in identifying and diagnosing this aggressive malignancy.

The case is unusual in its presentation of a "metastatic storm" phenotype with unique systemic arterial supply to the pulmonary tumor—prompting consideration for alternative differential diagnoses such as pulmonary arteriovenous malformation and sequestration. This underscores the diagnostic complexity

posed by the unique vasculogenic mimicry characteristic of choriocarcinoma.

Early radiological recognition, prompt intervention, and multiple therapies are crucial for effective management of aggressive conditions like metastatic choriocarcinoma. Our case emphasizes the importance of cross-sectional imaging in establishing the diagnosis of metastatic choriocarcinoma, even in the absence of histopathological confirmation. As demonstrated in our case, hemorrhagic complications can be managed successfully with emergent embolization followed by systemic chemotherapy, underscoring the need for integrated therapeutic approaches.

To the best of our knowledge, this case is one of the few reported cases in which CT angiographic findings, transcatheter arterial embolization, and early biochemical response monitoring have been integrated and illustrated. It highlights the importance of including choriocarcinoma in the differential diagnosis for females of reproductive age with prior gestational events who present with unexplained hemoptysis, elevated β-hCG levels, and hypervascular lesions–regardless of the time interval since the gestational event.

Patient Presentation

A 22-year-old nulliparous woman with a history of multiple miscarriages and a molar pregnancy evacuated two years earlier presented with sudden, life-threatening hemoptysis. On admission, she was hemodynamically stable. Lung auscultation revealed clear breath sounds without rales or pleural rubs. Laboratory studies showed a serum β-human chorionic gonadotropin (β-hCG) concentration of 225,000 mIU/mL. Other tumor markers, including α-fetoprotein, were within reference limits. Inflammatory indices were elevated (lactate dehydrogenase 428 U/L; C-reactive protein 1.38 mg/dL; erythrocyte sedimentation rate 42 mm/h).

Imaging Findings

Contrast-enhanced computed tomography (CT) of the chest revealed a large, lobulated, hypervascular soft-tissue mass occupying the right lower lobe and extending into the middle lobe, measuring approximately 9.5 × 8.5 × 7.0 cm. Axial reformats in the arterial (Figures 1A,1B) and venous (Figure 1C) phases demonstrated marked peripheral enhancement alongside prominent, dilated vessels with imaging features of arteriovenous shunting. These findings were highly suggestive of a hypervascular lesion, raising strong suspicion for metastatic choriocarcinoma.

A detailed anatomical delineation of the systemic arterial supply is shown in (Figure 2), which revealed abnormal vessels adjacent to the mass, particularly the right bronchial artery shown in (Figures 2A,2C) and the right phrenic artery shown in (Figure 2B). Figure 2D was generated using a Maximum Intensity Projection (MIP) reformat with digital magnification to enhance the visualization of three major systemic feeders: (1) hypertrophied branch of the subclavian artery (2) right bronchial artery from the descending thoracic aorta. (3) branch from the right phrenic artery. These abnormal systemic vessels converged on the pulmonary lesion and were interpreted as non-pulmonary arterial feeders, supporting the diagnosis of metastatic choriocarcinoma.

Subsequent digital subtraction angiography (DSA) further confirmed these vascular findings. As shown in (Figure 3), selective arteriography demonstrated four distinct arterial feeders supplying the right lung mass: (1) a branch from the right subclavian artery, (2) the right internal mammary artery, (3) the right bronchial artery and (4) the right inferior phrenic artery. These are indicated by white arrows (1-4) on the angiographic image. The extensive systemic arterial involvement, with atypical and redundant supply, mimicked congenital vascular anomalies such as pulmonary sequestration or arteriovenous malformation, complicating the radiological differential.

Pelvic ultrasound and abdominopelvic CT showed no uterine or adnexal abnormalities. Brain magnetic resonance imaging (MRI) disclosed a right occipital lesion, which measured 2 x 2 x 2 cm, with central hyperintensity on T2-weighted imaging/fluid-attenuated inversion recovery (T2/FLAIR), susceptibility blooming on susceptibility-weighted imaging (SWI), and avid post-contrast enhancement, consistent with hemorrhagic metastasis (Figure 4A, B, C and D) respectively.

Management and Follow-Up

Given the markedly elevated β-hCG level, prior molar pregnancy, and characteristic hypervascular pulmonary and cerebral lesions, metastatic choriocarcinoma was diagnosed. Selective transarterial embolization of three of the pulmonary tumor feeders achieved immediate hemostasis (Figure 5). The right inferior phrenic artery was not embolized due to difficult access. Systemic chemotherapy followed one cycle of etoposide–cisplatin and one cycle of EMA-CO (etoposide, methotrexate, actinomycin-D, cyclophosphamide, and vincristine). Post-treatment β-hCG declined from 225,000 mIU/mL to 10,090 mIU/mL, indicating a robust early biochemical response. The patient remains under close oncologic and radiologic surveillance for further β-hCG regression and imaging evidence of remission.

DISCUSSION

Etiology & Demographics

Gestational trophoblastic neoplasia (GTN) comprises a spectrum of malignant trophoblastic disorders, of which choriocarcinoma is the most aggressive subtype [1]. Epidemiologically, malignant transformation follows 15–20 % of complete moles and ≈ 2% of partial moles [2]. True choriocarcinoma is rare, occurring in about 1 in 40,000 pregnancies in Western countries and up to 9.2 in 40,000 in parts of Southeast Asia [3,4]. Risk is heightened at maternal ages < 20 years or > 40 years and after previous molar gestation, as in our patient [5]. Although presentation typically occurs

within the first post-molar year, intervals of several years—even decades—have been reported [6,7].

Clinical & Imaging Findings

Choriocarcinoma metastases early and hematogenously. Pulmonary spread occurs in ≈ 80% of patients and may precipitate paraneoplastic crises such as thyroid storm [8]. Systematic reviews confirm that lung involvement, brain metastasis and high FIGO score predict adverse outcome [9]. Vascular invasion is promoted by trophoblastic vasculogenic mimicry, producing exuberant neoplastic channels and "cannon-ball" nodules rimmed by hemorrhage on CT [10]. Fulminant hemorrhagic events ("choriocarcinoma syndrome") are classically associated with marked β-hCG elevation (> 50,000 mIU mL) [11,12].

Angiography commonly reveals hypervascular metastases supplied by systemic feeders, sometimes from unusual territories such as hepatic or phrenic branches [13,14]. Such anatomy can mimic pulmonary arteriovenous malformation or sequestration, while massive hemoptysis may be misattributed to tuberculosis in endemic regions [15,16]. Multiphase CT and MRI therefore remain critical for staging and treatment planning [17]. Brain metastases complicate 10–20 % of cases and are characteristically hemorrhagic [18]. Contemporary imaging algorithms recommend chest CT, pelvic imaging and brain MRI for all patients with proven GTN [19,20].

Treatment & Prognosis

Modern management is risk-stratified. High-risk disease (FIGO score ≥ 7) mandates multi-agent chemotherapy; EMA-CO yields overall survival ≈ 85–90% [21]. Rare metastatic sites, including vagina [22] and liver [23], may bleed catastrophically, necessitating selective trans-arterial embolization (TAE) before systemic therapy. Long-term reproductive and obstetric outcomes remain excellent after chemotherapy, as demonstrated in meta-analyses [24,25]. Prognosis worsens with chemoresistance, liver metastasis or FIGO score >12; nonetheless, five-year survival still approaches 70% in large high-risk cohorts [26]. Aggressive treatment—including emergency craniotomy or stereotactic radiosurgery—achieves remission rates of 80–93 % in patients with brain metastases [27,28]. Global experience highlights that timely diagnosis, access to chemotherapy and interventional radiology determine outcome, particularly in low-resource settings [29].

Differential Diagnoses

Massive hemoptysis in a reproductive-age woman with lung nodules has a broad differential. Infective pseudo-aneurysm, cavitating tuberculosis and primary pulmonary neoplasms are far more common but lack β-hCG elevation [30]. The "burned-out" phenomenon—in which the uterine primary regresses while metastases persist—may further obscure the diagnosis [31]. Systematic reviews underline that unusual presentations (e.g. gastrointestinal bleeding, renal crisis) account for ≈ 15% of reported cases [32]. Extremely delayed presentations up to 12 years after pregnancy and FIGO stage IV disease are well documented [33]. Intervals of 5–7 years, as in other post-molar cases, underscore the need for prolonged vigilance [34]. Pulmonary arteriovenous fistulas acquired within metastatic lesions [35] and collision metastases invading pre-existing cerebral arteriovenous malformations (AVMs) [36] exemplify additional vascular mimics.

In summary, the present case highlights a classic triad—remote molar pregnancy, markedly elevated β-hCG and hyper-vascular pulmonary/brain lesions. Awareness of its protean vascular manifestations permits prompt embolization and curative chemotherapy of this otherwise lethal but highly treatable malignancy.

Clinical Points and Further Considerations

This case reinforces several key principles for clinical practice. **Choriocarcinoma should be considered in any reproductive-age woman presenting with pulmonary and cerebral hypervascular lesions, regardless of the interval since her last pregnancy.** According to the 2021 Society of Gynecologic Oncology guidelines, women of childbearing age who present with pulmonary or cerebral lesions of uncertain origin warrant evaluation for potentially life-threatening conditions associated with elevated β-hCG levels.

The remarkable decline in β-hCG—from 225,000 to 10,090 mIU/mL—further supports the diagnosis of choriocarcinoma in our patient. Notably, this diagnosis was made clinically, without the need for histopathological confirmation, aligning with established guidelines that discourage biopsy in high-risk cases due to the risk of hemorrhage.

Transcatheter arterial embolization (TAE) remains an indispensable interventional technique in the setting of massive hemorrhage. In this case, TAE effectively halted blood loss and provided a critical window for systemic chemotherapy to be initiated. This emphasizes the importance of rapid embolization followed by urgent initiation of chemotherapy as a highly effective combined approach in managing life-threatening choriocarcinoma-related bleeding.

Additionally, this case underscores the importance of **ongoing surveillance after molar pregnancy.** While most cases arise within the first year post-evacuation, delayed presentations—even decades later—are well documented. Clinicians and patients alike must remain vigilant over the long term. The absence of an identifiable uterine primary lesion should not preclude the diagnosis, as the "burned-out" tumor phenomenon is a well-recognized feature of metastatic choriocarcinoma.

Ultimately, the resolution of this complex clinical presentation was achieved through the coordinated efforts of diagnostic radiology, interventional radiology, gynecologic

oncology and medical oncology. This highlights the need for **multidisciplinary collaboration** in the management of advanced gestational trophoblastic neoplasia.

TEACHING POINT

Choriocarcinoma can present years after a molar pregnancy with elevated β-hCG and hypervascular pulmonary or cerebral metastases that mimic vascular malformations. Metastatic choriocarcinoma must be considered in the differential diagnosis of any woman with a prior gestational event who presents with elevated β-hCG, even in the absence of a detectable primary tumor or histopathological confirmation.

CONCLUSION

This case illustrates a rare "metastatic storm" presentation of choriocarcinoma, manifesting as massive hemoptysis two years after a molar pregnancy. The complex hypervascular pattern—characterized by multiple systemic arterial feeders resembling congenital arteriovenous malformations—exemplifies the diagnostic challenge posed by the tumor's vasculogenic mimicry.

The absence of a primary uterine lesion and the lack of histological confirmation can further obscure the diagnosis. Therefore, **a constellation of radiologic findings, comprehensive imaging, elevated β-hCG, and detailed obstetric history must be integrated** for timely recognition. **Long-term follow-up is essential** to ensure disease resolution and to monitor for recurrence.

QUESTIONS AND ANSWERS

Applies to article: *Ngan HYS, Seckl MJ, Berkowitz RS, et al.: Diagnosis and management of gestational trophoblastic disease: 2021 update. Int J Gynecol \& Obstet. 2021, 155:86–93. https://doi.org/10.1002/ijgo.13877*

Question 1: Which of the following answer choices is true:

(1) (β-hCG) stands for beta-human chorionic gonadotropin (applies).

(2) Choriocarcinoma is usually not associated with elevated β-hCG.

(3) Molar pregnancy produces low β-hCG levels.

(4) Progression from complete hydatidiform and partial hydatidiform moles to gestational trophoblastic neoplasm is about 10%

(5) β-hCG levels can be monitored for disease response (applies)

Explanation for question (1):

(1) (β-hCG) stands for beta-human chorionic gonadotropin. (The appropriate sentence in the article is cited in squared brackets [Follow-up with human chorionic gonadotropin (hCG)]).

(2) Choriocarcinoma is usually associated with elevated β-hCG levels. (The appropriate sentence in the article is cited in squared brackets [(hCG) is essential for early diagnosis of gestational trophoblastic neoplasia (GTN)]).

(3) Molar pregnancy produces elevated β-hCG levels. (The appropriate sentence in the article is cited in squared brackets [a plateaued or rising hCG level enables the early detection of progression of complete hydatidiform mole and partial hydatidiform mole to GTN]).

(4) Progression from complete hydatidiform and partial hydatidiform moles to gestational trophoblastic neoplasm is about 15-20% and 0.5-5% respectively. (The appropriate sentence in the article is cited in squared brackets [rising hCG level enables the early detection of progression of CHM and PHM to GTN that occurs in 15%−20% and 0.5%−5% of cases, respectively]).

(5) β-hCG levels can be monitored for disease response. (The appropriate sentence in the article is cited in squared brackets [hCG is an excellent biomarker of disease progression, response, and subsequent post-treatment surveillance]).

Applies to article: *Ngan HYS, Seckl MJ, Berkowitz RS, et al.: Diagnosis and management of gestational trophoblastic disease: 2021 update. Int J Gynecol \& Obstet. 2021, 155:86–93. https://doi.org/10.1002/ijgo.13877*

Question 2: Which of the following answer choices is false:

(1) Low risk gestational trophoblastic neoplasms (GTN) is FIGO <7.

(2) High risk gestational trophoblastic neoplasms (GTN) is FIGO <7 (applies).

(3) High risk gestational trophoblastic neoplasms (GTN) is FIGO ≥7.

(4) Choriocarcinoma is a hemorrhagic tumor with necrotic components.

(5) Choriocarcinoma can be found in extra gestational organs.

Explanation for question (2):

(1) Low risk gestational trophoblastic neoplasms (GTN) is FIGO <7. (The appropriate sentence in the article is cited in squared brackets [Low-risk GTN (FIGO Stages I–III: score <7) is treated with single-agent chemotherapy]).

(2) High risk gestational trophoblastic neoplasms (GTN) is FIGO ≥7. (The appropriate sentence in the article is cited in squared brackets [High-risk GTN (FIGO Stages II–III: score ≥7 and Stage IV) is treated with multiagent chemotherapy]).

(3) High risk gestational trophoblastic neoplasms (GTN) is FIGO ≥7. (The appropriate sentence in the article is cited in squared brackets [High-risk GTN (FIGO Stages II–III: score ≥7 and Stage IV) is treated with multiagent chemotherapy]).

(4) Choriocarcinoma is a hemorrhagic tumor with necrotic components. (The appropriate sentence in the article is cited in squared brackets [The tumor is bulky with hemorrhagic and necrotic areas]).

(5) Choriocarcinoma can be found in extra gestational organs. (The appropriate sentence in the article is cited in squared brackets [Apart from the uterus, it can be found in tubes, ovaries, lung, liver, spleen, kidneys, bowel, or brain]).

Applies to article: *Bruce S, Sorosky J: Gestational Trophoblastic Disease. 2025.*

Question 3: Which of the following is true:

(1) Molar pregnancy usually originates from the placenta (applies).

(2) Aggressive malignant forms of gestational trophoblastic disease (GTD) can occur weeks to years following molar pregnancy (applies).

(3) Molar pregnancy contains only one form: partial mole.

(4) GTD is caused by abnormal trophoblastic proliferation (applies).

(5) Hydatidiform moles (HM) are considered malignant.

Explanation for question (3):

(1) Molar pregnancy usually originates from the placenta. (The appropriate sentence in the article is cited in squared brackets [Hydatidiform moles (HM), or molar pregnancy, originates from the placenta]).

(2) Aggressive malignant forms of gestational trophoblastic disease (GTD) can occur weeks to years following molar pregnancy. (The appropriate sentence in the article is cited in squared brackets [These malignancies can occur weeks or even years following any pregnancy but occur most commonly after a molar pregnancy]).

(3) Molr pregnancy can be characterized as a complete or partial mole. (The appropriate sentence in the article is cited in squared brackets [HM is categorized as a complete or partial mole and is usually considered the noninvasive form of GTD]).

(4) GTD is caused by abnormal trophoblastic proliferation. (The appropriate sentence in the article is cited in squared brackets [Gestational trophoblastic disease (GTD) is a group of tumors defined by abnormal trophoblastic proliferation]).

(5) Hydatidiform moles (HM) are considered benign with potential to become malignant. (The appropriate sentence in the article is cited in squared brackets ([Although HMs are usually considered benign, they are premalignant and can potentially become malignant and invasive]).

Applies to article: *Bruce S, Sorosky J: Gestational Trophoblastic Disease. 2025.*

Question 4: Which of the following is true:

(1) Choriocarcinoma is a rare and aggressive cancer (applies).

(2) Choriocarcinoma is benign.

(3) Choriocarcinoma can occur in females as well as males (applies).

(4) There is only one type of choriocarcinoma

(5) Choriocarcinoma is a common neoplasm

Explanation for question (4):

(1) Choriocarcinoma is a rare and aggressive cancer. (The appropriate sentence in the article is cited in squared brackets [Choriocarcinoma is a rare and aggressive neoplasm]).

(2) Choriocarcinoma is not benign. (The appropriate sentence in the article is cited in squared brackets [Choriocarcinoma is a rare and aggressive neoplasm]).

(3) Choriocarcinoma can occur in females as well as males. (The appropriate sentence in the article is cited in squared brackets [Choriocarcinoma predominately occurs in women but can also occur in men, usually as part of a mixed germ cell tumor]).

(4) There are two types of choriocarcinoma. (The appropriate sentence in the article is cited in squared brackets [The 2 significant choriocarcinoma subtypes, namely gestational and nongestational]).

(5) Choriocarcinoma is rare. (The appropriate sentence in the article is cited in squared brackets [Choriocarcinoma is a very rare neoplasm with varied incidence worldwide]).

Applies to article: *Mandava A, Koppula V, Kandati M, Reddy AK, Rajappa SJ, Rao TS: Multimodality Imaging in the Diagnosis and Staging of Gestational Choriocarcinoma. Indian J Radiol Imaging. 2025, 35:148–58. 10.1055/s-0044-1788590*

Question 5: Which of the following is true:

(1) Hemorrhagic complications can be managed successfully with angiographic embolization (applies).

(2) Hemoptysis serves as an indication for the pulmonary metastasis of choriocarcinoma (applies)

(3) Cerebral metastatic choriocarcinoma can present with intracranial hemorrhage (applies).

(4) Choriocarcinoma cannot produce hypervascular and hemorrhagic lesions.

(5) Metastatic choriocarcinoma usually present with "Cannonball" lesions (applies).

Explanation for question (5):

(1) Hemorrhagic complications can be managed successfully with angiographic embolization. (The appropriate sentence in the article is cited in squared brackets [Angiographic embolization is emerging as a successful procedure to control the severe hemorrhage of vaginal tumors]).

(2) Hemoptysis serves as an indication for the pulmonary metastasis of choriocarcinoma. (The appropriate sentence in the article is cited in squared brackets [a few asymptomatic patients can directly present with signs and symptoms of metastatic lesions in the lung (cough, chest pain, dyspnea, hemoptysis]).

(3) Cerebral metastatic choriocarcinoma can present with intracranial hemorrhage. (The appropriate sentence in the article is cited in squared brackets [a few asymptomatic patients can directly present with signs and symptoms of metastatic lesions in the lung (cough, chest pain, dyspnea, hemoptysis, pulmonary arterial hypertension), brain (intracranial bleeding or raised intracranial tension]).

(4) Choriocarcinoma cannot produce hypervascular and hemorrhagic lesions. (The appropriate sentence in the article is cited in squared brackets [Choriocarcinomas produce placental and epidermal growth factors causing exuberant and aberrant neo-angiogenesis, resulting in hypervascular and hemorrhagic lesions: the hallmark of choriocarcinoma and its metastases]).

(5) Metastatic choriocarcinoma usually present with "Cannonball" lesions. (The appropriate sentence in the article is cited in squared brackets [Pulmonary lesions are usually seen as well-defined rounded nodules and multiple nodules may exhibit a "cannonball" appearance, which is typical of hematogenous dissemination]).

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FIGURES

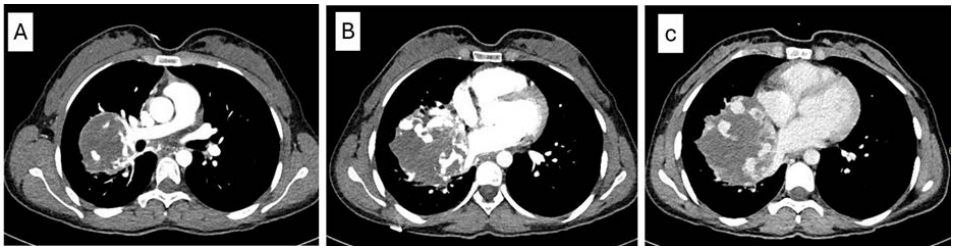

Figure 1: Enhanced CT chest demonstrating a hypervascular pulmonary mass with prominent feeding arteries and draining veins.

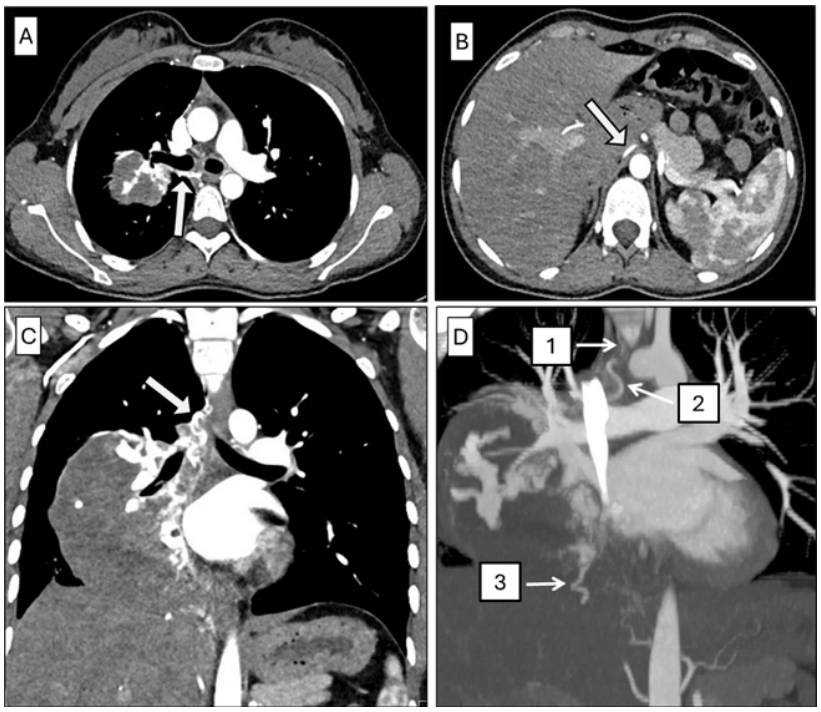

Figure 2: Multiple systemic arterial feeders to right lung choriocarcinoma metastasis

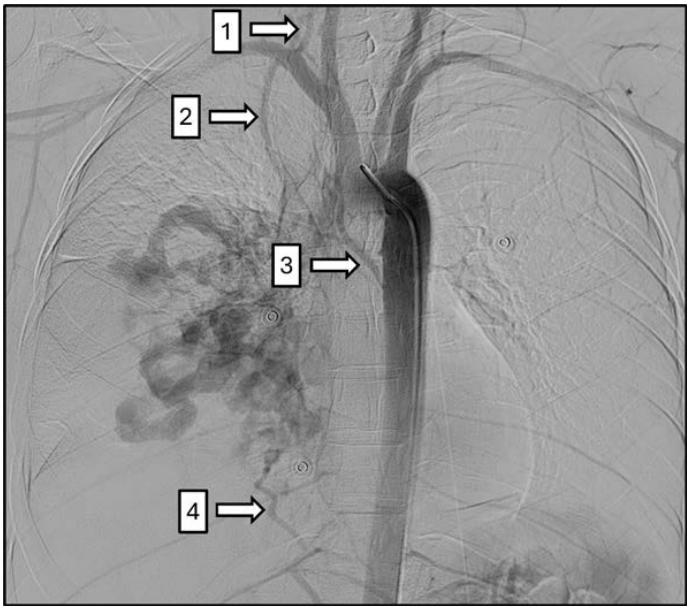

Figure 3: Angiography showing four-vessel arterial supply to the pulmonary mass

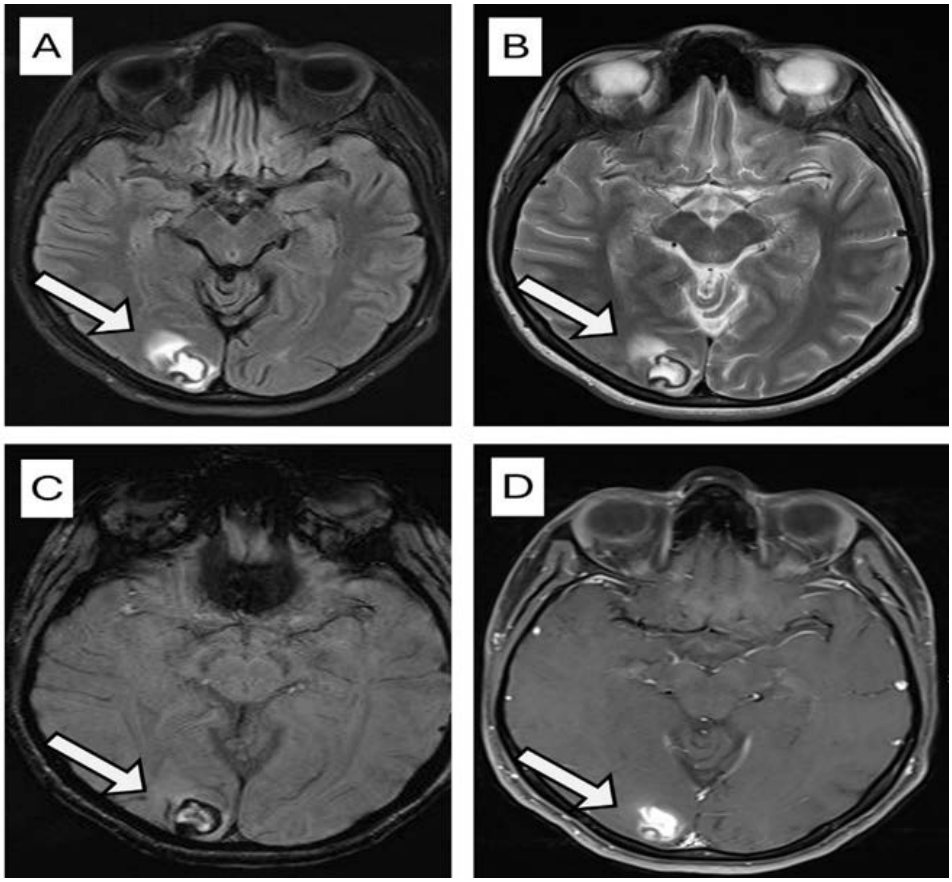

Figure 4: Hemorrhagic brain metastasis in the right occipital lobe on MRI

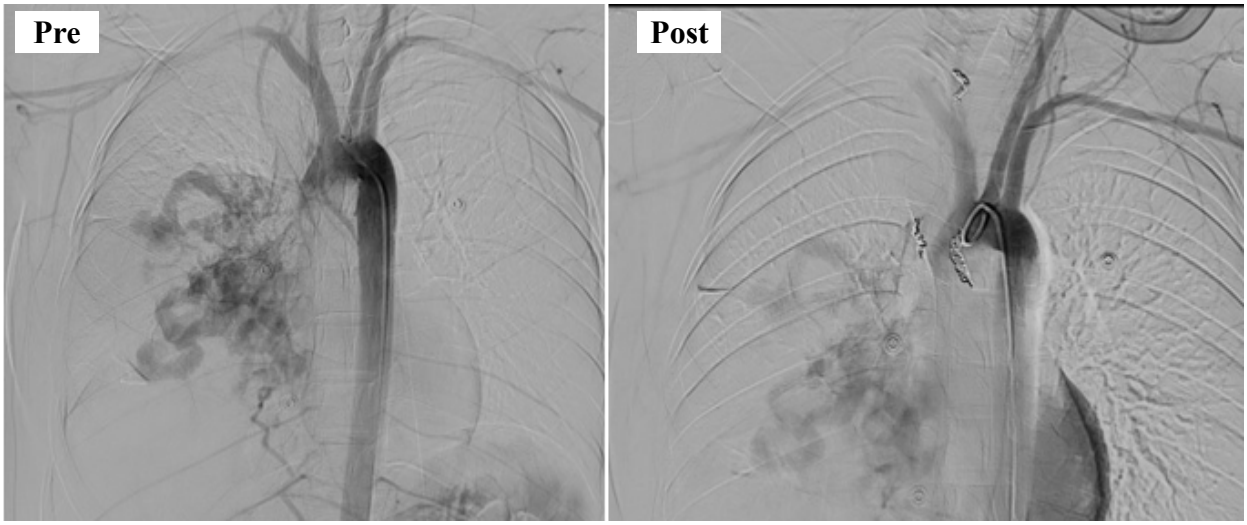

Figure 5: Pre- and post-embolization arteriograms demonstrating complete occlusion of the embolized arteries. Only the right phrenic artery was not embolized due to difficult access. *TECHNIQUE*: Anterior Posterior (AP) view of digital subtraction angiography (DSA), radiation does: 532 mGy, 450 cc contrast (Visipaque).

KEYWORDS

choriocarcinoma, gestational trophoblastic neoplasia, hemoptysis, arteriovenous malformation, metastatic storm.

ABBREVIATIONS

β-hCG = Beta-Human Chorionic Gonadotropin
CT = Computed Tomography
DSA = Digital Subtraction Angiography
EMA-CO = Etoposide, Methotrexate, Actinomycin-D, Cyclophosphamide, And Vincristine
FLAIR = Fluid-Attenuated Inversion Recovery
GTN = Gestational Trophoblastic Neoplasia
MRI = Magnetic Resonance Imaging
MIP = Maximum Intensity Projection
SWI = Susceptibility Weighted Imaging
TAE = Transcatheter Arterial Embolization
FIGO = International Federation of Gynecology and Obstetrics
AVM = Arteriovenous Malformation

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