Case Report
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Invasive hydatiform mole: a case report and review of the literature

Abstract

Background: Invasive mole is a subtype of gestational trophoblastic disease (GTD) that usually develops from malignant transformation of trophoblastic tissue after evacuation of the mole. Invasive mole occurs mainly in women of childbearing age, whereas it is extremely rare in postmenopausal women.

Observation: This is a 42-year-old cholecystectomized G3P2 patient who was referred for suspected GTD and whose pelvic echo and abdominal-pelvic CT scan revealed a hypervascularized image filling the uterine cavity, invading the myometrium to the serosa without invasion. The patient underwent total interannexal hysterectomy and chemotherapy with satisfactory clinical and biological follow up.

Conclusion: Our case highlights the difficulties of diagnosing invasive moles in the absence of a significant history of gestational trophoblastic disease. This study reviews the diagnostic methods, histological features and therapeutic recommendations.

Keywords: abnormal uterine bleeding, HCG, doppler, invasive mole, hysterectomy, methotrexate, follow up

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Abbreviations: GTTs, gestational trophoblastic tumors; GTD, gestational trophoblastic disease; BHCG, biological kinetics of the human chorionic hormone

Introduction

Invasive mole refers to a common manifestation of gestational trophoblastic tumors (GTTs) that complicate hydatidiform moles. invasive moles can invade the myometrium and cause massive, life-threatening bleeding. Previous studies have shown that the main distant metastatic site of invasive moles is the lungs, and in 5% of cases the vagina, pelvis, liver and brain.[1–3] Although invasive mole is sensitive to chemotherapy and highly curable,[4] late management is associated with serious complications, such as uterine perforation and hemoperitoneum. In this article, we report the case of a patient with invasive mole who benefited from optimal medical and surgical management.

Observation

Patient aged 42 years, cholecystectomy, G3P2: 2 children delivered by vaginal route, she reports a spontaneous miscarriage 2 months before having consulted for persistent bleeding which was related to a retention image where she benefited from an aspiration without anatomopathological study. The evolution was marked by an amenorrhea of two months before the reappearance of the bleedings motivating her consultation of which she benefited from a biological kinetics of the human chorionic hormone (BHCG) to 10 days of interval returned respectively to 64000 then 65000 from where its reference in our center for suspicion of TTG. The examination on admission found: a conscious patient, normo colored conjunctiva, hemodynamically and respiratory stable, apyretic. Abdominal examination: soft abdomen, normal breathing, no sensitivity to palpation, the gynecological examination reveals a macroscopically normal cervix, blackish bleeding from the endocervix with a soft, enlarged uterus. A BHCG assay was performed and came back to 72573, pelvic ultrasound revealed a heterogeneous hypervascularized echogenic image filling the entire uterine cavity from the isthmus to the uterine fundus, invading the myometrium up to the serosa with a doubt on a serosa effusion in some places, the 2 ovaries seen without abnormalities, no effusion (Figure 1).

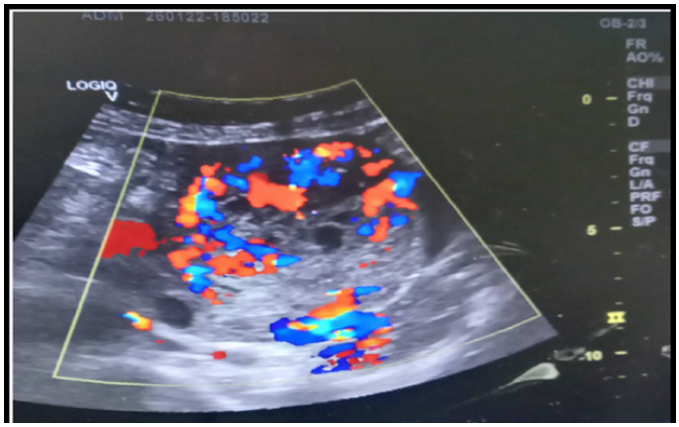

Figure 1 Ultrasound appearance of the highly vascularized invasive mole.

In view of the clinical, biological and echographic picture, we suspected an invasive mole, although there was no anatomopathological study of the product of conception expelled or issued during aspiration.

The patient underwent a thoraco-abdomino-pelvic CT scan which revealed a globular, enlarged uterus measuring 12.5 x 6cm in diameter, with heterogeneous, hypervascular intra-cavity material measuring 40 x 64mm, invading the myometrium at the fundial, anterior corporal and right lateral levels with focal invasion of the serosa at the right lateral corporal level. Both ovaries were of normal size, with a cystic lesion on the right measuring 3 cm in length, peri-uterine varices, permeable.

The patient underwent a total hysterectomy with bilateral salpingectomy, the anatomopathological study coming back in favor of chorionic villi of variable size, with a hydropic axis, bordered by a proliferating trophoblast. Cytotrophoblastic cells have an abundant eosinophilic cytoplasm and an atypical nucleus with heterogeneous

chromatin. Presence of mitoses. These cells are accompanied by multinucleated cells of syncytiotrophoblastic type with minimal atypia. Presence of large foci of necrosis. These villi infiltrate 2/3 of the myometrium, histological aspect of an invasive mole. On the other hand, the uterine cervix, the anterior and posterior vaginal collars, the right and left fallopian tubes, the parametrium and the paracervix are healthy.

Fortunately, the extension study did not reveal any distant metastases. After discussion in a multidisciplinary consultation meeting, the patient received adjuvant methotrexate-based monochemotherapy at a rate of 1mg/kg, with rigorous clinical and biological monitoring until negativation, and then maintenance of both clinical and biological monitoring. The evolution was marked by the rapid negativation of the BHCG level thanks to the hysterectomy, she presented some benign clinico-biological manifestations related to the chemotherapy resolved, but the evolution is satisfactory with a 12 months follow-up.

Discussion

Hydatidiform mole is a benign tumor with malignant potential. Malignant progression occurs in 15% of complete moles, and in 1.5% of partial moles.[5] The risk of progression to invasion is greater in multiparous patients than in those with a history of spontaneous abortion or previous mole.[6]

Gestational trophoblastic neoplasia (GTN) arises when the regulatory mechanisms of trophoblastic proliferation and invasion control are lost. Originating in placental tissue, they are characterized by a distinct tumor marker (beta-HCG), and have variable tendencies towards local invasion and distant metastasis.[7]

Invasive hydatidiform mole is a form of NTG that results from abnormal proliferation of the placental trophoblast. It most often occurs after molar evacuation, and is characterized by the presence of chorionic villus edema with trophoblastic proliferation that invades the myometrium or adjacent structures such as the vagina, vulva, broad ligament and may also invade the uterine vessels. It is important to distinguish between choriocarcinoma and invasive mole, as the latter has a more favorable prognosis.[8] Invasive mole usually occurs in women of childbearing age and is extremely rare in perimenopausal women. Only 5 cases of hydatidiform mole in postmenopausal women have been reported in the literature since 2004.[9]

The classic clinical presentation of NTG has decreased due to early diagnosis in the first trimester screening. However, the risk of developing a postmolar NTG remains unchanged.[10] Spontaneous presentation o f an invasive mole is extremely rare. It is preceded b y a hydatidiform mole in about 95% of cases with an interval of less than 6 months between presentation and diagnosis of invasive mole.

In our case, progression to invasive mole occurred within 3 months of evacuation. Follow-up of these patients is essential for the early detection of malignant trophoblastic tumors and to reduce the mortality rate.[11] The usual clinical manifestation of invasive mole includes vaginal bleeding, uterine enlargement and an elevated serum beta-HCG level.

The most common locations of invasive mole metastases are the vagina, lung, and brain, due to invasion of mole tissue into the venous system. Other sites of metastasis, including the epidural space and bladder, have been reported rarely.[12]

Seckl et al, report that a locally invasive NTG develops in 15% of patients and a metastatic form in 4% of patients after evacuation of a complete mole, and rarely after a partial mole.[13] In our case the histological evidence of the first evacuation was not available, and the diagnosis of invasive mole was only made on the hysterectomy specimen.

Ultrasound is the gold standard for the diagnosis of molar invasion. On ultrasound, invasive hydatidiform mole, implantation site tumors and choriocarcinomas usually present as a heterogeneous, hyperechoic solid mass with cystic vascular spaces, located in the myometrium. Color Doppler helps to evaluate the angiogenesis and neovascularization characteristic of these tumors, represented by large multidirectional blood flows suggestive of arterial and venous flows.[14] The FIGO Gynecologic Oncology Committeé has made recommendations for screening for metastatic forms of NTGs, which includes chest radiography, abdominal CT or body-scan in case of pulmonary localization, and brain MRI (or CT) in case of suspected brain metastases. Pathological diagnosis of invasive mole is rarely made, as most cases are treated conservatively, without the need for hysterectomy. In addition, biopsy of metastases is not recommended because of the risk of bleeding (Figure 2).

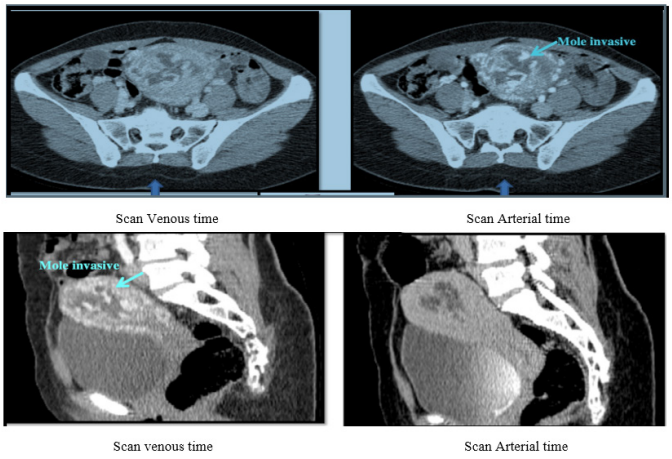

Figure 2 Scan images of invasive mole.

After the introduction of chemotherapy, the cure rates of invasive moles increased significantly. Because of their remarkable chemosensitivity, cure rates are nearly 100% in the low-risk group and nearly 80% in the high-risk group with current multidrug therapy protocols. Medical treatment is the best option and even histological evidence is not mandatory for initiation of chemotherapy because diagnosis is confirmed by diagnostic imaging and serum BHCG assay.

Alternatively, hysterectomy may be necessary in cases of uncontrolled vaginal or intra-abdominal bleeding (another option is uterine vessel embolization, but this is only considered if the patient is hemodynamically stable), resistance to chemotherapy, or neoplastic gestational disease. Surgical treatment could be a valid first-line treatment mainly in women who do not wish to preserve their fertility, but it does not prevent the development of metastases. On the other hand, hysterectomy could considerably reduce the trophoblastic tumor load and the number of cycles of chemotherapy, thus reducing their toxicity,[12] as was the case in our patient who benefited from surgical treatment given the risk of hemorrhage without the presence of distant metastases with a one-year follow-up. Recently, selective arterial embolization (SEA) has proven to be a safe and highly effective alternative or pre-surgical procedure to manage massive bleeding This technique offers several advantages, including avoidance of major surgical procedures, heavy general anesthesia, fertility preservation, and surgical comfort when hysterectomy is indicated.[15]

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Conclusion

We wish to add to the literature regarding the early diagnosis of invasive mole and its complications, to report unusual clinical presentations of invasive mole, and to outline the available treatment options. Although the first line of treatment is chemotherapy, we must consider the option of selective arterial embolization, and the surgical option in patients who present with uncontrollable bleeding.

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Conflicts of interests

The authors declare that there are no conflicts of interest.

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