

Case report

A rare case of a gestational and metastatic choriocarcinoma



Abderrahim Siati^{1,&}, Imane Benmouna¹, Anas Nah¹, Amina Lakhdar¹, Abdelaziz Baidada¹, Aicha Kharbach¹

¹Gynecology and Obstetrics Department, Maternity Souissi, University Hospital Center Ibn Sina, University Mohammed V, Rabat, Morocco

[&]Corresponding author: Abderrahim Siati, Gynecology and Obstetrics Department, Maternity Souissi, University Hospital Center Ibn Sina, University Mohammed V, Rabat, Morocco

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Abstract

Gestational choriocarcinoma is a malignant and rare trophoblastic tumor. It belongs to the group of gestational trophoblastic diseases (GTD) whose common denominator is hypersecretion of the gonadotrophic chorionic hormone. It is a highly malignant tumor made from the juxtaposition of cytotrophoblast and syncytiotrophoblast cells with complete disappearance of chorionic villi. It is most often metastatic and is followed in most cases by a hydatiform mole, but may also occur after spontaneous abortion or normal pregnancy. We present a case of a 50-year-old Moroccan patient, G2P2, admitted to the gynecological emergency department for vaginal bleeding and dyspnea. Speculum examination showed an invasive vaginal lesion. Thoracic-abdominal-pelvic CT showed bilateral pulmonary nodules and cystic lesions in the uterus and vagina. Through a vaginal biopsy, the diagnosis of choriocarcinoma was confirmed. Taking into account the usually very chemo-sensitive character of these diseases, EMA-CO type chemotherapy (Etoposide, Methotrexate and Actinomycin D alternating at weekly intervals with Vincristine and Cyclophosphamide) has been introduced. The patient received 8 cycles of EMA-CO including 2 cycles after normalization of beta - Human chorionic gonadotropin (β -hCG). The evolution was very favorable and regular and control was scheduled.

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Introduction

Gestational choriocarcinoma is a malignant and rare trophoblastic tumor [1,2]. It belongs to the group of gestational trophoblastic diseases whose common denominator is hypersecretion of the gonadotrophic chorionic hormone (hCG - human chorionic gonadotrophin) [3]. Its frequency in Europe and North America is estimated at 0.2-0.7 per 1000 pregnancies [1]. It occurs as a result of abnormal conception (hydatiform moles) or normal (pregnancies). Thus, half of the gestational choriocarcinomas appear after a hydatiform mole, 25% after abortion and 22% occur after normal pregnancy [2]. In some exceptional cases, choriocarcinoma can occur outside a gestational state. It is characterized by its strong metastatic potentiality and chemosensitivity. We present here a rare case of a gestational and metastatic choriocarcinoma with a good evolution under chemotherapy.

Patient and observation

A case of 50-year-old Moroccan woman, G2P2 (2 alive children), was presented to our emergency department for vaginal bleeding. The principal complaint on arrival was lower abdominal pain and dyspnea. According to the patient, there was an unencrypted weight loss and genital bleeding for the last 7 months. On physical examination, she had a heart rate of 110 beats/min, her blood pressure was 125/75mmHg, a respiratory rate of 19 breaths/min and temperature of 37.4 °C. Speculum examination showed a budding and infiltrating lesion in relation to the anterior vaginal wall (Figure 1). Pelvic ultrasound showed an enlarged uterus containing heterogeneous formation with cystic lesions (Figure 2). The plasma rate level of β -hCG (human chorionic gonadotrophin) was 470.000 IU. The thoracic-abdominal-pelvic CT was objectified bilateral pulmonary nodules and cystic lesions in

the uterus and vagina (Figure 3). Finally, the biopsy of the vaginal lesion was performed and the pathological report was confirmed the diagnosis of choriocarcinoma. Taking into account the usually very chemo-sensitive character of these diseases, EMA-CO type chemotherapy (Etoposide, Methotrexate and Actinomycin D alternating at weekly intervals with Vincristine and Cyclophosphamide) has been introduced. The patient received 8 cycles of EMA-CO including 2 cycles after normalization of β -hCG. The evolution was very favorable and regular controle was scheduled.

Discussion

Gestational choriocarcinoma is a malignant tumor of the villous trophoblast, free of placental villi and molar vesicle [1,4]. The frequency of choriocarcinoma is high in Asia, Africa and Latin America, ranging from 23 to 335 per 100,000 pregnancies, but rare in North America, Europe and Australia with rates ranging from two to seven per 100,000 pregnancies [5,6]. The average age of onset of choriocarcinoma is between 25 and 39 years [6,7]. Some authors suggest that the frequency increases beyond 39 years [6]. Metrorrhagia is the most common sign of call according to most authors [8,9]. In contrast, some choriocarcinomas that have already invaded the myometrium may be asymptomatic [9,10]. There are also cases where metastases, particularly vaginal, pulmonary and cerebral, inaugurate the clinical picture [11,12]. In our study, all patients had metrorrhagia. Gestational choriocarcinoma can complicate any pregnancy status. It can occur after a hydatiform mole in 50% of cases, miscarriage (25%) or normal pregnancy (25%) [6,13,14]. The diagnosis of choriocarcinoma is biological and relies to a large extent on the hormonal assay of plasma β -hCG [1,12]. Pelvic ultrasound often shows an uneven, heterogeneous image, with hyperdense zones and an attack of several tunics without respecting their

architecture [11]. Choriocarcinoma is a richly vascularized tumor; pulsed or color doppler demonstrates hypervascularization of the lesion [5]. Computed tomography is essential for the extension assessment to search for distant metastases [1]. Pelvic Magnetic Resonance Imaging (MRI) is not a first-line test; it keeps its place in the study of the penetration of the myometrium and the attack of the pelvic structures. Pelvic MRI may be useful in patients who had contraindication for the injection of iodinated contrast media. It may also be useful for assessing the tumor response to chemotherapy [15]. Currently, the role of positron emission tomography (PET) scans in gestational trophoblastic disease has not been established [15]. Macroscopically, choriocarcinoma presents itself as a nodular tumor, necrotic, well circumscribed and very haemorrhagic, significantly affecting the myometrium, measuring a few millimeters and can completely fill the uterine cavity [5, 6]. Histologically, it is composed of dimorphic cells, cyto and syncytiotrophoblastic, invading tissues, including vessels [6]. The tumor cells may be difficult to find in the hematoma always very extensive. The existence of a mature trophoblast is a factor of good prognosis [4,6].

However, histological examination is not required to make the diagnosis: choriocarcinoma is a tumor with a biological definition. Confirmation of choriocarcinoma can be histological. It can be obtained by aspirating curettage, echo-guided biopsy or hysteroscopy, or even on a hysterectomy [6]. Histological confirmation is not required to start treatment [1]. Choriocarcinoma is a highly metastatic tumor; a complete extension assessment is essential for the therapeutic decision. This lesional assessment is initially clinical, in particular the examination of the vagina with the speculum in search of metastases vaginal [1,6], then paraclinic looking for pulmonary metastases, which are the most frequent (80%). The cerebral and hepatic localizations are the most serious [4]. Chemotherapy has supplanted surgery and has become the treatment of choice for gestational trophoblastic tumors, with

the exception of the placental implantation site tumor, which is chemoresistant and where surgery remains [11,16]. Monochemotherapy is indicated in low-risk patients and multidrug therapy (MA-CO) is required for high-risk patients according to the FIGO-WHO 2000 classification [17]. The therapeutic efficacy is judged on the evolution of the rate of β -hCG plasma levels, falling by half after each treatment. The surgery keeps precise indications according to the evolutionary stage and the state of each patient. It is indicated in case of peritonitis, uncontrollable haemorrhage or chemoresistance [14,16]. Radiation therapy is indicated for the treatment of choriocarcinoma or its metastases; it is proposed as an adjunct to chemotherapy. Post-treatment surveillance should be both clinical and paraclinical based essentially on the regular determination of β -hCG [1,16]. During surveillance, effective contraception should be prescribed for at least one year [1]. Any subsequent pregnancy should be considered at risk of developing a gestational trophoblastic tumor and therefore requires ultrasound as soon as possible. After delivery, the placenta should be sent to the pathologist and the β -hCG should be dosed at six weeks postpartum. Healing is defined by normalization of β -hCG levels for three consecutive weeks. The existence of late recurrences warrants prolonged surveillance.

Conclusion

A good knowledge of this group of trophoblastic tumors makes it possible to propose more coherent and adequate therapeutic protocols. Early management of gestational trophoblastic tumors allows the detection of low-risk forms, and thus has high efficacy in chemotherapy and resistance. Chemotherapy has radically changed the prognosis of these cancers. The relative rarity of the condition and the complexity of the therapeutic protocols make it necessary to create

reference centers for the establishment of registers and for the consensual treatment of these tumors.

Competing interests

The authors declare no competing interests.

Authors' contributions

All the authors have read and agreed to the final manuscript.

Figures

Figure 1: representing a budding and infiltrating lesion on the anterior vaginal wall

Figure 2: pelvic ultrasound showed an enlarged uterus containing heterogeneous formation with cystic lesions

Figure 3: A) showing bilateral pulmonary nodules; B) representing cystic lesions in the uterus and vagina

References

1. Boufettal H, Coullin P, Mahdaoui S, Noun M, Hermas S, Samouh N *et al.* Les môles hydatiformes complètes au Maroc: étude épidémiologique et clinique (Complete hydatiforme mole in Morocco: epidemiological and clinical study). *Journal de Gynécologie, Obstétrique et Biologie de la Reproduction.* 2011;40(5):419-429. **Google Scholar**
2. Ramos AJ, Cantero MR, Zhang P, Raychowdhury MK, Green A, MacPhee D *et al.* Morphological and electrical properties of human trophoblast choriocarcinoma. *Placenta.* 2008 Jun;29(6):492-502. **PubMed | Google Scholar**
3. Cheung AN, Zhang HJ, Xue WC, Siu MK. Pathogenesis of choriocarcinoma: clinical, genetic and stem cell perspectives. *Future Oncology (London, England).* 2009 Mar;5(2):217-31. **PubMed | Google Scholar**
4. Behtash N, Behnamfar F, Hamed B, Ramezanzadeh F. Term delivery following successful treatment of choriocarcinoma with brain metastases, a case report and review of literature. *Archives of Gynecology and Obstetrics.* 2009 Apr;279(4):579-81. **PubMed | Google Scholar**
5. Dalenda Chelli, Dimassi K, Bouaziz M, Chiraz Ghaffari, Zouaouia B, Emiliano Sfara *et al.* Imagerie des maladies trophoblastiques gestationnelles (Imaging of gestational trophoblastic disease). *Journal de Gynécologie, Obstétrique et Biologie de la Reproduction.* 2008;37(6):559-567. **Google Scholar**
6. Cisse CT, Lo N, Moreau JC, Fall-Gaye C, Mendez V, Diadhiou F *et al.* Choriocarcinome au Sénégal: épidémiologie, pronostic et prévention (Choriocarcinoma in Senegal: epidemiology, prognosis and prevention). *Gynécologie, Obstétrique & Fertilité.* 2002;30(11):862-869. **Google Scholar**
7. Pisal N, North C, Tidy J, Hancock B. Role of hysterectomy in management of gestational trophoblastic disease. *Gynecologic Oncology.* 2002 Nov;87(2):190-2. **PubMed | Google Scholar**

8. Buckley JM. The epidemiology of molar pregnancy and choriocarcinoma. *Clinical Obstetrics and Gynecology*. 1984 Mar;27(1):153-9. **PubMed | Google Scholar**
9. Goldstein DP, Berkowitz RS. Current management of gestational trophoblastic neoplasia. *Hematology/Oncology Clinics of North America*. 2012 Feb;26(1):111-31. **PubMed | Google Scholar**
10. Bracken MB, Brinton LA, Hayashi K. Epidemiology of hydatidiform mole and choriocarcinoma. *Epidemiologic Reviews*. 1984;6:52-75. **PubMed | Google Scholar**
11. Altieri A, Franceschi S, Ferlay J, Smith J, La Vecchia C. Epidemiology and aetiology of gestational trophoblastic diseases. *Lancet Oncology*. 2003 Nov;4(11):670-8. **PubMed | Google Scholar**
12. Oranratanaphan S, Wongwathanavikrom R, Lertkachonsuk R. Correlation levels of serum placental growth factor and human chorionic gonadotropin in gestational trophoblastic disease patients. *Journal of Reproductive Medicine*. 2011 Jul-Aug;56(7-8):333-8. **PubMed | Google Scholar**
13. Berkowitz RS, Goldstein DP. Chorionic tumors. *New England Journal of Medicine*. 1996 Dec 5;335(23):1740-8. **PubMed | Google Scholar**
14. Lurain JR, Chapman-Davis E, Hoekstra AV, Schink JC. Actinomycin D for methotrexate-failed low-risk gestational trophoblastic neoplasia. *Journal of Reproductive Medicine*. 2012 Jul-Aug;57(7-8):283-7. **PubMed | Google Scholar**
15. DeBaz BP, Lewis TJ. Imaging of gestational trophoblastic disease. *Semin Oncol*. 1995 Apr;22(2):130-41. **PubMed | Google Scholar**
16. Turan T, Karacay O, Tulunay G, Boran N, Koc S, Bozok S *et al*. Results with EMA/CO (etoposide, methotrexate, actinomycin D, cyclophosphamide, vincristine) chemotherapy in gestational trophoblastic neoplasia. *International Journal of Gynecological Cancer*. 2006 May-Jun;16(3):1432-8. **PubMed | Google Scholar**
17. FIGO Oncology Committee. FIGO staging for gestational trophoblastic neoplasia 2000. *International Journal of Gynaecology and Obstetrics: the Official Organ of the International Federation of Gynaecology and Obstetrics*. 2002 Jun;77(3):285-7. **PubMed | Google Scholar**



Figure 1: representing a budding and infiltrating lesion on the anterior vaginal wall



Figure 2: pelvic ultrasound showed an enlarged uterus containing heterogeneous formation with cystic lesions

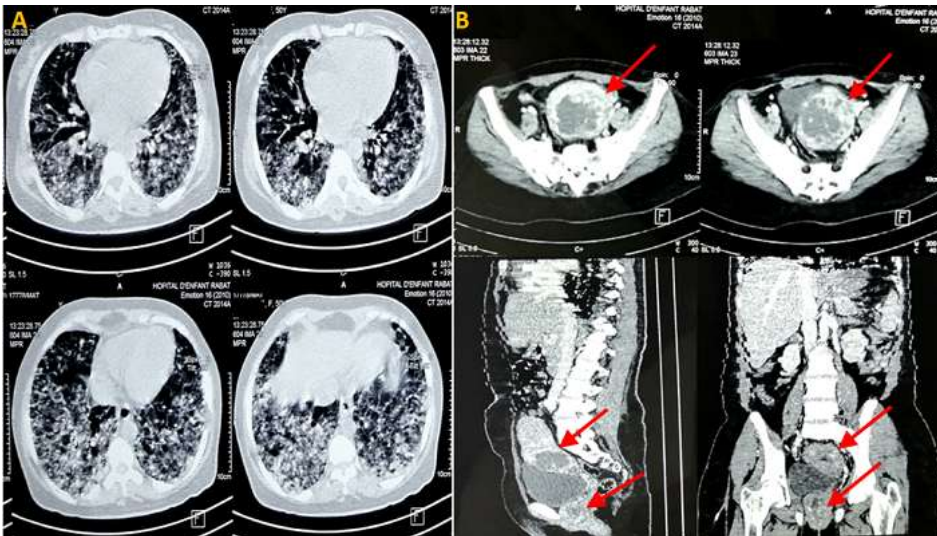


Figure 3: A) showing bilateral pulmonary nodules; B) representing cystic lesions in the uterus and vagina