

Case Report

Gestational Choriocarcinoma of the Uterus with Concurrent Metastasis and an Atraumatic Rupture of the Spleen: A Rare Association and First Description in Africa

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Abstract

A gestational choriocarcinoma of the uterus with concurrent metastasis and an atraumatic rupture of the spleen is rare disease and has been rarely reported in African women. We report here the case of a thirty-eight-year-old women who was referred to our hospital with a previous medical history of hydatidiform mole. Ultrasonography (US) revealed a non-pregnant uterus of normal size and a left adnexal pseudo-occupation of oval shape hypoechoic with a heterogeneous structure. Laparotomy revealed a hemoperitoneum, splenic rupture and the presence of a fleshy mass in the left iliac fossa. Histopathology of uterus revealed choriocarcinoma and those of the spleen and the fleshy mass revealed metastatic choriocarcinoma. Her Human Chorionic Gonadotropin (HCG) level was elevated. Our patient presented gestational choriocarcinoma of the uterus with concurrent metastasis and an atraumatic rupture of the spleen. This is a rare complication described in woman living in sub-Saharan Africa. Genetic and molecular investigations play an important role in diagnosis but were not available in poor resources settings in developing countries such as the Democratic Republic of Congo.

Keywords: Choriocarcinoma; Woman; Hydatidiform mole; Human chorionic gonadotropin; Metastasis; Lubumbashi; Democratic republic of congo; Africa

Introduction

Choriocarcinoma is a highly malignant tumor of trophoblastic cells characterized by a proliferation of cytotrophoblasts and syncytiotrophoblasts and extensive angio-invasion and by a propensity to metastasize to various sites including lungs, vagina, brain, liver, kidney, and gastrointestinal tract, in descending order of frequency [1]. Gestational choriocarcinoma is a rare complication of pregnancy. Gestational choriocarcinomas have been observed in the ovaries but rarely the uterus [2]. Gestational choriocarcinoma of the uterus with concurrent metastasis and an atraumatic rupture of the spleen have been rarely reported in the medical literature [3]. This paper reports a case of a gestational choriocarcinoma of the uterus associated with an atraumatic rupture of the spleen in a 38-year-old woman who presented this malignant tumor diagnosed in the University Hospital of Lubumbashi, Democratic Republic of Congo.

Case Presentation

A thirty-eight-year-old gravida 4 para 5 women, was admitted on the eight weeks post-menses for diffuse abdominal pain for 24 hours. Her previous medical history revealed a hydatidiform mole (when she was 37 years old) and follow-up was marked by the negativity of the beta-hCG assay after curettage associated with administration of methotrexate (100 mg/week for 3 weeks). During the initial exam, the patient reported physical asthenia. The abdominal palpation revealed a diffuse high sensitivity and a sloping dullness at the percussion. Per vaginal examination was normal. Rapid immunological pregnancy test was positive. With these findings, she was suspected to have an ectopic pregnancy. Ultrasonography (US) revealed a non-pregnant uterus of normal size (56 mm × 47 mm) with a thickened endometrium, a left adnexal pseudo-occupation of oval shape hypoechoic with a heterogeneous structure (35 mm × 44 mm). An effusion of average abundance that surrounds the uterus and upper abdomen with normal appearance was noted. An urgently exploratory laparotomy was conducted and revealed a hemoperitoneum, a normal uterus and both adnexa were normal (Figure 1). Furthermore, splenic rupture and the presence of a fleshy mass in the left iliac fossa were noted (Figure 2). An endometrial biopsy and splenectomy were performed. Histopathology of uterus revealed choriocarcinoma and those of the spleen and the fleshy mass revealed metastatic choriocarcinoma. Histological data of the spleen show a spleen whose white pulp is hyperplastic with tumor invasion made of undoubtedly malignant trophoblastic tissue without placental villi. Chest X-ray was normal. Her Human Chorionic Gonadotropin (HCG) level was elevated. Weekly biological monitoring by the beta-HCG urine assay was initiated and showed a perfect regression curve as soon as treatment

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was initiated. A semi-quantitative method previously described by Nsambi et al. [4] (Figure 3) have been used. The patient was given 7 cycles postoperative chemotherapy with methotrexate (100 mg/week) and folic acid (5 mg/day). This treatment was administered for 7 weeks and used the American methotrexate protocol which suggests giving methotrexate once a week. Methotrexate has been used because it was the only antimetabolic available on the market in Lubumbashi. She was on weekly follow-up with HCG titer, which gradually fell. After improving her condition, the patient was lost to follow-up and abandoned the treatment. After 2 months, she presented back to us with a coma and was admitted in the Intensive care unit. Death occurred 24 hours after her admission and was due to multiple organ failure.

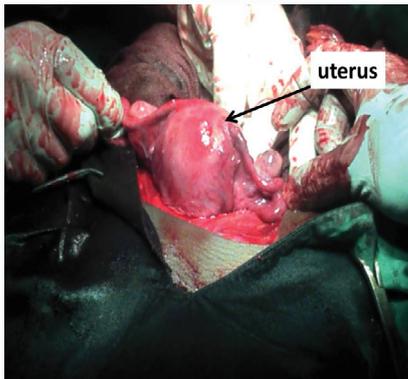


Figure 1: Exploratory laparotomy was conducted and revealed a hemoperitoneum, a normal uterus and both adnexa were normal.

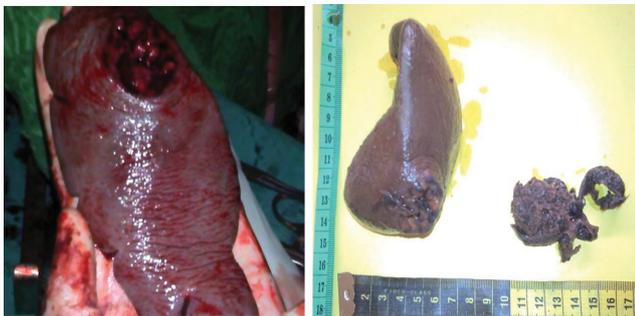


Figure 2: Splenic rupture and the presence of a fleshy mass in the left iliac fossa were noted.



Figure 3: HCG Urinary Assay.

Discussion

Choriocarcinoma is a malignant gestational trophoblastic disease which it usually develops in the aftermath of a pregnancy. The

medical literature showed that this choriocarcinoma occurred after a hydatidiform mole (50% of cases), abortion (25% of cases), normal pregnancy (22.5% of cases) or rarely after ectopic pregnancy (2.5% of cases) [5,6]. The incidence of hydatidiform mole varies worldwide from 1 in Europe and North America, to 3 in every 1500 gestations in the Far East and Eastern South of Asia [7,8]. In sub-Saharan Africa, this choriocarcinoma with concurrent metastasis in the spleen was not previously noted and this observation is the first description of a gestational choriocarcinoma of the uterus with concurrent metastasis and an atraumatic rupture of the spleen in African population. It is the first description of choriocarcinoma with concurrent metastasis in the spleen in Sub-Saharan countries after a previous medical history of hydatidiform mole. The pathogenesis of choriocarcinoma in uterus may be explained by several hypotheses such as the transport of chorionic cells from a previous pregnancy that undergo malignant transformations after a dormant period [3]. In this case, the age of the patients close to 40 years, the woman had medical history of molar abortion with preservation of the uterus, these situations were found to be risk factors of choriocarcinoma [9]. The diagnosis of choriocarcinoma is based on the curettage specimen. In the present case, histopathological analysis was compatible with choriocarcinoma as described in the literature, despite that the diagnosis was made in context of low resources, especially echographic associated with a proportioning of plasmatic β -hCG or prolans urinary. Immunohistochemistry analysis is useful in differential diagnosis of choriocarcinoma. Strong diffuse β -hCG immunoreactivity confirms the diagnosis of choriocarcinoma [10]. Metastasis of choriocarcinoma usually occur through blood, preferably reaching the lung (80% of cases), vagina (30% of cases), pelvis (20% of cases) [1]. Splenic localization is rare. To the best of our knowledge, only five cases were found [6,11-14]. Splenic rupture due to choriocarcinoma of the spleen is also rarely described in the medical literature. Less than 10 cases could be found with the use of available computer-assisted medical literature search programs [6,11-14]. The treatment of choriocarcinoma is surgical excision, if possible, followed by polychemotherapy (various chemotherapeutic agents, for example, methotrexate, cisplatin, mitomycin C, fluorouracil, futrafur) [15]. Recently, methotrexate has subsequently revolutionized the treatment of gestational trophoblastic neoplasia and ectopic pregnancy [16]. This drug was favored for the treatment in this case. In the present case, chemotherapy was not given regularly according to established standard treatment guidelines. The main reasons for this are resource deficiencies, the unavailability and high cost of some chemotherapeutic agents in Democratic Republic of Congo. This situation contributes to lack of treatment adherence and patients being lost to follow-up in sub-Saharan African countries [17].

Conclusion

A gestational choriocarcinoma of the uterus with concurrent metastasis and an atraumatic rupture of the spleen is rarely described in sub-Saharan countries. An implementation of molecular and genetic laboratories is advocacy in tertiary institutions of the Democratic Republic of Congo to elucidate cases of gestational choriocarcinomas for a contribution to knowledge and progress of science in Tropical environment.

References

1. Stevens FT, Katzorke N, Tempfer C, Kreimer U, Bizjak GI, Fleisch MC, et al. Gestational Trophoblastic Disorders: An Update in 2015. *Geburtshilfe Frauenheilkd.* 2015;75(10):1043-50.
2. Mittal S, Aird I, Haugk B. Gestational choriocarcinoma in liver mimicking ruptured ectopic pregnancy. *J Obstet Gynaecol.* 2012;32(5):499.

3. Seckl MJ, Sebire NJ, Berkowitz RS. Gestational trophoblastic disease. *The Lancet*. 2010;376(9742):717-29.
4. Nsambi JB, Mukuku O, Kinenkinda XK, Kakoma JB. Gestational Trophoblastic Diseases: Alternatives to Biological Monitoring in Under-resourced Settings (two clinical observations). *Afr J Health Issues*. 2017;1(1):4.
5. Ozaki Y, Shindoh N, Sumi Y, Kubota T, Katayama H. Choriocarcinoma of the ovary associated with mucinous cystadenoma. *Radiat Med*. 2001;19(1):55-9.
6. Ramarajapalli ML, Rao NAR, Murudaraju P, Kilara NG. Ovarian Choriocarcinoma with Concurrent Metastases to the Spleen and Adrenal Glands: First Case Report. *J Gynecol Surg*. 2012;28(2):153-5.
7. Ghaemmaghami F, KarimiZarchi M. Early onset of metastatic gestational trophoblastic disease after full-term pregnancy. *Int J Biomed Sci*. 2008;4(1):74-7.
8. Oranratanaphan S, Lertkhachonsuk R. Treatment of extremely high risk and resistant gestational trophoblastic neoplasia patients in King Chulalongkorn Memorial Hospital. *Asian Pac J Cancer Prev*. 2014;15(2):925-8.
9. Cisse CT, Lo N, Moreau JC, Fall-Gaye C, Mendez V, Diadhiou F. Choriocarcinoma in Senegal: epidemiology, prognosis and prevention. *Gynecol Obstet Fertil*. 2002;30(11):862-9.
10. Tempfer C, Horn LC, Ackermann S, Beckmann MW, Dittrich R, Einkenkel J, et al. Gestational and Non-gestational Trophoblastic Disease. Guideline of the DGGG, OEGGG and SGGG (S2k Level, AWMF Registry No.032/049, December 2015. *Geburtshilfe Frauenheilkd*. 2016;76(2):134-44.
11. Challis DE, Rew KJ, Steigrad SJ. Choriocarcinoma complicated by splenic rupture: an unusual presentation. *J Obstet Gynaecol Res*. 1996;22(4):395-400.
12. Kristoffersson A, Emdin S, Jarhult J. Acute intestinal obstruction and splenic hemorrhage due to metastatic choriocarcinoma. A case report. *Acta Chir Scand*. 1985;151(4):381-4.
13. Carr AJ, Jacob G, Glanfield PA, Rogers K. Male choriocarcinoma of the spleen: a case report. *Eur J Surg Oncol*. 1987;13(1):75-6.
14. Ghinescu C, Sallami Z, Jackson D. Choriocarcinoma of the spleen--a rare cause of atraumatic rupture. *Ann R Coll Surg Engl*. 2008;90(3):W12-4.
15. May T, Goldstein DP, Berkowitz RS. Current chemotherapeutic management of patients with gestational trophoblastic neoplasia. *Chemother Res Pract*. 2011;2011:806256.
16. Skubisz MM, Tong S. The evolution of methotrexate as a treatment for ectopic pregnancy and gestational trophoblastic neoplasia: a review. *ISRN Obstet Gynecol*. 2012; 2012:637094.
17. Dim CC, Ezegwui HU. Choriocarcinoma in Enugu, South east Nigeria: a need for a shift from mortality to survival. *Niger J Med*. 2013; 22(3):252-6.