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Unusual gestational choriocarcinoma arising in an interstitial pregnancy

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ABSTRACT

INTRODUCTION: Choriocarcinoma is a highly malignant trophoblastic neoplasm. Its association with ectopic pregnancy is very rare and usually with aggressive behavior.

PRESENTATION OF CASE: We report a new case arising in an interstitial pregnancy occurring in a 46-year-old woman. The patient was admitted for severe pelvic pain and abundant metrorrhagia. One month ago, she had had a laparoscopic resection of an interstitial pregnancy subsequent to failure of chemotherapy by methotrexate. The raise of serum β hCG level and the hyperechoic intrauterine mass were in favor of gestational trophoblastic disease. Urgent laparotomy was performed for circulatory collapse. Hysterectomy was done. Histological examination revealed a choriocarcinoma. The patient underwent chemotherapy. Two years later, neither metastasis nor recurrence was detected.

DISCUSSION: Clinical diagnosis of primary interstitial choriocarcinoma is difficult, since it is rare and manifesting by non-specific abnormal vaginal bleeding. Imaging findings are also not helpful in ectopic location. The frequency of metastasis is related to the delayed diagnosis. Serial measurement of β hCG level was the most useful marker of diagnosis and follow up. Histopathological examination remains the only tool of the precise diagnosis. Choriocarcinoma has a very good prognosis even in advanced stages, since it is very chemosensitive.

CONCLUSION: The current trend of the treatment of ectopic pregnancy by conservative surgery requires adequate monitoring of β hCG and careful examination of pathologic specimens to avoid misdiagnosis of ectopic gestational trophoblastic disease.

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1. Introduction

Choriocarcinoma is a rare malignant gestational trophoblastic tumor. Although it is most commonly due to malignant transformation of a molar pregnancy, it can develop after all types of pregnancy including normal term pregnancy, spontaneous abortion or ectopic pregnancy.¹ However, choriocarcinoma arising in ectopic pregnancy is an extremely rare event. Its incidence is one in 5333 tubal pregnancies and one in 1.6 million normal intrauterine pregnancies.¹ To our knowledge, two cases of choriocarcinoma arising in an interstitial pregnancy were reported in the literature.^{2,3} We report here a new case.

2. Case report

A 46-year-old woman, gravida 4, para 3, was admitted for abdominal pain. She had had an amenorrhea of 7 weeks. Physical examination revealed severe right adnexal pain and tenderness. Transvaginal sonography showed an empty uterus with a thin endometrium and a 3 cm intramural echogenic mass on the left side of the fundus. There was neither fluid in the Douglas pouch, nor adnexal masses nor “ring of fire” signs. Human chorionic gonadotropin (β hCG) was at 6320 mUI/ml. These findings were consistent with ectopic interstitial pregnancy. According to the department’s protocol, the patient received a single dose of methotrexate.⁴ β hCG decreased to 3010 mUI/ml after one week, then raised to 7020 mUI/ml two weeks later. Therefore, the patient underwent a laparoscopy. A 3 cm mass located on the left side of the uterine fundus was found. The fallopian tubes and ovaries were normal. The mass with portion of myometrium and serosa were excised. Histological examination showed a hemorrhagic infiltration of the myometrium without trophoblastic cells. Thus,

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a monthly monitoring of plasmatic β hCG level was decided till its negativity. However, earlier to her first appointment, the patient presented in the emergency room with severe acute pelvic pain and vaginal bleeding. Ultra sonography revealed a hyper echoic intrauterine mass and β hCG titer was very high (4,000,000 mUI/ml) suggestive for a persistent trophoblastic disease. Promptly, our patient had unstable hemodynamic status, so she underwent emergency hysterectomy. Histological examination revealed a proliferation of atypical trophoblastic cells, both syncytiotrophoblastic and cytotrophoblastic cells with numerous mitoses. The stroma was hemorrhagic and the tumor cells infiltration is limited to the myometrial wall. There were no placental villi. Thus, the diagnosis of choriocarcinoma was proposed. Imaging investigation of the chest, abdomen and pelvis did not show any metastasis. The patient was treated with 3 cycles of monochemotherapy based on Methotrexate. The patient was followed by β hCG test weekly in the first month, then monthly during the first year. Two years later, she was free from recurrence and metastasis.

3. Discussion

Choriocarcinoma is a highly malignant tumor originating in trophoblastic tissue. Usually presents within a hydatidiform mole, it primarily occurs during the procreation period.¹ It is a biphasic malignant proliferation of cytotrophoblast and syncytiotrophoblast without chorionic villi. The age has consistently been identified as an important risk factor. Age-specific incidence reports usually reveal a “J curve” and younger <18 year old and women >40 have higher rates.⁵

Although the tumor may arise from any type of prior gestational events as normal pregnancy or spontaneous abortion, it more frequently complicates hydatidiform mole regardless of its intrauterine or ectopic location.⁶

The development of the tumor varies between 5 weeks and 15 years after gestation or even after menopause.^{6,7} Its association with ectopic pregnancy is extremely rare and in general with aggressive behavior. Indeed, only 0.76–4% of choriocarcinomas developed in ectopic location.^{8,9} Its specific occurrence in interstitial pregnancy is exceptional with, in our knowledge, three cases being reported in the literature including the present one.^{2,3}

The diagnosis of primary extra-uterine choriocarcinoma is challenging because the clinical symptoms are often nonspecific and can mimic hemorrhagic ovarian cyst, tubo-ovarian abscess, ovarian torsion, and ectopic pregnancy.¹⁰ The frequency of metastatic disease with resultant increased morbidity and mortality is related to the delayed diagnosis.

The typical sonographic appearance of intra-uterine choriocarcinoma is a large echogenic irregular mass occupying the uterine cavity, hyper vascularized in color-flow doppler. However, imaging findings are not helpful in extra-uterine location. Serial measurement of β hCG level was the most useful marker of diagnosis and follow up.¹¹

When nonmetastatic choriocarcinoma occurs in young patients who desire to preserve their fertility, it should be treated by local resection of the tumor followed by chemotherapy.³ At the new England trophoblastic disease center, all the women with tubal choriocarcinoma (6/6) achieved complete remission with chemotherapy even those with metastases (4/6).⁹ The two cases

of choriocarcinoma arising in an interstitial pregnancy reported were also successfully treated with chemotherapy based on methotrexate.^{2,3}

4. Conclusion

The increasing tendency to treat ectopic pregnancy by conservative management suggests the crucial role of pathological examination associated with β hCG level follow up, not only to diagnose persistent ectopic pregnancy but also to avoid missing malignant trophoblastic disease.

Conflict of interest

No author has any conflict of interest to declare.

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Ethical approval

Patient's consent was obtained.

Author contributions

Sawsen Meddeb made substantial contributions to conception, design, and acquisition of data. Mohamed Salah Rhim was involved in the analysis and interpretation of data and drafting the article. Wissal Zarrouk participated in drafting the article. Mohamed Bibi critically revised the manuscript. Mohamed Tahar Yacoubi and Hedi Khairi gave final approval of the version to be submitted.

References

- Rettenmaier MA, Khan HJ, Epstein HD, Nguyen D, Abaid LN, Goldstein BH. Gestational choriocarcinoma in the Fallopian tube. *J Obstet Gynaecol* 2013;**33**(8):912–4.
- Venturini PL, Gorlero F, Ferraiolo A, Valenzano M, Fulcheri E. Gestational choriocarcinoma arising in a cornual pregnancy. *Eur J Obstet Gynecol Reprod Biol* 2001;**96**(1):116–8.
- Rotas M, Khulpateea N, Binder D. Gestational choriocarcinoma arising from a cornual ectopic pregnancy: a case report and review of the literature. *Arch Gynecol Obstet* 2007;**276**(6):645–7.
- Comparison of single and multiple dose methotrexate therapy for unruptured tubal ectopic pregnancy: a prospective randomized study. *Acta Obstet Gynecol Scand* 2010;**89**(7):889–95.
- Smith HO, Kohorn E, Cole LA. Choriocarcinoma and gestational trophoblastic disease. *Obstet Gynecol Clin North Am* 2005;**32**(4):661–84.
- Hassadia A, Kew FM, Tidy JA, Wells M, Hancock BW. Ectopic gestational trophoblastic disease: a case series review. *J Reprod Med* 2012;**57**(7–8):297–300.
- Soper JT, Mutch DG, Schink JC. American College of Obstetricians and Gynecologists diagnosis and treatment of gestational trophoblastic disease: ACOG practice bulletin no. 53. *Gynecol Oncol* 2004;**93**(3):575–85.
- Chen MJ, Yang JH, Lin MC, Ho HN, Yang YS. An unusual gestational choriocarcinoma occurring primarily on the surface of a subserous leiomyoma. *BJOG* 2004;**111**(2):188–90.
- Salleh S, Arthur I. Persistent peritoneal trophoblastic implantation following salpingotomy, salpingectomy and methotrexate for ectopic pregnancy. *Gynecol Surg* 2005;**2**(3):195–6.
- Muto MG, Lage JM, Berkowitz RS, Goldstein DP, Bernstein MR. Gestational trophoblastic disease of the fallopian tube. *J Reprod Med* 1991;**36**(1):57–60.
- Gerson RF, Lee EY, Gorman E. Primary extrauterine ovarian choriocarcinoma mistaken for ectopic pregnancy: sonographic imaging findings. *AJR Am J Roentgenol* 2007;**189**(5):W280–3.

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