Primary ovarian choriocarcinoma mimicking ectopic pregnancy managed with laparoscopy - case report

Y.X. Chen[1], *M.D., Ph.D.*; **J. Xu**[2], *M.D.*; **W.G. Lv**[1], *M.D.*; **X. Xie**[1], *M.D.*

[1]*Department of Gynecologic Oncology,* [2]*Department of Reproductive Medicine, Women's Hospital, School of Medicine, Zhejiang University, Hangzhou (P.R. China)*

Summary

Nongestational ovarian choriocarcinomas are extremely rare and pose diagnostic challenges in reproductive-aged patients because of elevated human chorionic gonadotrophin (hCG). A 23-year-old nulliparous Chinese woman with nongestational ovarian choriocarcinoma escaped diagnostic testing and was initially treated for an ectopic pregnancy. Three months after her first visit, a diagnostic laparoscopy demonstrated a nongestational ovarian choriocarcinoma. Comprehensive surgical staging was performed by laparoscopy. The tumor was confined to the left ovary. The patient was categorized as FIGO Stage IA. She was given four courses of combined chemotherapy after laparoscopic surgery and has been disease-free for 36 months.

Key words: Nongestational ovarian choriocarcinoma; Human chorionic gonadotrophin; Laparoscopy; Chemotherapy.

Introduction

Nongestational ovarian choriocarcinoma is an extremely rare tumor presumably arising from the transformation of a single germ cell. In a recent report by Lai and colleagues, only one choriocarcinoma was identified among 84 ovarian germ cell malignancies (GCMs) [1]. Notwithstanding its sensitivity to chemotherapy, nongestational choriocarcinoma is associated with an unfavorable prognosis especially in advanced stage. Therefore, early diagnosis and timely initiation of therapy is important and present unique challenges in reproductive-aged women. This report describes the clinical features and outcome of a case diagnosed and managed with laparoscopic ipsilateral salpingo-oophorectomy and surgical staging followed by the administration of a multiagent chemotherapeutic regimen.

Case Report

A 23-year-old sexually active woman, gravida 0, was admitted to our hospital on April 9, 2004. She presented with ten days of vaginal spotting after 50 days of amenorrhea. The hCG level increased from 256.0 U/l (normal range below 10U/l) to 363.1 U/l during a ten-day interval. Considering ultrasound failed to identify an intrauterine pregnancy, an ectopic pregnancy was suspected and the patient was treated with 75 mg methotrexate (MTX). The hCG nadir of 58 IU/l was recorded two weeks after therapy but rose to 202 IU/l within 15 days without detectable imaging abnormalities by both ultrasound and abdominal computer tomography. The patient received a second course of MTX (100 mg). Again, a prompt decrease in hCG with a nadir of 26.6 IU/l two weeks after the second medication was observed, but followed by a sharp rebound to 878 IU/l and 18,703 IU/l within 30 and 45 days from the day of nadir, respectively. Transvaginal color Doppler ultrasound revealed a left ovarian mass measuring 1.5 cm x 2.2 cm x 2.0 cm with a lower resistance index (RI) of 0.43 (Figures 1a and 1b). Recorded hormone assay indices included LH: 0.41 IU/l, FSH: 2.2 IU/l, TTE: 1.4 nmol/l, E2: 499.8 pmol/l, P: 9.7 nmol/l, PRL: 26.2 ng/ml. Tumor markers including CA125, CEA and α-fetoprotein were within the normal range except for hCG. With an indeterminate diagnosis, a diagnostic laparoscopy was performed on July 14, 2004. Intraoperative assessment revealed a normal appearing uterus and right adnexal structures and a small solid mass 2.0 cm in diameter with a smooth surface in the left ovary (Figure 2). Inspection of all other viscera and peritoneal surfaces failed to identify any suspicious lesions. The solid mass was enucleated and submitted for frozen section analysis. Frozen section examination reported a primary ovarian choriocarcinoma. The patient underwent laparoscopic ipsilateral salpingo-oophorectomy and peritoneal multipoint biopsy and pelvic lymph node sampling.

A detailed pathological examination reported a mixed germ cell tumor consisting predominantly of choriocarcinoma with smaller components of dysgerminoma and immature teratoma. Surgical pathological stage was IA. Adjuvant chemotherapy consisted of one course of EMA-EP and three courses of BEP, completed in October 2004 without major complications. The hCG level normalized within 28 days after the operation and the patient has been without clinical or biochemical evidence of disease for 22 months. The pattern of alterations in the hCG levels during the total clinical course is shown in Figure 3.

Discussion

Nongestational choriocarcinoma is an extremely rare malignant tumor of the ovary. While occasionally detected as a homogeneous lesion, other germ cell components are frequently present. Unfortunately, this biologically aggressive tumor is commonly found in young woman prior to age 20. Early-stage ovarian choriocarcinoma presents a significant diagnostic challenge in the reproductive-aged patient because of elevated hCG. While the differential diagnosis includes both an ectopic pregnancy and gestational trophoblastic disease, seldom

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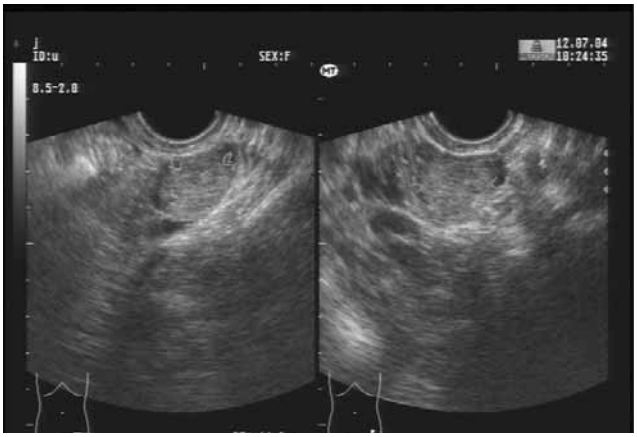
Fig. 1a



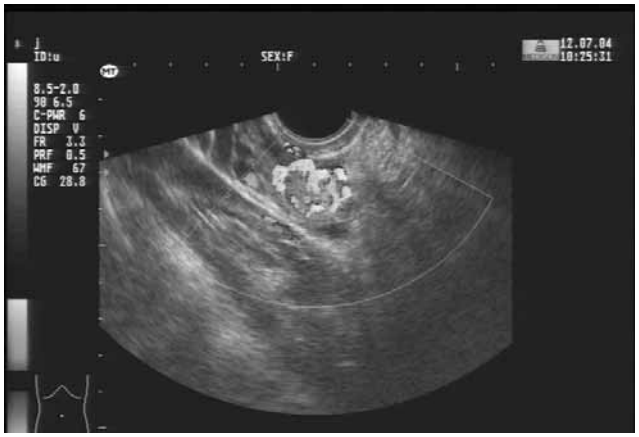


Fig. 1b

Figure 1. — (a) Transvaginal ultrasound showed a solid, heterogeneous lesion on the left ovary; (b) Color Doppler flow image showed multicolored abundant blood flow.

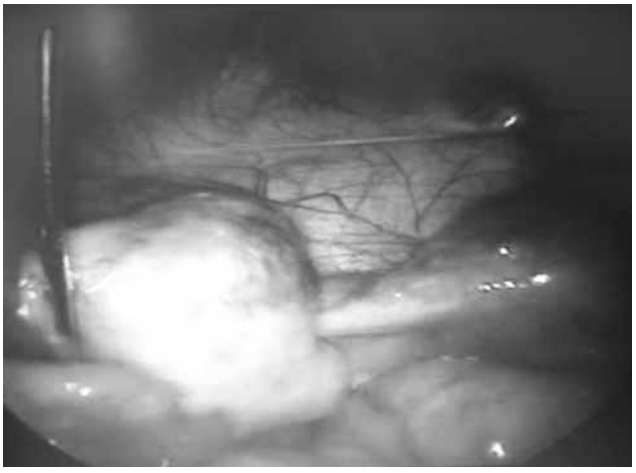


Figure 2. — Laparoscopy showed a solid mass 2 cm in diameter on the left ovary.

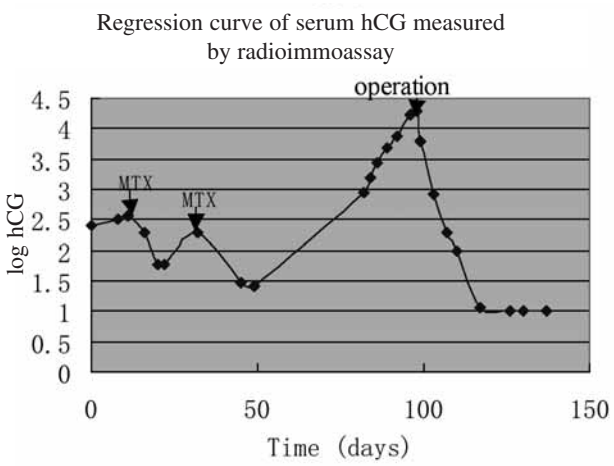


Figure 3. — The level of hCG went up and down during treatment with a single dose of MTX. After surgery and combined chemotherapy, hCG decreased sharply to normal within 28 days.

is nongestational choriocarcinoma a consideration. Irregular menses and vaginal bleeding may be common but with nonspecific complaints and signs that only reflect high hCG levels. Isosexual precocity has been reported to occur in about 50% of patients whose lesions appear before menarche [2]. The hCG may be the most sensitive measurement in diagnosing and monitoring the response to treatment. In the case reported here, the hCG was elevated before the abnormal image finding was detected. Laparoscopy would provide an additional important diagnostic tool in securing histologic definition. While laparoscopic management of benign ovarian tumors has become the treatment of choice, controversy persists regarding the management of malignant ovarian tumors that appear localized. There is justifiable concern about the risk of tumor rupture. Notwithstanding some studies that have demonstrated no adverse sequelae following intraoperative rupture for Stage I cancer, other reports have shown compromised outcomes [3, 4]. Laparoscopic retroperitoneal lymph node dissection has been applied widely to the staging and treatment of different kinds of cancer [5]. Some researchers consider that this minimally invasive procedure can fully duplicate the open technique and adhere to established strict oncologic principles [6]. A Gynecologic Oncology Group (GOG) study showed that laparoscopic staging of gynecologic malignancies can be successfully undertaken in selected patients [7]. In this reported case, with an indeterminate diagnosis, laparoscopic enucleation of the ovarian tumor was performed before pathological diagnosis was made. We think it is reasonable to complete the surgical staging procedure by laparoscopy when a lesion is confined to the ovary.

Only a limited number of reports exist evaluating the efficacy of the use of chemotherapy for nongestational choriocarcinoma. However, complete responses have been reported with the EMA/CO regimen (etoposide, methotrexate, actinomycin D, cyclophosphamide, vincristine) for pure nongestational choriocarcinoma [8]. The BEP regimen (bleomycin, etoposide, cisplatin) may be preferred as an alternative chemotherapy protocol [9]. An overall 5-year relative survival for ovarian germ-cell

tumors of 73% has been reported [10]. In this report, two courses of single-dose MTX were not effective although the pretreatment hCG level was low. The lack of response suggests that a single dose of MTX is not an appropriate choice for nongestational ovarian choriocarcinoma even though our case was in early stage with very low volume.

In conclusion, diagnostic laparoscopy is invaluable in the differential diagnosis between early stages of primary ovarian choriocarcinoma and other diseases with elevated hCG in reproductive-aged women. If the tumor is confined to the ovary, continuing the staging procedure by laparoscopy followed by an adjuvant BEP chemotherapy regimen is feasible when evidence-based therapeutic options are limited or non-existent.

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Address reprint requests to:
X. XIE, M.D.
Department of Gynecologic Oncology
Women's Hospital
School of Medicine, Zhejiang University,
No. 2 Xueshi Road,
Hangzhou 310006 (P.R. China)