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Case Report

Term pregnancy with choriocarcinoma presenting as severe fetal anemia and *postpartum* hemorrhage

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ABSTRACT

Objective: Term pregnancy with choriocarcinoma is a rare condition that can be a serious health threat to both the mother and the fetus. We present a rare case of term pregnancy with choriocarcinoma presenting as severe fetal anemia and *postpartum* hemorrhage.
Case Report: A 34-year-old woman, gravida 3 para 2, was admitted for profuse vaginal bleeding 2 weeks after cesarean delivery of a full-term anemic baby. Transvaginal sonography revealed a 4.7-cm × 10.6-cm heterogenous lesion in the endometrial cavity. Dilatation and curettage was done and a pathologic report revealed choriocarcinoma. Metastatic workup showed lung metastasis. The patient achieved remission after eight cycles of chemotherapy in the form of etoposide, methotrexate, actinomycin D, cyclophosphamide, and vincristine. There was no evidence of recurrence in the subsequent 3 years of regular follow up.
Conclusion: Although fetomaternal hemorrhage is a rare form of presentation of choriocarcinoma, its presence should alert the physician to investigate the cause further. This chemotherapy regimen was effective in our case and the patient needed to be followed up carefully.

Introduction

Term pregnancy with choriocarcinoma is a rare condition [1–10] that may present as hydrops fetalis [11], intrauterine fetal death, fetomaternal hemorrhage (FMH), and *postpartum* hemorrhage [2]. *Postpartum* treatment with multiagent chemotherapy is successful in most cases of choriocarcinoma [12,13], and timely diagnosis is prognostically important, both for the mother and the infant.

Case Report

A 34-year-old woman (gravida 3 para 2) presented to our hospital with a chief complaint of decreased fetal movements in the 37th week of gestation. Ultrasonography showed a normal fetus with oligohydramnios (amniotic fluid index = 4.1 cm). Fetal monitoring revealed a sinusoidal heart rate pattern (Figure 1). Per vaginal examination showed cervical dilation of 0.5 cm. A male infant (2650 g) was delivered by emergency cesarean with Apgar scores of 2 and 3 at 1 minute and 5 minutes, respectively. The neonate was pale with marked anemia (hemoglobin level = 3.7 g/L) and was admitted to the intensive care unit for intubation and blood transfusion. The mother was subsequently discharged 4 days after the cesarean delivery, but returned 2 weeks *postpartum* because of profuse vaginal bleeding. Transvaginal ultrasonography

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revealed a 4.7-cm × 10.6-cm heterogenous endometrial lesion (Figure 2A) with increased vascularity (Figure 2B). Dilatation and curettage was performed and pathological analysis revealed choriocarcinoma. Metastatic workup was performed. Computed tomography revealed a heterogenous mass measuring 16 cm × 12 cm × 9 cm in the uterine cavity (Figures 3A and 3B) and multiple lung metastases (Figure 3C). The level of serum β human chorionic gonadotropin was 538,312 mIU/mL. The patient was classified as Stage III in the International Federation of Gynecology and Obstetrics anatomic scoring system and had a score of 13 in the World Health Organization prognostic scoring system. She achieved complete remission (CR) after eight cycles of chemotherapy in the form of etoposide, methotrexate, actinomycin D, cyclophosphamide, and vincristine (EMA-CO). There was no evidence of recurrence in the subsequent 3 years of regular follow up.

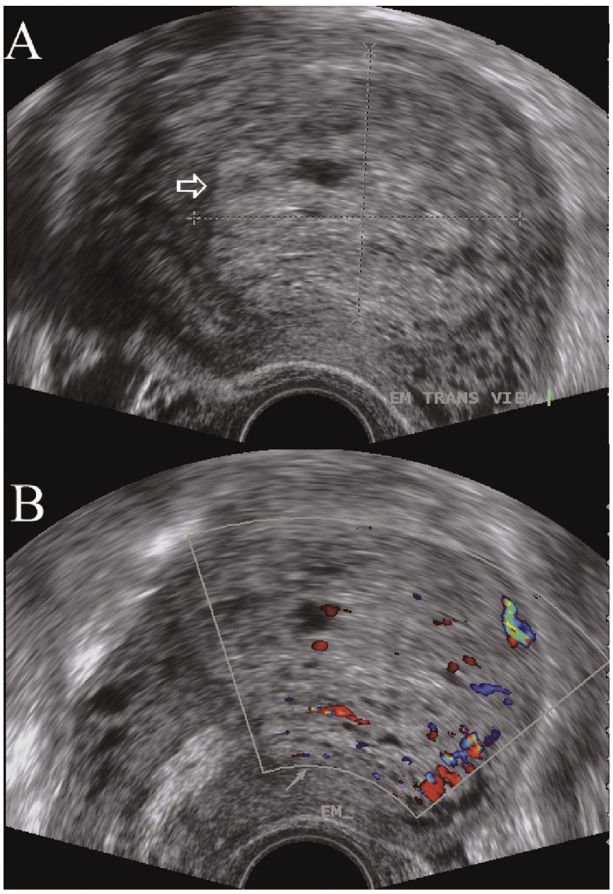

Figure 2. Ultrasonography at 2 weeks *postpartum* showing (A) a heterogenous lesion 4.7 cm × 10.6 cm (indicated by arrowhead) in the uterine cavity with (B) increased vascularity.

Discussion

Term pregnancy with choriocarcinoma is a rare condition [1–10]. The diagnosis is usually confirmed by the histopathological examination of the placenta after delivery or from samples of dilatation and curettage for *postpartum* hemorrhage. These cases may present as hydrops fetalis [11], intrauterine fetal death, FMH, and postpartum hemorrhage [2]. The last two symptoms were evident in the present case.

Previous studies have shown that choriocarcinoma is often incidentally discovered on histopathological examination. In a report of five cases of term pregnancies with choriocarcinoma [1], placentas of three of the cases were grossly similar with a single small lesion suggesting a small infarct. These lesions were confirmed to be choriocarcinoma on microscopic examination. The placenta of the fourth case contained a large marginal lesion microscopically identified as choriocarcinoma. The placenta of the fifth case had rare microscopic foci of choriocarcinoma and sheets of necrotic choriocarcinoma were identified in a blood clot submitted for analysis with the placenta. In another report of four cases of 3rd-trimester pregnancy with choriocarcinoma [8], the placentas in all cases were unremarkable macroscopically with small nondescript lesions, which were considered to be fresh infarcts or intervillus thrombi. These findings suggest that choriocarcinoma lesions in term pregnancy are easily overlooked by the gross examination of the placenta [1,4,7,8]. Therefore, it is highly recommended to submit the placenta for microscopic examination in suspicious cases.

Maternal and fetal complications in pregnancy are increased by choriocarcinoma and include vaginal bleeding, hydrops fetalis [11], FMH [10,11,14,15], intrauterine fetal death [6], and *postpartum* hemorrhage [12,13]. FMH can lead to preterm labor [16], decreased or absent fetal movements [11,16,17], fetal distress with a sinusoidal heart rate pattern, and neonatal death [17–19]. The most common presenting symptoms of FMH are anemia at birth (35.2%), decreased or absent fetal movements (26.8%), and unexpected stillbirth (12.5%) [16]. Prenatal findings of decreased or absent fetal body movements, sinusoidal heart rate pattern, or hydrops fetalis are usually late signs of FMH [16], making early detection relatively difficult.

The prognosis of patients diagnosed with choriocarcinoma is good, if an appropriate chemotherapeutic regimen is started early. A retrospective study reported CR in 87.8% of patients following the

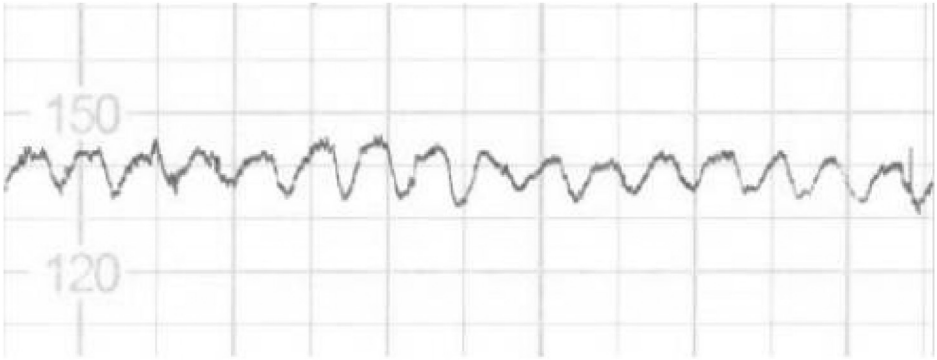

Figure 1. Sinusoidal pattern of fetal heartbeat.

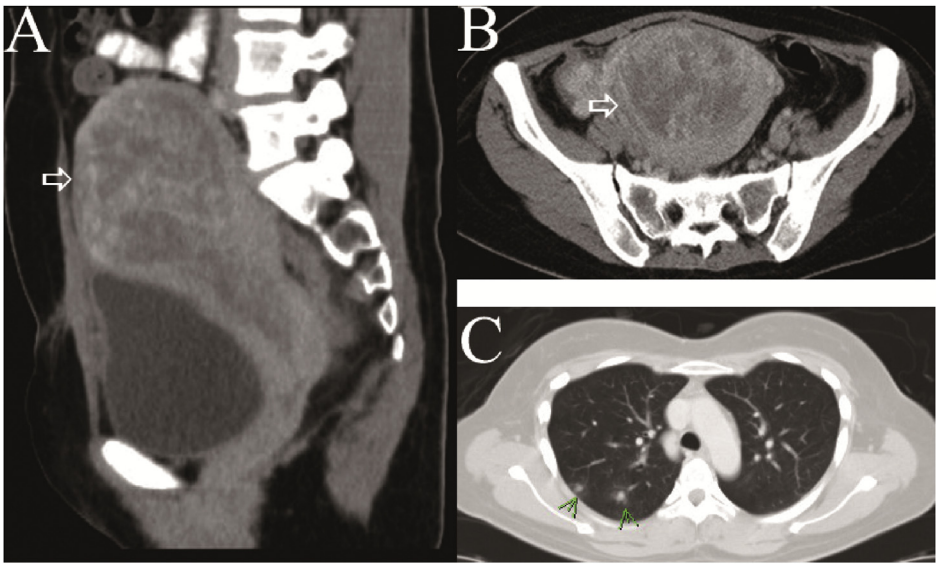

Figure 3. Computed tomography revealed (A, B) a heterogenous mass lesion measuring 16 cm × 12 cm × 9 cm in the uterine cavity and (C) multiple small nodules in lung fields.

administration of an average of 8.5 cycles of chemotherapy combined with comprehensive therapy [20]. The CR rates for patients diagnosed as FIGO (International Federation of Gynecology and Obstetrics) Stage I, II, III, and IV were 100%, 100%, 91%, and 62.5%, respectively. A shortened time interval between the antecedent pregnancy and treatment was correlated with improved prognosis. EMA-CO is the most commonly used combination chemotherapy to treat high-risk gestational trophoblastic neoplasia because it has the best efficacy-to-toxicity ratio. In a series of 272 women with high-risk gestational trophoblastic neoplasia who were treated with EMA-CO, 78% achieved CR [21]. A 16-year retrospective study of post-term choriocarcinoma patients at the New England Trophoblastic Diseases Center (Boston, USA) reported a survival rate of 87.5% [22].

In cases refractory to EMA-CO or relapse from EMA-CO chemotherapy, researchers reported that EMA-EP (etoposide and cisplatin) is an effective but moderately toxic regimen [23,24].

In summary, pregnancy with choriocarcinoma can be a serious health threat to both the mother and the fetus. Early diagnosis depends on a combination of detecting an unusually high level of serum β human chorionic gonadotropin, hydrops fetalis, fetal anemia, and *postpartum* hemorrhage. Although FMH is a rare form of presentation of choriocarcinoma, its presence should alert the physician to investigate the cause further. Chemotherapy with EMA-CO is effective in most cases. However, patients must be followed up carefully and any recurrent disease should be aggressively managed.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

Acknowledgments

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