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CASE REPORT

A Rare Case of Choriocarcinoma following a Normal Pregnancy and Delivery

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ABSTRACT

Introduction: Choriocarcinoma is one of the rarest entities encountered in routine obstetric practice. It is usually diagnosed when patient is symptomatic, presenting with either abnormal bleeding per vaginum or symptoms of distant metastases. The presentation of choriocarcinoma as secondary postpartum hemorrhage is a least common scenario. Choriocarcinoma is mostly seen following an abnormal pregnancy, either a partial or complete mole.

Case report: A 25-year-old para 3 live 3 patient presented with secondary postpartum hemorrhage on 4th week postpartum. She had two normal full-term vaginal deliveries that were uncomplicated with normal antenatal period. As she presented with secondary postpartum hemorrhage with the thought of retained products of conception, an emergency diagnostic dilatation and curettage was done. Histopathology was suggestive of gestational choriocarcinoma. Patient was given single regimen of methotrexate and subsequently decision of total abdominal hysterectomy was taken and patient was kept in close follow-up. Presently, beta-human chorionic gonadotropin (hCG) is below 2 mIU/mL.

Conclusion: The concern is the time interval or delay between the onset of symptoms and diagnosis with subsequent treatment. Although there are a number of reasons of postpartum hemorrhage, simple curettage for histological study and titer serum beta-hCG may lead to early diagnosis and initiation of treatment. Obstetricians and pathologists should have increased awareness regarding the consequences of choriocarcinoma.

Keywords: Beta-hCG, Choriocarcinoma, Secondary postpartum hemorrhage.

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INTRODUCTION

Postpartum choriocarcinoma is a rare complication of pregnancy. Partial moles and persistent trophoblastic tumors rarely give rise to choriocarcinoma. Choriocarcinomas can occur after any type of pregnancy – but usually occur following an abnormal pregnancy, either a complete mole or partial mole. The incidence of choriocarcinoma following term delivery without a history of complete mole is approximately 1 case in 50,000 births in the United States.[1,2]

The incidence varies geographically, with highest incidence in South-East Asian countries.[3] Studies from the United States and England have found that women with a history of one molar pregnancy (partial, complete, or persistent gestational trophoblastic neoplasia) have an approximately 1% chance of recurrence in subsequent pregnancy compared with a 0.1% incidence in the general population of the United States. The true incidence may actually be much higher as histology is often difficult to obtain, but interestingly choriocarcinoma following a full-term pregnancy is more often associated with aggressive form of disease and the presentation is similar to those of hydatidiform moles.

CASE REPORT

A 25-year-old patient visited the Department of Obstetrics and Gynecology at Acharya Vinoba Bhave Rural Hospital, Datta Meghe Institute of Medical Sciences, Sawangi, India, with complaint of bleeding per vaginum (secondary postpartum hemorrhage 15 days) during outpatient department hours. She delivered her third baby 15 days earlier in a private hospital. Her previous two deliveries were also full-term normal vaginal deliveries and were uncomplicated in antenatal, intranatal, and postnatal phases. Patient was conscious, cooperative. She was a B.Com graduate. She was thin-built, averagely nourished with height of 156 cm and weight 45 kg. General examination revealed severe pallor and edema feet, pulse was 110/minute, and blood pressure 110/78 mm Hg, systemic examination was normal followed by local examination, per abdomen soft uterus was well retracted. On per speculum examination, passage of blood clots was seen, and active bleeding through cervical os was present. Patient was advised admission for further workup. Ultrasound of pelvis was suggestive of enlarged uterus due to postpartum phase, with normal ovaries and thickened endometrium of 20 mm. She was posted for diagnostic dilatation and evacuation in view of retained products of conception. The correction of anemia was done by blood

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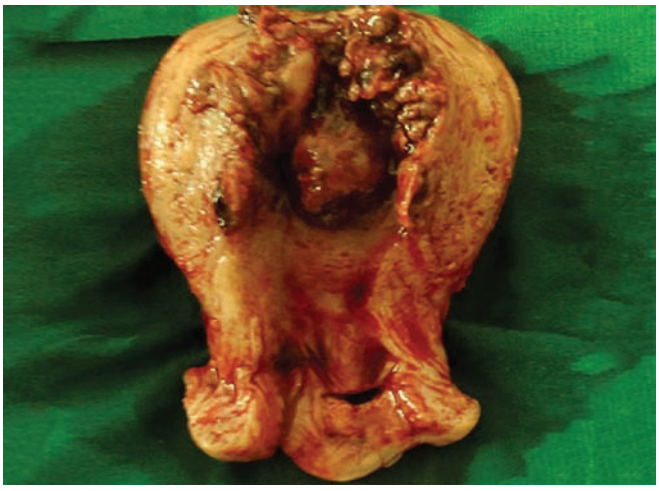

Fig. 1: The gross appearance of cut section of the uterus and cervix after total abdominal hysterectomy with malignant mass at the uterine fundus

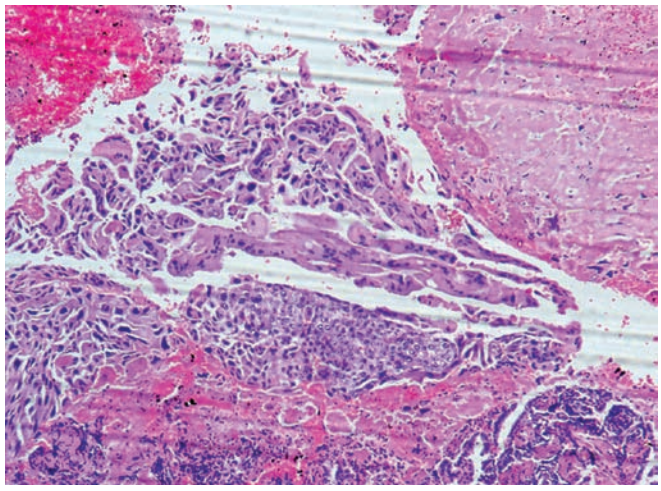

Fig. 2: Histological presentation of choriocarcinoma with syncytiotrophoblasts and cytotrophoblasts

transfusion simultaneously. After 7 days, histopathology was suggestive of choriocarcinoma with syncytiotrophoblast and cytotrophoblast cells (Fig. 1). Malignancy workup was done using chest X-ray, magnetic resonance imaging brain, and ultrasonography abdomen and pelvis; thyroid profile and serum beta-human chorionic gonadotropin (beta-hCG) were s/o 35, 156 mIU/mL. Patient had continuous bleeding per vaginum even after giving one cycle of methotrexate 1 mg/kg along with inj folinic acid 0.1 mg/kg, and therefore, decision of total abdominal hysterectomy was taken (Fig. 2). Postoperatively, patient was given one dose of chemotherapy with methotrexate on days 1, 3, 5, 7 and folinic acid on days 2, 4, 6, 8. The postoperative period remained uneventful. Patient was discharged on day 25 of surgery. Patient was informed regarding the follow-up and serial beta-hCG. Till date, she is in follow-up with serum beta-hCG less than 2 mIU/mL.

The patient in this case report presented with secondary postpartum hemorrhage; her previous two deliveries were normal, with no history of molar pregnancy either complete or partial. The presentation of choriocarcinoma with secondary postpartum hemorrhage is a very rare scenario.

DISCUSSION

Gestational trophoblastic disease most commonly follows molar pregnancy but may also occur following normal or ectopic pregnancies, spontaneous or therapeutic abortions. Its incidence varies with figures as high as 1 in 120 pregnancies in some areas of Asia and South America, compared with 1 in 1,200 in the United States. Metastatic disease occurs in 4% patients after local management of hydatidiform mole. The incidence of choriocarcinoma after complete hydatiditorm mole is about 1,000 times greater than after a normal pregnancy.[4]

Choriocarcinoma is a rare tumor. In Western countries, the incidence is 1 in 45,000 pregnancies. Higher incidence is reported from Africa, Asia, and South America.[5] Majority of cases occur in women aged less than 35 years.[6]

Postpartum choriocarcinoma rarely affects a viable pregnancy, with an incidence of only 1 case in 50,000 births in Western countries.[1,2] Choriocarcinoma, also known as chorioblastoma or trophoblastic tumor, is a rare form of cancer that occurs in the female genital tract. It may develop after a normal pregnancy; however, it is usually associated with antecedent history of molar pregnancy (50%), ectopic pregnancy (5%), miscarriage or abortion, or normal pregnancy (20%). Gestational choriocarcinoma is a highly malignant tumor with a very high propensity to metastasize to various sites including lungs, vagina, brain, liver, kidney, and gastrointestinal tract, in descending order of frequency. These cases present with vaginal bleeding, anemia, hyperemesis gravidarum, hyperthyroidism, uterine and ovarian enlargement, and pregnancy-induced hypertension.[7,8]

The precise molecular pathogenesis of gestational trophoblastic disease is yet to be elucidated. Genetics has a well-established role. The ultimate cause of gestational trophoblastic disease is claimed to be genetic in origin. No environmental etiological factor has been implicated up to now apart from deficient vitamin A precursor carotene in the diet. Obstetrically, the majority of the reported cases till now had a normal prenatal course. Choriocarcinoma was often diagnosed after the mother developed symptoms of metastasis postpartum. The symptoms included vaginal bleeding, chest pain, and neurological signs, such as seizures and stroke. In terms of maternal outcome, mothers who developed metastasis were often treated with chemotherapy, which consisted of etoposide, methotrexate, actinomycin D, cytoxan, or

vincristine (EMA-CO regime). The response rate after chemotherapy is high. Kodama et al[9] reported a case where the mother underwent partial lobectomy after pulmonary lesions were identified in 1994. Four cases of hysterectomy were reported by Brewer et al,[8] Ollendorff et al,[10] and Flam et al.[11]

Nugent et al[12] determined the clinical presentation and outcome of postpartum choriocarcinoma. The study was conducted from 1997 to 2005, and all the women who were histologically confirmed were included. The study was conducted in Sheffield Trophoblastic Treatment Center. There were 751 cases of trophoblastic disease. Out of these, 99 received chemotherapy for choriocarcinoma, 64 received chemotherapy following a miscarriage, ectopic pregnancy, or molar pregnancy, and 35 patients had viable pregnancy. In 33 patients, the presenting symptom was vaginal bleeding. In 10 cases, continuous abnormal bleeding per vaginum was seen just after the delivery. At the time of malignancy workup, 20 out of 35 patients had evidence of metastatic disease. The most common metastatic site was pulmonary (17 cases) for which lobectomy was also performed. Regarding fertility after chemotherapy, only 6 required hysterectomy, 8 had subsequent pregnancy after high-risk chemotherapy, and out of 11 patients who received high-risk chemotherapy, 9 had normal offspring without congenital anomalies. The present study concluded that although postpartum choriocarcinoma is an extremely rare condition, it is important for obstetricians and gynecologists to remain aware of this possibility in patients with persistent postpartum vaginal bleeding.

CONCLUSION

The concern is the time interval or delay between the onset of symptoms and diagnosis with subsequent treatment. Although there are a number of reasons of postpartum hemorrhage, simple curettage for histological study and titer serum beta-hCG may lead to early diagnosis and initiation of treatment. There should always be some index of suspicion if abnormal bleeding per vaginum persists after therapeutic intervention. Delay in diagnosis may result in increased morbidity, thereby increasing the chance of distant metastasis. In conclusion, the important message is that this condition is extremely uncommon, but when diagnosed is potentially treatable and curable.

Although postpartum choriocarcinoma is extremely uncommon, there is need for obstetrician to be aware of this possibility in cases of persistent postpartum vaginal bleeding.

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