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Choriocarcinoma following live birth: is it a delay in diagnosis or ignorance of the disease?

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Summary

Objective: Molar pregnancies and choriocarcinomas are derived from villous trophoblast and placental site trophoblastic tumor (PSTT) and epithelioid trophoblastic tumor (ETT) are derived from extravillous trophoblast. Of all forms of gestational choriocarcinoma, placental choriocarcinoma is the most rare and is usually diagnosed in symptomatic patients with metastases. The incidental finding of a choriocarcinoma confined to the placenta with no evidence of dissemination to mother or infant is the least common scenario. Choriocarcinoma coexisting with or after a "normal" pregnancy has an incidence of one per 160,000 pregnancies. The authors report the first case diagnosed in their department. *Case Report:* A 34-year-old woman was diagnosed with abundant vaginal bleeding after term pregnancy at three months after the delivery of a healthy baby. Abdominal ultrasonography revealed an intracavitary uterine tumoral mass with signs of myometrial invasion to the uterine serosa. The pretreatment hCG level was 1,500,000 IU/Ml. Computed tomography scan showed a pelvic mass on the uterus that invaded the neighborhood organ, extending to posterior retro-peritoneal organ (large vessels), but excluded extrapelvic tumoral masses. Removal of the tumor was impossible due to its extension to the digestive system and the large vessel, necrosis, and hemorrhagic character, therefore the authors were limited to a biopsy of the tumor. Histopathological examination revealed uterine choriocarcinoma after four courses of chemotherapy, and the patient experienced severe hemorrhagic shock in the general surgery department during surgery due to acute intestinal obstruction. *Conclusion:* Although postpartum choriocarcinoma is extremely uncommon, there is a need for obstetricians to remain aware of this possibility in patients with postpartum vaginal bleeding with a healthy newborn. Early diagnosis by histopathological examination of the placenta, the precocity of the diagnosis influencing the prognosis, and tumor response to chemotherapy are important.

Key words: Choriocarcinoma; Term gestation; Villous trophoblast; Placental site trophoblastic tumor.

Introduction

Choriocarcinoma is the most aggressive form of gestational trophoblastic disease (GTD) owing to their rapid growth and metastatic potential. They most commonly develop after a complete mole (CM); the incidence of an antecedent history of CM has been reported in 29–83% of choriocarcinoma cases in various studies across the world [1]. It is a malignant proliferation of syncytial trophoblast cells that do not form placental villi.

Choriocarcinoma coexisting with or after a "normal" pregnancy has an incidence of one per 160,000 pregnancies [2]. It is associated with an unfavorable outcome especially because of delayed diagnosis. If the interval from index pregnancy to initial treatment is less than four months, the remission rate is 87.5% [5]. Most of the cases reported in the literature known as choriocarcinoma after term pregnancy were diagnosed through macroscopic and histopathological examination of the placenta [2].

In this paper, the authors report the case of a 34-year-old woman diagnosed with choriocarcinoma after term pregnancy at three months after the delivery of a healthy baby with a review of the literature of this pathology.

Case Report

A 34- year old woman, gravida 1, para 1, without particular history, had delivered about three months ago (a caesarian delivery) at term after a normal pregnancy, and gave birth to a male healthy baby weighting 3,200 grams. The placenta was normal macroscopically at delivery. The woman breast-fed her baby and did not use any contraception. Approximately three months later, she returned to the present unit with abnormal vaginal bleeding. Indeed, there was no history of fever, abdominal pain, nausea or vomiting. On examination, the uterus was increasing in size on bimanual examination. B-hCG level was obtained and was found to be superior to 1,500,000 mUI/ml. A transvaginal ultrasound scan revealed a heterogeneous mass of 12×12 cm in the Douglas pouch.

CT scan revealed a solid cystic mass 105×118×124 mm lateralized to the right with undermined outline, invading:

The greasy structure, the anterior rectal wall (Figure 1), the posterior vesical wall (Figure 2), the bowel loops wall (Figure 3), primitive right iliac artery (Figure 4), the sacral nerve roots (Figure 5), both sacroiliac ligaments (more marked on the right) (Figure 6) are all shown

During surgery (Figures 7-9), the authors found a uterine mass of 12 cm, with cystic areas, adherent to surrounding structures, invading the Douglas pouch and even the posterior peritoneum vessels, with shielding. This mass was bleeding on contact. The authors' opted for abstention with mass biopsy. The pathological examination concluded with a choriocarcinoma.

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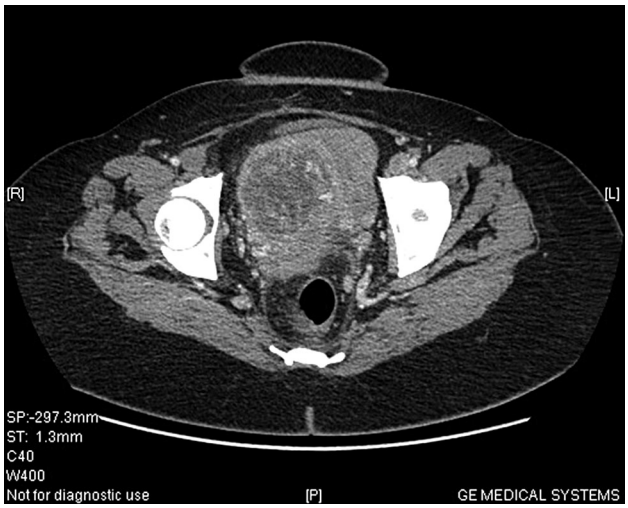


Figure 1. — Invasion of the anterior rectal wall.

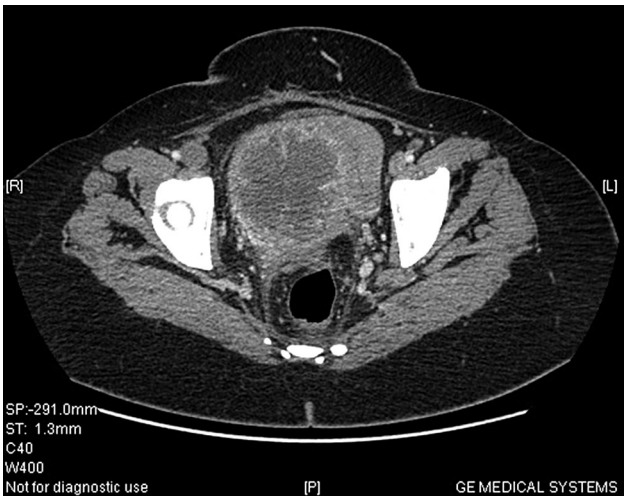


Figure 2. — Invasion of the posterior vesical wall.

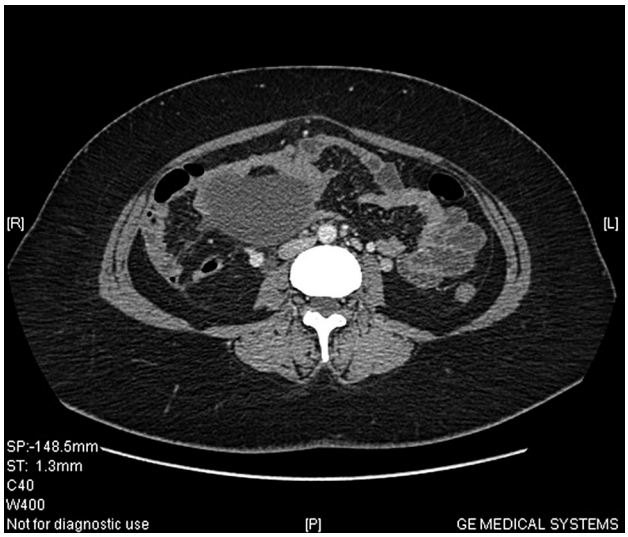


Figure 3. — Invasion of the bowel loops wall.

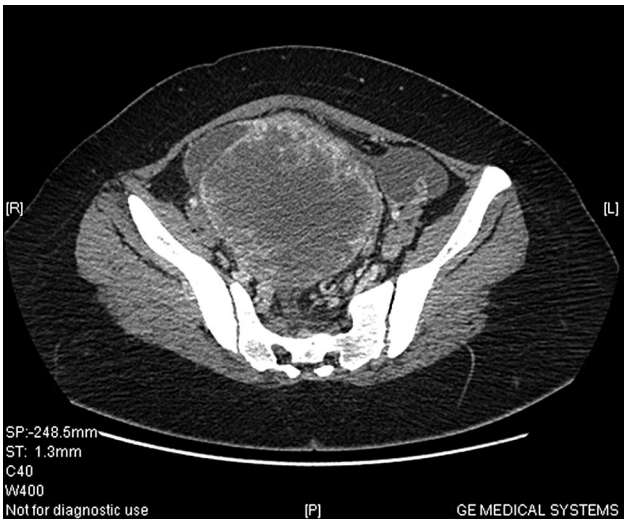


Figure 4. — Invasion of iliac artery primitive right.

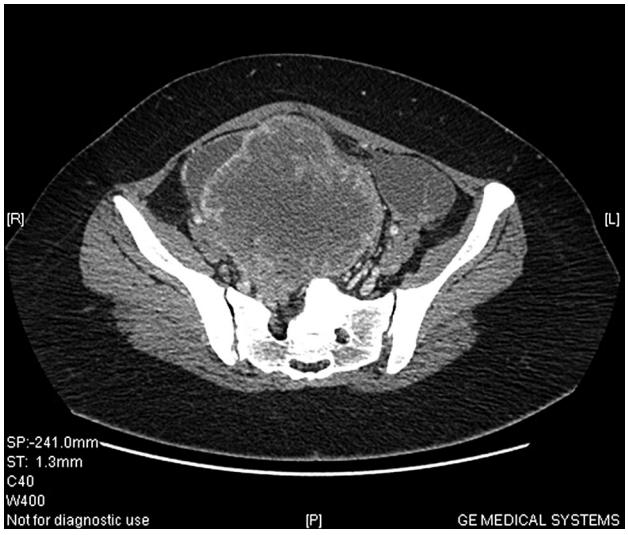


Figure 5. — Invasion of the sacral nerve roots.

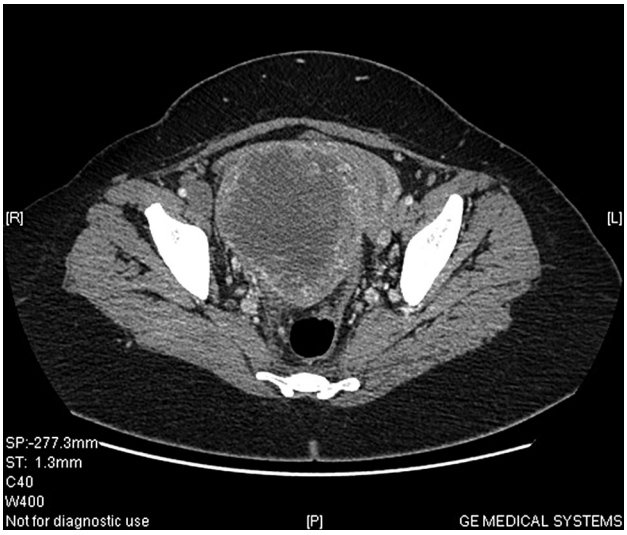
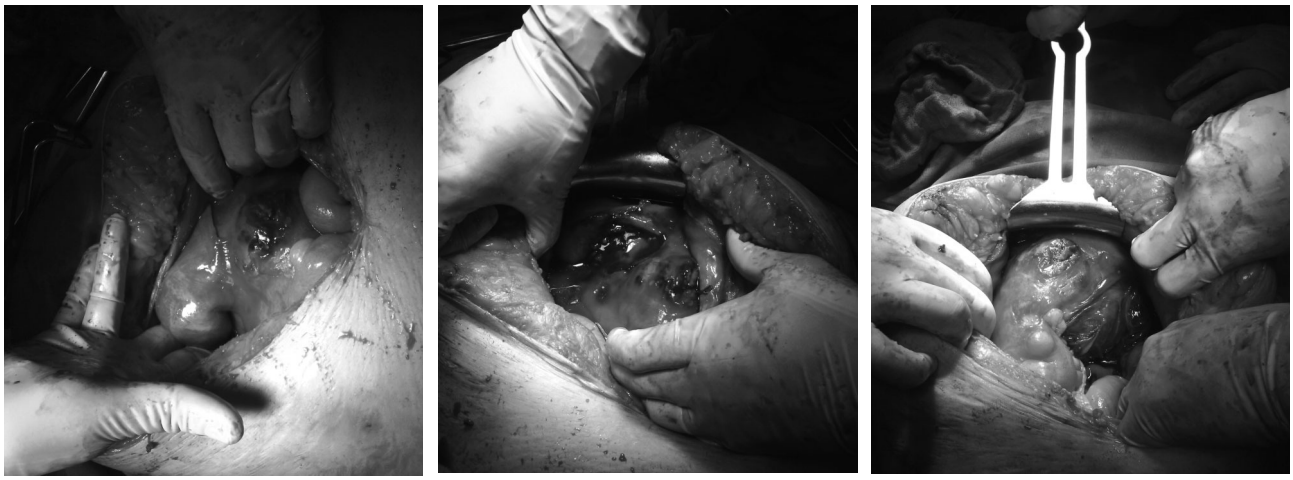


Figure 6. — Invasion of both sacroiliac ligaments.



Figures 7, 8, and 9. — Exploratory laparotomy.

The patient was transferred immediately after diagnosis to a center of chemotherapy where she received four cycles. The evolution was unfavorable and the woman died following a severe hemorrhagic shock in the general surgery department during surgery for acute intestinal obstruction. The newborn was healthy, had neither anemia nor liver masses. He is now one-year-old and is asymptomatic.

Discussion

Postpartum choriocarcinoma is a very rare complication of pregnancy with an incidence of one per 160,000 pregnancies [2]. The rare occurrence of choriocarcinoma after a live birth or molar abortion often leads to symptoms and signs of this disease which is ignored [3, 4]. Although the most common cause of postpartum hemorrhage is complication of delivery, but it should be remembered that GTN may be occur [3].

Choriocarcinoma appeared to have a propensity for early dissemination in liver and brain [4]. However, despite this aggressive behaviour, the opportunity for cure in patients may still be favorable with early diagnosis [5].

Since choriocarcinoma develops rapidly and is relatively aggressive, it is suggested that the cellular transformation did not occur during pregnancy and that the tumor arose from placental cells.

It is supposed that a few trophoblast cells might remain in the body after every pregnancy but they degenerate naturally. In rare instances, if this process is delayed, the transition of normal cells to malignant proliferation seems possible [6].

Generally, the main presenting complaint of the patient is vaginal bleeding three to four weeks after delivery. Late presentation of choriocarcinoma has been reported, even after 15 years after gestation and even after menopause [3]. In the present case, it was discovered three months after delivery.

Choriocarcinoma is a very aggressive malignancy and death may result from delays in diagnosis; therefore, its diagnosis should be considered in any woman in the reproductive age group presenting with abnormal vaginal bleeding or unexplained systemic symptoms. Clinical suspicion and any gross placental anomaly should mandate a thorough pathological examination of placenta. The usefulness of routine examination of tissue from uterine evacuation (placenta) has been questioned, but the recommendations for UK practice is that tissue should be sent routinely [7].

Diagnosis of choriocarcinoma is made by the raised concentration of hCG and appropriate histology and this was the case in the present patient. Final histopathology of choriocarcinoma requires the juxtaposition of cytotrophoblastic and syncytiotrophoblastic cells with complete disappearance chorionic villus with invasion of the myometrium. Many reports concluded that choriocarcinoma occurring after a live birth carries a worse prognosis than after a premature delivery or a miscarriage [7].

Infantile choriocarcinoma is even more rare, with less than 30 cases described in the literature [8]. Newborn infants tend to present with a characteristic clinical picture of anemia, hepatomegaly, and precocious puberty [8]. Without appropriate treatment, death usually occurs within three weeks of initial presentation [8]. Here the authors reported the case of a maternal choriocarcarcinoma, with a healthy infant with no symptoms and whose neonatal examination was normal. The infant is now one-year-old and is apparently healthy.

Conclusion

The authors conclude that although postpartum choriocarcinoma is extremely uncommon, there is a need for obstetricians to remain aware of this possibility in patients

with postpartum vaginal bleeding with a healthy newborn.

The incidence of placental choriocarcinoma may actually be higher than expected since it is not routine practice to send placentas for pathological evaluation after a normal spontaneous delivery [9]. Early diagnosis by histopathological examination of the placenta, the precocity of the diagnosis influencing the prognosis, and tumor response to chemotherapy are important.

Acknowledgements

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