CASE REPORT

Missing choriocarcinoma may be easy but not suspecting it in a high-risk case can be potentially fatal

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Accepted 20 March 2015

SUMMARY

A 27-year-old woman, fourth gravida, with three prior consecutive vesicular moles was diagnosed with a recurrent vesicular mole on ultrasonography (USG) and had very low β -human chorionic gonadotropin (HCG) values. During suction evacuation no vesicles were seen and on repeat USG the patient was diagnosed to have fibroid uterus. She was discharged at request and advised to undergo MRI to rule out choriocarcinoma. The MRI was interpreted as fibroid uterus with degeneration. After 3 weeks of suction evacuation, the patient presented with acute abdomen. She underwent emergency laparotomy for haemoperitoneum and was diagnosed as invasive mole with perforation; total hysterectomy was performed. Her β -HCG after laparotomy was more than 200 000 mIU/L, and the histopathological examination revealed choriocarcinoma. When methotrexate, adriamycin and cyclophosphamide (MAC) therapy was advised, the patient initially received methotrexate monotherapy; after three cycles her β -HCG started rising after an initial drop, and the patient required four cycles of EMACO to achieve remission.

BACKGROUND

Choriocarcinoma is fatal if not diagnosed and treated early. The diagnosis is difficult when β -human chorionic gonadotropin (HCG) levels are low, as there are no characteristic features on ultrasonography (USG) when compared with vesicular mole. When a mass lesion is present in the uterus, misdiagnosis occurs. A high index of suspicion can save the uterus and fertility potential as well as the life of the patient.

CASE PRESENTATION

A 27-year-old nulliparous woman with a history of three consecutive vesicular moles (G4V3) was hospitalised with a diagnosis of recurrent vesicular mole in August 2013. Her last pregnancy was 6 months ago, and was managed elsewhere with dilation and evacuation. She had no history of receiving chemotherapy.

During this pregnancy she had 3 months of amenorrhoea; on examination, the uterus was 18 weeks in size and USG showed a snowstorm appearance. Her β -HCG was 351.6 mIU/L and she underwent suction evacuation on 27 August 2013 under general anaesthesia. No vesicular structures were sucked out and only minimal product was obtained, hence a consultant was called in and suction evacuation was attempted under USG

guidance. The patient was diagnosed with a right lateral wall fibroid and the procedure was given up. β -HCG was repeated after a week and was 612 mIU/mL. The patient was advised MRI to rule out the possibility of choriocarcinoma. But the patient was discharged at request the next day after fixing the date for MRI. The material obtained during suction evacuation was reported as blood clots on histopathological examination.

Subsequently, the patient presented to the out-patient department on 18 September 2013, 10 days after discharge, with acute onset of abdominal pain, giddiness and three episodes of vomiting. There was no history of bleeding per vaginum. On examination, she had severe pallor, her pulse was 130/min and blood pressure was 110/80 mm Hg. Her abdomen was rigid and there was guarding and rigidity; the uterus was 22 weeks in size and tender on palpation. Per speculum examination revealed brownish discharge through os, and the cervix and vagina were pale. Per vaginal examination, the cervix was soft and the mass was felt to be uterine as it was continuous with the abdominal mass, and the movements were transmitted to the cervix. USG repeated by the same consultant along with a radiologist was reported as fibroid uterus measuring 9×7 cm and cysts in both ovaries, which on the right measured 7.3×6.8 cm and on the left, 4.6×4.7 cm; there was free fluid. Diagnostic aspiration revealed frank blood and the patient underwent emergency laparotomy with a provisional diagnosis of uterine perforation with a high risk consent for hysterectomy.

INVESTIGATIONS

22/8/13

USG: Uterus enlarged with snowstorm appearance/vesicular mole

β -HCG—351.6 mIU/mL

27/8/13

USG: Right lateral wall fibroid

2/9/13

β -HCG—612 mIU/mL

5/9/13—MRI showed a 10.3×10 cm well defined heterogeneous mass involving the fundus, and anterior and posterior myometrium, with a central non-enhancing area suggestive of central necrosis and fibroid uterus with degeneration (figures 1 and 2).

18/9/13—USG: Fibroid uterus 9×7 cm; right ovary 7.3×6.8 cm; left ovary 4.6×4.7 cm; free fluid present

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To cite: Dasari P. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-208526

Figure 1 Sagittal section of the pelvis showing a heterogeneous mass lesion occupying the fundus anterior and posterior myometrium with central non-enhancing area.

HPE (histopathological examination number 1340/13): Large areas of haemorrhagic necrosis; biphasic pattern of intermediate trophoblast, cytotrophoblast and syncytiotrophoblast. The intermediate trophoblasts are chronic type (p63) with high mitosis (Ki 67–80%) invading and proliferating the uterine wall, indicative of choriocarcinoma

21/9/13— β -HCG >225 000 mIU/mL
 17/11/13—>1000 mIU/mL
 7/11/14—1540 mIU/mL
 18/11/14—22 851 mIU/mL
 18/11/14—500 mIU/mL
 10/12/14—1504 mIU/mL

Figure 2 Coronal section of pelvis showing the heterogeneous mass lesion at the fundus of the uterus along with theca lutein cysts.

17/1/14—1540 mIU/mL
 25/2/14—13.6 mIU/mL
 21/3/14—2 mIU/mL
 Fourth cycle of EMACO—17/5/14; 3/7/14— β -HCG –1 mIU/mL.

DIFFERENTIAL DIAGNOSIS

Fibroid uterus.

TREATMENT

Emergency laparotomy showed 2 L of haemoperitoneum and uterus enlarged to 20 weeks in size. Bilateral theca lutein cysts were present. Multiple vascular infiltration in the myometrium was seen as blebs on the fundus and a perforation of 2×2 cm from which blood was oozing was seen. Perforated invasive mole was diagnosed and total hysterectomy was performed. The patient's β -HCG after 48 h of surgery was more than 200 000 mIU/L and she was advised to receive MAC therapy, but initially only methotrexate monotherapy was given.

OUTCOME AND FOLLOW-UP

The histopathological report was choriocarcinoma and after three cycles of methotrexate monotherapy, β -HCG plateau occurred. Subsequently, the patient was admitted and treated with EMACO. After three cycles her β -HCG normalised to 2 mIU/L and she was given one more cycle of EMACO; she was in good health when last seen in July 2014. She was advised follow-up for a period of 2 years and counselled for adoption. The patient gave permission to use the MRI pictures and details of the case for publication.

DISCUSSION

The incidence of choriocarcinoma in southeast Asia is 9.2/40 000 pregnancies when compared with 1 in 40 000 pregnancies and 1 in 40 molar pregnancies in Europe. The risk is approximately 1000 times higher after a molar pregnancy than after a normal pregnancy.¹ The risk of choriocarcinoma after repeated molar pregnancy is much greater. Choriocarcinoma usually occurs 50% of the time following vesicular mole, 25% of the time following abortion, 20% of the time after a normal pregnancy and 5% of the time following an extrauterine pregnancy. The diagnosis of choriocarcinoma is difficult, especially when β -HCG levels are low, as in this case. Mortality occurred in a case of choriocarcinoma that was clinically diagnosed as postabortal sepsis, as the urinary as well as serum β -HCG were negative and the USG picture was interpreted as that of a degenerating intramural fibroid. The authors state that the reason for the negative HCG report was due to the less sensitive HCG kits, which cannot detect nicked HCG and free β -HCG, and, of the available sensitive methods, DPC immulite assay is the most sensitive.² There are no specific ultrasonographic features described to diagnose choriocarcinoma, unlike vesicular mole, which has a characteristic snowstorm appearance on USG. A retrospective analysis of sonographic and Doppler findings for diagnosis of gestational trophoblastic disease concluded that the abnormal sonographic findings confined to the endometrial cavity in all cases of complete hydatidiform moles and soft tissue invasion, and cystic vascular spaces within the myometrium, were seen in invasive mole and choriocarcinoma. The resistive indices were high for complete mole and very low for invasive mole and choriocarcinoma.³ Sonographic features in cases of proven choriocarcinoma were described as being variable: nodular, submucosal, macrocystic, multicystic, compact and microcystic, with involvement of surrounding areas. They were also found to mimic other diseases of the endometrium

and myometrium.⁴ Fibroids appear as well defined solid masses with a whorled appearance, usually of similar echogenicity to myometrium; they can sometimes be hypoechoic. However, degenerated fibroids have a complex appearance with cystic change.⁵ The sensitivity of transvaginal USG for early diagnosis of molar pregnancy is only 50–86%, and 70% of partial moles can be missed.⁶ Kirk *et al* reported only 44% sensitivity and 48% positive predictive value for the USG diagnosis of histologically confirmed molar pregnancy.⁷

In choriocarcinoma, MRI of the uterus usually shows a high signal pattern outside and an irregular low signal pattern inside the mass lesion on T2-weighted images. The lesion is heterogeneous and hypervascular, and distorts the normal anatomy, sometimes extending to the parametrium.⁸ MRI is highly sensitive in diagnosing fibroids; non-degenerated fibroids are well-defined masses of low signal intensity as compared with the myometrium on T2-weighted images, and isointense on T1-weighted images; but they can also have a high intensity rim on T2-weighted images. All types of degeneration in fibroid produce low signals on T2-weighted images and can have a heterogeneous appearance.⁵ MRI findings of a degenerated fibroid closely mimic those of the choriocarcinoma. However, in this case, presence of a mass lesion with cystic spaces in the myometrium on ultrasound and MRI with the history of three previous vesicular moles pointed to a choriocarcinoma rather than a fibroid uterus, as the risk of choriocarcinoma developing even after one vesicular mole is 20%.

Learning points

- ▶ Every effort should be made to rule out choriocarcinoma with a prior history of recurrent vesicular mole.
- ▶ It is essential to know the imaging features of choriocarcinoma.
- ▶ Early diagnosis can be made with a high index of suspicion, and chemotherapy (EMACO) is the first-line treatment to achieve a cure in cases of choriocarcinoma.
- ▶ It is essential to understand that non-metastatic choriocarcinoma is 100% curable, with chemotherapy preserving fertility.

Spontaneous uterine perforation is reported as an initial clinical presentation⁹ and also when resolution occurs while the patient is on follow-up subsequent to chemotherapy.¹⁰ Patients with perforation invariably present with shock, as in the present case. Mortality can occur if the patient does not present early.¹¹ Management includes resection of the mass lesion followed by chemotherapy or hysterectomy. Successful pregnancy can be achieved after resection and chemotherapy.¹² Hysterectomy was undertaken in this case as the mass lesion was very large, occupying more than two-thirds of the uterus, and was highly vascular. Hysterectomy is a life-saving procedure in such cases.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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