Rare disease

A case of true tubal hydatidiform mole and literature review

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Summary

Tubal hydatidiform mole is an uncommon condition with about 40 confirmed cases in the accessible literature. The patient usually presents with symptoms and signs of a classical ectopic pregnancy and it is only after histological examination and DNA ploidy analysis of the conceptus that a hydatidiform mole is diagnosed. Management requires complete removal of the conceptus and follow-up needs to be arranged with an appropriate supraregional centre. The authors present a case of complete molar tubal pregnancy and a review of the literature.

BACKGROUND

This case demonstrates an unusual presentation of a hydatidiform mole. It illustrates the importance of histological examination of products of conception following surgical removal of the conceptus in ectopic pregnancies; only then it is possible to reach the correct diagnosis and offer appropriate counselling and follow-up to the patient.

CASE PRESENTATION

The patient presented in A&E with postmenopausal vaginal (PV) bleeding and abdominal pain.

She initially had PV spotting which became progressively worse ('slightly heavier than a period'). Her pain was intermittent, mild to moderate in severity and was localised in the lower abdomen and left iliac fossa with no radiation. She thought her last menstrual period was only 2 weeks earlier, but a pregnancy test was positive in A&E. She was not using any contraception as she was actively trying to conceive with her long term partner.

Past gynaecological history included one normal vaginal delivery and two episodes of bacterial vaginosis (but no history of pelvic inflammatory disease). She had also been seen twice in the colposcopy clinic following one mild dyskaryosis and one borderline smear test, but no treatment had been carried out and the patient had been discharged.

Past medical history was not contributory to her illness at the time.

INVESTIGATIONS

On examination, the patient had a blood pressure of 130/80 and a pulse of 100 bpm. She was well orientated, afebrile and had no pallor. She had some lower abdominal discomfort with more tenderness at the left iliac fossa, but there was no rebound or guarding.

Blood tests were carried out in the form of full blood count, β -human chorionic gonadotropin (β -HCG) and progesterone.

Hb was 13.0 g/l, β -HCG was 3352 U/l and progesterone was 21 nmol/l.

A decision was made to have an ultrasound scan performed in the early pregnancy assessment unit when the first available scan slot was available (2 days later).

The patient was advised to seek medical advice if she had worsening symptoms and was discharged from A&E with simple analgesia.

She was reviewed 2 days later as planned and her pain at the time was getting worse.

The ultrasound scan failed to identify an intrauterine pregnancy, while the endometrial thickness was measured at 19.1 mm. There was a small 2.5 cm mass adjacent to the left ovary and there was also some free fluid in the pouch of Douglas.

A repeat β -HCG measurement came back as 2994 U/l and Hb was 13.3 g/l.

DIFFERENTIAL DIAGNOSIS

Ectopic pregnancy (until proven otherwise).

TREATMENT

A diagnostic laparoscopy was performed the same day.

During the procedure 500 ml of blood were found in the abdomen while a left ectopic pregnancy which was bleeding was easily identified.

The laparoscopy was abandoned due to technical difficulties and a mini laparotomy with left partial salpingectomy was carried out without complications.

OUTCOME AND FOLLOW-UP

The patient made an uneventful recovery and was discharged home 2 days after the procedure.

The histology report confirmed the presence of a complete molar pregnancy.

'Clearly abnormal chorionic villi with focally hydropic change, collapsed vessels in villous stroma which also shows abundant nuclear debris in well preserved villi with focal myxoid stromal change and there is moderate abnormal circumferential trophoblast hyperplasia with sheets of pleomorphic extravillous trophoblast'.

The patient was registered with one of the three suprar-egional centres in the UK for follow-up of gestational trophoblastic disease.

She was reviewed in the gynaecology clinic 6 weeks later when she was found to be making an excellent recovery. The events surrounding her hospital admission were revisited and she was advised to continue her follow-up with the registration centre.

She had initially falling levels of HCG until they started rising up again. A pelvic ultrasound confirmed the presence of a viable intrauterine pregnancy 4 months after the diagnosis of the molar pregnancy.

DISCUSSION

Hydatidiform moles arise as a result of the fertilisation of an abnormal ovum. Complete moles are entirely paternal in origin and arise either from the fertilisation of an 'empty' ovum by a haploid spermatozoon that subsequently duplicates or from the fertilisation of an 'empty' ovum by a diploid spermatozoon.^{1 2}

The incidence of ectopic pregnancy is approximately 1% of all pregnancies in the UK.¹ The incidence of hydatidiform mole varies worldwide from 1 in 100 in Southeast Asia to 1 in 2000 in the USA.³ The combined incidence therefore of a molar ectopic pregnancy should be somewhere between 1 in 10 000 and 1 in 200 000 pregnancies. In the UK, however, the incidence of a true molar ectopic pregnancy is approximately 1.5 in 1 000 000 births as documented in a 15-year study in one of the three Trophoblastic Disease Screening and Treatment Centres in the UK.¹

There are few confirmed cases of tubal ectopic pregnancies in the world literature – with 40 being the latest estimation^{2 4–6} – as many of the earlier thought tubal moles are not universally accepted as 'true' ones since they do not conform to standard pathological criteria used to reach the diagnosis.^{4 7} It is widely believed that some of the earlier documented 'molar pregnancies' have actually been non-molar hydropic abortions. Therefore, diagnosis depends on the correlation of clinical and histological features which can be complimented by ploidy evaluation through DNA flow cytometric analysis.^{2 8}

It is now accepted that patients with ectopic molar pregnancies are clinically indistinguishable from patients with 'traditional' tubal pregnancies, with the possible exception of higher tendency of rupture at the time of presentation.^{1 2}

The management of ectopic molar pregnancies consists of surgically removing the conceptus with no obvious preference for laparoscopy or laparotomy provided that the whole trophoblast is removed.^{1 9}

Learning points

- Molar pregnancies can present outside the uterus and histological diagnosis of the surgically removed conceptus is mandatory.
- The incidence of ectopic gestational trophoblastic disease in the UK is around 1.5 per 1 000 000 births.
- Suspected molar ectopic pregnancies are usually proved not to be true moles after close scrutiny.
- The presenting features of molar ectopic pregnancies are indistinguishable to those of non-molar ectopic pregnancies.
- Appropriate follow-up with a supraregional centre is of paramount importance since inappropriately treated molar pregnancies may progress to choriocarcinoma.

The histological examination of the surgically removed conceptus in ectopic pregnancies is essential for appropriate follow-up to be arranged (which includes registration with an appropriate centre).

The prognosis of ectopic molar pregnancies is the same as for other forms of gestational trophoblastic disease.¹

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Competing interests None.

Patient consent Obtained.

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