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A Case of Heterotopic Intrauterine Complete Molar Pregnancy and Tubal Ectopic Pregnancy

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Abstract

Introduction: We describe clinical case of a woman who presented with a very rare diagnosis of heterotopic pregnancy with a coincident finding of intrauterine complete molar pregnancy and a tubal unruptured ectopic pregnancy. She was asymptomatic at the first presentation with features of a vesicular mole in the uterus and high suspicion of ectopic pregnancy on the left side of the uterus by transvaginal ultrasound scan (TVS). The serum Beta Human Chorionic Gonadotrophin (HCG) was 96915 U/L which was significantly high. She also had mild anemia. It was challenging to offer the best of treatment modalities as we did not find a similar case in the literature.

Outcome: After full counseling, the patient underwent suction curettage of the uterus and medical treatment with Methotrexate. The serum BHCG level was monitored and it reached zero in a few weeks. She made a good recovery without any complication.

Conclusion: Surgical evacuation for intrauterine complete mole followed by medical methotrexate therapy for coincident unruptured tubal ectopic pregnancy is optimal management for such cases.

Keywords: Ectopic; Molar; Ultrasound; Heterotopic pregnancy; Methotrexate therapy

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Introduction

Hydatidiform molar change is mainly due to genetically abnormal conceptions, in which an excess of paternally derived genetic material results in abnormal fetoplacental embryogenesis and placental villous trophoblast hyperplastic changes. Complete hydatidiform mole harbors a higher risk of invasive trophoblastic disease. Clinical factors that have been associated with the risk of malignant changes are advanced maternal age, high levels of hCG (>100,000 mIU/mL), eclampsia, hyperthyroidism, and bilateral theca lutein cysts [1]. Hydatidiform molar pregnancy and ectopic pregnancy are two uncommon, however, clinically significant complications of pregnancy. The simultaneous presence of these two abnormal pregnancies has been reported to occur but with very rare reporting [2,3]. The incidence of ectopic pregnancy by itself occurs at a rate of 12.3 per 1000 live births and seems the numbers are increasing [4] and is a leading cause of maternal mortality in the first trimester.

On other hand, the overall molar pregnancy incidence is around 1 for every 607 conceptions (complete mole 1:1,423; partial mole 1:1,058), but with major variations with age [5]. For complete molar pregnancy, the risk varied from < 1:1,000 for ages 18-40, to 1:156 for women aged 45 and 1:8 for those aged 50 and above. Moreover, heterotopic pregnancy, the coexistence of intrauterine and ectopic pregnancies, occurs in one of 2600 pregnancies in the United States [6]. In natural conceptions, the incidence of heterotopic pregnancy has been estimated to be 1 in 30 000 pregnancies [7], while with fertility treatments, the incidence of heterotopic pregnancy increases to as high as 3% [8]. This case report describes a patient with an intrauterine complete molar pregnancy with coexistent tubal ectopic pregnancy confirmed by examination, imaging, and pathologic analysis.

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Case Report

A 49-year-old multiparous woman (G6p3A2, all spontaneous vaginal deliveries, one dilatation, and curettage), presented to GYN consultation for a missed period with no symptoms and with approximately 10 weeks gestation. It was an unplanned pregnancy. She had a history of irregular menstruation. She did not use contraception). She is known to have Pituitary Microadenoma with hyperprolactinemia for which she was taking Dostinex 0.25 mg twice per month. Her BMI was >30She is allergic to penicillin. The diagnosis of molar pregnancy was made based on the ultrasound findings of snowstorm appearance and a high BHCG level of 87000 U/L. There were additional ultrasound suggestive features of ectopic pregnancy. Upon arrival, she had no pain and no bleeding. Her examination revealed: afebrile with a heart rate of 78 beats/min, blood pressure of 116/75 mm Hg. Abdominal examination revealed soft lax with no tenderness or masses with no guarding and rebound tenderness. The pelvic examination was remarkable for red blood at the closed cervical OS, minimal cervical tenderness, and no appreciable adnexal mass or fullness.

Lab on admission 15/9/20: Hb 9.9, BHCG 96.915 U/L. TVS report: Urinary bladder is empty. The uterus is Anteverted and bulky, measures 11.3 × 8.2 × 6.9 cm. Myometrium shows a homogenous echotexture. No focal lesions were noted. Endomyometrial differentiation maintained. The endometrium is grossly thickened measures 45 mm. The uterine cavity is distended and filled with multiple small cystic structures. No evidence of intrauterine gestational sac or fetal parts was seen, both ovaries are normal in size, normal echogenicity. The right ovary measures 21 × 12.9 mm. Left ovary measures 22 × 16 × 14 mm, 2.7 mL in volume.

A corpus luteum is seen in the left ovary measuring 18 × 13 mm. There is a left adnexal mass lesion adjacent but separate from the left ovary measuring 13 × 13 mm with a well-defined anechoic cystic structure within represents gestational sac measuring 7 mm corresponding to 5 weeks 3 days. No evidence of the yolk sac or fetal pole was seen within this cystic structure in the present study. There is a small clear cyst in the left adnexa separate from the left ovary measuring 10.6 × 7.9 mm. No fluid in Pouch of Douglas present. Conclusion: 1. Bulky uterus, 2. Thickened endometrium with multiple cystic structures within--findings is in favor of complete hydatiform mole, 3. Left adnexal mass as described raise suspicion of left adnexal ectopic gestation, 4. A small left adnexal clear cyst **(Figure 1).** A diagnosis of heterotrophic pregnancy- molar pregnancy with ectopic was made, which is very rare. The patient was counseled about the management of molar pregnancy as suction curettage and for ectopic pregnancy either laparoscopic salpingectomy or expectant management with Methotrexate therapy with serial HCG measurements. The patient and her husband chose to go with the second option of suction curettage followed by MTX therapy and expectant management. laparoscopy and wanted only suction curettage for suspected molar pregnancy, despite knowing the risks associated with ectopic pregnancy including rupture and massive intraperitoneal bleeding. The refusal form has been signed. Suction curettage was done with no complications, macroscopic examination confirmed molar pregnancy. She received Methotrexate 91.5 mg in IM after checking for liver function test which was within normal limits.

On 17/9/20: WBC 10.34 K/uL, RBC 4.75 M/uL, Hemoglobin: 9.6 g/dL, Hematocrit: 32.50 L, MCV: 68.40 fL, MCH: 20.20 pg, MCHC: 29.50 g/dL, RDW: 17.00%, Platelet: 448.00 K/uL, LFT: AST 20 U/L, and ALT 24 U/L, blood group O positive. Two days after the BHCG was 42.487 U/L, the patient was stable and was discharged. On the follow-up visit on 19/9/20: BHCG was 6.922 U/L. On day 4 post Methotrexate on the 21/9/20: BHCG 2.151 U/L and the patient had no complaints, after 30 days the β-HCG levels become 0 **(Figure 2).** A follow-up TVS showed: Anteverted uterus measures 8.0 × 4.9 × 5.6 cm. Inhomogeneous myometrial echotexture but no definite focal lesion is seen. Endometrial thickness of 0.33 cm. Rt ovary = 2.56 × 1.61 × 2.05 cm. Lt ovary = 2.82 × 1.42 × 1.59 cm, adjacent to it there is a hypoechoic cystic lesion approximately measures 1.14 × 0.77 × 0.95 cm, most probably par-ovarian cyst. No vascularity was seen. Multiple Nabothian cysts were seen at the cervix. No free fluid was seen at the POD **(Figure 3).**

Discussion

This case report is presented because it demonstrates the simultaneous occurrence of two rare clinical diagnoses of pregnancy in the same patient. The incidence of heterotopic pregnancy with a coexistent molar and ectopic pregnancy is

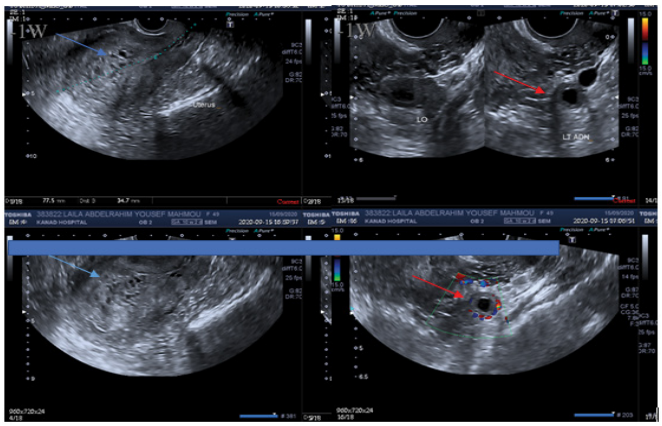

Figure 1 TVS showing snowstorm appearance representing molar pregnancy (blue arrow) and tubal ectopic pregnancy (as a 'tubal ring' or 'bagel sign') (red arrow).

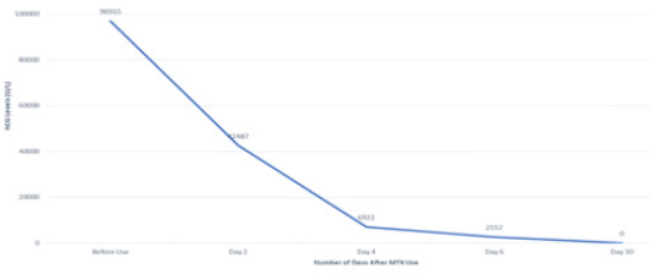

Figure 2 β-HCG levels before and after evacuation curettage & Methotrexate therapy.

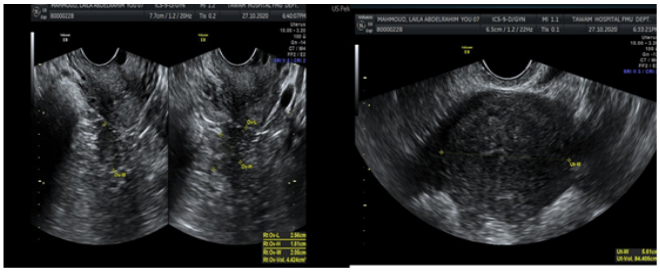

Figure 3 Follow up TVS with no evidence of neither Molar nor ectopic pregnancy.

extremely rare, and the true incidence of this rare occurrence is unknown. To our knowledge, this is the only case to be reported. There are two case reports [2,3] in the literature of a combined ectopic tubal pregnancy along with a partial intrauterine molar pregnancy, and several reported cases of a concurrent intrauterine pregnancy with heterotopic molar pregnancies [9].

The decision not to do laparoscopy for the ectopic was made based on the following facts: small diameter of the ectopic gestational sac, no fetal heart detected in the ectopic and the fact that molar pregnancies are associated with a higher risk for bleeding as factors released by the molar tissue could trigger the coagulation cascade and patients should be monitored for disseminated intravascular coagulopathy (DIC) and trophoblastic embolism which can cause acute respiratory insufficiency [10]. However, with Methotrexate (MTX) the trend of beta HCG and response to MTX would be very difficult to interpret with an existing molar pregnancy, and she will still have the risk of failed medical management may still need surgical intervention; if ectopic the risk of rupture is high which is life-threatening.

One can challenge that in our case there was no ectopic pregnancy at all as no histological and endoscopic proof of ectopic; however, we are confident enough that there was an ectopic due to the following reasons: the scan was done three times before the procedure by three different expert sonographers who suggested with high confidence the diagnosis of ectopic pregnancy based on the typical finding in ectopic of a left adnexal mass lesion adjacent but separate from left ovary measuring 13 × 13 mm with a well-defined anechoic cystic structure within represents gestational sac measuring 7 mm corresponding to 5 weeks 3 days. Moreover, TVS sensitivity in detecting ectopic pregnancy reaches up to 93% and is dependent on gestational age and operator expertise [11]. Nonetheless, transvaginal ultrasonography can identify two-thirds of EPs before rupture [12] and finally, the disappearance of the left adnexal mass after the therapy. In a complete mole, the level of serum beta HCG is often abnormally elevated, and the patient, in this case, had a beta HCG of 96.915 U/L that exceeded the expected level for gestational age of 10 weeks as calculated by her last menstrual period.

The management of a complete molar pregnancy includes close follow-up because 20% of patients with a complete mole may have the residual disease even after D&C treatment. The ultimate diagnoses of both an intrauterine molar pregnancy and an ectopic pregnancy demonstrate the importance of appropriate vigilance for the presence of ectopic pregnancy even in the setting of other uncommon abnormalities of pregnancy.

Conclusion

In conclusion, in such cases which are extremely rare a conservative management of ectopic pregnancy combined with the surgical evacuation of mole pregnancy and proper patient's counseling is the optimal management plan.

Acknowledgement

We thank the patient for permitting us to publish her case.

Disclosures

The authors declare no conflict of interest and there was no violation of human rights and we gained written informed consent from patient. This article does not contain any studies with human and animal subjects performed by the any of the authors.

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