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Case Report

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HYDATIDIFORM MOLE WITH THECA LUTEIN CYSTS OR HYPERREACTIO LUTEINALIS, A CASE REPORT

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INTRODUCTION

Hydatidiform mole with hyperreactio luteinalis is a rare case and can be misdiagnosed without adequate clinical history. In hydatidiform mole, elevated human chorionic gonadotropin (hCG) hormone levels are found. This can lead to hyperractio luteinalis, which is characterized by enlargement of both the ovaries with mulitple thin walled cysts. These can be easily mistaken to be malignant pathologies. A good clinical history and knowledge are required to avoid mistake.

CLINICAL REPORT

Mrs EE, a 26 year-old pregnant female from India.

The Patient (G2P1), presented to Ob and Gyn doctor with complaints of mild vaginal spotting accompanied by increasing abdominal pain and reduced urine output since 2-3 week. She also had severe nausea and vomiting as compared to last pregnancy. Her at home UPT was positive. She was married for 5 years. The Patient didn't have any significant past history of major illness or surgery. She denied any history of taking any medication for infertility or PCOD. She was referred to our centre by the Ob and Gyn Doctor as a case of acute abdomen in pregnancy for an ultrasonography of abdomen and pelvis to confirm pregnancy and to rule out causes of abdominal pain and reduced urine output.

Imaging Findings

The patient had refused for transvaginal ultrasonography (USG) so transabdominal USG of pelvis was then performed. This demonstrated an enlarged uterus with 12.35 x 5.6 x 11 cm (vol 455.15ml) sized multiple small cystic lesions having a cluster of grapes appearance within the endometrium without any definite fetal parts. (Fig.1 and 2). This extended from the fundus of the uterus to the upper cervix.

USG evaluation of both the adnexa revealed large bilateral multilocular cystic masses measuring 9.65 × 4.32 × 8.13 cm (vol 177 ml) in the right ovary and 8.65 × 3.93 × 8.36 cm (vol 148 ml) in the left ovary. Both the masses contained more than 10 locules with rather thick (>3 mm) septa and anechoic cyst fluid. The cyst wall was regular and thin. (Fig. 3 and 4). Normal arterial and venous doppler vascularity was noted in both (Fig. 5). No obvious evidence of ovarian torsion in presence scan.

USG of the rest of the abdomen was within normal limits and no ascites was noted. USG and X - ray examination of the chest was within normal limits and no pleural effusion was noted.

Differential Diagnosis List

- Theca lutein cysts or Hyperreactio luteinalis
- Ovarian hyperstimulation syndrome (OHSS)
- Polyscystic ovaries
- Ovarian cystic neoplasms

Diagnosis

Hydatidiform mole with theca lutein cysts or hyperreactio luteinalis.

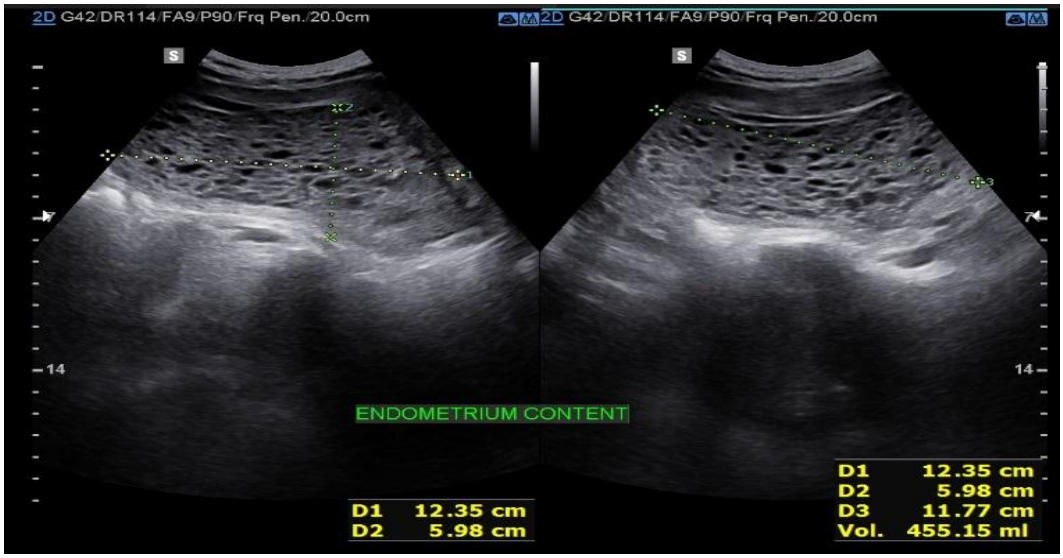

Fig. 1: USG shows enlarged uterus with multiple grape-like clusters mass lesions.

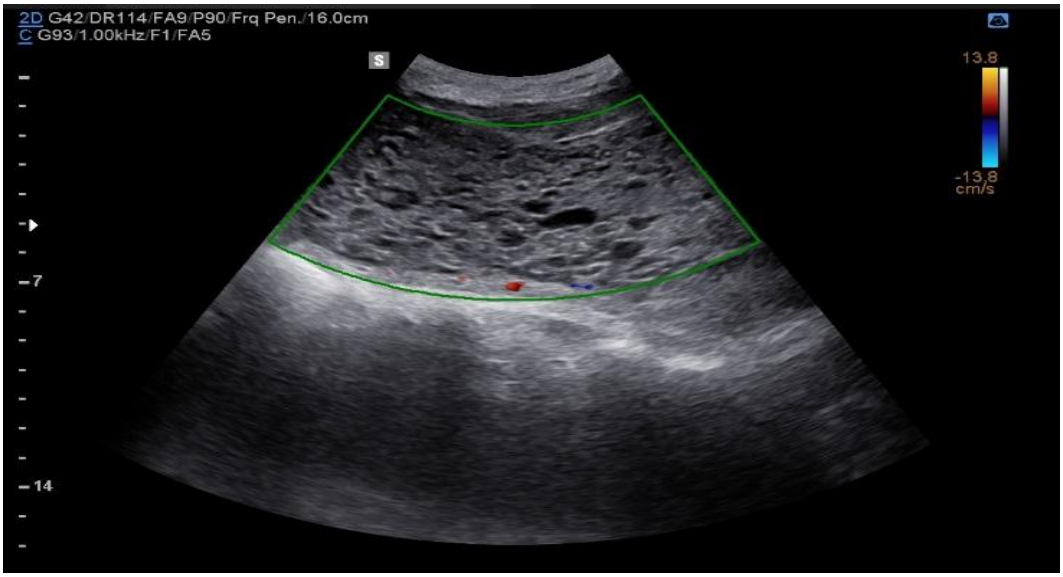

Fig. 2: A transabdominal USG shows enlarged uterus with multiple grape-like clusters mass lesions.

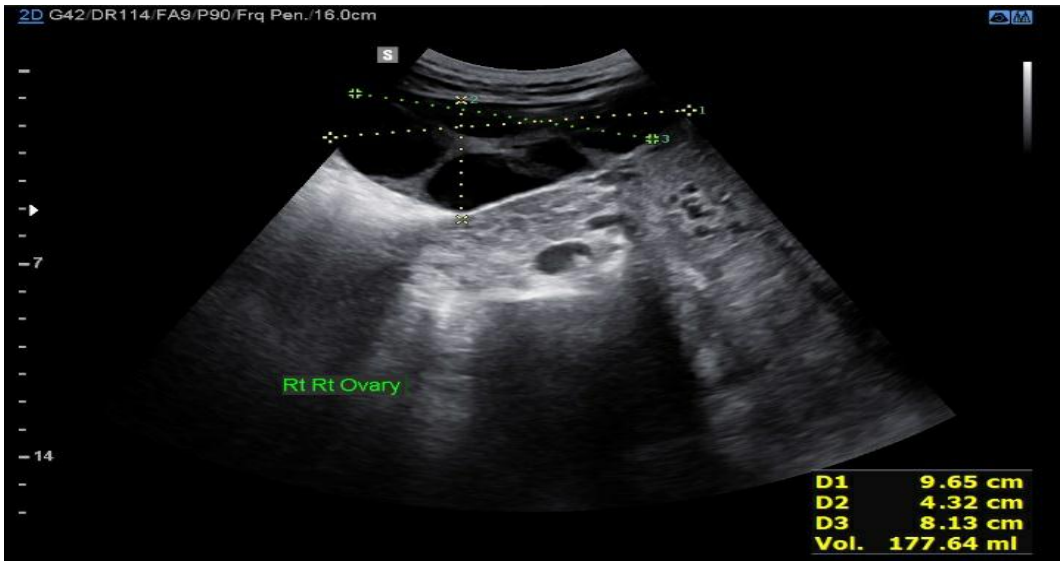

Fig. 3: USG shows enlarged right ovary with multilocular masses, thick septa and anechoic cyst fluid. The cyst wall are regular and thin.

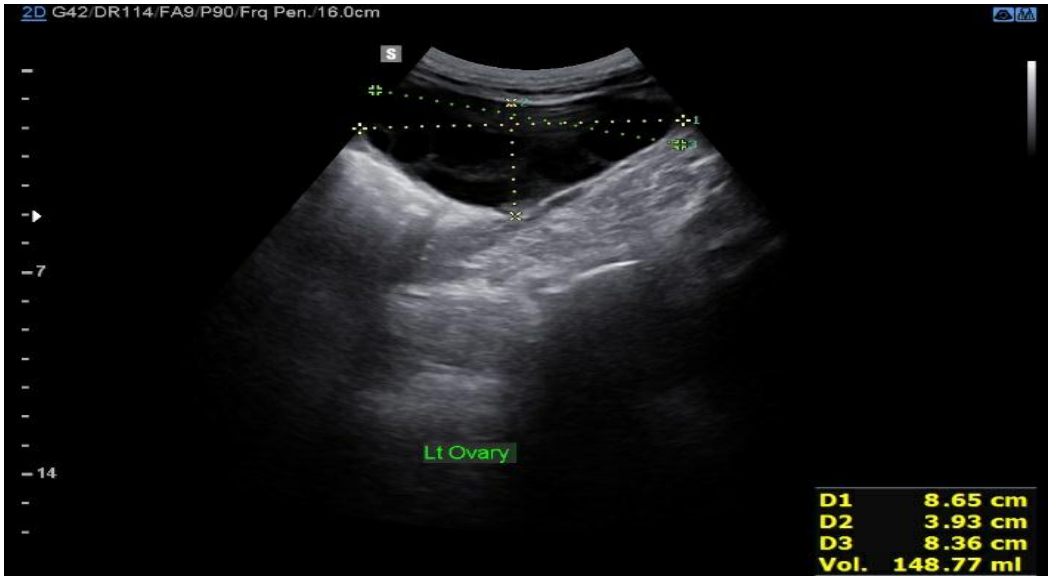

Fig. 4: USG shows enlarged left ovary with multilocular masses, thick septa and anechoic cyst fluid. The cyst wall are regular and thin.

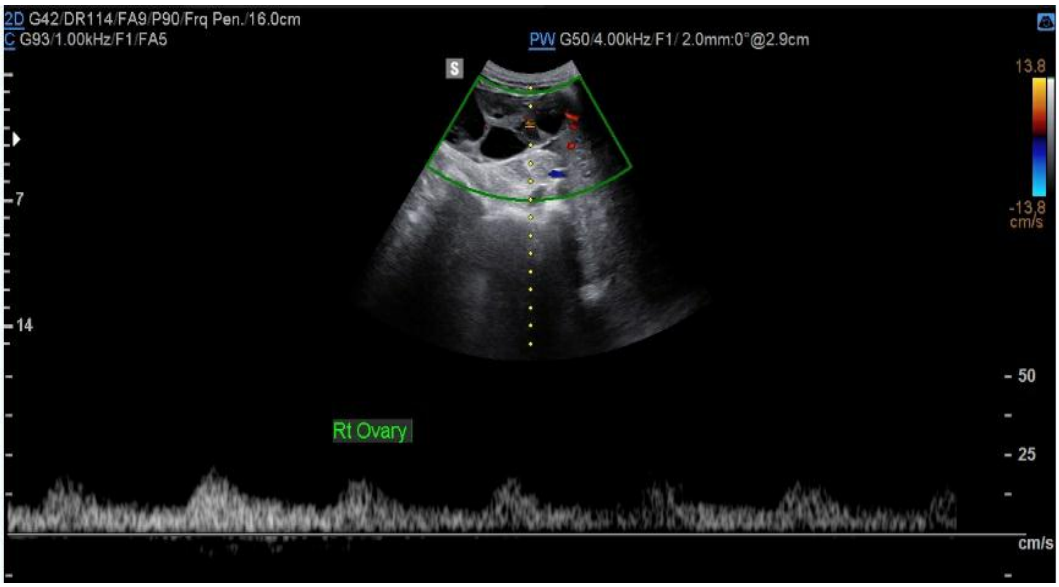

Fig. 5: Normal arterial and venous doppler vascularity was noted in right ovary.

DISCUSSION

This is a rare case of Hydatidiform mole with theca lutein cysts or hyperreactio luteinalis. Hydatidiform mole is part of a spectrum of gestational trophoblastic disease, which involves the abnormal fertilization of maternal ovum by spermatozoa that can range from a benign to an invasive condition. The hydatidiform mole can be partial (69 XXX or XXY, containing fetal tissue), or complete (46 XX or XY, both derived from paternal chromosomes with a lack of fetal tissue).1 Molar pregnancy is more common in extremes of reproductive age.[2]

Vaginal bleeding tends to be the most common symptom of a molar pregnancy. Quantitative beta-hCG levels higher than 100,000mlU/mL should raise suspicion for a molar pregnancy. However, molar pregnancy with normal beta-hCG levels can exist.[3,4,5]

USG is the standard imaging modality for identifying molar pregnancy. Classically, a 'snowstorm pattern' has been described, resulting from the presence of a complex vesicular intrauterine mass containing many 'grape-like' cysts. USG evaluation of the adnexa can also reveal theca lutein cysts, due to ovarian stimulation by abnormally elevated beta-hCG levels.[6]

Work up of a molar pregnancy includes obtaining a chest radiograph, a complete blood count, liver panel, thyroid function tests, coagulation studies, blood type and urinalysis.[3,6,7]

This case demonstrates the utility of USG in the evaluation of the early pregnant patient presenting with vaginal bleeding.

Hyperreactio luteinalis is a rare condition that can occur at any stage of pregnancy, but is typically seen in the third trimester.[1] In almost all cases it is triggered by very high endogenous or exogenous β-hCG stimulation.[2] Therefore most publications report on the presence of hyperreactio luteinalis in a multiple or molar pregnancy, in association with choriocarcinoma and fetal hydrops, or after fertility treatment. An abnormally rapid rise in β-hCG in the first trimester or abnormal sensitivity of the hCG receptor due to a gene mutation can lead to the exceptional case of hyperreactio luteinalis in a spontaneous singleton pregnancy.[3] Burger described the first case of hyperreactio luteinalis not associated with trophoblastic disease, since when a few cases have been reported in spontaneous singleton pregnancies.[4–6] Depending on the size of the masses either patients are asymptomatic or they present with pain due to intraabdominal pressure, torsion or intracystic hemorrhage. Virilization due to hyperandrogenism can occur in as many as 25% of affected patients.[7,8] Symptoms of hyperemesis gravidarum or hyperthyroidism have been reported and are usually not related to hyperreactio luteinalis but are provoked by the underlying problem that is causing the high β-hCG level (trophoblastic disease, multiple pregnancy). The prognosis of this benign condition is good and in the postpartum period theca lutein cysts usually regress spontaneously.[7]

On USG hyperreactio luteinalis is characterized by large adnexal masses that consist of many thin-walled small theca lutein cysts, giving it the appearance of a 'spoke wheel'. Ascites can be present. Owing to their large size and morphology they are hard to distinguish from ovarian hyperstimulation syndrome (OHSS).[4,5,7,8,10] OHSS almost exclusively occurs in patients following fertility treatment although, very rarely, it can occur in spontaneous singleton pregnancies. By contrast with hyperreactio luteinalis, OHSS presents at the beginning of the first trimester and is associated with more severe symptoms involving acute fluid imbalances that impair the natural pregnancy course. With their large number of locules, hyperreactio luteinalis can even mimic a malignancy, in particular a mucinous borderline tumor of the intestinal type, and lead to unnecessary surgery.[8] However, when compared with hyperreactio luteinalis, mucinous intestinal borderline tumors have smaller thin-walled locules that are not as round and tend to have less solid tissue than in cases of hyperreactio luteinalis.

Contrast-enhanced computed tomography (CECT) or magnetic resonance imaging (MRI) also shows similar findings but is rarely performed, except when it is done to rule out other causes of complex cystic ovarian lesions.[8]

Bilateral, symmetrically enlarged ovaries containing multiple variably sized cystic lesions ("spoke wheel" appearance) representing enlarged follicles or corpus luteum cysts in the presence of ascites are the typical imaging findings.[9] Corpus luteum cysts and ascites can be echogenic on US or dense on CT from hemorrhage. Additional imaging findings that can be seen on US, CT or MRI include pleural effusion and thromboembolism.[10]

CONCULSION

In summary, our case report on hyperrreactio luteinalis, which is a benign condition causing enlargement of the ovaries. Recognition of this is important, since misinterpretation has resulted in unnecessary surgery, often with sterilization. Follow-up with serial Beta-hCG is crucial in managing these cases. Complications are by rupture, torsion or hemorrhage, so careful monitoring is required.

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