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Hyperreactio luteinalis in a monochorionic twin pregnancy complicated by preeclampsia: A case report

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ABSTRACT

Hyperreactio luteinalis (HL) is a rare benign complication of pregnancy that is characterized by progressive ovarian enlargement and hyperandrogenism. We present a case of a 30-year-old woman with a spontaneous monochorionic diamniotic twin pregnancy who presented with early-onset preeclampsia, concern about possible twin-twin transfusion syndrome, and bilateral enlarged ovarian masses. Both ovaries had multiple thin-walled unilocular cysts; one ovary measured 17.9 × 17.5 × 9.1 cm and the other 12.5 × 11 × 12.3 cm. After extensive counseling, the patient underwent an uncomplicated dilation and evacuation. Postoperative assessment indicated elevated androgen levels, which spontaneously resolved, supporting the clinical diagnosis of HL. It is important to consider HL in the differential diagnosis of adnexal masses in pregnancy. HL spontaneously regresses after delivery and is managed expectantly. HL has been associated with gestational trophoblastic disease, multiple gestations, preeclampsia, and twin-twin transfusion syndrome.

1. Introduction

The incidence of ovarian masses in pregnancy is rare, complicating 1–2% of all pregnancies [1]. The differential diagnosis is vast and some masses require intervention during pregnancy. Hyperreactio luteinalis (HL) is a rare but benign condition. Its hallmarks include bilateral, marked cystic enlargement of the ovaries with associated overproduction of ovarian androgens. We present a case of a patient with a monochorionic diamniotic twin pregnancy, fetal anomalies, early-onset preeclampsia, concern for twin-to-twin transfusion syndrome (TTTS) and enlarging ovarian masses. The uniqueness of this case is the simultaneous association of TTTS, HL and preeclampsia. Our review of the literature suggested that a trifecta of this nature has not been previously reported.

2. Clinical Case Report

A 30-year-old woman, gravida 2 para 1001, presented for ultrasound assessment of her spontaneous monochorionic diamniotic twin pregnancy. She had had a previous uncomplicated term pregnancy and no significant medical history. At 16 weeks' gestation, a fetal anatomy evaluation indicated no apparent anomalies and normal bilateral adnexa. Two weeks later, an ultrasound showed twin A with a large ventricular septal defect but normal amniotic fluid volume; an umbilical artery Doppler study (UAD) was also normal. Twin B had bilateral ventriculomegaly with a non-visualized corpus callosum and oligohydramnios; UAD showed absent end diastolic flow (AEDF). There was 41% discordance between the twins' estimated fetal weights based on biometric parameters. The patient was counseled about the possibility of twin-twin transfusion syndrome (TTTS). A right complex ovarian mass measuring 7 × 5.7 × 7.5 cm (Fig. 1) was noted by the uterine fundus. To rule out ovarian malignancy, a cancer antigen125 was drawn and was normal (<35/value). The beta human chorionic gonadotropin level was >200,000. She had a normal complete blood count and liver enzymes. A basic metabolic panel was within normal limits except for mild hyponatremia at 130 mEq/L.

At 19 weeks' estimated gestational age, the patient complained of increasing abdominal pain. Given new-onset pain, ultrasound and MRI were performed for evaluation and gynecology oncology consultation was planned. The ovaries were markedly enlarged, with numerous small follicular cysts (right adnexa 6.8 × 10.1 × 9.4 cm and left adnexa 7 × 12 × 10cm) and concurrent ascites was noted. Twin A had normal amniotic fluid volume and UAD. Twin B had anhydramnios, a non-filling bladder, and persistent AEDF. At this point the patient declined intervention despite being counseled that the prognosis was dire.

The patient returned four days later with shortness of breath and lower-extremity edema. She was found to have new-onset hypertension but her preeclampsia work-up was only significant for proteinuria

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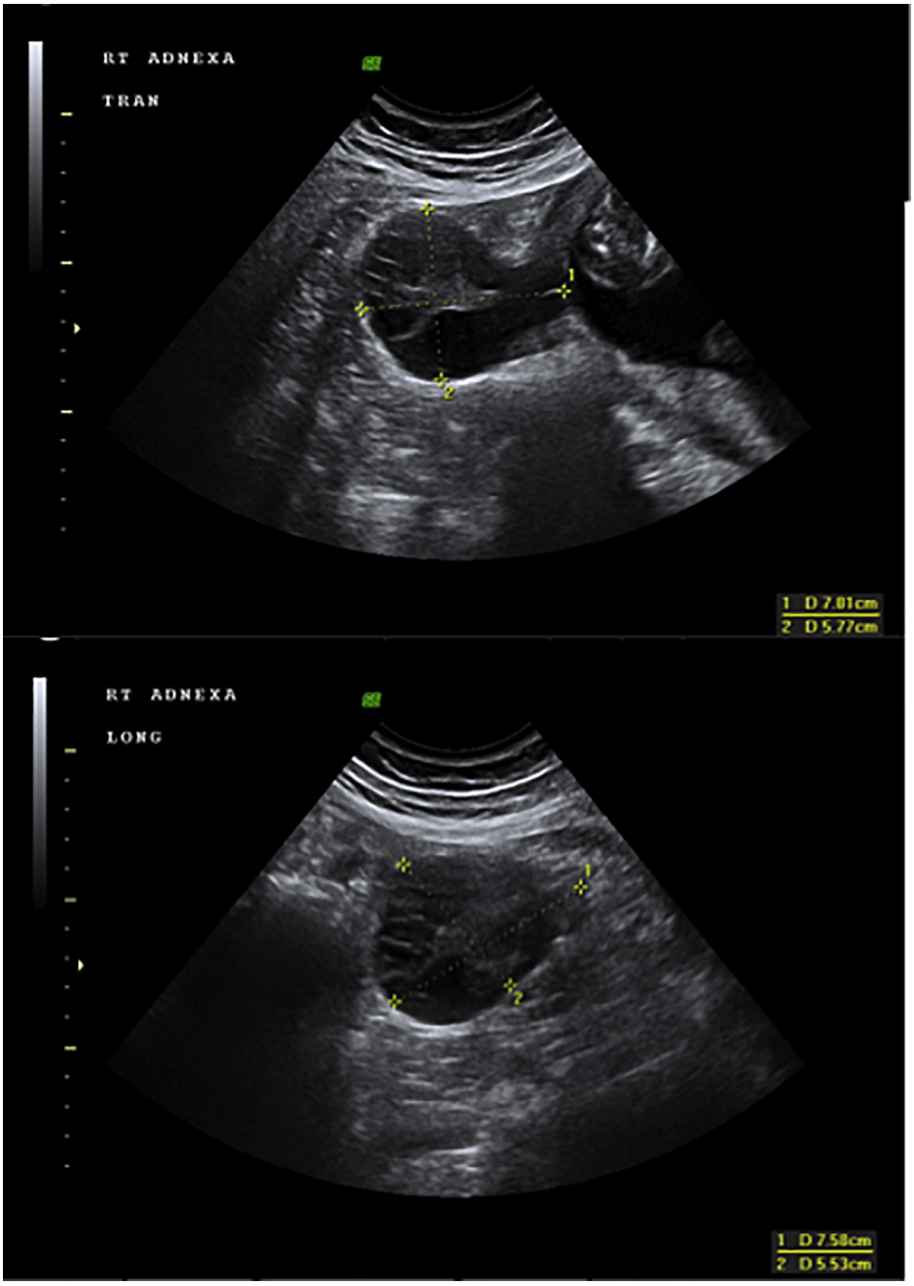

Fig. 1. Ultrasound scan of the right ovary at 18 weeks' gestation. Upper image: Transverse plane. Lower Image: Longitudinal plane.

(1958 mg/24 h). Due to persistent hypertension, she met diagnostic criteria for preeclampsia without severe features. After extensive counseling about her evolving preeclampsia and risk of respiratory distress due to abdominal distention, and in light of the fetal anomalies and TTTS, the patient elected for termination of pregnancy via dilation and evacuation (D&E). At 19 5/7 weeks, a standard D&E was performed under ultrasound guidance without complications. Pathology examination confirmed TTTS characterized by 65% discordant biometrics, marked pallor in the donor twin and plethoric recipient twin. Both fetuses were male. Placenta pathology was consistent with a monochorionic-dichorionic immature placenta, notably with abnormal anastomosis consistent with TTTS.

On postoperative day one, she was normotensive and complained only of abdominal fullness. On ultrasound, markedly enlarged ovaries were noted, the left measuring 17.9 × 17.5 × 9.1 cm and the right 12.5 × 11 × 12.3 cm (Fig. 2). The differential diagnoses included ovarian hyperstimulation syndrome (OHSS) and hyperreactio luteinalis (HL). An androgen panel revealed elevated levels of androstenedione at 15.9 ng/mL (0.26–2.14 ng/mL) and testosterone at 11.78 ng/mL (<0.2–0.73 ng/mL), but normal didehydroepiandrosterone (DHEA) at 1.79 ng/mL (1.33–7.78 ng/mL). She did not exhibit any maternal virilization. Given her characteristic ultrasound findings and lack of biochemical features supporting OHHS, our final diagnosis was HL. The decision was made to manage her course expectantly. The androgen panel was repeated one month later and showed a decline in androstenedione to 9.62 ng/mL (0.3–2.5 ng/mL) and testosterone to 2.84 ng/mL (0.02–0.45 ng/mL). The patient was asymptomatic and no longer complained of abdominal fullness.

3. Discussion

The differential diagnoses of an ovarian mass in pregnancy may include corpus luteum cyst, follicular cyst, endometrioma, malignancy, luteoma and OHSS. OHSS and HL have similar ultrasound