Vaginal delivery in a spontaneously conceived singleton pregnancy complicated with hyperreactio luteinalis: A case report

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Hyperreactio luteinalis (HL) is a rare entity caused by presence of numerous luteinized follicle cysts resulting in variable degrees of ovarian enlargement.1 The etiology of this condition is unknown, but it is believed to be caused by either prolonged or excessive stimulation of the ovaries by gonadotropins or increased ovarian sensitivity to gonadotropins.2 It is known to be typically associated with gestational trophoblastic disease and ovarian stimulation for ovulation. Association with hyperemesis gravidarum, multiple pregnancy, and fetal hydrops has also been reported. However, it is very rare in a spontaneously conceived normal singleton pregnancy.1,2

HL sometimes is revealed accidentally at the time of routine pelvic ultrasound examination because patients with HL may be asymptomatic. Unnecessary laparotomy during pregnancy can be performed by physicians because of its rarity and unfamiliarity. Since Burger3 described the first case of HL unassociated with trophoblastic disease in 1938, about 80 reports have been published. Still, only sporadic cases of HL, especially conditions managed conservatively, have been reported. We present a case of HL managed non-surgically resulting in successful vaginal delivery.

Case Report

A 29-year-old woman, gravida 1, para 0, was admitted to the hospital complaining discomfort of left lower quadrant of the abdomen. She was referred from a local clinic with diagnosis of benign ovarian tumor. She had no history of diabetes mellitus, hypertension, and chronic renal failure. She...
had no signs of hyperandrogenism or virilization. The patient had never taken drugs for ovulation induction. Ultrasound showed a singleton pregnancy at 9 weeks with bilateral ovarian cysts of diameters reaching 6×4×7 cm (right ovary) and 8×7×4 cm (left ovary) without ascites (Fig. 1A).

Laboratory results of serum levels at 9 weeks of gestation were as followings: hematocrit 39.1%, liver enzymes and coagulation profile, normal; β-hCG, 220,000 mU/mL; CA-125, 65.4U/mL; luteinizing hormone, 2.3 mIU/mL; follicle-stimulating hormone (FSH), 0.6 mIU/mL; thyroid-stimulating hormone 3.1 μIU/mL (normal, 0.4-4.5); free thyroxin 0.44 ng/dL (normal, 0.7-2); blood urea nitrogen (BUN) 7.3 mg/dL (normal, 8-23); creatine 0.5 mg/dL (normal, 0.5-1.3). Laboratory findings of glucose, BUN, and creatine serum level throughout the admission were within normal limit ranging from 69-96 mg/dL, 6.1-7.3 mg/dL, and 0.5 mg/dL respectively. Her blood pressure was stable through admission days as well, ranging from 100/70-120/80 mm Hg.

This case showed a very unusual clinical course. After the first detection of HL at 9 weeks of gestation, both ovarian cysts were increased in size week by week. At 15 weeks of gestation the patient complained abdominal discomfort (left ovary: 18×8×18 cm), so 160 mL of fluid was aspirated from left-sided ovarian cyst with disappearance of her discomfort. But even after then the size of both ovarian cysts was increased again week by week. At 31 weeks of gestation the size increased to 12×9×8 cm (right ovary) and 28×12×22 cm (left ovary). Three thousand and five hundred mL of fluid was aspirated through abdominal needle (19 G) due to marked abdominal discomfort and rupture risk of the huge cysts (Fig. 1B). Both ovarian cysts were increased in size again until 36 weeks of gestation, after then the size was decreased gradually. At 40 weeks of gestation, a healthy female infant weighing 2,910 g was delivered vaginally. The ovarian cysts were decreased in size markedly at 16 days postpartum and normal-sized ovaries and normal value of serum β-hCG were observed at 55 days (7 weeks) postpartum.

**Discussion**

This case is meaningful in the aspect of the fact that it is one of few reports of hyperreactio luteinalis occurring in a spontaneously conceived pregnancy, and moreover, was managed expectantly throughout complete gestation followed by successful vaginal delivery. In this case, the ultrasonography revealed a ‘spoke wheel’ appearance from the first trimester of pregnancy without iatrogenic induction of ovulation. On the basis of the ultrasonographic finding of large simple thin-walled cysts, along with intensely compressed stroma with physiological vasculature, differential diagnosis was spontaneous ovarian hyperstimulation syndrome (OHSS) vs. HL. Although