CASE REPORT

Ovarian Hyperstimulation Syndrome Complicating a Spontaneous Singleton Pregnancy: A Case Report

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It has been known that most cases of ovarian hyperstimulation syndrome (OHSS) are associated with the use of exogenous gonadotropins to induce multiple ovulation. However, OHSS is infrequently associated with a spontaneous ovulatory cycle, usually in the case of multiple gestations, hypothyroidism, or polycystic ovarian syndrome. We report a case of severe OHSS in a spontaneously pregnant woman with no underlying disease.

KEY WORDS: Ovarian hyperstimulation syndrome; normal spontaneous pregnancy.

INTRODUCTION

Ovarian hyperstimulation syndrome (OHSS) has been known as the most severe complication associated with ovulation induction or controlled ovarian hyperstimulation (COH). Its clinical manifestations are ovarian enlargement, ascites, oliguria, abdominal pain, electrolyte imbalance, hemoconcentration, and even thrombosis in severe cases. This phenomenon is believed to be an iatrogenic disease caused by the drugs used for ovulation induction but infrequently has been described in normal spontaneous pregnancy

(1–5). A case of severe OHSS in spontaneously conceived gestation is reported.

CASE REPORT

A 35-year-old Korean woman, gravida 1, para 1, visited our hospital initially at the sixth week of gestation for an antenatal check without a specific problem. Ultrasonographic examination revealed an intrauterine gestational sac with a fetal pole and normal heartbeat. Seven years ago the patient delivered a healthy normal male baby at full term. Her medical and family history was unremarkable. She received no medication from the three months before pregnancy. Menarche occurred at the age of 15 years, and the menstrual period was regular, at an interval of 30 days. At the 12th week of gestation, the patient complained of low abdominal pain, abdominal distension, and dyspnea. The vital signs were as follows: blood pressure, 110/70 mm Hg; pulse rate, 72 beats per min; body temperature, 36.2°C; and respiration rate, 17/min. The general appearance was not specific, but the patient had low abdominal tenderness, with no rebound tenderness. The uterus was enlarged to 3 months' gestation on pelvic examination, and ultrasonography revealed an intrauterine pregnancy, with a fetal crown–rump length (CRL) of 37.3 mm, which corresponded to the 10th week of gestation. Both adnexa were enlarged, measuring 15 × 11 × 11 cm in diameter on the right and 12 × 9 × 12 cm in diameter on the left, were multilobulated, and contained cystic structures (Fig. 1). A large amount of

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Fig. 1. Ultrasonographic findings for the patient on admission day. (A) right ovary; (B) left ovary; (C) fetus in the uterus.

ascitic fluid was observed in the peritoneal cavity. On CBC at admission, the white blood cell count was $11,900/\text{mm}^3$; hemoglobin, $12.9\text{ g\%}$; hematocrit, 37.5% ; and platelet count, $348,000/\text{mm}^3$. Blood coagulation test, liver function test, renal function test, and blood electrolytes were normal. Urinalysis was not specific. Blood estradiol (E_2) was $>5000\text{ pg/ml}$; progesterone, $>50\text{ ng/ml}$; and renin, 8.1 ng/ml/hr (normal, $0.68\text{--}1.36\text{ ng/ml/hr}$). Chest X-ray and EKG were normal.

The patient was managed with 1000 ml of 5% dextrose/sodium (Choongwae Pharmaceuticals, Seoul, Korea), 100 ml of 20% albumin (Dongshin Pharmaceuticals), and 500 ml of dextran (Rheomacrodex-D; Ilsung Pharmaceuticals) intravenously daily and carefully monitored by her symptoms, body weight, abdominal circumference, ultrasonography, and laboratory tests. At 13^{+2} weeks of pregnancy, 10 days after admission, her complaints of low abdominal pain, discomfort, and dyspnea were decreased, and

the laboratory findings remained stable; she was discharged.

At 22^{+1} weeks of pregnancy, the patient visited the outpatient department. Ultrasound examination revealed that the right adnexus had decreased to $9 \times 4 \times 5\text{ cm}$ in diameter and the left to $8 \times 4 \times 6\text{ cm}$ in diameter, and also there was only a small amount of ascites. The pregnancy progressed to term and at 41^{+4} weeks of pregnancy, a healthy normal female baby of 3045 g was delivered. Four weeks after delivery, on ultrasound examination at postpartum checkup, both ovaries were normalized.

DISCUSSION

OHSS is an iatrogenic complication caused by the various drugs used for the purpose of ovulation induction or ovarian hyperstimulation for the treatment of infertile patients. However, some cases of severe ovarian hyperstimulation syndromes have been reported

Fig. 1. (Continued).

in spontaneously pregnant patients without the use of such drugs (1–5).

The etiologic and pathophysiologic mechanisms of OHSS are still poorly understood, but it has been suggested that various factors, especially vasoactive substances which exist within the ovarian follicular fluid, might play an important role in the development of the condition. These vasoactive substances, which might be stimulated by exogenous gonadotropins used in ovarian hyperstimulation, might increase the capillary permeability and, as a result, cause fluid shift from the intracellular to the extracellular space (6–8).

There are some benign ovarian pathologic conditions complicating pregnancy, such as hyperreactio luteinalis, luteinized follicle cyst, ectopic decidua, cyst adenoma, luteoma of pregnancy, and granulosa cell tumor (9). The clinical presentation of OHSS may mimic hyperreactio luteinalis. Hyperreactio luteinalis is characterized by moderate or severe cystic ovarian

enlargement resulting in the development of benign theca lutein cysts (10). It occurs in 10–37% of gestational trophoblastic disease but has been reported in normal pregnancy without association of gestational trophoblastic disease (5). However, hyperreactio luteinalis is usually not associated with a complication such as ascites caused by fluid shift to the third space as in severe OHSS (11). Also, its symptoms are usually not severe (5) and develop later in gestation. Above all, it often leads to unnecessary surgery, whereas OHSS is treated conservatively. Finally, hyperreactio luteinalis is confirmed by histologic diagnosis after surgery, which might prove to be unnecessary.

In the present case, the patient had abdominal distension, discomfort, a large amount of ascites, and cystic ovarian enlargement categorized as severe OHSS by Golan's classification (12). As this disease regresses spontaneously with time or delivery, conservative management may be an initial approach; surgery should be done only in cases of ovarian

Fig. 1. (Continued).

rupture, torsion, intraperitoneal hemorrhage, or ectopic pregnancy.

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