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An ovarian torsion in severe spontaneous ovarian hyperstimulation syndrome associated with a singleton pregnancy

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Introduction

Ovarian hyperstimulation syndrome (OHSS) is a life-threatening condition, usually associated with pharmacological ovarian stimulation. Spontaneous OHSS is a rare condition, usually appearing in multiple gestation, hypothyroidism or polycystic ovarian syndrome (Baksu *et al.* 2004). Pharmacologically-induced or spontaneous, OHSS is characterized by ovarian enlargement along with a fluid shift into extravascular spaces causing ascites, pleural and/or pericardial effusion, hypovolemia and oliguria. A clear pathogenesis of this condition is still not understood. We report a case of spontaneous OHSS in a singleton pregnancy complicated with ovarian torsion. Another similar case has been reported recently (Baksu *et al.* 2004).

Case report

A 30-year-old woman, gravida 5, para 3, +1, was admitted at 11 weeks gestation because of abdominal pain for 1 week. Her medical history was unremarkable. Her last menstrual period (LMP) occurred 11 weeks before admission. Her previous pregnancies were spontaneous. The abdomen was severely distended and diffusely tender. Vaginal examination revealed a normal cervix, an enlarged uterus in size of 10-11 weeks gestation and enlarged ovaries (15 cm each), which were tender.

Laboratory results included: human chorionic gonadotropin (hCG) level; $19\,500$ mIU/ml, total protein; 4.2 g/dl (normal: 6-8 g/dl), albumin; 2.3 g/dl (normal: 3.5-5.5 g/dl); alpha fetoprotein (AFP) <4.0 ng/ml; carcino-embryonic antigen (CEA) <1 ng/ml; carcinogenic antigen 125 (Ca 125): 295 U/ml (normal range: 0-35 U/ml).

Ultrasonography revealed a single live intrauterine 10-week gestation with bilateral multilobular ovarian cysts that had smooth surfaces and clear contents. The right ovary was 140×148 mm in size and the left ovary was 137×97 mm. Fluid in pelvis and abdomen (ascites) was detected (Figure 1).

She was maintained at bed rest with fluid restriction. Due to difficulty in breathing, a diagnostic/therapeutic paracentesis was done. The extracted fluid proved to be exudative, the microbial cultures were negative and no malignant cells were found.

Because of hypoalbuminemia, she received 100 g albumin. She was discharged after 35 days with a live 15-week fetus. The right ovary decreased in size to 76×74 mm and the left to 79×96 mm. There were no ascites in the time of discharge. Total protein and albumin increased to 9.1 g/dl and 4.8 g/dl, respectively.

After 2 weeks, she was re-admitted with complaints of right lower abdomen pain and tenderness along with nausea and vomiting. In physical examination, the right lower quadrant of the abdomen was severely tender with rebound tenderness. Surgical exploration was carried out with a primary diagnosis of right ovarian torsion. In operation, it was observed that the right ovary was twisted through 180°. The ovary was unwound and several large ovarian cysts were aspirated. The patient was discharged 1 week after surgery. Her condition was then checked every week for 1 month and followed at normal prenatal check-

ups. She had a normal delivery of a healthy female infant weighing 3100 g at term.

Discussion

The complex clinical picture of severe ovarian hyperstimulation is an infrequent but well-described condition associated with



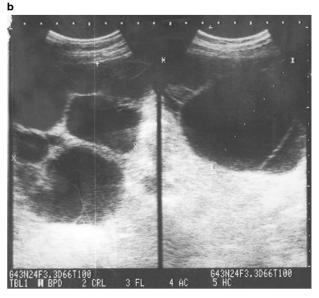


Figure 1. Ultrasonographic appearance of (a) an intrauterine singleton pregnancy and (b) an ovarian enlargement associated with multilubolar ovarian cystic structure.

ovulation induction especially in patients with polycystic ovarian syndrome, probably because of a higher sensitivity of follicles to gonadotropin stimulation (Baksu et al. 2004). Mild forms are infrequently associated with spontaneous ovulation and conception, primarily in the cases of multiple-pregnancies. Severe forms of this syndrome have been reported in severely hypothyroid pregnant women. This might be due to the fact that thyroid stimulating hormone (TSH) and follicle-stimulating hormone (FSH) can bind to each other's receptor and in hypothyroidism TSH is increasing significantly. Also, hCG plays a crucial role in the development of OHSS. Several cases of OHSS have been associated with multiple pregnancies or hydatidiform moles, conditions with very high levels of hCG. It has been shown that hCG can stimulate TSH and FSH receptors in vitro. A recent study identifies a mutant follitropin receptor responsible for OHSS (Smits et al. 2003).

OHSS rarely happens in a singleton pregnancy in women with no underlying disease. Only a few such cases have been reported (Baksu et al. 2004; Abu-Louz et al. 1997). This rare condition should be kept in mind in any pregnant woman showing clinical features of OHSS. The possibility of

ovarian rupture and torsion should always be considered in these patients. If surgery is necessary, only homeostatic measures should be undertaken and if torsion of the adnexa is encountered, unwinding the adnexa is possible, even when the adnexae are already ischemic.

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Ovarian hyperstimulation associated with a spontaneous pregnancy

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Case report

A 19-year primigravidida was admitted to the hospital because of abdominal pain, bloating and mild brown vaginal discharge. She had an 12-weekperiod of amenorrhea. She denied the recent use of an ovulation inducing drug. There were no signs of chronic anovulation, hyperandrogenism or hypothyroidism. She had a history of regular menses.

Laboratory testing showed normal haemoglobin (12.1 g/dl), hematocrit (35.6%), BUN (10 mg%), creatinine (0.6 mg%), uric acid (3.4 mg%), thyroxine 8.0 ng/ml, TSH 1.0 μ U/ml levels and slight hyponatremia (133 mEq/ml). Triiodotyronine resin uptake was 24%. Liver function tests were normal.

Transvaginal ultrasonography showed a 7 weeks' gestation without cardiac activity (Figure 1). There was massive ascites in the abdomen (Figure 2). Enlarged ovaries measuring 102×108 mm on the right and 99×80 mm on the left sides were seen to contain a great number of cysts between 15 mm and 40 mm (Figure 3).

Dilatation and curettage was performed because of a missed

Histopathological examination of uterine contents showed no evidence of gestational trophoblastic disease. The patient was discharged from hospital after 3 days of hospitalization advising weekly examination. Unfortunately, she was admitted to a private hospital and the ascites and ovarian cysts were aspirated at laparotomy. She was invited to our hospital 7 days after discharge from this hospital. Ascitie fluid and ovarian cysts had disappeared. Ovarian diameters were normal.

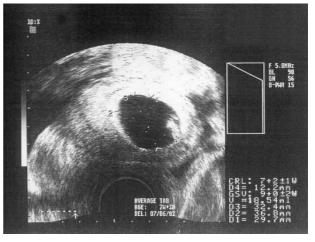


Figure 1. Transvaginal ultrasonography showing 7 weeks' gestation without cardiac activity.

Discussion

Ovariant hyperstimulation syndrome (OHSS) is characterized by a continuum of disorders ranging from a mild set of findings to a severe and potentially life-threatening entity and is most often associated with the administration of exogenous gonadotropins. OHSS is very rarely seen in pregnancy where ovulatory drugs or