CASE REPORT

Spontaneous ovarian hyperstimulation mimicking an ovarian tumour

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Ovarian hyperstimulation syndrome in a spontaneous singleton pregnancy is exceedingly rare. We report a case of ovarian hyperstimulation presenting as bilateral ovarian masses in association with spontaneous pregnancy, occurring in a woman with disturbed liver function. A possible mechanism is discussed.

Key words: ovarian hyperstimulation/ovarian tumour/spontaneous pregnancy

Introduction

The finding of a complex cystic and solid adnexal mass associated with ascites in the first trimester of a spontaneous pregnancy is highly suggestive of ovarian malignancy. This is intensified by the suspicion of neovascularization in the mass. We report a case of ovarian hyperstimulation in a spontaneous pregnancy that presented with a large adnexal mass and ascites.

Case report

A 34 year old primigravida was admitted with complaints of abdominal fullness and lower abdominal pain of 2 weeks duration. Her medical and gynaecological history were unremarkable. Menarche occurred at the age of 11 years, and subsequent menses were regular. Her last menstrual period occurred 11 weeks prior to admission. The patient had used oral contraceptives for 14 years, and was on no other medications.

Physical examination revealed normal vital signs and a severely distended abdomen with evidence of ascites, but without tenderness or palpable masses. Vaginal examination revealed a normal cervix, a uterus enlarged to 10 weeks gestation, and a cystic mass filling the entire pelvis.

Laboratory studies disclosed the following values: haemoglobin, 12.6 g/dl; haematocrit, 36%; blood electrolytes and coagulation profile, normal; β-human chorionic gonadotrophin, 106 000 mIU/ml; CA125, 2035 IU/ml (normal <35); α-fetoprotein and carcinoembryonic antigen concentrations, normal range; liver enzymes mildly elevated: serum glutamic-oxaloacetic transaminase (SGOT) and serum glutamic–pyruvic transaminase (SGPT) 52 IU/l and 80 IU/l respectively (normal 7–40 IU/l and 6–45 IU/l respectively). The patient was found to be a hepatitis-B carrier, without evidence of active viral replication.

Ultrasoundographic examination revealed a viable intrauterine pregnancy of a size consistent with dates, massive ascites and bilateral 10X10 cm multiloculated, complex cystic and solid adnexal masses. Doppler evaluation was suspicious of neovascularization (resistance index = 0.37). Chest X-ray was normal. Ultrasonographic evaluation of the upper abdomen disclosed a normal liver and pancreas, cholelithiasis and massive ascites. Surgical exploration was decided upon, because of a suspicion of malignant ovarian disease.

On laparotomy, 2500 ml ascitic fluid was removed. The uterus was of 12 weeks gestation in size, soft and mobile, and the ovaries were multilobulated, each measuring 10 cm in diameter, and containing cystic structures. Two mounds of 1 cm in diameter were found on the surface of the liver. There was no lymphadenopathy. Resection of several cysts from both ovaries was performed. Frozen section examination did not disclose malignancy and therefore no further radical procedure was undertaken. The liver modules were also sampled. Pathological examination of the cysts of both ovaries revealed fragments of corpus luteum cysts. The liver biopsies showed nodular features, fibrosis, proliferation of bile ducts and mild chronic inflammation. The ascitic fluid contained no malignant cells.

The patient was seen at the outpatient clinic 4 weeks postoperatively and was in good health, with no clinical or ultrasonographic evidence of ascites. An impressive regression of the ultrasonographic findings of the ovaries was noted. The serum liver enzymes were unchanged.

The patient delivered at term. The pelvic examination, including transvaginal ultrasonography, was normal 6 weeks after delivery.

Discussion

Ovarian hyperstimulation syndrome is a well-described entity that usually represents the most serious complication associated with ovulation induction. The incidence of the moderate-to-severe form is 1–2% (Speroff et al., 1994). Mild forms are infrequently associated with spontaneous ovulation and conception, primarily in the case of multiple gestations. Only three anecdotal case reports of severe ovarian hyperstimulation associated with a spontaneously conceived singleton pregnancy...
Spontaneous ovarian hyperstimulation have been reported (Rotmensh and Scommegna, 1989; Rosen and Mitchel, 1991; Zalel et al., 1992). One case report was related to severe hypothyroidism (Rotmensh and Scommegna, 1989), and the others to polycystic ovary disease (Rosen and Mitchel, 1991; Zalel et al., 1992).

The present case represents a condition of severe hyperstimulation syndrome (a combination of enlarged multilobulated ovarian cysts, abdominal pain and massive ascites) in a spontaneous pregnancy. Although the presenting symptom complex may have resembled a luteoma of pregnancy of hyperreactive lutealis, the ultrasound-Doppler characteristics, suggestive of neovascularization, and the extremely elevated CA125 value were highly suggestive of ovarian tumour. For this reason, explorative laparotomy was performed. The pathological examination ruled out a neoplastic process. Furthermore, this was supported by the resolution of the clinical and ultrasonographic findings. In retrospect, the operative procedure was unnecessary. The increased CA125 concentration has been described previously to be associated with ovarian hyperstimulation (Jager et al., 1987), probably because of increased mesothelial expression of the antigen (Lin et al., 1992).

Although polycystic ovary syndrome could not be excluded as a possible aetiological factor, it seems reasonable to speculate on an alternative aetiology in the present case. Our patient was a hepatitis-B carrier, with some degree of disturbed liver function, expressed by the mildly elevated liver enzymes. Although the liver damage was not severe enough to cause cirrhosis and portal hypertension (usually these patients are infertile), it might have disturbed the hormonal balance, mainly because of the possible decrease in the concentration of the sex hormone-binding globulin and the resulting increase in the concentration of the free hormones, oestrogens and androgens. This may present a clinical picture similar to that of women with polycystic ovarian syndrome, who are known to be more susceptible to hyperstimulation syndrome (Speroff et al., 1994). In these patients, the endogenous follicle stimulating hormone (FSH) is sufficient to stimulate the development of an abnormally large follicular cohort, which may then be luteinized by a spontaneous luteinizing hormone surge, and furthermore by the endogenous chorionic gonadotrophin secreted by the trophoblast (Zalel et al., 1992). Moreover, high oestrogen concentrations induce the secretion of less sialylated molecules of FSH, with higher receptor affinity and an increased clearance rate. The continuous subthreshold FSH concentrations cause multifollicular development and increase the risk of ovarian hyperstimulation (Ben-Rafael et al., 1995).

In summary, our report emphasizes that ovarian stimulation may occur in spontaneous pregnancy. This should be considered in a pregnant patient with ascites, bilateral enlarged multicystic ovaries, and pathological Doppler evaluation. Further studies should be performed in order to improve the characterization of the blood flow pattern by colour Doppler ultrasonography in hyperstimulated ovaries, and to avoid unnecessary intervention due to the misleading Doppler ultrasonography results.

References