Case Report

Severe Ovarian Hyperstimulation Syndrome in A Spontaneous Pregnancy with Normal Singleton Fetus: A Case Report

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Abstract

Most cases of Ovarian Hyperstimulation Syndrome (OHSS) are associated with the use of exogenous gonadotropins for the induction of multiple oocytes. However, OHSS is rarely associated with a spontaneous ovulatory cycle in women with multiple gestations, hypothyroidism, polycystic ovary syndrome, or molar pregnancies. Herein, we report a case of OHSS in a woman with spontaneous pregnancy, without any underlying diseases or risk factors. The clinical findings showed abdominal pain, nausea, dyspnea, and amenorrhea. After imaging and laboratory tests, the final diagnosis was established. The patient was managed successfully without any complications. In conclusion, although spontaneous ovarian hyperstimulation is a rare condition, physicians should be able to diagnose this condition. In fact, early diagnosis and successful management can potentially prevent serious complications, which are likely to develop rapidly in patients.

Keywords: *Fetus, ovarian hyperstimulation syndrome, pregnancy*

Introduction

Ovarian Hyperstimulation Syndrome (OHSS) is the most severe and potentially life-threatening iatrogenic complication, associated with ovulation induction.[1,2] This syndrome involves ovarian enlargement, along with the growth of multiple follicular cysts and acute fluid shifts from the intravascular to the extravascular space due to vascular hyperpermeability.[3]

The clinical manifestations of OHSS include ovarian enlargement, ascites, oliguria, abdominal pain, electrolyte imbalance, adult respiratory distress syndrome, and even thrombosis and death in severe cases.[1,2] OHSS, as an iatrogenic condition, often occurs following the use of drugs for ovulation induction. However, in extremely rare cases, it has been identified in normal spontaneous pregnancies.[4]

OHSS has been rarely described in non-pregnant women with primary hypothyroidism and gonadotroph pituitary adenoma.[5] On the other hand, spontaneous occurrence of OHSS has been reported in cases of multiple pregnancies or hydatidiform moles because of abnormally high serum levels of Human Chorionic Gonadotropin (HCG). In addition, high levels of Thyroid Stimulating Hormone (TSH) seem to stimulate the ovaries in hypothyroidism.[5,6] Herein, we describe the case of a woman with spontaneous singleton pregnancy, complicated by OHSS without any underlying diseases. The goal of this study was to raise awareness about this rare clinical condition.

Case Report

A 28-year-old primigravida woman at 8 weeks of gestation presented with severe generalized abdominal pain and distention 1 week before admission to the hospital. Her vital signs were as follows: respiratory rate, 24/min; pulse rate, 110 beats/min, and blood pressure 110/80 mmHg. No fever was detected in the patient, and the results of tilt test were negative. She complained of nausea without vomiting, while she had no medical history of diseases. Her clinical examination revealed abdominal distention with generalized tenderness and shifting dullness in the abdomen. There were also rales in the base of the lungs. She had not taken any drugs for ovulation induction, and her legs were edematous.

The laboratory tests showed hematocrit of 48% and mild leukocytosis of 14000/mL.

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The hCG level was 96103 U/L. The serum concentration of creatinine and blood urea nitrogen, electrolyte, estradiol, and follicle stimulating hormone (FSH) were normal. The thyroid function tests were also found to be normal. The CA-125 level was 86 U/mL (normal <35 U/mL), while the levels of CA15-3, CA19-9, and Carcinoembryonic Antigen (CEA) were normal.

Abdominopelvic ultrasonography showed a single viable intrauterine pregnancy at 8 weeks of gestation according to the fetal Crown-Rump Length (CRL) with normal amniotic fluid. Both ovaries were enlarged and multicystic. The left ovary was measured at 120 × 110 mm with multiple cysts; the largest cysts were 65 × 49 mm, and 40 × 39 mm, respectively. Also, the right ovary was measured to be 124 × 85 mm with multiple cysts, the largest cysts were 59 × 60 mm and 40 × 49 mm, respectively. Free fluid was detected in the pelvic cavity, as well as the right and left paracolic gutters.

The patient was under careful observation following her admission to the hospital. Diagnosis of ovarian cancer was not established considering the patient's age, pregnancy status, normal tumor markers, and detection of bilateral, thin-walled, and multiloculated ovarian cysts without papillary projection. Diagnosis of OHSS was established, and inpatient conservative management was performed. During hospitalization, vital signs, weight, abdominal circumference, fluid intake/output, serial hematocrit, and electrolytes were measured, and renal and liver function tests were carried out.

Finally, the patient was discharged after 1 week when her signs had partially regressed. During the outpatient follow-up at 18 weeks of gestation, the signs and symptoms of OHSS had completely resolved. Based on the ultrasound scan, the size of enlarged ovaries had decreased, and free fluid was not detected in the abdominal cavity. The duration of OHSS was 10 weeks. The patient's pregnancy continued until term uneventfully without any complications. She was followed-up during her pregnancy, and vaginal delivery was performed at 39 weeks of gestation. A healthy male newborn was delivered with an Apgar score of 10 and weight of 3,100 g. Three weeks after delivery, both ovaries were normal on the ultrasound in the postpartum checkup.

Ethical considerations

This study was approved by the Ethics Committee of Mashhad University of Medical Sciences (IR.MUMS.REC.1395.522). The patient's consent was obtained prior to preparing this case report. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Discussion

OHSS is typically associated with the use of gonadotropins for ovulation induction or ovarian hyperstimulation in the treatment of infertile patients. Its development after spontaneous pregnancy is extremely rare, although it is somewhat more common in multiple gestations due to the high level of HCG or hydatidiform moles related to abnormally high levels of serum hCG. It has been also suggested that high levels of TSH could stimulate the ovaries in women with hypothyroidism.[5,6]

In the spontaneous form of OHSS, follicular recruitment and growth occur later through promiscuous stimulation, and the clinical manifestations usually develop 8-14 weeks after amenorrhea. This form of OHSS culminates at the end of the first trimester of pregnancy, unlike the pharmacologic type, which usually develops 3-5 weeks after amenorrhea.[6,7] Management of OHSS is dependent on its severity. Evidence shows that early diagnosis and appropriate treatment can prevent serious sequelae in patients.[8]

Severe OHSS requires hospital admission and prompt management to replace the lost intravascular volume and prevent potentially fatal complications, particularly renal failure and thromboembolic events.[9] These patients should be closely monitored to ensure that they do not progress into the critical phase. In patients with significant ascites, paracentesis can be helpful due to reduction in intraabdominal pressure and improvement of renal blood flow with a subsequent increase in urine production.[10]

In a review of the literature, different cases of spontaneous pregnancy were reported in association with OHSS. Also, hypothyroidism was reported due to spontaneous OHSS with molar pregnancies or familial spontaneous OHSS. In this regard, Bassam *et al*. (2003) reported the case of a hypothyroid patient with OHSS and concluded that regression of ovarian cysts can be expected with initiation of thyroxin replacement.[11] Rachad *et al*. (2011) also presented a case of spontaneous OHSS with an invasive molar pregnancy.[9]

Arora *et al*. reported OHSS in a woman, who developed severe OHSS 3 days after a molar pregnancy evacuation.[12] However, our patient had a normal pregnancy. For the first time, Davoudian (2015) reported a pregnant woman with placentomegaly and enlarged multicystic ovaries. He described Placental Mesenchymal Dysplasia (PMD) associated with spontaneous OHSS and hypothesized that the most likely pathogenesis is ovarian stimulation by PMD-derived vascular endothelial growth factors;[13] however, this was not the case in our patient.

In another study, OHSS was diagnosed in a 25-year-old woman after delivery;;[14] nevertheless, our patient was in the first trimester of her pregnancy. Our patient is an example of OHSS, without evidence of thyroid or Polycystic Ovary Syndrome (PCOS) or factors involved

in spontaneous OHSS, as mentioned earlier. It should be noted that other cases of OHSS described in the literature were recurrent and familial.

We described an interesting case of OHSS in a primigravida woman, with no family history of OHSS during pregnancy. Since conservative management is usually the initial approach, surgery should be only used in the event of ovarian rupture, torsion, intraperitoneal hemorrhage, or ectopic pregnancy. It should be noted that case reports do not provide solid evidence, and the medical condition should be evaluated by professionals in each case.

Conclusion

OHSS is a very rare, but potentially fatal complication in spontaneous pregnancies. Early diagnosis and management of spontaneous OHSS are very important in the prevention of severe complications. Therefore, clinicians should be familiar with the differential diagnosis of OHSS if a patient presents with massive ascites and other symptoms of ovarian cancer, such as enlarged ovaries.

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Conflicts of interest

Nothing to declare.

References

1. Kasum M, Orešković S, Ježek D. Spontaneous ovarian hyperstimulation syndrome. Coll Antropol 2013;37:653-6.
2. Alper MM, Smith LP, Sills ES. Ovarian hyperstimulation syndrome: Current views on pathophysiology, risk factors, prevention, and management. J Exp Clin Assist Reprod 2009;6:3.
3. Sansone P, Aurilio C, Pace MC, Esposito R, Passavanti MB, Pota V, *et al*. Intensive care treatment of ovarian hyperstimulation syndrome (OHSS). Ann N Y Acad Sci 2011;1221:109-18.
4. Cabar FR. Ovarian hyperstimulation syndrome in a spontaneous singleton pregnancy. Einstein (Sao Paulo, Brazil) 2016;14:231-4.
5. Kim SJ, Yoon JH, Kim HK, Kang HC. Spontaneous ovarian hyperstimulation syndrome in a young female subject with a lingual thyroid and primary hypothyroidism. Korean J Intern Med 2016;32:559-62.
6. Lovgren TR, Tomich PG, Smith CV, Berg TG, Maclin V. Spontaneous severe ovarian hyperstimulation syndrome in successive pregnancies with successful outcomes. Obstet Gynecol 2009;113:493-5.
7. Oztekin O, Soylu F, Tatli O. Spontaneous ovarian hyperstimulation syndrome in a normal singleton pregnancy. Taiwan J Obstet Gynecol 2006;45:272-5.
8. Vloeberghs V, Peeraer K, Pexsters A, D'Hooghe T. Ovarian hyperstimulation syndrome and complications of ART. Best Pract Res Clin Obstet Gynaecol 2009;23:691-709.
9. Rachad M, Chaara H, Zahra Fdili F, Bouguern H, Melhouf A. Ovarian hyperstimulation syndrome in a spontaneous pregnancy with invasive mole: Report of a case. Pan Afr Med J 2011;9:23.
10. Davis M, Kennedy R. Ovarian hyperstimulation syndrome: Aetiology, prevention and management. Rev Gynaecol Perinatal Pract 2006;6:26-32.
11. Taher BM, Ghariabeh RA, Jarrah NS, Hadidy AM, Radaideh AM, Ajlouni KM. Spontaneous ovarian hyperstimulation syndrome caused by hypothyroidism in an adult. Eur J Obstet Gynecol Reprod Biol 2004;112:107-9.
12. Arora R, Merhi ZO, Khulpateea N, Roth D, Minkoff H. Ovarian hyperstimulation syndrome after a molar pregnancy evacuation. Fertil Steril 2008;90:1197.e5-7.
13. Davoudian P. Placental mesenchymal dysplasia associated with spontaneous ovarian hyperstimulation syndrome. BMJ Case Rep 2015;2015. doi: 10.1136/bcr-2014-207420.
14. Chae HD, Park EJ, Kim SH, Kim CH, Kang BM, Chang YS. Ovarian hyperstimulation syndrome complicating a spontaneous singleton pregnancy: A case report. J Assist Reprod Genet 2001;18:120-3.