Ovarian hyperstimulation syndrome complicating spontaneous molar pregnancy: a case report

Abhishek Mishra[1], Sushila Gaire[2], Bipana KC[1], Pradeep Mahat[1], Jeevan Neupane[1], Mandip Chaubey[3]
[1]Resident, Dept. of General Practice and Emergency Medicine, Maharajgunj Medical Campus, Institute of Medicine, Tribhuvan University, Kathmandu, Nepal; [2]Resident, Dept. of Gynaecology and Obstetrics, Pokhara Academy of Health Sciences, Pokhara, Kaski, Nepal;[3]Medical Officer, Nepal Bharat Maitri Hospital, Kathmandu, Nepal

ABSTRACT

Ovarian hyperstimulation syndrome (OHSS) is a rare finding that occurs in early pregnancy. There is a rapidly increasing ovarian size secreting vasoactive substances that lead to fluid shift into third spaces. It can be associated with a spectrum of other clinical findings, including ascites, hemoconcentration, hypercoagulability, and electrolyte imbalances. OHSS most commonly occurs as a complication of treatment with in vitro fertilization medications, such as human chorionic gonadotropin (hCG) or gonadotropin-releasing hormone agonists. OHSS has infrequently been reported to be caused by high hCG levels in molar pregnancies. We present a case of OHSS complicating spontaneous molar pregnancy. A 25-year gravida 3, para 2+0, living 2 lady presented with complaints of continuous vaginal bleeding for 2 weeks and lower abdominal pain at 17 weeks' gestation. A bulky uterus containing a large hyperechoic structure with multiple cystic spaces suggestive of complete molar gestation and enlarged ovaries containing multiple cysts were seen on ultrasound imaging. She was managed successfully with conservative and supportive treatment.

Keywords: Beta-hCG, hydatidiform mole, ovarian hyperstimulation syndrome

CORRESPONDENCE
Dr. Abhishek Mishra
Dept. of General Practice & Emergency Medicine, Maharajgunj Medical Campus, Institute of Medicine, Tribhuvan University, Kathmandu, Nepal
Email: abhishek.gm.mishra@gmail.com

INTRODUCTION

Ovarian hyperstimulation syndrome (OHSS) is the most serious complication of controlled ovarian hyperstimulation (COH) for assisted reproduction technologies (ART). It is a broad spectrum of signs and symptoms that include abdominal distention and discomfort, enlarged ovaries, ascites, and other complications of enhanced vascular permeability. The syndrome can be strictly defined as the shift of serum from the intravascular space to the third space, mainly to the abdominal cavity, in the context of enlarged ovaries due to follicular stimulation. In its very severe form, OHSS is a life-threatening condition. [1,2] We report a case of OHSS complicating spontaneous molar pregnancy- a rare but important complication; which in its severe form can be life threatening.

Case Report

A 25-year-old female gravida 3, para 2+0, living 2, lady presented to the outpatient clinic with complaints of continuous vaginal bleeding for 2 weeks which increased in amount for last 2 days rendering her to change 3 fully soaked pads in the last 24 hours along with passage of fleshy mass and intermittent lower abdominal pain. She is approximately 17 weeks pregnant dated by her last menstrual period. This was a spontaneous conception on background of regular menstruation pattern. Her last menstrual period was 17 weeks and 6 days ago. In each of her previous two pregnancies, she had given birth to one living child. Her previous pregnancies were uneventful with no history of ovarian stimulation in the index pregnancy. There is no history of previous gynecological abnormality. She had not yet received an ultrasonography for this pregnancy. On the 2nd day of admission, she developed increasing abdominal pain, distention, and nausea. On physical examination, she was normotensive, non-tachycardic, had no lid lag, and thyroid gland was non-tender and not-enlarged, and normal S1 and S2 heart sounds were heard with no murmurs and bilateral clear lung fields. On Obstetrical examination, her uterus was bigger than expected for the gestational age, measuring 23 cm from the symphysis pubis to the uterine fundus with doughy consistency, adnexa were nontender bilaterally. A bulky, anteverted uterus measuring 14.28 cm × 6.54 cm × 10.26 cm containing large hyperechoic lesion with multiple small-sized cystic spaces within its cavity was visualized on ultrasound. There was no demonstrable fetal part, increased vascularity, or myometrial invasion [Fig 1]. The ovaries were bulky and measure 7.4 cm × 4.2 cm × 5.1 cm and 6.4 cm × 3.2 cm × 5.9cm. They consist of multiple thin-walled moderate-sized cysts, echogenic stroma without solid component giving a spoke wheel pattern. Mild ascites without evidence of pleural effusion was also noted. [Fig 2]. Her level of beta human chorionic gonadotropin (beta-hCG) was greater than 10,00,000 mIU/mL. Her blood urea nitrogen level, serum creatinine level and TSH were within normal limits. A diagnosis of complete molar gestation with moderate OHSS was made. Frequent monitoring of vital signs, strict fluid balance, daily monitoring of abdominal girth, hematocrit and electrolytes were done. Abdominal ultrasound examination did not show any large fluid pockets that could be drained.

A chest radiograph was normal. Dilation and curettage was performed. A single dosage of intravenous Ceftriaxone 1gm was given

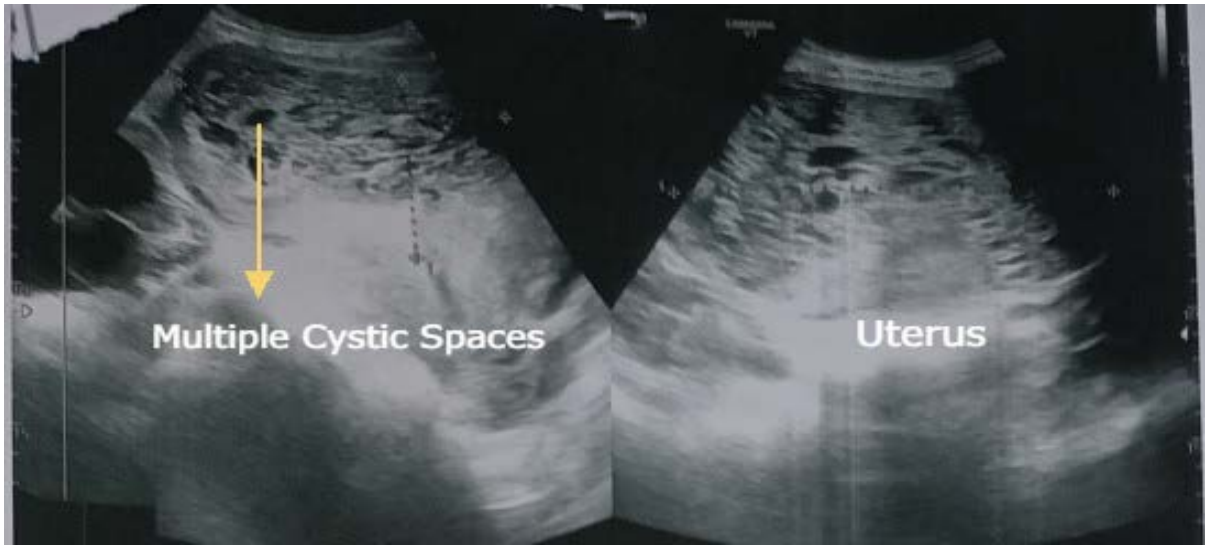

Figure 1. Ultrasonography of abdomen showing bulky uterus containing a hyperechoic structure with multiple cystic spaces within its cavity

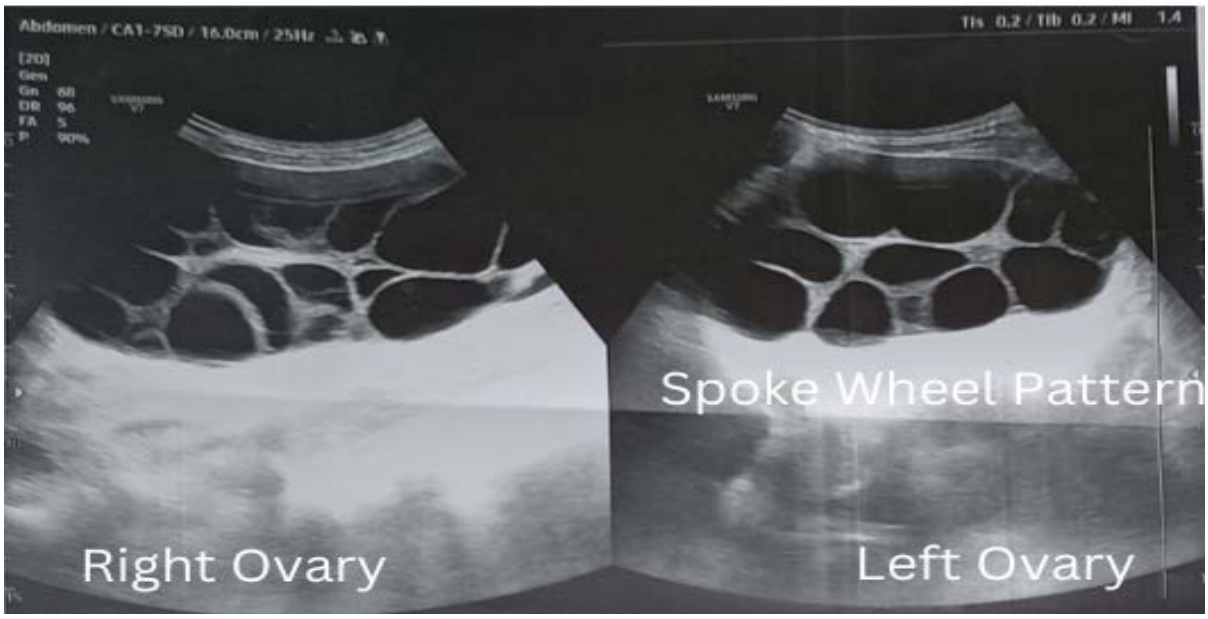

Figure 2. Bulky ovaries with thin-walled anechoic cysts and a thickened echogenic stroma giving a spoke wheel appearance

approximately an hour prior to the procedure. The postoperative course was uneventful. She received restricted intravenous fluids based on intake/output charting and analgesics- intravenous Paracetamol 1gram q6h for first 24 hours then for subsequent days oral Paracetamol 1gram q6hr and oral Pantoprazole 40 mg q12hr. The condition of the patient improved over the next 2 days; and she started to diurese, lost weight and her distension resolved. She was discharged home on day 6 of admission with counselling regarding contraceptive use, planning of future pregnancy, follow up timing.

Unfortunately, the patient has been lost to follow-up after discharge.

DISCUSSION

Ovarian hyperstimulation syndrome (OHSS) is the most serious complication of controlled ovarian hyperstimulation (COH) for assisted reproduction technologies (ART). It occurs when the ovaries are hyper stimulated and enlarged due to fertility treatments (or rarely, mutations in the follicle-stimulating hormone [FSH] receptor), resulting in the shift of serum from the intravascular space to the third space, mainly to the abdominal cavity. In its severe form, OHSS is a life-threatening condition because it can cause venous or arterial thromboembolic events, including stroke and loss of perfusion of an extremity.[1,2] In its mild form, it is a common complication, seen in 5% to 10% of patients undergoing ovulation induction; the moderate form is reported in 2% to 4% of patients undergoing ovulation induction, and the severe form in 0.1% to 0.5%.[3] There are a number of risk factors for OHSS, including Previous episode of OHSS[2], Polycystic ovary syndrome (PCOS)[2], Potential biomarkers of risk – Basal serum anti-müllerian hormone (AMH) concentration >3.3 ng/mL and an antral follicle count (AFC) >8.[4] Secondary risk factors related to ovarian response include: number of follicles[5], high serum estradiol concentration[6], number of oocytes retrieved in in-vitro fertilization (IVF) cycle[6], administration of human chorionic gonadotropin (hCG)[7], pregnancy.[8]

In our patient, OHSS was secondary to molar pregnancy and markedly elevated hCG levels. Hydatidiform mole or Molar pregnancy is part of a group of diseases classified as gestational trophoblastic disease (GTD), which originate in the placenta and have the potential to locally invade the uterus and metastasize.[9] The pathophysiology of OHSS is not fully understood; Elevated circulating hCG is thought to lead to ovarian enlargement and multiple cysts; this stimulates the ovaries to secrete vasoactive substances, increasing vascular permeability, leading to fluid shifts and the accumulation of extravascular fluid, resulting in renal failure, hypovolemic shock, ascites, pleural and pericardial effusions.[2] This acute shift produces hypovolemia, which may result in multiple organ failure, hemoconcentration, thrombosis, and disseminated intravascular coagulation from the increased viscosity of the blood. There are various classifications. The one discussed below is based on symptoms, imaging, and test results.[10] OHSS is classified into mild grade 1/2, moderate grade 3, and severe grade 4/5/6. The classification system is:

1) Mild OHSS
- Grade 1: Abdominal distention and discomfort
- Grade 2: Grade 1 plus nausea, vomiting, ovarian size of 5-12 cm, with or without diarrhea

2) Moderate OHSS
- Grade 3: Mild OHSS plus imaging evidence of ascites

3) Severe OHSS
- Grade 4: Moderate OHSS plus clinical evidence of ascites, with or without hydrothorax
- Grade 5: Additional hypovolemia, hemoconcentration (hematocrit > 45%), coagulation abnormalities, and oliguria
- Grade 6: Hemoconcentration (hematocrit > 55%), anuria, renal failure, venous thrombosis, and adult respiratory distress syndrome.

Based on the upper criteria, we diagnosed the case presented in this study as moderate OHSS.

Treatment is generally conservative. Mild OHSS can be treated on an outpatient basis with bed rest, oral analgesics, limited oral intake, and avoidance of vaginal intercourse, and usually resolves in 10 to 14 days. Moderate and severe OHSS require admission, bed rest and aggressive fluid resuscitation. Depending upon the severity, patients may require intensive care monitoring. Ultrasound-guided culdocentesis or paracentesis may be performed in those with tense ascites, orthopnea, rapid increase of abdominal fluid, or any other sign that may indicate progression of illness. Prophylactic anticoagulation with warfarin, heparin, or low molecular- weight heparin is indicated in women with a high tendency for thrombotic events who develop moderate to severe OHSS.[11]

Our patient was treated conservatively with supportive care and experienced a full recovery. Histopathological evaluation, and adequate follow-up with imaging and biochemical βHCG assay to rule out choriocarcinoma would have been part of our management protocol for the case, however,patient was lost to follow-up. This case indicated that OHSS should be considered in molar pregnancies, although the underlying reasons for the development of OHSS in these patients require further investigation.

CONCLUSION

The case assumes significance as it portrays the rare possibility of a molar pregnancy causing OHSS. It goes without saying that most obstetricians and gynecologists will encounter molar pregnancies during their career, so it is important for physicians to be aware of the signs, symptoms, and complications associated with molar pregnancies, one of which is OHSS. A rare but an important complication, which in its severe form can have life threatening implications Our purpose in presenting this case is to improve physician awareness of this complication in order to influence patient counseling and clinical management on this aspect.

Consent

A signed consent was taken from the patient regarding the publication of the case report.

Conflict of Interests

None

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