International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Neha et al. Int J Reprod Contracept Obstet Gynecol. 2024 Jul;13(7):1881-1884
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20241796

Case Report

A rare case on ovarian hyperstimulation syndrome in a spontaneous singleton pregnancy

Neha[1]*, Madhubala Chauhan[1], Divya Chaudhary[2]

[1]Department of Obstetrics and Gynaecology, RNT Medical College, Udaipur, Rajasthan, India
[2]Department of Obstetrics and Gynaecology, Geetanjali Medical College and Hospital, Udaipur, Rajasthan, India

Received: 23 May 2024
Accepted: 10 June 2024

***Correspondence:**
Dr. Neha,
E-mail: nehaverma.arya@gmail.com

ABSTRACT

Spontaneous form of ovarian hyperstimulation syndrome in pregnancy (OHSS) is extremely rare and characterized by gastro-intestinal symptoms and complication of shift of fluid in the third space. It is often associated with multiple gestations, hypothyroidism, polycystic ovarian syndrome and molar pregnancy. Treatment depends on the patient's clinical condition. The aim of this case report is to bring into light a case of spontaneous OHSS in a healthy Indian pregnant woman that presented with pain abdomen, abdominal distension and vomiting at 14 weeks of gestation without any of the above-mentioned risk factors. Ultrasonography showed a single viable intrauterine pregnancy along with bilateral enlarged cystic ovaries, ascites and raised CA-125 levels. There was no history of ovulation induction in present and previous pregnancy. We successfully managed the patient conservatively with Dopamine agonist Cabergoline and she delivered a healthy baby at 33 weeks. Although spontaneous OHSS is a rare entity, it should be included in differential diagnosis of acute abdomen in pregnant women. OHSS can lead to life threatening complications, early diagnosis is required for proper management.

Keywords: Ascites, CA-125, Dopamine agonist, OHSS

INTRODUCTION

Ovarian hyperstimulation syndrome (OHSS) in pregnancy is a rare but serious iatrogenic complication associated with assisted reproductive technology (ART). OHSS is characterized by cystic enlargement of ovaries and a fluid shift from intravascular compartment to extravascular space due to increased capillary permeability and ovarian neo angiogenesis in early pregnancy. Vascular endothelial growth factor (VEGF) plays an important role by increasing vascular permeability along with various other prostaglandin mediators like renin-angiotensin-aldosterone system (RAS), histamine and other inflammatory mediators.[1] The clinical manifestations of OHSS includes ovarian enlargement, ascites, oliguria, abdominal pain, electrolyte imbalance, adult respiratory distress syndrome and hyper- coagulability. Symptoms are often classified as mild, moderate, severe and critical on the basis of their clinical and laboratory features.[2] OHSS is usually seen as a complication of ovulation induction. Spontaneous OHSS during pregnancy is extremally rare. Spontaneous OHSS has been reported in pregnant women affected by mutation in follicular stimulating hormone (FSH) gene mutation, hypothyroidism, polycystic ovarian syndrome, molar and multiple pregnancies but the exact etiology is still unknown.[3]

We report a case of spontaneous OHSS in an Indian woman with a singleton pregnancy conceived naturally and without any of the above-mentioned risk factors.

CASE REPORT

A 20-year-old Indian female $G_2P_1L_0$ presented in emergency with amenorrhea of 4 months, pain abdomen, abdominal distension, nausea and vomiting since 3 days.

She was referred in acute distress with suspicion of malignant ovarian disease. She was 14±3 weeks pregnant; her last menstrual period was 09 November 2022. She had one previous normal vaginal delivery; the baby had died immediately after birth. She had no significant medical/ surgical history, with unremarkable gynaecological history.

Figure 1: Sonogram showing intra-uterine fetus.

On examination, she was conscious, oriented to time, place and person, with no evidence of pallor, icterus, oedema, afebrile on touch and maintaining SpO_2 of 98% on room air. Her pulse was 120/min, blood pressure 90/60 mmHg with unremarkable cardiovascular system (CVS) and respiratory system findings. Her abdominal examination revealed moderately distended, tense abdomen with no signs of guarding and rigidity. Her uterus size couldn't be assessed due to abdominal distension. She denied taking treatment for infertility or ovulation induction in past 6 months. We ran a battery of laboratory investigations and ultrasonography. Her magnetic resonance imaging (MRI) brain revealed normal brain parenchyma with normal pituitary gland.

Figure 2: Sonogram showing multiple ovarian follicles.

Table 1: Routine ante-natal investigations.

Investigations	At time of admission	After one week	At 28 weeks of gestation
Hemoglobin (gm%)	14.7	11.8	11.4
Hematocrit (%)	43.6	32.9	34.6
TLC	16,000	14,000	8,800
Platelet count (lakhs)	3.70	4.02	2.07
Blood sugar	71	103	72
Creatinine	0.65	0.4	0.42
Total bilirubin	0.29	0.15	0.2
SGPT/SGOT	24/38	40/58	35/28
PT/INR	13.4/1.02	13.8/1.07	
Na^+/ K^+	131/4.8	143/4.2	134/4.0
TSH		2.2	2.3
Blood group	A positive		
HIV, HBsAg, HCV	Non-reactive		

Table 2: Serum markers.

Markers	At time of admission
CA-125	79
S. prolactin	1804
LDH	150
β-hCG	>10,000
Testosterone	1.61

A diagnosis of moderate to severe ovarian hyperstimulation syndrome with single intrauterine pregnancy was made. She was managed conservatively with IV antibiotics, antiemetics and with 2 litres of IV fluids. She was given dopamine agonist bromocriptine in tablet form 2.5 mg daily for 8 days but she got no relief, hence tab. Cabergoline 0.25 mg twice weekly was started and continued till delivery. Ultrasound guided abdominal paracentesis was done twice and cytology of fluid confirmed benign effusion. She remained stable and her abdominal girth decreased from 80 cms to 67 cms during hospital stay. She was discharged in stable condition on tab. Cabergoline, iron and calcium supplementation. She was called for regular follow-ups. The pregnancy proceeded without any complications and she delivered a male child weighing 1.8 kgs at 33+4 weeks of pregnancy. The baby was discharged in good health after 2 weeks of NICU admission. Her postpartum ultrasound showed no signs of ascites, pleural effusion with normal ovarian size.

Table 3: Ultrasound studies.

Parameters	Findings	Ovaries	Other significant findings
At the time of admission	SLIUF of 13±3 weeks, FHR-166/min	R- 87×79×110 mm, L-75×61×101mm	Moderate ascites with mesenteric fat stranding, spleen - altered echotexture
After one week 01 March 2023	Uterus - 14±3 weeks	R- 58×41 mm, 540 cc, L-58×38 mm, 360 cc	Gross ascites, bilateral pleural effusion
After ascitic tap 04 March 2023	Ut.- 14±6 weeks with cephalic, placenta-posterior, AFI- normal	R-117×69×93 mm, 375 cc, L-106×79×91 mm, 381 cc	Mild ascites, mild amount of right-side pleural effusion
At 28 weeks 01 June 2023	SLIUF of 28±4 weeks, cephalic presentation, placenta- post, AFI-normal, fetal weight-1.2 kgs	R-39×19 mm, L-33×27 mm	
Postpartum	Ut.- postpartum in size	R-20×37×37 mm, 12 cc, L-24×24×29 mm, 9 cc	Normal study

DISCUSSION

OHSS is typically associated with the use of gonadotropins for ovulation induction or ovarian hyperstimulation in the treatment of infertility. Its development during spontaneous pregnancy is rare. Under certain circumstances such as multiple gestation, hypothyroidism, molar pregnancy, or polycystic ovarian syndrome possibility of its existence may be higher as per the reports available.[3] Spontaneous OHSS with a singleton pregnancy and no associated risk factors is extremely rare.

Spontaneous form of OHSS usually develops after 8 weeks of amenorrhea, differing from iatrogenic OHSS, that starts between 3 and 5 weeks of amenorrhea.[3] The hCG level in blood reaches maximum level at 8th to 10th weeks of gestation and starts to fall. However, our case was referred at 14 weeks of gestation on the suspicion of ovarian tumour owing to clinical features, raised CA-125 levels and ultrasonographic features of an unstimulated ovary. However, the diagnosis of spontaneous OHSS was confirmed with the help of senior radiologist.

For the cases of OHSS associated with assisted reproductive technology, preventive measures can be undertaken to avert the occurrence. But this is not the situation with spontaneous form as cases are recognised only after appearance of symptoms. Thus, effective management with early recognition is crucial to prevent any drastic events. Management is dependent on degree of severity.[2] In mild and moderate cases of OHSS, management of patient should be conservative with strict monitoring of all parameters keeping a keen eye on signs of haemoconcentration, hypovolemia, and coagulation disorders. Severe OHSS requires hospital admission and replacement of lost intravascular volume to prevent grave complications such as renal failure and thromboembolic events.[2] For patients with significant ascites, paracentesis is beneficial by decreasing intra-abdominal pressure and improving renal blood flow with a subsequent increased production of urine. Other helpful interventions that may be necessary include thoracocentesis for pleural effusion, heparin for thromboembolism, surgical laparotomy for torsion or haemorrhage of the cyst. However, in our case we managed the patient conservatively with cabergoline and ascitic fluid drainage. Cabergoline is a dopamine agonist which partially inhibits the VEGF receptor 2 phosphorylation levels and the associated vascular permeability without affecting luteal angiogenesis and reduces the 'early' (within the first 9 days after hCG) onset of OHSS.[1] A meta-analysis by Tang et al concluded that cabergoline efficiently reduces the rate of moderate OHSS, with no significant effect on clinical pregnancy rates and miscarriage rates.[4]

Our case emphasizes the importance of thorough evaluation of all women with ovarian mass/ascites complicating pregnancy and the need for clinicians to bear the differential diagnosis of OHSS. Furthermore, it also highlights the safety and beneficial effect of cabergoline in OHSS.

CONCLUSION

OHSS is an extremely rare, but potentially fatal complication in spontaneous pregnancy. Hence, early recognition and appropriate management are very important to prevent complication and to ensure good outcome.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Namavar Jahromi B, Parsanezhad ME, Shomali Z, Bakhshai P, Alborzi M, Moin Vaziri N, et al. Ovarian Hyperstimulation Syndrome: A Narrative Review of Its Pathophysiology, Risk Factors, Prevention, Classification, and Management. Iran J Med Sci. 2018;43(3):248-60.
2. The Management of Ovarian Hyperstimulation Syndrome (Green-top Guideline No. 5). Available at: https://www.rcog.org.uk/globalassets/documents/gui

delines/green-top-guidelines/gtg_5_ohss.pdf. Accessed on 09 March 2024.
3. Nwafor NN, Nyoyoko NP. Spontaneous Ovarian Hyperstimulation Syndrome: A Report of Two Cases from Different Pathogenesis. Niger Med J. 2020;61(5):269-72.
4. Manalai G, Shirzai A, Aalemi AK. High Dose Cabergoline in Management of Bilateral Ovarian Hyperstimulation Syndrome: A Case Report. Int Med Case Rep J. 2021;14:557-61.

Cite this article as: Neha, Chauhan M, Chaudhary D. A rare case on ovarian hyperstimulation syndrome in a spontaneous singleton pregnancy. Int J Reprod Contracept Obstet Gynecol 2024;13:1881-4.