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Case Report

Ovarian hyperstimulation syndrome with a twist: a rare case of adnexal torsion complicating severe ovarian hyperstimulation syndrome

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ABSTRACT

Ovarian hyperstimulation syndrome (OHSS) is a serious complication of ovarian stimulation protocol. This rare case highlights a patient with severe OHSS complicated by adnexal torsion. A 24-year-old married woman with history of oocyte donation presented in tertiary care centre with complaints of progressive abdominal pain, distension and shortness of breath. Clinical examination and investigations confirmed diagnosis of severe OHSS and right sided enlarged ovary (1190 cc) with torsion. Emergency laparotomy with right salpingo-oophorectomy was done. Adnexal torsion is common due to ovarian enlargement is common amongst patients undergoing ovulation induction. Doppler ultrasound aids the diagnosis, and surgical intervention is warranted. Early recognition of patient at risk of OHSS is essential for its prevention. Surgical management of adnexal torsion in OHSS is crucial to avoid complications.

Keywords: Adnexal torsion, Ovarian hyperstimulation syndrome, Enlarged ovaries, Emergency laparotomy

INTRODUCTION

Adnexal torsion occurs when the ovary, with or without the fallopian tube, rotates along its vascular pedicle, which can result in partial or total blockage of the blood supply potentially leading to necrosis of ovarian tissue and an acute abdomen upon clinical presentation.¹ Ovarian hyperstimulation syndrome (OHSS) is a rare complication of ART with incidence of moderate to severe OHSS being 1-5% of in vitro fertilisation cycles and adnexal torsion in such cases is even rarer with dearth of related data.²

Ovarian enlargement more than 12 cm, oliguria, hemoconcentration, clinical ascites with or without pleural effusion, hemoconcentration (hematocrit >45%), hypercoagulability, hypoproteinemia, oliguria and electrolyte imbalance are features of severe OHSS.² OHSS arising following ovulation induction, lead to increase

ovarian volume thereby elevating the risk of ovarian torsion. Ours is a very rare case of severe early onset OHSS presenting with adnexal torsion.

CASE REPORT

A 24-year-old Mrs XYZ para 1 living 1 hailing from lower socioeconomic background presented with complaints of abdominal distension and shortness of breath over past 3 days. She had history of undergoing ovulation induction with medications following which she underwent oocyte retrieval procedure in a different city, documentation of which not available. Her past menstrual cycles were regular and she had no past history of medical or surgical co-morbidities. General examination revealed severe pallor, pulse rate of 140 beat per minute, blood pressure 100/60 mmHg, oxygen saturation 97% on room air and body mass index (BMI) of 22. Chest examination revealed

decreased air entry on right side. On per abdomen examination a firm globular mass arising from pelvis and extending till umbilicus approximately corresponding to 22 weeks size with restricted mobility and tenderness over it was noted. Bimanual pelvic examination revealed a normal size anteverted uterus deviated to left with right sided fornical fullness and a mass separate from the uterus in right adnexa. Left ovary was bulky and palpable. A bedside urine pregnancy test was negative.

Transvaginal ultrasound revealed right ovarian volume of 1160 cc and dimensions of 15.9×9.3×14.9 cm with evidence of internal haemorrhage (Figure 1). The left ovary measured 8.2×3.9×7.3 cm with normal uterine size and endometrial thickness of 3.2 mm. Colour Doppler showed reduced arterial flow with mono-phasic waveform and absent venous flow in right ovary with normal findings in left ovary. There was moderate free fluid with internal echoes in pouch of Douglas and Morrison's pouch. Blood investigations revealed leucocytosis (total WBC count of 33,300), severe anemia (haemoglobin of 4.8 gm/dl) and hypokalemia.

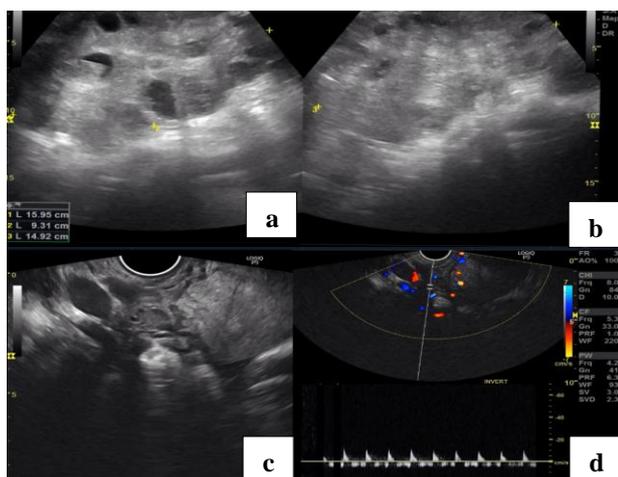


Figure 1 (a-d): Upper left and right - trans vaginal ultrasound in B mode shows bulky ovary with multiple peripherally arranged follicles and heterogeneously hyperechoic stroma, lower left-transvaginal ultrasound on b mode shows twisted, thickened and inflamed fallopian tube and lower right - on spectral Doppler there is absent venous flow and thumping monophasic waveform and low velocity from the artery.

A diagnosis of right adnexal torsion with severe anemia in a case of severe OHSS was made. Patient was shifted for emergency exploratory laparotomy with second packed red cell on flow. Intraoperatively approximately 100 ml of ascitic fluid was aspirated, a right multiloculated hemorrhagic necrosed ovarian mass of 15×12 cm with adjacent oedematous fallopian tube and one and half turns of torsion at its pedicle was noted (Figure 2 a and b). The left multicystic ovary with left fallopian tube appeared normal (Figure 2c). After obtaining intraoperative consent

patient underwent right salpingo-oophorectomy with specimen sent for histopathological examination.

Postoperatively patient was shifted to intensive care unit managed with blood transfusions, injectable antibiotics and injection LMWH. HRCT done on day 3 revealed mild bilateral pleural effusion not warranting any treatment as patient was maintaining oxygen saturation on room air. She was shifted to ward on day 4 and discharged on postoperative day 7 with normal radiological findings on chest X-ray and normal AMH value. Histopathological report was suggestive of areas of hemorrhage and necrosis along with oedema and interspersed with congested and distended blood vessels in the ovarian parenchyma (Figure 2d and e).

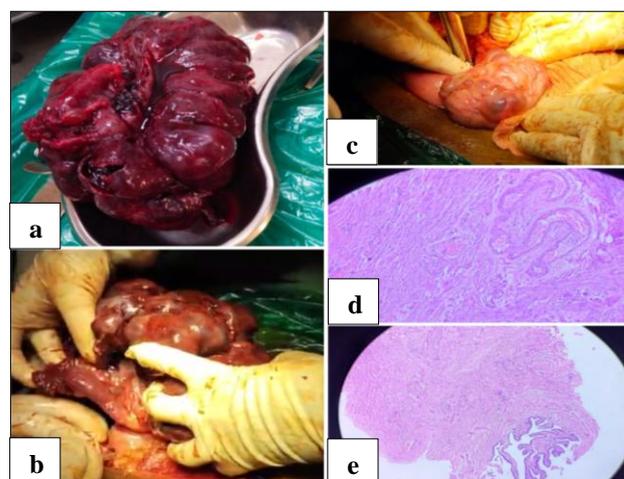


Figure 1: (a) Gross specimen of enlarged right ovary and fallopian tube, (b) intraoperative picture showing enlarged necrosed multicystic ovarian mass with thickened necroses fallopian tube on right side, (c) intraoperative picture of enlarged multicystic ovary of left side, and (d and e) histopathology slide suggestive of ovarian tissue with signs of hemorrhage and necrosis in stroma.

DISCUSSION

In past 20 years the incidence of OHSS and its complications have increased owing to late reproductive age and increasing incidence of ART. The overall incidence of ovarian torsion is 5.9 per 100,000 and that among women of reproductive age (15–45 years) is 9.9 per 100,000.¹ One in fifty women admitted with severe OHSS experienced ovarian torsion, and 10% underwent oophorectomy according to one study in USA.⁴

Emergency Doppler and grey scale ultrasonography (USG) are the preferred imaging techniques for diagnosing adnexal torsion but surgery is frequently carried out before a definitive diagnosis is reached. Laparoscopy is favoured because of its benefits of shorter hospital stay and fewer postoperative pain management needs. The conventional method in cases of ovarian necrosis is salpingo-

oophorectomy, may lead to decrease in desired fertility. Cystectomy following adnexal detorsion have also been practiced based on the postulation that even those ovaries that seem black or blue may continue to function after detorsion on USG follow-up, according to studies.

In severe cases of OHSS, hospitalization is necessary and treatment with low molecular weight heparin is recommended for all hospitalised patients as risk of thromboembolism is significantly increased in such patients.

CONCLUSION

Surgical intervention is only indicated for complications of OHSS like torsion or ovarian rupture. Such diagnosis requires clinical suspicion and USG with colour doppler is a reliable diagnostic modality. Immediate surgical intervention is treatment of choice. Early recognition timely intervention and multidisciplinary approach with individualised supportive care is the cornerstone of management of OHSS and its complication as was in our case.

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Ethical approval: Not required

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