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Case Report

A rare case of ovarian torsion in a pregnant patient

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ABSTRACT

Torsion of the ovary is the total or partial rotation of the adnexa around its vascular axis or pedicle. It is an uncommon cause of acute abdominal pain in females, and it is a gynecologic emergency. The majority of the cases present in the pregnant (22.7%) than in non-pregnant (6.1%) women. Diagnostic delay can result in loss of the ovary. This twisting initially obstructs venous flow, which causes engorgement and edema. The engorgement can progress until arterial flow is also compromised, leading to ischemia and infarction. The increased use of ovarian stimulation and assisted reproductive technology has led to an increase in the risk of adnexal torsion, particularly in pregnant women or women with ovarian hyperstimulation syndrome (OHSS). The differential diagnosis of adnexal torsion is particularly difficult in combination with OHSS or pregnancy, as abdominal pain, nausea and vomiting can be presenting symptoms of hyperstimulation or pregnancy as well. Here, we report a case of ovarian torsion occurring in pregnancy in which diagnostic delay occurred due to confusion with OHSS leading to oophorectomy. Fertility conservation may have been possible in case of earlier diagnosis and prompt treatment.

Keywords: Ovarian torsion, Hyperstimulation, Oopherectomy, Oopheropexy

INTRODUCTION

Torsion of the ovary is the total or partial rotation of the adnexa around its vascular axis or pedicle.[1] Although the exact etiology is unknown, common predisposing factors include moderate size cyst, free mobility and a long pedicle.[1] The classic presentation of adnexal torsion is sudden onset of unilateral lower abdominal pain which is initially visceral in character (vague and poorly localized) and may be accompanied by nausea and vomiting. It may radiate to the groin or flank.[3] There is a 5-fold increased risk of ovarian torsion during pregnancy, with an incidence of 5 per 10,000 pregnancies.[4] The majority of the cases presented in pregnant (22.7%) than in non-pregnant (6.1%) women.[2] Ovarian stimulation for infertility management further increases the risk, particularly in pregnant women or women with ovarian hyperstimulation syndrome (OHSS). Mashiach et al studied adnexal torsion in 201 women with OHSS over a period of 10 years and reported an incidence of 2.3% in non-pregnant women and an incidence of 16% in pregnant women.[3]

The differential diagnosis is particularly difficult in combination with OHSS or pregnancy, as abdominal pain, nausea and vomiting can be presenting symptoms of hyperstimulation or pregnancy but also of adnexal torsion.[3] Physical examination may reveal lower abdominal tenderness, adnexal tenderness or an adnexal mass. Fever is uncommon, and usually low-grade if present.[3]

Ultrasound is the diagnostic modality of choice for detecting torsion. The absence of blood flow within the ovary on Doppler exam is a useful finding in establishing the diagnosis. However, the presence of Doppler blood flow does not exclude the diagnosis of torsion. The rate of false-negative Doppler flow results is considerable, with a reported rate of 61% for pregnant women and 45% for non-pregnant women. Hence, the absence of Doppler

blood flow is a helpful finding (high specificity), but this finding has a low sensitivity.[5] Other common ultrasound findings in torsion include enlargement or edema of the ovary, an ovarian mass or cyst, and free pelvic fluid.[5] The most common finding in adnexal torsion on both ultrasound is simply enlargement of the ovary in question.[6] However, ultrasound may help find other pathology to explain a patient's pain (example: kidney stone, appendicitis).[6]

Here, we report a case of ovarian torsion occurring in pregnancy in which diagnostic delay occurred due to confusion with OHSS leading to oophorectomy. The aim of our case report is to draw attention to this complication of ovarian stimulation during infertility treatment and to emphasize the importance of early diagnosis for decision-making to preserve fertility.

CASE REPORT

A 24 year old female presented to our clinic with a history of infertility of two years. She gave a history of irregular menses. Semen parameters showed sperm count of 36 million, 55% motile, and 45% progressively motile with 3% normal morphological form. The patient had not menstruated for three months so withdrawal bleeding was induced with norethisterone (tablet feminine 5 mg po ds for 5 days). The antral follicular count assessed on day 2 of periods showed polycystic ovary syndrome (PCOS) (>12 follicles small follicles) bilaterally and arranged peripherally. The ratio of luteinizing hormone: follicle stimulating hormone (LH: FSH) was approximately 3:1. Hysterosalpingography on day 7 of the cycle was done which showed bilateral tubes to be patent with free spillage on both sides. Ovulatory dysfunction probably due to PCOS was diagnosed.

The patient was kept on low dose oral contraceptive pills i.e. ovral L for 21 days and tablet folvin 5 mg OD (folic acid) was started. In the next month from day 2 of the cycle, the treatment was started. She was given tablet letrozole 5 mg po od for 5 days (D2, D3, D4, D5, D6) and called on D7 to see for dominant follicle size. On day 7 no dominant follicle was noted. So injection human menopausal gonadotrophin (HMG) 75 mg subcutaneous (sc) was given for 5 days and she was called on day 12 for reassessment. On day 12 five follicles were noted of size 16 mm, 18 mm, 17 mm, 16 mm, and 14 mm respectively. Patient was advised to convert to in vitro fertilization (IVF) with a frozen cycle as the ovaries looked hyperstimulated or to cancel this cycle. However, in spite of the explained risks, the patient wanted to continue treatment as her husband was going abroad next month and for monetary reasons they could not afford IVF. So after taking consent, injection decapeptyl 0.2 mg was given sc and she was advised to have intercourse 36 hours after injection and then on alternate days for 6 days. Tablet duphaston 10 mg po bd was started after 36 hours. After 2 weeks she was advised to do urine pregnancy test which came positive. Two weeks after that, she was called to the clinic for a gestational sac scan. Gestational sac was seen intrauterine and it corresponded to 6 weeks 2 days of pregnancy. Repeat transvaginal scan (TVS) was done after 2 weeks which showed pregnancy of 7 weeks and 6 days with fetal pole and fetal cardiac activity. On that that day, the patients vitals were within normal limit and she had no complaints except occasional nausea. However, the same night around 11 pm the patient developed severe abdominal pain and vomiting and presented to the emergency. Her blood investigations showed haemoglobin (Hb): 12.5 gm% total leucocyte count (TLC): 11,700/cmm^3, platelet: 3,06,000/mm^3, sodium (Na): 134 meq/l, and potassium (K):4.0 meq/l. Renal function test and liver function test were also within normal range. Ultrasound showed an enlarged right ovary 12×10 cm with minimal fluid in the POD and left ovary was normal in size. She was evaluated by the surgical team and appendicitis and renal causes were ruled out. She was given a variety of pain medications including antispasmodics, tramadol, pethidine, but the pain was not relieved. She was advised for admission but refused.

The next morning with the same complaint of severe abdominal pain and vomiting patient was admitted. We suspected late OHSS and started conservative management. Antispasmodics was given for pain relief, ondensetron 4 mg iv TDS for nausea/vomiting and fluid replacement was also done. We discussed the possibility of ovarian torsion and did a Doppler ultrasonography. However, Doppler showed normal blood flow to both the ovaries. Only enlargement of the right ovary and minimal fluid in POD was noted. Pain medications and fluid replacement was continued and the patient was kept under observation for 24 hours. Symptoms were not decreasing at any cost so a diagnostic laparoscopy was done. Laparoscopy revealed an ischemic, bluish black and enlarged (13×9 cm) right ovary twisted on its pedicle two times. Right sided oophorectomy was performed. Post operatively, the patient's condition was stable and the pain resolved. She was discharged on the third postoperative day with antibiotics and heavy progesterone support.

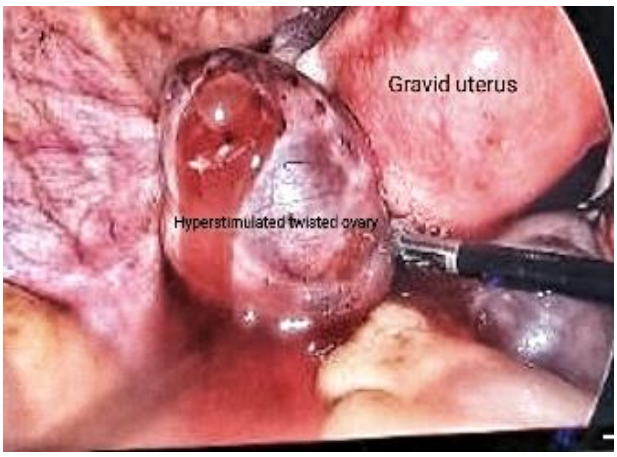

Figure 1: Laparoscopic image showing ischemic hyperstimulated ovary.

DISCUSSION

The exact incidence of adnexal torsion is unknown. In a 10-year study review ovarian torsion was 2.7 percent of emergency surgeries at a women's hospital and it was the fifth most common surgical emergency.[7] Adnexal torsion rarely occurs in spontaneous pregnancies (1–5 torsions per 10000 pregnancies). However, 12–25% of all adnexal torsion occur in pregnant women, often in combination with assisted reproduction and its complications (OHSS).[3] In our clinical experience, this was the first such case we came across at our infertility center.

The majority of adnexal torsion occur in the 1st or 2nd trimester of pregnancy and only around 10% occur in the 3rd trimester.[3,8] In the index case, the patient also had manifested in the first trimester (7 weeks 6 days) of pregnancy. The diagnosis of adnexal torsion is difficult if OHSS is also present because the symptoms are not specific. Abdominal pain, nausea and vomiting are often suspicious for torsion. Acute abdominal pain occurs in more than 80% of cases, often starting suddenly at night and persisting for more than 24 hours.[3,6,8,9] Such were the exact features in our case. However, as we had not seen many cases of adnexal torsion as a complication of ovarian stimulation, we kept late OHSS as our provisional diagnosis. Another mistake made on our part was that we relied heavily on Doppler flow investigation for diagnosis and ignored the other obvious findings like ovarian enlargement. Hasson et al recommended not to base the decision for surgical evaluation only on the results of Doppler flow investigation.[10] Arena et al also said the same and advised to take the patient's past medical history, clinical appearance, and laboratory assessment into account.[6] The size of the ovary on the affected side in patients ranged between 57 and 175 mm (mean 87.2 mm).[6,10]

Laparoscopic de-rotation of adnexal torsion is recommended as the first-line treatment, even for ovaries which are already ischemic, because in 73% of cases de-rotation is sufficient to preserve ovarian function.[6] For Arena et al complete absence of blood flow in the ovarian vessels is an indication for adnexectomy.[6] Laparoscopic fixation of the adnexa (ovariopexy) or shortening of the utero-ovarian ligament can be done to avoid recurrence of adnexal torsion.[11] In the present we opted for oophorectomy rather than de-rotation as we noticed a severely ischemic ovary. However, retrospectively we thought that maybe we could have tried derotation and observed for improvement. That might have been better for the patient's future fertility outcome.

Smith et al reported a reduced fertilization rate (FR) of 40% for oocytes aspirated from a de-rotated ovary, while the FR for the unaffected ovary was 93%. 75% of oocytes from the unaffected side and 64% of oocytes from the affected side developed into blastocysts. A reduced FR had been previously described in earlier reports in connection with reduced flow in the ovarian artery. Oelsner et al also did a study in which he retrieved oocytes from de-rotated ovaries in 6 patients and these oocytes could be subsequently fertilized.[12,13] This further supports the fact that our approach was incorrect. Had we only derotated, the chance of preserving fertility would be more. We undertook an outdated mode of management in which we went for oophorectomy in case of torsion.

Treatment of ovarian torsion is limited to surgery, either by laparoscopy approach or laparotomy. Pregnancy loss seems to be very rare and progesterone supplementation is recommended when the corpus luteum is removed prior to 7 to 9 weeks of pregnancy.[14-16] As we too removed the right ovary, supplementation with micronized, injectable and oral progesterone was given. The pregnancy of the patient is ongoing so we are unable to comment on the ultimate pregnancy outcome.

CONCLUSION

Ovarian torsion is an urgent gynecological surgery and can occur during pregnancy. Surgical techniques should be considered in the development of the adnexal torsion regardless of the gestational age. Adnexal torsion should be kept in mind in patients presenting with acute abdominal pain, nausea, vomiting and sonographically enlarged, polycystic ovaries. It is better to opt for diagnostic laparoscopy sooner than later, as diagnostic delay can lead to compromise of future fertility. Laparoscopic investigation and primary de-rotation should be attempted, even in cases where the organ is already livid and discolored and Doppler flow is absent. In most cases it will be possible to preserve the affected adnexa. We hope that through this case report we are able to outline the proper diagnosis and management of ovarian torsion in pregnancy and encourage others to not repeat the same mistakes we made.

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