

Laparoscopic detorsion for bilateral ovarian torsion in a singleton pregnancy with spontaneous ovarian hyperstimulation syndrome

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ABSTRACT

A 26-year-old primigravida with a singleton pregnancy of 9 weeks gestation presented with severe lower abdominal pain, following spontaneous hyperstimulation of the ovaries in a natural conception. Emergency laparoscopy was done and bilateral ovarian torsion with retained vascularity was noted. Bilateral detorsion with ovarian puncture and ovariopexy was performed. A review of international literature suggests that this is the first case reported with bilateral ovarian torsion following spontaneous ovarian hyperstimulation syndrome in a singleton pregnancy.

KEY WORDS: Bilateral ovarian torsion, laparoscopic detorsion, laparoscopic ovariopexy, spontaneous ovarian hyperstimulation syndrome

INTRODUCTION

Ovarian hyperstimulation syndrome (OHSS) is a life-threatening condition consequent to ovulation induction in assisted reproductive technology (ART) cycles.^[1] Although the majority of OHSS cases are iatrogenic, spontaneous OHSS (SOHSS), a very rare event, has also been reported.^[2] Although SOHSS almost always occurs in conjunction with pregnancy, the actual incidence still remains obscure. SOHSS usually develops between 8 and 14 weeks of gestational age, possibly due to a mutated follicular stimulating hormone (FSH) receptor, which shows abnormal sensitivity to human chorionic gonadotropin (hCG) or due to wild type of FSH with abnormally high levels of hCG.^[3]

Ovarian torsion, a major complication of OHSS, is estimated to occur in 12-25% of pregnant women.^[4]

We report a very rare case of bilateral ovarian torsion complicating SOHSS in a singleton pregnancy, which was successfully managed by laparoscopic detorsion.

CASE REPORT

The present case report is about a 26-year-old

primigravida at 9 weeks gestation presented with the complaints of severe abdominal pain. Pain was sharp, continuous in nature and not associated with vomiting and fever. She gave a history of pain during ovulation since several years and had undergone laparoscopic surgery twice in the past: For persistent left ovarian cyst in 1999 and for bilateral ovarian cysts in 2000. Cytological examination of fluid aspirated from the enlarged cystic ovaries was normal both times. She was diagnosed with recurrent hemorrhagic cysts following repeated admissions to hospital for abdominal pain and was managed conservatively. Diagnostic laparoscopy performed in 2012 for primary infertility revealed bilateral elongated ovarian ligaments.

Subsequently, the patient had a natural conception but developed severe abdominal pain due to SOHSS. Ultrasound was suggestive of 7 weeks intrauterine gestation with bilateral enlarged ovaries [Figure 1a-c]. The right ovary with retained vascularity, measured 81 mm × 42 mm × 64 mm and the left ovary had a corpus luteal cyst measuring 23 mm × 32 mm, with increased vascularity. She was re-admitted at 2 weeks later with similar complaints. An ultrasound revealed bilaterally enlarged cystic ovaries with absent blood flow.

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Emergency laparoscopy confirmed bilateral ovarian torsion. The ischemic-hemorrhagic appearing right ovary was enlarged and measured 7-8 cm with three twists [Figure 2]. The left ovary measured 3-4 cm and had undergone one twist [Figure 3]. Bilateral detorsion was performed [Figure 4] along with ovarian puncture to aid size reduction. Bilateral ovariopexy was also performed to secure the ovaries in their normal anatomical position. The post-operative period was uneventful and the patient was discharged on the 3rd post-operative day. Follow-up with an ultrasound showed successful continuation of the intrauterine pregnancy and normal ovarian blood flow.

DISCUSSION

Ovarian accidents, such as torsion and rupture, which complicate OHSS are known but rare events.^[5] Torsion of the right adnexa is more common than left adnexal torsion.^[6] However, bilateral adnexal torsion has also been reported.^[4]

A search of articles from 1857 to 2013 in the PubMed database was conducted to find the number of reported

cases with SOHSS complicated by bilateral ovarian torsion in singleton pregnancies. To the best of our knowledge, this is the first case with bilateral ovarian torsion complicating SOHSS in an early singleton pregnancy. So far, only two cases with unilateral torsion complicating SOHSS in singleton pregnancies have been reported.^[7,8] Even though torsion of the right adnexa is more frequent, in the current case, the left ovarian torsion may be attributed to the presence of a corpus luteal cyst, the most common risk factor for torsion during the first trimester of pregnancy.^[9]

An assessment of the extent of ovarian ischemia is crucial in deciding the mode of management, which is either untwisting for ischemic adnexa or adnexectomy if gangrene has developed. The macroscopic ischemic-hemorrhagic appearance of the affected ovary simulating gangrene could be attributed to venous and lymphatic stasis. In the current case, both the ovaries were twisted and enlarged with several hemorrhagic spots giving a gangrenous appearance. On performing detorsion and ovariopexy of the ovaries, significant color change was noted. This implies impeded blood supply from either uterine or ovarian arteries, but not complete cessation of the same. In addition, a study by

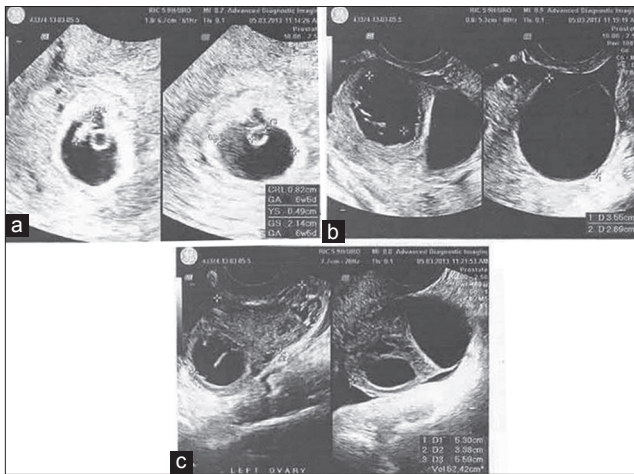


Figure 1: (a) Ultrasound image showing intrauterine gestation. (b) Ultrasound image showing enlarged right ovary with cysts. (c) Ultrasound image showing enlarged left ovary with cysts



Figure 2: Laparoscopic view of right ischemic torsed ovary



Figure 3: Laparoscopic view of left ovary with single twist



Figure 4: Laparoscopic view of left ovary undergoing detorsion and the detorsed right ovary

Oelsner *et al.*, which involved a retrospective analysis of 102 patients managed with surgical interventions for adnexal torsion, reported that 91.3% of patients with bluish-black ovary regained normal function. None of the patients developed pelvic or systemic thrombo-embolism, the risk presumed to occur following detorsion of the adnexa.^[10]

Oelsner *et al.* concluded that laparoscopic detorsion with ovarian salvage helps regain normal blood flow and at the same time retain normal ovarian function and reserve. The study reported that restoration of normal ovarian function was almost similar with laparoscopy (93.3%) and laparotomy (90.6%) in terms of macroscopic appearance, follicular development and fertilization of eggs retrieved from the detorsed ovary. However, laparoscopy was preferred owing to the reduced risk of adhesions, less post-operative pain, decreased use of analgesics, shorter hospital stay and early restoration of normal activity.^[10]

In summary, this case highlights the importance and appropriateness of early laparoscopic detorsion of bilateral ovarian torsion complicating OHSS in early pregnancy. Although SOHSS is a very rare event, it may be a life-threatening complication if associated with adnexal torsion and therefore necessitates a clear understanding of the syndrome.

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