Case Report

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Adnexal Torsion During Controlled Ovarian Stimulation: A Case Report

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Abstract:

A 34-year-old nulliparous woman with polycystic ovarian syndrome presented with mild discomfort in her right abdomen three hours before her scheduled retrieval. She underwent a transvaginal oocyte retrieval. Oocytes were obtained from the ovary; however, the right ovary was inaccessible as it was located above the uterine fundus. Consequently, the decision was made to abandon retrieval from the right ovary. After the procedure, the pain continued, and a physical examination indicated signs of peritonitis. Right ovarian torsion was suspected, leading to laparoscopic detorsion with transabdominal oocyte retrieval from the untwisted ovary. One mature oocyte was retrieved from the torsion-affected ovary, resulting in an embryonic arrest at the morula stage. Additionally, we reviewed the literature on the outcomes of in vitro fertilization cycles in women with ovarian torsion before oocyte retrieval.

Keywords: in vitro fertilization, ovarian stimulation, ovarian torsion

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Introduction

Ovarian torsion during in-vitro fertilization (IVF) is a rare event, with an incidence of 0.024%–0.2%[1,2]. Women with ovarian hyperstimulation syndrome (OHSS) and pregnancy after assisted reproduction are at a higher risk of torsion due to increased ovarian size[2]. Delays in diagnosis and management can lead to compromised ovarian function and future fertility[3]. Therefore, ovarian torsion should be suspected and ruled out in women undergoing fertility treatment that present with sudden-onset severe abdominal pain.

Data on ovarian torsion occurring before oocyte retrieval regarding outcomes of surgery and IVF treatment are limited[4–9]. To our knowledge, this is the first case report of ovarian torsion during an IVF cycle in which laparoscopic detorsion was performed, followed immediately by oocyte retrieval from the affected ovary within the same procedure. Despite a delay in the timing of the retrieval, a morula-stage embryo was successfully obtained.

Case report

This study was approved by the Institutional Review Board of the Faculty of Medicine at the Prince of Songkla University (approval number: REC. 66-272-12-1). Patient approval for publication was obtained. A 34-year-old nulliparous woman with a BMI of 28 kg/m^2 presented at the clinic with a 3-year history of primary infertility related to polycystic ovarian syndrome. She had history of irregular menstruation, with an interval of 2–3 months. Pelvic ultrasonography showed an antral follicle count of >20 bilaterally and an ovarian volume of 15 and 10 mL on the left and right sides, respectively, consistent with polycystic ovarian morphology. Other investigations and her medical history were unremarkable.

Choice of fertility treatment was offered, and she proceeded with her first IVF cycle, using progestin-primed ovarian stimulation protocol. Follitropin alfa (r-hFSH-alfa, GONAL-f®; Merck, KGaA, Darmstadt, Germany) at a daily dose of 150 IU was started on day 2 of progestin-induced withdrawal bleeding. A 10-mg dose of medroxyprogesterone acetate per day was administered to prevent premature luteinizing from cycle day 3 onwards. The peak estrogen level on the day of the trigger (cycle day 10) was 10,124 pg/mL, GnRH-agonist (Diphereline, France, Epson) upon which 0.2 mg was administered for final oocyte maturation. She complained of mild discomfort in the right side of her abdomen three hours before her planned retrieval. A differential diagnosis of premature ovulation and ovarian torsion combined with OHSS was determined. She underwent transvaginal oocyte retrieval, and an ultrasound performed during the procedure revealed an anteverted uterus, bilateral enlarged multicystic ovaries, and minimal free fluid in the cul-de-sac. The left ovary was in a cul-de-sac, and the right ovary was positioned above the uterine fundus. Color Doppler ultrasonography was inconclusive in diagnosing adnexal torsion. Eleven oocytes were retrieved from the left ovary. The transvaginal approach to aspirate oocytes from the right ovary was attempted; however, only two follicles were aspirated due to the abnormal position of the ovary. Thus, oocyte retrieval from the remaining follicles of the right ovary was abandoned.

After oocyte retrieval, the pain persisted and she had severe abdominal pain on her right side, with increasing severity within the next hour. Her vital signs were as follows: pulse rate, 92 beats/min; body temperature, 36.8 °C, and blood pressure, 110/85 mmHg. Physical examination revealed moderate abdominal distension, guarding, and rebound tenderness around the lower right abdomen. Laboratory investigation revealed leukocytosis, with an elevated white blood cell count of 16,100 per microliter. Transabdominal ultrasound showed the left ovary measuring 7.6×6.5 cm in a cul-de-sac and the right ovary measuring 8.5×8.6 cm positioned high in the midline above the uterine fundus. Color Doppler ultrasonography was inconclusive in

diagnosing ovarian torsion. However, right ovarian torsion was diagnosed through a combination of history and physical examination. Hence, an emergency laparoscopy for adnexal detorsion was performed 6 hours after symptom onset (3 hours after oocyte retrieval).

Laparoscopic approaches revealed a right enlarged ovarian cyst, measuring 8×8 cm, with an area of hemorrhage (Figure 1). The right adnexa had undergone 540° torsion around the utero-ovarian ligament. The twisted ovarian cyst was gently grasped with atraumatic forceps and detorsion was performed. Under laparoscopic vision a 17-gauge, single-lumen needle connected to an aspiration pump was used for transabdominal oocyte retrieval. The ovarian fluid was aspirated; however, only a minimal amount of follicular fluid was collected. At the end of the procedure, both adnexa were re-anatomized into a cul-de-sac.

Postoperatively her abdominal pain had improved, and as there were no postoperative complications, the patient was discharged 4 days after the operation. The follicular fluid from the two follicles of the right ovary was blood-stained and yellowish in color, and no oocytes were retrieved. One mature oocyte was retrieved from the torsion-affected ovary after detorsion was performed, which resulted in an embryonic arrest at the morula stage. Eleven oocytes were retrieved from the left ovary (9 MII and 2 GV), yielding three high-quality blastocysts; these were cryopreserved on day 5.

Discussion

The patient experienced ovarian torsion during ovarian stimulation after the trigger, and underwent transvaginal oocyte retrieval with subsequent laparoscopic detorsion in addition to transabdominal oocyte retrieval from a twisted ovary.

Adnexal torsion during fertility treatment is rare; however, it is a gynecological emergency requiring urgent surgical management to preserve ovarian function[1,2]. Adnexal torsion can be diagnosed when taking into account the patient's history, physical examination, and imaging studies. Common presenting signs and symptoms include: sudden onset of severe abdominal pain, nausea, and vomiting. Color Doppler ultrasound findings such as enlarged

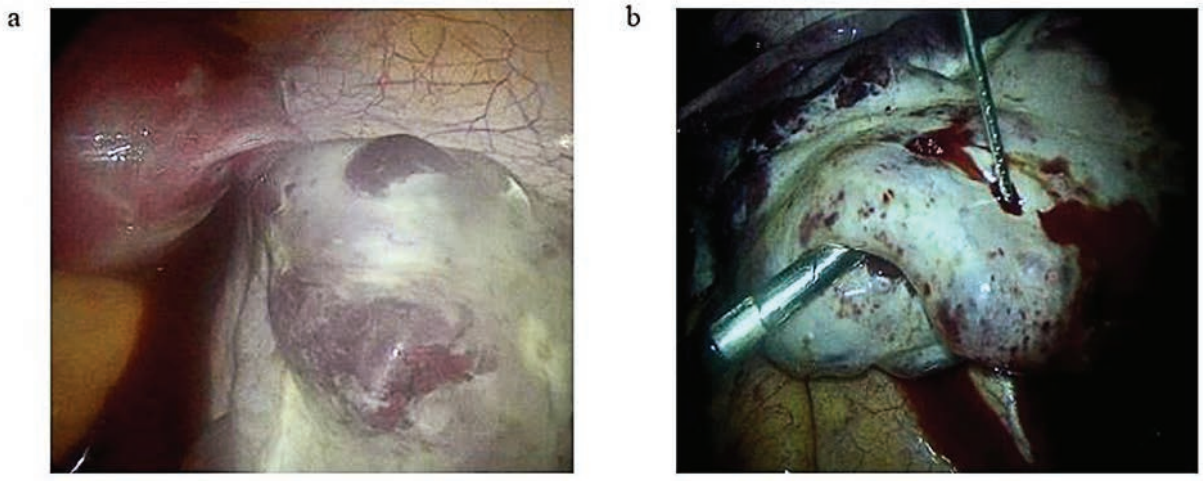

Figure 1 (a) Intraoperative findings demonstrating an enlarged edematous right ovary, with focal area of hemorrhage. (b) A single-lumen needle was used for transabdominal aspiration of the follicular fluid

or edematous ovaries, decreased or absent venous flow, follicular ring sign and whirlpool sign, are suggestive of torsion[10]. However, normal Doppler flow cannot reliably rule out adnexal torsion due to its low sensitivity. This limitation arises because arterial flow might still be preserved in cases with early presentation[11,12]: as was observed in our case. Hence, adnexal torsion is a clinical diagnosis, and a high index of suspicion for torsion is essential.

The majority of reports of ovarian torsion during ovarian stimulation occurred after oocyte retrieval, especially after embryo transfer, and patients usually have OHSS[2,13–15]. Data on ovarian torsion occurring before oocyte retrieval regarding outcomes of IVF treatment are limited[4–9] (Table 1). Cases where torsion occurs before retrieval show a decrease in the number of oocytes retrieved as well as the fertilization rate of oocytes retrieved from a twisted ovary. Several mechanisms leading to decreased fertilization rates of oocytes from twisted ovaries have been proposed; including a decreased arterial blood supply to the ovary, leading to a reduced concentration of essential hormones in folliculogenesis; venous and lymphatic stasis contributing to dysfunctional folliculogenesis[6], and ischemic injury to oocytes[4].

The outcome of IVF, where torsion occurs before the planned retrieval and oocytes are retrieved prior to performing laparoscopic detorsion, is poorer than for those who undergo laparoscopic detorsion followed by oocyte retrieval. This aligns with our case. Stefanidis et al[6]. reported a case of a 33-year-old female who presented with abdominal pain one day before her scheduled retrieval. The patient proceeded directly to oocyte retrieval as planned. Twelve follicles were aspirated from the twisted ovary; however, no oocytes were obtained. The patient's symptoms worsened postoperatively, leading to a laparoscopy performed six hours after oocyte retrieval. Robson et al[5]. also reported a case of a 37-year-old female patient whose symptoms arose one hour before her planned retrieval. Oocyte retrieval was conducted to aspirate all follicles and reduce ovarian volume, which led to the resolution of the patient's symptoms. The fertilization rate from the affected side was lower than that of the contralateral side (86% vs. 33%). The patient's pain intensified, prompting a laparoscopy two hours later.

Conservative surgical management with detorsion under laparoscopy represents the best option among women with adnexal torsion that occurs before the time of planned retrieval. Detorsion should be performed before proceeding directly to oocyte retrieval. Proceeding directly to oocyte retrieval with the aim of reducing ovarian volume to obliviate the surgery was unsuccessful in two previous studies, resulting in the need for subsequent laparoscopic detorsion[5,6]. Similar to our case, this approach necessitated subsequent surgery after ovum pick-up. Moreover, the outcome of IVF showed a decrease in the fertilization rate[5] and empty follicle syndrome from the twisted ovary[6]. In contrast, one study reported success in reducing ovarian volume by aspiration when ovarian torsion was suspected two days before oocyte retrieval. This relieved the patient's pain and allowed for successful oocyte collection two days later. Notably, laparoscopy was not performed to confirm torsion in this instance[14].

This case raises important points. First, ovarian torsion during IVF is a rare but serious complication. A high index of suspicion and early diagnosis are crucial for preserving ovarian function. Second, laparoscopic detorsion should be performed before proceeding directly to oocyte retrieval if torsion occurs prior to the scheduled retrieval time. Finally, in situations where diagnosis delays result in postponed oocyte retrieval, aspiration of any remaining follicles to reduce ovarian volume might lower the chance of re-torsion. Hence, the possibility of obtaining viable oocytes could still be attempted.

Table 1 A summary of in vitro fertilization outcomes from published case reports regarding ovarian torsion before oocyte retrieval

Case report	Patient profile and cause of infertility	Onset of symptoms	Management	Cycle outcome	
				Affected ovary	Unaffected ovary
Robson, et al. 2000	– 37 year–old nulligravida – Male factor	One hour before the planned retrieval	Oocyte retrieval followed by laparoscopic detorsion two hours after oocyte retrieval	– 7 oocytes were retrieved – 33% fertilization rate	– 12 oocytes were retrieved – 86% fertilization rate – 3 embryos were transferred (data on transfer outcome are unavailable)
Stefanidis, et al. 2002	– 33–year–old nulligravida – Male factor	One day before the planned retrieval	Oocyte retrieval followed by laparoscopic detorsion six hours after oocyte retrieval	– 12 follicles were aspirated but no oocytes were retrieved	– 18 oocytes were retrieved – Transfer of 4 embryos; however pregnancy did not ensue
Smith, et al. 2010	– 27–year–old nulligravida – Husband carried a balanced translocation	One day before the planned retrieval	Laparoscopic detorsion followed by oocyte retrieval on the next day	– 10 oocytes were retrieved – 4⁄10 (40%) were fertilized – 3⁄4 (75%) developed into blastocysts – All embryos were biopsied and revealed unbalanced translocations – None were transferred	– 15 oocytes were retrieved – 14⁄15 (93%) were fertilized – 9⁄14 (64%) developed into blastocysts – All embryos were biopsied and revealed unbalanced translocations – None were transferred
Inoue D. and Asada Y. 2020	– 34–year–old nulligravida	Two days before oocyte retrieval	Ovarian volume reduction by aspiration two days before oocyte retrieval and successful oocyte collection two days later	– 6 MII oocytes were retrieved – 14⁄15 (93%) were fertilized – 8 two–pronuclear (2PN) embryos were cryopreserved, while 6 were cultured to the blastocyst stage. – Two blastocysts were transferred, resulting in a successful pregnancy.	– 9 MII oocytes were retrieved
Naert, et al. 2023	– 32–year–old nulligravida – Polycystic ovarian syndrome	On cycle day 13 (one day before trigger)	Laparoscopic detorsion followed by trigger on the next day, and oocyte retrieval 36 hours after trigger	– 3 mature oocytes were retrieved – 2 blastocysts; one with high quality for cryopreservation	– 8 mature oocytes were retrieved – 5 blastocysts were cryopreserved on day 5 – 2 blastocysts were transferred resulting in live birth
Mortimer, et al. 2023	– 34–year–old nulligravida – Polycystic ovarian syndrome	12 hours before the planned retrieval	Laparoscopic detorsion followed by oocyte retrieval on the next day	– A total of 72 oocytes were retrieved, of which 70 were metaphase II (MII) oocytes. – Out of the 70 MII oocytes, 36 were cryopreserved, and 34 were inseminated using conventional IVF, resulting in a fertilization rate of 79.4% (27 out of 34). – 16 blastocyst–stage embryos were cryopreserved. (The author did not specifically report the outcomes based on the affected side.)	
Our case	– 34–year–old nulligravida – Polycystic ovarian syndrome	Three hours before the planned retrieval	Oocyte retrieval followed by laparoscopic detorsion and second oocyte retrieval three hours later	– 2 Follicles were aspirated in the first retrieval but no oocytes were retrieved – 1 mature oocyte was retrieved from the second retrieval and developed into a morula–staged embryo	– 9 mature oocytes were retrieved – 9⁄9 (100%) were fertilized – 3⁄9 (33%) developed into blastocysts

Conclusion

A high index of suspicion is required for diagnosing ovarian torsion occurring during IVF prior to ovum pick-up; so that surgery can be promptly performed to improve the outcome of IVF cycles. However, if there is a delayed diagnosis, viable oocytes can still be obtained from the affected ovary after laparoscopic detorsion; despite potential delays in the timing of retrieval.

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Conflicts of interest

The authors report that there are no competing interests to declare.

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