

# Management of Bilateral Adnexal Torsion in a Case of Ovarian Hyperstimulation Syndrome

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## Abstract

Adnexal torsion (AT) is usually unilateral and traditionally treated by oophorectomy. We present a case of bilateral AT, developed consequent to ovarian hyperstimulation syndrome (OHSS), managed by detorsion of the ovaries without oophorectomy. A 27-year-old female patient who underwent in vitro fertilization was hospitalized with acute abdomen. On ultrasonography, multifollicular ovaries, displaying continuity with each other on both sides with a size of 25×12 cm, which are consistent with OHSS and no apparent blood flow in the right adnexal area, were observed. In laparotomy, the right ovary (10×8 cm), left ovary (16×14 cm), and adnexa were bilaterally torsioned. Adnexa were detorsioned. Although it is assumed that blood flow did not improve in the ovaries, considering the patient's fertility desire, oophorectomy was not performed. On sonography at 6 months, the size of the ovaries and blood flow were normal. Conservative surgery should be the first choice of treatment in AT to preserve fertility in patients who underwent artificial reproductive technologies.

**Keywords:** Adnexal torsion, in vitro fertilization, ovarian hyperstimulation syndrome

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## INTRODUCTION

Adnexal torsion (AT), which occurs due to strangulation of blood supply in a twisted ovary and sometimes fallopian tube, is a surgical emergency for women with an incidence rate of 2.7% (1). In addition, the incidence rates of AT are 9.9/100,000 among reproductive age and 16% among pregnant women (2, 3). Therefore, pregnancy and conditions associated with assisted reproductive technologies (ARTs), such as ovarian hyperstimulation syndrome (OHSS), could be risk factors for AT (4). The presenting symptoms, such as nausea, vomiting, and abdominal pain that most pregnant women who underwent ART and/or developed OHSS experience, are not specific for AT (5). Although determination by clinical findings only is not easy, it is crucial to consider the possibility of AT in pregnant women, in patients who underwent ART, and in cases with OHSS in the differential diagnosis to avoid ovarian necrosis and preserve fertility.

Ovarian hyperstimulation syndrome has a wide spectrum of clinical outcomes from self-limiting to life-threatening. It is more common in young women, multiple pregnancies, and also patients with polycystic ovaries. The incidence of severe OHSS is reported to be 3.1%-8% in in vitro fertilization (IVF) cycles and is increasing in recent years due to increased availability of ART procedures (6, 7).

Adnexal torsion after IVF treatment is quite rare and is reported to be 0.08%-0.2% (6). We present a case of bilateral AT who developed OHSS and biochemical pregnancy following an IVF cycle and was managed successfully by detorsion of both ovaries without oophorectomy.



Figure 1. Right adnexa



Figure 2. Left adnexa

## CASE PRESENTATION

A 27-year-old, gravida 1 parity 0, female patient with no other medical or gynecologic history except polycystic ovarian syndrome (PCOS) underwent IVF treatment due to primary infertility. She was admitted to the emergency room at 23:00 pm with a sudden onset of severe abdominal pain, difficulty in micturition, and dyspnea persisting >24 h. On physical examination, rebound, defense, and abdominal tenderness were present in all quadrants. In addition, gynecologic examination revealed painful cervical movements and pain in the bilateral adnexal area that was more apparent on the left side. On transvaginal ultrasonography, the size of the uterus was normal, the endometrial thickness was 13 mm, and the multifollicular ovaries were observed, displaying continuity with each other on both adnexal sides, representing kissing ovaries, with a total size of 25×12 cm, which are consistent with OHSS. On pelvic Doppler ultrasonography, normal blood flow was observed on the left adnexa, and 15 mm of free fluid in the deepest pocket of the paraovarian region and Douglas was measured, although there was no apparent blood flow in the right adnexal area. The complete blood count values were as

follows: hemoglobin: 7.9 mg/dL, hematocrit: 24.1, white blood cell (WBC): 11,400/mm<sup>3</sup>, platelet: 156,000/mm<sup>3</sup>, albumin: 1.9 g/dL, total protein: 3.5 g/dL, and beta-subunit of human chorionic gonadotropin ( $\beta$ -HCG): 73. There was no pathological finding in biochemical and coagulation parameters. Laparotomy was performed due to analgesia resistant, strong, generalized abdominal pain with a suspicion of right AT. During exploration, the uterus was normal in size, and torsion was observed on both adnexal sides. The right twisted ovary was 10×8 cm in size, whereas the left twisted ovary was 16×14 cm in size. There was an apparent congestion in both ovaries, resulting in hemorrhage caused by ruptured areas on the surface of the ovary and also livid discoloration (Figure 1 and 2). The bilateral ovaries were detorsioned, and hemostasis was achieved as much as possible. Although it is assumed that blood flow did not improve in both ovaries after detorsion due to the persisting livid color, owing to the patient's fertility desire, operation was terminated without performing oophorectomy, keeping in mind the possibility of re-exploration. On clinical follow-up,  $\beta$ -HCG values were found to be decreased. On sonographic examination, although the size of the ovaries

was larger than normal, blood flow was normal on both sides in ovarian Doppler velocimetry. No pathological findings were detected in the size and blood flow of both ovaries at 6 months of follow-up.

## DISCUSSION

In this case report, we present a case of bilateral AT who developed OHSS and biochemical pregnancy following an IVF cycle and was managed successfully by detorsion of both ovaries without oophorectomy.

When OHSS is not accompanied by pregnancy, it is generally self-limiting, but otherwise, it should be evaluated carefully. Although treatment approaches may differ according to the clinical condition, our primary aim was to preserve fertility and to establish hemodynamic stabilization even with surgery. Although AT is uncommon in spontaneous pregnancies, the incidence is increasing due to ART regimens and related complications, such as OHSS (3). In the literature, the incidence of AT is approximately 6% even after ART treatment in the absence of OHSS, whereas it increases up to 16% in the presence of OHSS. Moreover, in the presence of pregnancy, OHSS itself increases the incidence of AT (3, 6). In our case, the patient underwent IVF treatment due to primary infertility with underlying PCOS and developed severe OHSS. Furthermore, biochemical pregnancy was detected without gestational sac in the uterine cavity on sonographic examination.

Nausea, vomiting, and abdominal pain are symptoms that are nonspecific, but acute abdominal pain generally starting at night and persisting for 24 h is an alarming symptom for AT (8). As in our case, the patient presented to the hospital with a sudden onset of pain persisting >24 h. Although mild leukocytosis is acceptable in pregnant women and in AT, it has a significant clinical importance in diagnosis when OHSS is accompanied by AT (9). We detected WBC count at 11,400/mm<sup>3</sup> that can be categorized as mild leukocytosis.

Conventionally, it is known that the gold standard diagnostic procedure is surgery. Generally, ultrasonography is a commonly used diagnostic tool in most adnexal pathologies. In AT, sonography is not specific or sensitive for diagnosis, but it can be helpful for detecting enlarged ovaries and absence of blood flow (10). In our case, sonography revealed radiological findings leading us to suspect only right AT, whereas bilateral AT was observed in surgical exploration.

In most of the cases presented in the literature, torsion was unilateral, and there was no superiority for neither the left nor the right ovary (3). Bilateral AT is uncommon, but its consequences are generally catastrophic. In our case, AT was bilateral with livid discoloration.

There are still controversies about the surgical approach in AT. Whether to choose laparoscopy or laparotomy under different clinical circumstances is the surgeon's decision. In our patient, laparotomy was used owing to the huge ovarian dimensions and hemodynamic instability. The traditionally recommended treatment approach for AT is adnexectomy (10). Similarly, Arena et al.

(11) offered adnexectomy in all cases if blood flow was not detected. On the other hand, Spitzer et al. (5) recommended adnexal derotating despite livid discoloration, a rather large size of torsioned adnexa, and absence of blood flow. In their case, similar to ours, the size of the ovaries was 57×175 mm. In addition to the huge dimensions of the ovaries, our case was different owing to the involvement of both adnexal sides. Despite bilateral adnexal detorsion, color of lividity did not disappear in our patient. Considering the patient's fertility desire, we avoided adnexectomy keeping in mind the possibility of re-exploration and any other complications, such as fever and thromboembolism, which is theoretical and very rare in practice.

## CONCLUSION

We want to emphasize that detorsion of adnexa must be performed as first-line treatment modality, in particularly in infertile patients, to preserve ovarian functions.

**Informed Consent:** Informed consent was obtained from the participant in the case.

**Peer-review:** Externally peer-reviewed.

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