

Adnexal Torsion in Pregnancy after Assisted Reproduction – Case Study and Review of the Literature

Adnex-Torsionen in der Schwangerschaft nach künstlicher Befruchtung

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Key words

- Adnexal torsion
- controlled ovarian hyperstimulation
- assisted reproduction
- laparoscopic derotation

Schlüsselwörter

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Abstract



Purpose: Aim of the study was to investigate the incidence, progress, management and outcome of adnexal torsion after controlled ovarian hyperstimulation in embryo transfer cycles.

Materials and Method: A retrospective analysis was done of 1007 patients of a private IVF centre. The literature on adnexal torsion is reviewed.

Results: In the literature, the incidence of adnexal torsion after assisted reproductive technologies (ART) is given as around 0.2%. A significant increase of up to 33% has been reported for cases with additional ovarian hyperstimulation syndrome (OHSS) and in pregnant women. In our retrospective analysis of 1007 women (incidence 0.46%) with 1411 fresh embryo transfer cycles, we found an incidence of 0.35% per embryo transfer. All adnexal torsions were treated by laparoscopic derotation to preserve fertility. All 5 cases with torsion were pregnant, 2 patients had mild OHSS. We recorded 3 term deliveries, 1 induced abortion for sirenomelia, and 1 missed abortion.

Conclusion: Adnexal torsion must be kept in mind after hyperstimulation and embryo transfer, especially when pregnancy or OHSS is also present. With early diagnosis, it should be possible to preserve fertility using laparoscopic derotation.

Zusammenfassung



Fragestellung: Häufigkeit, Verlauf, Therapie und Outcome von Adnex-Torsionen nach kontrollierter ovarieller Hyperstimulation in Embryotransfer-Zyklen.

Material und Methodik: Literatursichtung und retrospektive Untersuchung von 1007 Patientinnen eines privaten IVF-Zentrums.

Ergebnisse: In der Literatur wird die Inzidenz von Adnex-Torsionen nach ART mit maximal 0,2% angegeben. Ein deutlicher Anstieg bis auf 33% wird bei gleichzeitigem OHSS und einer Schwangerschaft beschrieben. In unserer retrospektiven Analyse an 1007 Frauen (Inzidenz 0,46%) in 1411 frischen Embryotransfer-Zyklen konnten wir eine Inzidenz von 0,35% pro Embryotransfer beobachten. Alle Adnex-Torsionen konnten mittels laparoskopischer Derotation fertilitätsertreu behandelt werden. Alle 5 Patientinnen waren schwanger, in 2 Fällen trat ein leichtes OHSS auf. Drei Termingerburten stehen 1 Abort-Induktion wegen Sirenen-Malformation und 1 Missed Abortion gegenüber.

Schlussfolgerung: An Adnex-Torsionen muss nach Hyperstimulation und Embryotransfer gedacht werden, vor allem, wenn eine Schwangerschaft eintritt oder zusätzlich ein OHSS besteht. Bei rechtzeitiger Erkennung sollte durch Laparoskopie und Derotation die Erhaltung der Fertilität möglich sein.

Introduction



With an incidence of 3%, adnexal torsion is a common cause of emergency gynaecological procedures [1]. The symptoms are mostly unspecific and diagnosis is therefore not simple. Early diagnosis is essential to preserve organ function and fertility. The increased use of assisted reproductive technologies such as controlled ovarian hy-

perstimulation (COH), in vitro fertilisation (IVF) and intracytoplasmic sperm injection (ICSI) has led to an increase in the risk of adnexal torsion, particularly in pregnant women or women with ovarian hyperstimulation syndrome (OHSS). Mashlach et al. studied adnexal torsion in 201 women with OHSS over a period of 10 years and reported an incidence of 2.3% in nonpregnant women and an incidence of 16% in pregnant

women [2]. 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 but also of adnexal torsion. The aim of our review article is to draw attention to this complication of assisted reproductive technology (ART) using case studies from our retrospective analysis of 1411 cases. Together with a review of the literature, we wish to emphasise the importance of early diagnosis for decision-making to preserve fertility.

Materials and Method

The incidence of adnexal torsion was retrospectively analysed in 1007 patients (1411 fresh embryo transfer cycles) who attended our private IVF centre between January 2006 and April 2011. A questionnaire was used to investigate the course and outcome of the pregnancy in 571 clinical pregnancies (pregnancy rate per embryo transfer 40.5%) including any maternal complications. The data of 461 births (birth rate 32.7%) after ART was collected. Adnexal torsion occurred in 5 cases and each case is presented below.

Case Reports

Case 1

A 35-year-old woman was stimulated prior to undergoing her 1st ICSI treatment which she required because of asthenozoospermia of her partner. Pituitary down-regulation consisted of long protocol (LP) with a GnRH agonist (GnRHa) (triptorelin 0.1 mg/d subcutaneously), starting at the midluteal phase. For COH, 225 IE of human menopausal gonadotropin (hMG) was injected daily intramuscularly until the largest follicle cohort had a diameter of 18–20 mm. Ovulation was induced with 10 000 IE human chorion gonadotropin (hCG). Five mature ovules were retrieved 35 hours after ovulation by transvaginal puncture and were fertilised. Two blastocysts were transferred. After a positive urine pregnancy test, the presence of an intact singleton pregnancy was recorded in the 6th gestational week (GW).

In the 10th GW the patient suddenly presented with strong abdominal pain over 24 hours and leukocytosis. The left ovary was enlarged and measured 64 mm (right ovary 34 mm). Laparoscopy found torsion of the left adnexa and polycystic ovaries with cyst rupture. After derotation of the left adnexa (▶ Fig. 1) and cyst puncture to relieve pressure, the adnexa could be preserved. Postoperative Doppler findings were unremarkable. In the 39th GW she was delivered of a healthy boy, born vaginally, weighing 3860 g and measuring 51 cm.

Case 2

A 24-year-old woman with polycystic ovarian syndrome (PCOS), hyperandrogenaemia, and protein S deficit had had 2 miscarriages in an earlier relationship. Her new partner had grade III oligo-asthenoteratozoospermia syndrome (OAT), making ICSI treatment necessary. Stimulation was done with a GnRHa LP (triptorelin 0.1 mg) and HMG 150 IE daily, together with metformin tablets 1000 mg and prednisolone tablets 5 mg daily. Eleven metaphase II oocytes were treated by intracytoplasmic morphologically selected sperm injection (IMSI). Fertilisation was confirmed in 7 oocytes. Two blastocysts were transferred, but further cryoconservation was not possible. In the 6th GW the patient

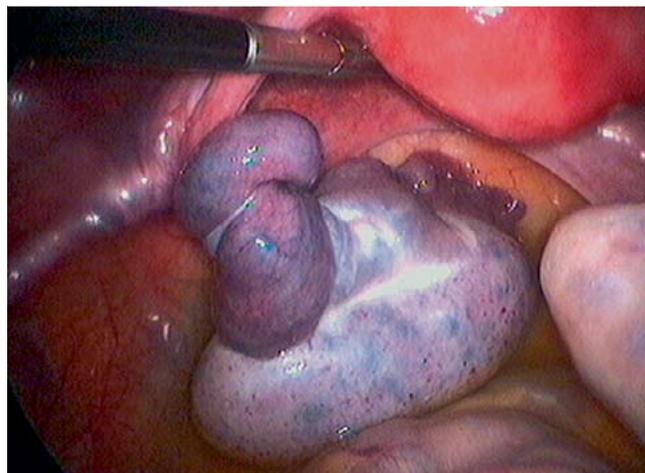


Fig. 1 Laparoscopy: torsion of enlarged left adnexa.

presented with persistent abdominal pain and nausea and increased β -HCG levels. Sonographically the ovaries were found to be enlarged, measuring 72 and 67 mm, respectively. Laparoscopy confirmed torsion of the right ovary, which was subsequently derotated. The patient was delivered vaginally of a healthy girl (3175 g, 46 cm) born in the 41st GW.

Case 3

The husband of a 40-year-old patient had undergone thyroidectomy followed by postoperative radiation therapy 16 years previously for thyroid carcinoma. He subsequently had OAT syndrome. Stimulation of the patient was done in accordance with a GnRHa LP (triptorelin 0.1 mg) and HMG 300 IE daily. Six of 8 mature oocytes could be fertilised and cultivated until the blastocyst stage. Two blastocysts were transferred. In the 5th GW the patient presented with strong abdominal pain and increased β -HCG levels. Sonography showed polycystic ovaries (right side 57 mm, left side 52 mm). Laparoscopy was indicated as the pain was resistant to analgesia. Both ovaries were enlarged, the right one showed livid discolouration and pelvic peritoneal adhesions. The right ovary was derotated and recovered quickly. Postoperative Doppler ultrasound was unremarkable. In the 10th GW, curettage was required for missed abortion. Eleven months later the patient was delivered of a healthy boy.

Case 4

A 31-year-old woman underwent IMSI because of asthenoteratozoospermia of her partner. Ten mature oocytes were retrieved after stimulation in accordance with GnRHa LP (triptorelin 0.1 mg) and HMG stimulation 150 IE daily. Nine oocytes could be fertilised and 1 blastocyst was transferred. Four more blastocysts were cryoconserved. After a positive urine pregnancy test, the patient underwent ultrasound examination for abdominal pain and tympanites. Ultrasound showed enlargement of both ovaries (175 mm on the right, 75 mm on the left), a 6-cm left-sided ovarian cyst, free intra-abdominal fluid and an eccentrically located, gestational sac. At this point no surgical intervention was required. In the 8th GW the pain suddenly returned and the torted right ovary was derotated laparoscopically. In the 19th GW the patient opted for induced abortion because sonography findings showed sirenomelia.

Case 5

IMSI was indicated in a 34-year-old woman due to asthenoteratozoospermia of her partner. Stimulation was done in accordance with a GnRHa LP (triptorelin 0.1 mg) and HMG 225 IE. Eight mature oocytes could be fertilised, 1 blastocyst was transferred, and 2 further blastocysts were cryoconserved. In the 10th GW, the patient suddenly presented with acute abdominal pain. On ultrasound the ovaries were found to be enlarged (68 mm and 48 mm, respectively). Torsion of the right adnexa was found on subsequent laparoscopy. The torsed ovary had ruptured, resulting in bleeding with hemoperitoneum. The adnexa was derotated, coagulation was used to stop the bleeding, and the hemoperitoneum was suctioned off. The adnexa could be preserved and subsequent ultrasound control found the pregnancy to be intact. The subsequent course of the patient's pregnancy was uncomplicated with the exception of gestational diabetes after GW 30. The patient was delivered of a healthy boy weighing 4090 g in the 40th GW.

Discussion

Incidence

The reported incidence of adnexal torsion is 2.3% in nonpregnant women and 16% in pregnant women [2]. Adnexal torsion rarely occurs in spontaneous pregnancies (1–5 torsions per 10000 pregnancies). However, 12–25% of all adnexal torsions occur in pregnant women, often in combination with assisted reproduction and its complications (OHSS). The reported incidence is low for oocyte donation cycles (0–0.2%) and IVF cycles (0–0.13%); however, the incidence increased to 6% under stimulation for ART and to 16% with OHSS [2–6]. But even when adnexal torsion occurs simultaneously with OHSS, the reported incidence varies between 1 and 33% [7]. 70% of torsions occur in multiple pregnancies [8]. The incidence in our study period with 1411 fresh embryo transfers was 0.35%; however, all 5 cases in our study were singleton pregnancies (5/461 births = incidence of 1.08% per birth after IVF/ICSI) and 2 patients additionally presented with mild OHSS.

Time of occurrence

The majority of adnexal torsions occur in the 1st or 2nd trimester of pregnancy and only around 10% occur in the 3rd trimester [2, 10]. If cysts are found in pregnant women on sonography, these very rarely (3.8%) result in torsion [6]. According to Mashlach et al., the incidence for both ovaries is approximately the same [2]. In all 5 cases from our ART collective, torsion occurred in pregnant women between the 5th and 10th GW, with 4 torsions found on the right side and 1 on the left side.

Diagnosis

Symptoms and laboratory parameters

The diagnosis of adnexal torsion is difficult if OHSS is also present because the symptoms are not specific. But late diagnosis increases the risk of having to excise the affected organ. 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 [2, 9, 10, 12].

Several authors have reported that patients had mild leukocytosis; however, while levels may be increased in nonpregnant women, levels may be within normal ranges for pregnant women

[7, 9, 12]. Leukocytosis is also one of the laboratory indicators which may change with OHSS.

Ultrasound diagnosis

Transvaginal ultrasound may often show enlargement of the ovaries and polycystic changes without this being evidence of torsion. A pathological result of Doppler flow ultrasound may indicate adnexal torsion, but even normal Doppler findings cannot exclude the possibility of torsion. The rate of false-negative Doppler flow results is considerable, with a reported rate of 61% for pregnant women and 45% for nonpregnant women [11]. Hasson et al. therefore recommended not to base the decision for surgical evaluation only on the results of Doppler flow investigation (as proposed by Arena et al.) but also to take the patient's past medical history, clinical appearance, and laboratory assessment into account [11, 12]. The differential diagnosis, particularly for right-sided adnexal torsion, includes appendicitis, renal colic, renal or urethral calculi, and obstructive bowel disease [12]. In all cases in our study, acute abdominal pain or gradually increasing pain, mostly accompanied by enlarged polycystic ovaries, led to the patient being admitted to hospital. The size of the ovary on the affected side in our patients ranged between 57 and 175 mm (mean 87.2 mm; [Table 1](#)).

Time between hospital admission and surgery

In the literature, the time between admission to hospital and surgery is between 6 and 15.5 hours and may be even significantly shorter in the 1st trimester of pregnancy. However, several days may pass between the start of symptoms and surgery [2, 13]. In our cases, acute symptoms or persistence of complaint meant that the mean time to laparoscopic evaluation and treatment was 6.4 hours (1–24 hours), and treatment was still in time to preserve fertility ([Table 1](#)).

Treatment

Laparoscopic derotation of adnexal torsion is recommended as the first-line treatment, even for ovaries which are already ischaemic, because in 73% of cases derotation is sufficient to preserve ovarian function. For Arena et al., complete absence of blood flow in the ovarian vessels is an indication for adnexectomy [12]. We were able to avoid adnexectomy in all cases and were therefore able to preserve fertility.

Risk of recurrence

Laparoscopic fixation of the adnexa (ovariopexy) or shortening of the utero-ovarian ligament can be done to avoid recurrence of adnexal torsion, but this should be the exception rather than the rule [7, 14–16]. In their retrospective case control study, Hasson et al. reported a recurrence rate of 19.5% for pregnant women and 9.1% for nonpregnant women; however 73.2% of pregnant women and 20.8% of nonpregnant women had been treated with ART prior to torsion [11, 17]. None of the 5 cases in our study experienced recurrence during the subsequent course of their pregnancy.

Outcome

Smith et al. reported a reduced fertilisation rate (FR) of 40% for oocytes aspirated from a derotated 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 blasto-

Table 1 Summary of 5 cases with adnexal torsion after COH and ET.

Age (years)	RO	TE	PR	OHSS	GW	US: right ovary*	US: left ovary*	Adm.-OP	Diagnosis	Treatment	Outcome of PR
35	5	2	yes	mild	10	38	64	24	left adnexal torsion	laparoscopic detorsion + puncture of ovarian cyst	vaginal delivery, 39th GW, boy (2 860 g, 51 cm)
24	15	2	yes	no	6	72	67	1	right ovarian torsion	laparoscopic detorsion	vaginal delivery, 41st GW, girl (3 175 g, 46 cm)
40	8	2	yes	no	5	57	52	2	right ovarian torsion	laparoscopic detorsion	missed abortion, 10th GW, curettage
31	13	1	yes	mild	5 8	175	75	3	left ovarian cyst (6 cm) right ovarian torsion	conservative laparoscopic detorsion	induced abortion, 19th GW, sirenomelia
34	8	1	yes	no	10	68	48	2	right adnexal torsion, ovarian rupture, hemato-peritoneum	laparoscopic detorsion + coagulation and suction	vaginal delivery, 40th GW, boy (4 090 g, 56 cm)

RO = number of retrieved oocytes, TE = number of transferred embryos (blastocysts), PR = pregnancy, OHSS = ovarian hyperstimulation syndrome, GW = gestational week at diagnosis, US = ultrasound, * = diameter (mm), Adm.-OP = time between admission to hospital and surgery (hours)

cysts. A reduced FR had been previously described in earlier reports in connection with reduced flow in the ovarian artery [18]. In a repeat IVF procedure, Oelsner et al. retrieved oocytes from laparoscopically derotated ovaries in 6 patients and these oocytes could be subsequently fertilised; Oelsner et al. therefore recommended derotation as the procedure of choice for ischaemic ovaries [19]. The subsequent course of a pregnancy after treatment for adnexal torsion is generally favourable; abortion rates of between 8.3% and 16.6% do not appear to be increased [2, 11]. In our cases, 3 pregnancies continued to term with the vaginal delivery of healthy children. One pregnancy was terminated in the 19th GW for medical reasons as ultrasound showed sirenomelia. There was 1 case of missed abortion in a 40-year-old patient; however 11 months later this patient gave birth to a healthy child.

Conclusion

Adnexal torsion after ART is not uncommon, particular if accompanied by OHSS and/or (multiple) pregnancy. Adnexal torsion should be kept in mind in patients presenting with acute abdominal pain, nausea, vomiting and sonographically enlarged, polycystic ovaries. The differential diagnosis is difficult, particularly if OHSS is present, and consultation with a reproductive physician should be considered. Laparoscopic investigation and primary derotation should be attempted, even in cases where the organ is already livid and discoloured and Doppler flow is absent. In most cases it will be possible to preserve the affected adnexa and thus preserve fertility.

Conflict of Interest

The authors declare that they have no financial connections to any company cited in this article.

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