Maternal ovarian torsion in pregnancy, which is caused by the rotation of the ovary and/or the tube around its supporting ligaments, is a rare complication estimated to occur in approximately 1:1000 pregnancies [1]. However, the rate of torsion may reach 16% in pregnancies resulting from assisted reproductive technology (ART) [2].

The most common underlying adnexal pathology in pregnant women with adnexal torsion is functional ovarian cysts, especially in pregnancies resulting from ovulation induction (OI) or in-vitro fertilization (IVF). Other adnexal pathologies include non-functional adnexal cysts, such as benign cystic teratoma and paratubal cysts, whereas some patients may experience torsion of otherwise normal adnexa [3-7]. During laparoscopy for adnexal de-torsion, non-functional adnexal cysts are usually removed by a cystectomy procedure. However, the management of functional ovarian cysts causing torsion is a topic of debate. Since these functional cysts often resolve spontaneously, the surgeon may opt for conservative management by de-torsion. Alternatively, intra-operative aspiration and drainage of functional cysts may be attempted, with the possible complication of uncontrolled bleeding. In addition, in cases of large multi-cystic ovaries in women who underwent OI or IVF, surgical drainage may be technically difficult because of the numerous cysts and their large size and vascularity.

Adnexal torsion in pregnancy has been associated with risk for recurrent torsion during the same pregnancy [3-6]. One prominent hypothesis is that recurrent torsion may occur in women with functional adnexal cysts who were managed by de-torsion alone, since the adnexal pathology causing torsion was not addressed in these cases. The aim of this study was to investigate the risk of recurrent torsion in pregnancy in women with functional ovarian cysts, and to compare the risk of recurrence in women who underwent de-torsion versus de-torsion and aspiration.

We analyzed all records of surgically diagnosed adnexal torsion cases occurring during pregnancy operated on in our department.
from January 2007 to April 2019. The demographic characteristics, clinical findings, preoperative ultrasound scans and laboratory tests, operative reports, surgical procedure, pathological diagnosis (when available), and torsion recurrence rate during the same pregnancy were retrieved from the hospital's computerized medical records and reviewed retrospectively.

We subsequently classified all cases of torsion occurring during pregnancy according to the underlying adnexal pathology. This classification was based on clinical, ultrasound and surgical findings, and the pathology results when available and included torsion cases caused by presumed functional ovarian pathologies (e.g., large multi-cystic ovaries associated with ovulation induction, follicular cysts, and corpus luteum cysts), torsion cases caused by non-functional adnexal pathologies (e.g., benign cystic teratoma and paraovarian cysts) and torsion cases labeled torsion of otherwise normal adnexa (examples where no adnexal pathology was diagnosed on the ultrasound or intra-operatively).

The preoperative ultrasound examinations were conducted using two-dimensional gray-scale imaging with a transabdominal and/or a transvaginal transducer (Voluson Expert, Voluson E8 and Voluson E10; GE Healthcare, USA). The Doppler flow analyses were only available for a few cases and thus not analyzed. The ovarian measurements included the largest ovarian diameter and the largest cyst diameter as recorded in the patient’s chart.

Gestational trimester was defined as follows:
- First trimester: 4 to 12 weeks of gestation
- Second trimester: 13 to 27 weeks of gestation
- Third trimester: 28 weeks of gestation and beyond

The duration of abdominal pain before arrival at the emergency room was calculated based on the patient’s descriptions of her complaints as recorded in the medical records. For descriptive purposes, pain over the previous 24 hours was defined as “short duration” and a duration of more than 24 hours as “long duration.” The cutoff was defined arbitrarily.

All patients underwent laparoscopy or laparotomy. After confirmation of adnexal torsion, three types of surgical procedures were performed by de-torsion alone, by de-torsion followed by drainage and aspiration of the adnexal cyst, and by de-torsion followed by resection of the adnexal pathology. All specimens were sent for pathological evaluation.

Statistical analyses were performed using IBM Statistical Package for the Social Sciences statistics software, version 24 (SPSS, IBM Corp, Armonk, NY, USA). Descriptive variables are presented as the mean ± the standard deviation. Frequencies are presented as percentages. One-way ANOVAs and Fisher's exact tests were used as appropriate. A P value < 0.05 was considered statistically significant. This study was approved by the institutional review boards.

RESULTS

The study cohort was comprised of 106 pregnant women with 113 episodes of adnexal torsion. Of these, 71 (67.0%) torsion cases were caused by presumed functional ovarian cysts, 5 (4.7%) cases were caused by non-functional adnexal pathologies (including 4 cases of benign cystic teratoma and one para-ovarian cyst), and 30 (28.3%) torsion cases were diagnosed as torsion of an otherwise normal adnexa. Most torsion cases (78, 73.6%) occurred in the first trimester of pregnancy, whereas 28 (26.4%) occurred in the second and third trimesters. Among the cases of torsion involving functional cysts, the mean gestational age at time of torsion diagnosis was 9.2 ± 4.6 weeks and most cases occurred in the first trimester of pregnancy (62, 87.3%). For the whole cohort, in-vitro fertilization (IVF) or ovulation induction (OI) was reported for 54 (50.9%) cases. Eight (7.5%) pregnancies were twin gestations. The clinical presentation included abdominal pain in all cases, and nausea or vomiting in 88 (83.0%) cases. Most women (87, 82.1%) presented to the emergency room within 24 hours of the initiation of symptoms.

The preoperative ultrasound findings and surgical characteristics of the cohort are presented in Table 1. On the preoperative ultrasound evaluation, the mean diameter of the involved ovary was 68.6 ± 20.6 mm. The diameter of the ovary involved in the torsion of functional cysts was larger than the non-functional cysts or an enlarged ovary without adnexal cysts (76.6 ± 19.7, 59.0 ± 24.5, and 55.9 ± 14.7 mm, respectively, P = 0.001). The
mean diameter of the contralateral ovary was 51.1 ± 21.9 mm. In terms of the surgical findings, the right adnexa was involved in 63 cases (59.4%). The mean number of adnexal twists was 1.7 ± 0.8. Most torsion cases were managed by de-torsion of the involved adnexa, with or without aspiration and drainage of the associated functional ovarian cysts. In 5 (4.7%) cases, a cystectomy procedure was required following de-torsion for removal of non-functional adnexal cysts such as ovarian cystadenoma, benign cystic teratoma, and paratubal cysts. The main surgical approach was laparoscopy. Two patients with non-functional adnexal cysts underwent laparotomy: a 29 gestational-week patient and a 6-week gestational patient, both with large benign cystic teratoma. No adnexal fixation procedures were performed.

There were no intra-operative complications. The post-operative complications included one case of abdominal wall hematoma at the incision of the left lateral trocar site managed conservatively, and one case of complicated urinary tract infection managed with intravenous antibiotic treatment. Specifically, there were no cases of intra- or post-operative intra-abdominal bleeding, and no cases of blood transfusion.

The rate of torsion recurrence during the same pregnancy for the whole cohort was 6.6% (n=7). Of these, 4 (57.1%) cases were caused by presumed functional ovarian cysts and 3 (42.9%) cases were caused by normal adnexa. The torsion recurrence rate in cases involving functional cysts was analyzed according to the surgical procedure performed for the first torsion event [Figure 1]. Among the patients who underwent de-torsion alone (n=28), 4 (14.3%) had recurrent torsion in the same pregnancy compared to no recurrence in the 43 patients who underwent aspiration and drainage of ovarian cysts (P = 0.021).

**DISCUSSION**

There is a general consensus that ovarian torsion in pregnancy is associated with the persistence of ovarian cysts and neoplasms, where larger masses are generally associated with greater potential for torsion [8]. Another reason for volume growth is the increased effect of gonadotropin, particularly with respect to iatrogenic overstimulation during infertility treatment [9-11]. In the current study, most cases of torsion caused by functional ovarian pathologies occurred in the first trimester. The diameter of the ovary involved in the torsion of functional pathologies was larger than the diameter of non-functional cysts or an enlarged ovary without adnexal cysts. The incidence of functional cysts and concurrently the risk of torsion are known to increase following ART treatment. We previously reported that cystic or multi-cystic ovaries can be visualized on the preoperative ultrasound in most cases (~85%) of first trimester torsion [4]. By contrast, in the current study only approximately 13% of all the torsion cases in the second and third trimesters were associated with functional ovarian pathologies. This finding suggests that first trimester torsion is usually associated with functional ovarian cysts (prevalent in patients who conceived by ART) and that the gradual regression of these ovarian cysts during the second and third trimester is connected to a lower risk of torsion.

Torsion recurrence during the same pregnancy has been described in case reports and case series [3-7,10,12-14], but has

---

**Figure 1.** Comparison of recurrence rate of adnexa torsion during the same pregnancy involving functional cysts treated by de-torsion alone or by aspiration and drainage of ovarian cysts

*P = 0.021, Fisher’s exact test*
not been directly compared according to the underlying adnexal pathology. In our previous reports [4-6], we noted that a multi-cystic ovary was significantly more common in patients with recurrent torsion during the same pregnancy than in those with a single torsion episode. We speculated that this was possibly due to a continuation of the underlying disorder causing torsion: enlarged and cystic ovaries. We suggested that fenestration of the functional cysts in the first operation might reduce the risk of recurrent torsion. As predicted, in the current study we found a statistically significant reduced risk of torsion recurrence in patients who underwent aspiration and drainage of ovarian cysts. Note, however, that fenestration has been associated with rare complications including uncontrolled bleeding from hyperstimulated and highly vascularized ovaries. Despite the theoretical concerns about the risks of fenestration, the current cohort had no intra-operative complications.

Three patients with recurrent torsion in our series had recurrent torsion of seemingly normal ovaries. We have described this condition in non-pregnant women elsewhere [6,15,16] and suggested that ovariopexy procedures should be considered. This procedure has been performed during pregnancy and described in two case reports in patients who experienced three episodes of torsion [17,18]. In the present study, none of the three patients with recurrent ovarian torsion had undergone ovariopexy. Although this procedure may impact the management of recurrent torsion during pregnancy, our findings tend not to support its routine use in all cases of recurrent torsion in pregnancy.

The treatment of adnexal torsion requires prompt surgery for adnexal de-torsion and the removal of associated adnexal pathologies (i.e., cyst aspiration for functional cysts and cystectomy for non-functional cysts). In standard practice, the surgical treatment of adnexal torsion is performed primarily by laparoscopy, since this procedure allows for early ambulation and reduced post-operative pain. Laparoscopy is also the primary surgical approach for adnexal torsion during pregnancy, but in these cases the unique intra-operative surgical risks as well as the long-term obstetrical outcomes should be considered.

LIMITATIONS
There are several limitations to the present study. This retrospective review precludes control of additional factors that may be associated with maternal torsion in pregnancy. Second, the number of patients experiencing recurrence was small. This may have influenced the results by either introducing selection bias and/or limiting the statistical significance of the analysis.

CONCLUSIONS
Pregnant women are at risk for recurrent torsion caused by functional ovarian cysts. Cyst aspiration and drainage are important in preventing further torsion episodes. Further multi-centric studies are required to validate our data prospectively.

Correspondence
Dr. Y. Melcer
Dept. of Obstetrics and Gynecology, Shamir Medical Center (Assaf Harofeh), Zerifin, 76300, Israel
Phone: (972-8) 977-9695
Fax: (972-8) 977-9089
email: ymelcer@gmail.com

References