

CASE SERIES / СЕРИЈА СЛУЧАЈЕВА

Adnexal torsion in pregnancy – relying on a six-year hospital experience in solving this problem

Dragana Maglić^{1,2}, Olivera Džatić-Smiljković^{1,2}, Milica Mandić², Ljubomir Srbinović², Rastko Maglić^{1,2}¹Narodni Front Clinic of Obstetrics and Gynecology, Belgrade, Serbia;²University of Belgrade, Faculty of Medicine, Belgrade, Serbia**SUMMARY**

Introduction Adnexal torsion is a serious complication in pregnancy that can lead to ovarian ischemia and necrosis. This retrospective study aimed to provide clinically driven guidelines for treatment of adnexal torsion during pregnancy.

Case report We analyzed data from 10 patients who underwent surgery for adnexal torsion between 2018 and 2023. The most common symptoms were pelvic pain, nausea, and vomiting. Laparoscopy and laparotomy were equally performed, with the choice depending on factors like trimester and tumor size. Unfortunately, adnexectomy was the most common surgery due to delayed presentation and advanced necrosis. Despite this, pregnancy outcomes were favorable, with most patients delivering live babies.

Discussion The most frequent adnexal tumor in pregnancy is the corpus luteum cyst. Several studies suggest laparoscopic management of adnexal torsion in pregnancy with excellent maternal and fetal outcomes. Both delayed diagnosis and intervention can lead to adnexal necrosis and hence increase the risk of miscarriage and maternal morbidity

Conclusion Expectant management is not recommended. Due to the increased risk of miscarriage and maternal morbidity, laparoscopy (detorsion and cystectomy) is the safest and most effective type of surgery in the first trimester. Laparotomy might be more appropriate in the third or late second trimester or with a very large adnexal mass.

Keywords: adnexal torsion; pregnancy; laparoscopy; laparotomy

INTRODUCTION

Torsion of the adnexa is an acute surgical condition and potentially lethal if left to be treated inappropriately [1]. It occurs due to rotation of the adnexa (the ovary and the fallopian tube) on their vascular axis, leading to partial or complete strangulation of the blood supply. The rotation may be either incomplete or complete, and in some cases, both the ovary and the fallopian tube are involved, leading to ischemia and necrosis [1]. Some of the causes of adnexal torsion are sexual intercourse, exercise, and sudden movement but the most common underlying cause is an ovarian cyst greater than 3 cm in size. Pregnancy and *in vitro* fertilization (IVF) are other risk factors. Pregnancy itself double the risk of torsion up to five times, and the rough estimate is five cases per 10,000 pregnancies [2, 3]. Ovarian torsion during pregnancy is an extremely emergent condition with dire consequences for both the mother and the fetus. Large ovarian cysts such as corpus luteum cysts and ovarian hyperstimulation syndrome as a result of assisted reproductive technologies (ART) are some other risk factors for adnexal torsion [4]. Torsion of the vascular pedicle leads to venous and lymphatic obstruction with resultant stasis, ischemia, and edema [5]. This ultimately results in necrosis of the tissue and can produce a local or pelvic inflammatory process. Early diagnosis

and timely surgery are required to save the adnexa and their function [1, 3, 5].

Symptoms and clinical presentation of adnexal torsion are typically non-specific, making it hard to diagnose. Common presentations in pregnant patients are subacute or acute severe pelvic discomfort, often accompanied by nausea and vomiting, and signs of acute abdomen [6]. These symptoms also occur with other diseases of the gynecologic and non-gynecologic etiology including ectopic pregnancy, rupture of a cyst, appendicitis, cholecystitis, ileus, and pelvic inflammatory disease [7], and therefore lead to diagnostic misdirection and treatment delays. It is particularly difficult to diagnose adnexal torsion during the second trimester of pregnancy since its symptoms may mimic premature labor or renal colic [8]. There are no tumor markers or blood tests that can differentiate between adnexal torsion and acute conditions during pregnancy [1].

Adnexal torsion during pregnancy is also made difficult by the enlarged uterus, especially after the first trimester [9]. The enlarged uterus pushes the adnexa posteriorly, making it difficult for physical and ultrasonographic assessment [9]. Ultrasonographic evidence of adnexal torsion consists of unilaterally swollen adnexa (large ovary with stromal edema and peripherally displaced follicles) and the presence of free fluid in the cul-de-sac. Doppler

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Beogradska 45
11000 Belgrade
Serbia
rastko.maglic@gmail.com

Table 1. Clinical manifestations associated with ovarian torsion in pregnancy

Clinical manifestation	n	%
Age (y)	27.1+/-3.9	
Parity	1.1	
Adnexal mass	7	70%
Pelvic pain	10	100%
Sudden onset	6	60%
Nausea and vomiting	5	50%
WBC count > 12 × 10 ⁹ /L	4	40%
Elevated CA 125	5	50%
Elevated CRP	4	40%
Fever	3	30%
Cul de sac fluid	7	70%

WBC – white blood cell; CA – cancer antigen; CRP – C-reactive protein

Table 2. Surgical results associated with ovarian torsion in pregnancy

Operative methods	n	%
Laparoscopy	5	50%
Laparotomy	5	50%
Mass size (cm)	7.8 ± 3.6 × 5.7 ± 3.1	(4.7–12) × (1.7 × 11)
Operative time (min)	40.1 ± 28.4	20–70
Blood loss (mL)	46.5 ± 34.3	10–20
Left side of adnexal mass	3	30%
Right side of adnexal mass	7	70%
Detorsion	1	10%
Cystectomy and detorsion	1	20%
Salpingectomy and detorsion	1	30%
Adnexectomy	6	40%
Salpingectomy	1	10%

Table 3. Comparison outcome between laparoscopy and laparotomy in pregnant women with ovarian torsion

Laparoscopy (n = 5)	Laparotomy (n = 5)	p	
Age (y)	32.4 ± 2.7	27.4 ± 5.98	0.127
Parity	1.20 ± 0.45	1.2 ± 0.45	1.00
BMI (%)	21.7 ± 3.1	22.8 ± 1.9	0.167
Tu mass size (cm)	6.92 ± 2.6	10.2 ± 3.39	0.123
Operative time (min)	28.2 ± 11.58	60 ± 10	0.02
Hospital stay (d)	4 ± 1.9	9.8 ± 7.1	< 0.001
Blood loss (mL)	41.8 ± 24.1	51.7 ± 58.4	0.241
Live term baby	4 (80%)	5 (100%)	0.347
Preterm delivery	1 (20%)	2 (40%)	0.312
Abortion	1 (20%)	0	

ultrasound may demonstrate diminished or no blood flow in the vascular pedicle but not always [10].

Ischemia and necrosis of the ovary and fallopian tube occur due to restricted or absent blood supply. Delayed diagnosis and surgical intervention can lead to permanent destruction of the ovaries, tissue necrosis, and signs of an acute abdomen, which may jeopardize the pregnancy with miscarriage or premature delivery [6].

Management of adnexal torsion is either expectant or, most often, surgical. They pose risks for both the fetus and the mother. Where adnexal torsion is incomplete, expectant management (wait-and-watch without treating) is acceptable because the adnexa will spontaneously untwist

and improve. However, in complete and multiple torsions, expectant management will lead to irreversible damage [11].

As mentioned, the pregnant uterus with distension, and, if any, maternal obesity, can hamper diagnosis by limiting visualization under transabdominal ultrasonography [12]. In the second trimester, visualization of the adnexa by transvaginal ultrasonography may not be sufficient due to their lateral position outside the pelvis. Computed tomography scans should be avoided lest there be fetal exposure to radiation [5]. Magnetic resonance imaging is helpful if it is available, but the most accessible imaging tool remains ultrasound [13]. Bloating, limited field of view, and altered anatomy during late pregnancy can all pose challenge to ultrasound visualization of the adnexa and contribute to the challenge of diagnosing adnexal torsion [9, 13].

Once emergency diagnosis, and short incurative episode of expectant management in some, has been achieved, adnexal torsion is treated by laparoscopic or open surgery [14]. The most common surgical procedures are cystectomy, detorsion, or, in the case of tissue necrosis, salpingo-oophorectomy [14]. Compared to laparotomy (open), laparoscopy is less invasive, involves less blood loss and thromboembolic consequences, and leads to faster patient recovery [15]. However, during the second and third trimester, the distended abdomen and intraperitoneal pressure caused by pneumoperitoneum (carbon dioxide gas insufflation of the peritoneum) can complicate laparoscopic visualization and surgical management and even carry greater risk for both the mother and fetus [14, 15].

The objective of this study was to provide clinically driven guidelines for the treatment of adnexal torsion during pregnancy, based on six-year single-center clinical data. Given the rarity of adnexal torsion during pregnancy, we aimed to analyze the conditions leading to it, report on symptoms, and evaluate treatment options and their effects on pregnancy outcomes.

CASE SERIES

This retrospective study was conducted at the Narodni Front Gynecology and Obstetrics Clinic in Belgrade, Serbia. We analyzed the medical records of pregnant patients diagnosed with adnexal torsion between 2018 and 2023. During this six-year period, 10 cases were admitted and treated. We excluded patients with a preoperative diagnosis of adnexal torsion that was not confirmed during surgery.

We analyzed demographic data, including age and parity, presenting symptoms and clinical signs, surgical approach (laparoscopy vs. laparotomy) and type of surgery (cystectomy, salpingectomy, detorsion, adnexectomy), duration of surgery, estimated blood loss, histopathological findings, and obstetric outcomes (miscarriage, gestational age at delivery, and mode of delivery). Additionally, we analyzed laboratory data, including white blood cell (WBC) count, C-reactive protein (CRP) levels, and cancer antigen (CA) 125 levels. We also reviewed ultrasound findings, including the size and location (left or right) of the adnexal mass, ovarian stromal blood flow [resistance index (RI)], and

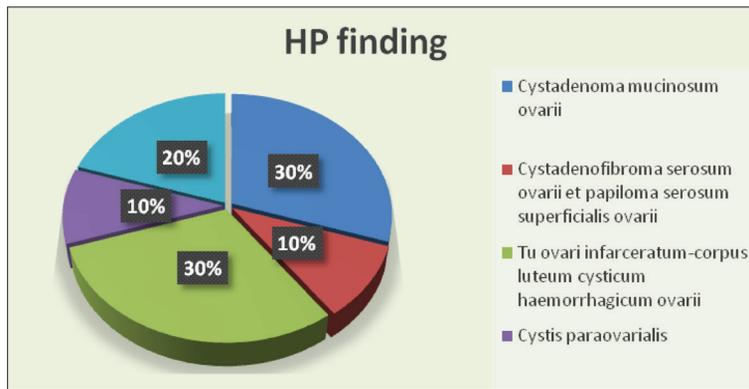


Figure 1. Histopathological (HP) finding

pregnancy status after surgery (gestational age at delivery, miscarriage, preterm delivery). Transvaginal and transabdominal ultrasound examinations were performed using the Acuson Sequoia 512 ultrasound system (Acuson, Mountain View, CA, USA; vaginal transducer 4-9 MHz; abdominal transducer 1.3 MHz. – 5.7 MHz).

Due to the small sample size, statistical analysis was limited to descriptive statistics, including mean, standard deviation (SD), standard error (SE), minimum-maximum range, median, and mode. Microsoft Excel (Microsoft, Redmond, WA, USA) was used for statistical analysis.

In 43,835 deliveries during the study period, there were 10 patients with preoperative diagnosis of adnexal torsion, which was proven by operation, with a rate of 2.3 per 10,000 pregnancies. Nine patients (90%) had adnexal torsion in the first and second trimesters, and one patient had torsion at the time of delivery. The mean age of the patients was 27.1 ± 5.1 years (18–36 years), and the mean parity was 1.1 ± 0.4 (1–3). Three (30%) of them had a history of pelvic surgery, two for ovarian cystectomy and one for an ectopic pregnancy.

The most common presenting symptom was pelvic pain (100%), which was of sudden onset in the majority, followed by vomiting and nausea (50%). Seven (70%) of the patients had palpable adnexal mass on gynecological examination, who were at the first and second trimesters of gestation. Normal blood reports were obtained in most of them, except for four of them with a mild elevation in WBC count ($> 12 \times 10^9/L$) and CRP levels. CA 125 was raised in 50% of the patients. Adnexal mass was present in all (100%) patients, right-sided in 70%, with a mean diameter of $7.8 \pm 3.6 \times 5.7 \pm 3.1$ cm. Seven patients had free fluid in the cul-de-sac. Eight patients had normal uterine artery (RI) and ovarian stromal blood flow on Doppler ultrasound. In two of them, there was an undetectable blood supply to the adnexa due to necrosis of tissue and severe strangulation of the vascular pedicle.

Equal frequency was applied in using laparoscopy and laparotomy. Patients in the first trimester, particularly with a small adnexal mass, were treated by laparoscopy as the procedure of choice. Laparotomy was reserved for patients in the third trimester or for bigger adnexal masses.

Adnexectomy was the most common procedure (60%), which likely reflects referral and diagnostic delays, and, as a result, more advanced disease at surgery.

As for comparison between laparoscopy and laparotomy, we noted that hospital stay and operative time were both significantly shorter in the laparoscopy group. There was no difference between the two groups regarding the outcome of pregnancy. There was only one miscarriage (10%).

Histopathological assessment was carried out that proved that 30% of the patients had an ovary mucinous cystadenoma, 30% had a hemorrhagic corpus luteum cyst with ovarian infarction, and 20% had a large follicular cyst as the cause for adnexal torsion. There were no postoperative infections (Figure 1).

Ethics: This study was approved by the Ethics Committee of the Narodni Front Obstetrics and Gynecology Clinic, Belgrade, Serbia (No: 22008/2024/022318).

DISCUSSION

The reported incidence of adnexal torsion varies in the literature, with rates of 5.9 per 100,000 in non-pregnant women and 1.6–5 per 10,000 in pregnant women [2]. Adnexal torsion is more common during pregnancy due to the presence of corpus luteum cysts and increased ovarian mobility caused by hormonal changes [5]. Our study found a lower incidence of 0.023%, which may reflect differences in population characteristics or reporting practices.

Eight of our patients (80%) also had in the first and initial second trimester (4th to 16th week gestation) a palpable adnexal mass on one side. The smaller uterine size during this period facilitated palpation. As was previously cited [6], the first trimester was the most common presentation time for adnexal torsion (60% of our study cases). The clinical presentation of adnexal torsion in our series was similar to that seen in non-pregnant women, with non-specific symptoms most being the most common. Sudden unilateral pelvic pain was the most common symptom, most commonly worsening with time and nausea and vomiting [16]. Fever and acute abdomen can be present as necrosis of tissue progresses [7]. Differential diagnosis in pregnancy is based on gestational age and may be miscarriage, hematoma retroplacental, and rupture of the uterus [8]. In the current study, all the patients presented with acute onset of pelvic pain on one side, and 50% presented with nausea and vomiting.

A palpable adnexal mass, typically larger than 5 cm, was detected on examination in 70% of our patients, most of whom presented in the first trimester. Palpation was painful and discomforting [1]. This relatively high rate of palpable masses likely reflects the more advanced gestational age of presentation in our series. While there is no specific laboratory test for adnexal torsion, we recorded subtle increases in WBC count, CRP, and CA 125 in some patients. It is thus conceivable that subtle CRP and CA 125 increases are non-specific but possibly evocative of adnexal torsion [10]. Ultrasound remains the gold standard

for adnexal torsion diagnosis in pregnancy, with a readily available non-invasive imaging modality. Color Doppler and measurement of RI in the ovarian stroma can be employed to assess the ovarian blood supply and monitor possible ischemia. The core of ultrasound diagnosis of adnexal torsion is an edematous swollen ovary (larger than the contralateral one), low or absent blood flow, and free fluid in the pelvis [10]. Incidence of ovarian torsion after stimulation of ovaries in IVF is 0.025% to 0.2% [17]. Adnexal torsion is enhanced by assisted reproductive techniques through risk of ovarian hyperstimulation syndrome and ovarian enlargement. In our study, 40% of patients had a history of IVF, which establishes the association between ART and adnexal torsion [17].

The most common adnexal tumor in pregnancy is the corpus luteum cyst, but dermoid cysts, and cystadenomas are more common in non-pregnant women [17]. The literature suggests that the most frequent histopathological diagnoses of adnexal torsion are mature cystic teratoma (46.3%), serous cystadenoma (17.5%), and mucinous cystadenoma (11.3%) [17]. Our findings were for the most part consistent with these reports, with corpus luteum cysts being found in 40%, mucinous cystadenomas being found in 30%, and serous cystadenomas in 10%. Greater adnexal masses (greater than 5 cm) have a higher risk of torsion. Risk appears to be highest between 10- and 17-weeks' gestation [7]. The average size of the tumor was 7.8×5.8 cm in our experience, and 40% of patients had torsion occur between 10 and 17 weeks, consistent with this. While a few reports have suggested that cysts larger than 15 cm will be malignant [8], in our population, we could not find any malignant adnexal tumors.

Several studies suggest laparoscopic management of adnexal torsion in pregnancy with excellent maternal and fetal outcomes [11]. A meta-analysis of 163 studies by Didar et al. [15] showed laparoscopy to be the most common surgical intervention (56.88%), and open surgery was performed in only 10% of patients [16]. Detorsion with cystectomy (29.06%), salpingo-oophorectomy (27.32%), and detorsion (18.31%) were the most frequent operations [14]. Laparotomy and laparoscopy were performed with the same frequency (50% each) in our study. Laparotomy was reserved for enormous adnexal masses or late gestational ages. Adnexectomy was the most common procedure (70%), perhaps due to delay in diagnosis and presence of advanced disease (tissue necrosis or gangrene) at the time of surgery. Cystectomy, salpingectomy, or detorsion was performed in the remaining 30% of patients. A retrospective study of 60 adnexal torsion patients found the likelihood of ovarian preservation was highest if surgery were performed within four hours of symptom onset (83% vs. 56% > 4 hours, $p = 0.39$) [5, 6]. This indicates the need for urgent early diagnosis and intervention to maximize the chances of preserving the ovary. Compared to open surgery, laparoscopy

has several advantages, including less invasiveness, less blood loss, less postoperative pain, faster recovery, lower risk of thromboembolism, and shorter hospital stay [15]. Pregnancy laparoscopy may also enhance visualization and minimize uterine manipulation, with potential reduction in the risk of miscarriage, preterm labor, and premature rupture of membranes. There are several studies that have established the safety and feasibility of laparoscopy during pregnancy [14]. American Society of Gastrointestinal and Endoscopic Surgeons recommendations prefer the use of laparoscopy in the management of acute abdominal conditions during pregnancy [12].

Our retrospective review of laparoscopy versus laparotomy for adnexal torsion in pregnancy revealed no marked difference in pregnancy outcome. However, patients who underwent laparoscopy experienced less pain, less blood loss, shorter hospital stay, and earlier recovery. Based on these findings, we recommend laparoscopy as the method of choice for adnexal torsion in pregnancy, especially during the first trimester.

CONCLUSION

Adnexal torsion is a pregnancy complication that, albeit unusual, is severe. The most common presenting symptoms of acute-onset unilateral pelvic pain, nausea, and vomiting should prompt urgent referral to a tertiary facility. Both delayed diagnosis and intervention can lead to adnexal necrosis and hence increase the risk of miscarriage and maternal morbidity [18, 19]. While there are no specific laboratory tests for adnexal torsion, an elevated WBC count and CA 125 can be suggestive. ART can increase the risk of adnexal torsion in pregnancy.

The most frequent underlying reason for adnexal torsion in pregnancy is the corpus luteum cyst. Expectant management is not typically recommended due to increased miscarriage and maternal morbidity. Laparoscopy appears to be the safest and most efficacious type of surgery in the first trimester [18]. Detorsion and cystectomy are the operations of choice. Laparotomy might be more appropriate in the third or late second trimester or with a very large adnexal mass. After 20 weeks of gestation, the distended uterus may complicate laparoscopic visualization and manipulation, and the Trendelenburg position may impair maternal respiratory function.

Our findings show that laparotomy and laparoscopy are equivalent in pregnancy outcome but that laparoscopy has some perioperative advantages. We would recommend a multicenter study of a larger group of patients to further evaluate the optimal management of adnexal torsion during pregnancy.

Conflicts of Interest: None declared.

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Торзија аднекса у трудноћи – ослањање на шестогодишње болничко искуство у решавању овог проблема

Драгана Маглић^{1,2}, Оливера Џатић-Смиљковић^{1,2}, Милица Мандић², Љубомир Србиновић², Растко Маглић^{1,2}

¹Гинеколошко-акушерска клиника „Народни фронт“, Београд, Србија;

²Универзитет у Београду, Медицински факултет, Београд, Србија

САЖЕТАК

Увод Торзија аднекса је озбиљна компликација у трудноћи која може довести до исхемије и некрозе јајника. Ова ретроспективна студија имала је за циљ да пружи клинички засноване смернице за лечење торзије аднекса током трудноће. Анализирали смо податке 10 болесница које су оперисане због торзије аднекса у периоду од 2018. до 2023. године.

Прикази болесника Најчешћи симптоми били су бол у карлици, мучнина и повраћање. Лапароскопија и лапаротомија су примењиване подједнако често, при чему је избор зависио од фактора попут триместра трудноће и величине тумора. Нажалост, најчешћи захват била је аднексектомија због касног јављања пацијенткиња и узапредовале некрозе. Упркос томе, исходи трудноће били су повољни, а већина пацијенткиња родила је живу новорођенчад.

Дискусија Најчешћи аднексални тумор у трудноћи је циста жутог тела. Више студија указује на то да лапароскопско лечење торзије аднекса у трудноћи даје одличне исходе по мајку и плод. Касно постављање дијагнозе и одлагање операције могу довести до некрозе аднекса и тиме повећати ризик од побачаја и морбидитета мајке.

Закључак Експективно лечење се не препоручује. Због повећане могућности побачаја и мајчиног морбидитета, лапароскопија (деторзија и цистектомија) најбезбеднија је и најефикаснија врста операције у првом тромесечју. Лапаротомија може бити прикладнија у трећем или касном другом тромесечју или код великих аднексалних маса.

Кључне речи: торзија аднекса; трудноћа; лапароскопија; лапаротомија