Isolated Fallopian Tube Torsion – A Challenge for the Timely Diagnosis and Treatment

Radoica Jokić1,2, Jovan Lovrenski1,3, Aleksandra Lovrenski4, Veličko Trajković2
1University of Novi Sad, Medical Faculty, Novi Sad, Serbia; 2Pediatric Surgery Department, Institute for Children and Adolescents Health Care of Vojvodina, Novi Sad, Serbia; 3Radiology Department, Institute for Children and Adolescents Health Care of Vojvodina, Novi Sad, Serbia; 4Pathology Department, University of Novi Sad, Medical Faculty, Novi Sad, Serbia

SUMMARY
Introduction Isolated torsion of the fallopian tube is a rare cause of acute lower abdominal pain and infrequent indication for surgical treatment.

Case Outline A 16-year-old girl was referred to the hospital due to the non-specific symptoms over period of a few months and pain acutization in the right infraumbilical region lasting for two days. Complete laboratory analyses were normal. Ultrasound examination revealed a round mass (23×14 mm) within the right fallopian tube with color Doppler whirlpool sign, normal ovary, and a simple ipsilateral paratubal cyst (50×40 mm). Laparoscopy showed a dilated and two times torquated right fallopian tube, as well as signs of chronic appendicitis. The tube was twisted about its longitudinal axis and it was livid, but not gangrenous. After appendectomy, fallopian tube was detorquated, cyst extracted and preservation of the tube was performed. Postoperatively, antibiotic therapy was administered based on antibiogram. On follow-up examinations within the next four months postoperative course was uneventful.

Conclusion Since there are no pathognomonic symptoms, clinical or laboratory findings, diagnosis of this condition is challenging. Familiarity with Doppler whirlpool sign can enable a timely diagnosis and treatment of isolated fallopian tube torsions. However, the diagnosis is rarely made before operation. Unlike in our case, surgery is often performed too late, and delay of intervention may result in failure to save the fallopian tube.

Keywords: fallopian tube; torsion; ultrasound; laparoscopy

INTRODUCTION
Isolated fallopian tube torsion (IFTT) is an uncommon cause of acute, lower abdominal pain in women of reproductive age with an incidence of one in one-and-a-half million [1]. The exact etiology of fallopian tube torsion is unknown. Since the symptoms are non-specific, diagnosis can be challenging and difficult. Ultrasonography and computed tomography (CT) can demonstrate changes strongly suggesting tubal torsion, but definite diagnosis, as well as an adequate treatment, require surgical exploration [2]. We report a case of IFTT in a 16-year-old girl with special emphasis on ultrasound characteristics and surgical treatment.

CASE REPORT
A 16-year-old girl was referred to our hospital due to the sudden symptoms in the form of acute pain in the right infraumbilical region lasting for two days. The patient reported un-specific symptoms over a period of several months. She had been hospitalized twice because of abdominal pain and bloating. Her menstrual history was as follows: the menarche was four years earlier, irregular menstruation (four to five per year), and the last period was one month prior to admission. Because of polycystic ovaries, she received hormonal therapy (combined oral contraceptives with anti-androgen progestin) which was ceased for evaluation of current hormonal status. She was not sexually active.

On physical examination abdomen was soft, but deep palpation revealed soreness in ileocecal region, without the presence of resistance and mass. At first, this finding raised suspicion of appendicitis, but complete blood count, and serum chemistries were normal.

Ultrasound examination showed normal uterus and both ovaries. A moderate amount of free fluid was present within the pelvis. Within the right fallopian tube a round mass (23×14 mm) slightly resembling a target was revealed. Color Doppler sonogram showed the circular vessels within this mass, presenting a whirlpool sign (Figure 1). Paratubal thin-walled cyst of 50×40 mm, with a clear fluid content, was also visualized on the right side (Figure 2).

The acute onset of pain, physical finding, and detection of an adnexal mass with whirlpool sign on ultrasonography raised suspicion of torsion of the right adnexal structures. It was decided that the patient should undergo urgent laparoscopy, which was therefore performed 48 hours after the beginning of acute abdominal symptoms.

Correspondence to:
Jovan LOVRENSKI
Doža Đerđa 17, 21000 Novi Sad
Serbia
jolo221177@gmail.com
The standard laparoscopic technique was performed. Creating a pneumoperitoneum of 12 mmHg and the placement of the first trocar were performed using an open Hasson technique. Two working iliac ports of 5mm were placed as well. Laparoscopy showed a dilated and isolated two times torquated right fallopian tube and signs of chronic appendicitis. The tube was twisted about its longitudinal axis, with edema and enlargement of the fimbrial end. It was livid, but not gangrenous. Along swollen fallopian tube paratubal cyst described on ultrasound, that was most likely causing the 720° torquation of the tube, was found (Figure 3).

Appendectomy, detorsion of the right fallopian tube, paratubal cyst extirpation and preservation of the tube were performed. The appendix and right paratubal cyst were sent to pathohistological analysis. The ovaries, as well as the left fallopian tube were normal in appearance. Cultures from the fallopian tube and peritoneal fluid were obtained for bacterial analysis. Pseudomonas species were isolated. Histopathology confirmed chronic appendicitis and simple paratubal cyst.

The patient was administered antibiotics therapy based on antibiogram, and on the fifth day after surgery she was discharged from the hospital in a good general condition. Postoperative course within the period of four months was uneventful.

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**Figure 1.** a) Gray scale sonogram showing a round mass faintly resembling a target (arrows) within the right fallopian tube, with a free fluid around it (asterisks). b) Color Doppler sonogram presenting a whirlpool sign within the described mass (arrows). Free fluid (asterisks).

**Figure 2.** Gray scale sonogram of the right-sided paratubal thin-walled cyst – 50×40 mm (C), with a clear fluid content. Next to the cyst, part of the edematous right fallopian tube was observed (arrows). Free fluid (asterisk).

**Figure 3.** a) Laparoscopically revealed two times torquated right fallopian tube (arrows), and a normal appearing right ovary (O). b) Laparoscopic photograph showing a paratubal cyst on the right (C), and a normal ipsilateral ovary (O).
DISCUSSION

IFTT is a rare gynecological cause of acute lower abdominal pain, especially in young teenage girls [3]. It primarily affects ovulating women, and is rarely seen in postmenopausal women [1]. Also, it is a very rare medical emergency in premenarchal girls [4].

It was first described in 1890 by Bland-Sutton [5] as a rare cause of lower abdominal quadrant pain and since then several hundred cases have been reported in the literature. Risk factors for IFTT can be divided to intrinsic factors (excessive length of tube or spiral course, hydroosalpinx, hematosalpinx, tubal ligation, tubal neoplasm, pelvic inflammatory disease, etc.), and extrinsic factors (adhesions, adnexal venous congestion, adjacent ovarian or paraovarian masses, uterine masses, gravid uterus, trauma, etc.) [6]. However, almost half of IFTT (47%) was reported not to have any associated pathology [7]. In our case, IFTT was most probably caused by a large paratubal cyst. Markhardt et al. [8] described one case of isolated torsion of the right fallopian tube occurring two days after onset of the first menses, thus pointing to a possible link between menstrual bleeding and IFTT.

The clinical presentation of a fallopian tube torsion is non-specific and therefore is a challenge for the clinician to recognize and differentiate from multiple other etiologies. Acute severe lower abdominal pain is always present. Nausea and vomiting may accompany the pain, but fever is rarely present. On clinical exam, findings include abdominal tenderness with or without peritoneal signs. Laboratory findings are usually non-specific, as they were in our patient. Necrosis can cause leukocytosis, and the sedimentation rate or C-reactive protein might be elevated [6, 9].

Differential diagnosis includes ovarian torsion, ruptured ovarian cyst, ectopic pregnancy, endometriosis, pelvic inflammatory disease, degenerative leiomyoma, acute appendicitis, and some urinary conditions [10, 11].

Harmon et al. [12] suggested that IFTT predominantly appears on the right side, possibly because of partial immobilization of the left tube by its proximity to the sigmoid mesentery, as well as the relatively reduced amount of venous flow on the right side. Also, according to the same authors, it is more likely that patients with right-sided lower abdominal pain will be operated on because of suspicion of appendicitis. However, Wong et al. [13] reported series of six cases in which isolated tubal torsion occurred on the left side in five patients.

Local necrosis of the tube can also result in irreversible damage to the ipsilateral ovary, but more serious complications include tube necrosis and gangrenous transformation, which can be followed by superinfection and peritonitis [2, 6].

The normal fallopian tube is rarely visible on ultrasonography because of its narrow diameter and lack of clear echogenic features. Occasionally, a fallopian tube can be seen if it is surrounded by fluid, but a tube that is visible on ultrasonography is probably abnormal. The spectrum of ultrasonographic findings varies depending on the adnexal pathology, the degree of severity, and the duration of adnexal torsion [14]. Reported ultrasonographic findings include a normal-appearing uterus and ovaries with normal blood flow, free fluid, a dilated tube with thickened, echogenic walls, and internal debris or a convoluted echogenic mass thought to represent a thickened, torse tube [2]. Harmon et al. [12] reviewed the hospital charts and imaging studies of the eight girls diagnosed with and treated for IFTT at Columbus Children’s Hospital between January 1995 and June 2006. The most common finding on ultrasonography was a complex cystic pelvic mass in the midline and normal uterus and ovaries. Three of the eight girls had CT imaging performed. In 63% of the girls, the uterus was deviated toward the side of torsion. The correct preoperative diagnosis was made in only one girl. On ultrasonography, the most common problem is to differentiate between IFTT and total adnexal torsion, and in these cases the use of color Doppler ultrasound is highly recommended. According to Origni et al. [15] an IFTT should be considered when a detailed Doppler flow ultrasound shows a normal ovary and presence of a pelvic cyst.

The whirlpool sign represents a tissue mass twisted around a central axis, which is visualized by moving the ultrasound probe back and forth along the axis of suspected torsion. A twisted vascular pedicle is shown, and Doppler sonogram reveals circular vessels within the mass. The whirlpool sign is a pathognomonic sonographic sign of midgut volvulus in newborns and young infants, and has also been described in patients with testicular and ovarian torsion [16, 17, 18]. It is also considered specific for IFTT, along with a presence of normal ovaries, which was confirmed in our patient [19]. However, all the authors claiming whirlpool sign being pathognomonic for diagnosis of IFTT used transvaginal approach, while we performed a standard ultrasound examination with a linear probe of 7–9 MHz [19, 20].

In our case, we did not perform CT, but reported CT findings of IFTT include an adnexal mass, a twisted appearance of the fallopian tube, dilated tube greater than 15mm, a thickened and enhancing tubal wall and luminal CT attenuation greater than 50 HU consistent with hemorrhage. Secondary signs include free intrapelvic fluid, peritubular fat stranding, enhancement and thickening of the broad ligament and even regional ileus [21]. However, occasionally, CT examination is not more conclusive than ultrasound [22], and therefore it is very important for radiologists to know ultrasonographic signs suggesting IFTT, because they often present the first line of diagnostic imaging. Magnetic resonance imaging (MRI) is also reported as a valuable diagnostic tool in recognizing adnexal torsion with most common findings of tube thickening, ascites, and uterine deviation to the twisted side [23]. Recently, Aydin et al. [24] first reported the whirlpool sign on MRI in patient with IFTT, which was observed using T2-weighted imaging.

MRI and ultrasound examination are especially helpful in the young or pregnant patients, because reliable information is provided without ionizing radiation.

Torsion of the adnexa is a surgical emergency [25, 26, 27]. Until recently, the common approach to the twisted ischemic adnexa was salpingo-oophorectomy [28]. Since
most of the patients are in their reproductive years, maximal efforts should be made to preserve fertility. Consequently, laparoscopy is currently the most specific diagnostic tool for evaluating torsion, and also a laparoscopic adnexal detorsion is the treatment of choice [6, 28-31]. Early laparoscopy is the reference standard in the diagnosis and treatment. Recovery after laparoscopy is faster, and laparoscopy also causes fewer pelvic adhesions, which is particularly important for women of reproductive age, who wish to preserve their fertility. A complete resection can be performed when the tissue is gangrenous, if there is a suspicion of tubal or ovarian neoplasm, or even when the woman has previously completed her family [6]. The delay in diagnosis of IFTT leads to irreversible consequences, such as the loss of the tube [32]. Laparoscopic differentiation of paratubal and parovarian cysts from ovarian cysts is usually easy. The characteristic laparoscopic finding is the crossing of blood vessels over the surface of the cyst. Different laparoscopic modalities have been used to extract paratubal or parovarian cysts. The recurrence of cysts which required laparoscopy or laparotomy has been reported [29]. In conclusion, although IFTT has already been reported it keeps surprising both clinicians and radiologists, so occasional reports of IFTT cases are important as a reminder and a way of saving tubes in ovulating women. Its diagnosis can rarely be made before operation, often due to an absence of characteristic sonographic whirlpool sign, or due to inexperience of radiologist or gynecologist to recognize it. For that reason, laparoscopy is often necessary to establish the diagnosis. Laparoscopic treatment in women of reproductive age is warranted as a means of preserving fallopian tube integrity, and thus maintaining fertility. Unfortunately, in many cases, unlike in ours, surgery is performed too late for tube preservation. Therefore, IFTT has to be considered in differential diagnosis of acute pelvic pain, both among clinicians (surgeons/pediatric surgeons/gynecologists) and radiologists.

REFERENCES

КРАТАК САДРЖАЈ

Увод
Изолована торзија јајовода је редак узрок акутног бола у доњем абдомену и ретка индикација за хируршко лечење.

Приказ болесника
Шеснаестогодишња девојчица упућена је у болницу због више месечних неспецифичних симптома и акутног бола у десној инфрауфралној регији који је трајао два дана. Напази целокупних лабораторијских анализи биле су нормалне. Упреважени преглед показао је округласту масу (23×14 mm) у десном јајоводу са колор доплер знаком вира, нормалан јајник и просту паратубарну цисту (50×40 mm). Лапароскопија је показала дилатиран и два пута торквиран десни јајовод, као и знаке хроничног апендицитиса. Јајовод је био увијен око своје лонгитудиналне осе, ливидан, али не и гангренозан. Након апендектомије јајовод је деторквиран, циста екстирпирана, а јајовод сачуван. После операције је, према антибиотичкој терапији, налази поред оперативних контролних прегледа у наредним четири месеци били су нормални.

Закључак
Будући да нема патогономоничних симптома, клиничких лабораторијских налаза, дијагноза овог стања представља изазов. Познавање знака вира на колор доплер у прегледу може омогућити постављање ране дијагнозе и правовремено лечење изоловане торзије јајовода. Међутим, дијагноза се ретко поставља пре операције. За разлику од приказаног случаја, хируршко лечење се често врши пре-десет дана, а закаснела интервенција може довести до немогућности да се јајовод сачува.

Кључне речи: јајовод; торзија; ултразвук; лапароскопија