


Isolated tubal torsion in pediatric ages by single surgeon's experience

Görkem Bulut 

While tubo-ovarian pathologies are considered to be more common among women in reproductive ages isolated fallopian tubal torsion (IFTT) is still a rare situation and challenging to diagnose. Isolated fallopian tubal torsion in pediatric age group has an estimated prevalence of 1 in 1.5 million women.^[1,2] It is usually published as case reports or case series in the literature.^[3-6] Schuch et al.^[7] reported a total of 102 cases with adnexal pathologies in which only 12 were cases of IFTT.

Patients typically manifest with complaints such as any acute abdominal situation: sudden onset of abdominal pain with or without; nausea, vomiting, or urinary problem. It is commonly diagnosed by performing laparoscopy or laparotomy, since Doppler ultrasound (DUS), computed tomography (CT), or magnetic resonance imaging (MRI) are insufficient to make the definitive diagnosis. Its rarity could cause a radiologist or surgeon to misdiagnose and IFTT can be overlooked. The delay in the management of this pathology can have important implications, since the torsion of the tubes can cause ischemia and necrosis.^[8] In the present study, we aimed to investigate rare situation of IFTT in pediatric patient population and to draw attention to these patients whose inner reproductive organs can be salvaged by minimally invasive surgery.

Abstract

Objectives: This study aims to investigate rare situation of isolated fallopian tubal torsion (IFTT) in pediatric patient population and to draw attention to these patients whose inner reproductive organs can be salvaged by minimally invasive surgery.

Patients and methods: Between January 2016 and December 2024, a total of 13 female patients (mean age: 14.1±1.5 years; range, 11 to 16 years) with IFTT were retrospectively analyzed. All patients were laparoscopically diagnosed and treated by a single surgeon. Data including demographic and clinical characteristics, imaging findings, six-month follow-up data, and postoperative outcomes were recorded.

Results: All patients except for one were postmenarchal and they were all sexually inactive. All patients had symptoms of severe abdominal pain and nausea. The mean time from onset of symptoms to admission was 9.6±21.6 (range, 6 to 72) h. Rebound and severe tenderness were found in all of their physical examinations. All except for one were treated with laparoscopic tubal detorsion and excision of the paratubal cysts. The remaining patient who was admitted on Day 3 following the onset of symptoms underwent laparoscopic salpingectomy, preserving the ovary.

Conclusion: Although rare, being vigilant and observant against IFTT even in premenarchal girls would help many surgeons diagnose and perform a prompt exploration avoiding organ loss, thereby preserving fertility.

Keywords: Adnexal torsion, isolated Fallopian tubal torsion, salpingeal torsion.

PATIENTS AND METHODS

This single-center, retrospective study was conducted at Aydın Gynecology and Pediatric Diseases Hospital, Department of Pediatric Surgery between January 2016 and December 2024. Medical data of a total of 69 female children who needed surgery due to tubo-ovarian diseases were collected. Inclusion criteria were as follows: Being diagnosed only with fallopian tubal torsions. Exclusion criteria

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Correspondence: Görkem Bulut, MD.

E-mail: drgorky84@hotmail.com

Department of Pediatric Surgery, Aydın Gynecology and Pediatric Diseases Hospital, Aydın, Türkiye

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were as follows: Having common adnexial diseases. A total of 13 female patients (mean age: 14.1 ± 1.5 years; range, 11 to 16 years) with IFTT who met the inclusion criteria were recruited. A written informed consent was obtained from the parents and/or legal guardians of the patients. The study protocol was approved by the Ege University Medical Research Ethics Committee (date: 09.05.2024, no: 24-5T/32). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Data including demographic and clinical characteristics, imaging findings, six-month follow-up data, and postoperative outcomes were recorded. All patients were laparoscopically diagnosed and treated. Surgery was performed under general anesthesia by inserting one 5-mm camera port through the umbilicus accompanying two other 5-mm work-ports placed on both lateral pelvic regions. The abdomen was insufflated with carbon dioxide with a pressure of 12 mmHg and flow of 4 L/min. White blood cell (WBC) counts and C-reactive protein (CRP) values were noted. A WBC of 4.5 to $11.0 \times 10^9/L$ and a CRP below 3 to 5 mg/L were considered normal. Pathological samples were taken during surgery to differentiate any malignancy or to define necrosis. The samples were analyzed using the hematoxylin-eosin staining.

Statistical analysis

Statistical analysis was performed using the IBM SPSS for Windows version 22.0 software (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in mean \pm standard deviation (SD), median (min-max) or number and frequency, where applicable. A p value of <0.05 was considered statistically significant.

RESULTS

All patients except for one were postmenarchal and they were sexually inactive. All patients had the symptoms of severe abdominal pain and nausea. The mean time from onset of symptoms to admission was 9.6 ± 21.6 (range, 6 to 72) h. Rebound and severe tenderness were found in all of their physical examinations. Three (25%) patients had a high WBC count, while the other patients' WBC counts were within the normal range. The CRP values were normal in all of the patients. All patients had DUS examinations and four of them also had a CT scan which showed pelvic homogeneous cystic masses.

These four patients did not have prior DUS at the time of their referral to our clinic, as their prior clinic lacked DUS imaging capabilities. Their DUS scans were taken after they were admitted to our clinic. The DUS scans were reported with ovarian simple cysts in 10 patients (77%) and with paratubal cysts in three (23%). Three DUS scans (23%) were reported with an ovarian torsion. Other scans indicated that the ovaries' blood circulations were normal.

There were no significant differences between the frequency of sides ($p > 0.05$). The right-to-left

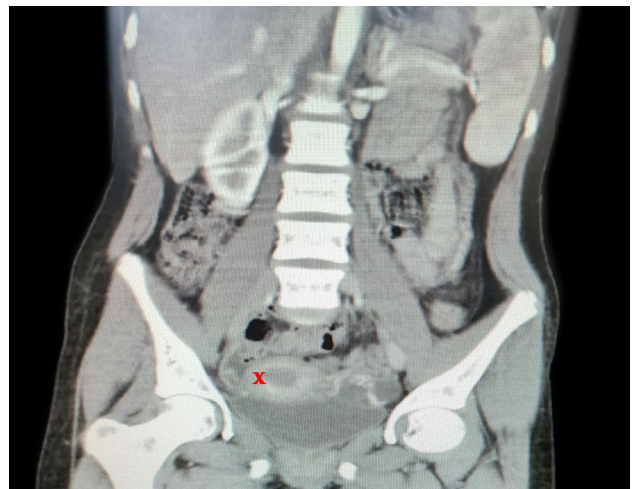


Figure 1. Computed tomography image of the patient with tubal necrosis (Right adnexal mass marked with an X).

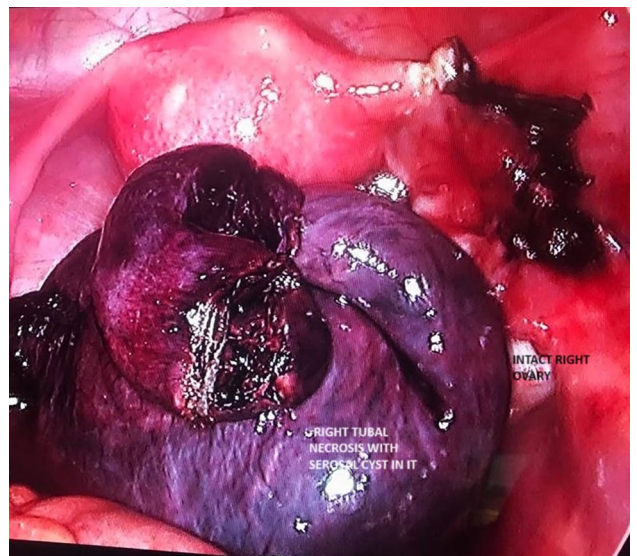


Figure 2. Laparoscopic imaging of the patient with tubal necrosis.

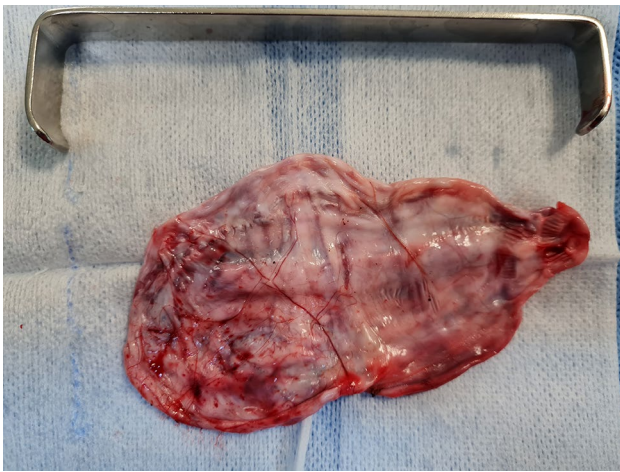


Figure 3. Intratubal serosal cyst (the retractor is 12-cm long).

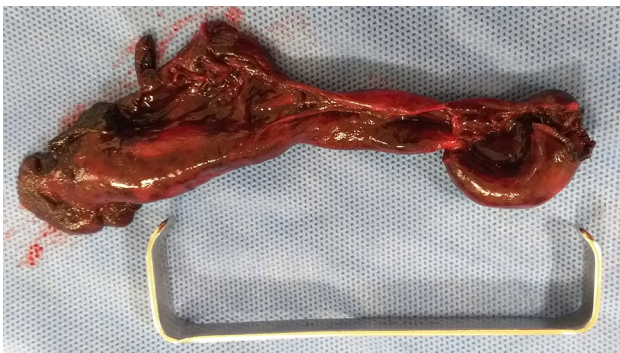


Figure 4. Necrotic tube after removing the cyst (the retractor is 12-cm long).

ratio of cyst location was 7/6. The cysts had a mean diameter of 60.5 ± 15.2 mm. During surgery, all patients had torsions. The mean torsion degree of the patients was at $570^\circ \pm 129^\circ$ due to paratubal cysts. All patients except for one were treated with laparoscopic tubal detorsion and excision of the paratubal cysts. The other patient applied on Day 3 following the onset of symptoms and had right tubal necrosis including a paratubal cyst. She underwent a laparoscopic salpingectomy while preserving the ovary (Figures 1-4). In the early DUS scan during follow-up (within the first six months after discharge), all patients had healthy ovaries with normal blood circulation. The radiologists considered the ovaries in normal sizes in relation to the patients age, without any masses while having normal blood circulation healthy. Pathological examinations were reported as

simple serous cysts in all patients. The patient treated with salpingectomy was also reported with a tubal necrosis. Table 1 shows the summary of our cases.

DISCUSSION

Although still rare, reports of pediatric IFTT are increasing in the literature. In a 20-year retrospective study, 45 cases of IFTT were reported.^[9] Currently, IFTT is usually reported as single case-reports.^[3-6,8]

In the literature, IFTT is reported to be more common in the right side.^[10,11] In our study, there were no significant differences between the frequency of sides. Several studies have shown that IFTT can be seen in premenarchal girls.^[5-8,12,13] In our study, there was only one premenarchal patient.

Except having only three patients with leukocytosis, there were no laboratory findings specific to IFTT in accordance to some studies.^[9,11,14]

For radiological diagnoses, our first preference is color DUS. Regarding the 20-year review study mentioned above,^[9] DUS, CT, and MRI showed a sensitivity of 22% (8/36), 14% (1/7), and 40% (2/5), respectively. In our study, we found that DUS had a sensitivity of 7% for the diagnosis of IFTT, 16% for pure adnexal cysts, and 84% for ovarian diseases. Four of our CT scans showed 100% sensitivity for adnexal masses; however, they could not rule out IFTT. These unreliable sensitivity results might be due to the radiologists' observation of any cysts, thereby reporting the most common pathological situation seen in adolescent girls.

Radiological testing is helpful, yet not reliable enough in the diagnosis of IFTT. Color DUS is useful and considered to be the first choice of diagnostic examination. However, a normal blood circulation does not always rule out torsion; therefore, it can be misleading for the surgeon. In addition, IFTT may mimic any acute abdominal situation; thus, surgical intervention can be delayed and salpingectomies may have to be performed.^[9,15] Meyer et al.^[16] indicated that salpingectomy was the most common treatment in 66.9% of cases; 33.1% of the patients were treated with detorsion of the tube (organ-sparing management) in a study with three of their IFTT patients combined with a review of 59 reports. Girls with an admission delay of 24 h or longer had a significantly higher rate of salpingectomy ($p=0.0323$).^[17] Mazouni et al.^[10] reported that waiting longer than 10 h after the onset

TABLE 1
Patients' overall characteristics

| Age (year) | Time of appliance | PE-symptom | Color Doppler USG | CT | Preoperative diagnosis | Side of IFTT | Perioperative additional or causing diagnosis | Procedure |
|------------|-------------------|---|---|--------------------|------------------------------|--------------|---|--|
| 15 | 72h | Localized, abdominal tenderness, nausea | 60 mm right ovarian cyst | - | Acute ovarian mass | Right IFTT | Paratubal cyst+tubal necrosis | Laparoscopic detorsion+ cyst excision+ salpingectomy |
| 11 | <24h | Localized, abdominal tenderness, nausea | 50 mm left ovarian cyst + torsion | - | Acute ovarian mass + torsion | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 14 | <24h | Localized, abdominal tenderness, nausea | 56 mm right ovarian cyst | - | Acute ovarian mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 12 | <24h | Localized, abdominal tenderness, nausea | 50 mm right ovarian cyst | - | Acute ovarian mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 15 | <24h | Localized, abdominal tenderness, nausea | 60 mm left ovarian cyst | - | Acute ovarian mass | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 15 | 24h | Localized, abdominal tenderness, nausea | 60 mm right ovarian cyst | Right ovarian cyst | Acute ovarian mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 13 | <24h | Localized, abdominal tenderness, nausea | 45 mm right ovarian cyst | - | Acute ovarian mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 16 | <24h | Localized, abdominal tenderness, nausea | 50 mm paratubal right cyst | - | Acute paratubal mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 14 | <24h | Localized, abdominal tenderness, nausea | 50 mm paratubal left cyst | - | Acute paratubal mass | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 15 | <24h | Localized, abdominal tenderness, nausea | 70 mm right ovarian cyst | Right ovarian cyst | Acute ovarian mass | Right IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 13 | <24h | Localized, abdominal tenderness, nausea | 75 mm left ovarian cyst | Left ovarian cyst | Acute ovarian mass | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 16 | 24h | Localized, abdominal tenderness, nausea | 100 mm left ovarian cyst + torsion | Left ovarian cyst | Acute ovarian mass+torsion | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |
| 13 | <24h | Localized, abdominal tenderness | 100 mm paratubal left cyst with torsion | Left ovarian cyst | IFTT | Left IFTT | Paratubal cyst | Laparoscopic detorsion+ cyst excision |

PE: Physical examination; USG: Ultrasonography; CT: Computed tomography; IFTT: Isolated fallopian tubal torsion.

of symptoms could lead to tubal necrosis. Qian et al.^[8] also found that patients with pain for more than 24 h were more likely to undergo salpingectomy, suggesting that longer periods of torsion might lead to more necrosis of the tissue. Gaied et al.^[9] reported that 88% of cases were treated by salpingectomy, and 12% were treated by tubal detorsion. In our study, despite a 24-h delay, the tube could be salvaged. However, one case who had 72 h of symptoms before surgery underwent salpingectomy. There are also some studies indicating that there can be healthy cilia cells left in a necrotic fallopian tube.^[14] Bertozzi et al.^[11] reported that conservative management for IFTT could also be considered in cases of necrotic tubes, since morbidity would not increase, but it could leave a non-functional tube.

The main strength of this study is its relatively large sample size (n=13) compared to the literature. Another strength is that all the patients were treated by a single surgeon.

In conclusion, despite being useful, radiological examination may not be precise in the diagnosis of IFTT. Being vigilant and observant against IFTT even in premenarchal patients would help many surgeons diagnose and perform a prompt exploration avoiding organ loss, thereby preserving fertility. Long-term follow-up should be done to assess the fertility rating of these patients.

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REFERENCES

- Crosier CJ, Ricca RL. Isolated fallopian torsion: A pediatric case series. *Am Surg* 2023;89:3917-9. doi: 10.1177/00031348231175118.
- Cobellis G, Torino G, Noviello C, Cruccetti A, Mastroianni L, Amici G, et al. Versatility of one-trocar surgery in children. *J Laparoendosc Adv Surg Tech A* 2011;21:549-54. doi: 10.1089/lap.2010.0063.
- Paganelli S, Seidenari A, Soncini E, Bruno R, Galassi MC. Isolated fallopian tube torsion associated with paratubal cyst in a young woman. *J Minim Invasive Gynecol* 2022;29:336-7. doi: 10.1016/j.jmig.2021.11.012.
- Noviello C, Romano M, Papparella A, Ciavattini A, Martino A, Cobellis G. The isolated tubal torsion: An insidious pediatric and adolescent pelvic urgency. *Pediatr Med Chir* 2018;40. doi: 10.4081/pmc.2018.206.
- Demirel BD, Hancioglu S, Bicakci U, Ariturk E, Bernay F. Isolated tubal torsion: A rare cause of acute abdomen in childhood. *Pediatr Rep* 2018;10:7604. doi: 10.4081/pr.2018.7604.
- Goktolga U, Ceyhan T, Ozturk H, Gungor S, Zeybek N, Keskin U, et al. Isolated torsion of fallopian tube in a premenarcheal 12-year-old girl. *J Obstet Gynaecol Res* 2007;33:215-7. doi: 10.1111/j.1447-0756.2007.00502.x.
- Schuh AM, Klein EJ, Allred RJ, Christensen A, Brown JC. Pediatric adnexal torsion: Not just a postmenarchal problem. *J Emerg Med* 2017;52:169-75. doi: 10.1016/j.jemermed.2016.08.020.
- Qian L, Wang X, Li D, Li S, Ding J. Isolated fallopian tube torsion with paraovarian cysts: A case report and literature review. *BMC Womens Health* 2021;21:345. doi: 10.1186/s12905-021-01483-2.
- Gaied F, Emil S, Lo A, Baird R, Laberge JM. Laparoscopic treatment of isolated salpingeal torsion in children: Case series and a 20-year review of the literature. *J Laparoendosc Adv Surg Tech A* 2012;22:941-7. doi: 10.1089/lap.2011.0530.
- Mazouni C, Bretelle F, Ménard JP, Blanc B, Gamberre M. Diagnosis of adnexal torsion and predictive factors of adnexal necrosis. *Gynecol Obstet Fertil* 2005;33:102-6. doi: 10.1016/j.gyobfe.2005.02.014.
- Bertozzi M, Magrini E, Riccioni S, Giovenali P, Appignani A. Isolated fallopian tube torsion with hydrosalpinx: Review of a debated management in a pediatric population. *J Pediatr Surg* 2017;52:1553-60. doi: 10.1016/j.jpedsurg.2017.07.005.
- Webster KW, Scott SM, Huguélet PS. Clinical predictors of isolated tubal torsion: A case series. *J Pediatr Adolesc Gynecol* 2017;30:578-81. doi: 10.1016/j.jpag.2017.05.006.
- Erikci VS, Hoşgör M. Isolated salpingeal torsion in children: A case series and review of the literature. *Ulus Travma Acil Cerrahi Derg* 2014;20:75-8. doi: 10.5505/tjtes.2014.26918.
- Boukaidi SA, Delotte J, Steyaert H, Valla JS, Sattonet C, Bouaziz J, et al. Thirteen cases of isolated tubal torsions associated with hydrosalpinx in children and adolescents, proposal for conservative management: Retrospective review and literature survey. *J Pediatr Surg* 2011;46:1425-31. doi: 10.1016/j.jpedsurg.2011.01.033.
- Verpalen IM, de Boer J, van den Hoed RD, Schutte JM, Teklenburg G, Vossenkaul R. Isolated fallopian tubal torsion; rare or overlooked?. *Ned Tijdschr Geneesk* 2018;162:D2670. Dutch.
- Meyer J, Steinhoff-Kellner A, Stehr M, Schäfer FM. IFTT in children and adolescents-single-center experience and systematic review of literature. *Pediatr Surg Int* 2024;40:143. doi: 10.1007/s00383-024-05719-8.
- Baracy MG Jr, Hu J, Ouillette H, Aslam MF. Diagnostic dilemma of isolated fallopian tube torsion. *BMJ Case Rep* 2021;14:e242682. doi: 10.1136/bcr-2021-242682.