

Isolated Fallopian Tube Torsion in Children: Is Salpingectomy Necessary?

A Şencan, G Özdemir, V Erikçi, M Hoşgör

ABSTRACT

Objective: Isolated tubal torsion (ITT) is a rare entity in which fallopian tube torses without ovarian torsion. The presenting symptoms are nonspecific. In this study, we aimed to report 6 pediatric cases of ITT treated with different surgical techniques in our instution and to suggest tubal conservation.

Methods: Six cases of ITT treated between 2000-2016 were retrospectively reviewed. Exclusion criteria were ipsilateral ovarian torsion together with tubal torsion, history of pelvic surgery, pelvic inflammatory disease, tubal/ovarian malignancies or pregnancy.

Results: The meanage of the patients was 10.16 years. Abdominal pain was the main complaint. Pelvic doppler ultrasonography was the first imaging modality. Salpingectomy was performed for 3 patients; one newborn with in-utero torsion with autoamputation, one patient with hydrosalpinx, one patient with torsion in the form of a necrotic mass. Detorsion was performed for 3 patients; one patient with ipsilateral ovarian cyst, one patient with paraovarian cyst and one patient with edematous ovary with follicule cysts. No recurrence was observed during follow-up.

Conclusion: ITT is a rare but an important surgical emergency that should be considered in the differential diagnose of acute abdominal pain in female children. Despite several therapeutic surgical options, main aim should be tubal conservation for future fertility.

Keywords: Children, isolated tubal torsion, salpingectomy, tubal conservation

From: Dr Behçet Uz Children's Hospital, Department of Pediatric Surgery, İzmir, Turkey.

Correspondence: Professor A Şencan, Dr Behçet Uz Children's Hospital, İsmet Kaptan Mh. Sezer Doğan Sok, No:11, Konak/İzmir, Turkey. Fax: +90 232 4892315, e-mail: arzusencan71@yahoo.com.tr

INTRODUCTION

Isolated tubal torsion (ITT) is a rare entity with an incidence of 1 in 1500000 females in which fallopian tube torsions without ovarian torsion (1,2). The presenting symptoms and physical examination findings are nonspecific and show similarities with the other causes of abdominal pain. Radiologic diagnosis of ITT is also limited. Prompt surgical intervention is mandatory to preserve the fertility capacity and to avoid serious complications. There are several surgical techniques for the treatment of tubal torsion. In this study, we aimed to report 6 pediatric cases of ITT treated with different surgical techniques in our institution and to suggest tubal conservation for the preservation of future fertility.

SUBJECTS AND METHODS

Three cases of isolated tubal torsion (ITT) treated in our institution between January 2000 and December 2012 have been previously reported (3). As it is a very rare pathology and the surgical therapeutic options vary, we aimed to expand our case series of ITT by combining the data of the original 3 cases with the additional 3 patients whom were treated between December 2012 and May 2016. Exclusion criteria were ipsilateral ovarian torsion together with fallopian tube torsion, medical history of pelvic surgery or pelvic inflammatory disease (PID), tubal or ovarian malignancies or pregnancy.

RESULTS

6 cases of ITT underwent surgical intervention. The mean age of the patients was 10.16 years (5 days-14 years). Abdominal pain was the main complaint in all of the patients (n:6/6, 100%). Vomiting associated pain in 3 patients (n:3/6, 50%) and palpable mobile abdominal mass in 1

new-born patient (n:1/6, 16.6%) . This 5-day-old baby had antenatal diagnosis. The mean duration of symptoms was 4.2 days (1-7 days). Two patients had the history of intermittent abdominal pain for a week. 3 patients were at menarche whereas the other 3 were at premenarchal period. All of the patients were afebrile and most of them had normal leukocyte levels and C-reactive protein (n:4/6, 66.6%). Except a newborn patient, tumor marker levels of ferritin, lactate dehydrogenase (LDH) and alpha-fetoprotein (AFP) were in normal range. Urinary tract infection was present in only one patient. Pelvic doppler ultrasonography (USG) was the first imaging modality in all of the patients.

Due to diagnostic difficulties, magnetic resonance imaging (MRI) (Picture 1) followed USG in 3 patients and computed tomographic scan (CT) in 2 patients. Sonographic findings were helpful to diagnose tubal torsion in only one patient preoperatively describing a whirlpool sign specific for ITT (16.6%). No further imaging was obtained for that patient and she underwent immediate laparotomy. Laparoscopy was performed in only one patient and it was converted to open surgery for easier removal of the necrotic tissue. At laparotomy, tubal torsion was observed on the right in 4 cases (66.6%). Torsion was 720 degrees counterclockwise in three patients and 360 degrees clockwise in two patients. Salpingectomy was performed in 3 cases (50%) and detorsion without salpingectomy in the other 3 patients (50%). In-utero tubal torsion and autoamputation was identified in the newborn patient. The right tube was 0.5cm in length with obliterated tip. Autoamputated tube 6x5 cm in diameter which was connected to the tip of inflamed appendix was removed together with the appendix.

Hematosalpinx associated tubal torsion in the second patient for whom salpingectomy was performed. In the third patient, the torsed left fallopian tube was in the form of a necrotic blue mass 10 cm in diameter. Histopathological examination of the resected fallopian tubes revealed hemorrhagic infarction and necrosis. In the first patient for whom right tubal detorsion without

resection was performed, the right ovary was 5x6 cm in diameter and it was edematous with milimetric follicule cyts (Picture 2). In the second patient, there was ipsilateral right ovarian cyst 9x10 cm in diameter. The cyst was excised preserving the right normal appearing ovary. In the third patient, there was a 10x10 cm left paraovarian cyst incorporated in the broad ligament close to the left fallopian tube and it was completely filling the pelvis (Picture 3). After aspirating clear fluid from the cyst, the left torsioned tuba was detorsioned and the cyst was totally excised. The left ovary was normal.

The postoperative period was uneventful in all of the patients. During a period of 7 years follow-up in one patient and the 14-year follow-up of the other two patients for whom salpingectomy was performed, no recurrence has been observed. In the postoperative US assessments of the three patients for whom detorsion was performed, no signs of tubal or ovarian pathologies were identified during the six-month follow-up period .It was not possible to evaluate the postoperative fertility of these patients due to their rather young age. Patient characteristics are summerized in the Table.

DISCUSSION

Isolated fallopian tube torsion refers to the complete or partial twist of the tube along its axis without ovarian torsion. It is a very rare but important cause of lower abdominal pain in premenstrual girls and postmenarchal teenagers younger than 18 years. Delay in surgical intervention results in ischemic tubal injury which adversely effects future fertility of the patient similar to infectious or adhesive diseases.

There may be intrinsic and extrinsic causes that predispose to ITT (4). Intrinsic causes include congenital or acquired pathologies of the fallopian tube such as long or spiral tube,

hydrosalpinx, paratubal or paraovarian cysts, neoplasms, tubal ligation, ectopic pregnancy, pelvic inflammatory disease, abnormal peristalsis or spasm of the tube. Extrinsic causes include local and mechanical factors such as adhesions and acceleration/deceleration of the body that is known as the Selheim theory (5).

A correlation between ITT and practicing sports involving sudden changes in the body position has been reported in children and adolescent females (6). Elevated follicle-stimulating hormone (FSH) level during early puberty may be the cause of abnormal tubal motility in the absence of tubal pathologies (7,8). In this study, hydrosalpinx was observed in one menarchal patient. Whether hydrosalpinx is the cause or the consequence of the torsion is an issue that should be considered in the pathogenesis (9). Ipsilateral ovarian cyst 9x10 cm in diameter was present in one premenarchal patient and ipsilateral paraovarian cyst 10x10 cm in diameter in another menarchal patient. No normal-looking fallopian tube was observed in the presented neonatal patient and it was speculated that it had presumably torsed in utero, resulting in autoamputation. To our knowledge, it was the only ITT of neonatal age reported in the English literature (3).

Torsion of the right fallopian tube is more common (10,11). This may be due to the presence of sigmoid colon presenting a preventing effect on the left, compared with the relative mobility of the cecum on the right. Another reason may be the fact that the right utero-ovarian ligament is physiologically longer than the left. In this study, tubal torsion was observed on the right in the majority of the cases (66.6%).

The clinical findings and presenting symptoms of ITT are nonspecific and are similar to those of ovarian torsion which include sudden onset of abdominal pain, nausea, vomiting and rarely, fever (12,13). Pain with duration of more than 10 hours before surgery was reported to be associated with an increased rate of tubal necrosis (14). Except from the antenatal diagnosed neonatal case, two patients for whom salpingectomy was performed admitted with the history of

pain for 3 days. On the other hand, however, two of the three patients for whom detorsion without salpingectomy was performed had the history of abdominal pain of seven-days' duration. In this study, the duration of pain was not a predictive factor for tubal necrosis. Laboratory findings are also nonspecific (15). Absence of leucocytosis and normal C reactive protein (CRP) values, as observed in most of the presented cases in this study, may help to distinguish tubal torsion from appendicitis.

Abdominopelvic doppler ultrasonography (USG) is the first imaging modality, but diagnostic property of radiology is limited in ITT. In a case series, the most consistent finding on USG was reported as a midline cystic mass with a normal ipsilateral ovary (16). Because of the dual vascular supply of the tubes, a normal doppler USG can not exclude the diagnosis of tubal torsion (17). A whirlpool sign has been described as a specific sonographic finding of ITT (18). USG revealed a whirlpool sign in the right fallopian tube in only one of the presented cases and she immediately underwent laparotomy with no further radiologic imaging. Computed tomographic scan (CT) and magnetic resonance imaging (MRI) can reveal the adnexial mass but no signs of ischemia. MRI associated USG in 3 of the patients and CT in the other two patients in this study, but revealed no pathognomonic signs for ITT. In one study, the sensitivity of US, CT and MRI in diagnosing ITT was reported as 22%, 14% and 40% respectively (19). The exact diagnose is made during exploratory laparotomy.

There are several surgical therapeutic options for the treatment of ITT. Salpingectomy was reported as the most common surgical technique (20,21,22,19). Although salpingectomy does not change subsequent patient fertility when the contralateral tube seems to be healthy (23), there is always a risk of infection or torsion for the remaining tube. Therefore, salpingectomy carries a risk of adverse effect on reproductive function of the patient when compared with tubal

conservation. Partial resection of the affected distal part of the tube is another option (7). However, distal part of the fallopian tube containing ampulla and fimbria is necessary for ovum captation (24). The remaining nonfunctional proximal part of the tube carries the risk of hydrosalpinx formation and a possible site for ectopic pregnancy. Total resection, instead of partial resection is therefore recommended, when necessary (7). In this study, salpingectomy was performed in 3 patients (50%) for totally necrotic ITT.

The future fertility of the pediatric patients is an important issue that should be taken into account when treating ITT. As ovarian salvage is opposed in ovarian torsion regardless of how necrotic the ovary appears (25,26), the same conservative approach can be applied in ITT. Therefore, detorsion without resection is another option. Recurrence after detorsion due to distal tubal occlusion which required salpingectomy has been reported (7). In the presented study, 50% of the patients were treated with detorsion and no recurrence has been observed during 6-month period of follow-up. Salpingoneostomy has been reported in premenarchal girls after detorsion of ITT as a second-look surgical procedure (7,27). After performing a salpingoscopic evaluation of the tube, an incision is made to create a new opening in the end of the occluded fallopian tube with a microsurgical needle electrode.

The tissue is folded over and stitched into place. The new ostium replaces the normal opening of the fallopian tube. Laparoscopic salpingoneostomy has been reported with good results for tubal infertility and hydrosalpinx treatment in adults (28). For better results, salpingoneostomy is suggested to be performed a few weeks after the inflammatory processes of the fallopian tube in adults (29). Salpingoneostomy has been reported in children after ITT with recurrence free follow-up (7).

The long-term results of tube-preserving and restorative surgery and its effects on future fertility are not exactly known. Spontaneous conception and successful in vitro fertilization-assisted

pregnancies have been reported after conservative tubal surgery in adults (30). On contrary, salpingoneostomy for hydrosalpinx treatment in adult infertile patients was not recommended due to the high risk of dismal spontaneous pregnancy and recurrence of hydrosalpinx (31).

Another controversial issue in the surgical treatment of ITT is the role of salpingopexy. It may prevent the recurrence of tubal torsion, but also may change the normal anatomic relation between the ovary and the distal part of the tube. Salpingopexy also shortens the mesosalpinx leading to impairment of blood supply to the adjacent ovary (32).

CONCLUSION

Isolated tubal torsion is a rare but an important surgical emergency that should be considered in the differential diagnosis of acute abdominal pain in female children. In the management of these patients, it should be aimed to preserve the future fertility. It is not possible to diagnose isolated tubal torsion based on the clinical and imaging findings. The exact diagnosis is possible only during exploratory surgery. There are several therapeutic surgical options for the treatment of ITT with some advantages and limitations of their own. Surgical techniques should be considered on a patient-by-patient basis, but conservative management should be the initial treatment modality. The results of most of these techniques are based on the surgical experience in adults. Therefore, more prospective studies showing the long-term results of salpingectomy or tubal conservation and their effects on future reproductive capacity of pediatric patients are needed.

AUTHORS' NOTE

A Şencan conceived paper, oversaw data collection, conducted data analysis, wrote manuscript and approved final version. G Özdemir participated in the data collection. V Erikçi and M Hoşgör participated in the study design, data analysis. The authors declare that they have no conflict of interest.

REFERENCES

1. Hansen OH. Isolated torsion of the Fallopian tube. *Acta Obstet Gynecol Scand* 1970; 49:3-6.
2. Provost MW. Torsion of the normal fallopian tube. *Obstet Gynecol* 1972; 39(1):80-2.
3. 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.
4. Youssef AF, Fayad MM, Shafeek MA. Torsion of the fallopian tube. A clinico-pathological study. *Acta Obstet Gynecol Scand* 1962; 41:292-309.
5. Blair CR. Torsion of the fallopian tube. *Surg Gynecol Obstet* 1962; 114:727-30.
6. Romano M, Di Giuseppe J, Serri M, Noviello C, Martino A, Cobellis G et al. A possible association between sports and isolated fallopian tube torsion in children and adolescent females. *Gynecol Endocrinol* 2015;31:688-92.
7. 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.
8. Merlini L, Anooshiravani M, Vunda A, Borzani I, Napolitano M, Hanquinet S. Noninflammatory fallopian tube pathology in children. *Pediatr Radiol* 2008; 38:1330-37.
9. Višnjić S, Kralj R, Zupančić B. Isolated fallopian tube torsion with partial hydrosalpinx in a premenarcheal girl: a case report. *J Med Case Rep* 2014; 8:197-201.
10. Peña JE, Ufberg D, Cooney N, Denis AL. Usefulness of Doppler sonography in the diagnosis of ovarian torsion. *Fertil Steril* 2000; 73:1047-50.

11. Warner MA, Fleischer AC, Edell SL, Thieme GA, Bundy AL, Kurtz AB et al. Uterine adnexal torsion: sonographic findings. *Radiology* 1985;154:773-75.
12. Krissi H, Shalev J, Bar-Hava I, Langer R, Herman A, Kaplan B. Fallopian tube torsion: laparoscopic evaluation and treatment of a rare gynecological entity. *J Am Board Fam Pract* 2001,14:274-77.
13. Seshadri S, Morris A, Uchil D, Joloaso A. Bilateral paratubal cysts with co-existent fallopian tube torsion in an adolescent. *J Obstet Gynaecol* 2009;29:564-65.
14. Mazouni C, Bretelle F, Ménard JP, Blanc B, Gamberre M. Diagnosis of adnexal torsion and predictive factors of adnexal necrosis. *Gynecol Obstet Fertil* 2005; 33:102-106.
15. Kisku S, Thomas RJ. An uncommon twist: isolated fallopian tube torsion in an adolescent. *Case Rep Surg* 2013; 2013:1-2.
16. Harmon JC, Binkovitz LA, Binkovitz LE. Isolated fallopian tube torsion: sonographic and CT features. *Pediatr Radiol* 2008; 38:175-79.
17. Huchon C, Fauconnier A. Adnexal torsion: a literature review. *Eur J Obstet Gynecol Reprod Biol* 2010; 150:8-12.
18. Vijayaraghavan SB, Senthil S. Isolated torsion of the fallopian tube: the sonographic whirlpool sign. *J Ultrasound Med* 2009; 28:657-62.
19. 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; 22:941-47.
20. Orazi C, Inserra A, Lucchetti MC, Schingo PM. Isolated tubal torsion: a rare cause of pelvic pain at menarche. Sonographic and MR findings. *Pediatr Radiol* 2006; 36:1316-18.

21. Terada Y, Murakami T, Nakamura S, Sato Y, Niikura H, Ito K et al. Isolated torsion of the distal part of the fallopian tube in a premenarcheal 12 year old girl: a case report. *Tohoku J Exp Med* 2004; 202:239-43.
22. Dadhwal V, Gupta N, Gupta B, Deka D, Mittal S. Laparoscopic management of isolated fallopian tube torsion in a premenarchal 13-year-old adolescent girl. *Arch Gynecol Obstet* 2009; 279:909-910.
23. Kennard EA, Karnitis JV, Friedman CI. Juxtaposition of contralateral ovary and fallopian tube to allow pregnancy in unicornuate uterine anomaly. *Am J Obstet Gynecol* 1994; 171:1387-89.
24. Perlman S, Hertweck P, Fallat ME. Paratubal and tubal abnormalities. *Semin Pediatr Surg* 2005; 14:124-134.
25. Oltmann SC, Fischer A, Barber R, Huang R, Hicks B, Garcia N. Cannot exclude torsion-- a 15-year review. *J Pediatr Surg* 2009; 44:1212-16.
26. Meynol F, Steyaert H, Valla JS. Adnexal torsion in children: plea for early laparoscopic diagnosis and treatment. *Arch Pediatr* 1997; 4:416-19.
27. Sozen I, Nobel PA, Nobel J. Partial tubal salvage through neosalpingostomy in a 12-year-old girl with combined ovarian and fallopian tube torsion. *J Pediatr Surg* 2006; 41:17-9.
28. Bontis J, Tarlatzis JV, Grimbizis G. Microsurgical and laparoscopic management of tubal fertility: report of 763 cases. *Middle East Fertil Soc* 1996; 1: 17-29.
29. Audebert AJ, Pouly JL, Von Theobald P . Laparoscopic fimbrioplasty: an evaluation of 35 cases. *Hum Reprod* 1998; 13:1496-1499

30. Chanelles O, Ducarme G, Sifer C, Hugues JN, Touboul C, Poncelet C. Hydrosalpinx and infertility: what about conservative surgical management? *Eur J Obstet Gynecol Reprod Biol* 2011; 159:122-26.
31. Bayrak A, Harp D, Saadat P, Mor E, Paulson RJ. Recurrence of hydrosalpinges after cuff neosalpingostomy in a poor prognosis population. *J Assist Reprod Genet* 2006; 23:285-88.
32. Lima M, Libri M, Aquino A, Gobbi D. Bilateral hydrosalpinx with asynchronous tubal torsion: an exceptional finding in a premenarcheal girl. *J Pediatr Surg* 2011; 46:27-9.

Isolated Fallopian Tube Torsion in Children

Table: Patient characteristics

	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient 6
Age	14years, menarchal	5 days, premenarchal	13years, menarchal	9years, premenarchal	12years, premenarchal	13years, menarchal
Side	Right	Right	Left	Right	Right	Left
Degree of torsion	360 clockwise	In-utero torsion	720 counterclockwise	720 counterclockwise	720 counterclockwise	360 Clockwise
Complaints	Pain, vomiting	Abdominal mass	Pain	Pain, vomiting	Pain, vomiting	Pain
Duration of symptoms	3 days	Antenatal diagnose	3 days	1 day	7 days	7 days
Laboratory findings	Normal	Ferritin ↑ LDH ↑ AFP ↑	Normal	Normal UTI (+)	WBC ↑ CRP ↑	WBC ↑ CRP ↑
Imaging modality	USG+CT	USG+CT	USG+CT	USG	USG+MRI	USG+ MRI
Associated pathology	Hydrosalpinx	Autoamputated tube+inflamed appendix	-	Edematous right ovary with follicule cysts	Right ovarian cyst(10x9cm)	Left paraovarian cyst(10X10cm)
Surgical treatment	Salpingectomy	Salpingectomy	Salpingectomy	Detorsion	Detorsion+ cystectomy	Detorsion+ cystectomy
Follow-up	No recurrence	No recurrence	No recurrence	No recurrence	No recurrence	No recurrence

LDH: lactate dehydrogenase, AFP: alpha feto-protein
WBC: white blood cell, CRP: C reactive protein

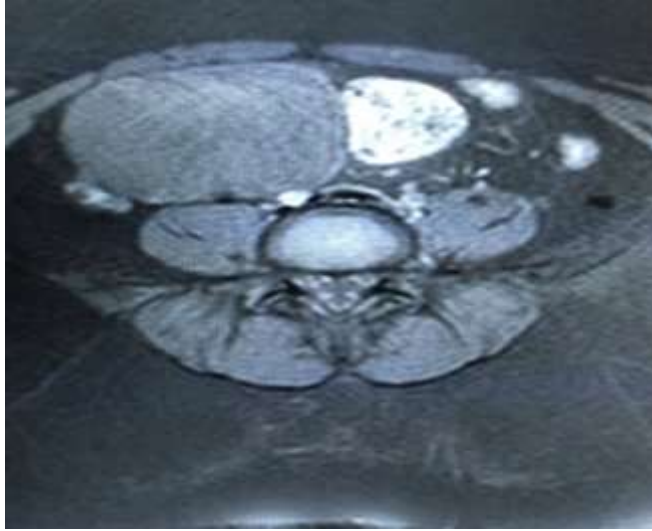


Fig 1. : Magnetic resonans imaging of left paraovarian cyst 10x10 cm in diameter which was reported as right ovarian cystic mass (Patient 6).



Fig. 2: Intraoperative image of ipsilateral edematous right ovary with follicule cysts (patient 4).