# A rare case of isolated fallopian tubal torsion in adolescent girls: A case report and system review of literature

SAGE Open Medical Case Reports
Volume 11: 1–12
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DOI: 10.1177/2050313X231215207
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### **Abstract**

Isolated torsion of a fallopian tube in adolescent girls is a very rare acute abdomen. How to early diagnose and treatment isolated torsion of a fallopian tube has always been one of key points. We reported a rare case of isolated torsion of a fallopian tube in a 14-year-old girl and conducted a systematic review of the literature over the past 21 years. We collected data on clinical characteristics, diagnosis and treatment, prognosis and follow-up outcomes. According to our research strategies, 43 reported cases were included. The results (Mean  $\pm$  SD) showed that the onset age was 14.09  $\pm$  2.49, the cycle torsion was 2.79  $\pm$  1.18, and the cyst size was 6.43  $\pm$  2.25. The main clinical symptoms (percentage) are 100% lower abdominal pain, 67.44% nausea, 51.16% vomiting, and 50.00% right side lesions. And the accurate rate of preoperative diagnosis was only 27.91%, in which laparoscopic exploration, tubal necrosis, and tubal preservation accounted for 79.07%, 41.86%, and 32.56%, respectively. Extremely difficult diagnosis of isolated torsion of a fallopian tube, because of non-specific clinical manifestations, led to the adverse clinical outcome of salpingectomy. However, the accuracy of preoperative diagnosis can be improved by improving acknowledge of ultrasound diagnosis and the awareness of clinical gynecologists. However, the accuracy of preoperative diagnosis can be improved by improving knowledge of ultrasound diagnosis and awareness of clinical gynecologists. The laparoscopic exploration is sole gold standard for diagnosis and treatment. The operation of preserving the fallopian tube may be the first choice, especially in the early stage of isolated torsion of a fallopian tube.

# **Keywords**

isolated tubal torsion, acute abdominal pain, salpingectomy, ultrasound, pubertal girls

Date received: 12 September 2023; accepted: 19 October 2023

## Introduction

Isolated fallopian tube torsion (IFTT) was first described in 1890 by Bland-Sutton. And IFTT is also an uncommon gynecologic acute abdomen.1 And it is often misdiagnosed due to the range of conditions which can mimic this diagnosis and lack of pathognomonic imaging method, specific symptom or characteristic laboratory findings.<sup>1,2</sup> The diagnosis is usually made during exploration surgery, delayed treatment exposes to risk of necrosis, irreversible tube damage, and infection.<sup>2-4</sup> To raise the awareness of this diagnosis among clinical gynecologists, we systematically describe the clinical manifestations, objective manifestations and surgical results of an IFTT secondary to a cyst at the fimbriate end of the fallopian tube that occurred in an adolescent girl, and systematically review the English literature reports of IFTT in pubertal girls from 2002 to 2023.

# Case presentation

A 13-year-old adolescent girl presented to our Emergency room with intermittent lower abdominal pain for 2 days. Her

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Figure 1. Color B-mode ultrasound shows that anechoic mass with a size of about 10.  $9 \, \text{cm} \times 9.0 \, \text{cm} \times 9.0 \, \text{cm}$  can be seen in the pelvic cavity, with clear boundary and sound transmission (left). Pelvic computed tomography can see a cystic homogeneous low-density lesion with a size of  $9.90 \, \text{cm} \times 9.25 \, \text{cm}$  and clear boundary (right).

medical history was no significant, and there was no previous significant medical or surgical history. Menarche age had occurred at 12 years of age. The last menstrual period is July 22, 2022. The primarily clinical manifestation were mild lower abdominal pain, mild nausea, no vomiting, and no fever. Therefore, pediatricians consider acute early gastroenteritis and provide corresponding spasmolysis targeted treatment. And her symptoms were obviously relieved. One day later, the abdominal pain became worse, and the nausea and vomiting became obvious again. So this patient revisited the gynecological clinic. On physical examination, tenderness was observed in her lower right quadrant abdomen and rebound pain was marked. However, her vital signs were normal. Pelvic and anal examination revealed an enlarged right adnexal tumor. Pelvic ultrasonography (USG) showed an enlarged mass with a size of  $109 \times 90 \times 90 \,\mathrm{mm}^3$  on the right side of uterus with clear boundary and uniformly low echogenicity. Pelvic computed tomography (CT) can see a cystic homogeneous low-density lesion with a size 9.90 cm × 9.25 cm and clear boundary (Figure 1). Laboratory results showed a white cell count of  $6.95 \times 10^3$  cells/mm<sup>3</sup> with 75.20% neutrophils, a hemoglobin concentration of 133.0 g/ dL. Level of C-reactive protein was significantly increased to 21 mg/dL. Levels of tumor markers including CA125, CA199, and CEA (carcinoembryonic antigen) were within normal limits. Results of other laboratory tests, such as liver function tests, kidney function tests, serum electrolyte levels, and her coagulation profile, were all within normal limits.

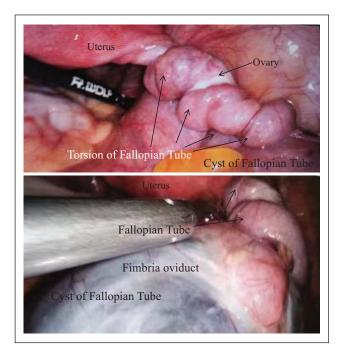
An emergency laparoscopic exploration was performed by the gynecologist. Right tubal isolated torsion was found possibly due to a giant cyst of fimbrial end of fallopian tube. The fallopian tube twisted from the cornu uteri to the fimbria for five circles, untwisted ovary was wrapped in twisted fallopian tube. However, the appearance of the fallopian tube is normal, and the color of the cyst is slightly black because of congestion mild accumulation of blood. No sign of complete necrosis of the tube and the normal healthy ipsilateral ovary was noted. The fallopian tube was completely preserved, and the paraovarian fimbrial cyst was removed (Figure 2). The patient recovered rapidly and was discharged home 3 days later. Postoperative pathology confirmed right fallopian tube paramesonephric duct cyst (Figure 3).

### **Review of literature**

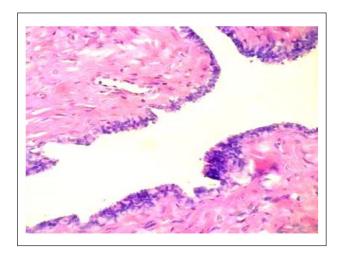
The study protocol was approved by the Ethics Committee of the Longquan People's Hospital affiliated to Lishui University and conformed to the ethical guidelines of the Helsinki Declaration (Approval Number 2023-04-001-EC-LQSRMYY). The patient and her mother signed informed consent before entering the study, and their anonymity was preserved.

A review of the literature was performed following the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) statement. In the last 21 years, we searched all the articles related to our topic from the international electronic bibliographic databases PubMed (from 2002 to 2023). The articles were found using comprehensive search criteria and a combination of MeSH (medical subject headings) terms. We used the following words for selection: "tube torsion," "diagnosis," "isolated fallopian tubal torsion," and "adolescent" (adolescent girls, teenagers, or pubertal girls). We selected the articles published between January 2002 and July 2023. The search was limited to studies reported in the English language. The references of the items chosen were also evaluated for related citations. Two independent researchers assessed the titles and abstracts retrieved to select the most relevant articles. The full text was obtained if the title and abstract did not provide enough information. Letters to editors, editorials, review articles,

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**Figure 2.** The isolated fallopian tube twists five cycles from the cornu uteri and untwisted ovary was wrapped in twisted fallopian tube (above). Giant cyst of fallopian tube can be seen at the distal end with a diameter of 10.0 cm. The cyst surface can be seen with fimbria fallopian tube (below).



**Figure 3.** The remnant of the accessory mesonephric duct beside the fallopian tube and the covered epithelium are typical fallopian tube-like manifestations, and the cyst was histologically recognized as a paramesonephric cyst.

duplicates, and meta-analysis were excluded. We only included items that followed our eligibility criteria represented by adolescent girls with a diagnosis of IFTT. We excluded those studies evaluating data from women with ovarian torsion, adnexal torsion, patients not in adolescence, and postmenopausal patients. We also included articles concerning single case reports of tubal torsion where both the

preoperative diagnosis and treatment were discussed. We collected data regarding general characteristics, clinical features, treatment, routine pathological results, and preoperative diagnosis.

## Results

We found 43 cases of IFTT in pubertal girls described in the English language literature in the past 21 years. <sup>5-37</sup> Recorded general characteristics including age, duration of abdominal pain (DAP), clinical manifestation, blood test, and image examination are shown in Table 1. And intraoperative and postoperative conditions of IFTT including preoperative diagnosis, size of the cyst, exploration mode of emergency operation, procedure, and routine pathology are also shown in Table 2.

From a systematic review of 43 cases of IFTT reported in recent 21-year English language literature, we found that the clinical data (Means ± SD) of adolescent IFTT showed  $14.09 \pm 2.49$ ,  $2.79 \pm 1.18$ ,  $6.43 \pm 2.55$ , and  $41.38 \pm 112.48$ in terms of onset age, number of torsion circles, size in the cyst, and DAP, respectively. And incidence of clinical symptoms was separately acute lower abdominal pain 100% (43/43), nausea 67.44% (29/43), vomiting 51.16% (22/43), fever 23.26% (10/43), lesion side (right: 50.00%, left: 45.24%, bilateral: 4.76%), and localized peritoneal irritation sign 44.19% (19/43). In addition, the accuracy of preoperative diagnosis is only 27.91% (12/43) by US combined with CT. Normal rate of WBC (white blood cell) and CRP (C-reactive protein) of 43 case patients in blood tests was respectively 51.16% (22/43) and 39.53% (17/43). The rate of laparoscopic exploration in exploration mode of emergency operation was 79.07% (34/43). The proportion of fallopian tube necrosis found during operation is 41.86% (18/43), and the proportion of routine pathology was respectively paraovarian and parafallopian cyst 39.53% (17/43), Hydatid of Morgagni 18.60% (8/43), hematosalpinx/hydrosalpinx 37.21% (16/43), and the other unknown pathology 4.65% (2/43). However, all cysts were showed 100% benign cyst. In addition, tubal preservation accounted for 32.56% (14/43). As for postoperative follow-up, except for one case recurrence, the data available in the literature is very limited, ranging from 2 months to 8 years, especially for the impact on later reproductive function.

# **Discussion**

IFTT is a more uncommon condition with an incidence estimated at 1 in 1.5 million women.<sup>38</sup> Although many possible risk factors for IFTT include prior tubal ligation, hydrosalpinx, pelvic inflammatory disease, a long or congested mesosalpinx, tubal tumors, Morgagni hydatids (HM), and trauma,<sup>1,2</sup> age may be a high-risk factors. Since Krstic reported the first case of adolescent IFTT in 1951, IFTT rarely occurs in adolescent girls.<sup>4,5</sup> Through this study, it is

 Table I. Characteristics of isolated fallopian tubal torsion cases reported in adolescent girls.

Case	Publish time	Age	DAP	Clinical manifestation	ıtion			Blood test		Image examination
(ii)	Authors (Reference)	(Year)	(week/day/ hour)	Model and site of pain R/L/B	Nausea +	Vomiting +	Fever	WBC (×10 <sup>9</sup> /l)	CRP (mg/L)	B-mode ultrasound (B-U)/CT/MRI
_	Okada et al. <sup>5</sup>	4	12-days	R-side/elastic soft	+	+	ı	6.2	0.2	B-U: A homogenous cyst, the right ovary showed a mixed pattern. CT:A twisted right fallopian tube in the adnexa.
7	Same as above	4	10-days	L-side/ tenderness	Z O	Z 5	Z	9.4	0.5	CT/MRI: A homogenous cyst close to the uterus and normal ovaries.
m	Rizk et al. <sup>6</sup>	17	4-h	R-side/guarding tenderness	+	+	+	9.2	Z S	B-U: a right ovarian unilocular cyst with no solid areas or ascites the pelvic cavity.
4	Matsuura et al. <sup>7</sup>	12	Z 5	L-side/colicky	N O	N O	Z S	N O	N S	B-U: A spherical fluid-filled mass adjacent to a normal shaped ovary.
72	Breitowicz et al. <sup>8</sup>	4	5-months	B-side/guarding tenderness	+	+	1	Z	Z 5	B-U: two cystic tumors, of size $5.5 \times 7.0$ cm and $5.5 \times 9.0$ cm, respectively. The tumors were single chambered, with no evidence of solid masses, and located behind a normal uterus.
9	Same as above		2-months	L-side/colicky	+	+	1	S	S	B-U: a cystic process of 4.5 cm connected to the left adnexa.
^	Lau et al.9	4	Z	R-side/guarding tenderness	ı	ı	ı	Normal	S	B-U: right ovarian cyst without ascites.
∞	Ho et al. <sup>10</sup>	61	l-year	L-side/tender	+	1	ı	N S	Z S	B-U: a cystic mass at the left adnexa.
6	Schollmeyer et al.''	15	I8-months	Left lower quadrant pain	+	1	ı	Normal	Z 5	B-U: Normal uterus and ovaries. A round, thick-walled, cystic structure, adjacent to the left ovary and mild fluid collection in the POD. MRI: a contrast-enhanced roundish structure.
0	Dadhwal et al. <sup>12</sup>	<u>3</u>	I-month	R-sided/tender	+	ı	ı	9.8	N S	B-U: the cyst with no solid areas and normal doppler findings. a right ovarian unilocular cyst.
=	Kopec et al. <sup>13</sup> 14	~ <del>4</del>	Z 5	L-side/mild	<u>z</u>	S	Z 5	Normal	Z 5	B-U: a large tubular midline structure with low-level echoes and a dilated tube. MRI: a spiral appearance of the portion of the left fallopian tube in proximity to the left ovary.
12	Lim and Roex <sup>14</sup>	4	3-weeks	L-sided/Flank	+	+	ı	Normal	Normal	B-U: right ovarian cyst, (CT): a large lesion from the pelvic cavity.
13	Pampal et al. <sup>15</sup>	<u>13</u>	2-days	L-sided/worsen guarding, tenderness	<u>z</u>	S	Z	Normal	Z	B-U/CT: a left tubal enlargement with increased free peritoneal fluid around the internal genitalia and bladder. MRI: hydrosalpinx.
4	Kisku and Thomas <sup>16</sup>	<u> </u>	5-days	L-sided/flank	+	+	1	10.60	S	B-U/CT/MRI: all unknown.
12	Same as above	<u>8</u>	l-week	L-sided/ moderate	Z 5	S	1	Normal	S	B-U: right adnexal cyst with internal echoes.
91	Rajaram et al.³	61	I-day	L-side/ tenderness	+	+	1	Normal	Normal	B-U: a heterogenous mass in the left ovary with absent vascularity.
	Blitz and Appelbaum <sup>17</sup>	4	S	L-side/ tenderness	+	+	+	89.	Z S	CT/MRI: A homogenous cyst close to the uterus and normal B-U: normal ovaries bilaterally, a 2-cm simple adnexal cyst adjacent to the right ovary and a normal uterus.

(Continued)

Table I. (Continued)

Case	Publish time	Age	DAP	Clinical manifestation	tion			Blood test		Image examination
(n)	Authors (Reference)	(Year)	(week/day/ hour)	Model and site of pain R/L/B	Nausea  +	Vomiting +	Fever	WBC (×10 <sup>9</sup> /l)	CRP (mg/L)	B-mode ultrasound (B-U)/CT/MRI
<u>&amp;</u>	Višnjić et al. <sup>18</sup>	=	7-days	R-side/moderate tenderness	+	+	N S	11.50	Normal	B-U: a hypoechogenic cyst with blood flow on the edges of the structure and absence in the central part.
<u>6</u>	Toyoshima et al. <sup>19</sup>	<u>o</u>	I-day	R-side/ tenderness rebound	+	+	+	21.60	Normal	B-U: right adnexal cyst with ascites. CT: a dilated and tortuous tubule-like structure in her central pelvis with a weakened contrast effect. MRI: normal ovaries and a well circumscribed mass with a thickened wall without obvious contrast enhancement.
20	Ormasa et al. <sup>20</sup>	0	I-day	R-side/ tenderness	+	+	ı	11.70	A-Nor	B-U: two cystic tumors, of size $5.5 \times 7.0\mathrm{cm}$ and $5.5 \times 9.0\mathrm{cm}$ , respectively . The tumors were single chambered, with no evidence of solid masses, and located behind a normal uterus.
2	Gunal et al. <sup>21</sup>	12	3-days	R-side/ tenderness Defense	Z 5	S	I	12.30	Normal	B-U: a left adnexal hemorrhagic cyst with internal echoes and simple cystic structure on ultrasound Doppler ultrasound revealed absence of blood flow on the left adnexa.
22	Same as above.	12	3-days	R-side/guarding tenderness	S	S	1	20.50	S	B-U: no significant sign.
23	Same as above	<u>3</u>	4 <del>-</del>	B-side/ tenderness defense	Z 5	S	ı	Normal	Normal	B-U: right adnexal cyst.
24	Mueller and and Tomita <sup>22</sup>	0	Z 5	R-side/persistent	1	1	ı	Normal	Normal	B-U: Adnexal cysts and normal ovaries bilaterally, with normal blood flow.
25	Same as above	<u>3</u>	Z 5	R-side/tender	ı	1	1	Normal	Normal	B-U: Adnexal cysts and normal ovaries bilaterally, with blood flow.
26	Zaat et al. <sup>23</sup>	<u>&amp;</u>	3-days	R-side/ Continuous	+	+	Z 5	9.90	Normal	B-U: an echogenic unilocular cyst, with possible relation to the right ovary, some free fluid in the pouch of Douglas.
27	Demirel et al. <sup>24</sup>	=	2-days	R-side/gradually tenderness.	1	1	1	Normal	Normal	B-U: normal blood circulation in both ovaries, an anechoic mass unrelated to the ovaries and uterus in the pelvis. MRI: a paraovarian, cystic lesion on the posterior side of the bladder which may tubal origin, considering IFTT.
28	Kolovos et al. <sup>25</sup>	9	3-days	Right-side/ Convulsive	+	+	+	16.40	98.00	B-U: a small unilocular in the right ovary, a little ascites in the pouch.
29	Martín- Vallejo et al.² <sup>6</sup>	12	3-weeks	First, left iliac fossa After 3 weeks: severely	+	1	I	11.30	Normal 24.00	B-U: hydrosalpinx adjacent to the left ovary and no ascites (free fluids); B-U: left para-adnexal formation, and moderate amount of free fluid.
30	lbirogba et al. <sup>27</sup>	<u>3</u>	3-days	R-side/severely	+	+	1	12.70	Normal	B-U: a large cyst in the right adnexal region. CT: the presence of a thinly walled, homogenous right adnexal cyst with mild free pelvic fluid. No acute appendicitis.

(Continued)

Table I. (Continued)

Case	Publish time	Age	DAP	Clinical manifesta	ation			Blood test		Image examination
(n)	Authors (Reference)	(Year)	(week/day/ hour)	Model and site of pain R/L/B	Nausea  +	Vomiting +	Fever +	WBC (×10 <sup>9</sup> /l)	CRP (mg/L)	B-mode ultrasound (B-U)/CT/MRI
31	Daniilidis et al.²8	17	2-h	R-side/convulsive tense and sensitivity	+	+	I	11.10	1.30	B-U: a large unilocular cyst in the left ovary with reduced vascularity. CT: confirmation of ovarian torsion.
32	Chappalley et al. <sup>29</sup>	15	3-months	L-side/ tenderness	+	+	+	13.90	S	B-U: a simple 9 cm ovarian cyst, without septa or solid mass in the left upper abdomen and moderate ascites.
33	Vadukkut et al.³¹	13	3-days	L-sided/sharp	+	+	1	Normal	S	B-U: a large unilocular cyst in the left adnexa region and POD. MRI: left-sided hydrosalpinx.
34	Ramadan et al.³¹	12	3-days	L-side/guarding rebound	+	+	I	16.10	17.80	B-U: normal-sized uterus, with multilocular adnexal complex of mixed echogenic components. CT: the absence of appendicitis, with minimal fluid in the pelvis and enlarged ovary with few cysts, ovarian torsion.
35	Same as above	4	2-days	R-side/ McBurney's sign	+	+	+	14.80	21.00	B-U: multicystic collection possibly.
36	Syed et al. <sup>32</sup>	6	24-h	L-side/ tenderness guarding	+	+	1	Normal	Normal	B-U: a left POC and both varies were of normal size, separately visualized, normal color and spectral flow in both ovaries.
37	Qian et al. <sup>33</sup>	<u>8</u>	10-days	R-side/ tenderness	Z	Z S	Z S	Normal	Z 5	B-U: Normal uterus and ovaries. A round, thick-walled, cystic structure, adjacent to the left ovary and mild fluid collection in the POD. MRI: a contrast-enhanced roundish structure.
38	Delacroix et al. <sup>34</sup>	13	N N	R-sided/ spontaneous	+	+	ı	Normal	Normal	B-U: an ovarian cyst with effusion in the recto-uterine pouch.
39	Same as above	<u>8</u>	Z S	L-sided (firstly)/ R-sided (6-year later)	+ +	+ +	ı +	Normal 16.00	UN 72.00	B-U: Left adnexal torsion was suspected. B-U: A normal-appearing ovary with a suspected POC.
9	O'Connell et al.³⁵	9	N N	R-side/ tenderness	+	1	I	Z 5	Z 5	B-U: A right adnexal cystic structure, normal-sized ovaries without edema and normal Doppler flow. CT: A large cystic structure within the pelvis, distinct from the right ovary.
4	Jacuzzi and Lodge <sup>36</sup>	<u>8</u>	4-days	L-sided/guarding	+	ı	+	16.00	167.00	B-U: A large right ovarian cyst, no obvious blood flow to the right ovarian cyst or parenchyma.
45	Khorshid and Tyson <sup>37</sup>	=	II-days	R-side/guarding rebound	Z	N N	+	S	Z 5	CT: A normal appendix and a 11.4 $\times$ 6.9 $\times$ 6.2 cm cystic structure in right abdomen as well as $1.3 \times 3.2 \times 2.8$ cm cystic structure in the left.
43	Present case	4	2-days	L-side/ tenderness	+	+	ı	6.95	Normal	B-U: A huge cyst in the pelvic cavity, with no echo, clear boundary, and sound transmission. CT: Left pelvic adnexal cyst.

-: negative manifestation; +: positive manifestation; A-Nor: Abnormal; B-U: B-ultrasound; CT: computed tomography; D: day; DAP: Duration of Abdominal Pain; H: hour; M: month; MRI: magnetic resonance imaging; N: number; POC: paraovarian cyst; R/L/B: Right/Left/Bilateral; UN: Unknown; Y: year.

 Table 2.
 Intraoperative and postoperative conditions of IFTT cases reported in adolescent girls.

Case (n) PD (A/I	PD (A/E)	EMEO TE/LE	Lesion side (L/R/B)	Torsion cycles (Circle)	Size of cyst (cm)	(cm) Procedure	Routine pathology	Follow-up (year/month)	Supplemental Material content (intraoperative exploration and other information)
02	∢ ш	22	<b>س</b> ک	2.0	$12 \times 10 \times 6.5$ $7.5 \times 5.5 \times 5.5$	R-SOO P-DPE-RT +Cy	A POC A POC	8-years 4-years	None A tense cyst in the broad ligament separated from the
03	ш	9	~	2.0	$5.0 \times 2.0 \times 1.0$	RS+Cy	Hydatid of Morgagni (HM)	2-months	A hemorrhagic and necrotic tube and a fimbrial cyst lined with flat cuboidal epithelium with features, origin from the hydarid of Morasani
40	ш	E	_	Z S	Large	RpS+Cy	Hydatid of Morgagni	Z S	The twisted distal portion of the left fallopian tube was resected.
90	ш	<b>"</b>	Ф	2.0 (L)/2.0 (R)	5.5 × 7.0(L) 5.5 × 9.0(R)	L-S R-S	paramesonephric cyst paramesonephric cyst	Z 5	both tubes were twisted twice and slightly discolored, but without necrosis. On both sides the tubes were distended by the cystic processes, and laterally the fubbes were adherent to the cysts.
90	ш	<b>"</b>	_	4.0	4.0 × 5.0	L-S	paramesonephric cyst	Z S	a cyanotic left tube twisted four times and in close proximity to a $4.0 \times 5.0$ cm large cystic process. Both ovaries were normal and no other abnormalities were found.
07	шш	<u> </u>	<b>~</b> _	3.0	4.1 × 4.2 6.24	RS LS+ BCv	Hematosalpinx (L) hydrosalpinx	Z Z	An excessively lengthy right fallopian tube. The left fallopian tube was detorsed and failed.
3	ı	ł	ı	9	-	()	(R) Paratubal cyst	5	
60	ш	빌	_	2.0	4.7 × 3.2	LS+ Cy	Hydrosalpinx	Z S	Mild fluid collection in PDF, Detorsed left fallopian tube with its middle part and a hick-walled cystic dilation at its distal portion.
0_	ш	끸	~	2.0	5.6 × 4.8	RS+ Cy	Hydrosalpinx	N S	A right-sided elongated hydrosalpinx, twisted twice on its pedicle with moderate congestion. Left fallopian tube, ovaries, uterus, and appendix all grossly normal.
=	ш	TE/LE	_	2.0	4.8	LS+ Cy	Pyosalpinx/ hematosalpinx	N S	An enlarged and edematous left fallopian tube that was filled with blood. The left ovary, right tube, and right ovary were normal.
12	ш	빌	_	2.0	9.0 × 1.1 × 1.2	LS+ Cy	A benign tubal cyst	Z	A urine dipstick revealed a tube with extensive hemorrhage. Large amount of blood.
13	∢	빌	_	2.0	4.0 × 6.0	LS+ Cy	Hematosalpinx	Z S	A dilated and twisted plum-colored salpinx on the left side.
4	ш	끧	_	0.5	6.5 × 6.1	P-DPE+RT +Cy	P-DPE+RT +Cy Simple tubal cyst Paratubal cysts	Z S	Tubal cyst was drained, and simply peel the cyst out of the viable fallopian tube. paratubal cysts in its distal.
15	ш	<b>H</b>	<u>«</u>	2.5	5.0 × 5.0	RS+Cy	Paratubal cyst (mesothelium)	N	Tumor markers were normal.
9	ш	E	_	2.5	2.0	LS+Cy	Paratubal cysts	N O	The right fallopian tube had congested, hemorrhagic, and infracted.

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Table 2. ((	

Case (n) PD (A/I	PD (A/E)	EMEO TE/LE	Lesion side (L/R/B)	Torsion cycles (Circle)	Size of cyst (cm)	(cm) Procedure	Routine pathology	Follow-up (year/month)	Follow-up Supplemental Material content (intraoperative (year/month) exploration and other information)
17	ш	끸	~	2.0	9.0	RS+Cy	Paratubal cyst	N S	Several torsion of the right tubal remnant and two paratubal cysts were noted.
<u>8</u>	ш	TE	~	Z 5	7.0	RpS+Cy	Fimbriectomy	N	Partial viable fallopian tubes have been preserved
61	⋖	쁘	~	N S	$7.3\times5.7\times5.6$	RS	Paramesonephric cyst	N	Dark-red, necrotic-appearing edematous right fimbriae.
20	⋖	쁘	~	3.0	4.I×4.2	P-DPE+RT +Cy	Paramesonephric cyst	N	The presumptive diagnosis was acute appendicitis.
21	ш	里	_	2.0	5.0 × 5.0	P-DPE+RT +Cy	P-DPE+RT +Cy Necrotic fallopian tube (UN)	3-years	A hemorrhagic paratubal cyst located just near to fimbria.
22	ш	끸	<b>∝</b>	4.0	<u>N</u>	RS+ Cy	Mesonephric paratubal cyst.	4-years	Hot isotonic solution application.
23	∢	쁘	~	Z S	5.5	P-DPE+RT +Cy	onephric paratubal	2-years	The right fallopian tube was detorsioned and a
24	ш	当	ď	Z 5	Z 5	P-DPE+RT +Cy	Cyst P-DPE+RT +Cy Hydatids of Morgagni (HMs)	N S	paratubal cyst was seen  Tubal torsion linked cystic structures which arise from the fimbriated to hydrosalpinx as well as paratubal cysts (HMs).
25	ш	빌	_	S	N S	P-DPE+RT +Cy	P-DPE+RT +Cy Hydatids of Morgagni (HMs)	Z 5	Tubal torsion has been linked to hydrosalpinx as well as paratubal cysts
26	ш	빌	<b>∝</b>	4.0	8.0 × 4.0	P-DPE+RT +Cy	VO	3-months	The fallopian fimbriae showing signs of necrosis. Retention of twisted fallopian tube.
27	∢	쁘	<b>∝</b>	4.0	5.0 × 4.0	RS	Tubal tissue with diffuse necrosis	Z 5	The prediagnosis of right tubal torsion by MRI.
28	ш	쁘	_	Multiple	3.5	RS+ Cy	The cystic conglomerate UN tumor	Z 5	Within the pelvis a conglomerate tumor was seen, consisting of a massively enlarged fimbrial funnel and a para-oval cyst.
29	ш	빌	_	S	$5.3\times3.5\times2.0$	LS	Existence of tissue with UN hemorrhagic	Z 5	None
30	ш	2	~	N N	5.4×4.4	P-DPE+RT/ RSOO Appendectomy	Benign serous ovarian cystadenoma	Z S	Recurrence after conservative treatment.
3	ш	삨	_	<u>S</u>	8.0	LS	Hemorrhagic necrosis and hydrosalpinx.	Z S	A massively enlarged fimbrial funnel and a POC tumor with torsion of the distal part of the tube.
32	∢	삨	_	2.0	5.8 × 3.2	SI	Hematosalpinx	Z	A black, necrotic-appearing, edematous left fimbriae were noted with left fallopian.
33	ш	끧	~	S	$7.0 \times 5.0$	RS+Cy	Hemorrhagic necrosis	N S	Right tubal torsion was found due to an adjacent POC.
34	ш	쁘	~	Z S	12.0	RS	POC	N N	Dark-red, necrotic-appearing edematous right fimbriae.
35	⋖	쁘	_	0.1	3.0	RS	Tubal congestion with	N S	Right tubal torsion was found possibly due to an
							tissue damage		adjacent 6 cm POC.

(Continued)

Table 2. (Continued)

Follow-up Supplemental Material content (intraoperative (year/month) exploration and other information)	UN A twisted, ischemic distal part of the right fallopian tube was seen. Right tubal torsion was found extraordinarily long of about 12 cm.	UN A torsion of the right fallopian tube twisted two times, complicated with hematosalpinx, pelvic hemoperitoneum.	UN The presumptive diagnosis was acute appendicitis.	6-years (First) the patient was treated conservatively by unwinding and preserving the left IFTT. (secondly) A large pyosalpinx was found requiring LS.	2-weeks The patient was found to have right-sided measuring, 5.0 cm in maximum dimension, and left-sided measuring $7\times5\times1$ cm.	UN A large fluid-filled hemorrhagic structure arising from left fallopian tube associated torsion, with 900° of rotation, of the left fallopian source of the serositis tube and an acutely inflamed Appendix.	UN A large mass in the right mid and upper abdomen and a confirmed right hydrosalpinx.	I-year A huge paratubal cyst between the distal end of the left fallopian tube and the fimbria end.
Routine pathology	Serous papillary cystadenoma	Tubal tissue necrosis, hemorrhagic, a small serous cyst	P-DPE+RT +Cy Paramesonephric cyst	P-DPE+RT +Cy (L) No specimen RS (R) Serous cyst.	Hydrosalpinx and bilateral POS	P-DPE+RT +Cy Hydrosalpinx and Appendicectomy inflamed appendix.	Hydrosalpinx	$10.9 \times 8.3 \times 8.9~\text{P-DPE+RT}$ +Cy Paramesonephric duct cyst, hydrosalpinx.
) Procedure	P-DPE+RT	RS	P-DPE+RT +C	P-DPE+RT +C RS	RS +BCy P-DPE+RT	P-DPE+RT +C Appendicecton	RSOO+LS	P-DPE+RT +C
Torsion Size of cyst (cm) Procedure cycles (Circle)	6.0	3.8 × 3.4	$5.6\times7.3\times5.7$	UN 2.6 × 2.6	(R) 5.0 (L) 7.0 × 5.0	Large	(R) 11.4 $\times$ 6.9 (L) 3.8 $\times$ 2.8	10.9 × 8.3 × 8.9
Torsion cycles (Circle)	3.0	2.0	3.0	<u> </u>	Z	2.5	S	4.5
Lesion side (L/R/B)	~	<b>∝</b>	8	L(First) 1 R(Second) 1	<b>∝</b>	_	~	_
EMEO TE/LE	里	<b>"</b>	끸	H	끸	쁘	끸	끸
Case (n) PD (A/E)	∢	ш	∢	∢	∢	ш	ш	ш
Case (r	36	37	38	39	40	4	42	43

A/E: accuracy/error; B: bilateral; BCy: bilateral cystectomy; Cy: cystectomy; EMEO: exploration mode of emergency operation; L: left; LE: laparoscopic exploration; LS: left salpingectomy; PD: preoperative diagnosis; PDF: pelvic Douglas fossa; POC: paraovarian cyst; P-DPE+RT: Preservation-Detorsion, Puncture and Evacuation of the affected fallopian tube + Release from Torsion; R.S. right partial salpingectomy; RS: right salpingectomy; R-SOO: right salpingo-oophorectomy; RT: release from torsion; TE: transabdominal exploration; UN: unknown; y/m/w/d: year/month/week/day.

found that the onset age of IFTT in pubertal girls is concentrated in  $14.09 \pm 2.48$ , which is the age of menarche. In addition to congenital causes, such as abnormal fallopian tube development,<sup>30</sup> we speculate that puberty may promote the pathogenesis of endocrine activity.<sup>28,31</sup> And the periodic activity of endocrine sex hormones may play a role in the residual fallopian tube accessory mesonephric epithelium, resulting in the progressive enlargement of mesosalpinx cyst, which increases the risk of fallopian tube torsion. This also may be one of the reasons why IFTT is more common in women of reproductive age. 34 In many cases, it was found that the pathological changes of the fallopian tube of IFTT were mostly caused by the torsion of the enlarged paraovarian or parafallopian cyst. Paraovarian cysts (POCs) also referred to as paratubal cyst or hydatid cyst of Morgagni, represent approximately 10% of adnexal masses. 39,40 And this study also showed except for 37.21% of hematosalpinx/ hydrosalpinx, <sup>28–30</sup> the POCs accounted for 58.14% of pathoanatomical etiology of IFTT. At the same time, this study found that the right side incidence rate of IFTT was 50.00%, slightly higher than the left side 45.24%, whereas the bilateral incidence rate was only 4.76%, of which one case<sup>34</sup> developed from the left side to the right side, with an interval of 6 years. This is also consistent with the results reported in the previous literature. 21,28,29 IFTT is more common on the right side, and this may be attributed to anatomical reasons that the sigmoid colon occupies the left pelvic cavity. And right IFTT is more often surgically explored suspecting appendicitis. 25,31,36 However, the specific epidemiological causes need to be furtherly studied.

Presenting symptoms of 43 case IFTT patients include acute onset of lower abdominal pain (100%) that can be accompanied by nausea (67.44%), vomiting (51.16%), and localized peritoneal irritation sign (44.19%). Fever is rare except for complication of infection or necrosis. 2,25 By analysis of the DAP of 10-case fever patients, 6,17,19,25,29,31,34,36,37 especially for one case which displayed no fever firstly, but fever after 3 weeks.<sup>26</sup> We found the DAP of fever patients showed obvious extension; however, the risk of fever increases significantly with the prolongation of DAP  $(\chi^2 = 15.57, p < 0.001)$ , and this report of Martin-Vallejo also confirmed it.<sup>25</sup> Moreover, the number of adverse outcomes such as fallopian tube necrosis and resection in febrile patients significantly increased (p < 0.05), but it seems to be unrelated to the number of tubal twists (p > 0.05), which fully indicates that the treatment of IFTT requires early detection and intervention.

Due to lacking typical specific symptoms, the preoperative diagnosis of IFTT is extremely difficult, which is usually determined during the operation. Sonographic diagnosis may be one of the most direct and fast methods. Based on the imaging performance of 43 cases and sonographic findings of IFTT reported by Ormasa, <sup>20</sup> the basic criteria for sonographic diagnosis of IFTT were (1) cyst mass separate from ovary (17/43) including paraovarian/parasalpinx cyst

(15/43); (2) A dilated tubular structure with a tapering end (peak sign, 5/43) adjacent to a normal ipsilateral ovary with normal blood flow (36/43); (3) or a midline cystic mass (either in the posterior cul-de-sac or superior to the uterus) associated with a normal ipsilateral ovary (12/43), and (4) pelvic effusion of different degrees (10/43). Although the sonographic whirlpool sign may be the specific for IFTT, sonographic diagnosis of IFTT was also very difficult, <sup>38,41</sup> as was CT. Enhanced magnetic resonance imaging (MRI) may be the most reliable imaging method for the preliminary diagnosis of IFTT.<sup>13</sup> Sakuragi et al.<sup>42</sup> have reported that there are several findings on MRI that can be used to support the diagnosis of IFTT, such as plicae tubariae of the twisted fallopian tube, whirlpool sign, and ovary on the affected side. Moreover, by analyzing 12 cases of IFTT with accurate preliminary preoperative diagnosis in this study, we found that the accuracy of preoperative diagnosis did not completely depend on imaging results, only two cases were indicated by ultrasound, four cases by enhanced MRI, and five cases by enhanced MRI combined with CT, but depend on whether the clinical gynecologists or pediatricians were aware and consider of IFTT. Durairaj and Gandhiraman<sup>43</sup> retrospective analysis of IFTT also fully confirmed the importance of clinicians' awareness and understanding of IFTT. At the same time, the acknowledge of sonographic diagnosis related to IFTT is extremely Insufficient, this may be the key point of accuracy pre-operation diagnosis of IFTT.

Although preoperative diagnosis can be suspected and considered based on imaging, IFTT is ultimately diagnosed during exploratory surgery. It is well known that with the prolongation of IFTT's DAP, the risk of secondary necrosis, hemorrhage, and infection of the twisted fallopian tube increases sharply, often causing irreversible damage to the fallopian tube, and even affecting the function of the ipsilateral ovary. 5,28,29 Without early surgical intervention, the fallopian tube may undergo irreversible vascular changes, requiring salpingectomy and possibly salpingoophorectomy.<sup>29,42</sup> This is of particular concern in adolescent girls. As for exploration mode of emergency operation, there is no doubt that laparoscopic exploration is preferred method and gold standard for the diagnosis and treatment of IFTT. 30,32,39 Although there is no sufficient research evidence on the improvement of fertility function after fallopian tubal reservation, fallopian tubal reservation surgery (P-DPE +RT)<sup>5,22,32,35,36</sup> is still the first choice at present, especially in the early stage of IFTT, 44 which may be the reason for the renewal of the surgical plan for ovarian torsion in adolescent girls. 45 This reservation surgery (P-DPE+RT) also confirmed that it was consistent with the two-step conservative surgical management proposed by Boukaidi et al.<sup>46</sup> However, it was controversial that the second-look laparoscopic and salpingoscopic surgery scheduled several weeks after the first surgical procedure. And it is not recommended to perform salpingopexy to prevent recurrence, because it may reduce the reproductive function of fallopian tube.<sup>27</sup>

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# Conclusion

IFTT in adolescent girls is a very rare acute abdomen. Because of its non-specific clinical manifestations and extremely difficult clinical accurate preoperative diagnosis led to the adverse clinical outcome of salpingectomy which seriously affected the future reproductive function. However, improving the knowledge of IFTT ultrasound diagnosis and the awareness of clinical gynecologists or pediatricians can improve the accuracy of preoperative diagnosis. The emergency laparoscopic exploration is the only gold standard for diagnosis and treatment. The operation of preserving the fallopian tube (P-DPE+RT) may be the first choice, especially in the early stage of IFTT.

### **Author contributions**

Initial drafting of the manuscript and subsequent revisions: Z.-J.L.; surgical and medical practices: Z.-J.L.; statistical analysis: Z.-J.L. and Y.-Z.; literature search and clinical data collected: W.P. and Y.-Q.Z. All authors have read and approved the final manuscript.

# **Declaration of conflicting interests**

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

### **Funding**

The authors received no financial support for the research, authorship, and/or publication of this article.

### **Ethical approval**

Ethical approval to report this case was obtained from \*the Ethics Committee of the Longquan People's Hospital affiliated to Lishui University (APPROVAL NUMBER/ID: 2023-04-001-EC-LQSRMYY)\*.

### Informed consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

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