

A Fifteen Year Analysis of Rare Isolated Fallopian Tube Torsions in Adolescent Children: A Case Series

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ABSTRACT

Background: Isolated fallopian tube torsions presenting to the emergency department are a very rare cause of childhood acute abdominal pain. Since the diagnosis to be made in the early period is of importance in terms of affecting tubal damage and fertility, it was aimed to evaluate the cases in the light of literature.

Materials and Methods: This study included 10 patients under 18 years of age presented to the emergency department with abdominal pain between January 2003 and December 2018. The mean age was 14.5 ± 1.43 years (range: 12-17 years). The demographic characteristics, surgical findings, and methods, concomitant pathology results of these patients were retrospectively evaluated.

Results: The reason for admitting to the emergency department of 10 patients included in the study was abdominal pain. The mean duration of hospital admission with pain was 4.97 days. The onset of pain was less than 24 hours in 7 patients (70%) and was more than 24 hours in three patients (30%). Of the patients, 9 (90%) had tenderness in the lower abdominal quadrant, 5 (50%) had the defense, and 3 (30%) had a rebound. Nausea, vomiting, and

leucocytosis were present in 50% of the cases. Right and left tubal involvement of the cases was equal. Seven (70%) of the isolated tubal torsions were accompanied by paraovarian cysts. Of the patients, 8 (80%) underwent open surgery and 2 (20%) underwent laparoscopic intervention. Detorsion was performed in 5 (50%) and salpingectomy was performed in 5 (50%) cases.

Conclusion: *Isolated tubal torsion should be considered in children presenting with acute abdominal pain in early adolescence. Early diagnosis is important for the maintenance of fertility.*

Keywords: emergency department acute abdominal pain; isolated fallopian tube torsion; detorsion; salpingectomy

1. Introduction

Isolated fallopian tube torsion (IFTT) is the rotation of the fallopian tube on its own axis without ovarian torsion. The incidence is estimated to be 1/1.500.000 women¹. The etiology is uncertain. Anatomical abnormalities such as long mesosalpinx, hydro/hematosalpinx, tubal mass/neoplasm, adnexal mass (ovarian/paraovarian tumor), and physiological abnormalities such as abnormal peristalsis or periovulatory spasm, hemodynamic abnormalities such as venous congestion, Sellheim theory indicating sudden body position changes, trauma, pelvic inflammatory disease, pelvic adhesion, previous surgery/disease such as tubal ligation, and enlarged uterus/uterine mass may play a role in the etiology^{2,3}. Specific symptoms, clinical findings, imaging, and laboratory characteristics could not be determined so far⁴. It is rarely diagnosed before surgery. Detorsion or salpingectomy is performed in the treatment.

The aim of this study was to present the results of 10 patients with IFTT in the light of literature and to contribute to the diagnosis and treatment.

2. Materials and Methods

Study design and population

Patients diagnosed with isolated fallopian tube torsion between January 2003 and December 2018 were retrospectively analyzed using the electronic medical record system. The study included girls aged between one month and 18 years and patients diagnosed with isolated fallopian tube torsion during operation. Patients under one month of age and above 18 years of age, and patients who had fallopian tube torsion with ovarian torsion were excluded from the study. The patients' age, characteristic and localization of abdominal pain, presence of nausea and vomiting, physical examination findings, preoperative laboratory and radiological imaging findings, affected side, surgical findings, method and procedure, presence of concomitant pathology and examination results were recorded.

3. Statistical Analysis

The data obtained from this study were collected from the hospital's electronic medical record system. They were then analyzed with the SPSS 15.0 software package. The descriptive statistics were shown as mean±standard deviation or median (minimum-maximum) for continuous variables and a number of cases and percentage (%) for nominal variables. Other data were analyzed using Microsoft Excel and simple descriptive statistics.

4. Results

The mean age of 10 patients included in the study was 14.5 ± 1.43 (range: 12-17) years. The reason for admitting to the emergency department of the patients was abdominal pain. The mean duration between the onset of pain and hospital admission was 4.97 days (six hours-one months). Of the patients, five had abdominal pain in the left lower quadrant, two had in the right lower quadrant, one had in the lower quadrant, and two had in the whole abdomen. All but one of the sexually inactive patients were menstruating. Of the patients, five had nausea and three had vomiting. Two of the patients had nausea and non-bilious vomiting. The patients had no trauma, constipation, diarrhea, vaginal discharge, or dysmenorrhea

complaint. Only one patient had dysuria. Of the patients, 90% (9) had lower quadrant tenderness, five had the defense, and three had a rebound on physical examination (Table I).

White blood cell (WBC) level was high ($>11000/\text{mm}^3$) in 5 patients, while it was within the normal range ($4000-11000/\text{mm}^3$) in 5 patients. The mean time to onset of symptoms was 8.4 days in the patients with a high WBC value, while it was found to be shorter in those with a WBC value within normal range with 0.95 days. Neutrophil dominance was detected in 70% of the patients. In the patients with relatively low lymphocyte percentage, platelet counts were within a normal range.

Urea, creatinine, amylase, aspartate transaminase, alanine aminotransferase, gamma glutamyl transferase, sodium, potassium and C-Reactive protein (CRP) values of all patients were normal. The values of three patients whose Alpha-fetoprotein (α -FP), β -Human Chorionic Gonadotropin (β -HCG), Cancer Antigen-125 (CA-125) were studied were found to be normal.

Direct abdominal X-ray and Doppler ultrasonography (US) were performed on all patients and magnetic resonance imaging (MRI) was performed on two patients. Direct abdominal X-rays of all patients were evaluated as normal. Paraovarian cyst/mass was detected in seven patients who had undergone the US, while two patients were evaluated as normal. Due to the presence of free fluid in the pelvic region of one patient, it was thought to be an ovarian cyst. Four patients were pre-diagnosed with ovarian torsion due to the absence or reduction of ovarian blood supply. Pelvic free fluid was identified in four patients and paraovarian cyst was detected in five patients. MRI was performed on one appendectomized patient with the diagnosis of paraovarian mass and this patient was diagnosed with IFTT preoperatively (Fig I).

Nine of the patients were operated on the day of admission. One patient was operated four days later due to non-healing pain, previous abdominal pain attacks and detection of

paraovarian solid mass on the US after her further examinations were performed. Of the patients, two were operated using the laparoscopic method, while eight underwent open surgical intervention. Torsioned tuba was equal on both sides. Five patients did not have any additional intervention because of normalization of blood supply after detorsion, while the other five patients underwent cystectomy /salpingectomy (Fig II). The pathological examination of the resected materials revealed changes of chronic salpingitis along with serosal cysts (Table II).

5. Discussion

Isolated fallopian tube torsion (IFTT) is one of the rare causes of acute abdominal pain in adolescent girls. In the literature, it is usually presented as case reports. In recent years, there are publications in the form of case series⁵⁻⁷. Tubal torsion more frequently arises on the right side. The reason for this is that the mobility of the left tuba with the sigmoid colon is more restricted and right lower quadrant pains are diagnosed by opening the abdomen with suspicion of acute appendicitis^{8,9}. In our study, it was present on both sides with the same frequency. It was also similar to the multicentre study by Bertozzi et al. which found an equal number on both sides⁸.

Paraovarian cyst/mass is blamed in the etiology of IFFT. Paraovarian cyst was detected in five of our cases. In addition, hydro/hematosalpinx was identified in one patient. There are studies that associate isolated fallopian tube torsion with some sports involving sudden body movements^{10,11}. In our study, two patients were actively engaged in sports.

Lower abdominal pain is common in IFFT; however, lower abdominal pain is not specific to IFFT, and similar manifestations may also arise in other surgical pathologies causing acute abdomen³. All patients had a complaint of abdominal pain. Of the patients, eight had abdominal pain in the lower abdominal region, while two had abdominal pain in the whole abdomen. The onset of pain ranged from six hours to one month from admission and was

similar to the study by Harmon et al¹². Whereas, the complaint of nausea and vomiting varied in the patients.

On physical examination, there is no specific examination finding suggestive of IFTT. Of the patients, nine had tenderness, five had the defense, and only three had a rebound. WBC was above the normal values in five patients, while it was within the normal range in other patients; however, seven patients had neutrophil dominance. The presence of neutrophil dominance in 70% of the patients was notable. Whereas, CRP was within the normal range in all of our patients. There was no specific laboratory finding of IFTT in our patients, and as stated by Bertozzi et al., the contribution of laboratory parameters to the diagnosis in IFTT patients was not clearly obvious⁶.

Preoperative diagnosis of isolated fallopian tube torsion is difficult since there is no pathognomonic imaging method, specific symptoms or characteristic laboratory findings¹³. The diagnosis is usually made during surgical intervention. In our series, one patient was preoperatively diagnosed, while other patients were diagnosed during surgery. The MRI of the patient diagnosed preoperatively revealed thickened fallopian tube walls, tubal rotation, hydro/hematosalpinx, which were evaluated in favor of IFTT. The preoperative diagnosis was ovarian torsion in four patients. Five patients were diagnosed with paraovarian cyst on USG. IFTT was identified on the side where these cysts were present during operation and torsioned fallopian tubes were removed by being evaluated to be necrotic. Paratubal cysts, called hydatid cysts of Morgagni, are very rarely neoplasia¹⁴. In the study, the cystic structures present in five patients were diagnosed with simple serosal cyst as a result of the pathological examination. In our patient preoperatively diagnosed with solid mass, the only cystic structure was found during the operation.

Pathologies such as appendicitis, ovarian torsion, ectopic pregnancy, pelvic inflammatory disease, the ruptured ovarian cyst should be considered in the differential diagnosis (9). In

terms of preserving fertility, early diagnosis and early surgical intervention may be possible by preserving the fallopian tube, avoiding possible salpingectomy. Saving the fallopian tube is a rare condition due to the difficulty in early diagnosis. Mazouni et al. emphasized that duration from onset of pain to surgery longer than 10 hours increases the risk of tubal necrosis¹⁵. In our study, half of the patients did not have tissue loss because of the normalization of blood supply to the fallopian tube after detorsion. Of these patients, three were operated within the first 24 hours from the onset of their complaints, while the other two were operated after more than 24 hours.

Salpingectomy or detorsion is the preferred methods in surgical treatment. The necrotic appearance of the tubes is taken as a reference when deciding on salpingectomy; however, in recent studies, the observation of healthy cilia cells in the pathological examination of the removed tubes leads on to a discussion of maybe leaving the fallopian tubes in place as in ovarian torsion¹⁶. However, in such a case, it is recommended to follow up patients with USG until adulthood. However, no matter how necrotic it appears, the conservative treatment of fallopian tube does not affect morbidity. This reveals the need for future studies in terms of the functionality of fallopian tubes.

The most important limitation of the study is the retrospective and single-center design. Another important limitation is the small sample size and the selection of very rare cases. Moreover, the difficulty in accessing the records and radiological imaging and inability to obtain adequate information on the patients' medications used and personal history were the main limitations.

6. Conclusion

In this study, no specific clinical, physical examination or laboratory finding was identified when the results of 10 patients with IFTT were analyzed over a period of 15 years. IFTT is a cause of acute abdomen which should be kept in mind in patients presenting with

lower abdominal quadrant pain. Although USG is not very helpful in the diagnosis, the presence of paraovarian cystic masses in the affected area is an important parameter in terms of reminding IFTT. In terms of fertilization, preservation of fallopian tubes is possible with early surgical intervention. Although there are views on the preservation of necrotic fallopian tubes in recent times, there is no adequate information about the long-term results. Further studies are needed to be conducted in this respect.

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Table I: Baseline characteristics, laboratory and clinic finding of study patients.

	Isolated fallopian tube torsion	
	All patients	n (%)
Baseline Characteristic		
Age, mean±SD, yr	14.5±1.43	
Laboratory Finding		
WBC, mg/dL	10.47±2.40	
Neutrophils, %	76.9 ±7.58	
Lymphocytes, %	18.1±5.94	
BUN, mg/dL	16.24±8.19	
Crea, mg/dL	0.73±0.24	
ALT, mg/dL	17.21±7.23	
AST, mg/dL	19.44±7.31	
ALP, mg/dL	86.24±48.51	
BS, mg/dL	94.11±18.82	
Clinic Finding		
Presence of Pain		10(100)
Time to start pain		

Less than 24h	7(70)
More than 24h	3(30)
Shape of pain	
Colic	3(30)
Continuous pain	7(70)
Nausea	5(50)
Vomiting	5(50)
Vaginal discharge	0(0)
Menarche	9(90)
Physical examination	
Abdominal tenderness	9(90)
Abdominal defender	5(50)
Abdominal rebound	3(30)
Imaging methods	
Direct abdominal radiography	10(100)
US	10(100)
MRI	2(20)
Tumor Markers	2(20)
Paraovarian cyst	7(70)
Doppler current reduction / absence	3(30)
Surgical intervention method	
Open surgery	8(80)
Laparoscopic	2(20)
Place of torsion	
Right	5(50)
Left	5(50)
Operation performed	
Detorsion	5(50)
salpingectomy	5(50)

Yr: year, WBC: White Blood Cell, BUN: Blood Urea Nitrogen, Crea: Creatinin, ALT: Alanine Aminotransferase, AST: Aspartate Aminotransferase, ALP: Alkaline Phosphatase, BS: Blood Sugar, US: Ultrasonography, MRI: Magnetic resonance imaging

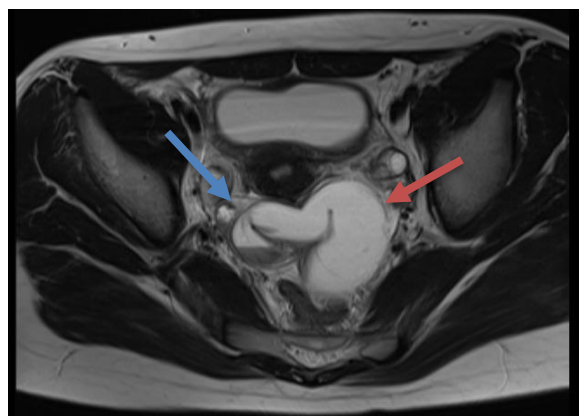
Table II: Characteristics of fallopian tube torsion cases

Age	Localization	RDUS	MRI	Preop Diagnosis	Postop Diagnosis	Accompanying Pathology	Procedure	
1	14	Lower left	Left paraovarian cyst Free liquid	None	Acute Abdomen	Left IFTT	None	Open Salpingectomy
2	13	Lower right	Normal	None	Acute Abdomen	Right IFTT	None	Open Detorsion, Cyst excision
3	14	Lower left	Right paraovarian mass	Semisolid cystic paraadnexial	Paraovarian mass	Right IFTT	Paratubal kist	Open Salpingectomy, Cyst excision
4	15	All Abdomen	Right paraovarian cyst Free liquid	None	Right ovarian torsion	Right IFTT	Paratubal kist	Open Salpingectomy, Cyst excision

5	17	Lower left	Left paraovarian cyst	None	Left ovarian torsion	Left IFTT	None	LAP Detorsion
6	16	Lower right	Right paraovarian cyst Free liquid	None	Right ovarian torsion	Right IFTT	None	LAP Detorsion
7	12	All Abdomen	Normal	Fallopian tube wall thickening Hidrohematos alpinx	IFTT	Right IFTT	None	Open Salpingectomy
8	15	Lower left	Free liquid	None	Ovarian cyst rupture	Left IFTT	Paratubal kist	Open Detorsion Cyst excision
9	14	Sub-dial	Left paraovarian cyst	None	Acute Abdomen	Left IFTT	Paratubal kist	Open Salpingectomy, Cyst excision
10	15	Lower left	Left paraovarian cyst	None	Left ovarian torsion	Left IFTT	None	Open Detorsion

RDUS: Color Doppler Ultrasonography, MRI: Magnetic resonance imaging, IFTT: Isolated fallopian tube torsion LAP: lymphadenopathy

Figure I: Magnetic resonance image of a patient diagnosed with preoperative isolated fallopian tube torsion



MRI; T1A sections show blue arrow torsion, red arrow indicates hydrohematosalpinx

Figure II: Isolated fallopian tube torsion material caused by hydrohematosalpinx



Ethical Statement: All subjects gave their informed consent for inclusion before they participated in the study. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of Project identification code :**2019-05/16-22/05/2019**.

CUMHURİYET ÜNİVERSİTESİ GİRİŞİMSEL OLMAYAN KLİNİK ARAŞTIRMALAR ETİK KURULU KARAR FORMU

ANIN AÇIK ADI: Çocuklarda Nadir Bir Akut Batın Nedeni: İzole Tubal Torsiyon

Belge Adı	Tarihi	Versiyon Numarası	Dili		
ARAŞTIRMA PROTOKOLÜ			Türkçe <input checked="" type="checkbox"/>	İngilizce <input type="checkbox"/>	Diğer <input type="checkbox"/>
BİLGİLENDİRİLMİŞ GÖNÜLLÜ OLUR FORMU			Türkçe <input checked="" type="checkbox"/>	İngilizce <input type="checkbox"/>	Diğer <input type="checkbox"/>
OLGU RAPOR FORMU			Türkçe <input type="checkbox"/>	İngilizce <input type="checkbox"/>	Diğer <input type="checkbox"/>

Belge Adı	Açıklama
SIGORTA	<input type="checkbox"/>
ARAŞTIRMA BÜTÇESİ	<input type="checkbox"/>
BIYOLOJİK MATERYEL TRANSFER FORMU	<input type="checkbox"/>
ILAN	<input type="checkbox"/>
YILLIK BİLDİRİM	<input type="checkbox"/>
SONUÇ RAPORU	<input type="checkbox"/>
DİĞER:	<input type="checkbox"/>

Karar No: 2019-05/16 Tarih: 22.05.2019

Yukarıda bilgileri verilen başvuru dosyası ile ilgili belgeler araştırmann/çalışmann gerekçe, amaç, yaklaşım ve yöntemleri dikkate alınarak incelenmiş ve uygun bulunmuş olup araştırmann/çalışmann başvuru dosyasında belirtilen merkezlerden gerekli izin alınarak gerçekleştirilmesinde etik ve bilimsel sakınca bulunmadığına toplantıda katılan etik kurul üye tam sayısının salt çoğunluğu ile karar verilmiştir.

KLİNİK ARAŞTIRMALAR ETİK KURULU

KURULUN ÇALIŞMA ESASI: Klinik Araştırmalar Hakkında Yönetmelik, İyi Klinik Uygulamaları Kılavuzu, Helsinki Bildirgesi, Cumhuriyet Üniversitesi Girişimsel Olmayan Klinik Araştırmalar Etik Kurul Yönergesi

ANIN UNVANI / ADI / SOYADI: Prof. Dr. Muhittin Sönmez

Unvanı/Adı/Soyadı	Uzmanlık Alanı	Kurumu	Cinsiyet		Araştırma ile ilişki		Katılım *		İmza
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Dr. Hatice Özer	Patoloji	Sivas Cumhuriyet Üniversitesi, Tıp Fakültesi	E <input type="checkbox"/>	K <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	HO
Dr. Ercan Özdemir	Fizyoloji	Sivas Cumhuriyet Üniversitesi, Tıp Fakültesi	E <input checked="" type="checkbox"/>	K <input type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	EO
Dr. Gülşay Yıldırım	Tıp Tarihi ve Etik	Sivas Cumhuriyet Üniversitesi, Tıp Fakültesi	E <input type="checkbox"/>	K <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	GY
Dr. Binnur Bağcı	Beslenme ve Diyetetik	Sivas Cumhuriyet Üniversitesi, Sağlık Bilimleri Fakültesi	E <input type="checkbox"/>	K <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	BB
Dr. Üyesi Mehmet Ataş	Farmasötik Mikrobiyoloji	Sivas Cumhuriyet Üniversitesi, Eczacılık Fakültesi	E <input checked="" type="checkbox"/>	K <input type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	MA
Dr. Üyesi Engin Altınkaya	İç hastalıkları	Sivas Cumhuriyet Üniversitesi, Tıp Fakültesi	E <input checked="" type="checkbox"/>	K <input type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	EA
Dr. Üyesi Melih Ülgey	Protetik Diş Tedavisi	Sivas Cumhuriyet Üniversitesi, Diş Hekimliği Fakültesi	E <input checked="" type="checkbox"/>	K <input type="checkbox"/>	E <input type="checkbox"/>	H <input checked="" type="checkbox"/>	E <input type="checkbox"/>	H <input type="checkbox"/>	MU

planında bulunma

Kurul Başkanının Unvanı/Adı/Soyadı: Prof. Dr. Muhittin Sönmez

Author Contributions: Study conceptualization involved, C.G. and A.C; Methodology, A.C; Validation, C.G. and A.C; Formal analysis, C.G; Investigation, All authors.; Resources, C.G, and A.C; Data curation, C.G.; Writing—original draft preparation, C.G and A.C; Writing—review and editing, All authors; Visualization, C.G and A.C; Supervision, C.G and A.C; Project administration, C.G and B.K.; Funding acquisition, C.G.,

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