



Case Series

Isolated tubal twist: A case series of a rare event occurring at different times in reproductive life

C. Delacroix^a, N. Hcini^{a,*}, E. Vintejou^b, S. Kedous^a, G. Carles^a^a Department of Obstetrics and Gynaecology, West French Guiana Hospital Center, Saint-Laurent-du-Maroni, French Guiana^b Department of Obstetrics and Gynaecology, Arnaud de Villeneuve Hospital, Montpellier, France

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Isolated fallopian tube torsion (IFTT) is a rare but potentially serious cause of pelvic pain in women. Despite being a surgical emergency, this diagnosis is often overlooked before surgery. To raise awareness of this diagnosis among clinicians, we describe here five cases, which occurred at different times in reproductive life.

CASES PRESENTATION: We present five cases of isolated fallopian tube torsion at different ages (13–54 years). It often manifests with sudden onset of acute pelvic pain in four cases and chronic pelvic pain in one case.

At admission, patients were suspected of adnexal torsion (3 cases), genital infection (1 case), and renal pain (1 case). CT-scan showed IFTT in only one patient. Laparoscopic surgical management, performed by experienced surgeons, consisted of salpingectomy in 4 cases and conservative treatment in one case. The latter was complicated with hydrosalpinx 6 years later. All patients were followed in outpatient clinic at least one time after surgery and had favorable outcomes.

CLINICAL DISCUSSION: Given the rarity of the pathology and the lack of pathognomonic imaging, IFTT is rarely diagnosed before surgery. Its etiology is still unknown but hydrosalpinx following an infectious process seems to be a major risk factor.

CONCLUSION: Increasing awareness of this rare entity is advocated, especially in woman of reproductive age. Torsion should be evoked in front acute pelvic pain in patients with hydrosalpinx or paratubal cyst. Conservative management must be privileged especially in women of childbearing age and in pediatric population.

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1. Introduction

Isolated fallopian tube torsion (IFTT) was first described in 1890 by Bland-Sutton [1,2]. IFTT is a gynecological cause of acute pelvic pain. 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 finding. The diagnosis is usually made during surgery. Delayed treatment exposes to risk of necrosis, irreversible tube damage, and infection. It is a surgical emergency.

To raise awareness of this diagnosis among clinicians, we describe in the following report five cases of IFTT occurring at different times of reproductive life stages. We aimed to analyze

the clinical, imaging features, and surgical management of this rare entity. The work is reported in line with the SCARE criteria [3].

2. Presentation of case

2.1. Case 1

A 13-years old woman, non-smoker, with no past medical or surgical history, went to the emergency department at university hospital with symptoms of lower abdominal pain. The pain occurred suddenly with uncontrollable vomiting. The patient reported no digestive or urinary signs. She was on day 21 of a 28-day menstrual cycle.

Clinical examination revealed a spontaneous and reproducible pain in the right iliac fossa. It was relieved by level 2 pain relievers. Pregnancy was eliminated by a negative hCG test and biological test was normal.

* Corresponding author at: Department of Obstetrics and Gynaecology, West French Guiana Hospital Center, Saint-Laurent-du-Maroni, French Guiana.

E-mail addresses: hcininajeh@gmail.com, hcininajeh@hotmail.fr (N. Hcini).

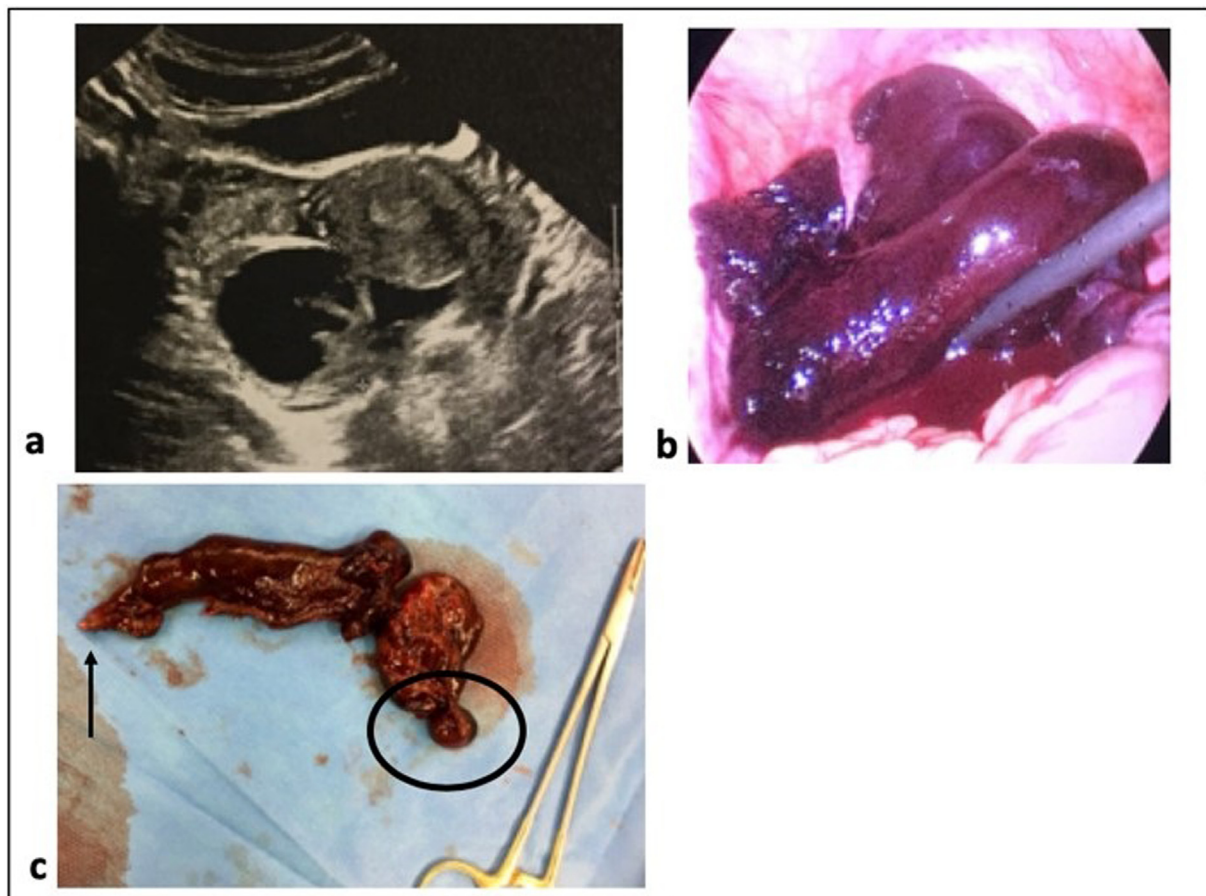


Fig. 1. a. Suprapubic ultrasound view: hypoechoic image evocative of tubal origin with incomplete septations. b. Laparoscopic view of the necrotic fallopian tube. c. Surgical specimen, right salpingectomy. The arrow indicates the ostium and the circle indicates the tubal cyst.

A suprapubic ultrasound showed an ovarian cyst of 3.8×3.4 cm in size with effusion in the recto-uterine pouch (56 mm), (Fig. 1a).

Adnexal torsion was suspected, and laparoscopy was performed (Fig. 1b). It revealed a torsion of the right fallopian tube twisted two times complicated with hematosalpinx, pelvic hemoperitoneum (100cc). Both ovaries and the left fallopian tube were normal. A detorsion of the right fallopian tube was tried, but it was necrotic and bulging. Consequently, a right salpingectomy was performed (Fig. 1c) with good postoperative evolution. Histopathological examination showed tubal tissue necrosis, haemorrhagic infarction with a small serous cyst within the necrotic mass.

2.2. Case 2

A 47-year-old woman with history of chronic pelvic pain reported suddenly increasing pain. Her menstrual periods had been regular with no relevant gynaecological history or medical problems.

On physical examination, at the time of admission, she was afebrile and normotensive. Blood tests were normal with negative hCG test. Pelvic ultrasound scan (Fig. 2a) revealed liquid partitioning in the anterior and lateral parts of the right side of the pelvis, covering an area of 11×7 cm with a poor vascularization. The right ovary was not visible. The uterus and the left ovary appeared normal. Tumor markers (CA-125, a-fetoprotein, CEA) were negative.

Abdominopelvic computed tomographic (CT) (Fig. 2b) showed a partitioned cyst measuring 93 mm adjacent to the right side of the uterus. A cyst of mucinous appearance was suspected, graded IOTA

B4. Thus, the patient was referred to a district general hospital and scheduled surgery was performed one month later (Fig. 2c).

The laparoscopy found a hydro-hematosalpinx of 10 cm with markedly dilated and tortuous fallopian tube. It was twisted two complete turns. Right salpingectomy was performed (Fig. 2d). The presence of false peritoneal cysts was suggested a chlamydia trachomatis infection. The right ovary and left adnexa were normal. The patient was treated by Azithromycin and the postsurgical outcome was favorable. Histopathological examination described a tube of 13 cm in length, with a dilated lumen (3.5 cm in diameter), flattened fimbriae and muscle atrophy and concluded to a hydrosalpinx with infectious origin.

2.3. Case 3

A 54-year-old woman with a past medical history of obesity (BMI 31) and hypertension consulted to the emergency department at a district general hospital with sudden onset of lower abdominal pain. It was localized in the right iliac fossa, radiating to the right flank and upper part of the thigh. The surgical history reported a bilateral tubal ligation ten years before, appendicectomy with ovarian cystectomy thirty years ago. The patient also reported similar pain six months before.

The urinary dipstick test reported two blood cross and the urine analysis was normal. The patient required level 3 pain relievers. Initially, acute renal colic was suspected.

CT uroscanner revealed no evidence of dilation or obstruction of the urinary tract. However, it revealed the presence of cystic

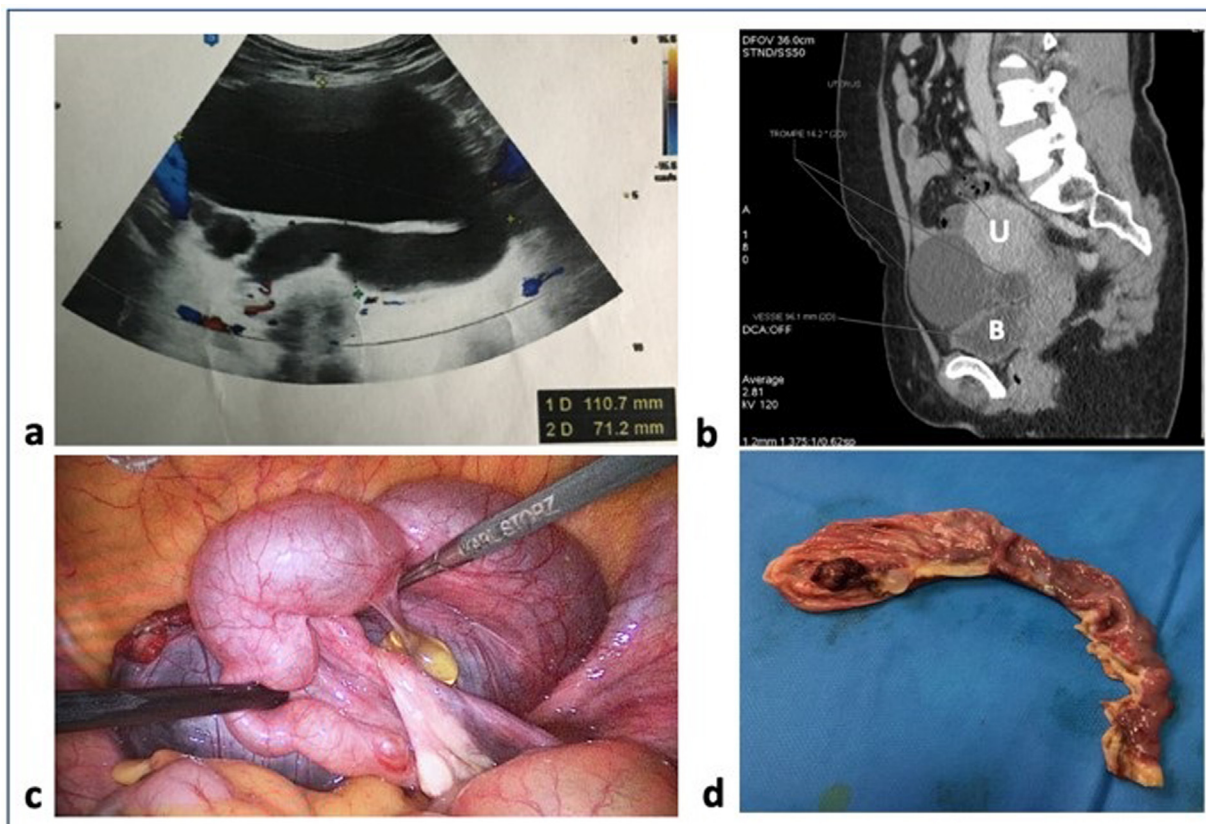


Fig. 2. a. Initial ultrasound view showing serpiginous fluid-fluid tubular mass. b. Abdominopelvic CT-scan show mass between the bladder and the uterus. c. laparoscopic view showing isolated tubal twist with hydrosalpinx. d. surgical specimen. The arrow indicates the ostium.

formations in the right ovary, the largest of which was 25 mm in diameter, and possible hydrosalpinx (Fig. 3a).

The patient was discharged home with pain medication. Two days later, she came back to hospital for increased pain. On the examination, the abdomen was soft but rebound tenderness was present in the suprapubic region with a strong pain. A vaginal examination triggered strong pain.

A pelvic ultrasound examination (Fig. 3b) revealed a cyst on the right side of the uterus. The cyst was unilocular, by 4 cm in diameter and vascularized. Additional to this, there was a probable right hydrosalpinx. Routine blood tests showed a raised CRP of 28 mg/L and 12×10^9 leukocytes/L.

Laparoscopy was performed and the diagnosis of isolated torsion was discovered (Fig. 3c). The right fallopian tube, seat of hydrosalpinx, twisted two times. It was edematous, haemorrhagic and seem non-viable. The right ovary was not included in the torsion. Right salpingectomy was necessary. Left adnexa appeared normal. The patient made an uneventful recovery. Histopathology report confirmed haemorrhagic, infarcted, and ischemic tube consistent with torsion. A malignant cause was excluded and not etiology was found.

2.4. Case 4

A 33-year-old patient with a history of Von Willebrand disease consulted at the emergency department at university hospital for sudden pain in the right iliac fossa that had woken her during the night. It was associated with an episode of vomiting.

She used a copper intrauterine device since her last delivery. The intensity of pain necessitated the introduction of opioid treatment.

The physical examination found a rebound tenderness in the right lower quadrant of the abdomen associated with fullness in the right recto-uterine pouch on vaginal examination. Laboratory tests were normal, and a pregnancy was excluded.

A vaginal ultrasound showed an oblong image an image suggestive of right hydrosalpinx (66 × 44 mm). Pelvic CT scan suspected IFTT (Fig. 4b).

Laparoscopy was performed and revealed a voluminous hematosalpinx, with bleeding associated with a three-turn torsion of the tube (Fig. 4c). The right ovary was not included in the torsion and appeared normal. The contralateral adnexa was normal. After detorsion and expectant attitude, the right tube remained blue. Therefore, salpingectomy was performed (Fig. 4d). She recovered uneventfully after surgery. Histopathological analysis confirmed a benign, infarcted hydrosalpinx resulting from infectious process.

2.5. Case 5

A 13-year-old woman without a significant past medical or surgical history presented to the emergency department at university hospital with pain in left lower quadrant of the abdomen that had occurred suddenly. Left adnexal torsion was suspected, and emergency laparoscopy was performed. Isolated fallopian tube torsion was discovered. Because of her age, the patient was treated conservatively by unwinding and preserving the tube. No immediate postoperative complications had occurred.

Six years later, the patient returned to the hospital with pain right lower quadrant of the abdomen. No digestive or urinary problems were noted. On examination, he was afebrile, with a normal cardiovascular examination. Spontaneous and reproducible pain

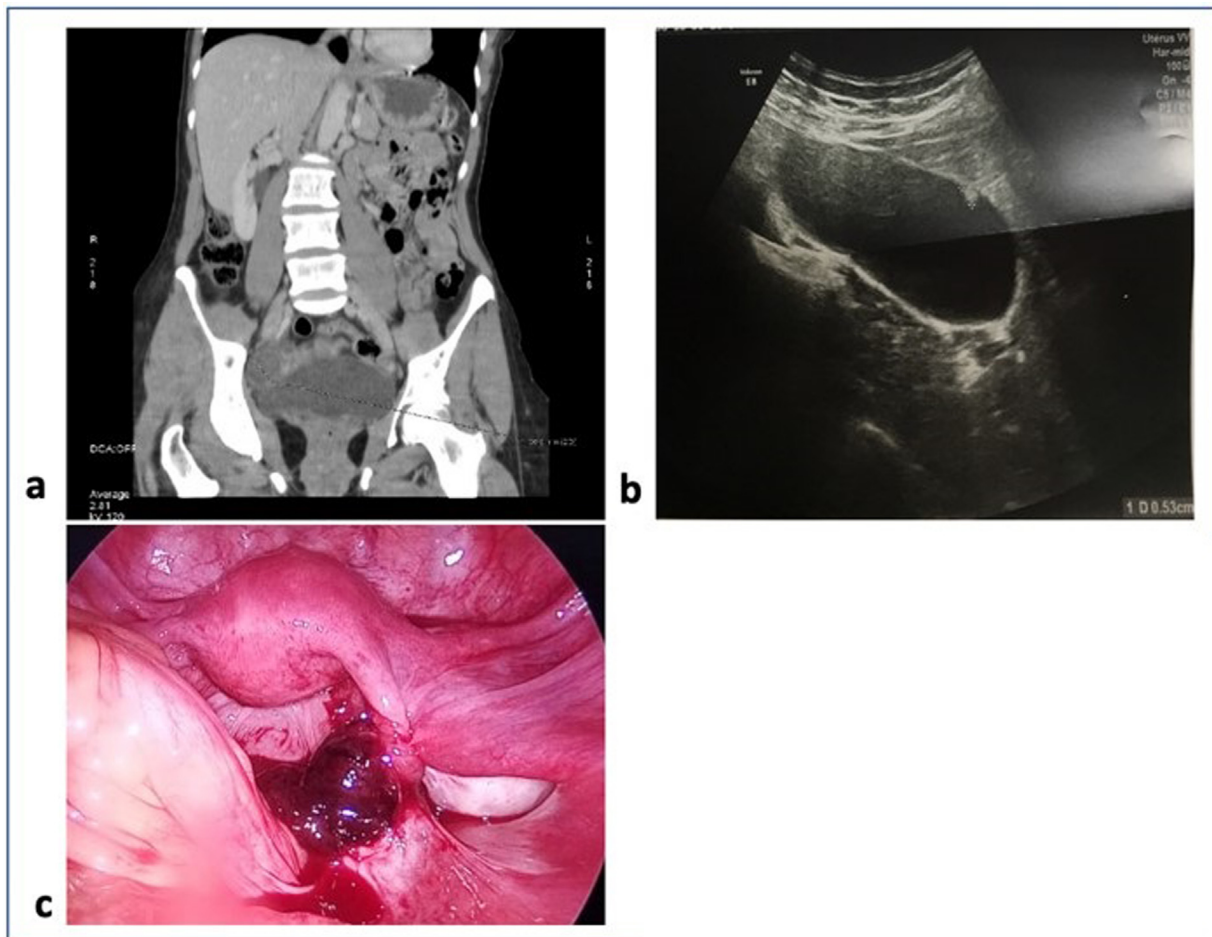


Fig. 3. a. Abdominopelvic CT-scan view, visualization of elongated structure interpreted as hydrosalpinx. b. Suprapubic US view showing a normal-appearing ovary with a suspected paraovarian cyst. c. laparoscopic appearance of twisted fallopian tube.

on palpation of the left iliac fossa, vaginal discharge, and painful uterine mobilization were founded.

Laboratory tests revealed an inflammatory syndrome: leukocytes = 16×10^9 leukocytes/L, CRP concentration of 72 mg/L and eliminated a pregnancy. Transvaginal ultrasound (Fig. 5a) showed elongated cystic mass measuring 26 × 26 mm in diameter, appearing to be a hydrosalpinx. A pelvic CT scan (Fig. 5b) confirmed it.

Patient received intravenous antibiotic therapy (metronidazole, doxycycline and Rocephin).

Due to a persistent pain with significant CRP increases, laparoscopy was performed. A large pyosalpinx (Fig. 5c) was found requiring a left salpingectomy. Right adnexa was normal. The outcome was favorable.

3. Discussion

Isolated fallopian tube torsion is uncommon cause of acute pelvic pain, rarely diagnosed before surgery. Women of childbearing age are most affected [4]. It is rare before puberty [5,2], in post-menopausal women [6,7], and even more rare during pregnancy [8]. The real incidence of IFTT is still unknown. It may be the consequence of an increase in risk factors, such as tubal ligation and sexually transmitted diseases. Many theories and predisposing factors have been cited in previous studies as tubal sterilization [9], tubal tumors [6] and paratubal cysts [10]. However, in most cases, no predisposing factor was identified [11].

3.1. Clinical features

Most of the time patients describe an acute abdominal/pelvic pain, often localized on the same side of the torsion. Several non-specific symptoms can be associated: nausea, vomiting, pain on urination, uterine pain, digestive problems, spotting [10]. The diagnosis is often not suspected clinically due to the range of conditions that can mimic it, such as appendicitis, adnexal torsion, and pelvic inflammatory disease. Patients with early stage have normal blood tests.

In our first three cases, the initial diagnosis was erroneous, because the patients did not have ovarian torsion, complicated ovarian cysts, or nephritic colic, respectively. The unusual diagnosis of IFTT, lack of awareness among clinicians, and inappropriate early clinical orientation lead to delayed and, thus, irreversible damage.

3.2. Imaging

The US diagnosis of isolated fallopian tube torsion is challenging. The commonest finding on pelvic ultrasound is a dilated fallopian tube secondary to impaired venous and lymphatic drainage, with thickened walls and intraluminal debris [12]. An enlarged tubular structure between the uterus and the ovary concomitant with normal-appearing ovaries is the most described sign in US imaging. It was cited in 4 of 5 cases of our case series. Endovaginal ultrasound provides a higher resolution but it is not feasible in young and sex-

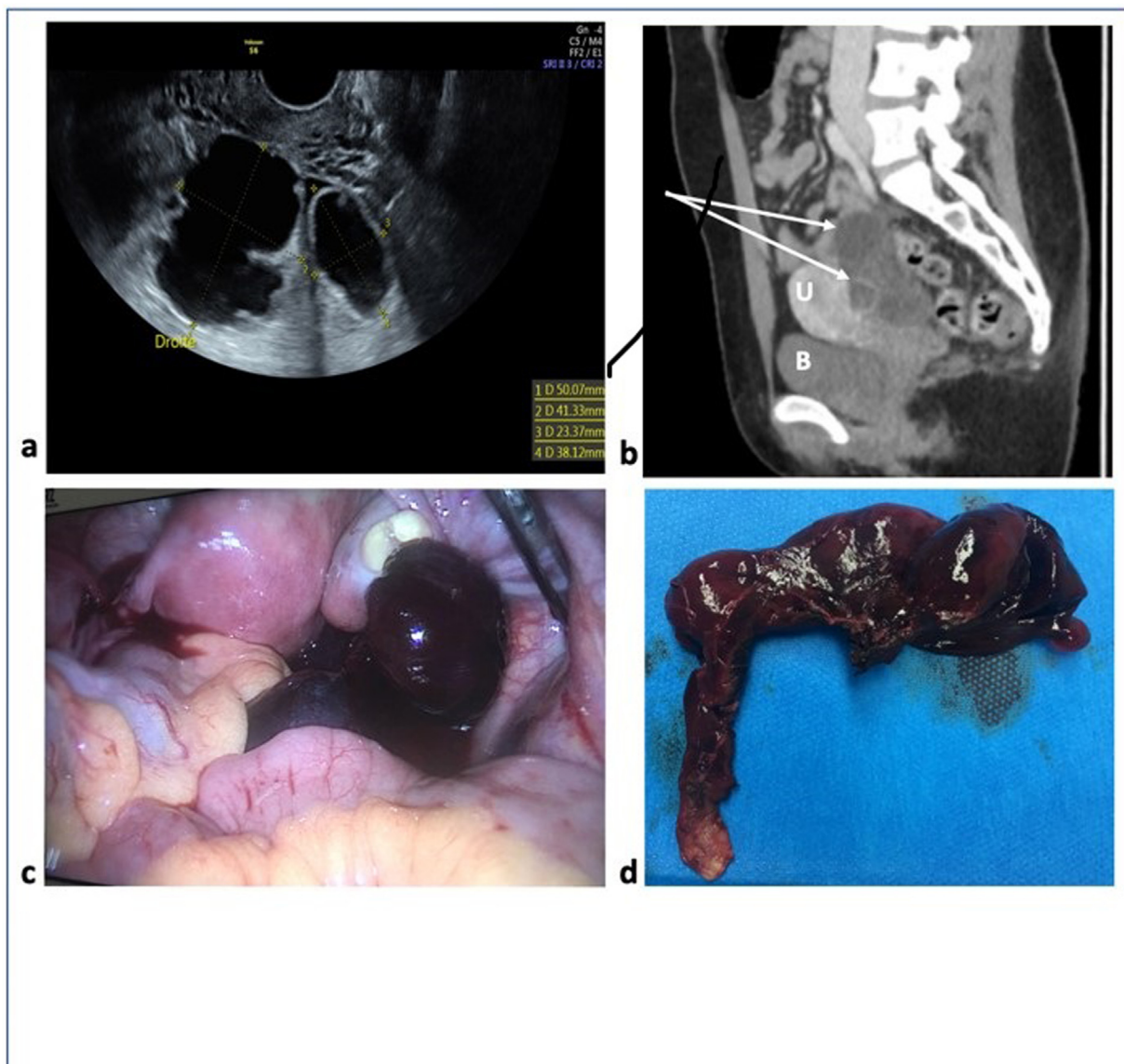


Fig. 4. a. Transvaginal ultrasound showed an aperistaltic echo-free tubular image with incomplete septations. b. CT-scan showing retro uterine lesion with extension to the retro-uterine pouch. c. Laparoscopic view of isolated twisted tube with hematosalpinx. d. Surgical specimen.

usually inactive women. In addition, a moderate volume of free fluid in the pelvis is often objectified.

Although US-Doppler is first recommended in women suffering from non-traumatic acute abdominal pain, the reported sensitivity for the diagnosis is reported to be low [13].

Among 27 surgically verified cases, Oded Raban et al. reported 29.6% of preoperative US diagnosis. In the remainder, US signs were often attributed to adnexal torsion [14]. The presence of a paraovarian cyst concomitant with normal-appearing ovaries was a warning sign for the diagnosis [11]. CT-scans helps to overcome these problems in diagnosis provided to be read by a trained team. MRI is often not readily available in the emergency context. The presence of a mass between the uterus and the ovary on CT-scan is a reliable characteristic associated with adnexal torsion in women presenting with abdominal pain and an adnexal mass. This sign is both sensitive (Se = 97%, 95% CI = 84–100%) and specific (Sp = 81%, 95% CI = 64–93%) [15].

3.3. Management

The literature describes different management techniques. Salpingectomy is the most frequently performed procedure (90%). Based on a histological study, the resected tubes showed the persistence of ciliated cells associated with signs of moderate ischemic infarction in half of patient [16]. Recently, as is commonly advocated for ovarian salvage in adnexal torsions, tubal conservation seems to be increasingly applied [4]. It should be performed, when possible, in woman of childbearing age and in a pediatric population. Bertozzi et al. reported 33% of conservative management in a Pediatric Population [4]. This attitude can expose to the risk of early recurrence necessitating repeat surgery. In order to reduce this risk, several authors have reported surgical technique associated with detorsion, such as puncture-drainage or cystectomy for associated bilateral cysts [3], ipsilateral salpingopexy [3], associated with paratubal cysts. According to the viability of the fal-

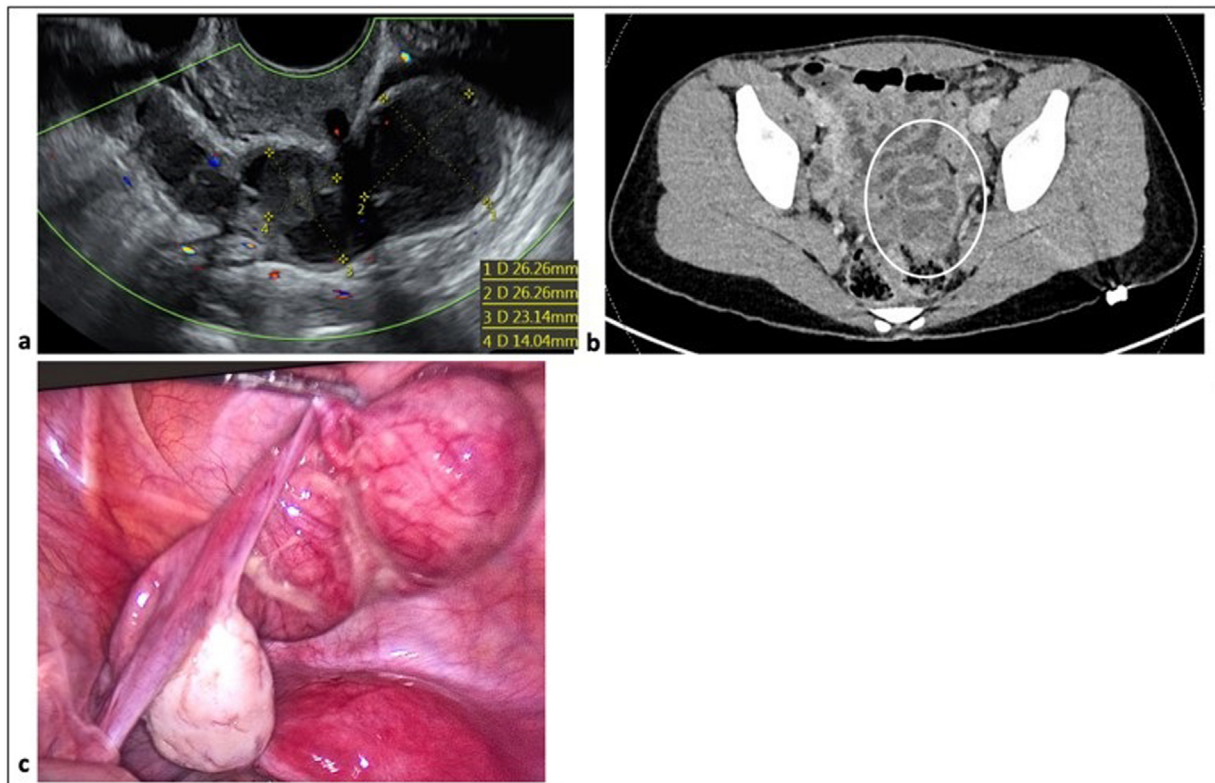


Fig. 5. Pyosalpinx, late complication of conservative traitement of Isolated torsion of fallopian tube. Transvaginal ultrasound (a) and CT scan (b) findings, with corresponding laparoscopic appearance (c).

lopian tube, salpingectomy or neosalpingostomy was performed [17]. Questions remain about the effects of these surgical techniques on female fertility and risk of recurrence. Unfortunately, authors did not collect long-term data [18].

However, the most common symptoms closely resemble those of adnexal torsion, and these two conditions are managed in the same way. Rate of successful conservative treatment decrease with increasing delay in the management [10]. As is commonly advocated for ovarian salvage in adnexal torsions [9,19] and considering negative impact of radical treatment on the future reproductive potential of young patients, conservative treatment must be considered particularly in child-bearing age and in pediatric population even in case of the twisted ischaemic fallopian tube [20]. Among conservative treatments, standardized treatment must be defined. Long-term fertility should be studied in greater detail, to orient the choice of optimal surgical management. Long-term fertility outcomes must be further assessed to orient these decisions.

4. Conclusion

IFTT is a rare but serious event which can occurs at different times in reproductive life stages. To further improve its clinical outcomes, growing awareness of this serious complication is advocated. Torsion should be evoked in front acute pelvic pain in patients with hydrosalpinx or paratubal cyst. Conservative management must be privileged especially in woman of childbearing age and in pediatric population. Only long-term follow-up with wide population can define a standardized treatment.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

The study was in accordance with the establishment's ethical charter of the CHOG.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

C. Delacroix: the main conceptual ideas, collect cases and wrote the majority of the manuscript.

E. Vintejou, S. Kedous, G. Carles: contributed to the analysis of the results and to the writing of the manuscript.

Dr N. Hcini : contributed to the analysis of the results and built the final version of the manuscript.

Registration of research studies

Not applicable.

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