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# Laparoscopic tubal salvage in an adolescent girl with bilateral isolated tubal torsion

N. Berlanda<sup>1</sup>, S. Bianchi<sup>2</sup>, C. Ferrero Caroggio<sup>1</sup>, N. Ciappina<sup>1</sup>, A. Bulfoni<sup>3</sup>, L. Fedele<sup>1</sup>

<sup>1</sup>Unità Operativa Dipartimentale Ginecologia Chirurgica e Endometriosi, Fondazione Istituto di Ricovero e Cura a Carattere Scientifico Ca' Granda Ospedale Maggiore Policlinico, Università degli Studi di Milano, Milan

<sup>2</sup>Dipartimento di Ostetricia e Ginecologia, Ospedale San Giuseppe, Università degli Studi di Milano, Milan

<sup>3</sup>Unità Operativa di Ostetricia e Ginecologia, Casa di Cura Humanitas-San Pio X, Milan (Italy)

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## Summary

*Purpose of investigation:* To report surgical treatment of a rare case of bilateral isolated fallopian tube torsion (IFTT). IFTT, especially when occurring bilaterally in adolescents, poses the dilemma of whether removing the salpinges, preventing the possibility of spontaneous conception, or restoring tubal patency, risking IFTT recurrence or a future ectopic pregnancy. *Case Report:* A 13-year-old, sexually inactive, presented after being discharged with the diagnosis of bilateral tubal ectasia from another hospital, where she received antibiotic treatment. At laparoscopy, bilateral IFTT was diagnosed. One tube was necrotic and a salpingectomy was performed. The contralateral tube presented a hydrosalpinx, but, once derotated and drained, appeared viable and salpingoneostomy and salpingopexy were performed. Six-month follow up was uneventful. *Conclusion:* in cases with an uncertain diagnosis, especially in young women in which tubal preservation is particularly important, laparoscopy allows an early diagnosis of tubal torsion and a conservative treatment before irreversible tubal necrosis occur.

*Key words:* Isolated tubal torsion; Laparoscopic salpingoneostomy; Pediatric laparoscopy.

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## Introduction

Isolated fallopian tube torsion (IFTT), without a torsion of the homolateral ovary, is a rare condition with an estimated prevalence of one in 1.5 million women [1]. The typical presenting symptom of IFTT is the acute onset of persistent or intermittent abdominal pain. Imaging techniques such as ultrasonography and MRI may reveal findings suggesting IFTT. However, due to its rarity and the aspecific information from clinical and imaging findings, the diagnosis of IFTT is difficult and therefore surgical treatment may be delayed, increasing the risk of irreversible tubal damage.

When dealing surgically with IFTT, especially in adolescent girls, the dilemma is whether to definitively treat the condition by removing the affected salpinx and possibly reducing the fertility potential of the young woman, or restore tubal patency and possibly increasing the risk of IFTT recurrence or of a future ectopic pregnancy. This dilemma is obviously emphasized in the exceedingly rare case in which IFTT occurs bilaterally. The authors present the first case of isolated torsion of the fallopian tubes, occurring bilaterally and simultaneously in an adolescent girl, in which the potential for spontaneous conception was preserved by means of laparoscopic monolateral salpingoneostomy and salpingopexy.

## Case Report

A 13-year-old caucasian girl went to her local hospital's emergency room because of the acute onset of pelvic pain, predominantly in the right iliac fossa, with irradiation to the right leg and associated with vomiting. Although she is a professional jockey, she denied any precipitating events. Her medical history was significant for the correction of a cardiac inter-atrial defect at the age of 12. She had menarche at the age of 12 and she reports irregular menses, in the form of oligomenorrhea and hypomenorrhea, during the last six months. She reported recurrent leucorrea during the last year. She denied previous sexual intercourses. She was admitted overnight, her symptoms resolved without intervention, and she was discharged the following day. Six days later, she presented again at the same hospital, for the acute onset of abdominal pain not associated with vomiting. After gynecologic evaluation and abdomino-pelvic ultrasonography, she was admitted in the Department of Surgery for suspected ovarian torsion. Upon admission, the physical examination was unremarkable, with absence of abdominal tenderness, rebound, or a palpable mass. The remarkable MRI findings were: absence of signs of ovarian torsion. Irregular bilateral dilatation of the tubal lumen and coiling salpinges. The left fallopian tube had a tortuous course with an endoluminal liquid-liquid level and a hematic peripheral rim, constituting a pseudo-mass compressing the posterior aspect of the uterus. Therapy consisted of intravenous penicillin. Vaginal swab was negative for infections. Because complete blood count and serum chemistry analyses were normal, the dimensions of the fallopian tubes were not modified at pelvic ultrasonography, and the patient did well during her hospital stay; she was discharged

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after five days with the diagnosis of bilateral tubal ectasia. A follow up visit was scheduled. Nine days later, this young woman came to this Institution for a second opinion. She was in good general condition and did not present abdominal pain. In the authors' Department, transabdominal ultrasonography confirmed bilateral tubal pathology. The right salpinx showed a 62×34-mm dilatation with anechoic content, consistent with hydrosalpinx. The left salpinx showed a 36×35-mm dilatation with homogeneous hypoechoic content, suggesting a hematosalpinx. The uterus and the ovaries were morphologically normal, both ovaries had regular arterial and venous flow, and no free fluid was present in the pouch of Douglas. The authors thoroughly counselled the young woman and her parents about the diagnosis of bilateral tubal dilatation of unclear origin and, after obtaining informed consent for a bilateral salpingectomy, they performed a laparoscopy for diagnosis and possible treatment of her condition.

Diagnostic laparoscopy confirmed a bilateral isolated fallopian tube torsion, with normal ovaries and uterus. The left fallopian tube was twisted twice at the level of its middle-third. The distal two-thirds of the salpinx were transformed in a 4-cm dark, apparently necrotic-hemorrhagic nodule adherent to the left pelvic sidewall. Tubal fimbriae, macroscopically normal, were visible at the end of the fallopian tube (Figure 1). On the right side, the fallopian tube was twisted four times, it was not patent because of the conglutination of the fimbriae, and its distal end was dilated. An accessory right tube was observed. (Figure 2). A conservative approach was attempted, but on the left side, after the healthy portion was separated from the necrotic portion of the fallopian tube, the tubal fimbriae were deemed not suitable for an end-to-end anastomosis and the proximal end of the fallopian tube was deemed inadequate for a salpingoneostomy. Therefore, a salpingectomy was performed on the left side. Histological evaluation confirmed the diagnosis of a left fallopian tube showing a necrotic-hemorrhagic segment. On the right side, after the salpinx was untwisted, a salpingoneostomy was performed because of the absence of necrosis, with a complementary salpingopexy in order to minimize the risk of recurrent tubal torsion. The accessory right tube was removed.

The post-operative course was uneventful and the patient was discharged after 48 hours. At a six-month follow up, she had no episodes of abdominal pain and no sonographic signs of right hydrosalpinx.

## Discussion

The authors report the first case of successful laparoscopic conservative treatment of bilateral isolated fallopian tube torsion in an adolescent girl. In this young woman, one fallopian tube already underwent necrosis and required salpingectomy. However, the contralateral tube, although being diagnosed as a hydrosalpinx at least two weeks prior, appeared viable and could be managed conservatively by means of salpingoneostomy and salpingopexy. The anatomical success of the procedure could be assessed at a six-month follow up, when there was no recurrence of hydrosalpinx and no recurrence of pain.

In cases of IFTT, the issue whether to perform a salpingectomy or a salpingoneostomy, is controversial. On one hand, salpingoneostomy might preserve the potential of spontaneous conception. On the other hand, this conservative procedure might result in suboptimal recovery of the

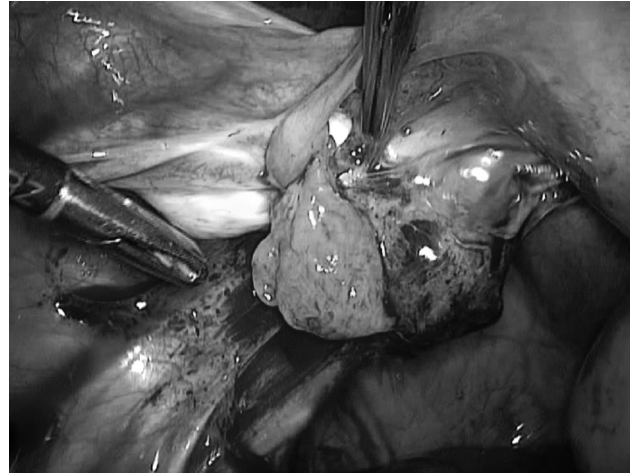


Figure 1. — Twisted and necrotic left fallopian tube.

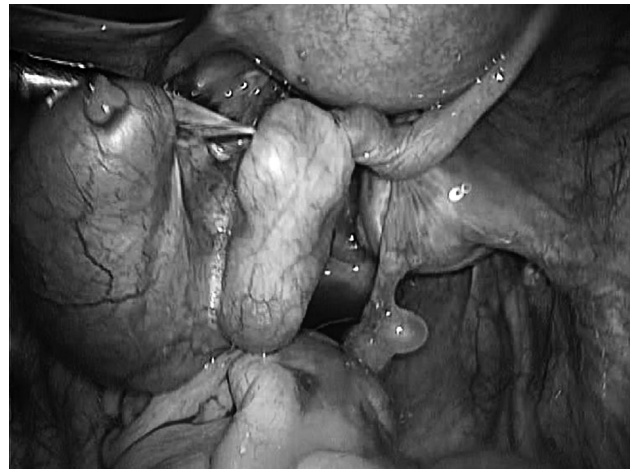


Figure 2. — Twisted right hydrosalpinx. An accessory right tube is noted.

tube, causing a future recurrence or an ectopic pregnancy. Similarly, a complementary salpingopexy could prevent torsion recurrence, but could also potentially result in additional surgical trauma on the fallopian tube.

Unfortunately, data on which to base the clinical decision of performing a demolitive versus a conservative treatment in cases of IFTT are limited. Among previously published paper, the present authors found only four cases of bilateral tubal torsion [2-5]. Three of them occurred in non-adolescent women, all of whom underwent bilateral salpingectomy [2-4]. The fourth case occurred in a premenarcheal 13-year-old girl who, similarly to the current patient, presented with a severely damaged fallopian tube, requiring salpingectomy, and a contralateral apparently vi-

able hydrosalpinx. A conservative approach was attempted by leaving the hydrosalpinx untreated, but acute pelvic pain recurred after five months and a second laparoscopy showed a twisted sactosalpinx, which, once derotated, maintained a necrotic appearance, requiring salpingectomy [5].

A previous review of the literature of monolateral IFTT associated with hydrosalpinx in children and adolescents reported that salpingectomy was performed immediately in eight out of 12 cases (67%) and in three out of four recurrences after a primary conservative treatment by means of detorsion and puncture. In the fourth patient, the recurrence was successfully treated by means of salpingoneostomy and follow-up at 26 months has been uneventful [6]. The authors observed a mean recurrence period of 68 days and they proposed a conservative surgical management of IFTT associated with hydrosalpinx in two steps. The first step was detorsion, puncture, and evacuation of the hydrosalpinx. The second step, approximately three weeks later, was salpingoscopy and salpingectomy or salpingoneostomy, depending on the tubal state. This protocol would allow performing cytologic and bacteriologic analysis of the fluid, ruling out tubal malignancy and infection. However, in the present authors' opinion, performing an immediate salpingoneostomy could prevent the worsening of tubal vascular supply and tubal edema that may occur during the period between the first and the second step. Moreover, malignancy is an exceedingly rare cause of hydrosalpinx and tubal infections are unfrequent in the mostly sexually inactive adolescent population and, furthermore, tubal fluid can be collect also when a salpingoneostomy is planned, allowing a successive timely salpingectomy in case of positive results.

The two largest series of IFTT in pediatric and adolescent females report a similar rate of underlying tubal pathology of 53% [6] and 44% [7], respectively. However, the rate of conservative procedures varied from 47% among 15 patients in the first study [7] to 12% among 45 patients in the second study [8] This is consistent with the fact that no guidelines are available and treatment is eventually dependent upon visual judgement and experience of the surgeon.

The possibility for tubal conservation is suggested by the encouraging pregnancy rates observed in adults treated by laparoscopic salpingoneostomy for tubal infertility and hydrosalpinx [9]. In a pediatric population, sexually inactive and without a history of PID, pregnancy rates after salpingoneostomy could be potentially higher as compared to the adult population. Accordingly, in one study, 50% of the resected tubes showed ciliated cells at histological evaluation [6]. However, prospective, randomized trials comparing salpingoneostomy with salpingectomy followed by in vitro fertilization would be necessary to disentangle this issue.

The analysis of the present patient's medical history did not highlight any clear etiologic cause for the occurrence of

IFTT. This adolescent girl did not have risk factors for pelvic infection or adhesions such a medical history suggestive of PID, appendicitis, or endometriosis, she was sexually inactive, and denied any previous abdominal surgery. Since the young woman is a professional jockey, a suggestive speculation is that the frequent up and down body movements that occur when riding horses have contributed to the tubal torsion. Accordingly, in a previous series of five adolescent women aged 12-16, a close association between IFTT and sports causing rapid body movements was found [10].

There are multiple factors that render diagnosis and treatment difficult in cases of IFTT. Firstly, although a presumptive diagnosis can be made, pathognomonic symptoms and imaging diagnostic findings are not available. Secondly, even when adnexal pathology is suspected, the diagnosis of IFTT is difficult due to its extreme rarity, and the condition might be mistaken for a tubal phlogosis, which is more frequent. Furthermore, if ovarian torsion has been ruled out by means of visualization of regular blood supply to the ovary, the severity of the clinical picture might be downgraded and a surgical procedure may be deemed not necessary. Watchful waiting may appear to be the best management for a very young girl that is overall in good clinical conditions.

## Conclusion

The present authors believe that laparoscopy should be considered a fundamental diagnostic tool when imaging techniques are inconclusive. In cases with an uncertain diagnosis, especially in young women in which tubal preservation is particularly important, the threshold for a diagnostic laparoscopy should be low. Laparoscopy is the only way to achieve an early diagnosis of tubal torsion, allowing a conservative treatment before irreversible tubal necrosis occur.

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Corresponding Author:

N. BERLANDA, M.D.

Unità Operativa Dipartimentale Ginecologia Chirurgica  
e Endometriosi, Fondazione IRCCS Ca' Granda  
Ospedale Maggiore Policlinico

Via della Commenda 12

20122 Milan (Italy)

e-mail: nicola.berlanda@gmail.com