Isolated Tubal Torsion: A Rare Cause of Acute Pelvic/Abdominal Pain among Adolescent Females

Mohamad K. Ramadan1,2*, Khouloud Demachkie1,3, Amani Mohsen1, Loubna Sinno4, Janoub Kaza’al1,2
1Department of Obstetrics and Gynecology, Rafik Hariri University Hospital, 2Department of Obstetrics and Gynecology, Lebanese University, 3Faculty of Medicine, Beirut Arab University, 4Research Unit, Makassed General Hospital, Beirut, Lebanon

Abstract

Tubal torsion usually occurs as a part of adnexal torsion that affects an ovary and the adjacent tube; however, isolated tubal torsion is an extremely rare condition. Usually, it presents as acute pelvic/abdominal pain but could also exhibit milder intermittent pain alternating with periods of relief (subacute). This condition has seldom been diagnosed preoperative and commonly results in tubal damage due to delayed management. We hereby, report the findings of two cases managed recently at our center. In both cases, the diagnosis was delayed 2–3 days and was only made intraoperative when the tubes could not be salvaged due to extensive necrosis. The extent of tubal damage is predominantly dependent on the duration of vascular insult; hence, the urgency for affecting early diagnosis and intervention to restore blood supply and preserve tubal integrity and function. We advocate the liberal and early use of laparoscopy in patients presenting with subacute unexplained pelvic/abdominal pain.

Keywords: Isolated tubal torsion, laparoscopy, subacute pelvic/abdominal pain

INTRODUCTION

Isolated fallopian tube torsion is an exceptionally rare condition, with a reported incidence of 1 in 1.5 million women.[1] The majority of cases present with acute pelvic pain,[1] but might as well present initially with less intense pain alternating with periods of relief to worsen gradually along a course of few days-weeks.[1] This latter group with a longer duration of pain was associated with a higher rate of oophorectomy/salpingectomy.[2] This condition lacks specific clinical presentation and constant diagnostic imaging features rendering its diagnosis exceptionally difficult, and is characterized of having predilection toward perimenarchial females. Most of the cases are not diagnosed preoperative or misdiagnosed for other pathologies.[3] When discovered at surgery, the tube is invariably necrotic and cannot be salvaged.[4] Hereby, we report two cases of teenage girls who presented with acute lower quadrant pain to our emergency department (ED) after seeking medical advice in two different neighboring hospitals 2–3 days earlier. Initial management was executed by the ED team. Computed tomography (CT) scan and ultrasonography were performed but were not helpful in the diagnosis of isolated tubal torsion (ITT). One case was misdiagnosed as ovarian torsion, while the second was suspected for acute appendicitis. In both instances, we were called to the operative theater only after the pediatric surgeons had discovered unexpected ITT. The tubes were immensely necrotic and could not be salvaged.

Case Reports

Case 1

This was a 12-year-old, previously healthy premenarcheal girl. She presented to the ED complaining of right lower quadrant pain to our emergency department (ED) after seeking medical advice in two different neighboring hospitals 2–3 days earlier. Initial management was executed by the ED team. Computed tomography (CT) scan and ultrasonography were performed but were not helpful in the diagnosis of isolated tubal torsion (ITT). One case was misdiagnosed as ovarian torsion, while the second was suspected for acute appendicitis. In both instances, we were called to the operative theater only after the pediatric surgeons had discovered unexpected ITT. The tubes were immensely necrotic and could not be salvaged.
Case 1

A 30-year-old woman presented to the emergency department with a 3-day history of right lower quadrant pain that had recently increased in intensity. She also reported vomiting and decreased oral intake. On examination, McBurney’s sign was equivocal with voluntary guarding, rebound tenderness, and mild rigidity. Preliminary laboratories disclosed leukocytosis of 16,100, neutrophils of 77%, and a C-reactive protein (CRP) of 17.8. Bedside transabdominal ultrasound revealed a normal-sized uterus, with multilocular adnexal complex of mixed echogenic components. Doppler studies showed positive flow to the ovary but was absent in the center of this complex adnexal mass. CT scan of the abdomen and pelvis showed the absence of appendicitis or other abdominal pathologies, with minimal fluid in the pelvis and enlarged ovary (7 cm × 5.5 cm) with few cysts suggestive of ovarian torsion [Figure 1]. Exploratory laparotomy was performed by the pediatric surgeon. Right tubal torsion was found possibly due to an adjacent 6 cm paraovarian cyst. Complete necrosis of the tube was noted with the normal healthy ipsilateral ovary. The fallopian tube and the paraovarian cyst were removed [Figure 2]. The patient recovered rapidly and was discharged home 2 days later.

Case 2

A 14-year-old, previously healthy girl presented to ED with 2 days onset of periumbilical pain radiating to the right lower quadrant and associated with undocumented fever and vomiting. McBurney’s sign and obturator sign were positive. Laboratories revealed leukocytosis of 14,800 with 70% neutrophils and a CRP of 51. Bedside transabdominal ultrasound disclosed multicystic collection possibly originating from the right adnexa. CT scan of the abdomen and pelvis showed twisted tubular fluid-filled structure in the right iliac fossa possibly continuous with the cecum and demonstrating an enhancing wall with adjacent fat stranding and fluid, suggestive of acutely inflamed appendicitis and less likely hydrosalpinx/pyosalpinx [Figure 3]. During laparotomy, a twisted, ischemic distal part of the right fallopian tube was seen. This was resected due to extensive necrosis. Both the appendix and the ipsilateral ovary looked normal and healthy. Of interest, the tube was judged to be extraordinarily long of about 12 cm. Pathology image showed tubal congestion with tissue damage and engorged blood vessels [Figure 4].

Discussion

These two cases exemplify an unusual clinical scenario where care providers tend to be reluctant to perform surgical interventions due to the subacute nature of pain, whereas, in severe and constant pain, the management is usually taken more serious with earlier surgical intervention.
Ramadan, et al.: Isolated tubal torsion

Unfortunately, these young patients are subjected to long periods of observation and hesitation to perform any surgery, pending deterioration in their clinical status. Both cases were subjected to delay of 2–3 days in the diagnosis and management. The majority of cases with adnexal/tubal torsion develop acute abdomen, while some cases present with subacute intermittent pain, which is difficult to diagnose.

Figure 4: Pathology image showing tubal congestion with tissue damage and engorged blood vessels for case 2

Tubal torsion is very rare and is estimated to affect 1 in 1,500,000 women.[4] The usual clinical presentation is similar to that of adnexal/ovarian torsion. Nonetheless, both have nonspecific signs and symptoms that can mimic each other together with other various etiologies of acute pelvic/abdominal pain. Invariably, certain associated pathology is found, while the adjacent ovary is mostly normal.[4] There are the number of case reports of this condition affecting young adolescents even before having menses.[1-5] Risk factors would include intrinsic tubal (e.g., cysts of Morgagni, hydro/pyosalpinx, congenital anomaly, or excessively long tubes) or extrinsic pathology such as ovarian mass, infection, pelvic congestion, pregnancy, or adhesions.[3] Furthermore, imaging has not been reported to be of major assistance in the early diagnosis because of the lack of constant features specific to this entity.[3] The absence of features characteristic of ovarian torsion should raise the suspicion of ITT, especially in young female patients. The most common reported sonographic findings were: (i) A dilated tubular structure with a tapering end (peak sign) adjacent to a normal ipsilateral ovary was present in 50% of cases. (ii) Cystic masses (paratubal cysts) separate from an adjacent normal ovary. (iii) A midline cystic mass (either in the posterior cul-de-sac or superior to the uterus) associated with a normal ipsilateral ovary was the most consistent finding in both pelvic ultrasound and CT. (iv) some cases were associated with ovarian pathology as hemorrhagic cyst or ovarian teratoma.[1]

Nevertheless, in a few cases, the torsed tube appeared-like an enlarged multicystic-appearing ovary, a picture that mimics ovarian torsion.[1] Most of these tubes were filled with sonolucent contents, a picture similar to hydro/pyosalpinx. Although Doppler has been found to improve the sensitivity of ultrasound for the diagnosis by some authors,[4] yet, similar to ovarian torsion, the use of the color Doppler is of limited benefit. None of these tools can confirm the diagnosis of tubal torsion.[6] Similar to ovarian torsion, direct visualization remains the most reliable method for diagnosis. In the majority of cases, torsion occurred in the midsegment of the fallopian tube, but it may, as well, occur around the ligamentous supports of the tube.[7] If the rotation is partial or intermittent; the venous and lymphatic congestion may subside quickly, along with related symptoms. If twisting is complete and prolonged, venous and arterial thrombosis ensues, eventually resulting in tubal infarction and necrosis. It was previously observed that torsion preferentially affects the right side due to the presence of the sigmoid colon on the left side and slow venous flow on the right side. This pattern was refuted in two recent reports.[1-5] Hence, torsion can affect either side equally. Acute pelvic/abdominal pain is approached differently by health-care providers. ED physicians tend to resort to imaging with CT scan, while gynecologists prefer ultrasonography as the primary imaging modality. CT scan is the standard imaging procedure for all cases with an acute abdomen seen by ED specialists. Nonetheless, ultrasonography is equally sensitive in depicting most gynecologic pathologies and is cheap, readily available, and does not need special preparations. ITT lacks specific features, rendering all imaging modalities of equally limited usefulness in diagnosis. Furthermore, most of these features are not constantly present in every case. Detorsion and preservation of the affected tube remain the objective of the sound management. Spinelli et al. suggested a practical guide for the surgical management of these cases.[8] The key factor remains early diagnosis and timely intervention. Even a delay of only 12 h was found to have devastating effects on future fertility.[4] Detorsion of the adnexa helps to preserve the function and maintain fertility. Removal of the tube should not be attempted depending merely on its color, but when it is found to be friable, necrotic and bleeds to touch, thence, removal might be unavoidable. Fluorescein injection was sometimes used to confirm the viability of the tissue before taking any further steps in the surgery.[9] We fully concur with the suggestion that the mere suspicion of any adnexal torsion should instigate gynecologists to perform diagnostic laparoscopy to have an early and definitive diagnosis.[10] Laparoscopy should become the gold standard for the diagnosis and management of this elusive, yet potentially damaging condition. Its use can benefit patients by avoiding unnecessary surgery or delay in diagnosis and treatment. Added merits include shortening of operative and hospital stay and offer minimally invasive intervention where detorsion can be done with ease, and it is
known also to diminish future adhesions and provide better fertility potentials. Not all cases with adnexal or tubal torsion develop acute surgical abdomen, and actually, in many cases, the episode of pain alternates with periods of relief, possibly due to temporary spontaneous detorsion. These patients, in particular, are usually subjected to periods of observation and denied early intervention, which might be detrimental to the vascular supply of the tissues. In fact, little is known about the natural history of tubal torsion. On suspicion of the condition, we encourage the avoidance of observation pending worsening of pain and advocate the liberal and early use of laparoscopy for diagnosis and possible intervention.

Acknowledgment
We are grateful to the radiologist, Dr. Ahmad Lakkis, for his contribution and help in providing and interpreting the CT images.

Ethical approval
IRB approval (approval letter obtained on 30th September 2019) for release of information and conditional consent of the guardians was acquired provided personal information was kept anonymous.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardians have given their consent for the images and other clinical information to be reported in the journal. The guardians understand that names and initials will not be published, and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES
1. Narayanan S, Bandarkar A, Bulas DI. Fallopian tube torsion in the pediatric age group: Radiologic evaluation. J Ultrasound Med 2014;33:1697-704.
2. Rossi BV, Ference EH, Zurakowski D, Scholz S, Feins NR, Chow JS, et al. The clinical presentation and surgical management of adnexal torsion in the pediatric and adolescent population. J Pediatr Adolesc Gynecol 2012;25:109-13.
3. Comerci G, Colombo FM, Stefanetti M, Grazia G. Isolated fallopian tube torsion: A rare but important event for women of reproductive age. Fertil Steril 2008;90:1198.e23-5.
4. Lau HY, Huang LW, Chan CC, Lin CL, Chen CP. Isolated torsion of the fallopian tube in a 14-year-old adolescent. Taiwan J Obstet Gynecol 2006;45:363-5.
5. Mueller C, Tomita S. Fallopian Tube Torsion as a Cause of Acute Pelvic Pain in Adolescent Females. Case Rep Pediatr 2016;2016:8707386.
6. Casey RK, Damle LF, Gomez-Lobo V. Isolated fallopian tube torsion in pediatric and adolescent females: A retrospective review of 15 cases at a single institution. J Pediatr Adolesc Gynecol 2013;26:189-92.
7. Li PC, Chen BC, Yeh BH, Kao SP, Ding DC. Hydrosalpinx with Adnexa Torsion Treated with a Salpingostomy in a Virgin Woman. Gynecol Minim Invasive Ther 2018;7:136-8.
8. Spinelli C, Piscioneri J, Strambi S. Adnexal torsion in adolescents: Update and review of the literature. Curr Opin Obstet Gynecol 2015;27:320-5.
9. Righi RV, McComb PF, Fluker MR. Laparoscopic oophoropexy for recurrent adnexal torsion. Hum Reprod 1995;10:3136-8.
10. Ben-Arie A, Lurie S, Graf G, Insler V. Adnexal torsion in adolescents: Prompt diagnosis and treatment may save the adnexa. Eur J Obstet Gynecol Reprod Biol 1995;63:169-73.