Case Report Eurasian Journal of Critical Care

A Rare Cause of Pediatric Acute Abdomen: Isolated Tubal Torsion

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Introduction

Isolated tubal torsion (ITT) is an extremely rare cause of acute abdomen frequently observed in women of reproductive age. Its diagnosis may become difficult due to its nonspecific clinical signs and symptoms including abdominal pain, nausea, vomiting, and fever. Its prevalence ranges between 1:500.000 and 1:1.500.000. It was first described by Bland-Sutton in 1890. The first pediatric case in the literature was described by Hansen in 1922; since then, a total of seventy-seven pediatric ITT cases have been described until 2019. Radiological imaging methods provide information supportive of the diagnosis of ITT ¹⁻⁵. In this paper, we aimed to report the clinical features and ultrasonographic (US) and magnetic resonance imaging (MRI) findings of a pediatric patient with ITT that was radiologically diagnosed and confirmed by surgery.

Case

A 10-year-old girl presented to emergency service with abdominal pain that had lasted for one week. She stated that the pain was cramp-like and particularly involved in the right lower quadrant. On abdominal examination, she had diffuse tenderness, rebound tenderness, and guarding at the lower abdomen, which was particularly prominent at the right lower quadrant. She had a stable general condition and was conscious, and she had no nausea, vomiting, loss of appetite, or fever. Her biochemical parameters, urinalysis, and coagulation parameters were within normal limits.

She underwent an abdominal US and MRI to detect the underlying pathology. On the transabdominal US, there was a regular contoured, thick-walled, cystic formation with a tubular appearance, internal echoes, and fluid-hemorrhage levels; it was located at the pelvic level at the right side and compressed the urinary bladder from its anterior aspect (Figure-1). Both ovaries were visualized clearly. A contrast-enhanced MRI was taken for lesion characterization and detection of coexisting findings. The MRI examination showed a dilated, thick-walled, tubular, cystic pelvic formation with contents suggestive of hydro-hematosalpinx. Additionally, there was the whirlpool sign representing vascular torsion at the right adnexal area (Figure-2 A, B, C). There was also fluid in Douglas pouch. Considered to have an adnexal torsion based on the available findings, the patient was taken to laparoscopic exploration, which showed that the uterus, both ovaries, and the left salpinx had a normal appearance but the ampullary region of the right salpinx was twisted around its axis. Based on the operative findings, the case was considered an ITT (Figure-2). The patient developed no complication during the postoperative period and was ultimately discharged with follow-up instructions. She had no residual sign or symptom at her follow-up appointment one month later.

Discussion

Adnexal torsion is most commonly observed with salpinxes and ovaries twisting around the broad ligament. However, twisting of a salpinx alone around the mesosalpinx creating an adnexal torsion is an extremely rare event in childhood. Its etiology may include congenital anomalies of the salpinxes, a long mesosalpinx, hydrosalpinx, hematosalpinx, and tubal mass lesions. Although ITT constitutes a rare cause of lower abdominal pain among women of reproductive age, it may also be observed in premenarchal girls and postmenopausal women. More rarely, ITT may also complicate pregnancy. Although ITT has no specific symptoms, lower abdominal pain is a common finding. There may also be nausea, vomiting, and signs of peritoneal irritation and an adnexal mass. ITT is commonly seen at the right side as a result of partially restricted motion of the left salpinx due to its close anatomical relationship with the sigmoid colon²⁻⁵.

Three types of torsion patterns based on operative findings have been described in the literature ⁶.



Figure -1. An ultrasonographic image depicts a thick-walled, dilated, cystic right fallopian tube (arrows).



Figure -2. (A) An axial T2-weighted MR image shows a dilated, thick-walled, tubular cystic formation that contains hemorrhagic content in its lumen and takes up contrast material (white arrow). Both ovaries appear normal (dashed white arrows). (B) A coronal T2-weighted MR image shows the whirlpool sign of the right fallopian tube (white arrow). (C) An axial post-contrast fat suppression T1-weighted MR image shows diffuse contrast uptake on the walls of the fallopian tube. (D) A photograph of the surgical specimen depicts a blackish-colored, edematous, ischemic, necrotic fallopian tube with torsion.

- Type 1, Long axis torsion with full-length dilation of the fallopian tube
- Type 2, Short axis torsion with full-length dilation of the fallopian tube,
- Type 3, long-axis torsion with distal dilation of a fallopian tube or a para-tubal cyst.

Based on the above classification, our patient had a type 3 pattern.

Although the US should be the first-line imaging modality by virtue of its rapid, readily available, and noninvasive nature, it, unfortunately, has a low specificity. Among the ultrasonographic signs of tubal torsion are tubal thickening, haemato-/hydrosalpinx, and an adnexal mass. Doppler US may prove useful in suspected cases. It may demonstrate a high impedance coupled with an absent or reversed diastolic vascular flow. However, one should also keep in mind that the presence of a normal flow pattern does not necessarily exclude the possibility of torsion. MRI is a useful technique with a better tissue contrast and the ability to detect the underlying disorder. MRI allows the detection and characterization of an underlying mass and its organ of origin. The whirlpool sign of adnexal torsion is characteristic for its diagnosis. The whirlpool sign was also confirmed by MRI in our case. Other cross-sectional imaging findings of ITT include dilated tubal structures wider than 15 mm, thickened and contrast-enhanced tubal wall, free pelvic fluid, thickened broad ligament, contamination of peri-tubal fat, and localized ileus ^{4, 6, 7}. As there are no specific radiological, clinical, and biochemical signs despite all of the above-mentioned signs, the preoperative diagnosis of ITT may be difficult. Thus, surgical exploration is considered the gold standard for its diagnosis and treatment. Early diagnosis of ITT is important. A study showed that a time window from the onset of pain to surgical exploration longer than 10 hours increased the risk of tubal necrosis in patients with adnexal torsion ^{1, 5, 8, 9}. Hence, we opted for surgical exploration since the time from the onset of our patient's symptoms to her hospital admission was one week. In ITT, the risk of interrupted blood supply to the ovaries is low since they receive blood supply from both the ovarian and uterine arteries.

The differential diagnosis of ITT should include acute appendicitis, ovarian cyst rupture or torsion, ectopic pregnancy, pelvic inflammatory disease, cystitis, endometriosis, leiomyoma degeneration, intestinal obstruction or perforation, diverticulitis, and renal colic ^{3, 6, 7, 8, 10}.

Conclusion

In conclusion, despite rare, ITT should be considered among the causes of acute abdomen in women of reproductive age and adolescents. ITT's nonspecific signs and laboratory findings may easily lead to a wrong diagnosis. Radiological imaging should be carried out to evaluate any meaningful finding of tubal torsion in suspected cases. Making a correct diagnosis of ITT and implementing its treatment early in its course is of paramount importance for the preservation of the fertility potential of affected patients.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial suppo

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