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Case Report

A rare cause of acute abdominal pain in children: Isolated tubal torsion; a case series

Yasemin Dere Gunal ^{a,*}, Gokhan Berktug Bahadir ^b, Ozlem Boybeyi ^c, Aylin Pelin Cıl ^d, Mustafa Kemal Aslan ^a

^a Kırıkkale University, Department of Pediatric Surgery, Kırıkkale, Turkey

^b Mersin University, Department of Pediatric Surgery, Mersin, Turkey

^c Hacettepe University, Department of Pediatric Surgery, Ankara, Turkey

^d Kırıkkale University, Department of Obstetrics and Gynecology, Kırıkkale, Turkey

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ABSTRACT

Isolated tubal torsion -a rare cause of acute abdomen in children-is usually difficult to diagnose because of non-specific findings. Surgical salpingectomy is required in delayed diagnosis in most cases. Three sexual inactive adolescents diagnosed in isolated tubal torsion (ITT) were discussed for its diagnostic features and surgical management. Laboratory tests and radiological studies including ultrasonography (US), color doppler ultrasound were performed in all patients after evaluation for acute lower abdominal pain in emergency department and they underwent surgical intervention with laparotomy (n:2) and laparoscopy (n:1). One of the patients in this study had salpingectomy. Detorsion of the fallopian tube and cyst excision were performed in the remaining two patients who also had paratubal cysts. There was no recurrence in these patients during the follow-up for 3 and 2 years. The isolated tubal torsion should be kept in mind and early surgical management is essential in order to preserve fallopian tube because of its importance in fertility.

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1. Introduction

Adnexal torsion is one of the most important causes of acute abdominal pain and requires prompt surgical treatment.¹ Although adnexal torsion including ovary with or without fallopian tube is a common condition, isolated torsion of fallopian tube without an ovarian torsion is extremely rare, with an incidence of 1 in 1.5 million women.¹ The incidence is considerably uncommon in the pediatric and adolescent population.^{1,2}

Since the clinical and imaging findings for ITT are non-specific, the diagnosis of ITT can be challenging.^{1–3} In most cases, surgical salpingectomy is the choice of treatment because of delayed diagnosis. Nevertheless, there are very few cases managed with detorsion of the fallopian tube.^{1,2,4,5}

Herein, we presented three cases of ITT patients referred to tertiary level pediatric surgery center due to paratubal cyst in order to discuss its diagnostic features and surgical management.

2. Case presentations

2.1. Case 1

A 12-year-old sexually inactive postmenarchal female patient was admitted to the emergency department with left lower abdominal pain lasting for 3 days and was referred to pediatric surgery department with an initial diagnosis of acute abdomen. She was admitted to another hospital with a diagnosis of functional ovarian cyst and suggested to be followed-up one year ago. There was no history of trauma and her family history was unremarkable.

The physical examination revealed tenderness and defense in the left lower quadrant. Laboratory tests were within normal limits except elevated white blood cell count (WBC) (12,300/ μ l). The left ovary was invisible and there was a left adnexal complicated mass measuring 70 × 35 × 30 mm in diameter composed of a

* Corresponding author. Department of Pediatric Surgery, Kırıkkale University Faculty of Medicine, Kırıkkale, 71450, Turkey.

E-mail address: drderegunal@yahoo.com (Y.D. Gunal).

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hemorrhagic cyst with internal echoes and simple cystic structures on ultrasound. Doppler ultrasound revealed absence of blood flow on the left adnexa. The patient underwent an emergent exploration.

At laparotomy; uterus, right adnexa and left ovary were normal, but left fallopian tube was twisted twice around itself at clockwise direction. There was a hemorrhagic paratubal cyst 5 × 5 cm in diameter located just near to fimbria. The twisted tube was edematous with purple patchy areas suggesting early signs of ischemia (Fig. 1). The tube was untwisted and the perfusion returned back after waiting several minutes with warm wet gauzes (Fig. 2). The tubal opening was checked with a stilette, and the paratubal cyst was excised totally. The postoperative course was uneventful. Two months later, ultrasound showed normal findings. Histopathological examination revealed simple cyst lined by single layer of tubal type epithelium with hemorrhages. There were no signs of tubal or ovarian pathology during the follow-up with ultrasound for three years.

2.2. Case 2

A 12-year old premenarche girl was admitted to the emergency department with complaints of abdominal pain, nausea and vomiting for three days and was referred to pediatric surgery department. Her physical evaluation revealed tenderness and defense in the right lower quadrant. She was found to have leukocytosis (20,500/ μ l). The sudden onset of intense pain and microscopic hematuria was detected. There was no significant sign in her ultrasonographic examination. Emergent laparotomy was planned since her complaints continued and her physical examination signs were significant. In her laparotomy, there were diffuse hemorrhagic fluid and proximal right fallopian tube was torsioned four times clockwise. Right fallopian tube was gangrenous. Right ovary, uterus and left adnexa were normal. Ischemic right fallopian tube was detorsioned. After 20 minutes of hot isotonic solution application,

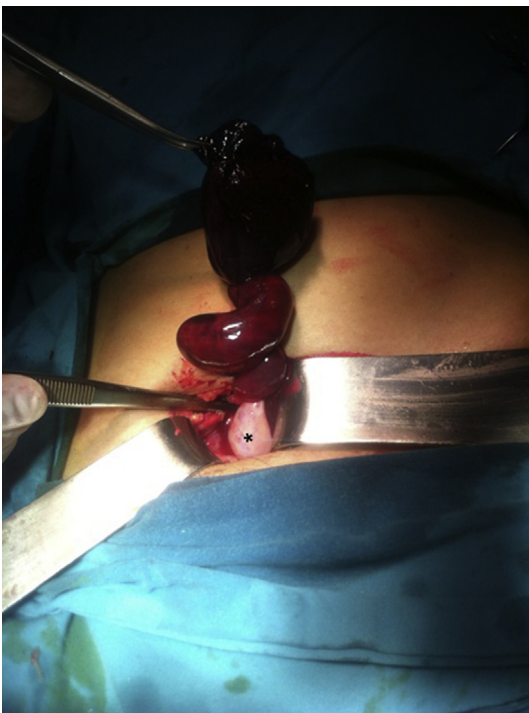


Fig. 1. Twisted fallopian tube with the hemorrhagic sessile paratubal cyst and normal ovary*. (case 1).



Fig. 2. Fallopian tube after detorsion and cystectomy. (case 1).

fallopian tube was still gangrenous. Ovary-preserving right salpingectomy was performed. Histopathological evaluation revealed necrotic fallopian tube. There was not any postoperative complication. There were no signs of ovarian pathology during the follow-up with ultrasound for four years.

2.3. Case 3

A 13-year old premenarche girl was admitted to the emergency department with a right lower quadrant pain lasting for 4 hours and was referred to pediatric surgery department. Physical examination revealed left and right lower quadrant pain both and guarding. Laboratory investigations were within normal limits. She had a history of follow-up due to a right ovarian cyst by another institute during last one year. There was a cyst with a diameter of 5.5 cm thought to be arising from right adnexia in pelvic ultrasound and the ovarian blood supply was normal. An emergent laparoscopy was performed due to acute abdomen with a diagnosis of suspected tubal torsion. The right fallopian tube was twisted around itself and the right ovary was normal (Fig. 3). The right fallopian tube was detorsioned and a paratubal cyst was seen and then the cyst was excised totally. The color of the tube was quite normal after detorsion. Although the left ovary was normal there was also a torsioned and pedunculated cyst (Morgagni Hydatide) in the left ovary and it was also excised. Postoperative course was uneventful and histopathologic examination revealed mesonefric paratubal cyst. There were no signs of tubal or ovarian pathology during the follow-up with ultrasound for two years.

3. Discussion

The torsion of fallopian tube without an associating ovarian torsion is called isolated tubal torsion (ITT) and is an uncommon cause of acute abdominal pain in adolescent girls.^{1–3} Although, ITT usually occurs spontaneously, it can also take place due to underlying adnexal pathology such as abnormally long tube/meso-salpinx, premenarchal hormonal activity leading to adnexal congestion and tubal motility, paraovarian/paratubal cysts.^{1–3} Two of three patients in this study were premenarche and the remaining was postmenarche. Although, a paratubal cyst is associated with ITT in two patients, hormonal activity leading to congestion and tubal motility might also be considered as a causative factor.

The diagnosis of ITT is difficult because of nonspecific clinical, laboratory and radiological findings.^{1–3} The presenting symptom is

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