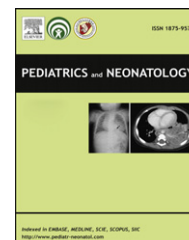


Available online at www.sciencedirect.com

SciVerse ScienceDirect

journal homepage: <http://www.pediatr-neonol.com>

ORIGINAL ARTICLE

Pediatric Ovarian Torsion in a Medical Center in Taiwan: Case Analysis

Jun-Kai Kao ^{a,*}, Chun-Chien Chiu ^b, Po-Yu Wang ^a, Meng-Kung Yu ^a^a Department of Pediatrics, Children's Hospital, Changhua Christian Hospital, Taiwan^b Department of Emergency Medicine, Show Chwan Memorial Hospital, Changhua, Taiwan

Received Mar 22, 2011; received in revised form May 30, 2011; accepted Jun 16, 2011

Key Wordsabdominal pain;
children;
ovarian torsion

Background: Abdominal pain is one of the most common complaints made by patients visiting emergency departments; however, ovarian torsion is an uncommon cause of abdominal pain, especially in young children. Early diagnosis is essential in order to salvage the ovaries. We performed a retrospective analysis of patients under 18 years of age who visited Changhua Christian Hospital with adnexal torsion between June 2003 and June 2010.

Methods: Medical records were reviewed for age, associated symptoms, past and present medical histories, physical findings, diagnostic tests performed, clinical course, pathological findings, and diagnoses.

Results: A total of 21 patients were identified and included in the analysis; their mean (SD) age was 13.62 (3.75) years. Abdominal pain was the universal symptom. Vomiting or nausea was the second most common symptom (33.3%). The average period between symptom onset and diagnosis was 3.29 (6.39) days. Right-side adnexal torsion occurred in 14 patients, and left-side torsion in seven patients. Ovarian cysts, most often dermoid cysts (e.g., teratomas), were found in 16 patients (76%). Except for one conservatively treated case and one malignant case (adult granulosa cell tumor), the other results of the pathological examination were as follows: eight dermoid cysts, two cystadenomas, one follicular cyst, and eight simple cysts.

Conclusion: Although it is uncommon, adnexal torsion should be included in the differential diagnosis of lower abdominal pain in all girls, regardless of age. Sonography is helpful for making such a diagnosis. Time is a critical factor, and early laparoscopy or laparotomy may save a viable ovary.

Copyright © 2012, Taiwan Pediatric Association. Published by Elsevier Taiwan LLC. All rights reserved.

* Corresponding author. Children's Hospital, Changhua Christian Hospital, 135 Nanhsiao Street, Changhua, Taiwan 500.
E-mail address: 96777@cch.org.tw (J.-K. Kao).

1. Introduction

Abdominal pain is one of the most common complaints of patients visiting emergency departments.^{1–3} Approximately one-third of children who present with abdominal pain at an emergency department do not receive a specific diagnosis.³ Diagnosis of abdominal pain in children can be difficult given the high prevalence of gastrointestinal disorders, especially in young children who may not be able to describe their symptoms particularly well or localize their discomfort. Failing to diagnose an emerging surgical condition, such as acute appendicitis, is the primary concern of physicians when treating patients with abdominal discomfort. However, ovarian torsion is an uncommon cause of abdominal pain, especially in young children, and early diagnosis is essential in order to salvage the affected ovaries. In this study, we analyzed the epidemiological characteristics of patients below 18 years of age with adnexal torsion at Changhua Christian Hospital (CCH). We retrospectively examined all patients with adnexal torsion who visited the pediatric emergency department or outpatient clinic of CCH between June 2003 and June 2010. We also reviewed relevant studies in the literature.

2. Materials and Methods

We reviewed the medical charts of all patients who treated between June 2003 and June 2010 who were diagnosed with adnexal torsion or ovarian torsion (International Classification of Diseases 620.5). Medical records were reviewed for age, associated symptoms, past and present medical histories, physical findings, diagnostic tests performed, clinical course, pathological findings, and diagnoses. Leukocytosis was defined as >10,000 white blood cells (WBCs)/mL in a peripheral blood sample, and pyuria was defined as >5 WBCs per high-power field (HPF) of a urine sample. The study was approved by the institutional review board of CCH.

3. Results

A total of 21 patients below 18 years of age with adnexal torsion were identified over the 7-year period and included in the analysis. Of these children, three had visited the outpatient department and the remaining had visited the pediatric emergency department of our hospital. All patients were eventually admitted to our hospital. Their mean (SD) age was 13.62 (3.75) years (Table 1). Four patients were below 10 years of age, and the youngest was 7 years old. Eight children had previously visited other private clinics or were transferred from other local hospitals. The incidence rate of adnexal torsion in the pediatric emergency room of CCH was one case per 10,000 visiting patients.

Abdominal pain presented in each child. The lower abdomen was the most common area where children described the location of their pain (Table 2). Among these children, one child mentioned radiating flank pain but did not have a urinary tract infection. Vomiting and nausea was the second most common symptom (33.3%). Two children presented with diarrhea, one had difficulty in urinating, and one presented with a fever. A palpable mass was detected by physical examination in one child, and one

Table 1 Epidemiological characteristics of patients with adnexal torsion.

	Ovarian teratoma							Ovarian simple cyst, serous cystadenoma	Ovarian simple cyst, follicular cystadenoma			Tubal serous cystadenoma, paratubal cyst	Adult granulose cell tumor
	7	9	11	13	18	18	18		14	16	17		
Age (y)	7	9	11	13	18	18	18	14	16	17	18	17	18
Counts	1	2	2	1	2	2	1	1	1	2	1	1	1
Location							5	5	3	2	1	3	1
							3	3	0	3	1	1	0
Surgical Technique							0	0	0	3	1	1	0
							2	1	2	1	1	0	1
							2	1	2	1	0	0	0
							2	3	2	3	0	0	0
							1 (cystectomy 3 mon later)	1	1	1	0	0	0
							Partial oophorectomy	2	1	2	0	0	0
Comorbidity							Hydrosalpinx (1) Corpus luteum hematoma (1)	2	1	2	0	0	0
							Ovarian hemorrhagic necrosis (1)	2	1	2	0	0	0

Table 2 Clinical manifestations of abdominal pain.

		Number of cases
Location	Lower abdomen	10
	Right lower abdomen	5
	Left lower abdomen	0
	Could not be localized	6
Types of pain	Progress from mild to severe	1
	Sharp pain	3
	Mild and intermittent	4
	No particular description	13

child presented with signs of peritonitis. The period between symptom onset and diagnosis varied from 4 hours to 30 days. On average, children were diagnosed 3.29 (6.39) days after the onset of symptoms.

In terms of medical history, one child had a known ovarian cyst (measuring 4.2 × 3.5 × 3.0 cm) on the side of torsion for 2 years. Two children had experienced the same symptoms for 3 months before this study, but the symptoms were resolved through conservative treatment at that time. One child had a history of intussusception. None had ever been pregnant.

Nine (45%) children demonstrated leukocytosis with neutrophils as the predominant cell. The average WBC (SD) count among all patients was 11.12 (3.65) × 10³ cells/μL. A mildly elevated C-reactive protein (CRP) level was noted, with an average of 2.45 (3.06) mg/dL and large variation across all patients (normal to 10.9 mg/dL). Urine analysis showed that one child had hematuria and three patients had pyuria, but none of these four patients had a positive urine culture. Tumor markers and hormone levels, including cancer antigen 125(CA125), α-fetoprotein (AFP), carcinoembryonic antigen (CEA), estradiol, testosterone, progesterone, β-human chorionic gonadotropin (β-HCG), and follicle-stimulating hormone (FSH), were tested in some patients, but the results were normal.

All patients underwent ultrasonography, and four patients underwent computed tomography (CT). The diameters of the identified cysts were between 3.4–14.3 cm: 7.77 (3.16) × 6.15 (2.25) (Figure 1).

One child received conservative treatment during hospitalization and never underwent an operation; the others received laparoscopic operations. Right-side adnexal torsion occurred in 14 patients, and left-side adnexal torsion occurred in seven. Ovarian cysts were found in 16 patients and parafallopian cysts were found in four. One child had a pseudocyst due to a hemorrhagic corpus luteum hematoma that was found in addition to an ovarian cyst during surgery. One 17-year-old patient had a solid ovarian tumor mass that was diagnosed as an adult granulosa cell tumor.

Eight patients underwent an oophorectomy (including five patients who underwent a salpingo-oophorectomy), and three underwent a partial oophorectomy. Six patients underwent an ovarian cystectomy and three underwent a salpingectomy (Table 1).

A Kaplan-Meier survival curve was constructed and analyzed to investigate the relationship between time of

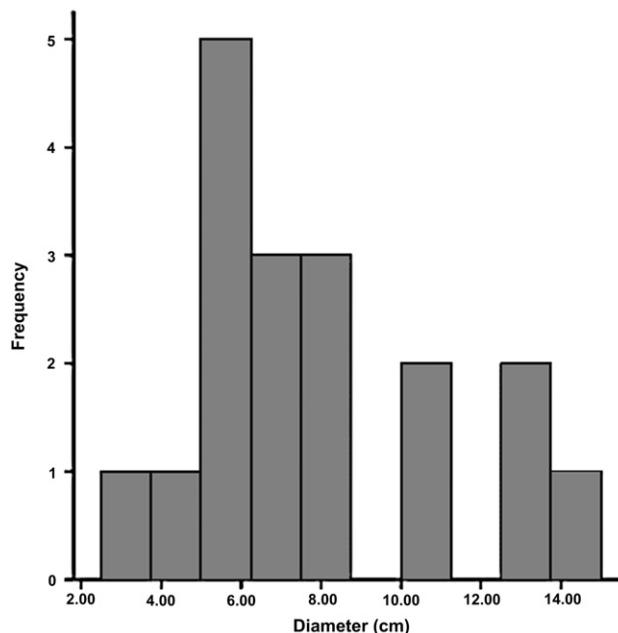


Figure 1 Size distribution of ovarian cysts in patients with torsion.

symptom onset, clinical presentation, and surgery. The median ovary-saving time was 3 days, which means that at the 3rd day half of the patients needed to receive an oophorectomy. If the surgery was performed within 1 day, 75% of patients would have their ovary saved, as shown by our results (Figure 2).

Except for one conservatively treated case and one malignant case, the other results of the pathological

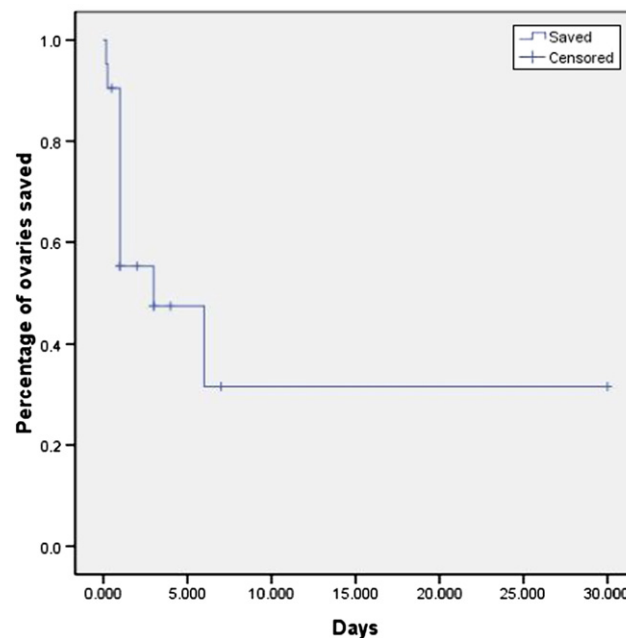


Figure 2 Kaplan-Meier analysis of the relationship between time from symptom onset until surgery and percentage of ovaries that were saved.

examination were as follows: eight children had dermoid cysts (teratomas), two had serious cystadenomas, one had a follicular cyst, and eight had simple cysts. Ovarian hemorrhagic necrosis was found in seven children, and hemorrhagic changes on micropathological examination were observed in another five patients.

The average length of hospital stay was 4.05 (1.95) d. One patient underwent an additional surgery 7 months later to treat a contralateral ovarian cyst and pelvic adhesion. One child who had initially undergone an ovarian cystectomy underwent another surgery 6 years later to treat a bilateral ovarian teratoma. One patient was later diagnosed with Graves' disease and received follow-up examinations at the outpatient department. The patient with the adult granulosa cell tumor was treated with chemotherapy and received regular follow-up examinations at our hospital.

4. Discussion

Gynecological disorders are not often considered in the differential diagnosis of prepubertal girls because of their infrequent occurrence. Acute ovarian torsion is uncommon in children and timely diagnosis is difficult. Adnexal torsion has been reported to occur from the antenatal period to as late as 53 years of age.⁴ Torsion of the right ovary was more common in this study, similar to the findings of other studies.^{1,3} A study of torsion in 20 girls below 14 years of age in Israel found that 30% had normal adnexa and 40% had diseased ovaries (mostly cysts).⁵ Bilateral adnexal torsion may occur simultaneously, or the unaffected side may become torsed years later, resulting in sterility.^{6,7} Although two of our patients (10%) had undergone other surgeries for the treatment of ovarian cysts on the contralateral side, no patient in our study presented with simultaneous bilateral torsions.

The onset of symptoms has been reported to range from 6 hours to 3 weeks before admission.⁵ In this study, the longest duration was 30 days. Abrupt onset or chronic abdominal pain due to occlusion of the vascular supply to the twisted ovary is the only common symptom.^{1,2,8} Physical findings and pain characteristics are highly variable. In contrast to the results presented by Houry and Abbott,⁹ which showed that 51% of torsion patients have radiating pain in the flank, back, or groin, only one patient in our analysis complained of flank pain. As in other studies, vomiting and nausea were the most commonly associated symptoms in our study. Two patients in our study experienced repeated symptoms in the months leading up to the operation, which suggests recurrent torsion and detorsion. Although previous studies have shown that the location of abdominal pain, as described by the patient, always corresponds to the side of torsion,¹⁰ two patients in our study reported pain on the side opposite to that of the torsion.

Torsion is more likely to occur in a diseased ovary.^{11,12} Younger children more commonly present with a mature cystic teratoma or without an underlying abnormality compared with older children, in whom torsion is more likely to result from a follicular or corpus luteum cyst.¹³ Although the occurrence rate of bilateral mature cystic

teratomas is approximately 10%, contralateral biopsy remains controversial.^{13,14} Solid ovarian masses in children are much more likely to be malignant. In this situation, preoperative tumor markers, such as AFP and β -HCG, may help postoperative management. According to a report by Warner et al, cysts greater than 5 cm in diameter rarely cause ovarian torsion,¹⁵ however only three patients in our study had cysts less than 5 cm in diameter.

An abnormal blood count may indicate mild leukocytosis. However, this finding, as well as the results of other routine laboratory studies, is too nonspecific to be helpful for the diagnosis of ovarian torsion. Ultrasonography is the imaging method of choice for evaluating and managing suspected ovarian torsion.^{5,16,17} The most common sonographic finding is an echogenic/hypoechoic pelvic mass with nonvisualization of the ipsilateral ovary because occlusion of venous and lymphatic flow often results in ovarian edema.^{5,6} Fifteen of our patients (75%) had a hypoechoic cystic mass and five (25%) had an echogenic or solid mass noted on sonography. Fluid was visualized in the cul-de-sac of one patient, and ovary hematomas were found in two other cases. In addition, if malignancy is suspected on ultrasonography, a CT scan may be helpful for assessing the spread of the tumor.

A laparotomy can also be used to make a definitive diagnosis. If the vascular supply to the ovary is thrombosed, the organ should be excised without detorsion in order to decrease the risk of pulmonary embolus.² The current findings and results of previous studies suggest that children usually present for organ salvage very late (more than 8 hours) after the onset of symptoms.^{1,2,5,18,19} Thus, treatment for acute ovarian torsion generally includes unilateral salpingo-oophorectomy. Our results indicate that 50% of patients with ovarian torsion can still have their ovary saved if they receive an operation within 3 days of symptom onset.

As a retrospective study, this study is limited because there are some inaccurate or missing data. There are also some unknowable factors that might have affected the surgeons' decisions regarding whether or not to remove the involved ovaries. For example, surgeons who believe that the ovaries have been torsed for a long time, and, thus, unable to be saved, would presumably be more likely to remove those ovaries.

5. Conclusion

Although uncommon, adnexal torsion should be included in the differential diagnosis of lower abdominal pain in any girl, regardless of age. Sonography is helpful when making such a diagnosis. Time is a critical factor, and early laparoscopy or laparotomy may be able to save a viable ovary.

References

1. Lee CH, Raman S, Sivanesaratnam V. Torsion of ovarian tumors: a clinicopathological study. *Int J Gynecol Obstet* 1989;28: 21–5.
2. Nichols DH, Julian PJ. Torsion of the adnexa. *Clin Obstet Gynecol* 1985;28:375–80.

3. Ward M, Frazier T. Torsion of normal uterine adnexa in childhood: case report. *Pediatrics* 1978;**61**:573–4.
4. Buss JG. Sequential torsion of the uterine adnexa. *Mayo Clin Proc* 1987;**62**:623–5.
5. Mordehai J, Mares AJ, Barki Y, et al. Torsion of uterine adnexa in neonates and children: a report of 20 cases. *J Pediatr Surg* 1991;**26**:1195–9.
6. Davis AJ, Feins NR. Subsequent asynchronous torsion of normal adnexa in children. *J Pediatr Surg* 1990;**25**:687–9.
7. Bower RJ, Adkins JC. Surgical ovarian lesions in children. *Am Surg* 1981;**47**:474–8.
8. Hibbard LT. Adnexal torsion. *Am J Obstet Gynecol* 1985;**152**:456–61.
9. Houry D, Abbott JT. Ovarian torsion: a fifteen-year-review. *Ann Emerg Med* 2001;**38**:156–9.
10. Meyer JS, Harmon CM, Harty MP, Markowitz RI, Hubbard AM, Bellah RD. Ovarian torsion: clinical and imaging presentation in children. *J Pediatr Surg* 1995;**30**:1433–6.
11. Brown MF, Hebra A, McGeehin K, Ross III AJ. Ovarian masses in children: a review of 91 cases of malignant and benign masses. *J Pediatr Surg* 1993;**28**:930–2.
12. Helvie MA, Silver TM. Ovarian torsion: sonographic evaluation. *J Clin Ultrasound* 1989;**17**:327–32.
13. Kokoska ER, Keller MS, Weber TR. Acute ovarian torsion in children. *Am J Surg* 2000;**180**:462–5.
14. Comerchi Jr JT, Licciardi F, Bergh PA, et al. Mature cystic teratoma: a clinicopathologic evaluation of 517 cases and review of the literature. *Obstet Gynecol* 1994;**84**:22–8.
15. Warner BW, Kuhn JC, Barr LL. Conservative management of large ovarian cysts in children: the value of serial pelvic ultrasonography. *Surgery* 1992;**112**:749–55.
16. Ledesma-Medina J, Towbin RB, Newman B. Pediatric case of the day. *Radiographics* 1992;**12**:199–200.
17. Graif M, Itzchak Y. Sonographic evaluation of ovarian torsion in childhood and adolescence. *Am J Roentgenol* 1988;**150**:647–9.
18. Shust NM, Hendricksen DK. Ovarian torsion: an unusual cause of abdominal pain in a young girl. *Am J Emerg Med* 1995;**13**:307–9.
19. Meyer JS, Harmon CM, Harty MP, et al. Ovarian torsion: clinical and imaging presentation in children. *J Pediatr Surg* 1995;**30**:1433–6.