Pediatric ovarian torsion: case series and review of the literature

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Background: Ovarian torsion in children is an uncommon cause of acute abdominal pain but mandates early surgical management to prevent further adnexal damage. The clinical presentation mimics other pathologies, such as appendicitis. We sought to more completely characterize ovarian torsion with respect to pain and ancillary studies, such as urinalysis.

Methods: We performed a retrospective review of hospital charts of all patients aged 0–18 years with a diagnosis of ovarian torsion at the Children's Hospital at London Health Sciences Centre, in London, Ont., from 1993 to 2008.

Results: We analyzed 13 charts of patients aged 7 months to 18 years. Most patients presented with peripheral leukocytosis, vomiting and right lower quadrant pain that did not radiate or migrate. On urinalysis, about half the patients demonstrated pyuria without bacteruria. Pelvic ultrasound revealed an ovarian cyst on the same side of the pain in 11 of 13 patients. Most were found to have a hemorrhagic cyst or ovary and underwent salpingo-oophorectomy or cystectomy within 48 hours of presentation.

Conclusion: Ovarian torsion should be considered in any female child with acute onset lower abdominal pain accompanied by vomiting. Pain can be characterized as constant or colicky, but unlike with appendicitis, does not typically migrate. Sterile pyuria is found in a substantial proportion of cases. Ultrasound is the most useful initial diagnostic modality, but the absence of flow on Doppler imaging is not always present. Conservative management with detorsion and oophoropexy is recommended.

Contexte : La torsion ovarienne est une cause rare de douleur abdominale aiguë chez l'enfant, qui commande un traitement chirurgical immédiat pour protéger l'ovaire touché. Le tableau clinique ressemble souvent à celui d'autres pathologies, comme l'appendicite. Nous avons voulu caractériser de manière plus précise la torsion ovarienne sur le plan de la douleur et des examens complémentaires, tels que l'analyse d'urine.

Méthodes : Nous avons effectué une revue rétrospective des dossiers hospitaliers de toutes les patientes âgées de 0 à 18 ans chez qui un diagnostic de torsion ovarienne avait été posé à l'Hôpital pour enfants du Centre des sciences de la santé de London, en Ontario, entre 1993 et 2008.

Résultats : Nous avons ainsi analysé 13 dossiers de patientes âgées de 7 mois à 18 ans. La plupart de ces patientes présentaient une leucocytose périphérique, des vomissements et une douleur au quadrant inférieur droit, sans irradiation ni migration. À l'analyse d'urine, environ la moitié des patientes présentaient une pyurie sans bactériurie. L'échographie pelvienne a révélé un kyste ovarien du côté douloureux chez 11 patientes sur 13. La plupart étaient porteuses d'un kyste hémorragique de l'ovaire et ont dû subir une salpingo-ovariectomie ou une kystectomie dans les 48 heures suivant leur arrivée.

Conclusion : Il faut envisager un cas de torsion ovarienne chez tout enfant de sexe féminin qui présente une douleur abdominale basse aiguë, accompagnée de vomissements. La douleur peut être constante ou sous forme de coliques, mais contrairement à l'appendicite, elle n'a pas tendance à migrer. La pyurie stérile s'observe dans une proportion substantielle de cas. L'échographie est la modalité diagnostique initiale la plus utile, mais l'absence de vascularisation n'est pas toujours observée à l'imagerie Doppler. On recommande un traitement conservateur qui consiste à détordre l'ovaire et à pratiquer une ovariopexie.

Pediatric ovarian torsion is an uncommon cause of acute abdominal pain in children. It is estimated to account for 3% of all cases of acute abdominal pain in adult women.[1] Most cases (71%) are seen in women

older than 20 years.[2] Ovarian torsion is an emergency that mandates early diagnosis and timely surgical exploration and detorsion to avoid the catastrophic consequence of further adnexal injury. However, because the signs and symptoms can mimic other acute abdominal conditions, the preoperative diagnosis often remains a challenge for primary care physicians.

The normal mobility of the fallopian tube can lead to rotation of the ovary along with its vasculature. This leads to the obstruction of venous outflow, infarction and eventual necrosis, infection, peritonitis or loss of the adnexa. This is especially dangerous in young children, as the entity can go unrecognized because of its rarity and the nonspecificity of its presentation. This can delay surgical intervention and lead to a greater risk of complications.

Most cases of torsion occur in ovaries containing masses, such as functional cysts and neoplasms. Torsion is reportedly more common on the right side (3:2). This is possibly due to either the sigmoid colon on the left limiting movement or a hypermobile cecum on the right that is more permissive to movement.[3]

To date, there are only a few large pediatric studies of ovarian torsion that have detailed the presentation, pathologic findings and management. The number of study participants has ranged from 12 to 97 patients, reflecting the relative rarity of this condition. Taken together, findings have shown that acute onset lower abdominal pain and vomiting are a consistent presentation of ovarian torsion. Ultrasound is the diagnostic modality of choice; however, the presence of vascular flow on Doppler imaging does not reliably exclude torsion.[4–8]

More work is needed, as no large pediatric studies have characterized the pain in detail, nor have they presented the results of ancillary tests, such as urinalysis. This information may prove helpful for primary care physicians in differentiating ovarian torsion from more frequent competing diagnoses, such as appendicitis. The diagnostic challenge of ovarian torsion is evidenced by a delay in diagnosis reported by several authors.[9,10] The objective of the present 15-year review of ovarian torsion in a large Canadian children's hospital was to more completely describe the nature of the pain in pediatric patients with ovarian torsion and detail findings on ancillary tests, such as urinalysis. We hoped this information would prove useful to front-line clinicians in identifying ovarian torsion as a consideration in the evaluation of a potentially surgical abdomen.

Methods

We undertook a retrospective review of hospital charts, including emergency department, operative, imaging and pathology reports, of all patients aged 0–18 years with a discharge or secondary diagnosis of ovarian torsion at the Children's Hospital, London Health Sciences Centre, London, Ont., from 1993 to 2008. The Children's Hospital is a postgraduate teaching site of the University of Western Ontario. There are about 37 000 annual pediatric emergency department visits. Patients seen initially at a peripheral centre were included in our chart review.

Patients with a preoperative diagnosis of torsion who did not have torsion confirmed operatively or sonographically were excluded. Patients with ovarian torsion diagnosed antenatally were excluded. Fever was defined as a temperature greater than 38°C. A white blood cell (WBC) count was deemed elevated if it was at least 10 $\times 10^9$/L (10 000 cells/mm^3).

A standardized chart review form with an anonymous patient identification number was used by the 2 authors (N.P. and C.P.) involved in data extraction. Missing data were not included in the analysis. The abstractors were not blinded to the patient's final diagnosis or to the study's purpose.

The information reviewed included the emergency resident or supervising physician's note; triage vital signs; surgical consultation admission note; laboratory and imaging results, as reported by the staff radiologist; surgical pathology; and operative notes. Data collected included demographic information; characteristics, chronology and duration of symptoms; gynecologic history; vital signs at presentation; pertinent physical examination findings; laboratory results, including complete blood count and differential, blood culture, pregnancy test, and urine culture; results of preoperative imaging, such as Doppler ultrasound or computed tomography; the duration of symptoms until definitive management; intraoperative findings; surgical management (e.g., resection or oophoropexy); and pathology reports.

Pain characteristics such as "sharp," "colicky" or "dull" were recorded as such if these words appeared in the chart. If not, a descriptor was not recorded. Other pain characteristics, such as lateralization, migration and associated symptoms, were recorded as coded responses.

Statistical analysis

We analyzed the data using univariate statistics with Microsoft Excel for Mac. The study was approved by the ethics review board of the University of Western Ontario.

Results

We identified 46 charts for review. Seven were for neonates with antenatal diagnoses and were therefore excluded. Twenty-six charts were excluded because ovarian torsion was either not confirmed operatively or sonographically or because an alternate diagnosis was evident on surgical exploration. No charts were excluded owing to illegibility. The charts of 13 patients remained for analysis.

The age range was 7 months to 18 years (mean 12 yr). Most patients were menarchal (5 of 13) or perimenarchal

(5 of 13), and most patients (11 of 13) presented with right lower quadrant pain that did not radiate (7 of 11) or migrate (9 of 11). All patients reported a pain duration of less than 48 hours before seeking medical attention. Nearly half of the patients (5 of 10) described the pain as colicky. Fever was mentioned in the history of 3 patients and identified at triage in 1 patient. Most patients (7 of 12) reported vomiting. Vital signs were stable with respect to blood pressure, heart rate and respiratory rate in all patients. Peritoneal findings were absent in all but 1 patient. None of the patients presented with vaginal bleeding, and 4 of 10 patients reported a loss of appetite.

In terms of hematologic parameters, 8 patients had a moderately elevated WBC count (10.3–17.6 $\times$ 10^9/L). All but 1 of these patients had a predominance of neutrophils. Hemoglobin and platelet counts were unremarkable in all patients. Results of dipstick urinalysis was variable. About half the patients were dipstick positive for leukocyte esterase (6 of 10), hemoglobin (6 of 10) and nitrites (7 of 10). However, none of the 5 patients for whom a urine culture was obtained demonstrated any bacterial growth.

The preoperative diagnosis was appendicitis in roughly half (7 of 12) of the patients. In 5 patients, the diagnosis of ovarian torsion was suspected preoperatively.

In 3 of 5 patients in whom an abdominal radiographic series was carried out, the radiologist interpretation revealed a radio-opaque soft tissue mass in the right lower quadrant. Computed tomography was performed in 2 patients, and an ovarian cyst was identified in both.

Pelvic ultrasound demonstrated an ovarian cyst ipsilateral to the pain in 11 of 13 patients. Doppler ultrasonography was carried out in 4 patients; 2 of them had venous flow, but only 1 of these 2 patients had arterial flow.

With regards to surgical management, most patients (11 of 12) underwent surgery within 48 hours of presentation. In 1 patient, the pathologist reported a right-sided enlarged ovary, which presumably had torsioned and then spontaneously detorsioned. Six of the 11 patients underwent ipsilateral salpingo-oophorectomy. Three patients underwent cystectomy. Only 2 patients underwent detorsion and oophoropexy. Three patients underwent detorsion only. The clinical records of only 2 patients who underwent detorsion were available for follow-up. The patient who underwent a detorsion and oophoropexy at age 2 years had normal ovarian volume and flow at 2-year follow-up. The other patient underwent a cystectomy and detorsion at age 12 and had a smaller affected ovary that contained follicles at follow-up. She went on to deliver a healthy infant several years later.

Pathologic findings were varied and available for 11 patients. Most (8 of 11) had a hemorrhagic cyst or ovary and underwent salpingo-oophorectomy or cystectomy. One patient who was discovered to have a hydrosalpinx secondary to pelvic inflammatory disease subsequently underwent detorsion and salpingostomy. In 1 patient, no abnormal pathology was found pertaining to the torsioned ovary. A necrotic ovary was found in 2 patients, both of whom underwent salpingo-oophorectomy. One of these patients was an infant in whom the diagnosis of ovarian torsion was suspected preoperatively. The infant had features suggestive of systemic inflammation with a fever and an elevated peripheral WBC count. The other patient was an 18-year-old who had symptoms for 96 hours and an elevated peripheral WBC count. The time to surgery was 9 and 24 hours, respectively. The presentation, diagnosis, pathologic findings and management for all 13 patients are presented in Tables 1 and 2.

Discussion

Acute ovarian torsion is a rare but important entity in children. In this retrospective chart review of pediatric patients with ovarian torsion in our centre, several similarities and key differences with previous studies are evident. A summary of the clinical presentation of ovarian torsion across pediatric studies can be found in Table 3.

First, the average age of our patient population was 12 years. Other studies of pediatric patients with ovarian torsion have found a mean age of presentation ranging 9 to 12.5 years.[4,5,9–13] The largest study of pediatric ovarian torsion reported a mean age of 9.2 years.[11] This may be explained by the preponderance of ovarian cysts among menarchal or perimenarchal girls. Second, almost all patients presented suddenly with right-sided lower abdominal pain of a duration less than 48 hours. Similarly, most patients reported vomiting.[4–7,9,12,13] Finally, fever was seen in a minority of patients.[4,5,7,11,13–15] However, in contrast to other pediatric reports,[4,5] a pelvic mass was not documented in any charts in our review.

Our chart review sought to further describe the presentation of patients with ovarian torsion, particularly in the

Table 1. Summary of presentation in 13 children with ovarian torsion

Patient	Age, yr*	Pain localization	Duration of pain, h	Vomiting	Fever
1	18	RLQ	12	No	No
2	18	RLQ	9	Yes	No
3	7 mo	RLQ	48	No	Yes
4	18	Lower back	120	No	No
5	2	RLQ	72	Yes	No
6	12	RLQ	12	Yes	No
7	18	RLQ	96	No	No
8	6	Periumbilical	16	Yes	No
9	11	RLQ	48	Yes	Yes
10	10	RLQ	24	No	No
11	13	RLQ	Unknown	Yes	Yes
12	14	RLQ	4	Unknown	No
13	14	RLQ	24	Yes	No

RLQ = right lower quadrant.
*Unless otherwise indicated.

characterization of pain. In most of the cases, the pain did not radiate or migrate. Half the patients described the pain as colicky, and the other half described it as constant. In contrast, Houry and Abbott[15] described radiation of pain in half of their predominantly adult patients with ovarian torsion. This may reflect more detailed reporting of pain by adults. As in previous studies, roughly half our patients received a preoperative diagnosis of appendicitis. Peritoneal signs were absent in all but 1 of our patients. This is consistent with a previous report in which only 3 of 87 women had peritoneal signs.[15] In contrast to appendicitis, in which perforation results in peritonitis, ruptured ovarian cysts tend to result in only transient peritoneal signs, which may or may not be manifest at the time of clinical presentation.[4]

Table 2. Summary of diagnostic modality, management and pathologic findings in 13 children with ovarian torsion

Patient	Age, yr*	Pathology	Diagnostic modality	Management	Time to surgery, h
1	18	Unknown	Doppler ultrasound	None; spontaneous detorsion	Unknown
2	18	Hemorrhagic cyst	Doppler ultrasound	Detorsion and oophoropexy; cystectomy	8
3	7 mo	Necrotic ovary	Ultrasound and abdominal radiograph	Salpingo-oophorectomy	9
4	18	Hemorrhagic ovary	Ultrasound	Salpingo-oophorectomy	13
5	2	Unknown	Doppler ultrasound	Detorsion and oophoropexy	26
6	12	Healthy ovary	Ultrasound and abdominal radiograph	Detorsion and cystectomy	8
7	18	Hemorrhagic and necrotic ovary	Ultrasound and computed tomography	Salpingo-oophorectomy	24
8	6	Hemorrhagic ovary	Doppler ultrasound	Salpingo-oophorectomy	9.5
9	11	Healthy ovary	Abdominal radiograph	Salpingo-oophorectomy	7
10	10	Hemorrhagic ovary	Ultrasound and abdominal radiograph	Salpingo-oophorectomy	72
11	13	Hemorrhagic cyst	Abdominal radiograph, computed tomography, and ultrasound	Detorsion alone	30
12	14	Hemorrhagic cyst	Ultrasound	Cystectomy	8
13	14	Hydrosalpinx	Ultrasound	Detorsion and salpingostomy	42

*Unless otherwise indicated.

Consistent with other studies,[4,6] we found a pleiocytosis with a preponderance of neutrophils in most patients. In the patient with the highest peripheral WBC count of 17.6×10^9/L (17 600 cells/mm^3), a necrotic ovary was found at laparoscopy. However, generalizations regarding pleiocytosis in ovarian torsion are difficult because of studies involving small samples and varied cutoff criteria. Although most patients have an elevated peripheral WBC count, a normal count does not mitigate against torsion; in fact, 5 of 13 patients in our study had a normal WBC count.

About half of our patients demonstrated findings on urinalysis suggestive of a urinary tract infection, except none was confirmed by urine culture. To our knowledge,

In terms of sexual development, 77% of the patients in our review were postmenarchal. In the largest pediatric series of ovarian torsion by Kokoska and colleagues,[4] roughly half of the patients were postmenarchal. In contrast, Meyer and colleagues[7] reported on 12 patients with ovarian torsion, only 3 of whom were postmenarchal. Torsion in otherwise normal ovaries is well described. However, it is postulated that menarchal girls are more prone to ovarian cysts due to anovulation. These cysts may act as a fulcrum for ipsilateral ovarian torsion. Ipsilateral cysts were found in most of our patients. Ovarian torsion on the contralateral side to the pain, which we noted in 2 of our patients, has been previously reported[7] and remains unexplained, but may represent the difficulty of pain localization in the pediatric population.

Table 3. Clinical presentation of ovarian torsion across pediatric studies

Study	No. patients	Age, mean (range) yr		Mean duration of symptoms before surgery	Right-sided, %	Vomiting, %	Fever, %	Diagnostic modalities used (%)
Meyer et al.[7]	12	10	(0–15)	4.6 d	58.3	50	16.6	Ultrasound (91.2)
Kokoska et al.[4]	51	12.5	(8–16)	4.8 h	57	73	22	Ultrasound (73) CT (10) OR (17)
Anders and Powell[13]	22	10.2	(3–15)	94.3 h	40.9	77	18	Ultrasound (86.4) CT (18.2)
Rousseau et al.[12]	40	11	(3–14)	NR	57.5	67.5	NR	Ultrasound (97.5) CT (2.5)
Galinier et al.[10]	45	11	(2–17)	3 d	NR	NR	NR	Ultrasound (84.4) OR (15.6)
Oltmann et al.[11]	97	10.9*	(0–17)	NR	68	NR	NR	NR

CT = computed tomography; NR = not reported; OR = operating room.
*Excluding infants (age < 1 yr).

our study represents the first to characterize urinalysis findings in patients with ovarian torsion. Given that no patient was found to have a urinary tract infection by culture, the finding of pyuria is likely nonspecific and may simply reflect irritation or inflammation in the pelvic region. This is important to the front-line clinician, as the presence of pyuria may lead to an incorrect diagnosis and delay timely treatment in patients with ovarian torsion.

The diagnosis of ovarian torsion is usually made with ultrasonography. The most consistent finding is an echogenic pelvic mass with nonvisualization of the ipsilateral ovary[16] or an adnexal mass.[11] Ultrasonography with or without Doppler remains the most feasible initial diagnostic modality because it is readily accessible and can differentiate cystic ovarian pathology from appendicitis without radiation exposure. Our finding that the absence of vascular flow on Doppler-enhanced imaging was not consistently demonstrated highlights a well-described finding that the presence of vascular flow does not rule out torsion.[17] There are 2 explanations for this phenomenon. A dual blood supply from ovarian and uterine arteries provides persistent arterial blood flow. In addition, torsion may cause symptoms related to venous engorgement before arterial blood supply is compromised. Therefore, ultrasound, with or without Doppler, may be misleading, contributing to a subsequent delay in management. If torsion is suspected clinically, laparoscopy is recommended.

All but 2 patients in our study were found to have ipsilateral cysts. Most surgical specimens showed a hemorrhagic ovary. Torsion is a known complication of ovarian cysts. Most of these are follicular or luteal cysts. Their timing with menarche is due to increased hormonal stimulation. Women and girls older than 12 years are more likely than younger girls to have an etiology that includes functional ovarian cysts, congenitally long tubes or supporting ligaments, and increased premenarchal activity leading to venous congestion.[8] Oltmann and colleagues[11] described 97 patients with ovarian torsion and found that pathology was benign in 98% of cases. Findings mainly consisted of infarcted or edematous tissue, benign cysts and benign neoplasms. In contrast, patients with torsion have a malignancy only 2% of the time.[6,7,11] Torsion is the most frequent complication of ovarian neoplasms in children, of which benign teratomas are the most common. Although previous studies have reported a significant proportion of teratomas in patients with a ovarian torsion,[4,6,9,15] our study failed to identify such pathology. This may have been owing to age-related reasons. In younger girls, most cases of torsion are idiopathic or secondary to a mature cystic teratoma. Only 3 patients were younger than 10 years in our study. Ovarian volume with an ultrasound-measured length greater than 5 cm has also been associated with torsion.[11] In 1 of our patients, no predisposing tubal or ovarian abnormalities were noted. Although various theories, such as congenitally long supportive ligaments, have been put forth to explain torsion in hosts with normal adnexa,[18] no such factors were identified in our patient.

Two patients in our review had pathologic findings of a necrotic ovary, suggesting that either time to management was prolonged or that disruption of ovarian circulation was prompt or initially asymptomatic. The infant showed signs and symptoms of systemic inflammation and, although torsion was suspected preoperatively and there was timely intervention,[9] the ovary was found to be necrotic. This begs the question of whether the disease process is accelerated or whether it is initially asymptomatic in very young children. We know of no reports examining this phenomenon in great detail among infants. The other patient had a late presentation with a prolonged duration of symptoms and time to surgery that likely contributed to the perioperative finding of a necrotic ovary.

In the past, management of ovarian torsion consisted of resection of the entire ovary without detorsion. It was believed that a hemorrhagic ovary represented nonviable tissue and that simple detorsion would lead to thromboembolism. A further fear was leaving a malignancy in situ.[9] Roughly half of the patients in our sample underwent this procedure. Given only 1 report of embolic phenomenon following detorsion[5] and the low frequency of malignancy,[4,5,9] a more conservative approach consisting of detorsion with or without cystectomy has been used over the last 20 years.[19] Normal ovarian function has been reported after up to 72 hours of torsion.[20] In several studies, follow-up evaluation has demonstrated sonographic evidence of apparently healthy and well-perfused ovaries following detorsion despite their gross appearances.[5,21–24] An ultrasound performed 6 weeks postoperatively can rule out any suspicion of neoplasm so that it may be resected without the technical difficulties associated with a hemorrhagic ovary.[9] Regarding the contralateral ovary, oophoropexy remains controversial but is often considered in cases of recurrent torsion. The favourable outcomes of the 2 patients in our study who underwent detorsion alone are promising, and it is likely that detorsion alone may become a universal standard of care.

Limitations

The limitations of this study arise from its retrospective nature involving a small number of patients in 1 medium-sized centre. However, these are limitations inherent to the study of an uncommon disease in a population that seldom seeks urgent care. These limitations precluded our ability to identify potential predictors and characterization of ovarian torsion with statistical certainty.

Conclusion

Ovarian torsion is a rare cause of abdominal pain and can result in infarction of the ovary and fallopian tube and

should be considered in any girl or woman with acute onset lower abdominal pain accompanied by vomiting. From all studies of ovarian torsion, the clinical and laboratory features can be quite varied; however, our study demonstrated that pain can be characterized as either constant or colicky, but unlike with appendicitis, does not typically migrate. To our knowledge, our study is the first to document sterile pyuria in a substantial proportion of patients with ovarian torsion, suggesting that urinary tract infection as an alternate diagnosis may delay timely intervention. Nevertheless, no single laboratory or urinalysis findings are consistently suggestive of torsion. Ultrasound is the most useful initial diagnostic modality, but the absence of flow on Doppler imaging is not always present. Young children may present with more advanced or more rapidly progressive disease and require a high index of suspicion. Conservative management consisting of detorsion and oophoropexy is currently advocated despite the macroscopic appearance of the ovary. Timely diagnostic imaging and surgical intervention are essential to preventing the complications of ovarian torsion.

Competing interests: None declared.

Contributors: N. Poonai, R. Lim and T. Lynch designed the study. N. Poonai and C. Poonai acquired the data and analyzed it together with R. Lim. N. Poonai and C. Poonai wrote the article, which R. Lim and T. Lynch reviewed. All authors approved publication.

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