

Original Research Article

Pediatric adnexal torsion - from presentation to outcomes: a 5 years experience of a tertiary center in Bahrain

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ABSTRACT

Background: This study aims to explore the clinical signs, diagnostic methods, and surgical treatments of adnexal torsion in children under 14. The study included 19 children, with an average age of 12, who presented with severe abdominal pain leading to a diagnosis of adnexal torsion.

Methods: We conducted a retrospective review of pediatric patients who underwent surgery for suspected adnexal torsion at a pediatric surgery center from January 2018 to December 2022. Data collected included age, symptoms, diagnostic tests, surgical details, and histopathology results.

Results: The predominant symptoms were abdominal pain, vomiting, nausea, and elevated white blood cell counts in about 50% of the cases. Laparoscopy was the initial approach in 68.4% of cases, with 21.1% requiring conversion to open laparotomy. Ovarian-sparing surgery was performed in 73.7% of the cases, while oophorectomy was necessary in 26.3%. Most histopathological examinations disclosed benign ovarian cysts, including simple cysts, serous cystadenomas, and mature cystic teratomas, often complicated by torsion and leading to ovarian infarction. Younger patients and those with higher leukocyte count found to have higher rates of oophorectomy.

Conclusions: Adnexal torsion, although rare and challenging to diagnose in children due to non-specific symptoms such as abdominal pain, vomiting, and nausea, necessitates prompt recognition and management to minimize severe complications. Awareness among clinicians is crucial for early diagnosis and treatment, significantly impacting long-term outcomes.

Keywords: Adnexal torsion, Pediatric, Tubal torsion, Ovarian torsion

INTRODUCTION

Adnexal torsion is a common gynecological condition, but rare in children where it produces abdominal pain. It accounts for roughly 15% of all torsion cases in pediatric and adolescent populations. It can occur at any age in children (infant to 18 years), but 52% of cases occur between the ages of 9 and 14 with a median age of 11. Neonatal ovarian torsion is rare with only 16% of cases occurring in girls below the age of 1.¹ Adnexal torsion is more common than isolated ovarian torsion and accounts for 67% of cases. Isolated tubal torsion is defined as tubal torsion in the absence of torsion of the ovary-it is rare

with an incidence of only 1.0 per 1.5 million.^{1,2} Adnexal torsion is a clinical diagnosis, and the most commonly used diagnostic imaging modality is ultrasonography with Doppler analysis.

In ovarian torsion, the ovary rotates around its own pedicle compromising its blood supply and leading to ischemia.^{1,2} Patients present with non-specific symptoms such as lower abdominal pain, fever, nausea, and vomiting. These symptoms may mimic other causes of abdominal pain such as acute appendicitis, gastroenteritis, and UTI leading to a delay in diagnosis and management.³⁻⁵

The objective of this study is to examine the clinical manifestations, investigations, including imaging and laboratory studies, and surgical management of adnexal torsion in children.

METHODS

This is a retrospective chart review study. Patients' data were collected from the files and electronic health record of our governmental hospital's center in Bahrain from January 2018 till the 31st of December 2022. This study was approved by the ethical committee of Bahrain government hospitals (research approval serial no. 45-150424). The study subjects included all patients who were diagnosed with or suspected to have adnexal torsion and underwent surgical management between the neonatal period and 14 years of age, as this is considered as the pediatric age group in our center. All the patients included in our study, who presented to our emergency department with abdominal pain, underwent an ultrasound evaluation. The ultrasound results showed confirmatory or suspicious findings of adnexal torsion, leading to subsequent emergency surgery. Exclusion criteria included individuals with a known diagnosis of isolated adnexal pathology without torsion.

The medical records were evaluated for patient's age, presenting signs and symptoms, physical examination findings, imaging and laboratory studies. Data were also obtained on the type of surgical approach (laparoscopic vs laparotomy (Pfannenstiel) approach, oophorectomy vs ovarian-sparing surgery, histopathology results and postoperative follow up for any recurrence.

A data collection sheet was constructed on excel, and a form was made where the data was entered immediately during collection. Basic demographic data including date of birth, age, and diagnosis were added to the excel sheet. More specific data like date of admission, date of surgery, symptoms during presentation, duration of the symptoms, radiological findings, laboratory investigations (WBC), histopathology findings, and the final surgical outcome (oophorectomy vs spared-ovary) were all entered on the form. A fever was defined as temperature greater than 38. Leukocytosis was defined as a white blood cell (WBC) count (greater than $10.0 \times 10^9/l$).

Statistical analysis

Descriptive statistics were employed to summarize the demographic and clinical characteristics of the study population. Categorical variables were reported as frequencies and percentages. Continuous variables were presented as medians and interquartile ranges (IQRs) due to their non-normal distribution and small sample size.

To assess the associations between categorical variables, chi-square tests or Fisher's exact tests were utilized as appropriate, depending on the expected cell counts. For

comparisons involving continuous variables between two independent groups, the non-parametric Mann-Whitney U test was employed due to the non-normal distribution of the data and small sample size.

The statistical significance level was set at $p < 0.05$ for all analyses. All the analyses were carried out using IBM SPSS v29.0.0

RESULTS

Presentation, physical exam, and objective characteristics

We included 19 cases of paediatric patients undergoing adnexal surgery. The median age of presentation was 12 years (IQR 9-12 years). Regarding the type of Pathology based on the intraoperative findings, ovarian torsion was the most common 9 (47.4%), followed by no torsion found 7 (36.8%), tubal torsion 2 (10.5%), and tubo-ovarian torsion 1 (5.3%).

The most common symptom was abdominal pain, followed by vomiting and fever. The most common location of abdominal pain was the right lower quadrant (63.1%), followed by the left lower abdomen (21.1%). The duration of pain was most frequently reported to be 2 days (36.8%) or 1 day (31.6%). The majority of patients presented with vomiting/nausea (94.7%), while fever was less common (15.8%). Around half of the patients had an elevated white blood cell count (47.4%) (Table 1).

On physical examination, tenderness was present in 68% of cases. The most frequent location of tenderness was the right lower quadrant (64.6%), left lower quadrant and suprapubic region (17.7% each). Guarding was noted in 16% of cases, while rebound tenderness, palpable mass, and distension/fullness were less common (4-8% each) (Table 2). All patients underwent ultrasonography, and two patients underwent computed tomography (CT).

When comparing patients aged ≤ 12 years (dichotomized on the base of median age) to those between 12-14 years of age, there were no significant differences between the clinical signs and symptoms or type of torsion (Table 3 and 4).

Surgical management

We evaluated laparoscopy vs laparotomy vs laparoscopic converted to laparotomy (open) approach. There were 13 laparoscopic cases, four laparoscopy cases converted to laparotomy, and two cases were laparotomy.

The most common surgical approach was laparoscopy accounting for 13 (68.4%) of cases. Of all laparoscopic surgeries, four cases required conversion to open (21.1%). Laparotomy was the least common approach accounting for 2 (10.5%) (Table 5).

Table 1: Demographics and the clinical presentation.

Variables		Median	IQR
Age (in years)		12	9-12
Abdominal pain localization	Generalized	1	5.3
	Left lower abdomen	4	21.1
	Generalized lower abdominal pain	2	10.5
	Right lower abdomen	12	63.1
Duration of pain (days)	1	6	31.6
	2	7	36.8
	3	4	21.1
	4	2	10.5
Vomiting/ nausea	No	1	5.3
	Yes	18	94.7
Fever	No	16	84.2
	Yes	3	15.8
WBC count	Elevated	9	47.4
	Normal	10	52.6

Table 2: Physical examination findings.

Findings ^{a,b}	N	Percentage (%)
Tenderness present	17	68
Guarding present	4	16.0
Rebound tenderness	1	4.0
Palpable mass	1	4.0
Distension/ fullness	2	8.0
Location of tenderness		
Right lower quadrant (RLQ) tenderness	11	64.6
Left lower quadrant (LLQ) tenderness	3	17.7
Suprapubic tenderness	3	17.7

^aN and N% calculated based on only positive findings, one of the patients had soft, lax, non-tender abdomen. ^b1 patient could have >1 positive findings, thus tally of findings >N.

Table 3: Type of torsion.

Type of torsion	N	Percentage (%)
Ovarian	9	47.4
Tubal	2	10.5
Tubo-ovarian	1	5.3
No torsion	7	36.8

Table 4: Comparison by age groups.

Variables		Up to 12 years		13 and above		P value
		N	%	N	%	
Duration of pain (days)	1	5	26.3	1	5.3	0.596
	2	6	31.6	1	5.3	
	3	2	10.5	2	10.5	
	4	2	10.5	0	0.0	
Vomiting/ nausea	No	1	5.3	0	0.0	1.000
	Yes	14	73.7	4	21.1	
Fever	No	12	63.2	4	21.1	0.211
	Yes	3	15.8	0	0.0	
WBC count	Elevated	8	42.1	1	5.3	0.303
	Normal	7	36.8	3	15.8	

Continued

Variables		Up to 12 years		13 and above		P value
		N	%	N	%	
Type of pathology	Ovarian	7	36.8	2	10.5	1.000
	Tubal	2	10.5	0	0.0	
	Tubo-ovarian	1	5.3	0	0.0	
	No torsion	5	26.3	2	10.5	

Table 5: Type of surgical approach.

Surgical intervention	N	Percentage (%)
Laparoscopy	13	68.4
Laparoscopy converted to laparotomy	4	21.1
Laparotomy	2	10.5

Table 6: Type of surgery being performed.

Type of surgery		N	Percentage (%)
Oophorectomy	Cyst	4	21.0
	Normal	1	5.3
Ovarian sparing	Cyst	12	63.2
	Normal	2	10.5

Table 7: Histopathology findings.

Histopathology finding	N	Percentage (%)
Benign serous cystadenoma/ cyst	5	26.32
Torsion/ovarian infarction	4	21.05
Unavailable	4	21.05
Mature cystic teratoma	2	10.53
Paratubal/fimbrial cyst	2	10.53
Hemorrhagic ovarian cyst	2	10.53

Table 8: Association between presentation and surgical management.

Variables		Type of surgery				P value
		Oophorectomy		Ovarian sparing		
		MDN	IQR (%)	MDN	IQR (%)	
Age (in years)		10	8-11	12	11-13	0.050
Duration of pain (days)	1	2	40.0	4	28.6	0.437
	2	3	60.0	4	28.6	
	3	0	0.0	4	28.6	
	4	0	0.0	2	14.3	
Fever	No	4	85.7	12	85.7	1.000
	Yes	1	14.3	2	14.3	
WBC count	Elevated	5	100.0	4	28.6	0.011
	Normal	0	0.0	10	71.4	

Oophorectomy was performed in 5 cases (26.3%) with four cases (21.0%) involving presence of a cyst and one case (5.3%) where ovary appeared bulky with no cyst. Ovarian-sparing surgery was most common approach and was performed in 14 cases (73.7%). This included the 12 cases (63.2%) where a cyst was present and 2 cases (10.5%) with a normal-appearing ovary (Table 6).

Regarding pathologic examination, it demonstrated a variety of ovarian cystic lesions, with benign simple cysts

(n=2, 10.5%) and benign serous cystadenomas (n=2, 10.5%) being the most common findings. Mature cystic teratomas or dermoid cysts were also frequently encountered (n=2, 10.5%), some of which exhibited associated hemorrhage (n=2, 10.5%). A notable proportion of cases involved ovarian cysts complicated by torsion (n=4, 21.1%), leading to ovarian infarction in one instance (n=1, 5.3%) (Table 7).

We completed further analyses to determine any associations between age, presentation and surgical management. Oophorectomy was done in younger patients with median age 10 years (IQR: 8-11) and ovarian sparing was done in relatively older patients with median age 12 years, (IQR: 11-13) and this difference was borderline significant ($p=0.050$).

Finally, assessing duration of pain, fever, and leukocytosis, patients who underwent oophorectomy had a higher rate of leukocytosis compared to those who had ovarian-sparing surgery (100% vs 28.6%, $p=0.011$) (Figure 2). All patients with a normal WBC count underwent ovarian-sparing surgery. However, there were no significant differences in duration of pain or presence of fever between the two surgical groups (Table 8).

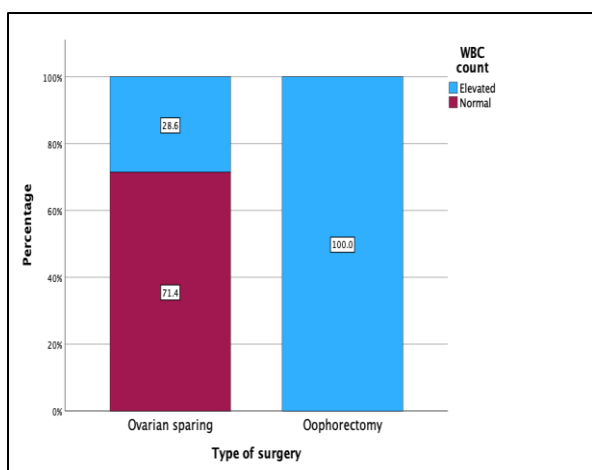


Figure 1: Comparing leucocytosis across type of surgery.

Follow up

Of the five patients who underwent oophorectomy, one patient failed to attend the follow ups, and two cases reported no post-surgical complications. One patient experienced recurrent left-sided abdominal pain, but ultrasound was normal, and the patient was advised for regular follow-up. By 2020, this patient was asymptomatic, and a repeat ultrasound showed a normal right ovary.

For the 14 patients who underwent ovarian-sparing surgery, four patients missed their follow up, while four cases reported no post-surgical complications or recurrences. Two patients were lost to follow-up, and four patients had a documented unremarkable one-year follow-up.

DISCUSSION

This study sheds light on adnexal torsion within the largest tertiary care center in our country. It offers essential insights into the characteristics, clinical presentations, and surgical approaches specific to adnexal

torsion. The diagnosis of adnexal torsion remains complex due to its non-specific symptoms, which often lead to misdiagnoses and delayed interventions.

The objective of our study was to identify the signs and symptoms exhibited by patients with adnexal torsion. The median age was 12 years, which also falls within the range reported in other studies. The most common symptoms were abdominal pain, vomiting, and nausea. These findings are consistent with previous studies as well.⁶⁻⁸

We found higher rates of right-side pain and torsion accounting for 12 cases (63.1%), which falls within the range of other studies.²⁻⁸ This observation is potentially explained by anatomical factors such as the presence of the sigmoid colon on the left iliac fossa, which aids in decreasing the mobility of the tubal structure, thus reducing the chance of left adnexal torsion.^{9,10}

Of the 19 patients who had a preoperative diagnosis or suspected adnexal torsion, a plurality ($n=9$; 47.4%) had confirmed intra-operative ovarian torsion; seven (36.8%) had negative torsion with existing adnexal pathology. Tubal and tube-ovarian torsion were the least common accounting for two (10.5%) and one case (5.3%), respectively. All patients underwent preoperative ultrasound evaluation as part of the diagnosis workup for abdominal pain in the emergency, although this information is not reflected in the statistics provided. The abdominal ultrasound conducted on these patients revealed the presence of a pelvic or abdominopelvic mass with either questionable vascularity or complete absence of vascularity-specifically in cases where torsion was confirmed. While ultrasound serves as a beneficial diagnostic tool, its effectiveness still hinges on the skills of the operator. Therefore, the final diagnosis of adnexal torsion ultimately depends on clinical judgment.¹¹⁻¹⁴

Predictive factors for adnexal torsion in girls with abdominal pain and a pelvic mass

There is no consensus in the literature regarding accurate predictive factors for ovarian torsion in terms of assessing the duration of pain, fever, and leukocytosis. Gasser et al concluded that only 20.1% of individuals experienced fever, while 51.4% demonstrated leukocytosis.¹¹ These observations did not improve the diagnostic process in other studies.^{4,8,9,11,15}

We further examined the association between oophorectomy cases and the rate of leukocytosis. Higher leukocytosis rates were evident in oophorectomy cases with a rate of 100% compared to the ovarian-sparing surgery patients with 28.6%, thus suggesting a potential correlation with adnexal torsion severity. However, the value of this predictive factor remains controversial in the literature.^{6,7,11,15} Recent literature underscores the importance of thorough pain assessment-particularly for pelvic masses exceeding 5 cm.

A recent study by Liu et al highlighted the significant importance of pain assessment, particularly in girls who have a pelvic mass size exceeding 5 cm.¹⁶ The study emphasized the importance of promptly addressing abdominal pain specifically when it is accompanied by acute pain characterized by sudden onset that persists without relief or that recurs. These cases have a high suspicion index of adnexal torsion.¹⁶

Surgical management

There are historical shifts in the surgical management from oophorectomy towards more conservative approach due to concerns about fertility and the minimal risks of malignancy and embolic events. Earlier theories suggested that coagulated blood might escape into the vessels following the reduction of torsion, thus leading to emboli formation. However, these theories were later contradicted by multiple case series reporting an incidence of only 0.2% of pulmonary emboli. None of the patients reported embolic disease after conservation surgery.¹² Furthermore, the incidence of malignancy is estimated to be approximately 2% in cases of ovarian torsion.¹²

Interestingly, several studies have indicated differences in management practices depending on the surgeon's specialty. For example, pediatric surgeons are more likely to perform oophorectomy than gynecologists. This can be attributed to the gynecologist's knowledge and understanding of the long-term risks to fertility that can arise from the complete removal of ovaries or fallopian tubes.^{6,13} Oltmann et al studied the pediatric literature and found salvage rates were around 0 to 50% regardless of surgeon specialty.^{3,9,10,17} In our cohort, conservative approaches accounted for 73.7% of the cases.

Pediatric surgeons at our center perform all surgeries for adnexal torsion in patients under the age of 14. There were a few cases where the gynecology team was involved to provide a second opinion. Laparoscopy is the most common method with detorsion and/or cystectomy accounting for 68.4%. Another 21.1% of those cases necessitated conversion to open surgery due to either the cyst's significant size causing limited working space or the surgeon's experience. Furthermore, the preferred approach was ovarian-tubal conserving approach that accounted for 14 (73.7%) of the cases. Oophorectomy was performed in 5 cases (26.3%). The operative notes indicated that these tubes/ovaries were blackish and gangrenous and did not improve after detorsion.

Finally, the pathology exam revealed a variety of ovarian cystic lesions. The most common findings were benign simple cysts (n=2, 10.5%) and benign serous cystadenomas (n=2, 10.5%). Mature cystic teratomas or dermoid cysts were also frequently seen (n=2, 10.5%); some of these showed associated hemorrhage (n=2, 10.5%). A significant number of cases involved ovarian

cysts complicated by torsion (n=4, 21.1%) resulting in ovarian infarction in one instance (n=1, 5.3%). These findings were not significantly different than other studies.^{4,6,17} Although there may be an underestimation, it is important to note that histological examinations were unavailable for four cases (21%). These cases most likely involved simple clear cysts in which the surgeon performed a cyst puncture without the need for histopathology.¹⁸

Our study has a few limitations. First, it is a retrospective study conducted at a single institution. Nevertheless, the findings may have value to other centers and contribute to the existing literature. A second limitation is the relatively small sample size, although this is reflective of the rare incidence of adnexal torsion in children. Another limitation is the lack of important clinical details in the database such as the nature of symptoms for each patient, e.g., (intermittent, constant pain, etc.) Finally, data on long-term adnexal viability was not feasible for analysis because some patients missed their follow ups.

CONCLUSION

This study emphasizes the critical need for prompt and accurate diagnosis of adnexal torsion. Early diagnosis of adnexal torsion is challenging due to the lack of specific clinical signs. Leukocytosis may be a potential correlation with adnexal torsion severity. Acute abdominal pain is a common symptom, and vomiting and nausea are common symptoms. Prompt diagnosis is essential because delayed intervention can lead to irreversible damage to the affected adnexa. Further research and standardized guidelines are needed to enhance our understanding and effectively manage this challenging condition.

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