Original Study

Adnexal Torsion in Children and Adolescents: A Retrospective Review of 245 Cases at a Single Institution

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Oluyemisi Adeyemi-Fowode MD [1,2,]*, Emily G. Lin MD [1], Fatima Syed BSc [1],
Haleh Sangi-Haghpeykar PhD [1], Huirong Zhu PhD [3], Jennifer E. Dietrich MD, MSc [1,2]

[1] Department of Obstetrics and Gynecology, Baylor College of Medicine, Houston, Texas
[2] Division of Pediatric and Adolescent Gynecology, Department of Obstetrics and Gynecology, Baylor College of Medicine, Houston, Texas
[3] Outcomes and Impact Service, Texas Children's Hospital, Houston, Texas

A B S T R A C T

Study Objective: Adnexal torsion (AT) requires urgent surgical intervention to preserve ovarian function. Historically, treatment for AT was oophorectomy because of concerns related to leaving a nonviable ischemic ovary. No published studies support these theoretical concerns and current literature supports conservative management. The aim of this study was to review the institutional outcomes for AT cases, including salvage rates and complications.
Design, Setting, Participants, Interventions, and Main Outcome Measures: This study was approved by the institutional review board at Baylor College of Medicine. A retrospective chart review on cases of AT from 2007 to 2016 at a single Children's hospital was performed on the basis of International Classification of Diseases, 10th revision and Current Procedural Terminology codes. A standardized chart review form was used in data extraction. Statistical analysis was performed using SAS version 9.4 (SAS Institute).
Results: Chart review identified 245 torsion cases in 237 patients. The mean age was 12.4 ± 3.29 years. Of the participants, 230 (94%) underwent minimally invasive laparoscopy with ovarian preservation in 233 (95%) of the cases. There were no complications due to detorsion of the affected adnexa. Intraoperatively, the right adnexa was affected in 134 (55%) cases and a lesion was noted in 193 (79%) cases, most commonly paratubal cysts and mature teratomas. The malignancy rate was low, noted only in 4/245 patients at (1.2%). Pediatric gynecology performed most of the cases (n = 214; 87%).
Conclusion: The findings of our study continue to support the conservative management of patients with AT.
Key Words: Adnexal torsion, Ovarian torsion, Tubal torsion, Pediatrics, Adolescents, Surgical management

Introduction

Adnexal torsion (AT) is an uncommon gynecologic disorder caused by the partial or complete rotation of the ovary and/or the fallopian tube on its vascular support, but it is the fifth most common gynecologic emergency.[1,2] It accounts for 2.7% of all cases of children with acute abdominal pain.[3,4] Although the actual incidence is unknown, studies have reported between 0.3% and 3.5% cases per year.[5,6] AT mainly occurs in adolescents and women of childbearing age, thus when suspected, urgent surgical intervention is indicated to preserve ovarian function, because delays in treatment can lead to impaired or lost fertility. The mechanism of torsion in women and girls with ovarian masses is likely related to the increased size and weight of the involved ovary.[6,7]

Historically, the treatment for AT was oophorectomy on the basis of theoretical concerns, such as leaving a nonviable severely ischemic ovary that might lead to infection or return to the operating room, missing a malignancy, and/or potential for thromboembolism from untwisting ovarian vessels.

No studies to date support these theoretical concerns and current literature suggests the standard of care should be conservative management.[8,9] With increasing access to laparoscopy in many health care institutions, there has also been an increase in the application of minimally invasive surgery techniques to address AT.[10–12] Lo and colleagues investigated laparoscopy vs laparotomy for surgical intervention for AT and noted that laparoscopy was associated with shorter hospital stay, fewer postoperative complications, and improved opportunities for ovarian preservation.[12]

The treatment paradigm is thus changing to favor adnexal detorsion with ovarian conservation in an effort to preserve ovarian function. Although some studies show a high rate of ovarian conservation (85%-95%),[13–15] oophorectomy is still a widely performed procedure despite good outcomes with conservative treatment. A recent literature review of 14 different studies showed an ovarian salvage rate of only 32%.[4] Another study used a nationwide database to review the national trend in the surgical treatment of AT in children and analyzed 2041 pediatric patients and noted that release of torsion alone without oophorectomy had increased from 9% in 1998 to 25% in 2011.[16]

The aim of this study was to review institutional outcomes for AT cases. The primary outcome to be assessed was the salvage rate. Secondary outcomes measures included

postoperative infections, reoperation rates, postoperative thromboembolism, and missed malignancies.

Materials and Methods

The institutional review board at Baylor College of Medicine approved this study. The medical records of all children with surgically diagnosed AT (including International Classification of Diseases, 10th revision codes for isolated tubal, isolated ovarian, and pedicle involving the ovary and tube) admitted to a single children's hospital between 2007 and 2016 were reviewed retrospectively. Patients seen initially at an outside institution and transferred to our center were also included in the chart review. The medical charts were reviewed for basic demographic characteristics, presenting clinical symptoms and signs, laboratory and imaging studies on presentation, surgical management, intraoperative findings, postoperative outcomes, and pathology findings. A standardized chart review form was used in data extraction. Missing data were not included in the analysis.

Statistical Analyses

Data were compiled into a computer database. For descriptive analyses, categorical data are expressed as frequencies (percentages), continuous data are expressed as means and standard error of the mean. When comparing proportions between 2 groups, Wilcoxon rank sum tests were used for continuous variables and Fisher exact tests for categorical variables. Statistical analysis was performed using SPSS and SAS statistical software (version 9.4, SAS Institute). A P value less than .05 determined statistical significance.

Results

Patient Demographic and Clinical Characteristics

The chart review identified 245 surgically confirmed torsion cases in 237 patients. The mean age was 12.4 ± 3.29 years (range, 0.4-19). 70% (n = 160) were menarchal, and 30% (n = 68) were premenarchal women. All 245 patients reported abdominal pain with most (n = 107; 47%) reporting pain in the right lower quadrant. On a visual pain scale, severity of pain of 8 or more was reported 77% (n = 122) of the time with 57% (n = 94) describing the pain as intermittent. Radiation was reported by only 32% of patients (n = 43); when present, the flank was the most commonly reported location of referred pain at 15% (n = 20). Associated symptoms of nausea and/or vomiting occurred in 196 patients (87%). Subjective fever was only reported by 6% of patients (n = 14). White blood cell count (WBC) was available for 194 patients. Mean WBC was $12.1 \pm 4.17 \times 10^3/$ μL, however, 43% (n = 84) had an elevated WBC greater than 13. Preoperative ultrasound was suggestive of torsion 69% of the time (n = 138; Table 1). Comparison between premenarchal and menarchal groups revealed no significant differences in the largest dimension (5.78 ± 2.39 cm vs 6.27 ± 3.76 cm; $P = .9$) or volume (78.4 ± 121 cm^3 vs 272 ± 145 cm^3; $P = .77$) of the affected adnexa as reported on preoperative ultrasound (Fig. 1).

Table 1
Presenting Signs and Symptoms of Premenarchal and Menarchal Girls Who Underwent Laparoscopy for Adnexal Torsion (N = 245*)

Presentation	n[†]	%[‡]
Symptoms	245	100
Abdominal pain	196	87
Nausea/vomiting	196	87
Fever	14	6
Pain description, severity		
Mild (0-4)	3	2
Moderate (5-7)	33	21
Severe (≤8)	122	77
Location		
RLQ	107	47
LLQ	76	34
Suprapubic/pelvic	37	16
Periumbilical	7	3
Quality		
Intermittent	94	57
Constant	72	43
Radiation		
None	91	68
Pelvis	14	10
Flank	20	15
Groin	9	7
Labs/imaging		
Elevated WBC (≥13)	84	43
U/S suggestive of torsion	138	69

LLQ, left lower quadrant; RLQ, right lower quadrant; U/S, ultrasound; WBC, white blood cell count.
* Total number of adnexal torsion cases.
† Nonmissing number value.
‡ Percentage of nonmissing number.

Intraoperative Findings

Ninety-four percent of the patients underwent laparoscopy (n = 230); 6% (n = 15) underwent laparotomy, of which 6 were described as mini laparotomy (incision < 4 cm). The most common reasons cited for laparotomy was size of the cyst (n = 9) and concern for malignancy (n = 4); 2 cases had unclear indication. The right adnexa was noted to be affected 55% of the time (n = 134). Isolated tubal torsion was noted in 10% of patients (n = 24) with 3 patients who underwent salpingectomy, all isolated tubal torsion cases involved an associated cystic lesion of the ipsilateral tube or ovary. There was 1 case of bilateral tubal torsion in a 14-year-old girl, which involved isolated torsion of the fallopian tubes bilaterally due to bilateral paratubal cysts. Overall, 79% of the cases (n = 193) involved a lesion (functional, benign, or tumor) of the tube and/or ovary. The affected adnexa was described as abnormal (edematous, purple, blue-black) 79% of the time (n = 146). Despite the high rate of abnormal findings, there was a high salvage rate of 95% (n = 233), with only 5% of patients (n = 12) who underwent oophorectomy because of concern for necrosis of the ovary (n = 11) or because of concern for malignancy (n = 1; Table 2). Intraoperative lesion size was also assessed. Most (n = 155; 91%) torsion cases involved lesions of 5 cm or larger (Fig. 2). Pediatric gynecology performed most of these procedures (n = 214; 87%).

Pathology Results

Data were categorized into premenarchal and menarchal groups, missing data were not included. On the basis of this algorithm, although there were 245 cases of
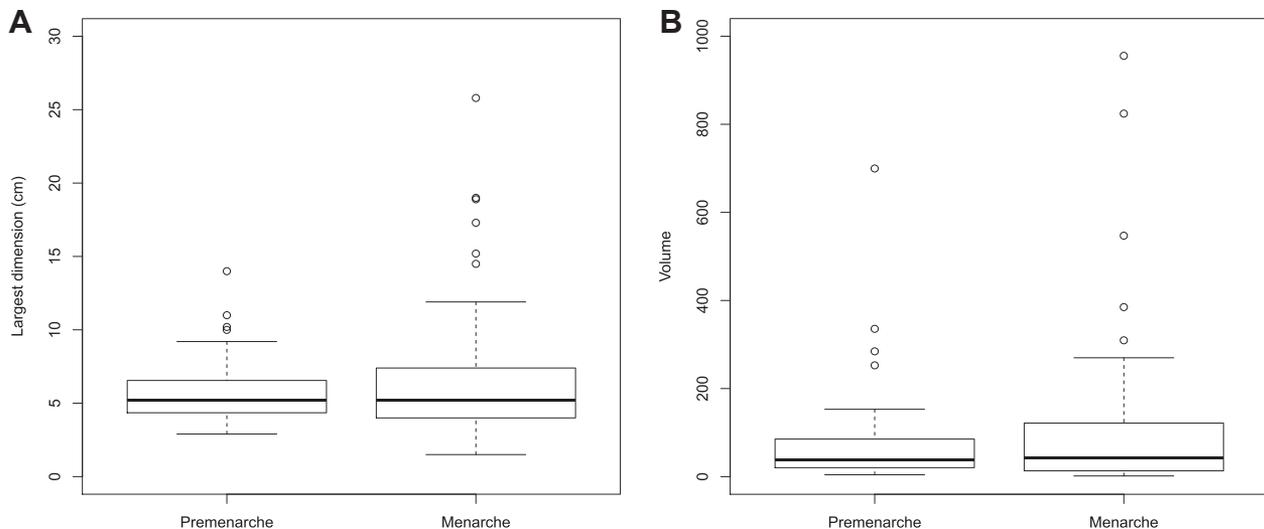
A

B

Fig. 1. (A) Largest dimension of affected adnexa on preoperative ultrasound on the basis of menarchal status (P = .9). (B) Volume (cm^3) of affected adnexa on preoperative ultrasound on the basis of menarchal status (P = .77).

ovarian torsion, menarchal status was not available for 17 cases thus only 228 cases were included in pathology result analysis. Actual tissue diagnosis was available for 87% (199 of 228) because 13% (29 of 228) of patients were treated with detorsion only.

Of the 199 cases with tissue analysis, 17% (33 of 199) were biopsied in the absence of a lesion and revealed only hemorrhagic ovarian stroma. In the premenarchal group (n = 68), no pathology was noted half of the time whether by visual inspection (n = 15) at the time of surgery or if biopsy was performed (n = 19). The most common pathological findings in this age group were paratubal/paraovarian cysts (n = 17; 32%) and mature teratomas (n = 11; 21%). In contrast, the menarchal group was noted to have pathology most of the time (n = 132; 90%), with the most common pathology noted to be paratubal/paraovarian cysts (n = 88; 60%), followed by mature teratomas (n = 24; 16%). Differences in pathology findings between groups was significant, showing that premenarchal girls were less likely to have pathology (P < .0001; Table 3).

The malignancy rate was low at 1.2% (n = 4 of 245 cases). Reported malignancies included immature teratoma (n = 2), adenocarcinoma (n = 1), and serous borderline tumor (n = 1; Table 4).

Recurrent Cases

Of patients who had recurrent AT (n = 8; 3.4%), recurrences were noted on the same side in all cases. Although none had pathologic lesions with the initial torsion, at the time of recurrence, 75% (n = 6) had pathologic masses. Oophoropexy was performed during the subsequent torsion on 62.5% of the patients (n = 5).

Follow-up

There were no reoperations to remove the torsed adnexa. There was 1 readmission in an open laparotomy case for small postoperative abscess vs wound cellulitis. This

infection resolved with antibiotics alone. There were no cases of thromboembolism after detorsion. Most (81%; n = 199) presented for clinic follow-up (mean postoperative day = 24.1 ± 28.8). Postoperative imaging was ordered at this time in 51% (n = 123). Patients followed-up for ultrasound examinations on average at 265 ± 389 days postoperatively, with most (67%; n = 83) of post-detorsion ultrasound examinations showing normal ovaries with active follicles on the side affected by AT.

Discussion

To our knowledge, we present the largest series of AT published to date in the pediatric population. Our primary outcome was to examine the salvage rate. Our rate of salvage (95%) surpasses the national average.[4,17,18] Secondary outcomes were focused on complications related to conservative management strategies: thromboembolism, infection, reoperation, missed malignancy. Our findings are consistent with other studies, showing that adnexal detorsion is not associated with an increased complication rate.[8,17] A recent systematic review by the American Pediatric Surgical Association Outcomes and Evidence Based Practice Committee examined evidence on ovarian torsion in the pediatric and adolescent population and concluded that there is no evidence to suggest an increased risk of pulmonary embolism, specifically attributable to ovarian detorsion; therefore, this theoretical risk should not influence surgical decision-making.[8] Additionally, we had only 1 readmission to the hospital for wound cellulitis vs possible abscess, however, this complication was not specific to adnexal detorsion. Finally, there were no reoperations after detorsion.

The preoperative diagnosis of AT is challenging because of the nonspecific clinical presentation. A systematic review of clinical data among patients with surgically diagnosed AT noted that 97.5% reported abdominal pain with associated nausea/emesis.[4] This is consistent with our study, because 100% reported acute abdominal pain symptoms, with associated emesis also noted to be common (87%). Elevated

Table 2
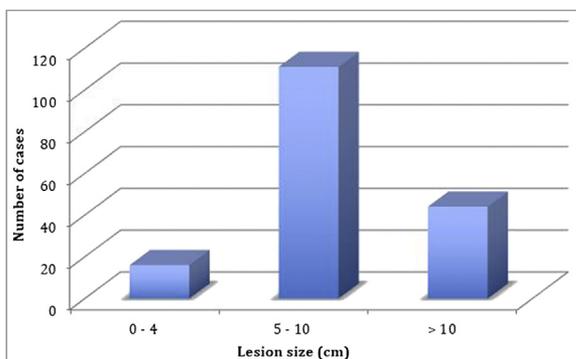Procedures Performed and Intraoperative Findings (N = 245)

	n	%
Procedure		
Laparoscopy	230	94
Laparotomy	15	6
Affected adnexa		
Left	109	45
Right	134	55
Bilateral	1	0.4
Isolated tubal torsion	24	10
Left	8	
Right	15	
Bilateral	1	
Type of lesion		
None	52	21
Paratubal	92	38
Ovarian/paraovarian	101	41
Description of color of ovary*		
Normal	38	21
Abnormal	128	70
Only fallopian tube abnormal†	18	9
Operating service		
Pediatric gynecology	214	87
Pediatric surgery	31	13
Oophorectomy	12	5
Laparoscopic oophorectomy	9	
Open oophorectomy	3	

* Description of color of the ovary on the affected side.
† Applicable only to cases of isolated tubal torsion.

Table 3
Pathology Results*

Pathology	Premenarche (n = 53)	Menarche (n = 146)	P
Hemorrhagic ovarian tissue	19 (36)	14 (9.6)	.0001
Paratubal/paraovarian	17 (32)	88 (60)	<.0001
Mature teratoma	11 (21)	24 (16)	.79
Serous/mucinous cystadenoma	2 (3.8)	8 (5.5)	.5
Functional cysts	1 (1.9)	11 (7.5)	.098
Immature teratoma	2 (3.8)	0 (0)	.12
Adenocarcinoma	1 (1.9)	0 (0)	.12
Serous borderline tumor	0 (0)	1 (0.7)	.52

Data are presented as n (%).
* Patients who had no pathology available due to detorsion only were excluded (15 premenarchal and 14 menarchal patients).

WBC was not a frequent finding in our study, consistent with previous studies.[4,13,19]

Ultrasound is the gold standard imaging choice for AT because of the ability to directly and rapidly evaluate ovarian anatomy and perfusion in a noninvasive manner.[20] On ultrasound examination, absent Doppler arterial flow and ovarian enlargement are reported to be the most helpful hallmarks of torsion, but these findings are not always present.[21,22] Even in the setting of acute torsion, preservation of Doppler arterial flow can often be seen because venous and lymphatic flow is affected first. This explains why normal Doppler findings in the setting of AT should not exclude torsion.[21,23] Findings on ultrasound examination suggestive of AT include: increased ovarian size, peripheral distribution of follicles, abnormal location of the adnexa compared with the uterus, and the presence of free fluid.[19,23,24] In our study, 69% of preoperative ultrasound examinations were suggestive of AT. Ultrasound is a useful tool, however, the diagnosis of AT is ultimately clinical.

Fig. 2. Intraoperative description of size of lesion noted on affected adnexa.

Intraoperatively, AT is overall reported to occur more commonly on the right side than on the left.[4,25−27] This was also noted to be consistent in our study, with 55% of AT cases occuring on the right. This has been attributed to the relative mobility of the cecum on the right side, allowing for more ovarian movement, compared with the fixed nature of the sigmoid colon on the left side.[1,28] Adnexal pathology has been reported to be associated with 51%-84% of AT cases.[1,5] In our cohort, 79% had adnexal pathology with the most common being paratubal cysts followed by dermoid cysts. Compared with the menarchal group, premenarchal patients were less likely to have associated pathology (50% vs 17% Respectively). The increased incidence of pathology during adolescence is likely secondary to hormonal influence and gonadal growth resulting in an increased frequency of physiologic and pathologic masses.[7,15,29,30] Our pathology findings were consistent with other studies that showed that the most frequently encountered ovarian and tubal lesions were benign, such as paratubal cysts, cystic teratomas, and hemorrhagic/follicular cysts. Oltmann and colleagues also showed a strong association between AT torsion and a pelvic mass larger than 5 cm.[26] This was consistent with our study in which 91% of AT cases were associated with pelvic masses greater than 5 cm diameter. Because of this risk, it is reasonable to detorse the twisted adnexa and perform a cystectomy simultaneously to prevent recurrence. Occasionally, the twisted adnexa can be severely edematous and friable, in which case, attempts at cystectomy can result in further ovarian tissue damage, bleeding, and need for emergent oophorectomy.[31] In these cases, consideration should be given to detorsion alone with the goal of ovarian conservation. Reimaging at a later time and performing an interval cystectomy if indicated can then be undertaken.

AT involving the ovary and tube is far more common than isolated torsion of the ovary or fallopian tube.[7,23] Tubal torsion is almost always associated with tubal pathology such as a hydrosalpinx or tubal cyst.[6,23] The incidence of isolated tubal or ovarian torsion in pediatric and adolescent patients is difficult to determine. Case series have collectively reported between 13 and 15 cases over 10 years.[28,32] In our study, isolated tubal torsion occurred in 10% of cases, all of which were associated with tubal pathology. Asynchronous and synchronous AT are very rare.[33−35] In our

Table 4
Patients with Ovarian Malignancy Presenting with Adnexal Torsion

Age, years	Menarche	Procedure	Affected Adnexa	Oophorectomy	Pathology	Stage	Spillage during Initial Surgery	Treatment	Recurrence
8	No	Laparotomy	Left	No	Immature teratoma	I	No	Cystectomy	No
10	No	Laparoscopy	Right	No	Immature teratoma	IC*	Yes	Cystectomy	No
12	Yes	Laparotomy	Right	Yes	Poorly differentiated adenocarcinoma	IIIA	No	Adnexectomy Complete staging	Unknown[†]
16	Yes	Laparoscopy	Right	No	Serous borderline tumor	—	Yes		Yes[‡]

* Stage upgraded because of intraoperative spillage.
[†] Refused any further treatment after diagnosis, lost to follow-up.
[‡] Recurrence on ipsilateral side 3 years later treated with cystectomy, spillage occurred during this procedure as well.

cohort, we had only 1 case of synchronous isolated tubal torsion due to bilateral paratubal cysts.

Malignant lesions are uncommonly associated with AT in pediatric patients, although rates between 1% and 6% have been reported.[10,30,36–39] The most common torsed malignant ovarian neoplasms in the pediatric population are of germ cell origin. Oltmann et al[36] noted only 4 malignancies (3.5%) among 114 cases, similar to what Stankovic and colleagues[39] noted (4%). Our study showed a low malignancy rate with only 4 malignancies (1.2%) among 245 cases.

With adnexal pathology, recurrence rates of up to 12% have been reported; it appears to be even higher among premenarchal girls found to have otherwise normal ovaries and fallopian tubes.[13,19,38,40,41] In our cohort, there was a recurrence rate of 3.4% and although no association could be made regarding pubertal status, it was noted that none had pathologic lesions with the initial torsion.

The benefits to oophoropexy are controversial. Some have cited fears that ovarian fixation could impair the ovary and compromise the anatomic relationship between the ovarian follicles and oviduct.[19,40–42] Although this is a theoretical concern, there are no prospective studies available to assist with decision-making in this situation. Nonetheless, experienced surgeons suggest that oophoropexy can be offered in the setting of recurrent torsion, absent contralateral ovary presence, elongated ovarian ligament, and torsion of normal adnexa.[19,40,43] In our cohort, 5 of the 8 recurrences (62.5%) received an oophoropexy during their second presentation, none of which had associated pathology during the initial or subsequent surgery.

The retrospective nature of this study is a limitation of this study and therefore a small amount of selection bias cannot be excluded. In addition, in rare cases, some data were missing or excluded because of limited follow-up information.

Conclusions

AT is a surgical emergency. Prompt diagnosis and early intervention can improve reproductive outcome. Urgent laparoscopic adnexal detorsion with ovarian conservation should be offered. The findings of our study continue to support use of conservative techniques with high rates of salvage and rare complications.

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