Is It Ovarian Torsion? A Systematic Literature Review and Evaluation of Prediction Signs

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Objectives: This study aimed to identify, through systematic literature review, the most reliable clinical, biological, and radiological signs of ovarian torsion in the pediatric population and to compare their diagnostic value.

Methods: This is a systematic review of the literature, searching MEDLINE, EMBASE, and Cochrane Databases for articles published between January 1990 and January 2014.

Results: From the 946 references initially identified, 14 retrospective publications fulfilled the inclusion criteria, involving a total of 663 episodes of ovarian torsion. Sudden onset abdominal pain with nausea and/or vomiting is the most frequent symptom of ovarian torsion. It can occur at any age, not only in menarchal or perimenarchal patients. Abdominal tenderness is present in 88.4% of patients, whereas only 24% have a palpable mass. Blood tests are commonly requested (51.4% of cases) but are not diagnostic. Abnormalities on plain abdominal radiograph include masses, calcifications, and ossified images. Ultrasound has a sensitivity for ovarian torsion of 79% and computerized tomographic scan of 42.2%. There is a significant diagnostic delay at 101.8 hours (median).

Conclusions: Abdominal pain in children and adolescents is difficult to evaluate, and the diagnosis of ovarian torsion remains a challenge. Because of its potential complications, we need effective clinical tools. From our review of the literature, it was not possible to develop a diagnostic algorithm. Further research is needed to improve our practice and shorten the delay to diagnosis. Considering the low incidence of ovarian torsion, a multicenter prospective study would be required.

Key Words: ovarian torsion, adnexal torsion, acute pelvic pain

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The incidence of ovarian torsion in the pediatric emergency department (ED) is 0.5 to 2 per 10,000 patients.[1,2] It accounts for 2% to 3% of all consultations for abdominal pain in EDs,[1,3,4] and its diagnosis is often delayed because of nonspecific clinical, biological, and radiological signs. A prolonged interval between the onset of pain and the diagnosis of torsion correlates with a decreased rate of ovarian salvage. It is difficult to influence the duration between the first symptoms and consultation in the ED. However, the duration of pain between first examination and operative intervention does influence the outcome as well,[2] with the ovarian salvage rate reported as low in most series. It is therefore essential to identify the features that rapidly point to a correct diagnosis, thus shortening the interval between onset of symptoms and operative detorsion.

Several studies have been published on this topic, but there are no large prospective series reported. This prompted us to perform a systematic literature review to identify and assess the most useful diagnostic tools.

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METHODS

Search Strategy

For this systematic review of the literature, we searched MEDLINE, EMBASE, and the Cochrane Databases for articles published between January 1990 and January 2014. We used the key words *ovarian torsion*, *acute pelvic pain*, and *adnexal torsion*, crossing them with *pediatric* and *children* to identify studies focusing on the pediatric population. There was no language restriction for the abstract search.

Studies Selection

Of the 946 publications initially identified, we first reviewed all titles and abstracts. Articles identified as relevant by title and/or abstract were all reviewed to exclude the following:

- Single case reports and single case discussions
- Studies that were not focused on the pediatric population (defined as birth to 21 years)
- Studies focusing only on management (surgical or hormonal)
- Unusable statistical data
- Studies based on a specific ovarian histopathology
- Full texts in languages other than English, French, Italian, Spanish, and German

We identified 14 studies[5–18] that provided data on children admitted in the ED with a suspicion of ovarian torsion, which documented history, physical examination findings, and/or investigations (laboratory and/or radiological), and confirmed or excluded a diagnosis of ovarian torsion by surgery. In 2 studies,[9,14] one ovarian torsion episode was diagnosed by radiology without surgical confirmation, as conservative management was chosen. We also reviewed all the publications cited in the bibliography of the 14 selected studies.

Data collected by a first investigator were then assessed independently by a reviewer. The quality of the included studies was evaluated through published guidelines. Results are presented in pooled total, calculated prevalence, and 95% confidence intervals (CIs) for differences between means.

Assessment of the Publications' Quality

The quality of the 14 selected studies was evaluated through the checklist of the STROBE [Strengthening the Reporting of Observational Studies in Epidemiology] statement (Table 1).[19] Published by von Elm et al in the *Lancet* in 2007, the STROBE statement provides guidelines for reporting observational studies. Although its aims and use are specifically developed for cohort, case-control, and cross-sectional studies, we were able to adapt its checklist to all 14 selected retrospective observational studies, giving an overall estimation of the publications' qualities. A majority of titles and abstracts provide informative summary and

TABLE 1. Publications' Quality Assessment Adapted From STROBE (von Elm et al, *Lancet* 2007)

		Anders et al[5]	Appelbaum et al[6]	Chang et al[7]	Galinier et al[8]	Kao et al[9]	Kokoska et al[10]	Meyer et al[11]	Oltman et al[12]	Piper et al[13]	Poonai et al[14]	Rossi et al[15]	Rousseau et al[16]	Servaes et al[17]	Stark and Siegel[18]
Title and abstract															
1a	Study design indicated	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Introduction															
3	Specific objectives stated	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	Yes	Yes	No	Yes	Yes
Methods															
5	Setting	Yes	No	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	No	No	No
5	Data collection mode	Yes	No	Yes	No	No	No	No	Yes	Yes	Yes	Yes	No	No	No
5	Follow-up	No	No	No	Yes	Yes	Yes	No	No	No	No	No	Yes	No	No
6a	Participants selection	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
6a	Control group	No	Yes	Yes	No	No	No	No	No	Yes	No	No	No	No	No
7	Outcomes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
8	Sources of data	Yes	No	Yes	No	Yes	No	No	Yes	Yes	Yes	Yes	No	Yes	No
9	Sources of bias	No	No	No	No	No	No	No	No	No	No	No	No	No	No
10	Study size	22	94	49	45	21	51	13	97	90	13	83	40	41	20
12a	Statistical methods described	yes	yes	yes	yes	yes	yes	No	yes	yes	yes	yes	No	yes	No
12c	Description of missing data	No	No	No	No	No	No	No	No	No	No	No	No	No	No
12e	Sensitivity analyses	No	no	no	no	no	no	no	no	yes	no	yes	no	yes	no
Results															
13a	No. individuals at each stage	yes	no	yes	yes	yes	no	no	yes	yes	yes	yes	yes	yes	no
13c	Flow diagram	no	no	yes	yes	no	no	no	no	yes	no	no	no	no	no
14b	No. participants with missing data	yes	no	yes	no	no	no	yes	no	no	yes	no	no	no	no
Discussion															
19	Limitations discussed	yes	yes	yes	no	yes	no	no	yes	no	yes	yes	no	yes	no
20	Cautious interpretation of results	yes	yes	yes	no	yes	no	no	yes	yes	yes	yes	no	yes	yes
21	Generalizability discussed	no	no	no	no	no	no	no	no	no	no	no	no	no	no
Other															
22	Funding/competing interest mentioned	no	no	no	no	no	no	no	no	no	yes	no	no	no	no
Total, 27		19	15	21	16	17	12	10	17	20	20	19	12	17	11

The following items are not described in the table because they were positive for all publications: 1b. Informative and balanced summary, 2. Scientific background and rationale, 4. Study design key elements, 5. Relevant dates, 7. Diagnostic criteria, 14a. Characteristics of participants, 18 Key results summarized.

TABLE 2. Publication Characteristics

Author	Year of Publication	Department	Country	Evidence Level	No. Torsion Episodes	Age	Patient Selection
Anders et al[5]	2005	Pediatrics	United States	III	22	3 to 15 y	Surgical diagnosis of adnexal torsion
Appelbaum et al[6]	2013	Gynecology	United States	III	45	4 to 18 y	Surgical diagnosis of adnexal torsion
Chang et al[7]	2008	Pediatrics	Taiwan	III	49	4 to 17 y	Surgical diagnosis of adnexal torsion
Galinier et al[8]	2009	Pediatric surgery	France	III	45	22 mo to 17 y	Surgical diagnosis of adnexal torsion
Kao et al[9]	2012	Pediatrics	Taiwan	III	21	7 to 18 y	Surgical or radiological diagnosis of adnexal torsion
Kokoska et al[10]	2000	Pediatric surgery	United States	III	51	8 to 16 y	Surgical diagnosis of adnexal torsion
Meyer et al[11]	1995	Radiology/ pediatric surgery	United States	III	13	1 d to 15 y	Surgical diagnosis of adnexal torsion
Oltman et al[12]	2009	Gynecology/ pediatric surgery	United States	III	97	2 d to 17 y	Surgical diagnosis of adnexal torsion
Piper et al[13]	2012	Pediatric surgery	Canada	III	90	1.7 to 19 y	Surgical diagnosis of adnexal torsion
Poonai et al[14]	2013	Pediatrics	United Kingdom	III	13	7 mo to 18 y	Surgical or radiological diagnosis of adnexal torsion
Rossi et al[15]	2012	Gynecology/ pediatric surgery	United States	III	83	3 to 21 y	Surgical diagnosis of adnexal torsion
Rousseau et al[16]	2008	Pediatric surgery	France/ Switzerland	III	40	3 to 14 y	Surgical diagnosis of adnexal torsion
Servaes et al[17]	2007	Radiology/ pediatric surgery	United States	III	74	1 mo to 21 y	Surgical diagnosis of adnexal torsion
Stark and Siegel[18]	1994	Radiology	United States	III	20	2 d to 16 y	Surgical diagnosis of adnexal torsion

clearly indicate the study design, but specific objectives were not always stated. Setting and data collection modes were missing in many studies. Only 3 studies included a control group, and outcomes were stated in all but one. Only 3 articles provided follow-up data. Study size ranged from 13 to 97 patients. Details of missing data were not specified in any study. Statistical methods were not always described, sensitivity analyses were included in 3 publications, and only 3 included a flow diagram. Key results were always summarized but limitations, cautious interpretation, and discussion of generalizability were often missing.

RESULTS

Description of the Studies and Settings

From the 946 articles initially identified, only 14 studies fulfilled our inclusion criteria and were used for this review (Table 2). Bibliographical references quoted in each of these 14 articles were also examined for suitability.

All included articles were retrospective studies. We found no prospective study. Published between 1994 and 2014, 8 studies were from the United States,[5,6,10–12,15,17,18] 1 from France,[8] 2 from Taiwan,[7,9] 1 from the United Kingdom,[14] and 1 from Canada.[13] A single study was multicenter (France, Switzerland).[16] All patients were first evaluated in a pediatric ED, and studies were conducted by pediatric surgeons in 8 series,[8,10–13,15–17] pediatricians in 4,[5,7,9,14] gynecologists in 3,[6,12,15] and radiologists in 3.[11,17,18] Four studies involved more than 1 department.[11,12,15,17]

The common inclusion criterion for all 14 studies is a documented surgical diagnosis of adnexal torsion. Two studies[9,14] also included 1 patient each who was diagnosed by radiology only and had a conservative management.

Description of Patients

All studies specifically included children, with age range from 1 day to 21 years, mean age being 11.6 years (Table 3). Nine studies excluded neonatal patients.[5–10,13,15,16] The total number of torsion episodes enrolled was 663 (median number, 47 per study, with range 13 to 97), involving 654 patients as 9 episodes were recurrences.[8,11,15,16] The proportion in premenarchal patients was 43.4% (95% CI, 36.3%–50.5%) and in postmenarchal patients was 56.6% (95% CI, 49.5%–63.7%). The overall incidence of adnexal torsion described in the pediatric ED was 0.5 to 2 per 10,000 patients.[9,13,15]

Clinical Data

Abdominal pain is, as expected, the commonest symptom, present in 97.5% (95% CI, 96%–99%), but with large reported ranges (65%–100%) (Table 3). Descriptions of location of pain differ between studies. Some use "lower abdomen," whereas others specify the side (right, left, bilateral). In studies specifying the side of pain, the lower right abdomen is the most common location (51%; 95% CI, 43.9%–58.1%), followed by lower abdomen with no side specified in 42.8% (95% CI, 29.8%–55.8%). Pain descriptions are highly variable. In some studies, it is reported it as sharp or sudden in 78.9 % (95% CI, 72.8%–85%) of

TABLE 3. Clinical Data

		Total	Calculated Prevalence	
		n/N	%	95% CI
Patients characteristics				
	Premenarchal	82/189	43.4	36.3–50.5
	Postmenarchal	107/189	56.6	49.5–63.7
	Previous similar symptoms	16/115	13.9	7.6–20.2
History				
Abdominal pain		425/436	97.5	96–99
Localization	Lower abdomen	24/56	42.8	29.8–55.8
	Lower right	97/190	51	43.9–58.1
	Lower left	51/177	28.8	22.1–35.5
Type	Sharp/sudden	135/171	78.9	72.8–85
	Severe	16/49	32.6	19.5–45.7
	Intermittent	78/171	45.6	43.2–48
	Constant	43/132	32.6	24.6–40.6
	Recurrent	7/49	14.3	4.5–24.1
	Unspecified	25/34	73.5	58.7–88.3
Vomiting		150/241	62.2	56.1–68.3
Nausea		56/83	67	56.9–77.1
Nausea and vomiting		70/137	51.1	42.7–59.5
Dysuria		11/121	9.1	4–14.2
Physical examination				
Abdominal tenderness		176/199	88.4	84–92.8
Guarding		20/96	20.8	12.7–28.9
Rebound		17/132	12.9	7.2–18.6
Palpable mass		50/248	20.2	15.2–25.2
Fever		42/208	20.1	14.7–25.5

the patients and severe in 32.6% (95% CI, 19.5%–45.7%). It is intermittent for 45.6% of the patients (95% CI, 43.2%–48%) and constant in 32.6% (95% CI, 24.6%–40.6%). It is however also unspecified in some studies for 73.5% (95% CI, 58.7%–88.3%) of the patients. Almost 14% of the patients had experienced similar symptoms in the past (95% CI, 7.6%–20.2%).

Other commonly reported symptoms were nausea (67%; 95% CI, 56.9%–77.1%), vomiting (62.2%; 95% CI, 56.1%–68.3%), or both (51.1%; 95% CI, 42.7%–59.5%). Less commonly reported were dysuria (9.1%; 95% CI, 4%–14.2%), periumbilical pain (7.7%), and back pain (3.2%).

On physical examination, abdominal tenderness was the main finding when mentioned (88.4%; 95% CI, 84%–92.8%). Peritoneal signs include guarding in 20.8% (95% CI, 12.7%–28.9%) and rebound in 12.9% (95% CI, 7.2%–18.6%). A palpable mass was present in only 20.2% (95% CI, 15.2%–25.2%), whereas a fever was seen in 20.1 % (95% CI, 14.7%–25.5%).

Investigations

Of the 663 episodes, 208 had blood tested for leucocytosis; a positive result (with positive limit varying between 10,000 and 12,000) was seen in 51.4%, but with a mean value of 11,860, which is not significant (range, 11,120–13,000) (Table 4). In 1 study, patients had a C-reactive protein tested, with a mean value of 34.7 mg/L (no range and normal limits documented). Thirty-one patients had their urine tested (dipstick and/or urine culture) with 25.85% testing positive for blood (95% CI, 10.4%–41.2%) and 29% positive for leucocytes (13%–45%). However, no urine cultures performed (26 of 31) were positive.

Plain abdominal radiograph was used in several centers. Of 438 patients, 45 had an abdominal radiograph (10.3%) documented in the publications. Abnormal findings described[7,8,10,11,14,16] included 3 soft tissue radio-opaque masses, 2 masses, 6 foci of calcification, 8 ossified images (all teratoma), and 7 nonspecified abnormalities. Abdominal ultrasound (US) was performed in 409 (73.4%) of 557 episodes. In studies specifying US findings, the sensitivity was 79% (95% CI, 73.7%–84.3%). A unilateral augmentation in the size of the ovary is often found (71.3%; 95% CI, 64.7%–77.9%), but measurement values vary between studies.[18,20–22] Ovarian volume depends on age and pubertal status; some authors use a volume ratio, based on comparison to the contralateral ovary.[17] Abdominal or pelvic masses were also documented by US scan. This was described as complex (not further specified) in 53.6% (95% CI, 45.6%–61.6%), cystic in 27% (95% CI, 20.8%–33.2%), solid in 15.1% (95% CI, 8.3%–21.9%), and calcified in 2.4%. Doppler studies confirmed vascular flow in 64.6% (95% CI, 51.1%–78.1%). However, as described elsewhere, normal Doppler evaluation result does not exclude torsion.[18,20,21] Pelvic free fluid was noticed in 36.8% (95% CI, 27.6%–46%). Computerized tomography (CT) was used in 158 (40.8%) of 387 patients, and in studies specifying the results, we found an overall sensitivity of 42.2% (95% CI, 31.6%–52.8%). Abnormalities described on CT results include 3 heterogeneous masses, 1 focus of calcification, and 2 cysts.[7,11,14]

Treatment and Outcome

The time between first physical examination and surgery varies greatly between studies (0–90 days), with a median of

TABLE 4. Investigations

	Total	Calculated Prevalence	
	n/N	%	95% CI
Blood test			
Leukocytosis	107/208	51.4	44.6 to 58.2
Urine test			
Blood +	8/31	25.8	10.4 to 41.2
Leukocyte +	9/31	29	13 to 45
Urine culture	0/26	0	—
Radiology			
Plain abdominal x-ray documented	45/438	10.3	7.5 to 13.1
Abnormal x-ray	26/45	42.2	27.8 to 56.6
US done	409/557	73.4	69.7 to 77.1
US positive	177/224	79	73.7 to 84.3
Increased ovarian size	127/178	71.3	64.7 to 77.9
Complex mass	81/151	53.6	45.6 to 61.6
Cystic mass	53/196	27	20.8 to 33.2
Solid mass	16/106	15.1	8.3 to 21.9
Calcified mass	1/41	2.4	−2.3 to 7.1
Doppler flow present	31/48	64.6	51.1 to 78.1
Doppler flow absent	17/48	35.4	21.9 to 48.9
Free pelvic fluid	39/106	36.8	27.6 to 46
CT scan done	158/387	40.8	35.9 to 45.7
CT scan positive	35/83	42.2	31.6 to 52.8

101 hours (Table 5). The ovary was salvaged in 32.1% (95% CI, 27.9%–36.3%), whereas the oophorectomy rate was 67.9% (95% CI, 63.7%–72.1%) and 54.2% of the patients required a salpingectomy (95% CI, 48.4%–60%). In publications where the question was documented, 13.9% of the patients had experienced similar symptoms in the past (95% CI, 7.6%–20.2%), but we could not extract a correlation between time to diagnosis or recurrent symptoms and ovary salvage rates.

Surgical and Pathology Findings

In studies specifying the surgical findings, a right-sided torsion is more common (60%; 95% CI, 53.2%–66.8%) than a left-sided torsion (40%; 95% CI, 33.2%–46.8%). Ovarian cysts were found in 29.9% (95% CI, 24.2%–35.6%) and paratubal cysts in 13% (95% CI, 7.9%–18.1%), and 47.5% had previously normal ovarian anatomy (95% CI, 35%–60%). An underlying tumoral pathology was described in 2.8% (95% CI, 1.4%–6%).

Pooling the Results

All studies having a limited number of patients, it would have been very interesting to pool the results. This would have helped identify the most relevant clinical, laboratory, and radiological signs for a prompt diagnosis of adnexal torsion. It was however impossible because most studies used different clinical complaint definitions and signs descriptions, different laboratory limits, and different US volume guidelines.

DISCUSSION

After reviewing the literature from the last 24 years, we only identified 14 studies that fulfilled our inclusion criteria. Except for

TABLE 5. Diagnosis and Outcomes

	Total	Calculated Prevalence	
	n/N	%	95% CI
Surgical findings			
Right-sided torsion	120/200	60	53.2–66.8
Left-sided torsion	80/200	40	33.2–46.8
Ovarian cysts	75/251	29,9	24.2–35.6
Paratubal cysts	22/169	13	7.9–18.1
Torsion of a normal ovary	29/61	47,5	35–60
Outcome			
Tumoral disease	9/324	2,8	1v4.6
Ovary salvaged	152/474	32.1	27.9–36.3
Oophorectomy	322/474	67.9	63.7–72.1
Salpingectomy	155/286	54.2	48.4–60

rare neonatal cases included in 3 studies, publications were based on ED visits of pediatric patients. In all studies, data were collected retrospectively after a surgical and/or radiological diagnosis of adnexal torsion. No prospective studies that were specifically based on a pediatric population were found.

Prevalence according to patient's age shows that ovarian torsion is not only a postmenarchal occurrence. However, because of lack of data, we could not identify a peak age.

The large differences in definition for clinical complaints and for physical examination signs between studies made it impossible to pool results and identify true relevant elements to assist in prompt clinical diagnosis. We could only identify trends because of a high discrepancy between publications.

Abdominal pain is the most common presentation. The commonest site is the lower abdomen, whether right, left, bilateral, or unspecified. Sharp, sudden onset pain is often described by the patients, but it is not quoted in all studies, and some only describe the pain as severe. The characteristics of the pain can be constant, intermittent, as well as recurrent or unspecified. The most common accompanying complaints are vomiting and/or nausea.

On physical examination, abdominal tenderness is the most relevant sign; some studies describe guarding or rebound, with some studies not mentioning the presence or the absence of peritoneal signs. Physical examination description is incomplete in many studies.

Laboratory tests, when used,[5–7,9–11,14] do not seem to aid the diagnostic process. Based on a predefined population having intraoperative confirmation of adnexal torsion and with no control population, sensitivity and specificity of laboratory tests are impossible to extract.

Although plain abdominal radiograph is still used by many, the most commonly used and most useful radiological investigation is ultrasound (79% sensitivity), followed by CT scan (42.2% sensitivity). An augmentation in the ovarian size, a complex mass, and free fluid can be signs of adnexal torsion. The presence of vascular flow on Doppler studies does not rule out torsion, but we could not identify any correlation between blood flow and ovarian outcome.

CONCLUSIONS

Implications for Practice

Because of few specific clinical signs, an early diagnosis of adnexal torsion remains difficult. The aims of our literature review were to identify reliable predictive signs to achieve a prompt diagnosis in the pediatric population and to develop an algorithm for the management of patients with suspected adnexal torsion, based on evidence-based medicine. This was, however impossible because of the many limitations in the literature review. All studies were retrospective and based on a predefined population. Most series were small, and the definitions used varied greatly. There was a large discrepancy in ranges, and data were often impossible to pool. Acute abdominal pain remains the major presenting symptom of adnexal torsion but is nonspecific and a common complaint in the ED. The diagnosis of adnexal torsion should be considered in any female pediatric patient presenting to the ED with sudden onset lower abdominal pain.

Implications for Research

A large, prospective, multicenter study, in which definitions of symptoms, clinical signs, and investigations are precisely predefined and quoted in a standardized questionnaire, is needed to further investigate this.

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