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GENERAL GYNECOLOGY

Clinical characteristics of adnexal torsion in premenarchal patients

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Abstract
Purpose To compare clinical characteristics of adnexal torsion in premenarchal patients as compared to postmenarchal ones.
Methods A retrospective 22-year cohort of all cases of surgically verified adnexal torsion in premenarchal and postmenarchal patients (excluding postmenopausal and pregnant patients). Data collected included symptoms, signs and imaging at presentation, surgical mode, and procedure and histology.
Results 16 cases of adnexal torsion among premenarchal patients were compared to 302 cases among postmenarchal ones. Clinical presentation was similar, notable only for more nausea and vomiting among the premenarchal group. Ovarian cysts were more commonly demonstrated among postmenarchal patients (82.4 and 30.8 %, $p < 0.001$), as opposed to a normal adnexal appearance on ultrasound, which was more common in premenarchal patients (69.3 vs. 17.2 %, $p < 0.001$). Absent Doppler flow, when preformed, did not differ statistically between the groups. Laparoscopic detorsion only was more commonly performed in premenarchal patients (56.2 vs. 19.8 %, $p = 0.001$), with a trend for detorsion and cystectomy/fenestration, which were more common in postmenarchal surgeries (25 vs. 50.6 %, $p = 0.06$). Histology was similar among both groups, with no cases of malignancy in premenarchal patients.
Conclusion Adnexal torsion in premenarchal patients is rare, presents similarly to older patients, but involves a normal adnexa in 69 % of cases, therefore requiring a high index of suspicion in any premenarchal patient with acute onset abdominal pain.

Keywords Adnexa · Torsion · Premenarchal · Laparoscopy

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Introduction

Ovarian torsion refers to the complete or partial rotation of the ovary, typically around both the infundibulopelvic and utero-ovarian ligaments. If the fallopian tube twists as well, as often occurs, it is referred to as adnexal torsion. Isolated torsion of the fallopian tube may also occur [1], as well as paraovarian [2] or paratubal [3] cyst torsion. Ovarian torsion was found in 2.5–7.4 % of adult patients undergoing surgery for acute pelvic pain [4, 5], with an estimated annual prevalence of 2–6 % [5]. However, occurrence in childhood is rare, and the estimated annual prevalence

among females aged 1–20 years is only 4.9 per 100,000 [6]. Although ovarian torsion is one of the most common gynecologic emergencies among reproductive aged women [7], it must also be considered in every young girl presenting with acute onset abdominal pain [8], as blood supply to the ovary is often compromised as a result of the torsion, and prompt diagnosis and intervention are required to preserve ovarian function and prevent additional morbidity [9].

Few reports of adnexal torsion have focused on the premenarchal girl [33, 34] as opposed to the adolescent or adult patient, and there are less data regarding the specific characteristics of adnexal torsion in this group of patients as compared to reproductive age women. The aim of our study was to deliver a comprehensive comparison of adnexal torsion in the two groups, including clinical characteristics, diagnostic process, surgical approach, and histology.

Materials and methods

Our study is a retrospective cohort study of patients with surgically verified adnexal torsion, treated in the Edith Wolfson Medical Center between March 1990 and October 2012. The study was approved by the institutional Helsinki Committee. Patient charts with an ICD9-coded diagnosis of adnexal torsion were retrieved and reviewed for data, including patient demographics, menarchal status, medical history, clinical and laboratory presentation and imaging at admission, surgical findings and procedure, and histological results. Torsion was defined as a rotation of the ovary/adnexa of at least 360 degrees. Reproductive aged controls were excluded if pregnant or postmenopausal.

Continuous variables were calculated as mean ± SD or median and range as appropriate, and compared with the use of Student *t*-tests. Categorical variables were calculated as rate (%) and compared using the Chi-square test or Fisher's exact test as appropriate. A p value of <0.05 was considered statistically significant.

Results

353 operations were performed for adnexal torsion during this 22-year period in non-pregnant women. 35 of these were performed in postmenopausal patients and therefore excluded. Of the remaining operations, 16 were performed in premenarchal patients and 302 in postmenarchal (reproductive age) ones, all meeting the inclusion criteria.

Patient presentation, including clinical, imaging, and laboratory findings, are noted in Table 1. As anticipated, the premenarchal group consisted of younger patients, with an average age of 9.5 as compared to 32.1 ($p < 0.001$).

The clinical presentation was similar for both groups, including similar rates of fever, abdominal pain, and peritoneal signs, although nausea and vomiting were more commonly noted among premenarchal patients (Table 1). Right-sided presentation was similarly noted in both groups (56.2 and 62.9 %). Patient history revealed a similar duration of pain prior to presentation in both groups (26.4 and 26.6 h, $p = 0.97$). Cases of recurrent torsion were similarly prevalent in both groups (6.2 and 3.9 %). The average ovarian diameters upon ultrasound examination at admission among premenarchal and postmenarchal patients were 4.8 and 6.8 cm, respectively ($p = 0.01$). Ovarian cysts were more commonly found among postmenarchal patients (82.4 and 30.8 %, $p < 0.001$), as opposed to a normal adnexal appearance, which was more common in premenarchal patients (69.3 vs. 17.2 %, $p < 0.001$). Pathological Doppler flow defined as Absent Doppler flow, when preformed, did not differ statistically between the groups. No differences were observed in the leukocyte count between study groups (Table 1).

The admission to surgical interval noted in our study did not differ substantially between the groups, with an average interval of 15.9 h in the premenarchal group, as compared to 24.4 in the postmenarchal one ($p = 0.45$) (Table 2). The main surgical indication for both pre- and postmenarchal patients, as noted by the performing surgeon, was suspected ovarian torsion (68.8 and 77.1 %, respectively, $p = 0.54$), although a trend towards a preoperative suspicion of appendicitis was noted among premenarchal patients.

Table 2 refers to surgical characteristics and intraoperative findings. Similar rates of laparoscopy were noted among both groups. Total adnexal torsion (of both ovary and fallopian tube) was similarly noted in both groups by the performing surgeon, although there was a tendency for cyanotic adnexal appearance in the premenarchal group (93.8 vs. 73.3 %, $p = 0.07$), which persisted among these premenarchal patients post-detorsion, as assessed by the attending surgeon during operation based on pale bluish appearance of the tissue, consistent with ischemia (23.1 vs. 5.6 %, $p = 0.03$). Detorsion only was more commonly performed in premenarchal patients (56.2 vs. 19.8 %, $p = 0.001$), with a trend for detorsion and cystectomy/fenestration, which were more common in postmenarchal surgeries (25 vs. 50.6 %, $p = 0.06$). Rates of unilateral salpingo-oophorectomy were similar for both groups, with a minority of postmenarchal patients undergoing more radical surgery such as bilateral salpingo-oophorectomy (BSO) (as detailed in the discussion). No "second look" laparoscopies were performed during the study period.

Table 1 Presenting symptoms and signs among pre- and postmenarchal patients with adnexal torsion

Characteristic	Premenarchal ($n = 16$)	Postmenarchal ($n = 302$)	p value
Demographics			
Age (years)[a]	9.5 ± 4.1	32.1 ± 14.9	<0.001
Recurrent torsion	1 (6.2 %)	12 (3.9 %)	0.49
Signs and symptoms			
Fever >38 °C (%)	1 (6.2 %)	23 (7.7 %)	1.0
Nausea and vomiting (%)	13 (81.2 %)	141 (46.7 %)	0.008
Abdominal pain (%)	16 (100 %)	260 (87 %)	0.23
Peritoneal signs—rebound (%)	7 (46.6 %)	82 (27.4 %)	0.13
Right side (%)	9 (56.2 %)	190 (62.9 %)	0.6
Duration of pain (hours)[a]	26.4 ± 23.0	26.7 ± 33.7	0.97
Ultrasound findings			
Ovarian diameter (centimeters)[a]	4.8 ± 1.4	6.8 ± 2.6	0.01
Normal appearance (%)	9/13 (69.3 %)	48/279 (17.2 %)	<0.001
Cyst (clear/echogenic) (%)	4/13 (30.8 %)	230/279 (82.4 %)	<0.001
Absent flow on Doppler (%)	3/4 (75 %)	41/94 (43 %)	0.32
Laboratory findings			
WBC ($_{cells/\mu L}$)[a]	9.4 ± 3.1	10.5 ± 3.4	0.2

[a] Data presented as mean ± standard deviation

Table 2 Operative characteristics

Characteristic	Premenarchal ($n = 16$)	Postmenarchal ($n = 302$)	p value
Admission to surgery interval (hours)[a]	15.9 ± 30.6	24.4 ± 40.6	0.46
Main surgical indication			
Suspected torsion (%)	11 (68.8 %)	232 (77.1 %)	0.54
Suspected appendicitis (%)	3 (18.8 %)	21 (7 %)	0.11
Duration of surgery (minutes)[a]	48.5 ± 28.3	60.2 ± 31.9	0.16
Surgical mode—laparoscopy (%)	11 (68.8 %)	230 (76.4 %)	0.54
Intraoperative findings			
Torsion of entire adnexa (ovary and tube) (%)	14 (87.5 %)	240 (80 %)	0.74
Cyanotic appearance of ovary (%)	15 (93.8 %)	217 (73.3 %)	0.07
Cyanotic appearance post-detorsion (%)	3 (23.1 %)	13 (5.6 %)	0.03
Surgical procedure			
Detorsion only (%)	9 (56.2 %)	60 (19.8 %)	0.001
Detorsion + cystectomy/fenestration (%)	4 (25 %)	153 (50.6 %)	0.06
Unilateral salpingo-oophorectomy (%)	3 (18.7 %)	30 (10 %)	0.22
Bilateral salpingo-oophorectomy ± TAH (%)	0 (0 %)	46 (15.3 %)	0.14
Salpingectomy (%)	0 (0 %)	11 (3.6 %)	1.0
Oophoropexy	1 (6.2 %)	10 (3.3 %)	0.43
Post-surgical course			
Hospitalization (days)[a]	3.1 ± 2.2	3.7 ± 3.4	0.48

TAH total abdominal hysterectomy

[a] Data presented as mean ± standard deviation

Finally, a histological sample was reviewed in 6 cases of premenarchal adnexal torsion (Table 3). Necrosis only with no additional histological finding of malignancy was noted in 3 of these patients, a rate significantly higher than that in the postmenarchal group (50 vs. 13 %, $p = 0.03$). The remaining histological finding among the other 3

Table 3 Histological findings

Characteristic	Premenarchal ($n = 6$)	Postmenarchal ($n = 208$)	p value
Cyst adenoma (mucinous, serous) (%)	1 (16.6 %)	45 (21.6 %)	0.2
Functional cyst (corpus luteum, follicular) (%)	1 (16.6 %)	46 (22.1 %)	0.2
Paraovarian (%)	1 (16.6 %)	37 (17.8 %)	0.31
Mature teratoma (dermoid) (%)	0 (0 %)	32 (15.4 %)	0.13
Necrosis only (%)	3 (50 %)	27 (13 %)	0.03
Other—hydrosalpinx, fibroma (%)	0 (0 %)	15 (7.2 %)	0.6
Cancer (%)	0 (0 %)	5 (2.4 %)	1.0

premenarchal patients were serous, functional, and paraovarian cysts, at rates similar to that of postmenarchal patients. Cancer was diagnosed in 2.4 % of postmenarchal patients with adnexal torsion, with no cases noted among premenarchal patients.

Discussion

Adnexal torsion represents a diagnostic challenge which requires prompt intervention, with the intent of ovarian tissue salvage and fertility preservation. It is especially uncommon in premenarchal girls, in whom the diagnosis may be elusive.

Ovarian cysts or neoplasm predispose to ovarian torsion as directly related to the mass size, unless the mass is relatively fixed (due to size or adhesions) or malignant [10]. Other risk factors described include a past history of adnexal torsion, polycystic ovarian syndrome, ovarian hyperstimulation syndrome, ovulation induction, tubal ligation, and pregnancy [11]. However, ovarian torsion may often occur in patients with no such findings [12], as is more common in premenarchal girls [13, 34]. This may be explained by an elongated utero-ovarian ligament noted in premenarchal patients [14, 15], increased venous congestion in the premenarchal period, or certain strenuous activity involving an intra-abdominal rise in pressure [14, 16].

Although torsion may occur bilaterally, there seems to be a predisposition for right-sided torsion both in pre- and postmenarchal patients [16–19], possibly explained by a longer right utero-ovarian ligament or proximity of the left ovary to the sigmoid colon. Symptoms and signs of adnexal torsion include acute onset abdominal pain, nausea, vomiting, and even peritonitis and fever [8, 16, 20], though non-specific findings upon physical examination [8, 16, 20] and laboratory results [8, 12, 16, 20, 21] make this a challenging diagnosis among all age groups. Pelvic ultrasonography has also shown partial success in diagnosing ovarian torsion in reproductive age women [21–23], with uncertain predictive value of Doppler flow [24]. This has also been demonstrated in premenarchal patients [8, 14, 16]. Eventually, adnexal torsion is often confirmed intraoperatively [25–27], when a conservative approach is the preferred choice of treatment [27–29], as determined by patient age, ovarian findings, and the possibility for malignancy, also considering recurrence rates for each treatment option [12, 19, 30].

The role of oophoropexy in the treatment of ovarian torsion remains unestablished [14, 31, 32]. There is a general concern for reduced vascularization to the ovary and future infertility, balanced by the higher recurrence rates of torsion in the pediatric population, which themselves endanger the ovary, and may possibly be prevented with oophoropexy. A study by Puchs et al. [35] of 7 cases whom underwent oophoropexy demonstrated recurrence in one case, with all cases followed notable for spontaneous menstruation and normal ovarian volume on follow-up. An additional report by Poonai and colleagues also supports this notion [36]. Due to the limited number of cases in reports to date, further investigations are needed in order to establish a routine treatment algorithm. In addition, different pharmacological treatments are being investigated as adjuvant means against ovarian ischemia–reperfusion injury in animal models [37, 38], none of which are in clinical use yet.

Few reports to date have investigated the specific characteristics of adnexal torsion in premenarchal patients. Tsafrir et al. [34] previously presented a series of 22 cases in 20 premenarchal patients, 3 of which involved recurrent torsion. The main presenting symptoms were pain and vomiting, with pre-surgical evaluation notable for abnormal Doppler examination, when preformed. Laparoscopic approach was the preferred mode in most cases in which detorsion and cystectomy were commonly performed. These results were not compared to any control group.

In our study comparing adnexal torsion in premenarchal patients to postmenarchal ones, we demonstrate a similar clinical presentation of similar duration prior to admission, with premenarchal patients exhibiting more nausea and vomiting. On ultrasound examination, cystic findings were more commonly demonstrated in postmenarchal patients,

in contrast to premenarchal ovaries, which were mostly normal appearing (82.4 vs. 30.8 %, $p < 0.001$). This further supports the notion [12, 13, 33] of a tendency for adnexal torsion with no apparent abnormal anatomical finding in premenarchal patients, possibly due to an elongated utero-ovarian ligament as discussed above. These premenarchal patients also exhibited edematous ovaries per imaging (38.5 vs. 12.5 %, $p = 0.02$). This may be related either to the adnexal torsion itself or possibly to physiological venous congestion during these years [14, 16], prone to the insult caused by twisting of the adnexa. This notion is further supported by a trend for cyanotic appearing adnexas in the premenarchal group and significant post-detorsion cyanosis during surgery. In contrast to previous studies, we did not demonstrate a significant difference in the rate of absent doppler flow between the groups, although this may very well be underpowered for such a distinction, seeing as Doppler imaging was performed in only 4 premenarchal patients. The admission to surgery interval was similar in both groups, reflecting similar concern regarding ovarian preservation and risk of future infertility as mandated. Cases of recurrent torsion were noted in 6.2 % of premenarchal and 3.9 % of postmenarchal patients ($p = 0.49$). Seeing as recurrence rate has been demonstrated to be higher for normal adnexas [12] as more common in premenarchal patients, this difference may be underpowered due to sample size.

The most common surgical indication was suspected adnexal torsion for both groups, although the premenarchal group demonstrated a slight tendency for suspected appendicitis. Indeed, a review of these surgical reports indicates that the initial operating surgeon in 8 of 16 premenarchal surgeries was a general pediatric surgeon rather than a gynecologist.

The main surgical mode for both groups was laparoscopy. Premenarchal patients underwent detorsion only at a significantly higher rate (56.2 vs. 19.8 %, $p = 0.001$), with a tendency of postmenarchal patients for detorsion with cystectomy/fenestration. This, again, reflects the underlying cystic findings associated with adnexal torsion in postmenarchal patients as opposed to premenarchal ones. While reviewing the 5 laparotomies performed in premenarchal patients, 2 were performed up to the year 1993, one was combined with hernioplasty and another one began in a laparoscopic approach and was converted to open surgery due to technical considerations. All 3 cases of unilateral salpingo-oophorectomy among premenarchal patients were performed up to the year 1994, and a review of the respective surgical reports indicated severe necrosis of the affected adnexa. More radical surgery performed in postmenarchal controls may be explained by the long study period in which surgical trends towards laparoscopic approach and minimal intervention have evolved. These postmenarchal controls may have also elected a more radical approach if approaching menopause, as more commonly offered in the beginning of the study period, or as a solution for additional comorbidities, such as a myomatic uterus. Indeed, 60 % of the 72 laparotomies performed in postmenarchal women occurred prior to the year 1996, and of the 29 preformed afterwards, 12 included an additional procedure (such as total abdominal hysterectomy or hernioplasty) and 5 additional surgeries began in a laparoscopic approach and were later converted to laparotomy. In addition, all surgeries for BSO and total abdominal hysterectomy (TAH) in the postmenarchal group were performed in women over their forties, 9 of which were performed up to the year 1997.

Comparison of additional histological results is limited due the small number of samples from premenarchal patients (6). We demonstrated a malignancy rate of 2.4 % among postmenarchal patients investigated, with no cases observed in the premenarchal group. This was not proven statistically significant, due to the overall low rate in both groups. A histological finding of necrosis was demonstrated in 3 of 6 premenarchal patients with no additional pathology. In the 3 other premenarchal patients, cysts were present, which were similar to those of postmenarchal patients. Functional cysts were the most common type in postmenarchal patients according to histological examination (22.1 %). However, this is probably an underestimation, seeing as a histological examination was not preformed in all patients, and we can assume that in cases of clear and simple appearing cysts, the performing surgeon elected to perform fenestration and waive pathological examination (as performed in 30 cases). Therefore, although the prevalence of histologically confirmed functional cysts did not differ statistically due to limited sample size, we believe they are probably more prevalent in postmenarchal patients.

Our study is limited by its retrospective nature, including the potential for underreporting and the limitation of lack of a uniform diagnostic process (such as Doppler imaging for all patients). In addition, the study may be subjected to selection bias resulting from inclusion of patients who were referred to surgery, as opposed to those whom received conservative treatment. However, it has the advantage of a rather large control cohort in comparison to the scarce previous series published to date on the subject and makes use of our ICD coded database in patients with proven intraoperative finding. Nevertheless, we suggest further research on the topic be performed to determine exact rates and specific guidelines.

In conclusion, our retrospective study of adnexal torsion among premenarchal patients as compared to postmenarchal ones depicts a picture of similar clinical presentation, with 69 % of cases of torsion in premenarchal patients

involving normal adnexas, and in whom detorsion, only via laparoscopy was therefore the preferred mode of treatment. Abdominal pain in a premenarchal patient requires a high index of suspicion due to lack of predisposing ultrasound findings or specific markers of ovarian torsion, and mandates urgent diagnosis and treatment.

Compliance with ethical standards

Conflict of interest None.

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