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Adnexal torsion with paraovarian cysts in pediatric and adolescent populations: A retrospective study☆

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ABSTRACT

Background: Determination of the clinical characteristics associated with adnexal torsion involving paraovarian cysts in pediatric and adolescent populations.
Methods: Retrospective review of all cases of paraovarian cysts operated on in our department between 2007 and 2019. Demographic characteristics, clinical and sonographic findings were reviewed.
Results: The cohort was composed of 39 pediatric and adolescent patients with an operative diagnosis of adnexal masses located in the paraovarian area. The patients were classified into two groups: 19 girls (48.7%) with a confirmed operative diagnosis of adnexal torsion and 20 girls (51.3%) without torsion. The preoperative diagnosis of adnexal torsion was correct in ≈70% of the cases. The mean BMI were similar in both groups. The preoperative sonographic detection rate of paraovarian cysts was also similar (11/19 [57.9%] vs. 14/20 [70.0%]; P = 0.514). The mean cyst diameter did not differ between groups, nor did the classification into cyst size groups (≤50 mm, 51–99 mm and ≥100 mm).
Conclusion: Adnexal torsion is a common complication diagnosed in girls undergoing surgery for paraovarian cysts, and is not associated with sonographic appearance or cyst diameter. In order to prevent torsion, surgical removal of paraovarian cysts should thus be considered in young girls undergoing surgery for paraovarian cysts.
Type of study: Retrospective case series.
Level of evidence: IV

Paraovarian cysts are found in the broad ligament between the ovary and the fallopian tube [1,2]. They may be either nonneoplastic simple cysts or cysts of neoplastic origin [1,3,4]. The true epidemiology of these cysts is difficult to evaluate because they are frequently diagnosed in asymptomatic patients. They constitute 3% to 20% of all adnexal masses and have been reported in all age groups, from the premenarcheal period to menopause [5,6].

Paraovarian cysts are also diagnosed in pediatric and adolescent populations evaluated for pelvic pain. It has been hypothesized that the size of these cysts is hormonally regulated at the onset of menarche, which is indicative of the maturation of the hypothalamic pituitary gonadal axis [7,8], and can lead to a large and clinically significant adnexal pathology. Some authors assume that as the mass increases in size, the risk of adnexal torsion also increases. Adnexal torsion involving paraovarian cysts may cause acute abdominal pain necessitating urgent surgery for detorsion and cyst removal to avoid ischemic damage to the ovary, especially in pediatric and adolescent patients [9,10]. Thus, because of the lack of spontaneous resolution given its origin in the mesonephric duct [3,4] and the risk of torsion, elective surgery for removal of paraovarian cysts has been recommended in pediatric and adolescent populations [10]. However, these recommendations are largely based on small cases series [10] and there appear to be no studies that have attempted to evaluate the prevalence of paraovarian cysts in association with torsion in pediatric and adolescent populations. In addition, there is scant if no literature comparing the clinical presentation of torsion and its ultrasonic and surgical characteristics in young patients with and without torsion of paraovarian cysts. In response to this need, the purpose of this study was to describe the demographic, clinical and surgical characteristics of paraovarian cysts causing adnexal torsion in pediatric and adolescent populations, and to compare these parameters in young girls with and without torsion of paraovarian cysts.

1. Materials and methods

A computerized database search was conducted for pediatric and adolescent patients (<19 years old) with an operative diagnosis of paraovarian cyst(s) who underwent surgery in our department between 2007 and 2019. The database was created a priori to include cases with an intraoperative diagnosis of paraovarian cyst regardless of the suspected preoperative diagnosis, and not as a secondary database using an existing

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adnexal cysts database. The World Health Organization's upper age cutoff for adolescence (age 19) was used. The reasons for surgery were a pelvic mass incidentally found by sonography as part of an abdominal pain workup, or suspected adnexal torsion. Data relating to demographic characteristics, clinical presentation, preoperative ultrasound scans, operative reports, and pathological diagnoses were retrospectively retrieved from the computerized medical records and reviewed. While information on menarcheal status (i.e., pre- or postmenarche) was available in all cases, the Tanner stage and the thelarche were not systematically reported in the patients' records.

After reviewing the medical records, the patients were classified into two groups. The first group was made up of cases with a surgical diagnosis of adnexal torsion of a paraovarian cyst. The second group was composed of cases without a surgical diagnosis of adnexal torsion of a paraovarian cyst (where surgery was conducted because of a pelvic mass identified incidentally by sonography as part of an abdominal pain workup, or cases with a suspected preoperative diagnosis of adnexal torsion that was not confirmed during surgery).

The final classification was restricted to cysts in a paraovarian location whose removal did not involve the dissection of the ovarian capsule (i.e., cysts that were clearly paraovarian as opposed to ovarian). The paraovarian masses were divided into benign epithelial paraovarian cysts and benign neoplastic paraovarian cysts, based on the pathological findings.

The preoperative ultrasound examinations were performed in three different locations in our department (gynecological emergency room, gynecology department and gynecological ultrasound unit) using different ultrasound machines (Voluson Expert, Voluson E8 and E10 and Logic V5, all manufactured by GE Healthcare, Milwaukee, WI, USA). All the ultrasound machines were equipped with a transvaginal probe (5–7.5 MHz, with a focal range of 6 cm from the transducer tip) and a transabdominal probe (3.5–5 MHz). The transabdominal examination was used mainly for virgins and was performed using the patient's full bladder as an acoustic window. Scans were performed by board-certified gynecologists and by physicians in training (residents) with varying levels of ultrasound expertise. Thus, color Doppler flow examinations were only available for a minority of patients and hence were not included in the study. Because our sonographers routinely measure two cyst diameters rather than three, we could not calculate cyst volume and instead used the largest cyst diameter as a measure of cyst enlargement.

The decisions as to surgical approach (i.e., laparoscopy or laparotomy) and procedures (detorsion, drainage and suction of the cyst, paraovarian cyst removal – cystectomy – or salpingo-oophorectomy) were at the discretion of the attending surgeon managing the case. All specimens obtained during cystectomy or salpingo-oophorectomy procedures were sent for pathological evaluation.

Statistical analyses implemented SPSS software (SPSS Inc., version 24 Chicago, IL, USA). The descriptive variables are presented as the mean ± the standard deviation. Frequencies are presented as percentages. One-way ANOVAs and Fisher's Exact tests were employed as appropriate. A p value of less than 0.05 was considered statistically significant. This study was approved by the Institutional Review Board of the hospital (#220/13; approval 06-01-2013).

2. Results

The cohort was composed of 39 pediatric and adolescent patients with an operative diagnosis of adnexal masses located in the paraovarian area. For the whole cohort, the median paraovarian cyst diameter for the whole cohort was 65 mm (range 40 mm–220 mm). The mean body mass index (BMI) was 26.8 kg/m^2. There was no correlation between BMI and the size of the paraovarian cyst ($r^2 = 0.087, P = 0.08$).

All patients were postmenarcheal. Of these, 27 (69.2%) patients underwent surgery for suspected adnexal torsion. The preoperative diagnosis of adnexal torsion was correct in 19/27 (70.4%) of the cases. The patients were classified into two groups: 19 (48.7%) patients with a confirmed operative diagnosis of adnexal torsion and 20 (51.3%) patients who underwent surgery because of a pelvic mass found by sonography ($n = 4$), as part of an abdominal pain workup ($n = 8$) or patients with the suspected preoperative diagnosis of adnexal torsion that was not confirmed during the operation ($n = 8$).

Table 1
Comparison of demographic and clinical characteristics of pediatric and adolescent patients with and without a surgical diagnosis of adnexal torsion of a paraovarian cyst.

Parameter	Torsion ($n = 19$)	No torsion ($n = 20$)	P value
Age (years)	15.4 ± 2.8	16.1 ± 2.0	0.351[a]
BMI (kg/m^2)	27.1 ± 7.4	26.9 ± 7.5	0.934[a]
Clinical presentation			
Abdominal pain	19 (100.0)	16 (80.0)	0.106[b]
Right lower quadrant pain	9 (47.4)	2 (12.5)	
Left lower quadrant pain	6 (31.6)	3 (18.8)	0.05[c]
Diffuse pain	4 (21.0)	11 (68.7)	
Nausea and/or vomiting	11 (57.9)	6 (30)	0.202[b]

Data are presented as the number (%) or as the mean ± standard deviation.
[a] Two tailed t-test.
[b] Fisher's Exact test.
[c] Pearson's chi-square test.

The demographic and clinical characteristics in the two groups are compared and summarized in Table 1. The mean age and BMI were similar in both groups and abdominal pain was the main presenting symptom. Torsion was significantly less common in girls who complained of diffuse abdominal pain. Nausea and/or vomiting was common in both groups.

Table 2 compares the preoperative ultrasound findings for the pediatric and adolescent patients with and without a surgical diagnosis of adnexal torsion of paraovarian cyst. Most sonographic examinations in both groups were conducted abdominally since the patients were young adolescent virgins. The preoperative sonographic detection rate of paraovarian cysts was similar in both groups (11/19 [57.9%] vs. 14/20 [70.0%]; $P = 0.514$). The mean cyst diameter was not different between groups, nor was the classification into cyst size groups (≤50 mm, 51–99 mm and ≥100 mm). The most common grayscale sonographic finding in the two groups was a unilocular simple cyst (found in 57.9% of girls with torsion, and 70.0% of girls without torsion). Bilocular or multilocular cysts were found in ≈10% in both groups. A unilocular-solid cyst presentation was more prevalent in girls with torsion (31.6% vs. 20%, respectively). However, these differences did not reach statistical significance.

The comparison of surgical findings, characteristics, procedures and adnexal pathologies is shown in Table 3. The main surgical approach in the two groups was laparoscopy. The time of surgery (i.e, the time period from initiation of anesthesia to transfer to the postoperative area) was similar. The paraovarian cysts were evenly distributed between the right and left sides. The majority of girls with torsion underwent a cystectomy procedure (84.2%), and the remainder underwent detorsion

Table 2
Comparison of ultrasound findings for pediatric and adolescent patients with and without a surgical diagnosis of adnexal torsion of a paraovarian cyst.

Parameter	Torsion ($n = 19$)	No torsion ($n = 20$)	P value
Transabdominal sonography	15 (78.9)	15 (75.0)	1.0[b]
Preoperative sonographic diagnosis of paraovarian cysts	11 (57.9)	14 (70.0)	0.514[b]
Largest cyst diameter (mm)	76.4 ± 36.3	80.2 ± 53.5	0.797[a]
Cyst diameter group			
≤50 mm	7 (36.8)	7 (35.0)	
51–99 mm	10 (52.6)	7 (35.0)	0.404[c]
≥100 mm	2 (10.5)	6 (30.0)	
Sonographic characteristics of the paraovarian cysts			
Unilocular simple cyst	11 (57.9)	14 (70.0)	
Bilocular or multilocular cyst	2 (10.5)	2 (10)	0.693[c]
Unilocular-solid cyst	6 (31.6)	4 (20)	

Data are presented as the number (%) or as the mean ± standard deviation.
[a] Two tailed t-test.
[b] Fisher's Exact test.
[c] Pearson's chi-square test.

Table 3
Comparison of surgical findings, characteristics, procedures and adnexal pathologies of pediatric and adolescent patients with and without a surgical diagnosis of adnexal torsion of a paraovarian cyst.

Parameter	Torsion ($n = 19$)	No torsion ($n = 20$)	P value
Surgical findings and characteristics			
Cyst side			
Right	11 (57.9)	12 (60.0)	1.0[b]
Left	8 (42.1)	8 (40.0)	
Duration of surgery (min)	52.9 ± 16.2	51.1 ± 14.2	0.712[a]
Laparoscopy	18 (94.7)	18 (90)	1.0[b]
Torsion of adnexa	12 (63.2)	0	
Torsion of fallopian tube	7 (36.8)	0	
Surgical procedure			
Cystectomy	16 (84.2)	20 (100)	0.106[b]
Detorsion with cyst drainage and biopsy	3 (15.8)	0	
Histology			
Benign epithelial paraovarian cyst	13 (68.4)	11 (55.0)	
Benign neoplastic paraovarian cyst	6 (31.6)	9 (45.0)	0.522[b]

Data are presented as the number (%) or as the mean ± standard deviation.
[a] Two tailed t-test.
[b] Fisher's Exact test.

with cyst drainage and biopsy (15.8%). Among pediatric and adolescent patients with an operative diagnosis of torsion involving the paraovarian mass, torsion of the ipsilateral adnexa was found in 12/19 girls (63.2%), whereas torsion of the fallopian tube was found in 7/19 (36.8%).There was no difference in the distribution of benign epithelial and neoplastic cysts between the two groups in the histological examinations. All neoplastic paraovarian cysts were of the benign serous type, including 7 serous cystadenomas and 8 serous cystadenofibromas.

3. Discussion

To the best of our knowledge, this is the first detailed study of the characteristics of paraovarian cysts causing adnexal torsion in pediatric and adolescent populations. The results point to several important findings that merit further discussion. The key observation is the relatively high torsion rate of 70% among pediatric and adolescent patients who underwent surgery for a paraovarian cyst. The pathophysiology of paraovarian cysts that cause adnexal torsion is unknown. One possible mechanism is the increased hypermobility of the adnexa owing to the increased length and laxity of the infundibulopelvic ligament, mesosalpinx or fallopian tubes in young women [11,12]. However, we believe that this theory explains cases of torsion of otherwise normal adnexa without cysts which are also prone to recurrent torsion. We do not believe that the laxity of the ovarian ligaments plays a significant role in torsion of paraovarian cysts, which do not tend to recur following surgical removal.

Some authors have recommended the removal of paraovarian cysts in young women to prevent torsion [10,13–16]. However, these studies do not provide conclusive information on the risk of torsion relative to the size of the paraovarian cyst [10,13–16]. Our results showed that adnexal torsion occurred in the presence of paraovarian cysts that are considered small (i.e., <50 mm), intermediate (51–99 mm) as well as large (>100 mm). In addition, there was no statistically significant difference between the mean cyst diameter in the torsion and nontorsion groups. Therefore, cyst size could not be used as a differentiating criterion for torsion here.

However, this observation is limited because our series only included surgical cases, where patients are often women with large cysts, whereas smaller cysts are usually monitored conservatively or remained undetected in asymptomatic women. Thus, the risk of torsion for smaller cysts is probably overestimated in our surgical case series.

Interestingly, an association between paraovarian cyst size and BMI was reported in pediatric patients [10]. Although our adolescent population had a higher BMI (95th centile) according to WHO growth references (https://www.who.int/growthref/who2007_bmi_for_age/en/), there was a no correlation between BMI and the size of the paraovarian cyst.

The clinical presentation of our patients was similar to the usual presentation of adnexal torsion; i.e., acute localized abdominal pain and nausea. One possible explanation is that significant necrosis had not yet occurred because of the short duration of torsion before surgical intervention, and that any existing necrotic tissue was relatively sparse [17].

The reported preoperative sonographic diagnosis of paraovarian cysts (Fig. 1) varies from 26% to 92% [10,18–20]. Savelli et al. showed a videoclip that the movement of a given mass in relation to the

Fig. 1. Transabdominal ultrasound image of the right adnexa showing a paraovarian unilocular anechoic cyst close to but separate from the ipsilateral ovary [19].

surrounding organs (ovary, bowel, uterus) during the dynamic sonographic evaluation of a patient (the "split sign") is a useful indication for discriminating between paraovarian masses [19].

Paraovarian cysts can take on a wide range of sonographic appearances. Similar to previous reports [5,20,21], the majority of our cases were anechoic unilocular cysts with a thin outer wall (simple cyst). Our data evidenced a similar distribution of benign epithelial and benign neoplastic paraovarian cysts for girls with and without torsion, and therefore could not be used as a differentiating criterion.

Note that this study is also limited by its sample size, retrospective design and the exclusive inclusion of surgical cases. Thus, the prevalence of torsion involving paraovarian cysts in our series is probably overestimated compared to its risk in asymptomatic women with paraovarian cysts who are followed conservatively. Furthermore, because of the retrospective design no power analysis was done to estimate the random chance of the study findings.

Overall, adnexal torsion is a common complication diagnosed in young patients undergoing surgery for paraovarian cysts, which is not associated with the sonographic appearance or the cyst diameter. In order to prevent torsion, surgical removal of paraovarian cysts should thus be considered in young girls undergoing surgery for paraovarian cysts.

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