doi:10.1111/jog.15159

J. Obstet. Gynaecol. Res. Vol. 48, No. 3: 838-842, March 2022



Pediatric and adolescent females are at higher risk of adnexal torsion recurrence—A large-scale retrospective study

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Abstract

Aim: Recurrence of adnexal torsion (rAT) is reported mainly in small series. Normal and small appearing ovaries are associated with an increased risk for rAT. Nevertheless, updated data of larger cohorts is lacking. We aimed to investigate the predictors for rAT in a cohort of women who had surgical intervention for primary adnexal torsion (pAT).

Methods: A retrospective case–control study from a single institution between 2011 and 2020. Women with a primary occurrence of surgically proven adnexal torsion were included. We compared those who had experienced rAT to those who had not. Univariate and multivariate analysis were performed to study independent predictors for rAT.

Results: Overall, 358 women were included. Of those, 35 (9.8%) had a rAT. Women who experienced rAT were younger (mean age 26 vs. 30 years, p = 0.01). Women experiencing rAT had smaller mean ovarian cyst diameter in the pAT episode (42 vs. 59 mm. p < 0.001). Performance of laparoscopic detorsion was only associated with rAT (odds ration [OR] 95% confidence interval [CI] 2.13 [1.02–4.42], p = 0.03), while the performance of additional cystectomy was negatively associated with rAT (OR 95% CI 0.10 [0.01–0.79], p = 0.006). Multivariate analysis demonstrated that age ≤15 and smaller cyst diameter at pAT were independently associated with the risk for rAT (aOR 95% CI 5.0 [1.09–23.2] and 1.47 [1.08–2.0], for every 10 mm decrease in cyst diameter, respectively).

Conclusions: Adolescents and pediatric females and women with smaller ovarian cysts at pAT are at higher risk for future recurrence of adnexal torsion.

Key words: adnexal torsion, adolescence, laparoscopy, pain, recurrence.

Introduction

Adnexal torsion (AT) accounts for 2.5%–7.4% of emergency surgeries for the indication of acute pelvic

pain.¹ Unwinding the twisted adnexa (without further surgical action carried out) is considered safe, even in cases in which the ovary appears necrotic.^{2–4} Recurrence of adnexal torsion (rAT) is reported to occur in

Abbreviations: AT, adnexal torsion; CI, confidence interval; OD, odds ratio; pAT, primary adnexal torsion; rAT, recurrent adnexal torsion

Received: November 10 2021. Accepted: January 11 2022.

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838 © 2022 The Authors. *Journal of Obstetrics and Gynaecology Research* published by John Wiley & Sons Australia, Ltd on behalf of Japan Society of Obstetrics and Gynecology.

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9%–28.6% of women when future follow up is performed, 1,5,6 however, these reports include mainly small series. Studies evaluating predictors of rAT have concluded that normal appearing ovaries and smaller ovarian size are associated with an increased risk for rAT. 5,7 Nevertheless, updated data of larger cohorts is lacking.

Ovarian fixation may be considered for women at risk for rAT^{8,9} as they may potentially decrease the risk for rAT.^{10,11} Therefore, it is paramount to identify which women will benefit from fixation at the primary adnexal torsion (pAT) surgery.

In the present study, we aimed to study predictors for rAT among a large cohort of pAT.

Material and Methods

Patients

A retrospective case—control study including women with a pAT, between March 2011 and January 2020, at a single medical center. We divided the cohort into two groups, cases and controls. We have identified women who had a second confirmed rAT (cases). Women with no future rAT composed the second group (controls). We excluded women in which oophoropexy was performed at the pAT.

Data collection

In all cases, physicians performed a thorough evaluation including medical history taking, clinical examination, laboratory, and sonographic studies.

Enlarged ovary was sonographically defined by the examiner when the involved ovary measured larger than the contralateral unaffected ovary (>10 mm).

Edematous ovary was defined in accordance with previous published work. 15

During laparoscopy, the involved ovaries were rotated back to the normal position. In some cases, cystectomy was performed at the discretion of the surgeon.

We manually reviewed all medical records, examination reports, and surgical report files to extract data and record the findings.

Statistical analysis

We compared women with rAT to those without using univariate analysis by the chi-square test and Fischer's exact test. T test and Mann–Whitney were performed for continuous variables. Multivariable regression analysis was performed, including variables resulting in p-values <0.05 in the univariate analysis. A value of p < 0.05 was considered statistically significant. The data were analyzed using Software Package for Statistics and Simulation (IBM SPSS version 27, IBM).

Ethics statement

Institutional Review Board approval was obtained for this study, waiving informed consent (5889-19-SMC, 20/03/2019).

Results

A total of 358 women were included in this study, of which 35 women (9.8%) had a rAT. Women with a future rAT were younger (mean age 26 vs. 30, p = 0.01) with higher proportion of age \leq 15 (odds ratio [OR] 95% confidence interval [CI] 4.4 [1.80–11.1], p < 0.001) (Table 1). Pregnancy rates during pAT did not differ

TABLE 1 Baseline characteristics of the recurrence vs. no recurrence groups

Characteristics	No recurrence, $n = 323$	Recurrent adnexal torsion, $n = 35$	OR (95% CI)	<i>p</i> -Value
Age	30 [24–37] (30)	27 [17–34] (26)		0.01
≤15	20 (6.2%)	8 (22.9%)	4.4 (1.80-11.1)	< 0.001
>40	62 (19.2%)	4 (11.4%)		0.35
Polycystic ovaries syndrome	32 (9.9%)	3 (8.6%)		1
Previous pelvic surgery	51 (15.8%)	9 (25.7%)		0.13
Premenarchal	10 (3.1%)	2 (5.7%)		0.33
Parity	0 [0–2] (1)	0 [0–1] (1)		0.03
Nulliparous	174 (53.9%)	22 (62.9%)		0.31
Pregnant	105 (32.5%)	10 (28.6%)		0.63
Gestational week	10 [7–14] (12)	10 [8–11] (9)		0.01

Note: Continuous variables are expressed as median [interquartile range] (mean). Categorical variables are presented as proportions. and Abbreviations: aOR, adjusted odds ratio; CI, confidence interval; IVF, pregnancies as denominator.

TABLE 2 Clinical characteristics of the recurrence vs. no recurrence groups

Characteristics	No recurrence, $n = 323$	Recurrent adnexal torsion, $n = 35$	OR (95% CI)	<i>p</i> -Value
Onset of pain, hours	27 (46.4)	25 (41.9)		0.89
Sonographic characteristics				
Affected side maximal diameter	66 (23.0)	64 (18.4)		0.66
Not affected side maximal diameter	43 (19.7)	42 (16.2)		0.94
Edematous ovary	79 (24.5%)	11 (31.4%)		0.36
Ovarian cyst	244 (75.5%)	28 (80.0%)		0.55
Cyst diameter, mm	59 (27.2)	42 (14.8)		< 0.001
Cyst diameter < 45 mm	51 (15.8%)	11 (31.4%)	2.44 (1.12-5.29)	0.02
Surgical characteristics				
Time from admission to surgery	704 (913.7)	577 (677.7)		0.42
Detorsion only	153 (47.4%)	23 (65.7%)	2.13 (1.02-4.42)	0.03
Cystectomy	70 (21.7%)	1 (2.9%)	0.10 (0.01-0.79)	0.006
Mature teratoma	21 (30.0%)	1 (100%)		
Benign cyst, other	24 (34.3%)	- '		
Paraovarian cyst	12 (17.1%)	-		
Unknown	17 (24.3%)	-		
Cyst drainage	58 (18.0%)	5 (14.3%)		0.81

Note: Continuous variables are expressed as median [interquartile range] (mean). Categorical variables are presented as proportions. and Abbreviations: CI, confidence interval; OR, odds ratio; PCO, polycystic ovaries.

between study groups (28.6% vs. 32.5% in the rAT and no recurrence groups respectively, p = 0.63).

Ovarian cyst diameter was smaller in the rAT group (mean 42 vs. 59 mm in the rAT and no recurrence groups respectively, p < 0.001) (Table 2). Among surgical characteristics, laparoscopic detorsion without further intervention was associated with rAT (OR 95% CI 2.13 [1.02–4.42], p = 0.03), and a performance of additional cystectomy was negatively associated with rAT (OR 95% CI 0.10 [0.01-0.79], p = 0.006). There were 21 (30.0%) mature teratomas, 24 (34.3%) other benign ovarian cysts, 12 (17.1%) paraovarian benign cysts, and 17 (24.3%) cases with unknown pathology reports in the non-rAT group and 1 (100.0%) mature teratoma in the rAT group. Among women of young age (<15 years old), the rate of detorsion-only during surgery did not vary between study groups (p = 0.807).

TABLE 3 Multivariate analysis of factors associated with recurrent adnexal torsion

Characteristics	aOR (95% CI)	<i>p</i> -Value
Age ≤15 years	5.0 (1.09–23.2)	<0.001
Cyst diameter ^a	1.47 (1.08–2.0)	<0.001
Parity	0.70 (0.40–1.23)	0.223
Detorsion only	2.00 (0.64–6.24)	0.229

Abbreviations: aOR, adjusted odds ratio; CI, confidence interval. and aFor every 10 mm decrease.

In multivariate regression analysis (Table 3), age \leq 15 [aOR 95% CI 5.0 (1.09–23.2), p < 0.001], and smaller cyst diameter [aOR 95% CI 1.47 (1.08–2.0), p < 0.001] were positively associated with the risk of rAT.

Discussion

This study demonstrates that younger age and smaller ovarian cysts at pAT episodes are independently associated with an increased risk for a future recurrence of AT. The rate of rAT in our study is 9.8% and the presence of one risk factor was associated with a 20.2% risk for rAT.

We have found that younger age (≤15 years) is an independent risk factor for rAT, with 1:4 recurrence rate in this age group. It was previously reported that women with rAT were 5 years younger than those without a rAT.⁷ It is possible that younger age enables longer follow-up period, in which rAT can be recorded. Notably, the follow-up duration in our retrospective study is influenced by the date of diagnosis rather than by the age of the women, therefore we advocate future prospective studies to elucidate this association. Furthermore, it is possible that AT among young women occurs following an inherent anatomical predisposition for AT (e.g., anatomical variations,

laxity of ligaments, etc.). Therefore, rAT could be more common in younger aged women. In our cohort, the proportion of adnexal cysts was comparable between groups, but cystectomy was performed significantly more in the non-rAT group, possibly due to lack of suspected pathology. This is in line with a previous study reporting higher rAT rate in normal appearing adnexas.⁵ There was one case of mature teratoma in the rAT group, compared with different benign pathological findings in the non-rAT group, in line with previous studies reporting low rate of malignant ovarian tumors in AT cases.¹

rAT is reported with rates varying 9%–28.6%.^{1,5,6} However, these reports are scarce and based on small cohorts. Furthermore, studies regarding independent risk factors for rAT are underreported. A previous report underlined 63% retorsion rate among postmenarchal women with normal appearing adnexa, and 9% in adnexa with pathology.⁵ However, only 62 women were included in this report. Another study compared 41 pregnant and 77 nonpregnant women and found 19.5% and 9.1% recurrence rates in both study groups, respectively.¹² In a larger study including 216 women of different ages, rAT occurred in 11% of cases.¹³ Our finding of 9.8% recurrence rate is in line with these publications.

A smaller cyst diameter was independently associated with rAT in our study. Previous studies reported increased risk of rAT in normal appearing and smaller ovaries.^{5,7} While ovarian size and the proportion of ovarian cysts in each group was comparable between study groups, cysts were smaller in the rAT group. Therefore, smaller cyst size may represent a more "normal ovarian appearance" as previously mentioned, conveying a higher risk for rAT.

The prevention of rAT, if possible, is paramount. As we have found that young girls, at their early reproductive years, are at increased risk for rAT, possible prevention means should be discussed and acknowledged. Oophoropexy during pAT has been suggested by some authors as a mean to decrease the risk of recurrence. ^{5,10,11,14}

Our study has several limitations. The retrospective nature carries bias, inherent to the retrospective nature, including information and selection bias. It is possible that other, nonstudied factors may account for some of the results observed. Furthermore, conducting the study in a single medical center may limit the generalizability of our results. Most importantly, we cannot account for the true prevalence of rAT in

our study population since it is possible that patients with rAT did not undergo surgery or underwent a surgery in another hospital, introducing a loss to follow-up bias. Therefore, the rate of rAT in every group may have been different, influencing the results. Furthermore, it is possible that some women in our study will suffer a rAT in the future. Therefore, importantly, as this is a retrospective study, we cannot account for the follow up period in both groups for all cases. The angle of torsion was not available in all cases—therefore it was not analyzed—this could be an important predictor of rAT. Nevertheless, our study includes a large, heterogenous cohort, increasing the generalizability of our results.

In summary, we have found that age ≤15 years and smaller ovarian cyst at pAT are independently associated with the risk for recurrence of AT. These factors should be acknowledged when considering measures at pAT to prevent AT recurrence.

Conflict of interest

The authors declare no conflict of interest.

Data availability statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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