

Premenarchal Ovarian Torsion: Ten Years of Experience of A Tertiary Center

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ABSTRACT

Ovarian torsion is a surgical emergency, as it could lead to tissue necrosis and loss of fertility in the long term when diagnosis and intervention are delayed. In this study, the effectiveness of sonographic ovarian appearance in the decision of surgery and the clinical value of ovarian size in the diagnosis of torsion were investigated in premenarchal girls whose diagnosis of ovarian torsion was surgically verified.

A retrospective cohort study design was utilized. Demographic characteristics, physical examination findings, laboratory findings, ultrasonography findings, operation records, pathology reports of 20 premenarchal girls who were admitted to Van Yuzuncu Yil University Medical Faculty Emergency Service between 2010-2020 and who were surgically confirmed to have torsion scanned and recorded.

The cases included in the study were divided into two groups: nine patients with normal ovarian torsion (group1) and eleven patients with mass or cystic ovarian torsion (group2). Regarding the duration passed between admission to the emergency department and undergoing surgery, the time was shorter in group 2 compared to group 1 (13.54 ± 6.02 hours and 77.0 ± 101.86 hours respectively; $p=0.053$). Conservative surgical treatment was realized in all patients in Group 1 (9/9; 100%) and 7 patients in Group 2 (7/11; 63.6%); patients in Group 2 underwent cystectomy. Furthermore, oophorectomy was performed on 4 patients in Group 2. In terms of the torsion side, right-sided torsion predominance was detected in the ovary in both groups (6 patients (66.6%) in Group 1 and 7 patients (63.6%) in Group 2.

Ovarian volume and Doppler flow could be considered as a potential predictive variable for ovarian torsion.

Keywords: Ovarian torsion, ovarian volume, premenarchal

Introduction

Ovarian torsion is a surgical emergency that might impact fertility potential adversely. Nonspecific clinical symptoms and variable sonographic images make diagnosis challenging (1). Thus, diagnosis and intervention are crucial. Albeit the reason for admission to the hospital is generally lower abdominal pain, nonspecific symptoms such as nausea, vomiting, fever, and leukocytosis might be present as well. However, the relatively high location of the reproductive organs in the abdomen among the pediatric population leads to difficulties in assessing the physical examination and thus reaching an accurate diagnosis (2).

The arterial and venous stems of the ovary are together with the peduncle part and the partial or full-twisting around the axis of the peduncle at a level that prevents the flow is called ovarian torsion. Concomitant torsion of the fallopian tube is termed adnexal torsion. Cases of torsion reported from antenatal period to postmenopausal age have been revealed in the literature. Its

etiology is not clear yet (3, 4). Comorbidity of it with benign mature cystic teratoma is common (5). On the other hand, torsion in the normal ovary is more common among the pediatric age group, and longer peduncle length is considered to be the cause (6).

There is no specific laboratory parameter that can be employed in the diagnosis of ovarian torsion. It has been revealed that 20% to 56% of patients with ovarian torsion have an increased white blood cell counts, but this is not a specific finding and might not be present at the first application. An elevation in CRP values might also be observed, but likewise, this finding is remarkably nonspecific and might not be observed if necrosis has not occurred (7-9).

In patients with ovarian torsion, the most sensitive sonographic sign is considered to be the vortex finding (tracking of torsioned peduncular vessels); however, this may not always be visible (10, 11). Color Doppler sonography might lead to confusion in diagnosis because there are numerous series of pediatric cases in which blood

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flow is monitored on Doppler sonography but surgically proven to have torsion (12).

The definitive diagnosis of torsion is direct observation of the torsioned ovary via laparoscopy or laparotomy. Clinical findings and laboratory parameters are not sufficient for diagnosis, though not specific for torsion. The obtained ultrasonographic data can be helpful in terms of torsion diagnosis or exclusion, but might also lead to error and delay in the intervention time. Early diagnosis and intervention are crucial to preserve ovarian function and fertility potential.

In our study, the adequacy of sonographic ovarian appearance in the decision of surgery and the clinical value of ovarian size in the diagnosis of torsion was investigated by evaluating the laboratory parameters and sonographic data of premenarchal girls who were admitted to the emergency department in the last 10 years in our clinic and were operated for torsion and confirmed to have torsion.

Material and Method

In this study, the files of 20 premenarchal female patients aged between 0 to 14 who were confirmed to have ovarian torsion and were operated on by Van Yuzuncu Yil University, Medical Faculty hospital Pediatric Surgery Clinic, and Gynecology and Obstetrics Clinic together between June 2010 and June 2020 were examined retrospectively. The ethical approval of the study was obtained from the medical science ethics committee of the university hospital (Ethics committee number: 2021/01-27). Demographic data, which was obtained from the hospital's computer-based data system, including the age and presentation symptoms of the patients, were examined. Data regarding the duration from ultrasonography to operation, surgical operation, intraoperative findings, and postoperative final histopathological results were assessed. Details of physical examination, clinical presentation, and laboratory findings were noted down, including the values of hematocrit (Htc), leukocytes (WBC), hemoglobin (Hb), and C-reactive protein (CRP). The exclusion criteria were as follows: patients in the post-menstrual, patients with tubal-ovarian torsion or isolated tubal, patients with a preoperative diagnosis of torsion and no intraoperative torsion, patients who underwent concomitant surgery such as appendicitis and patients with incomplete data records. Color Doppler ultrasonography and trans-abdominal ultrasonography (US) were performed on all

patients. Ultrasonographic findings of ovarian cyst or and vascular coding of suspected ovarian torsion were noted. Moreover, the size of the ovaries was measured in three planes. We calculated the volume of the ovaries by considering these organs as ellipsoid and using the formula of " $V = D1 \times D2 \times D3 \times \pi / 6$ " (D1=transverse diameter, D2=antero-posterior diameter, D3=longitudinal diameter, $\pi = \text{Pi}$).

Based on the intraoperative and histopathological examination, patients were categorized as follows: nine patients with normal ovarian torsion (group1) and eleven patients with mass or cystic ovarian torsion (group2). Clinical data and demographic variables and were compared between the two groups.

Statistical Analysis: The SPSS (IBM SPSS for Windows, ver.20) statistics package program was used for data analysis. Descriptive statistics such as average, standard deviation, minimum and maximum values was used for continuous variables, and number and percentage were given for categorical variables. Independent sample T-test was used to compare group means in terms of continuous variables.

Results

Twenty premenarchal girls who were operated for ovarian torsion over a ten-year period were included in the study. The clinical and demographic characteristics of all cases included in the study are presented in table 1. The mean age of all girls included in the study was 107.90 months (ranged between 7 to 163 months), while the mean values for hemoglobin was 12.56, for hematocrit 37.20, for leukocyte 12.23, for thrombocyte 326.62, and mean ovarian size was 107.97, and the mean duration between the diagnosis time and operation was 42 hours.

In our study, patients were divided into two groups based upon the histopathological examination: Group 1 included 9 girls with torsion of a normal ovary and group 2 included 11 girls with ovarian torsion with any mass or cyst. The mean ages of Group 1 and Group 2 were 108.0 ± 40.98 months and 107.81 ± 54.33 months, respectively (ranged between 7 to 163 months). Laboratory findings and the demographic of the two groups are summarized in Table 2. No statistically significant difference was determined between the two groups in terms of laboratory data including, WBC, Htc, Hb, Htc, Plt and CRP ($p > 0.05$). All patients had complaints of

Table 1. The clinical and demographic characteristics of the girls with ovarian torsion

	Mean \pm SD	Minimum	Maximum
Age (weeks)	107.90 \pm 47.55	7	163
Hemoglobin (g/dl)	12.56 \pm 1.41	9.76	15.40
Hematocrit (%)	37.72 \pm 4.26	29.50	45.30
Leukocyte (10 ³ / μ l)	12.23 \pm 4.69	4.86	23.40
Trombosit (10 ³ / μ l)	326.62 \pm 130.17	146	649
CRP (mg/L)	22.73 \pm 51.57	3.02	237.73
OS* (cm ³)	107.98 \pm 276.57	9.79	1256

OS Ovarian size on ultrasound; CRP: C-reactive proteine; * Mean time from admission to the emergency unit to the operation

Table 2. Clinical data and demographic characteristics of patients with normal ovarian torsion (group1) and mass or cystic ovarian torsion (group2).

	Group1	Group2	p
Age (weeks)	108.0 \pm 40.98	107.81 \pm 54.33	0.181
OS (cm ³)	15.06 \pm 5.95	184.00 \pm 362.18	0.993
Hemoglobin (g/dl)	12.87 \pm 1.48	12.31 \pm 1.37	0.389
Leukocyte (10 ³ / μ l)	12.15 \pm 6.83	12.21 \pm 2.12	0.985
Trombosit (10 ³ / μ l)	284.10 \pm 97.40	361.41 \pm 147.12	0.194
CRP (mg/L)	14.11 \pm 14.41	29.78 \pm 69.04	0.514
Time to operation* (hours)	77.0 \pm 101.86	13.54 \pm 6.02	0.053

OS: Ovarian size on ultrasound; CRP: C-reactive proteine; * Mean time from admission to the emergency unit to the operation; Group1: Normal ovarian torsion; Group2: mass or cystic ovarian torsion

abdominal pain. 3/9 (33.3%) patients in group 1 and 4/11 (36.3%) patients in group 2 had pain with vomiting and nausea. As radiological imaging, transabdominal and color Doppler ultrasound examinations of all patients were performed. Ultrasonographic examination of the patients was performed in the emergency department by radiology department residents with at least two years of ultrasonography experience. Ovarian torsion was demonstrated sonographically by a unilateral enlarged ovary, peripheral follicles, a complex adnexal mass, medialization of the ovary, loss of current in Doppler ultrasonography, displacement of the uterus out of the midline, whirl eddy signs in the vessels with torsion and free pelvic fluid. In Group 2, histopathological examination revealed simple cysts in 4 (36.3%) patients, including follicular and hemorrhagic cysts, corpus luteum cysts in 2 (18.2%) patients, and mature cystic teratoma in 5 (45.5%) patients.

A significant difference was determined between the groups in terms of the duration that passed from admission to the emergency department to undergoing surgery (77.0 \pm 101.86 hours and 13.54 \pm 6.02 hours, respectively; p=0.053). Furthermore, ovarian dimensions were measured in 3 planes in all patients, and ovarian volume was calculated. Group 1 had lower ovarian size

compared to Group 2, though not statistically significant (15.06 \pm 5.95 cm³ and 184.00 \pm 362.18 cm³, respectively; p=0.993). In terms of surgical treatment, the laparoscopic approach was utilized in 2 patients in Group 1 and 2 patients in Group 2. Conservative surgical treatment (detorsion) was performed in all patients in Group 1 (9/9; 100%) and 7 patients in Group 2 (7/11; 63.6%); plus, cystectomy was performed on patients in Group 2. Furthermore, oophorectomy was performed on 4 patients in Group 2. These cases were as follows: two had progressive necrosis, and the other two were suspected of malignancy. Regarding the torsion side, right-sided torsion predominance was determined in the ovary in both groups (6 patients (66.6%) in Group 1 and 7 patients (63.6%) in Group 2). In color Doppler examinations of torsioned ovaries, decreased or absent ovarian arterial blood flow was detected in 3 patients among Group 1 and in 4 patients among Group 2. Furthermore, it was observed that there was no venous blood flow in two patients in Group 1 and three patients in Group 2.

Discussion

Pediatric ovarian torsion constitute for nearly 15% of all ovarian torsion cases (12). Diagnosis is often

difficult in children due to the confusing symptoms and a wide differential diagnosis that includes urinary, genital, and gastrointestinal etiologies. Timely diagnosis is very important to avoid irreversible ischemic ovarian damage. The radiologists play a vital role in this. In our study, ultrasonographic examination of all patients was performed in the emergency department by radiology department residents with at least two years of ultrasonography experience, and ovarian torsion was demonstrated sonographically by a unilateral enlarged ovary, peripheral follicles, a complex adnexal mass, loss of current in Doppler ultrasonography, displacement of the uterus out of the midline, medialization of the ovary, free pelvic fluid, , and whirl eddy signs in the vessels with torsion. Adnexal torsion is much more prevalent than pure isolated ovarian torsion and is available in up to 67% of cases (5). The cases in this study included both adnexal torsions and pure isolated ovarian. Torsion of the ovarian pedicle impacts venous flow first, resulting in thrombosis and stasis, ovarian enlargement and progressive ovarian edema, and leading to clinical symptoms. Arterial flow is preserved initially, thanks to the thicker muscular walls of these vessels. Yet upon persistent torsion, arterial thrombosis occurs, and ultimately it ends up with necrosis and ovarian infarction. The infection can develop an untreated ovarian infarction into peritonitis. Localized peritonitis, which ultimately ends up with ileus or intestinal obstruction, might occur as well (13, 14). In our study, decreased or absent ovarian arterial blood flow was observed three patients in Group 1 and four patients in Group 2 in color Doppler examinations of torsioned ovaries. Furthermore, it was observed that there was no venous blood flow in two patients in Group 1 and in three patients in Group 2. Oophorectomy was performed in three patients due to the occurrence of progressive necrosis. In adult series, there is a factor that leads to ovarian torsion in 64-82% of cases.

Pediatric ovarian torsion can be seen at any age up to 18 years of age. However, more than half of the cases occur between the ages of 9-14., with a median of 11 years (12, 15). In a study with 504 cases, the mean age was determined to be 9.2 years.(16). In our study, the patients aged ranging between 7 months to 163 months. The median age was 120 months, and the mean age was 107 months.

Similar predisposing conditions are present in 51-84% of pediatric cases, albeit the incidence of ovarian torsion is lower in children compared to adults. (17). The most prevalent ovarian lesions

that cause torsion in children are mostly benign, and cystic teratomas (31%) and follicular or hemorrhagic cysts (23–33%) are more prevalent than paratubal cysts, hydrosalpinx or cystadenoma (17). In our study, 55% (11/20) of our patients had ovaries containing cysts or masses that could be predisposed to torsion. These patients constitute Group 2 in our study. In Group 2, histopathological examination revealed simple cysts in 4 (36.3%) patients, including follicular and hemorrhagic cysts, corpus luteum cysts in 2 (18.2%) patients, and mature cystic teratoma in 5 (45.5%) patients. The torsion mechanism in pediatric cases with benign ovarian masses is likely associated with the enlarged weight and size of the involved ovaries, which serve as a fulcrum for torsion. Malignant lesions associated with torsion are extremely rare, and account for only 2% of cases (18); it is considered that in these cases, the malignancy invades adjacent structures and some adhesions limit the mobility of adnexal structures (19). In our study, one patient was suspected of malignancy, and oophorectomy was performed; however, histopathological result was reported as mature cystic teratoma. In this case, the dimensions of the ovary were 20cmX12cmX10cm, and the ovary volume was calculated as 1256 cm³.

Ovarian volume is impacted by the age of the girls and the pubertal state (20, 21) Ovarian volume is calculated using the formula of the prolate ellipsoid: Length x width x height x 0.523 (1). It has been revealed that ovarian ascites are common among children with ovarian torsion (12). The average ovary volume is 1.06 cm³ in infants above three months of age and can rise to 3.6 cm³ (22). It has been revealed to be 1.05 cm³ in girls aged 4 to 12 months and 0.67 cm³ among girls aged 13 to 24 months. (22) Mean ovarian volume is 0.7 to 4.2 cm³ in premenarchal girls aged between 2 and 13 years, and mean ovarian volume is 9.8 cm³ in menstruating women (23). Servaes et al. (12) determined that the ovarian volume was greater than 20 in the ultrasound of pediatric patients who presented with ovarian torsion over 11 years and suggested that it was associated with adnexal mass. In our study, the mean ovarian volume of all patients was 107.97 cm³. These measurements were larger than normal ovarian sizes. When the groups were compared, it was determined that the size of the ovary in Group 1 was smaller compared to Group2 (15.06 ± 5.95 cm³ and 184.00 ± 362.18 cm³, respectively; p=0.993). However, no statistically significant difference was determined.

Group 1 had lower ovarian size compared to Group 2, though not statistically significant ($15.06 \pm 5.95 \text{ cm}^3$ and $184.00 \pm 362.18 \text{ cm}^3$, respectively; $p=0.993$). The reason that there was no statistically significant difference between Group 1 and Group 2 in terms of ovarian size might be due to the ovarian volume of 1256 cm^3 in one patient in Group 2, and this value might have increased the standard deviation in Group 2. The assumed mechanism for normal ovarian torsion without any cysts or mass in the ovary is the increased mobility of the adnexal structures. It is considered to occur due to a range of adnexal ligament length and laxity. As assumed in cases of asynchronous multiple torsion, above-average ligament laxity could predispose some girls to torsion (24). There are two putative mechanisms for ovarian torsion in neonates and infants. Prenatal descent of the ovaries from the abdomen might allow for the first increase in gonadal motility (25). Alternatively, ovarian enlargement and cyst development, with maternal hormone-induced stimulation, might serve as a fulcrum for torsion (26). Besides, ovarian ascites might display a sensitivity and specificity for ovarian torsion. These ovarian ascites can be a point for torsion and allows an enlarged ovary to twist along the fixation axes, resulting in impaired venous outflow, vascular occlusion, and edema in the ovary. (16)

In our study, one of our patients was 7 months old, her ovarian size was $4\text{cm} \times 2.7\text{cm} \times 3.3\text{cm}$, and there was a cyst. Torsion might not be evaluated clearly due to the general absence of specific symptoms in infants and limitations for evaluating pain in infants (27). The presence of feeding intolerance, an abdominal mass or ovarian abnormalities on prenatal imaging has been identified as the most common clinical characteristics that are associated with ovarian torsion in this age group (15). Unilaterally increased of ovary volume is the most common finding in ovarian torsion (12). Albeit the higher limit of normal ovarian length of 4 cm in adults has been reported (5), a similar measurement of length has not been fully established for children. But, Oltmann et al. (15) determined that the present of a pelvic mass greater than 5 cm in the pediatric population aged older than 1 year had the highest diagnostic sensitivity for ovarian torsion. Comparing with the contralateral is considerable when assessing ovarian torsion. It has been found in the study by Linam et al. (20) that the size of the ovary was a good indicator of torsion. In their series, ovarian torsion was not

detected in ovaries smaller than 20 ml, and they suggested ovarian size as the best predictor of torsion (20).

In our study, there was a statistically significant difference between the groups in terms of the time passed from admission to the emergency department until undergoing surgery (77.0 ± 101.86 hours and 13.54 ± 6.02 hours, respectively; $p=0.043$). Surgical intervention decision was taken earlier for patients with cysts or masses (Group 2). In our study, 3 of the patients in group 1 underwent oophorectomy due to progressive necrosis, which might be due to the delay in the decision of early intervention in torsion cases without ovarian cysts and masses. Lastly, a recent case report has demonstrated that normal ovarian volume and volume ratios may even be present in patients with ovarian torsion, though very rare (28).

The limitations of this study are its retrospective design and the fact that it is single-centered. Moreover, it may depend on the operator and there are potential variations between sonographers, regarding performed techniques.

When the ovarian torsion case developed in 20 premenarchal girls was examined, it was concluded that torsion could be detected in ovaries without cysts and masses, and that the patients with ovarian torsion in the presence of cysts and masses in the USG examination were surgically intervened earlier. At this stage, normal assessment of the ovaries on USG and the absence of cysts or masses might lead to diagnosis delay. Ovarian volume ratio and doppler flow could be considered as potential predictive variables for ovarian torsion. At the diagnosis stage, the decision of the clinician to intervene should be made primarily based on clinical findings. It should be noted that the predictive value of the assessment of bilateral ovarian volume by comparison and determining unilateral volume increase in the obtained USG data is more valuable than the Doppler sonography data in cases of partial torsion.

Conflict of interest: The authors declare that they have no conflict of interest

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