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CLINICAL RESEARCH





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# Background

Ovarian torsion is an emergency condition affecting 4.9/100,000 females aged 1–20 years [1]. It is caused by the twisting of the ovary on its vascular pedicle, leading first to lymphatic and venous congestion and then to the arterial blood supply becoming compromised, resulting in tissue necrosis. Torsion may occur in cases of any age, but the majority of cases are accompanied by the presence of an ovarian mass or cyst [2]. However, it has been reported that normal ovaries may twist around on their axis, with an incidence varying from 16% to 49% [3]. In the pediatric and adolescent age group, the twisting of a normal ovary is more common, and this may be caused by the greater length of the ovarian pedicle [4].

The usual presentations of an adolescent girl with ovarian torsion include abdominal pain lasting a few hours, nausea, vomiting, and fever. Because the signs and symptoms are non-specific, the diagnosis of ovarian torsion is a great challenge in the adolescent age group in the emergency department for both pediatric surgeons and adolescent gynecologists. Transabdominal ultrasound examination of pelvic organs in adolescents in whom the transvaginal route is not possible is mostly the crucial diagnostic tool. In spite of using laboratory analysis, Doppler flow studies, and sonography, the misdiagnosis of ovarian torsion is still frequent [4,5]. Fortunately, the presence of an ovarian mass in an adolescent girl complicated with abdominal pain alerts the physician to the possibility of ovarian torsion. Hence, it may be difficult to diagnose ovarian torsion in the absence of an enlarged ovary due to a mass or cyst, which will contribute to the delay in the diagnosis [6]. It has been reported that about 46% of ovarian torsions in children occurred in normal-appearing ovaries [5].

Ovarian torsion is predominately seen on the right side due to the possible effect of sigmoid colon in the left iliac fossa, which reduces the motility of tubal structure [7]. The rightsided presentation of torsion may be misdiagnosed as acute appendicitis in adolescent girls. It has been reported that mature cystic teratoma followed by corpus luteum cyst and follicular cyst are frequently found in torsion cases. Also, the examination with ultrasound in cases of intra-ovarian mass makes the diagnosis more feasible. The possible mechanisms for torsion of a normal ovary are sudden acceleration/deceleration movements, severe vomiting, and coughing that lead to an increase of intraabdominal pressure and hypoplasia of the uterus or Mullerian agenesis [4,8].

To date, a considerable number of studies that include large pediatric case series have detailed the presentation, intra-ovarian pathologic findings, and management options. However, the previous studies did not delve more completely into associated factors related to the torsion of a normal ovary in adolescent age groups. In this study, we aimed to analyze the ovarian torsion cases in a postmenarchal adolescent age group with regards to presentation, diagnosis, and management, and also to investigate whether normal-appearing ovaries on ultrasound affected the diagnosis of ovarian torsion or not.

## **Material and Methods**

This was a retrospective chart review of all postmenarchal adolescent girls who were operated on and surgically verified to have ovarian torsion between January 2010 and May 2016. The university hospital's ethical committee for medical sciences approved the study. The demographic data from the hospital's computer-based data system, including patient's age, menarche age, and presenting symptoms, were reviewed. Data regarding surgical operation, intraoperative findings, and postoperative final histopathological results were evaluated. Details of the clinical presentation, physical exam, and laboratory findings including hemoglobin (Hb), hematocrit (Htc), platelets (Plt), leucocytes (WBC), C-reactive protein (CRP), and alpha-fetoprotein (AFP) were noted. The exclusion criteria were as follows: patients who were in the premenarchal or adult age group, patients with isolated tubal or tubal-ovarian torsion, patients with incomplete data records, patients with concomitant surgical operation like appendicitis, and patients with a preoperative diagnosis of torsion who did not have torsion confirmed intra-operatively. Trans-abdominal ultrasonography (US) and color Doppler ultrasonography were performed in all patients, while selected patients underwent magnetic resonance imaging (MRI). The ultra-sonographic findings regarding ovarian mass or cyst and vascular coding of suspected ovarian torsion were noted. Also, the longitudinal axis of uterine size measured from the fundus uteri to the isthmus uteri on coronal section as millimeters (mm) was noted in all patients. This measurement was done at a longitudinal plane where the internal cervical os and endometrium were seen tallest and at a distance from the projection point of endometrium on uterine corpus to the internal cervical os. Due to the algorithm in the emergency radiology department, only uterine longitudinal axis size was measured. The data about uterine width were not available in medical records.

The patients were grouped according to the final intraoperative and histopathological examination as follows: either torsion of normal ovary (group 1) or torsion of ovary including any mass or cyst (group 2). The demographic variables and clinical data were compared between two groups.

#### Statistical analysis

Descriptive statistics for studied variables (characteristics) were presented as median, mean, and standard deviation values.

	Group 1 (n=8)	Group 2 (n=21)	P value
Age, years (mean ±SD)	14±1.7	14.1±1.8	0.808
Menarchial age, years (mean ±SD)	12.1±0.6	12.1±0.8	0.852
Hemoglobin, g/dL (mean ±SD)	11.4±1.3	12.5±1.1	0.051
Hematocrit,% (mean ±SD)	33.4±4.2	36.9±3.6	0.066
Leucocytes (×1000/mm³) (mean ±SD)	10.1±2.3	10.5±4.9	0.573
Platelets (/mm³) (mean ±SD)	238.8±76.2	266.3±56.2	0.283
CRP (mean ±SD)	15.4±33.8	15.7±40.1	0.750
AFP (mean ±SD)	2.0±1.9	1.3±1.0	0.534
Time to operation*, hours (mean ±SD)	34.5±24.3	19.5±9.2	0.001
Uterine size on ultrasound <sup>#</sup> , mm (mean ±SD)	34.3±2.9	47.6±4.5	0.001

**Table 1.** The demographic and clinical data of the study and control groups.

P<0.05 indicates statistical significance. Group 1: ovarian torsion in an otherwise-normal ovary; group 2: ovarian torsion in an abnormal ovary including a cyst or mass. \* Mean time from admission to the emergency unit to the operation; # measurement of the longitudinal axis of the uterus from the fundus uteri to the isthmus uteri. CRP – C-reactive protein; AFP – alpha-fetoprotein.

Continuous variables were compared among the two groups using the Mann-Whitney U test. Fisher's exact test was used to examine the association between categorical variables. Statistical significance levels were considered as 5%. The SPSS statistical program (IBM SPSS Statistics for Windows, Version 22.0, released 2013; IBM Corp, Armonk, New York, USA) was used for all statistical computations.

#### Results

Over a 6-year period, 29 patients in the postmenarchal adolescent age group who were operated on for ovarian torsion were included in the study. Based on the final histopathological examination, the patients were divided into two groups: group 1 (study) included 8 girls with torsion in a normal ovary, and group 2 (control) included 21 girls with torsion of an ovary with any mass or cyst. The mean age and menarchal age of groups 1 and 2 were 14.0±1.7 and 12.1±0.6 years, and 14.1±1.8 and 12.1±0.8 years, respectively. The demographic and laboratory findings of the two groups are summarized in Table 1. There was no statistically significant difference with regard to laboratory data including Hb, Htc, Plt, WBC, CRP, and AFP (p>0.05). Abdominal pain was seen in all patients in both groups. Pain with nausea and vomiting was seen in 3/8 (37.5%) in group 1 and 9/21 (42.8%) in group 2. Imaging studies of patients with ovarian torsion included transabdominal and color Doppler ultrasound examination in all patients and MRI examinations in 1 patient in group 1 and 2 patients in group 2. The ultrasonographic examination of the cases were performed in the emergency department by assistants from the radiology department with at least two years of experience in ultrasonography. The sonographic appearance of torsion in a normal ovary showed an enlargement and the presence of small cystic structures with increase in density of stroma around the periphery of the ovary. In group 2, the histopathological examination showed 4 (19%) simple cysts including follicular and hemorrhagic cysts, 4 (19%) cases with corpus luteum, 8 (38.1%) mature cystic teratomas, 2 serous cystadenomas, 1 mucinous cystadenoma, 1 juvenile type granulosa cell tumor, and 1 case with ovarian fibroma.

There was a statistically significant difference between the groups regarding the time from admission to the emergency unit to the operation (34.5±24.3 hours and 19.5±9.2 hours, respectively; p=0.001). Also, the longitudinal axis of uterine size was measured in all cases, and group 1 had a statistically significantly lower uterine size than group 2 (34.3±2.9 mm and 47.6±4.5 mm, respectively; p=0.001). With regards to the surgical management, a laparoscopic approach was used in one patient in group 1 and three patients in group 2. Conservative surgical management (detorsion) was performed in all cases (100%) in group 1 and 18 cases (85.7%) in group 2. Three patients in group 2 underwent oophorectomy. These cases were as follows: one had progressive necrosis, and the other two had a suspicion of malignancy. In terms of the side of torsion, both groups showed a predominance of right-sided torsion of the ovary (5 cases in group 1 [62.5%] and 18 cases in group 2 [85.7%]). When analyzing the color Doppler examinations of the torsioned ovaries, decreased or absent arterial blood flow to the involved ovary was observed in 4 patients in group 1 and 6 patients in group 2. Also, the absence of venous blood flow was observed in 2 patients in group 1 and 6 patients in group 2.

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## Discussion

This current retrospective study showed that ovarian torsion in the adolescent age group is rare but is an important issue. Although it frequently occurs with the presence of an ovarian mass, a normal ovary can twist around its pedicle. Our study population was restricted to the postmenarchal age group. The present study showed that the adolescent girls with torsion of a normal ovary were found to have a smaller uterus with regards to the measurement of the longitudinal axis of the uterus when compared to adolescent girls with torsion of an ovary with a mass or cyst. Also, our study showed that the time from first admission to the operation in cases of ovarian torsion was shorter in the patients with an ovary including any mass or cyst.

The most common presenting symptom with ovarian torsion is lower abdominal pain, which can have other causes such as acute appendicitis or urinary tract abnormalities in the adolescent age group, so it poses a diagnostic challenge [9]. The majority of our cases had acute-onset abdominal pain along with nausea and vomiting. This may lead to delay in diagnosis of torsion; it is reported that the delay may range from several hours and days to even months [10,11]. Fortunately, torsion cases with ovaries including a mass or cyst may help to raise suspicion and prompt the diagnosis. However, in cases of torsion with ovaries of normal size and appearance, diagnosis and timely operation are more difficult.

Tsafrir et al., who evaluated a 12-year experience with ovarian torsion in children, reported a 60% rate of normal-sized ovaries in adolescent girls with ovarian torsion [12]. Similarly, a study on premenarchal girls including 38 torsion cases reported 71% normal-sized ovaries [11]. These authors stated that ovarian torsion with normal-appearing ovaries is more characteristic of the younger age group. They reported that the median interval from symptom onset to emergency department admission was 24 hours and from emergency department admission to operation was 9.5 hours [11]. In our study, we evaluated the time from admission to the emergency unit to the operation in cases of torsion of a normal ovary and torsion of an ovary including any mass or cyst. We found that this time was longer in the group with normal-appearing ovaries (median, 24 hours vs. 18 hours, respectively). This can lead us to speculate that an ovarian mass or cyst can help to prompt earlier suspicion and diagnosis of ovarian torsion in adolescent girls who present with lower abdominal pain.

It has been reported that the presence of an abnormally long tube, meso-salpinx, or mesovarium may be a reason for the development of ovarian torsion in the normal-appearing ovary [13]. Also, another explanation for torsion of an otherwisenormal ovary in the pediatric population is abrupt changes in the intraabdominal pressure with vomiting, coughing, and sudden acceleration/deceleration movements [8]. Consistent with these data, the procedure of oophoropexy, which shortens the utero-ovarian ligament, reduces the ovarian laxity [14]. The most interesting finding of our study was that the longitudinal axis of uterine size, which was measured from the fundus uteri to the isthmus uteri, showed a significantly smaller uterine size in the group of patients with torsion of otherwise-normal ovaries. We can speculate that the hypoplastic or small uterine size may create and lead to long utero-ovarian pedicles and an appropriate space for the torsion of ovary. Sullivan et al. reported the case of a 14-year-old girl with ovarian torsion who had Mullerian agenesis, and they stated that the patients with Mullerian agenesis may be at an increased risk of ovarian torsion due to the absence of the utero ovarian ligament [15]. However, in contrast to our finding, there were several reports stating that the enlarged uterus due to a fibroid or gravid uterus may be a strong cause for the torsion of an otherwise-normal ovary or adnexa [16,17].

The diagnosis of ovarian torsion is usually made by the clinical examination findings and diagnostic imaging modalities, in which ultrasound examination with color Doppler analysis takes great part. However, use of color Doppler imaging has some controversies as the presence of vascular flow does not rule out torsion [18]. This is because of the dual blood supply from uterine and ovarian arteries. Moreover, torsion symptoms may occur due to the venous congestion before arterial supply ceases. Several papers reported that arterial and venous Doppler flows are often present in cases of surgically proven torsions [18,19]. Shadinger et al. reported that 54% of the torsion cases had arterial flow and 33% had venous flow [19]. In our study, consistent with the literature, 34.4% of torsion cases had arterial blood flow and 27.5% had venous flow.

The main limitations of this study were its retrospective nature and the small number of cases in each group. The other limitation is the uncertainty about data regarding the recurrence rate of torsion in normal adnexa and the future fertility outcomes on follow-up examinations. However, the strength of our study was that the preoperative examinations of all cases with color Doppler ultrasonography and measurement of uterine size as a routine procedure in the radiology department in the same center gave us the chance to present more comprehensive data.

### Conclusions

Ovarian torsion is a rare but important surgical emergency because of its potentially ominous consequences. The presence of ovarian torsion should be considered in a postmenarchal adolescent girl with lower quadrant abdominal pain. Ovarian torsion in an otherwise-normal ovary on ultrasound examination in the emergency department may lead to delay in diagnosis, and the clinicians should be alert about this issue.

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However, this needs to be clarified in future studies including larger case series.

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