Adnexal torsion: is there a familial tendency?

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Summary

Objective: To investigate whether patients diagnosed with adnexal torsion report a family history of adnexal torsion in a first degree relative. Materials and Methods: All women with a surgical diagnosis of adnexal torsion operated from 2008 to 2016 were contacted for participation in the study. The study was conducted by a telephone questionnaire. In addition, the medical records were retrospectively reviewed and information on demographic characteristics, surgical findings at time of torsion, and history of recurrent torsion events were collected. Results: Two hundred thirty women with a surgical diagnosis of adnexal torsion were identified. Of these, 147 women were reached and agreed to participate in the study. A history of adnexal torsion in a first degree relative was reported by seven (4.8%) patients: three cases involved an adnexal torsion event in a sister, and four cases involved an adnexal torsion event in a mother or daughter. Patients with a positive family history of adnexal torsion were more likely to experience torsion of otherwise normal adnexa without adnexal cysts compared to those with a negative family history (71.4% vs. 27.1%, p = 0.009). Conclusion: A family history of adnexal torsion in a first degree relative is found in a minority of cases and is significantly associated with torsion of otherwise normal adnexa and with recurrent torsion events. Thus, patients with a family history of torsion may be considered a high-risk subgroup of patients.

Key words: Adnexal torsion; Ovarian torsion; Recurrent torsion.

Introduction

Adnexal torsion is relatively rare cause of acute pelvic pain, with an estimated incidence of 4.9 cases per 100,000 [1]. In some cases, especially in those cases involving torsion of otherwise normal adnexa without any cysts or masses, recurrent torsion has been observed [2]. The etiology of adnexal torsion is to date unknown. Some authors hypothesized that adnexal torsion could be associated with abnormally long ovarian ligaments, more so for cases of torsion involving otherwise normal adnexa [3]. The familial tendency of adnexal torsion is to date unknown. A family history of torsion was elicited in a case report describing a 7 years-old girl with recurrent contralateral torsion of normal adnexa, whose mother was also operated for recurrent adnexal torsion as a pre-menarchal girl [4]. The aim of the current study is to investigate whether a family history of adnexal torsion exists in first degree relatives of pre-menopausal patients surgically diagnosed with adnexal torsion.

Materials and Methods

A computerized search of the hospital’s records was performed for pre-menopausal patients with the surgical diagnosis of adnexal torsion (ICD-9 code 620.5, “Torsion of ovary, ovarian pedicle and fallopian tube”) operated in the present department between 2008 and 2016. The authors reviewed the medical records of these patients and abstracted information on their demographic parameters, menstrual status, ultrasound and surgical findings, and pathology results. Additional torsion events were similarly retrospectively analyzed. Based on this information, cases of torsion were classified according to the adnexal pathology recognized at time of torsion, i.e., torsion of otherwise normal adnexa versus torsion involving adnexal cysts or masses.

All patients were prospectively contacted by telephone for participation in the study. Following verbal informed consent, the participants were asked to answer the telephone questionnaire, which included questions on the occurrence of torsion in first degree female relatives (i.e., sisters, mothers or daughters). The study’s flow chart is shown in Figure 1.

The statistical analysis was performed at the Tel-Aviv University statistical laboratory using the SPSS software. Descriptive variables are presented as mean ± standard deviation. Frequencies were compared with the Chi square test or with the Fisher’s exact test. Means were compared with the Student t-test. A two-tailed p value < 0.05 was considered statistically significant. The study was approved by the Assaf Haroフェ Medical Center Institutional Review Boards (IRB number 0258-16-ASF).

Results

During the study period, 230 pre-menopausal patients were surgically diagnosed with adnexal torsion. Of those, 150 (65.2%) were reached by telephone and 147 (63.9%) agreed to participate in the study (Figure 1). The comparison of the demographic and adnexal torsion characteristics between the entire torsion cohort (n=230) and the participants included in the study (n=147) is shown in Table 1. There were no statistically significant differences between...
the groups in patients’ age and menarchal status at time of torsion, rates of torsion involving otherwise normal adnexa versus torsion involving adnexal cysts or masses, and rates of recurrent torsion.

A history of adnexal torsion in a first degree relative was reported by seven (4.8%) patients: three patients reported an adnexal torsion event in a sister, and four cases reported an adnexal torsion event in a mother or daughter (Table 2). In five (71.4%) of these cases, the adnexal torsion involved otherwise normal adnexa, while one patient had torsion of a dermoid cyst and another had torsion of a corpus luteum cyst. Four (57.1%) patients with a family history of torsion reported recurrent torsion events. Among patients who experienced torsion as pre-menarchal-girls and adolescents, the rate of familial torsion was 3/49 (6.1%), while it was 4/65 (6.2%) for reproductive age patients (p = 0.3). There were no patients with a family history of torsion among the 33 patients diagnosed with adnexal torsion during pregnancy.

| Age (years) | Menarchal status | Adnexal pathology | Recurrent torsion | Familial history of torsion |
|------------|-----------------|-------------------|------------------|----------------------------|
| 6          | Pre-menarchal   | Torsion involving normal adnexa | Yes              | Sister                     |
| 9          | Pre-menarchal   | Dermoid cyst       | No               | Mother                     |
| 13         | Post-menarchal  | Corpus luteum cyst | No               | Mother                     |
| 22         | Post-menarchal  | Torsion involving normal adnexa | Yes              | Sister                     |
| 22         | Post-menarchal  | Torsion involving normal adnexa | Yes              | Sister                     |
| 23         | Post-menarchal  | Torsion involving normal adnexa | Yes              | Mother                     |
| 32         | Post-menarchal  | Torsion involving normal adnexa | Yes              | Daughter                   |

The comparison of patients with and without a family history of adnexal torsion is shown in Table 3. There were no statistically significant differences in the mean patients’ age or in their menarchal status. Patients with a positive family history of adnexal torsion were more likely to experience torsion involving otherwise normal adnexa compared to those with a negative family history (71.4% vs. 27.1%, p = 0.01). Similarly, recurrent torsion events were more prevalent among patients with a positive family history of torsion (57.1% vs. 17.1%, p = 0.009).

**Discussion**

The etiology of adnexal torsion is to date unknown. In cases of adnexal torsion involving adnexal cysts or masses, the probable etiology is the presence of those cysts, especially benign cystic teratomas (dermoid cyst) and paratubal cysts [5, 6]. In cases of adnexal torsion involving otherwise normal adnexa without adnexal cysts or masses, the likely etiology is abnormally long and mobile adnexal ligaments, which appear to be more common in pre-menarchal girls and adolescents [2]. In accordance with this hypothesis, the recurrence of adnexal torsion is more common in cases of...
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Table 3. — Comparison of demographic and adnexal torsion characteristics between patients with and without a family history of adnexal torsion.

| Parameter                     | Positive family history of adnexal torsion (n=7) | Negative family history of adnexal torsion (n=140) | p value |
|-------------------------------|-------------------------------------------------|---------------------------------------------------|---------|
| Age (years)                   | 18.1 ± 9.2                                      | 24.8 ± 11.4                                       | 0.1     |
| Menarchal status              |                                                 |                                                   |         |
| Pre-menarchal                 | 2 (28.6)                                        | 22 (15.7)                                         | 0.3     |
| Post-menarchal                | 5 (71.4)                                        | 118 (84.3)                                        |         |
| Age group                     |                                                 |                                                   |         |
| Girls and adolescents         | 3 (42.9)                                        | 46 (32.9)                                         | 0.3     |
| Reproductive-age              | 4 (57.1)                                        | 61 (43.6)                                         |         |
| Pregnancy                     | 0 (0)                                           | 33 (23.6)                                         |         |
| Torsion type                  |                                                 |                                                   |         |
| Torsion involving normal adnexa| 5 (71.4)                                        | 38 (27.1)                                         | 0.01    |
| Torsion involving adnexal cysts/mass | 2 (28.6) | 102 (72.9)                                      |         |
| Recurrent torsion episodes    | 4 (57.1)                                        | 24 (17.1)                                         | 0.009   |

Data is described as mean ± standard deviation or as number (%).

torsion involving otherwise normal adnexa [3]. The prevention of recurrent torsion of normal adnexa is also derived from this theoretical etiology, as the most common surgical approach for prevention of recurrent torsion is currently the shortening of the utero-ovarian ligaments [7].

The present authors’ hypothesis was that abnormally long adnexal ligaments leading to adnexal torsion may be linked with a familial tendency. Indeed, they found a family history of adnexal torsion in a small minority of patients with adnexal torsion, in the range of 5%. The familial occurrence of adnexal torsion was significantly associated with recurrent torsion and with torsion of otherwise normal adnexa, and both conditions are thought to be associated with abnormalities of the adnexal ligaments. Future research may discover genetic variations in specific connective tissue proteins which could be responsible for this condition.

The limitations of this study are its retrospective design and reliance on information obtained by telephone questionnaire, which could lead to recall bias. However, because adnexal torsion is an uncommon medical condition, the authors believe patients are less likely to falsely report this condition in family members. Another limitation of this study is the lack of control group, i.e. the information on the familial occurrence of adnexal torsion among women who did not experience adnexal torsion themselves. Nevertheless, although the findings are limited due to lack of control group, the authors believe these preliminary results may help to highlight a newly described familial association of adnexal torsion.

In conclusion, a family history of adnexal torsion in a first degree relative is found in about 5% of torsion cases. Patients with a positive family history are more likely to experience torsion of normal adnexa and recurrent torsion events. Thus, patients with a family history of torsion may be considered a high-risk subgroup of patients.

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