Uterine torsion in non-gravid women: A case report and review of cases reported in the last 20 years

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Abstract
Uterine torsion is defined as a rotation of >45° around the long axis of the uterus. Uterine torsion is an uncommon event but is even rarer in non-gravid women, with only 25 cases reported in the last 20 years. Here, we report a case of uterine torsion associated with multiple pedunculated subserosal uterine leiomyomas in an 83-year-old woman. She presented at the hospital with lower abdominal pain, and a computed tomography scan revealed multiple uterine leiomyomas with calcifications. Subsequent magnetic resonance imaging raised suspicion for torsion of pedunculated subserosal uterine leiomyomas. Emergency laparotomy was performed, and the patient was diagnosed with uterine torsion with multiple pedunculated subserosal uterine leiomyomas. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. The patient’s postoperative course was uneventful. Although difficult to diagnose due to its rarity, uterine torsion can be life-threatening and may cause infertility. Therefore, early diagnosis with imaging and surgical intervention are crucial to avoid serious complications.

Keywords
Uterine torsion, early diagnosis, imaging, surgical intervention, non-gravid women, postmenopause

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Introduction
Uterine torsion is defined as a rotation of >45° around the long axis of the uterus.1,2 Although uterine torsion is rare, it has been reported in women of all ages from premenarcheal to postmenopausal stages. Gravid women account for most of the reported cases, and uterine torsion in non-gravid women is extremely rare.1,3–9 Only 25 cases of uterine torsion in non-gravid women have been reported in the English literature available on PubMed over the last 20 years. Although uterine torsion is difficult to diagnose due to its rarity and non-specific symptoms and laboratory data, it can be life-threatening and cause infertility.4,5,9–16 Therefore, early diagnosis and surgical intervention are critical. Here, we report a case of uterine torsion associated with pedunculated subserosal uterine leiomyomas in a non-gravid older woman and present a review of all cases of uterine torsion in non-gravid women reported in the last 20 years.

Case
An 83-year-old woman presented at a hospital with acute lower abdominal pain. Acute enteritis was suspected, and antibacterial drugs were prescribed. Since her symptoms did not improve, she consulted the doctor again after 5 days. This time, appendicitis was suspected, and she was referred to another hospital. Contrast-enhanced computed tomography (CT) scan and abdominal X-ray were performed, which revealed uterine leiomyomas with multiple calcifications. Abdominal pain due to gynecological disease was then suspected, leading to her referral to our department. She had a medical history of untreated uterine leiomyomas, and she stopped visiting her gynecologist after menopause. On admission, she was generally stable with normal vital signs. Her abdomen was soft with no rebound tenderness, mass, or vaginal bleeding. Ultrasonography did not show clear findings.
due to artifacts caused by calcifications. Blood tests showed signs of inflammation with an elevated white blood cell count (10,040/μL) and C-reactive protein (1.61 mg/dL), and low hemoglobin levels (9.8 g/dL). Other laboratory findings were normal. Subsequently, contrast-enhanced magnetic resonance imaging (MRI) revealed multiple pedunculated subserosal uterine leiomyomas with calcifications (Figure 1(a)); peritonitis was similarly suspected. In retrospect, the MRI findings implied uterine torsion, considering the “whirl sign” of the uterine cervix (Figure 1(b)). Prior to surgery, we were not aware of the MRI findings of uterine torsion and suspected torsion of the pedunculated subserosal uterine leiomyomas. Two days after admission, laparotomy was performed. Multiple pedunculated subserosal uterine leiomyomas with calcification were identified, and the uterus was rotated 90° counterclockwise. The uterine body and bilateral uterine adnexa appeared congested and necrotic (Figure 2(a)). The stalks of the pedunculated subserosal uterine leiomyomas appeared to wrap around the junction between the uterine cervix and body, pulling the uterine body counterclockwise (Figure 2(b)). Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. Three pedunculated subserosal uterine leiomyomas (3–6 cm) were observed emerging from around the tubal cornua. The right adnexa and uterine body were stretched and appeared congested and necrotic. In addition, the right side of the uterine body was particularly stretched (Figure 2(c)). The final pathology of the specimen was uterine leiomyomas with diffuse calcification and hemorrhagic necrosis. Both ovaries and fallopian tubes and the uterine body were necrotic, suggesting total tissue infarction due to axial rotation of the uterus. No signs of malignancy were observed. The patient remained stable, and her postoperative course was uneventful, except for transient paralytic ileus. She was discharged on postoperative day 9 and has since been followed up at our department as an outpatient. No postoperative complications have been detected 6 months postoperatively.

Discussion

A literature search was conducted using the search engine PubMed and the search terms “uterine torsion” OR “torsion of the uterus.” Publications from 1 April 2001 to 31 March 2021 were searched, and the search was exclusively restricted to literature published in English. Since it was difficult to exclude gravid cases, cases of torsion of uterine leiomyoma and uterine adnexa or non-human cases based on the search strings alone, we manually checked the abstracts of all the articles returned by the search and extracted non-gravid uterine torsion cases, despite it being inefficient. Twenty-five abstracts were identified by the search. Of the 26 cases, including the present case, 5 involved premenarcheal girls, 7 involved menstruating women (including 2 cases of amenorrhea due to abnormalities of the reproductive organs), and 14 involved postmenopausal women. In uterine torsion cases, postmenopausal cases, such as the present case, are relatively common, with 14 cases out of 26 in this study (53.8%).

Etiology

The causes of uterine torsion differed by age group. In premenarcheal and young menstruating women, the cause of uterine torsion was abnormalities of the reproductive organs or ovarian tumors. In older menstruating or postmenopausal women, uterine torsion was caused by uterine leiomyomas or ovarian tumors, with uterine leiomyomas (including leiomyosarcoma in one case) being the cause in the majority of cases (n = 13/18, 72.2%; Table 1).
Complications
Regardless of age, uterine torsion can progress rapidly and become life-threatening. Cases complicated by progressive renal failure, hemorrhagic shock, or blood transfusion need have been reported. Of the eight cases involving premenarcheal and young menstruating women, five (62.5%) lost their fertility after surgery. Since the symptoms were often vague or non-specific, most patients were monitored for a few days to several months until surgical intervention was performed, with or without a preoperative diagnosis of uterine torsion (Table 1). Had uterine torsion been diagnosed earlier in these cases, life-threatening situations could have been avoided, and fertility could have been preserved.

Diagnosis and management
Although challenging, early identification of uterine torsion is important to avoid severe complications. The clinical features of uterine torsion in non-gravid women vary (Table 1), with no observed pattern in symptoms by age group. A previous report suggested that peritoneal signs are frequently absent in patients aged $\geq 60$ years. However, our review revealed that peritoneal signs were also frequently absent in cases involving premenarcheal or young menstruating women (Table 1). Regarding laboratory data, there are currently no known biomarkers that specifically indicate uterine torsion. A case exhibiting gradual elevation of lactate dehydrogenase and creatinine phosphokinase levels after the onset of symptoms has been reported. Similarly, there is a reported case wherein the patient developed coagulopathy. However, these findings were not common in uterine torsion based on our review of recent cases (Table 1). Therefore, elevated muscle enzyme levels and coagulopathy are not particularly useful for early diagnosis. The only common findings in the recently reported cases of uterine torsion were non-specific inflammation and/or anemia.

In this case, uterine torsion might have been caused by calcified pedunculated subserosal leiomyomas emerging from the fundus of the uterus because such uterine leiomyomas are more likely to cause uterine torsion. However, previously reported locations of uterine leiomyomas and the presence or absence of calcification vary; therefore, these factors are not predictive of uterine torsion. However, except for the present case, the diameters of the uterine leiomyomas or whole uterine bodies were $\geq 13$ cm in all uterine torsion cases caused by uterine leiomyomas.

Some reports have suggested that CT scan and MRI findings help diagnose this condition. Specifically, several features of CT scan and MRI, such as ischemia or hemorrhagic infarction of the pelvic mass, X-shaped configuration of the upper vagina, gas in the uterine cavity, or changes in the position of the pelvic mass, suggest uterine torsion. However, the most characteristic and common finding is the “whirl sign” of the uterine cervix. “Whirl sign” might be labeled as “whorled structure” or “whirled appearance,” or “twisted cervix” in some reports; however, these descriptions refer to the same finding. The “whirl sign” cannot be observed on ultrasonography; it can only be observed on CT

Figure 2. (a) Findings from laparotomy. Multiple pedunculated subserosal uterine leiomyomas with calcification are identified, and the uterus is rotated 90° counterclockwise. The uterine body and bilateral uterine adnexa appear to be congested and necrotic. (b) Stalks of the subserosal uterine leiomyomas appear to twine around the junction between the cervix and the uterine body and pulled the uterine body counterclockwise. (c) Resected uterus and both adnexa. Three pedunculated subserosal uterine leiomyomas (3–6 cm) arising near the tubal cornua are observed. The right adnexa and uterine body are stretched and seem to be both congested and necrotic.

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Table 1. Summary of reported cases of uterine torsion in non-gravid women in the last 20 years (surveyed from English literature listed in PubMed).

| Authors         | Age (years) | Symptoms (peritoneal signs) | Time from symptom onset to surgery | Laboratory data | Preoperative diagnoses (image inspections) (whirl sign) | Cause(s) of uterine torsion (major axis of the tumor) | Torsion degree (°) | Surgery | Remarks |
|-----------------|-------------|-----------------------------|-----------------------------------|-----------------|---------------------------------------------------------|------------------------------------------------------|-------------------|---------|---------|
| Holbrook et al. | 3           | Lower abdominal pain and distention Vomiting (+) | 10 days                          | Inflammation    | Pelvic tumor (US, CT scan) (-)                          | NA                                                  | NA                | TAH + RSO |         |
| Mohapatra et al. | 5           | Vaginal bleeding Abdominal girth (−) | 3 months                         | NA              | Ovarian tumor (US, CE-CT scan) (-)                      | Ovarian juvenile granulosa cell tumor (14.5 cm)     | 180               | LSO + pelvic LN | Fertility preservation |
| Ramasamy et al. | 9           | Lower abdominal pain (+)     | 3 days                            | Normal          | Ovarian torsion (US) (NA)                               | Ovarian cyst (6 cm)                                 | 180               | Subtotal hysterectomy + RSO |         |
| Dutra et al.    | 10          | Abdominal pain (−)           | 1 day                             | Normal (CBC)    | Abdominal cyst (CT scan) (−)                            | Ovarian cyst (NA)                                   | 360               | Laparoscopic partial hysterectomy + LSO |         |
| Grover et al.   | 10          | Lower abdominal pain (+)     | > 4 days                          | NA              | Uterine outflow obstruction (US) (NA)                  | Ovarian cyst (NA)                                   | 270               | Subtotal hysterectomy + RSO + LS |         |
| Omurtag et al.  | 13          | Cyclic pelvic pain (NA)      | 9 months                          | NA              | Hematometra Cervical agenesis (US, MRI) (−)            | Complete cervical and partial vaginal agenesis      | 180               | Hysterectomy | Horizontal torsion |
| Grover et al.   | 14          | Dysmenorrhea (NA)            | NA                                | NA              | Accessory non-communicating uterine horn (US) (NA)     | Septate uterus with obstructed right uterine horn   | 90                | Metroplasty after correction of uterine torsion | History of abdominal pain and admission Fertility preservation |
| Grover et al.   | 15          | Amenorrhea (NA)              | NA                                | NA              | Obstructed left unicornuate uterus (US, Radiological diagnosis) (NA) | Left unicornuate uterus                             | 180               | Surgery to anastomose cervix and uterus SAH + BSO | History of abdominal pain and admission Fertility preservation |
| Collinet et al.| 35          | Lower abdominal pain         | NA                                | Normal (CBC)    | Ovarian carcinoma (US) (NA)                             | Ovarian cyst (25 cm)                                | 360               |         |         |
| Grover et al.   | 36          | Lower abdominal pain (NA)    | 2 months                          | NA              | Broad ligament leiomyoma (CE-MRI) (−)                  | Uterine leiomyoma (13 cm whole uterine body)        | 90                | Myomectomy | Fertility preservation |
| Grover et al.   | 37          | Urinary retention (−)        | >1 day                            | Inflammation    | Uterine leiomyoma (US) (NA)                            | Uterine leiomyomas (13 cm)                          | 60                | TAH + BSO | History of myomectomy Myotonic dystrophy |
| Varras et al.   | 41          | Abdominal pain Vaginal bleeding (NA) | NA                                | NA              | Uterine leiomyoma (US) (NA)                            | Uterine leiomyomas (20 cm whole uterine body)       | 45                | TAH     |         |
| Grover et al.   | 52          | Lower abdominal pain (NA)    | 2 days                            | Normal          | Uterine torsion (CE-CT scan) (+)                       | Uterine leiomyomas (25.1 cm whole uterine body)     | 720               | TAH + BSO |         |
| Hashimoto et al.| 54          | Lower abdominal pain Genital bleeding Decreased urine output (−) | 4 days                            | Inflammation    | Ruptured ovarian endometriotic cyst                    | Ruptured ovarian endometriotic cyst (20 cm)         | 180               | TAH + BSO |         |

(Continued)
| Authors                  | Age (years) | Symptoms (peritoneal signs) | Time from symptom onset to surgery | Laboratory data | Preoperative diagnoses (image inspections) (whirl sign) | Cause(s) of uterine torsion (major axis of the tumor) | Torsion degree (°) | Surgery          | Remarks              |
|-------------------------|-------------|----------------------------|-----------------------------------|-----------------|--------------------------------------------------------|--------------------------------------------------|------------------|------------------|---------------------|
| Havaldar and Ashok⁶      | 55          | Abdominal pain Vomiting Flatus (NA) | 2 days                            | NA              | Ovarian cyst with hemorrhage (US) (NA) | Ovarian cyst (20 cm) | 180              | TAH + BSO       |                     |
| Yap et al.⁷             | 57          | Lower abdominal pain Nausea Vomiting (NA) | 1 days                            | NA              | Uterine torsion Adnexal cyst (US, CE-CT scan) (+) | Ovarian cyst (29 cm) | 180              | TAH + BSO       | History of CS and tubal ligation |
| Nagose et al.¹³         | 57          | Abdominal pain Vaginal spotting (+) | 3 days                            | Inflammation    | Uterine torsion Uterine leiomyoma (US, CT scan) (+) | Uterine leiomyoma (18 cm) | 270              | SAH + BSO       | Blood transfusion |
| Lük et al.⁵             | 61          | Lower abdominal pain and distension (NA) | ≥ 4 days                          | Anemia          | Uterine torsion Pelvic mass (uterine leiomyoma) (US, CE-CT scan) (+) | Uterine leiomyoma (15.5 cm) | 720              | TAH + BSO       | Hemorrhagic shock |
| Sikora-Szczeńiak et al.¹⁰ | 67         | Abdominal pain (−) | 5 days                            | Inflammation Coagulopathy | Abdominal mass (US, CE-CT scan) (−) | Uterine leiomyomas (35 cm whole uterus body) | 180              | TAH + BSO       | Blood transfusion Periumbilical hernia |
| Halassy and Clarke⁶     | 70          | Abdominal pain (−) | NA                                | Anemia          | Degenerated uterine leiomyoma (US, CE-CT scan) (−) | Leiomyosarcoma (16 cm) | 180              | TAH + BSO       | Blood transfusion |
| Oda et al.²¹            | 73          | Abdominal pain (−) | NA                                | Inflammation Anemia Elevated muscle enzyme | Uterine torsion Uterine leiomyoma (US, CE-CT scan) (+) | Uterine leiomyoma (15 cm) | 540              | TAH + BSO       | Umbilical hernia |
| Matsumoto et al.²²     | 73          | Lower abdominal pain (−) | NA                                | Inflammation   | Torsion of ovarian tumor (US, CT scan, CE-MRI) (−) | Ovarian tumor (23 cm) | 360              | TAH + BSO       |                     |
| Chua et al.²²           | 73          | Lower abdominal pain radiating to back (−) | 14 days                          | NA              | Uterine torsion Pelvic mass (US, CE-CT scan) (+) | Uterine leiomyoma (16 cm) | NA              | TAH + BSO       |                     |
| Present case            | 83          | Lower abdominal pain (−) | 7 days                            | Inflammation Anemia | Torsion of subserosal uterine leiomyoma (US, CT scan, CE-MRI) (−) | Uterine leiomyomas (3-6 cm) | 90               | TAH + BSO       |                     |
| Wang et al.¹¹           | 86          | Pelvic mass Uterine bleeding Urinary discomfort (−) | >9 days                           | Inflammation Anemia Renal dysfunction | Degeneration and infection of uterine leiomyoma (US, CT scan) (−) | Uterine leiomyomas (27 cm whole uterus body) | 360              | TAH + BSO       | Renal failure Undilical hernia |
| Jeong et al.²           | 87          | Abdominal pain Fever (NA) | NA                                | Inflammation    | Uterine torsion Uterine leiomyoma (US, CE-CT scan) (−) | Uterine leiomyomas (13 cm) | 360              | TAH + BSO       |                     |

US: ultrasonography; CT: computed tomography; NA: not available; TAH: total abdominal hysterectomy; RSO: right salpingo-oophorectomy; LSO: left salpingo-oophorectomy; LN: lymphadenectomy; CBC: complete blood count; LS: left salpingectomy; MRI: magnetic resonance imaging; SAH: supravcerval abdominal hysterectomy; BSO: bilateral salpingo-oophorectomy; CE: contrast-enhanced; CS: cesarean section.
scan and MRI. CT scan and/or MRI were performed in 18 of the 26 reported cases. Of these 18 cases, the “whirl sign” was observed in eight (44.4%), all of which were eventually diagnosed as uterine torsion. Accordingly, the sensitivity of the “whirl sign” for uterine torsion diagnosis was 44.4%, with a positive predictive value of 100% (specificity could not be calculated). Of the eight cases wherein the “whirl sign” was reported, six cases were detected by contrast-enhanced CT scan, one case by contrast-enhanced MRI, and one case by non-contrast-enhanced CT scan. Thus, due to the small number of reported cases, it is difficult to discuss whether the use of contrast media is beneficial for detecting the “whirl sign.” However, the use of contrast media might be useful for detecting other characteristic findings—such as ischemia or infarction of the uterus. Therefore, it is advisable to perform a contrast-enhanced CT scan or MRI whenever possible if the patient’s condition permits it. Despite its rarity, non-specific symptoms, and laboratory data, uterine torsion was diagnosed in 7 of the 18 (38.9%) cases based on the “whirl sign” of the uterine cervix preoperatively.\(^2,5,7,8,13,21,23\) In our case, the “whirl sign” of the uterine cervix was observed on preoperative MRI. However, we could not diagnose uterine torsion at that time as we had never encountered the “whirl sign” of the uterine cervix preoperatively.\(^2,5,7,8,13,21,23\) In our case, the “whirl sign” of the uterine cervix was observed on preoperative MRI. However, we could not diagnose uterine torsion at that time as we had never encountered a patient with this condition; thus, uterine torsion was not considered in the differential diagnosis. However, uterine torsion should be considered a differential diagnosis for acute abdominal pain in women of all ages, as early imaging and surgical intervention may preserve fertility and prevent life-threatening complications.

**Conclusion**

Uterine torsion in non-gravid women is rare and often diagnosed late, resulting in serious complications. According to the review of recent cases, uterine torsion in non-gravid women is typically associated with non-specific symptoms and laboratory data; however, CT scan and MRI often reveal characteristic findings; specifically, the “whirl sign” of the uterine cervix may be useful for early diagnosis. Thus, physicians should consider uterine torsion as a differential diagnosis when a woman of any age presents with acute abdominal pain.

**Declaration of conflicting interests**

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**Ethical approval**

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**Informed consent**

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