

# A case of uterine torsion concurrent with a ruptured ovarian endometrial cyst

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## Abstract

The patient was a 54-year-old female presented with severe abdominal swelling and intermittent pain. On emergent CT, massive ascites with thickened peritoneum and intra-pelvic cystic mass approximately 20 cm in diameter were observed. The cyst wall showed redundant irregular shape. The uterus was enlarged with intramural mass located at its right anterior fundus. MRI showed this pelvic cyst as high intensity on T1-weighted images, so that it was suspected as an endometrial cyst. The operation revealed the rupture of endometrial cyst and the uterine torsion of 180° around the long axis. Retrospectively, the X-shaped configuration of the upper vagina was observed on MRI and both the adnexa, including right ovarian cyst, were connected to the opposite side ovarian vein. Torsion of a non-gravid uterus is rare. In this case, the uterine torsion seemed to be caused by enlarged uterine body and ruptured endometrial cyst. Radiologists should be aware of this potential complication of huge ovarian mass and enlarged uterus and its appearance.

**Key words:** Uterine torsion—Ruptured endometrial cyst—Degenerated leiomyoma

Uterine torsion is defined as rotation of more than 45–720° on its long axis [1–4]. Dextrorotation occurs in two-third, and levorotation in the remaining one-third cases at the level of the uterine isthmus [4–6]. Uterine torsion is a rare condition, and many human cases occur during pregnancy [1, 2]. Non-gravid uterine torsion is extremely rare, but has been described due to the recent develop-

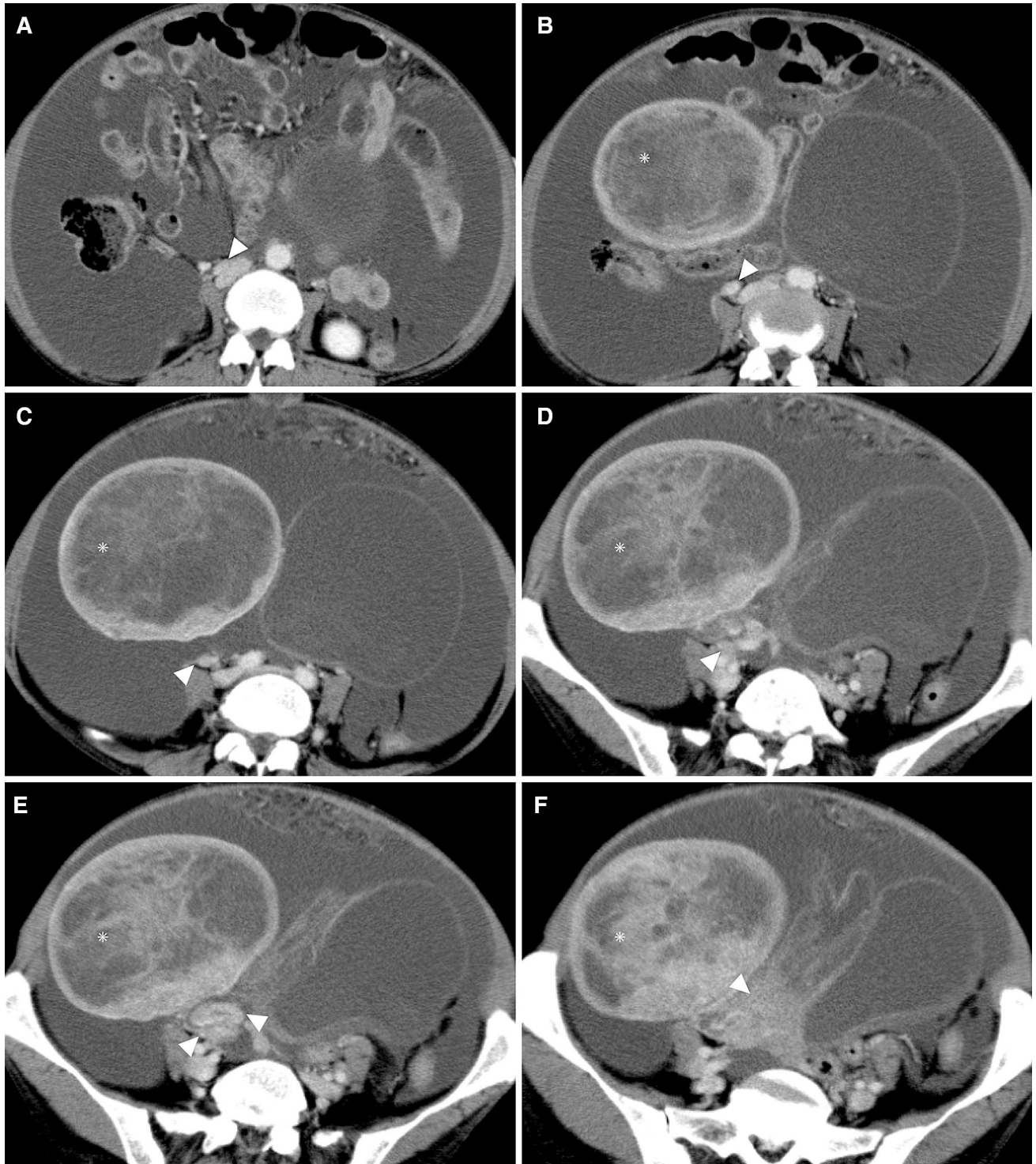
ment of examination and diagnostic imaging such as ultrasonography scanning and magnetic resonance imaging (MRI) [7–9]. Uterine torsion is associated with several conditions, such as abnormal fetal condition, previous cesarean section, leiomyoma, adnexal mass, malformation of uterus, and adhesion [3, 7, 8].

Abdominal pain is the major symptom, although it varies from mild to severe with shock. The difficulty with diagnosing uterine torsion is that the symptoms are non-specific, so that the majority of cases are not correctly diagnosed preoperatively [3, 4, 7–9]. This report describes a postmenopausal woman of non-gravid uterine torsion with a ruptured ovarian endometrial cyst based on the imaging findings. To the best of our knowledge, this is the first reported case of uterine torsion concurrent with ruptured ovarian endometrial cyst.

## Case report

The patient was a 54-year-old woman (Gravid 0, Para 0), who presented with a 4-month history of abdominal swelling and intermittent abdominal pain. The lower abdominal pain had worsened over the last 4 days with irregular genital bleeding and decreased urine output. She was admitted to the hospital as an emergency. Her past medical history was unremarkable. She had attained menopause at the age of 50 years. On abdominal examination, there was distension of lower abdomen, but it was neither tense nor tender. Her laboratory examination showed anemia (hemoglobin: 9.5 g/dL) and elevated C-reactive protein levels (5.25 mg/dL). Tumor markers (carbohydrate antigen 19-9: 1299.4 U/mL, cancer antigen 125: 5180.9 U/mL) were elevated.

Contrast-enhanced computed tomography (CT) showed an intra-pelvic multicystic mass, 20 cm in diameter, massive ascites, and thickened peritoneum.



**Fig. 1.** A–J Contrast-enhanced CT images (A–G axial view, H–J coronal view). CT images show intra-pelvic multicystic mass of 20 cm in dimension with redundant irregular wall. The multicystic mass locates to the *left side* of the uterus, though connects to right ovarian vein passed through dorsal uterus

(*arrow head*). The uterine body and fundus is dislocated to the *right side* in the pelvic space. The upper vagina shows an X shape (*arrows*). CT also shows the intramural mass that is suspected degenerated leiomyoma (*asterisk*) and massive ascites.

Ascites had a higher density than the bladder urine. The cystic mass located to the left and dorsal side of the uterus. The cystic wall revealed redundant irregular

without mural nodule or solid parts. The cystic wall and the uterus were clearly enhanced. Although the cystic mass was located on the left side, the dilated right

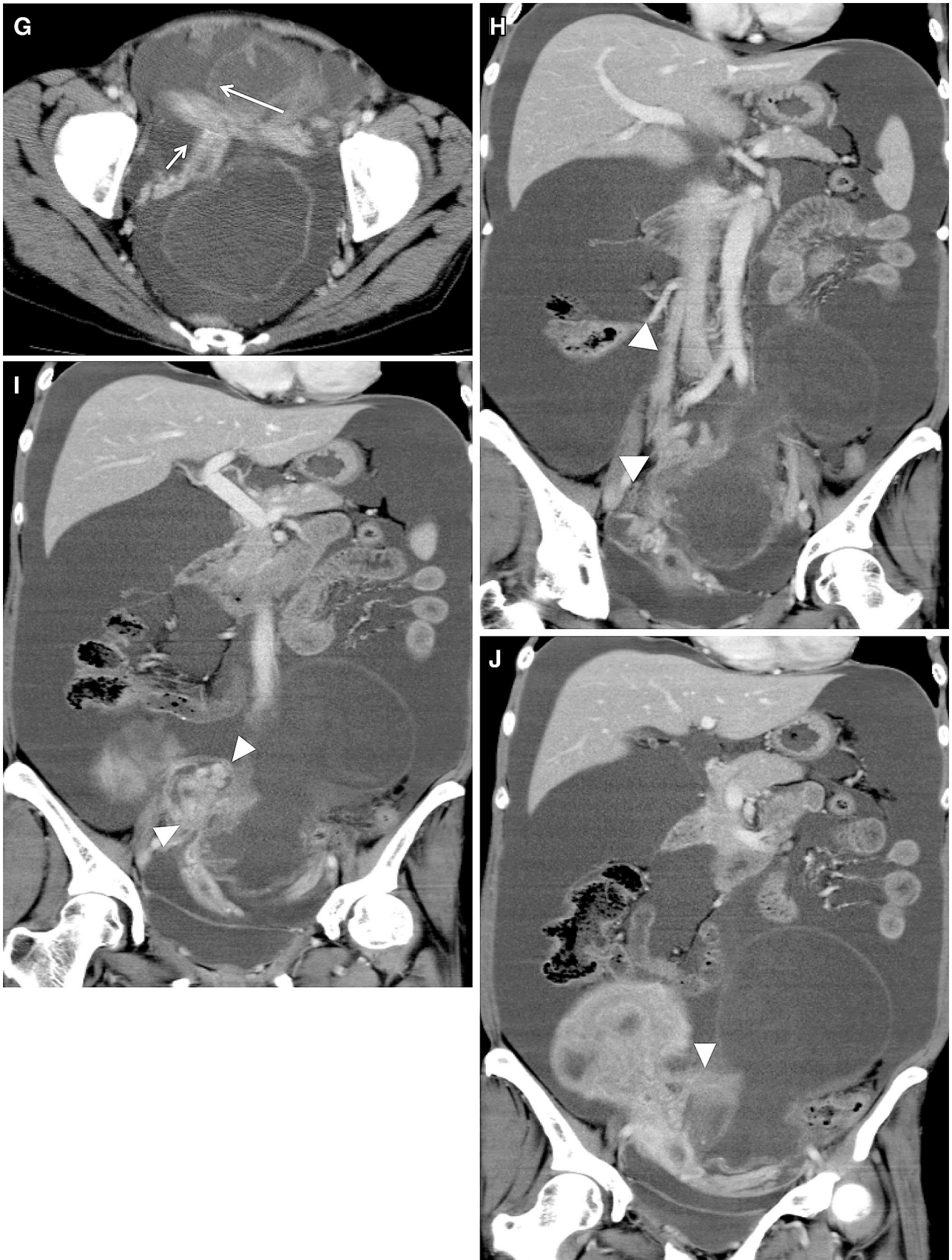
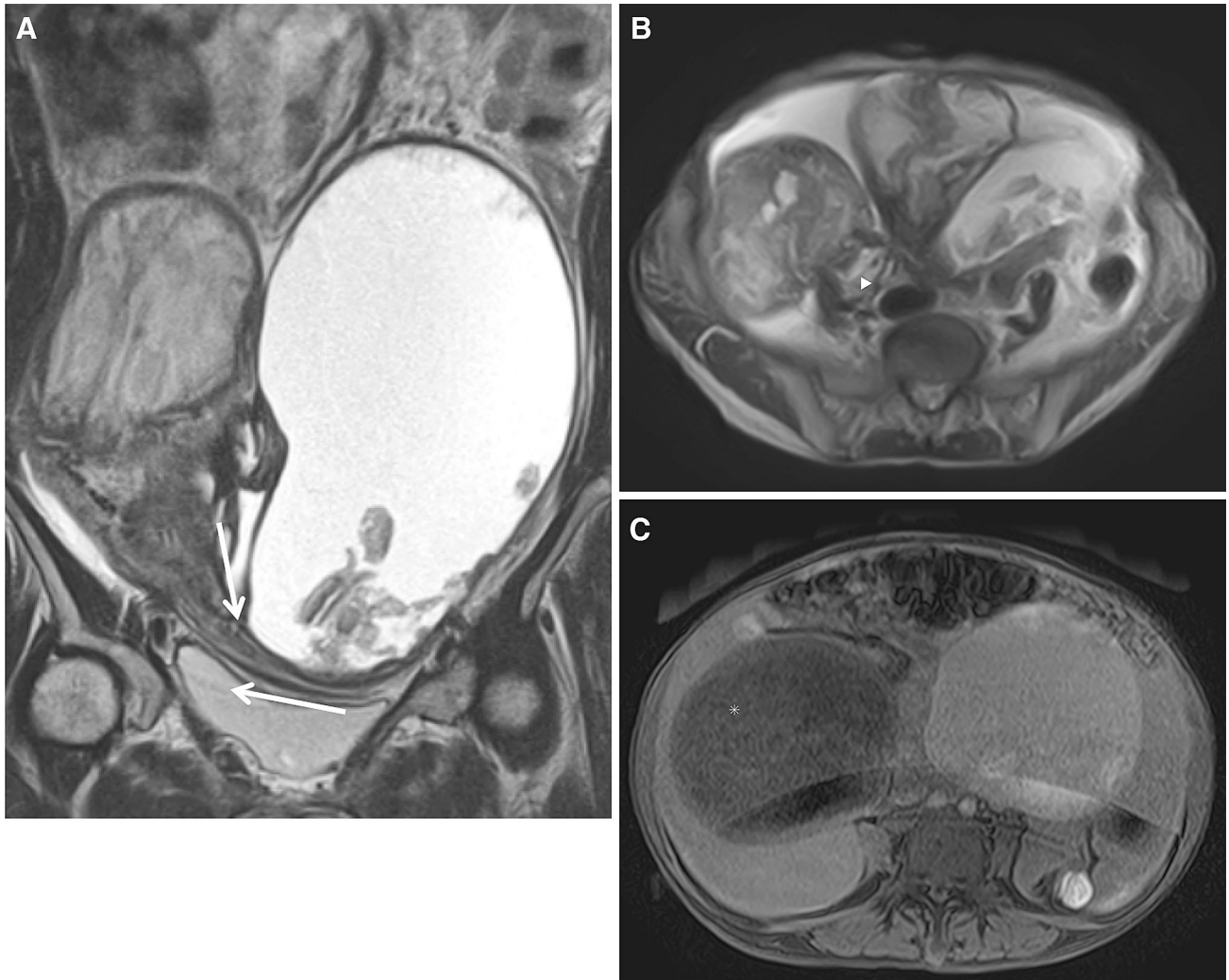


Fig. 1. continued.



**Fig. 2. A–C** Non-contrast-enhanced MR images (**A, B** coronal and axial T2-weighted images, **C** axial T1-weighted images). The multicystic mass with debris connects to right ovarian vein (*arrow head*). The contents of cystic mass demonstrate high intensity on T1-weighted images. Massive

bloody ascites is also detected. The uterus twists at the level of the isthmus, and the uterine body locates across the cystic mass (*arrows*). As CT images, the well-defined mass localizes in myometrium of uterine fundus (*asterisk*).

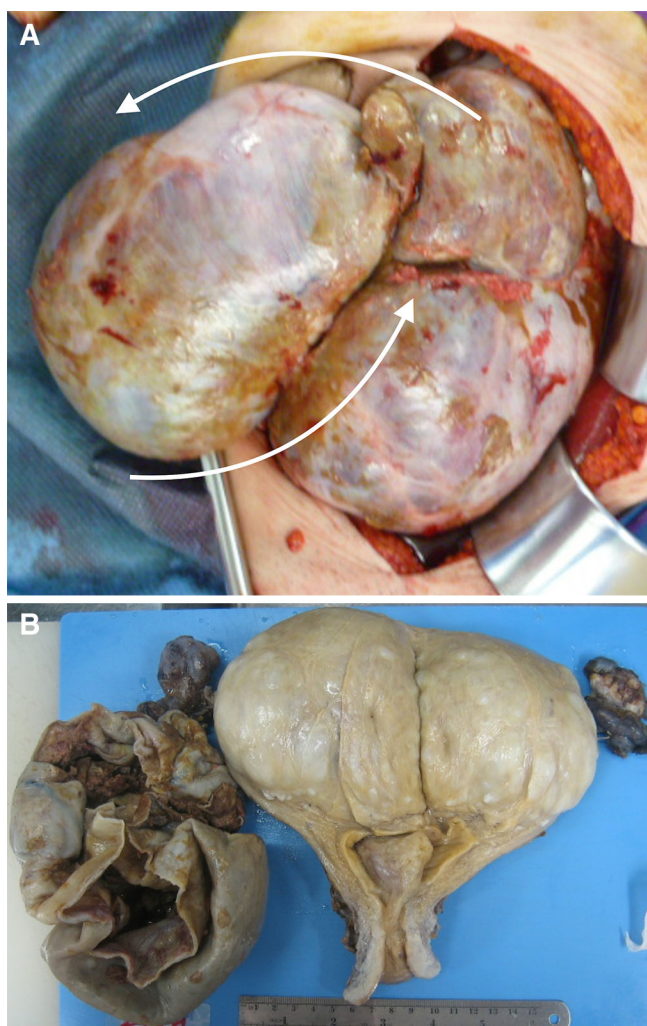
ovarian vein was twisted and connected to the cystic mass. An intramural mass, 13 cm in size, of the uterine fundus with heterogeneous enhancement was also seen. CT showed thickened uterine endometrium of about 2.5 cm (Fig. 1).

MRI showed the multicystic mass as high intensity with debris on T2-weighted images. Massive ascites and the contents of the cystic mass were high intensity on T1-weighted images. The uterine body was dislocated to the right side of intra-pelvic space with the mass of the uterine fundus. The well-defined mass of uterine fundus showed a heterogeneous high intensity, and was localized in myometrium (Fig. 2). Uterine endometrial thickening was observed, but it was not high intensity on diffusion-weighted imaging.

The multicystic mass was suspected to be a ruptured endometrial cyst, which was thought to be due to bloody ascites and peritonitis. A degenerated leiomyoma and endometrial hyperplasia were also suspected. There were no findings suggestive of metastases, though ovarian cancer arising from endometriosis or uterine endometrial carcinoma could not be ruled out. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed electively.

At operation, intra-pelvic coagulation, adhesions, and peritoneal thickening were observed. The enlarged uterus had rotated 180° anticlockwise at the level of isthmus. A right ovarian multicystic mass with a ruptured endometrial cyst was dislocated to the left side of uterus (Fig. 3). The multicystic mass adhered to the





**Fig. 3. A, B** Intraoperative photos. The enlarged uterus is rotated toward the *left* by 180° and right ovarian multicystic mass with ruptured cyst is located to the *left side* of uterus.

uterine body and the pouch of Douglas, which was easily freed. The left Fallopian tube and ovary were normal, but located to the opposite right side of the uterus.

The pathological diagnosis was a right ovarian endometrial cyst without malignancy, myxomatous degenerated leiomyoma, and endometrial polyps.

The patient's postoperative course was uneventful.

## Discussion

Uterine torsion is a rare condition, and its mechanism is unclear. However, it can be explained by laxity, elongation, and weakness of the parametrial tissues following menopause, and by asymmetrical uterine enlargement and abnormal movement or position with a leiomyoma or an adnexal mass [7–12]. In the present case, the enlarged uterus with huge leiomyoma of fundus was elevated, and the ovary endometrial cyst also may have become a trigger of the uterine torsion.

Acute abdominal pain is the major symptom, and some patients may present with vaginal bleeding, vomiting, and urinary problems [4, 7, 9–11]. In the present patient, the cause of abdominal swelling, worsened abdominal pain, and anemia was likely to have been intra-abdominal hemorrhage and peritonitis caused by rupture of the ovarian endometrial cyst followed by uterine torsion.

Ultrasonography scanning is insufficient to make a correct diagnosis of the uterine torsion. CT and MRI have shown a change in the position of leiomyoma or adnexal mass, twisting of cervix or ovarian vessels across the uterus, an X-shaped configuration of the upper vagina, and the difference in contrast enhancement between uterine corpus and cervix [12–15]. CT/MRI is also important for the different diagnosis of conditions such as appendicitis, degeneration of leiomyoma, adnexal torsion, ectopic pregnancy, and placental abruption. In the present case, considering the preoperative CT and MRI findings retrospectively, the key diagnostic findings were the following two points: first, the ruptured endometrial cyst was connected to the right ovarian vein, whereas the adnexa were located to the opposite side of the uterus; second, the uterine body and fundus were dislocated to the right side and the upper vagina showed an X shape. These findings are identical to those of the previous reports.

Uterine torsion often requires diagnosis laparotomy. If the preoperative diagnosis is delayed, a case with persistent or worsening abdominal pain demands immediate surgery, because the uterus may have undergone infarction, congestion, and necrosis with thrombosis [3, 9, 12]. The surgical management is influenced by the patient's age, fertility preservation, pathology, and viability of the uterus and adnexa. Since the present patient was postmenopausal, total hysterectomy with bilateral salpingo-oophorectomy was performed. Due to expected operative difficulty with orientation and ensuring a visual field due to massive ascites and peritonitis, the present patient underwent elective surgery. The histopathological report did not confirm the presence of infarcts or necrosis in both the uterus and adnexa, as shown by preoperative imaging diagnosis. There was not evidence of malignancy and, therefore, the elevated tumor markers were thought to be due to bloody ascites and peritonitis.

Uterine torsion should be considered an important different diagnosis in a patient presenting with abdominal pain. Even if massive ascites, a huge adnexal mass, or a leiomyoma are seen on CT/MRI, it is necessary to diagnose an enlarged or displaced uterus and adnexa exactly. The findings of a twisted or whorled structure of the uterus, adnexa, and ovarian blood vessels with or without contrast enhancement could be helpful in the diagnosis of uterine torsion.

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*Compliance with ethical standards*

*Conflict of interest* Aya Hashimoto, Junko Takahama, Naoya Harada, Shinsaku Maeda, Hiroshi Anai, Akio Fukusumi, Shunsuke Imai, and Kimihiko Kichikawa declare that they have no conflict of interest.

*Ethical approval* All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

*Research involved in animal participants* This article does not contain any studies with animals performed by any of the authors.

*Informed consent* Informed consent was obtained from the patient included in this study.

## References

- Piot D, Gluck M, Oxorn H (1973) Torsion of gravid uterus. *Can Med Assoc J* 109(10):1010–1011
- Corona M, Bachrach I (1984) Pathologic torsion of the pregnant uterus. *Acta Obstet Gynecol Scand* 63:375–376
- Hawes CH (1935) Acute axial torsion of the uterus. *Ann Surg* 102:37–40
- Dua A, Fishwick K, Deverashetty B (2006) Uterine torsion in pregnancy: a review. *Int J Gynecol Obstet* 6:1
- Corr JE (1943) Axial torsion of the gravid uterus in two successive pregnancies. *Am J Obstet Gynecol* 46:749–751
- Collinet P, Narducci F, Stien L (2001) Torsion of a nongravid uterus: an unexpected complication of an ovarian cyst. *Eur J Obstet Gynecol Reprod Biol* 98:256–257
- Qureshi S, Singh U, Bansal B, et al. (2013) Torsion of preterm gravid uterus: a case report. *IJCRI* 4(7):392–395
- Sharma D, Usha MG (2013) Torsion of a non-gravid uterus: a rare cause of acute abdomen. *Int J Reprod Contracept Obstet Gynecol* 2(2):234–236
- Havaldar N, Ashok K (2014) Torsion of non-gravid uterus with ovarian cyst—an extremely rare case. *Pan Afr Med J* 18:95
- Moores KL, Wood MG, Foon RP (2014) A rare obstetric emergency: acute uterine torsion in a 32-week pregnancy. *BMJ Case Rep*. doi:10.1136/bcr-2013-202974.
- Nielsen TF (1981) Torsion of the pregnant human uterus without symptoms. *Am J Obstet Gynecol* 141(7):838–839
- Jeong YY, Kang HK, Park JG, et al. (2003) CT features of uterine torsion. *Eur Radiol* 13:249–250
- Matsumoto H, Ohta T, Nakahara K, et al. (2007) Torsion of a nongravid uterus with a large ovarian cyst: usefulness of contrast MR image. *Gynecol Obstet Invest* 63(3):163–165
- Nicholson WK, Coulson CC, McCoy MC, et al. (1995) Pelvic magnetic resonance imaging in the evaluation of uterine torsion. *Obstet Gynecol* 85(5–2):888–890
- Simms-Stewart D, Hardie J, Mitchell P, et al. (2012) Torsion in a perimenopausal non-gravid uterus with infarction and gangrene of uterus and adnexa: a proposed means of making the diagnosis clinically. *J Obstet Gynaecol* 32(3):312–313