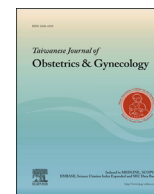


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Research Letter

Spontaneous rupture and massive hemoperitoneum from uterine leiomyomas and adenomyosis in a nongravid and unscarred uterus



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Uterine leiomyomas (fibroids or myomas) are benign monoclonal tumors arising from the smooth muscle cells of the myometrium [1]. The symptoms attributed to myomas include heavy or prolonged menstrual bleeding, pelvic pressure and pain, and reproductive dysfunction. Acute or chronic pelvic pain may be a result of mass bulk-related symptoms, dysmenorrhea, dyspareunia, or degeneration or torsion of the myoma [2]. Adenomyosis refers to a disorder in which endometrial glands and stroma are present within the uterine musculature (uterine adenomyomatosis). Heavy menstrual bleeding, painful menstruation, and chronic pelvic pain are the major symptoms of adenomyosis, and they occur in approximately 25% of cases [3]. Although leiomyomas and adenomyosis are common gynecological disorders, hemorrhage, internal bleeding, hemoperitoneum, and uterine rupture due to these disease etiologies are rare and can easily be missed in emergency conditions. Furthermore, delayed diagnosis and management may increase the risk of morbidity and mortality [4,5]. Herein, we report an extremely rare case presenting as spontaneous rupture from uterine leiomyomas with adenomyosis in a nongravid and unscarred uterus.

A 39-year-old female presented to our emergency department in shock and near fainting condition accompanied with acute diffuse lower abdomen pain. Her medical history was unremarkable, and she denied undergoing any surgery such as laparoscopy or laparotomy for uterine tumors. She denied being pregnant, having given birth, or undergoing an abortion in the past and denied having an

active sexual life in the past few years. A physical examination revealed rebound tenderness and peritoneal signs, unstable clinical vital signs (blood pressure 95/67 mmHg, pulse rate 110 beats/min, and normal body temperature), and no vaginal bleeding. A urine pregnancy test yielded a negative result. Laboratory test results showed a decreased hemoglobin level (Hb = 7 g/dL) with low hematocrit (Ht = 31%) and coagulopathy with prolonged prothrombin time (INR (international normalized ratio) = 1.38). Gynecological ultrasound and emergency computerized tomography demonstrated several huge uterine leiomyomas, the largest of which was approximately 8 cm × 9 cm × 9 cm in size, and posterior uterine wall adenomyosis (Fig. 1). A massive amount of intraperitoneal fluid was also noted, leading to the suspicion of internal bleeding (Fig. 2), with a high density area over the cervical–uterus junction, raising the concern of a uterine rupture with active bleeding (Fig. 3). Under the impression of hemorrhagic shock of uncertain origin (no other specific vascular bleeding origin was noted on the computerized tomography scan), exploratory laparotomy was arranged. Massive internal bleeding of about 4500 mL and a huge, ruptured uterus were noted intraoperatively, which were compatible with our suspicion. With the consent of the patient's family, total abdominal hysterectomy was performed. The gross specimen of the uterus measured 17 cm × 11 cm × 11 cm and weighed 888 g with several intramural, submucosal, and subserosal types of leiomyomas, the largest of which was 8.5 cm in size, and the posterior uterine wall showed adenomyosis pathologically. The serosa layer over the uterus was rough and irregular, and a spontaneous laceration wound was found at the posterior cervical–uterus junction (Fig. 4). Blood transfusion with whole blood and packed red blood cells (12 units) were given during the operation. The patient had an unremarkable perioperative period and was discharged on postoperative Day 4 in a stable condition.

Leiomyomas and adenomyosis are common in women of reproductive age. The incidence of leiomyomas is approximately 12–25% in women of reproductive age, and are found on pathological examinations in approximately 80% of surgically excised uterus specimens [6,7]. The diagnosis of adenomyosis is confirmed by microscopic examination of the uterus with ectopic endometrial

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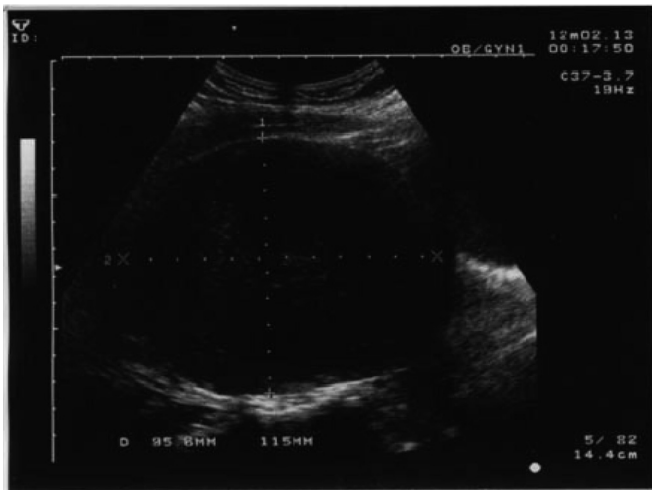


Fig. 1. Transabdominal sonography. Large myoma uteri is noted.



Fig. 3. Postcontrast computerized tomography. High-density area over the cervical–uterus junction raise concerns of active bleeding origin (red arrow).

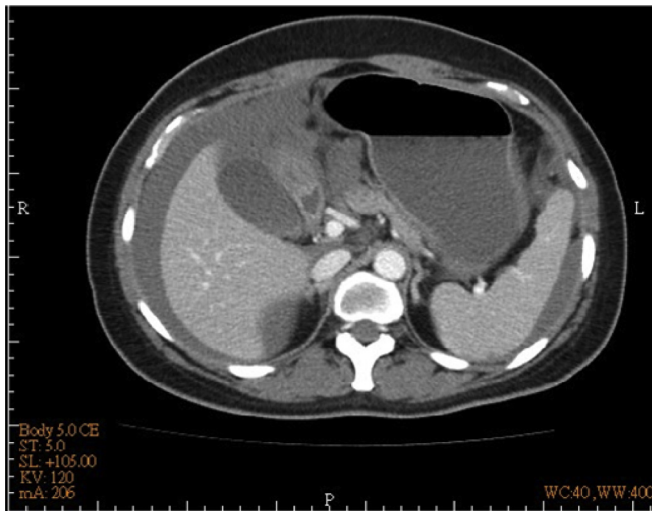


Fig. 2. Computerized tomography. Massive amount of intraperitoneal fluid suspicious of internal bleeding is noted.

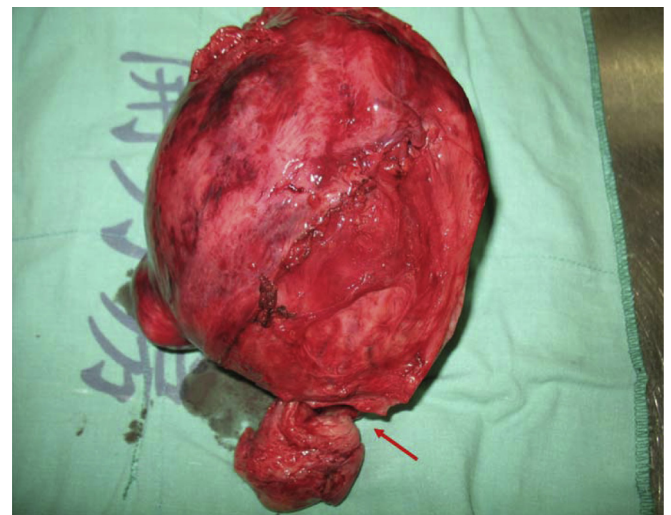


Fig. 4. Gross picture of the surgical specimen. A spontaneous laceration wound is located at the posterior cervical–uterus junction (red arrow).

glands appearing in the uterine muscle layer. It is estimated to affect 25–65% of women [8] and often coexists with leiomyomas. Both diseases mainly arise in women of reproductive age and typically present with classic symptoms of uterine enlargement, abnormal vaginal bleeding, pelvic pain, pressure, and reproductive dysfunction such as subinfertility or adverse pregnancy outcomes [9].

Rupture, internal bleeding, and hemoperitoneum are rare and atypical complications of leiomyomas and adenomyosis. A leiomyoma may hemorrhage owing to ruptured superficial vessels, either an engorged artery or a congested vein, and can potentially lead to hemorrhagic shock [10,11]. Leiomyomas > 10 cm in size and venous congestion have been reported as risk factors for the rupture of superficial vessels [12,13]. Other predisposing risk factors for uterine rupture from leiomyomas and adenomyosis include trauma, striking force on the abdomen, leiomyoma degeneration, multiparity, uterine distension with or without multiple pregnancies, prolonged steroid use, infection, and a scarred uterus owing to hysterectomy, laparotomy, or laparoscopy for myomectomy. Cases of spontaneous perforation of leiomyoma after red or cystic

degeneration have been reported in pregnant and nonpregnant patients [11,14,15]. A uterine rupture in pregnancy is usually associated with previous laparotomy or laparoscopy for myomectomy, which is an obstetrical emergency with increased maternal and neonatal morbidity and mortality. Rupture can occur in any pregnancy trimester and stage of labor, although it most commonly occurs during the third trimester or the intrapartum period. Twelve cases of rupture in a gravid uterus related to adenomyosis have been reported, with the locations of the rupture being the fundus, isthmus, or posterior cervical–uterus junction [16].

The clinical signs and symptoms of a uterine rupture include the sudden onset of lower abdominal pain with or without peritoneal irritation, nonreassuring fetal heart rate such as fetal bradycardia during pregnancy, hypotension, and hemorrhagic shock. In our case, the uterine rupture occurred without obvious risk factors, including no history of trauma, striking force on the abdomen, gravid status, leiomyoma degeneration, or previous surgical scars on the uterus. The patient presented with diffuse lower abdominal

tenderness complicated with hemorrhagic shock and coagulopathy. Ultrasound and computerized tomography were helpful in the tentative diagnosis of internal bleeding with hemoperitoneum as demonstrated by the massive amount of free fluid in the abdominal cavity. The management of a uterine rupture requires an emergency surgical intervention with concurrent resuscitation to stabilize the vital signs. We explained the risks and benefits of the surgical intervention to the patient, and quickly arranged for an exploratory laparotomy to confirm our suspicions. With the patient's consent, total hysterectomy was performed. In conclusion, although extremely rare, a uterine rupture can still occur in a nonpregnant and unscarred uterus with leiomyoma and adenomyosis. Clinical physicians should keep in mind this unusual differential diagnosis of acute abdominal pain in female patients of reproductive age. Early surgical intervention via laparoscopy or laparotomy may be considered in cases of uncertain acute abdominal pain with internal bleeding and hemoperitoneum.

Conflict of interest

The authors have no conflicts of interest statement.

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