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ABOUT COVER

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CASE REPORT

Uterine rupture due to adenomyosis in an adolescent: A case report and review of literature

Nah Ihm Kim, Ji Shin Lee, Jong Hee Nam

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Abstract

BACKGROUND

Uterine rupture is a fatal medical complication with a high mortality rate. Most cases of uterine rupture occur in late pregnancy or during labor and are mainly related to uterine scarring due to previous surgical procedures. Adenomyosis is a possible risk factor for uterine rupture. However, spontaneous uterine rupture due to severe adenomyosis in a non-gravida-teenaged female has not been reported in the literature to date.

CASE SUMMARY

A 16-year-old girl was referred to our hospital for acute abdominal pain and hypovolemic shock with a blood pressure of 90/50 mmHg. Radiologic studies revealed a huge endometrial mass with multiple nodules in the lung, suggesting lung metastasis. The patient underwent an emergency total hysterectomy and wedge resection of the lung nodules. Histologically, the uterus showed diffuse adenomyosis with glandular and stromal dissociation. Lung nodules were endometrioma with massive hemorrhage. Immunohistochemistry demonstrated that the tumor cells were positive for PAX8, ER, and PR expression, leading to a final diagnosis of pulmonary endometriosis and uterine adenomyosis. Following surgery, the patient remains in good condition without recurrence.

CONCLUSION

This is the first case of spontaneous uterine rupture due to adenomyosis in a non-gravida adolescent.

Key Words: Uterus; Adenomyosis; Malignancy; Endometrioma; Case report

Core Tip: Uterine adenomyosis is rare in adolescents but can lead to massive menorrhagia. Differential diagnoses, early detection, and therapeutic care must be provided to avoid hysterectomy in adolescents.

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INTRODUCTION

Adenomyosis is a commonly encountered estrogen-dependent disease in women across the lifespan, causing heavy menstrual bleeding, intense pelvic pain, and infertility[1]. Adenomyosis is associated with an increased risk of many obstetrical complications, such as uterine rupture, postpartum hemorrhage and fetal growth restriction[2].

Spontaneous uterine rupture due to adenomyosis in an adolescent and non-gravida female is extremely rare, with no cases reported in the literature. Here, we describe a unique case of uterine rupture due to adenomyosis with coexisting pulmonary endometriosis and review previously reported, similar cases[3-11].

CASE PRESENTATION

Chief complaints

A 16-year-old female visited the emergency department in hypovolemic shock.

History of present illness

The patient was obese (150 cm, 78 kg, body mass index 34.7 kg/m^2) and complained of dysmenorrhea that suddenly occurred a month ago.

History of past illness

The patient was suffering from irregular menstrual period, heavy menstrual bleeding and was on ferrous sulfate medication for anemia (hemoglobin levels 9.2 g/dL). She had no history of taking other medications, including oral contraceptives or hormonal agents.

Personal and family history

The patient was a virgin. She had no other significant personal or family history or previous surgical history. Menarche began at the age of 12.

Physical examination

Physical examination revealed a distended abdomen with diffuse abdominal tenderness. A palpable mass was not detected.

Laboratory examinations

Laboratory findings showed decreased hemoglobin levels (6 g/dL). LDH (1476 U/L), CA-125 (1063 U/mL), and CA19-9 (1347 U/mL) levels were significantly increased. An hCG blood test was negative.

Imaging examinations

An enhanced computed tomography (CT) scan of the entire abdominal pelvic cavity revealed a 13 cm × 12.5 cm × 10 cm heterogeneously enhancing mass in the uterine corpus, suggesting a uterine malignancy (sarcomatous change of uterine myoma with lung metastasis), such as rhabdomyosarcoma or leiomyosarcoma (Figure 1). The chest CT also revealed enhancing lesions in the right lower lung (3.5 cm) and left lower lung (1.5 cm), suggesting metastatic lesions.

FINAL DIAGNOSIS

The preoperative differential diagnosis was uterine malignancy, such as rhabdomyosarcoma or leiomyosarcoma, with lung metastasis. However, there was no histologic evidence of malignancy in the resected surgical specimen. The final diagnosis was diffuse adenomyosis with extensive hemorrhage. Histopathology after a wedge resection confirmed

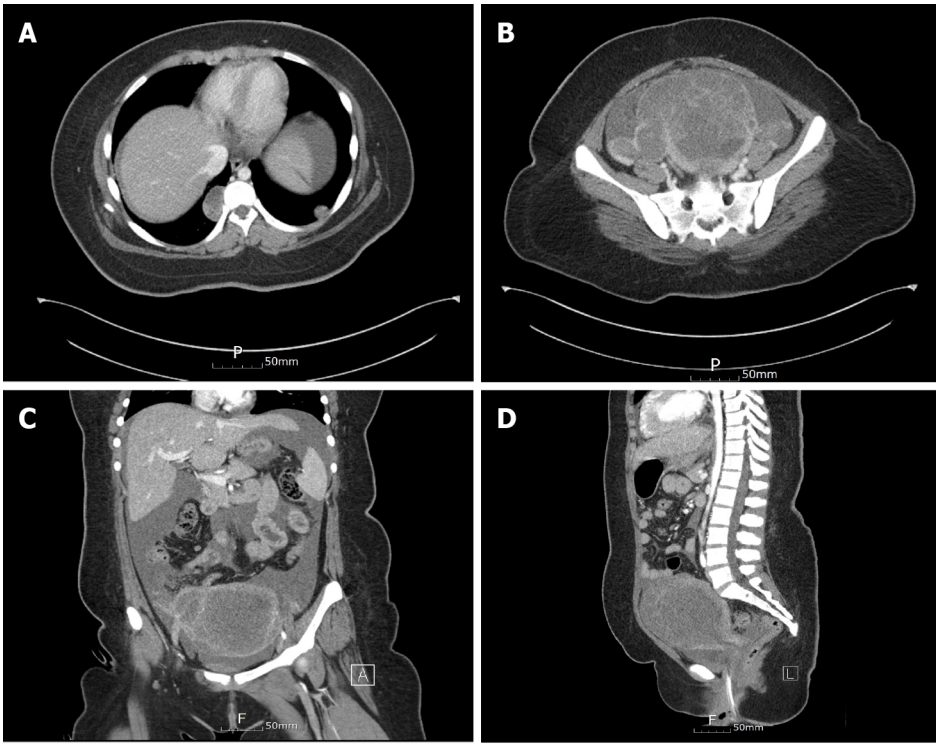

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Figure 1 Radiologic findings. A: Chest computed tomography scan revealed two nodules, suggesting metastatic lesions; B-D: Abdominal computed tomography scan demonstrated a huge mass in the uterine corpus, suggesting a uterine malignancy (B: Axial; C: Coronal; D: Sagittal).

pulmonary endometrioma of both lung nodules.

TREATMENT

The patient underwent a total hysterectomy with bilateral salpingectomy and wedge resection of the lung nodules. Intraoperative findings revealed a 4 L blood-filled abdominal cavity, a 15 cm sized huge necrotic tumor filling the uterus with blood, and multiple uterine perforations (Figure 2).

Macroscopic examination of the resected surgical specimen demonstrated an enlarged uterus with extensive hemorrhagic necrosis and hematoma. There were no definite mass-like lesions in the uterine corpus. Under low-power microscopy, the uterus showed extensive necrosis from the endometrium to the entire myometrium due to hemorrhage (Figure 3). Pathologic evaluation revealed diffuse adenomyosis with hemorrhagic necrosis and glandular and stromal dissociation.

While there was no evidence of malignancy in the uterine specimen, histology of the two lung nodules showed hemorrhagic cystic lesions lined by the cuboidal epithelium and surrounded by hemosiderin-laden macrophages. The cuboidal epithelium showed no cytologic atypia or mitotic activity. Immunohistochemistry was positive for CD10, PAX8, ER, and PR and negative for TTF-1. Ectopic endometrial glands outside the uterine cavity confirmed a final diagnosis of pulmonary endometriosis.

OUTCOME AND FOLLOW-UP

All elevated blood tests normalized after surgery. Following surgery, radiologic studies revealed no specific abnormalities. The patient is currently taking Diengest (Visanne) and is doing well without any side effects or recurrence.

DISCUSSION

Adenomyosis is a gynecologic disorder, associated with a high risk of obstetric complications and adverse pregnancy outcomes[12]. Uterine rupture is an obstetric emergency with a high incidence of morbidity and mortality. It mostly occurs during the third trimester of pregnancy or delivery, with a prevalence rate of 0.05% in pregnant women[13]. A history of surgery, such as a cesarean section or myomectomy, is the most common risk factor for uterine rupture[14]. Advanced maternal age, multiparity, uterine malformation, excessive uterine pressure, and rare intrauterine manipu-

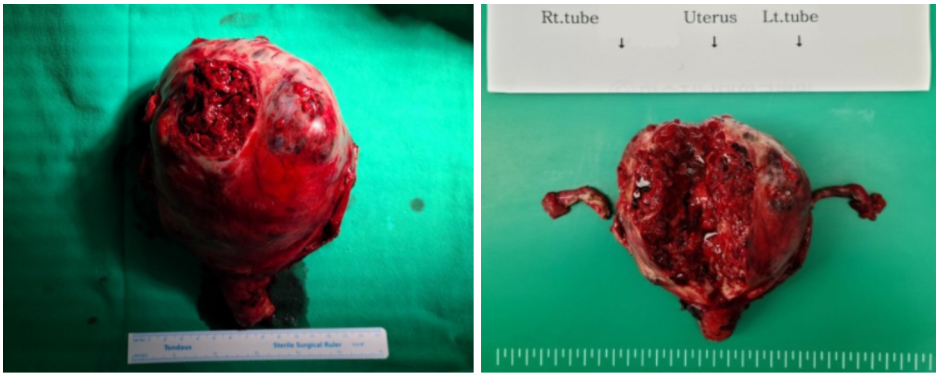

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Figure 2 Operative findings. Intraoperative findings revealed a huge necrotic mass with multiple uterine perforations.

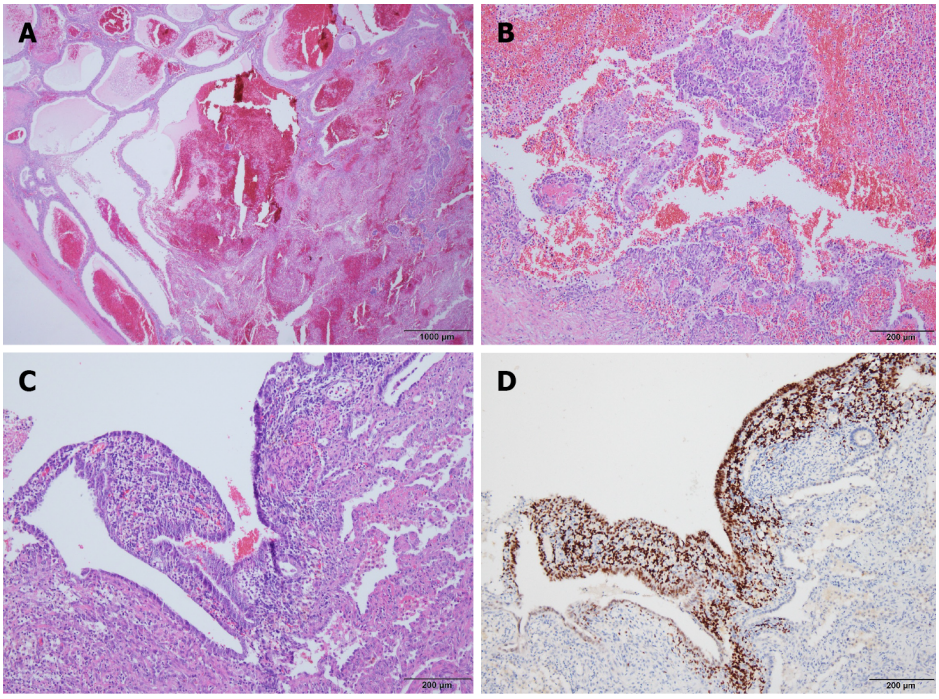

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Figure 3 Microscopic findings. A: Under low-power microscopy, the uterus showed diffuse adenomyosis with extensive hemorrhagic necrosis [Hematoxylin-and-eosin stain (H&E), × 20]; B: At a higher magnification, pathologic evaluation revealed glandular and stromal dissociation with necroinflammatory exudates (H&E, × 100); C: Histology of lung nodules demonstrated Müllerian type epithelium surrounded by endometrial stroma (H&E, × 100); D: Immunohistochemistry revealed positivity for ER in the tumor cells (immunohistochemistry, × 100).

lation are other important risk factors that may precipitate uterine rupture[15]. Spontaneous uterine rupture of an unscarred primigravid uterus is an extremely rare event[13,16]. Nikolaou *et al*[8] reported that nine of 12 cases of spontaneous uterine rupture were associated with adenomyosis.

Uterine adenomyosis involves the endometrial tissue growing into the uterine muscle wall of the uterus. This can cause painful menstrual periods, heavy bleeding, and pelvic pressure or discomfort. While adenomyosis mostly occurs in adult life, it can also involve adolescents in a mild to moderate form[17-23]. The exact cause of adenomyosis is unknown, but hormonal imbalances, uterine abnormalities, and certain medical conditions may increase the risk of this condition. Exacoustos *et al*[17] suggested using ultrasound as a diagnostic tool for adenomyosis could avoid the need for histologic diagnosis and facilitate appropriate management. Adenomyosis treatment may include medication or surgery in severe cases[1,22,24,25].

Obesity is associated with a higher risk of endometriosis and adenomyosis, although the exact relationship is unclear. The increasing incidence of adenomyosis and endometriosis in adolescents may be due to obesity[26]. Adenomyosis and endometriosis may increase the risk of obstetric complications[12]. Obesity can increase the risk of uterine rupture by increasing uterine pressure and may complicate the diagnosis of adenomyosis due to increased estrogen levels. Hormonal imbalances and inflammation may play a role in the development of both of these conditions in obese individuals[27].

Table 1 Cases of spontaneous uterine rupture due to adenomyosis

Ref.	No.	Age	Gravida/Para	Endometriosis	Pregnancy	Hysterectomy
Azziz[3] (1986)	1	41	NA/P10	NA	Yes	NA
	1	NA	NA	NA	Yes	NA
	1	25	NA/P0	NA	Yes	NA
	1	38	NA/P1	NA	Yes	NA
	1	33	NA/P0	NA	Yes	NA
	1	25	NA/P1	NA	Yes	NA
	1	26	NA/P3	NA	Yes	NA
Bensaid *et al*[4] (1996)	1	22	G1/P1	NA	Yes	No
Mueller *et al*[5] (1996)	1	30	G1/P0	No	Yes	Total hysterectomy
Pafumi *et al*[6] (2001)	1	30	G3/P2	No	Yes	Total hysterectomy
Villa *et al*[7] (2008)	1	30	G1/P1	Rectovaginal endometriosis	Yes	Total hysterectomy
Nikolaou *et al*[8] (2013)	1	33	G1/P1	Ovarian endometriosis	Yes	Subtotal hysterectomy
Indraccolo *et al*[9] (2015)	1	37	G2/P0	No	Yes	No
Li *et al*[10] (2021)	1	32	G1/P0	No	Yes	No
Vimercati *et al*[11] (2022)	1	27	G0/P0	No	Yes	Total hysterectomy
Present case	1	16	G0/P0	Pulmonary endometriosis	No	Total hysterectomy

NA: Not available.

However, hormones could cause different reactions to the adenomyotic stroma, especially in pregnant individuals. The adenomyotic stroma has two response patterns to pregnancy-related hormones. One is the superficial foci of adenomyosis, which are located within the endometrium's basal layer with no to minimal decidualization. In the second pattern, deeper adenomyosis foci could exhibit prominent decidualization[3]. Higher expression of progesterone receptors in the stromal components of adenomyosis is likely related to stromal decidualization, supporting the theory that adenomyosis is a response to progesterone during pregnancy[8]. Furthermore, abundant decidual transformation of stromal cells in adenomyosis results in atrophy and necrosis of muscle cells[7]. Necrosis of the uterine muscles causes atony and muscle cell separation, leading to life-threatening rupture[28].

This could explain the spontaneous uterine rupture due to adenomyosis in pregnant women. About 15 cases of spontaneous uterine rupture due to adenomyosis have been reported to date (Table 1). Azziz[3] reviewed 11 cases of uterine rupture, seven of which were associated with adenomyosis. Uccella *et al*[29] reviewed the literature and found that 1 in 25 reported cases of prelabor spontaneous uterine rupture involved adenomyosis. Mueller *et al*[5] reported a primigravida woman who experienced spontaneous uterine rupture at 18 wk of gestation due to heavily decidualized adenomyosis. Nikolaou *et al*[8] reported a case of rupture of an unscarred uterus caused by multiple foci of adenomyosis with a marked decidual reaction in the adenomyotic stroma. Indraccolo *et al*[9] also reported a woman with uterine rupture caused by adenomyosis.

However, all previously reported cases were related to pregnancy, and the current case in a nulligravida juvenile patient is the first reported to date. The spontaneous uterine rupture in this nulligravida adolescent girl may be due to increased uterine pressure and changes in estrogen due to her obesity. We believe that transmural adenomyotic foci with significant hemorrhage and subsequent splaying of the myometrial smooth muscle fibers may have weakened the myometrium, ultimately rupturing the uterus.

Uterine rupture, a rare adenomyosis complication, can be fatal if not treated immediately. It can be difficult to diagnose adenomyosis, as the symptoms are similar to those of other conditions, such as endometriosis[30,31]. A preoperative diagnosis of spontaneous uterine rupture is also challenging, especially in a juvenile patient with nulligravida. Unrecognized adenomyosis is particularly problematic in younger patients[32]. The standard workup for all women who present with severe dysmenorrhea and heavy menstrual bleeding should include an evaluation for adenomyosis, regardless of their age and health conditions. Regular monitoring is key to managing the risks associated with adenomyosis, especially in obese, nulliparous teenagers. Early detection may lower the risk of associated infertility and adverse obstetric outcomes, including uterine rupture[11].

The present case highlights the importance of considering a spontaneous uterine rupture diagnosis in women with a history of adenomyosis, regardless of their parity. Extensive adenomyosis may contribute to uterine wall weakness and increase the risk of uterine rupture, even in women who are not pregnant. If adenomyosis is detected early, fertility can be preserved with medical treatment and hysterectomy avoided.

CONCLUSION

Here we present an extraordinary case of spontaneous uterine rupture due to adenomyosis in a nulliparous adolescent. Uterine rupture should be considered for all female patients with adenomyosis, regardless of gestational status and history. It should be distinguished from other neoplastic conditions and early detection may lower the risk of adverse obstetric outcomes, including uterine rupture.

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FOOTNOTES

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