



Contents lists available at ScienceDirect

## Taiwanese Journal of Obstetrics &amp; Gynecology

journal homepage: [www.tjog-online.com](http://www.tjog-online.com)

## Research Letter

## A rapidly growing adenomyosis associated with preterm delivery and postpartum abscess formation

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## ARTICLE INFO

## Article history:

Accepted 5 February 2015

## Dear Editor,

Adenomyosis usually occurs in women of reproductive age, and poor pregnancy outcomes in women with adenomyosis have been reported. The incidence of adenomyosis associated with pregnancy appears to be 17% [1]. Recent reports showed that pregnant women with adenomyosis are at elevated risks of miscarriage, preterm delivery, preterm premature rupture of membranes, spontaneous uterine rupture during labor, and postpartum hemorrhage [2].

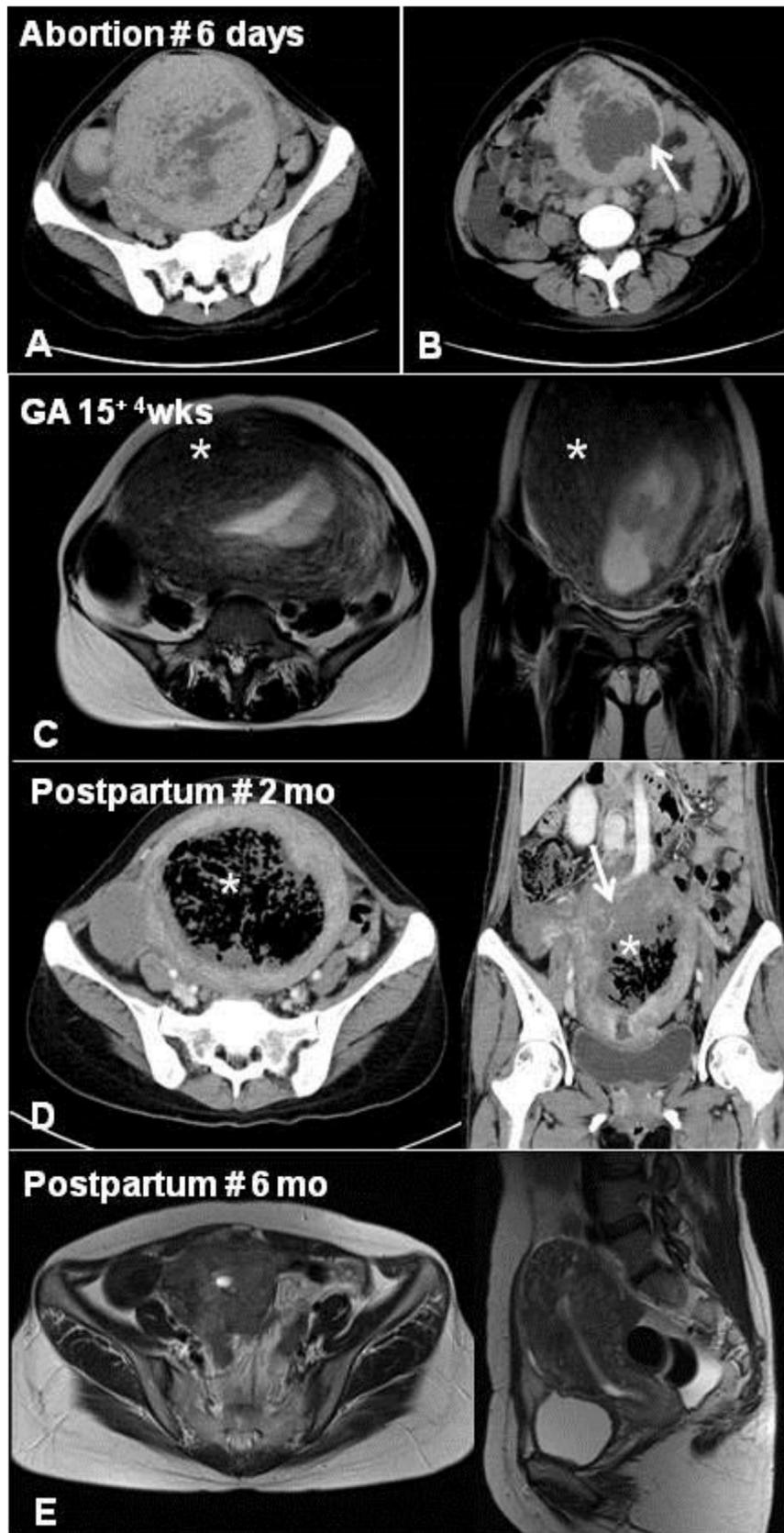
Here, we report a rare case of a rapidly growing adenomyosis associated with preterm delivery and postpartum abscess formation within adenomyosis. A 34-year-old nulliparous woman visited our hospital for antenatal care at 6<sup>+4</sup> weeks (6 weeks and 4 days) of pregnancy. She presented with a previous history of menorrhagia and dysmenorrhea. She had been diagnosed with adenomyosis during a prior pregnancy and previously underwent uterine curettage for septic abortion (Figure 1A and B). Magnetic resonance imaging (MRI) at 15<sup>+4</sup> weeks demonstrated diffuse enlargement of the uterus with adenomyosis, which was of homogeneous, low-signal intensity according to T1-weighted images (Figure 1C). At 19 weeks, she was admitted due to irregular uterine contractions and a reduced cervical length (1.9 cm). At 22<sup>+4</sup> weeks and 29<sup>+2</sup> weeks, cervical length was further reduced to 0.95 cm and 0.2 cm, respectively. At 30 weeks, membrane bulging into the vagina was noted. Cesarean delivery was performed due to breech presentation with powerful contractions and remarkable cervical changes. She had an

uneventful postoperative course, and was discharged on Post-operative Day 5. During follow-up visits at 2-weeks and 6-weeks postpartum, uterine enlargement was still evident. At approximately 8 weeks after delivery, she developed a high fever and experienced a profuse, foul, odorous discharge after completing her first menstruation. Computerized tomography depicted an intra-uterine, necrotic, degenerated cystic mass of 12 cm in diameter containing air bubbles (Figure 1D). The mass resembled one cystic space with multiple septa and papillae within the uterus and was connected to the endometrial cavity. It was presumed to be an abscess within the myometrium. Gram-positive bacteria (nonspore forming bacilli) were cultured from the vaginal discharge. Intravenous antibiotics were administered, and transabdominal catheter drainage was recommended, but she refused the procedure. During subsequent observation following antibiotic administration, she became afebrile. The purulent materials drained spontaneously for approximately 2 weeks, and she was discharged at 10<sup>+5</sup> weeks after delivery. At 6-months postpartum, pelvic MRI revealed no more evidence of abscess accumulation (Figure 1E).

Although a few reports have described abscess formation in endometriotic foci, reports on abscess formation arising within adenomyosis are rare. Erguvan et al [3] reported the first case of abscess in postmenopausal woman (Table 1) [3–5], although the authors did not demonstrate how the abscess formed. Functional changes of adenomyosis during pregnancy and after delivery have also been reported. These cases displayed decidualization and hemorrhage within adenomyosis. In those cases, a relative ischemic condition might develop after childbirth due to rapid decrease in blood flow to an enlarged adenomyosis [6]. Here, we report for the first time development of an abscess within adenomyosis during postpartum involution. Contributory factors, such as hemorrhage inside the adenomyotic lesions or ischemia after childbirth, might play roles in the development of postpartum abscess. We suggest that the resumption of menstruation might be another factor. Menstruation is considered an inflammatory process [7], during which intramyometrial menstrual flow and endometrial shedding from the ectopic endometrium within adenomyosis might precipitate abscess formation. In our case, abscess occurred 3 days after completing the first menstruation after delivery.

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**Figure 1.** (A) Contrast-enhanced CT performed 6 days after abortion and showing a diffusely enlarged uterus and multiple cystic lesions within the myometrium of the uterine body suggestive of uterine adenomyosis. (B) Contrast-enhanced CT at the upper level showing a large cystic lesion (arrow) suspicious of abscess formation within the myometrium of the uterine body. (C) At 15 weeks and 4 days of gestation, axial (left) and coronal (right) T2-weighted MRI showed marked diffuse enlargement of the uterine myometrium (\*) with low signal intensity. Rapid enlargement of the fundus and body displaced the gestational sac to the lower segment. (D) At 2-months postpartum, axial (left) and coronal-reformatted (right) contrast-enhanced CT showed a large abscess containing air bubbles (\*) in the endometrial cavity. The abscess communicated with adenomyosis (arrow) in the uterine fundus. (E) At 6-months postpartum, axial (left) and sagittal (right) T2-weighted MRI depicted diffuse uterine adenomyosis and no abscess formation in a well-involved uterus. CT = computed tomography; MRI = magnetic resonance imaging.

**Table 1**  
Clinicopathologic findings of abscess formation arising within adenomyosis.

Authors (Year)	Age (y)	Symptoms	Procedures
Erguvan et al [3] (2003)	54	Inguinal pain, night sweats, hot flashes	Total abdominal hysterectomy, bilateral salpingo-oophorectomy
Weng et al [4] (2013)	50	Fever, persistent vaginal bleeding, lower abdominal pain	Total abdominal hysterectomy
Wu et al [5] (2014)	49	Fever of unknown etiology	Subtotal hysterectomy
Present case (2015)	34	Fever, foul odorous discharge	No surgical procedures, spontaneous drainage

Surgical intervention was performed, because malignancy was suspected or response to antibiotics was lacking in previous cases. We do not know how spontaneous drainage of the abscess occurred in our case. Communications between the endometrial cavity and adenomyotic lesions were previously detected by sonohysterography [8]. We speculate that these communications became more apparent during the postpartum period and that the diffuse abscess drained spontaneously through these communications.

Our study has a limitation. No pathologic examination was performed for a definitive diagnosis; however, diagnosis was based on imaging findings and clinical suspicion of adenomyosis. In summary, the present case suggests that adenomyosis can rapidly enlarge during pregnancy and be associated with inflammatory consequences, such as septic abortion, preterm delivery, and postpartum abscess formation. Physicians should be aware of the potential complications of adenomyosis in pregnancy. Further studies are needed to elucidate the causes of abscess formation within adenomyosis and to explore its inflammatory nature.

#### Conflicts of interest

The authors have no conflicts of interest relevant to this article.

#### Acknowledgments

This work was supported for two years by Pusan National University Research Grant.

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