

Septic shock caused by abscess formation inside cystic adenomyosis after responsive treatment of pelvic inflammatory disease

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Summary

Objective: To report on septic shock in a patient with an abscess inside cystic adenomyosis after responsive treatment of pelvic inflammatory disease. **Case report:** A 51-year-old woman with known cystic adenomyosis was admitted to the hospital due to fever and low abdominal pain. In suspicion of pelvic inflammatory disease, intravenous antibiotics were administered. As her symptoms and signs of inflammation improved, she was discharged from the hospital after five days. However, after completing 14 days of oral antibiotics, her symptoms aggravated with decreased blood pressure. Magnetic resonance imaging indicated abscess formation inside cystic adenomyosis, and total hysterectomy was performed. Postoperatively, she fell into shock condition. Intravenous antibiotics were administered for 11 days and she was discharged from the hospital with no adverse event. **Conclusion:** Physicians should be aware of the possibility of septic shock caused by abscess formation inside known cystic adenomyosis.

Key words: Abscess; Adenomyosis; Pelvic inflammatory disease, Sepsis; Shock.

Introduction

Sepsis is life-threatening organ dysfunction caused by a dysregulated host response to infection [1]. Septic shock is a subset of sepsis in which the circulatory and cellular metabolism abnormalities are so severe that they can cause death [1]. Infection that can lead to septic shock occurs through various routes, such as the respiratory, gastrointestinal, and genitourinary tracts or soft tissue. Severe sepsis associated with pregnancy or pelvic inflammatory disease has been reported [2, 3], but it has rarely been associated with adenomyosis. Here, we report on septic shock in a patient with a significant abscess inside cystic adenomyosis after responsive treatment of pelvic inflammatory disease.

Case Presentation

A 51-year-old woman was admitted to Chungnam National University Hospital due to fever and low abdominal discomfort in May 2019. She had one cesarean delivery and no abortion history. She had been taking hypertensive medication for four years. She was diagnosed with adenomyosis in 2006 by ultrasonography and had regular follow up at 2-year intervals. She was menstruating regularly without menorrhagia or dysmenorrhea. Pelvic sonography, as well as pelvic computed tomography, showed a cystic adenomyosis lesion increased in size compared to a pelvic computed tomography performed in January 2017 (from 35 mm × 16 mm to 49 mm × 27 mm) (Figure 1A and 1B).

Fever, low abdominal discomfort, and turbid bloody dis-

charge started two days prior to admission. At the time of admission, the patient's leukocyte count was 11,440/ μ L with 88% neutrophils and the C-reactive protein 11.8 mg/dL (Table 1). Her blood pressure was 130/80 mmHg, body temperature 38.5 °C, pulse rate 88/minutes and respiratory rate was 18/minutes (Figure 2). Cervical motion and adnexal tenderness were noted by bimanual examination. Intravenous antibiotics (ceftriaxone 1g twice a day and metronidazole 500 mg three times a day) were administered for six days for the suspicion of pelvic inflammatory disease. Blood samples and vaginal discharge were cultured but no microorganisms were identified. The patient was afebrile for four days and her abdominal discomfort, as well as vaginal discharge, improved. The leukocyte count was decreased to 9,110/ μ L with 70.0% neutrophils and the C-reactive protein decreased to almost normal. With the clinical and laboratory improvements, the patient was discharged from the hospital. Oral antibiotics (cefditoren pivoxil 100 mg three times a day and metronidazole 250 mg three times a day) were prescribed for seven days to complete 14 days of treatment.

Three days after completing two weeks of antibiotic treatment, the patient revisited the outpatient clinic for follow-up. Her body temperature was 38.8 °C and the uterus was enlarged with severe tenderness. Her pulse rate was 88/minute, respiratory rate was 20/minute, and her blood pressure was 99/63 mmHg despite omission of hypertensive medication for two days. Shock index was elevated to 0.89, but there was no sign of vaginal bleed-

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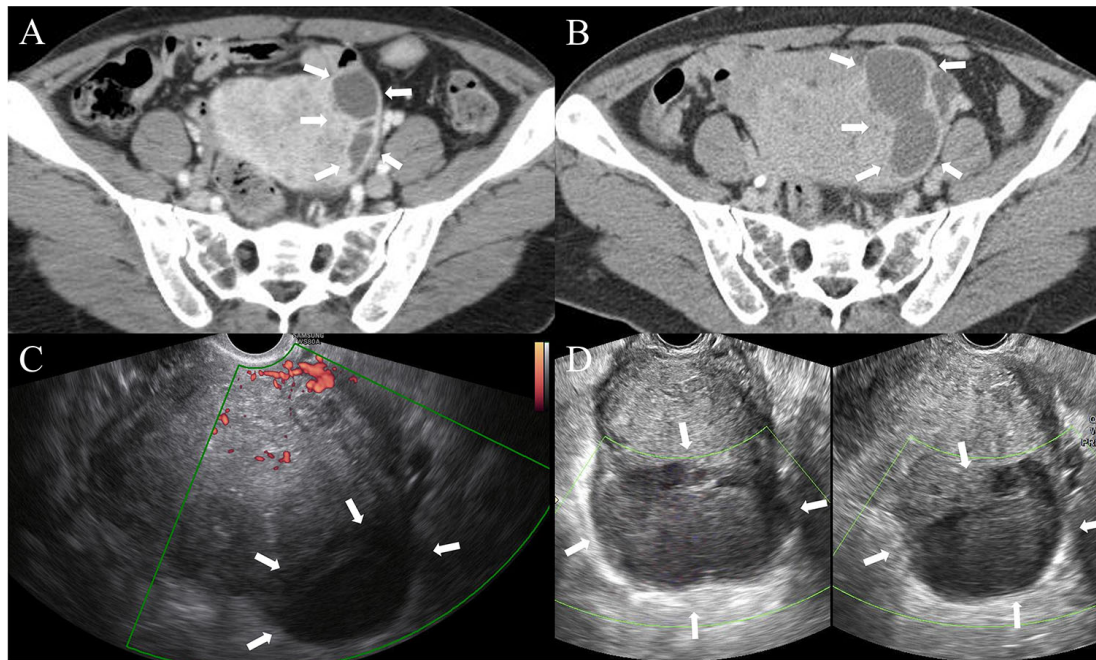


Figure 1. — Computed tomography imaging of cystic adenomyosis (A) in January 2017 and (B) at initial admission in May 2019 (white arrows indicate the cystic adenomyosis). Ultrasonographic imaging of cystic adenomyosis (C) at initial admission in May 2019 and (D) at second admission in June 2019 (white arrows indicate the cystic adenomyosis).

Table 1. — *Laboratory data of the patient.*

Laboratory data	HD 1 of initial admission	HD 5 of initial admission	HD 1 of readmission	HD 3 of readmission (OP day)	HD 15 of readmission
WBC ($\times 10^3/\mu\text{L}$)	11.44	9.11	17.5	20.99	5.49
Neutrophils (%)	88.0	70.0	88.1	90.2	61.8
Hb (g/dL)	13.3	12.6	11.4	10.6	11.6
Platelet ($10^3/\mu\text{L}$)	328	368	461	293	375
AST (U/L)	11	21	13	38	24
ALT (U/L)	58	13	9	14	24
BUN (mg/dL)	12.0	10.0	9.0	8.1	16.0
Creatinine (mg/dL)	0.94	0.92	0.75	0.64	0.64
CRP (mg/dL)	11.8	4.2	15.4	14.3	0.6

ALT, alanine aminotransferase; AST, aspartate aminotransferase; BUN, blood urea nitrogen; CRP, C reactive protein; Hb, Hemoglobin; HD, hospital day; OP, operation; WBC, white blood cell.

ing or hemoperitoneum [4]. Transvaginal ultrasonography showed the cystic mass within the uterus had increased in size from 50 mm \times 32 mm to 85 mm \times 53 mm (Figure 1C and 1D). The patient was re-admitted and intravenous antibiotics (piperacillin 4 g/tazobactam 500 mg three times a day) were administered. Her leukocyte count was 17,500/ μL with 88.1% neutrophils and the C-reactive protein was 15.4 mg/dL at the time of re-admission. Magnetic resonance imaging showed increased cystic lesion size inside the left side of the myometrium with diffusion restriction without significant enhancement, suggesting abscess formation from the previous adenomyosis with inner cystic degeneration (Figure 3A and 3B). Two days after admission, her fever and abdominal pain persisted despite treat-

ment with antibiotics, her blood pressure remained around 90/50 mmHg without hypertensive medication, and laboratory studies showed persistent severe infection.

A total hysterectomy was performed three days after re-admission and a large, cystic adenomyotic uterus was discovered (Figure 4A). Small amounts of pus inside the abdominal cavity and severe adhesions were identified. Both ovaries and fallopian tubes were normal. The frozen section revealed an abscess within an adenomyosis lesion. Purulent discharge was present inside the adenomyotic lesion and endometriotic cavity. The estimated blood loss during surgery was 400 mL and the patient's vital signs remained stable throughout the operation. Histopathologic findings of the myometrium section revealed diffuse thick-

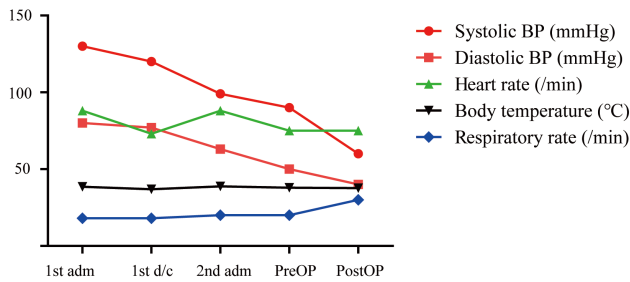


Figure 2. — Patient's blood pressure, heart rate, body temperature and respiratory rate. *1st adm, first admission; 1st d/c, first discharge; 2nd adm, second admission; PreOP, Pre-operation; PostOP, Post-operation

ening from adenomyosis and cystic change with purulent inflammation (Figure 4B and 4C). A tissue section obtained during surgery was cultured but neither aerobic nor anaerobic microorganisms were identified. No microorganisms were identified from two subsequent blood samples.

Postoperatively, the patient's blood pressure dropped to 60/40 mmHg and her respiratory rate increased to 30/minute without response to aggressive crystalloid fluid loading. She was conscious with no significant cold sweat or peripheral cyanosis and her pulse rate remained between 70–80/minute. Her quick sequential organ failure assessment score exceeded 2, and her serum lactate level was 4.9 mmol/L [1]. She was transferred to the intensive care unit for close monitoring and management. After continuous infusion of norepinephrine her vital signs normalized the next day. With intravenous antibiotic treatment, her fever slowly subsided and she remained afebrile from three days after surgery. Mild pleural effusion was identified by a simple chest X-ray due to excessive fluid loading during shock and resolved after administration of diuretics for two days. The patient was discharged from the hospital 14 days post-operatively.

Discussion

Here, we report a rare case of septic shock caused by abscess formation inside cystic adenomyosis. Abscess formation inside adenomyosis is extremely rare. To our knowledge, only two such cases have been reported [5, 6]. Abscess formation in adenomyosis was first described by Erguvan in 2013 [5]. A 54-year old postmenopausal woman with inguinal pain underwent hysterectomy in suspicion of uterine malignancy. She was afebrile but histopathology revealed an abscess inside adenomyosis. Results from tissue culture were inconclusive. The second case was reported in 2013 by Weng *et al.* [6]. A 50-year-old woman with persistent vaginal bleeding, lower abdominal pain and fever underwent hysterectomy due to persistent symptoms despite intravenous administration of antibiotics. Histopathology revealed microabscess in the foci of adenomyosis. Microorganism that caused the infection were not mentioned in the manuscript. Similarly, no microorganism has been

identified despite significant abscess formation in our case. However, infection leading to abscess formation in adenomyosis were not severe enough to cause septic shock in both two previous reports. Our patient had an extremely rare case of septic shock caused by abscess formation inside a previously identified degenerative cystic lesion of adenomyosis despite responsive treatment of pelvic inflammatory disease.

Sepsis is a life-threatening condition caused when the body's response to an infection causes injuries to its own organs [1]. Benign gynecologic disease has been rarely related to sepsis or septic shock. Pelvic inflammatory disease itself is rarely associated with sepsis. However, tubo-ovarian abscess that arise from pelvic inflammatory disease can progress into sepsis, especially when ruptured [7]. Ruptured tubo-ovarian abscesses, pyometra or pyomyoma that led to sepsis or septic shock have been reported in some case reports or case series [8, 9]. In this case, the patient progressed to septic shock without ruptured abscess, and despite successful treatment of pelvic inflammatory disease with intravenous antibiotics for the first five days.

Most common pathogens associated with PID are sexually transmitted pathogens such as *Neisseria gonorrhoeae*, *Chlamydia trachomatis* and *Mycoplasma genitalium* [10]. However, in our case all cultures including abscess tissue, pus inside abscess pocket, urine and blood came out negative. Two previous reports on abscess inside adenomyotic foci could not find any specific microorganism isolated from the abscess either [5, 6]. Negative culture results have been reported in brain abscess and liver abscess [11, 12]. Gene analysis such as next generation sequencing can be the alternative diagnostic tool for identifying pathogens in such culture negative abscess [11]. Following mechanism is our hypothesis regarding pathogenesis of abscess formation inside cystic adenomyosis and septic shock. Initially pelvic inflammation developed due to ascending infection via vagina. At first admission, first antibiotics treatment was successful because pelvic inflammatory disease did not affect cystic adenomyosis. However, remnant infection that was not treated with 14 days of antibiotics remained and spread into cystic adenomyosis and formed abscess. Once abscess has been formed, antibiotics were not successfully delivered because of poor perfusion inside abscess cavity. Although the patient developed septic shock right after the operation, she recovered quickly from the shock. We think it was not only because of the removal of the abscess itself, but also because of extensive peritoneal lavage. Intraoperatively, peritoneal cavity was washed with 4 liters of normal saline. Extensive intraperitoneal lavage has been proven to be effective in peritoneal infections [13].

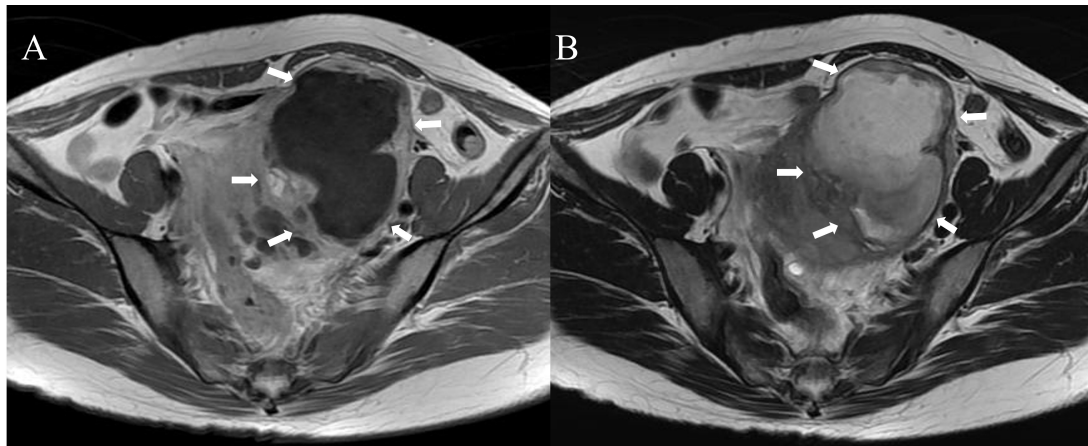


Figure 3. — Magnetic resonance imaging of abscess formation inside cystic adenomyosis. (A) T1 weighted image after enhancement and (B) T2 weighted image (white arrows indicate abscess formation inside cystic adenomyosis).

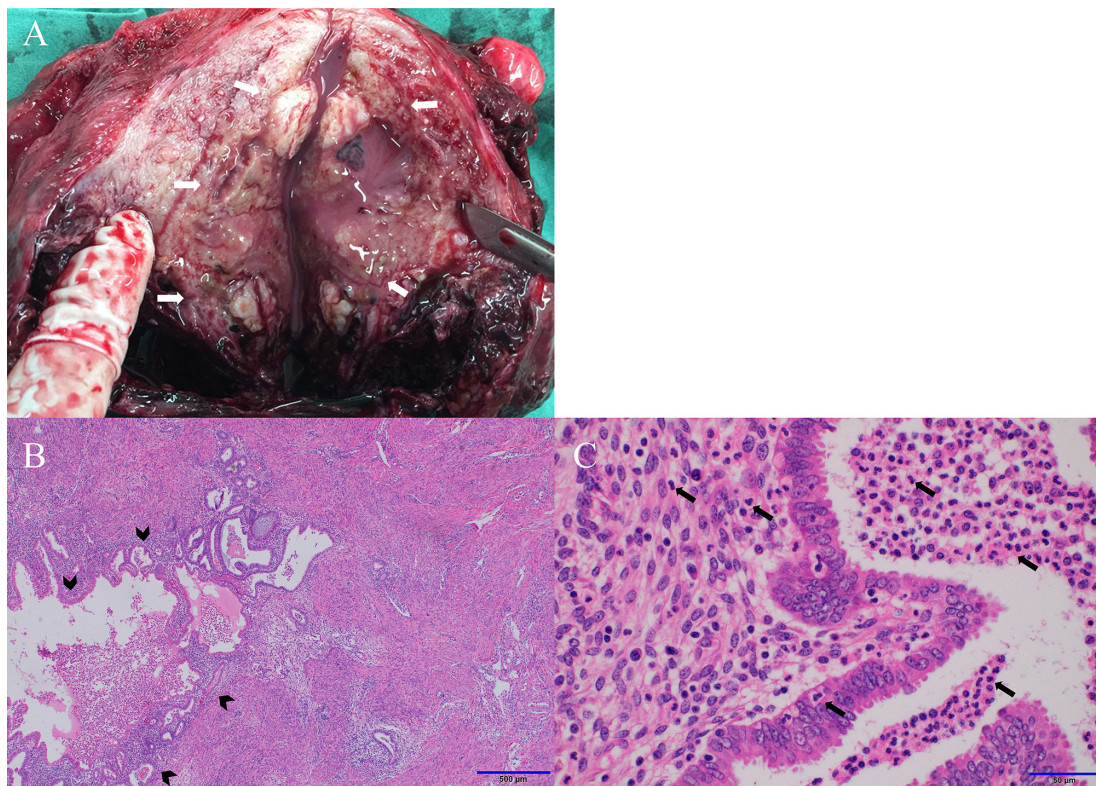


Figure 4. — Uterus after hysterectomy and bilateral salpingo-oophorectomy (A) Gross imaging of the uterus after surgery (white arrows indicate abscess formation inside cystic adenomyosis) (B and C) Histopathologic finding of the uterus (B) adenomyotic glands inside myometrium (C) neutrophil infiltration in myometrium, glands, and exudates.

Most adenomyosis presents with diffuse or focal thickening of the myometrial junctional zone. Less common forms of adenomyosis include adenomyoma, adenomyomatous polyps, and cystic adenomyoma [14]. Cystic adenomyosis or adenomyotic cysts are rare forms of adenomyosis that have been reported in less than 100 case reports and small case series [15, 16]. Cystic adenomyosis is most common in adolescents and women under the age of 31. Symptoms associated with cystic adenomyosis, like those

of other types of adenomyosis, are dysmenorrhea, menorrhagia, and chronic pelvic pain [17]. We had a rare case of a 51-year-old patient who had been asymptomatic for ten years with cystic adenomyosis.

To our knowledge, this is the first case of septic shock caused by abscess formation inside a previously identified degenerative cystic lesion of adenomyosis despite responsive treatment of pelvic inflammatory disease. Physicians should be aware of the possibility of septic shock caused by

abscess formation inside cystic adenomyosis.

Ethics Approval and Consent to Participate

The patient gave her informed consent for this case report before her medical chart was reviewed. This case report was conducted in accordance with the Declaration of Helsinki, and was approved by the Ethics Committee of Chungnam National University Hospital (2020-06-072).

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Conflict of Interest

The authors declare no conflict of interest.

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