PYOMYOMA: A RARE AND LIFE-THREATENING COMPLICATION OF UTERINE LEIOMYOMA

Zoe Hui-Ying Chen, Horng-Der Tsai, Mou-Jong Sun*

Department of Obstetrics and Gynecology, Changhua Christian Hospital, Changhua, Taiwan.

Despite the high prevalence of pelvic inflammatory disease and uterine leiomyomas in women, an intramyometrial abscess is a rare gynecological infection. Since 1945, only 22 cases of pyomyoma have been documented in the literature and confirmed by postmortem or postoperative pathological examinations. All patients had predisposed risk factors such as a gynecological operation, pregnancy, vascular insufficiency, or immunodeficiency [1]. We herein present a case of a woman with no known risk factors for tumor-associated infections and briefly review other cases previously reported in the literature.

A 46-year-old woman who had experienced three cesarean sections, G3P3, was admitted for a hysterectomy. She had a history of an asymptomatic uterine leiomyoma for 10 years without regular follow-up examinations. The last clinical evaluation was done in 2004, 1 week following a cesarean delivery, at which time her ultrasound examination showed no specific findings except for a homogenous uterine tumor, 9 cm in diameter. According to the patient's statements and previous medical records, she formerly had a regular menstrual cycle with normal flow. Two weeks before this admission, she reported an intermittent low-grade fever which was accompanied by mild rhinorrhea without abdominal pain or pelvic discomfort. She suspected that it was due to a common cold and so she did not take any medication. The patient sought evaluation at our hospital due to an episode of heavy menstrual flow and severe dysmenorrhea. Pelvic examination revealed a non-tender, fixed mass at 16 weeks' gestation, with no abnormal discharge from the cervix. Transabdominal ultrasound revealed a tumor, measuring $14.3 \times 12 \times 8$ cm, including a cystic configuration located over the anterior lower segment of the uterus (Figure 1). Lab data from the first visit to our clinic also showed mild leukocytosis (white blood cells $= 13 \times 10^9$/L) and anemia (hemoglobin = 7.9 mg/dL). Due to diagnosis of a uterine leiomyoma and abnormal uterine bleeding, a total abdominal hysterectomy was arranged and performed in November 2006.

During the laparotomy, adhesions between the uterus and abdominal wall were observed. A tumor was located over the anterior lower segment of the uterus, which was tightly adherent to the bladder and pelvic sidewall. The tumor mass was well circumscribed with a capsule without extension or communication to the endometrial cavity. While dissecting the mass, the tumor ruptured and emitted a malodorous purulent fluid. A culture of the purulent discharge was obtained intraoperatively. Due to the necrotic character of the mass, malignancy was suspected, and the specimen was immediately sent for pathological frozen section examination. However, the preliminary pathological examination indicated a uterine abscess without neoplastic changes (Figures 2 and 3). The abdominal cavity was irrigated with a large amount of normal saline solution before the wound was surgically closed. After the operation, the patient presented with a persistent temperature elevation to 38°C and a low systolic pressure of around 95 mmHg. Blood cultures were done before experimental antibiotic treatment was begun. The complete blood cell count with differential classification (white blood cells $= 22 \times 10^9$/L; segment = 86%, band = 2%) and C-reactive protein (12.37 mg/dL) were determined during hospitalization. The fever subsided 3 days after intensive care, a blood transfusion, and broad-spectrum antibiotic treatment using cefazoline, gentamycin, and clindamycin. The cultures from the pus demonstrated growth of *Escherichia coli* 5 days after the operation and we adjusted the antibiotics based on a bacterial sensitivity test. The patient responded well to surgery and antibiotic treatment, and was discharged on the eighth day after the operation. The patient returned to our outpatient department 10 days after the operation for follow-up. The lab data were all within normal limits and the surgical wound was healing well.

*Correspondence to: Dr Mou-Jong Sun, 135, Nanhsiao Street, Changhua 500, Taiwan.
E-mail: sun.mj@msa.hinet.net or 13212@cch.org.tw
Accepted: July 22, 2009

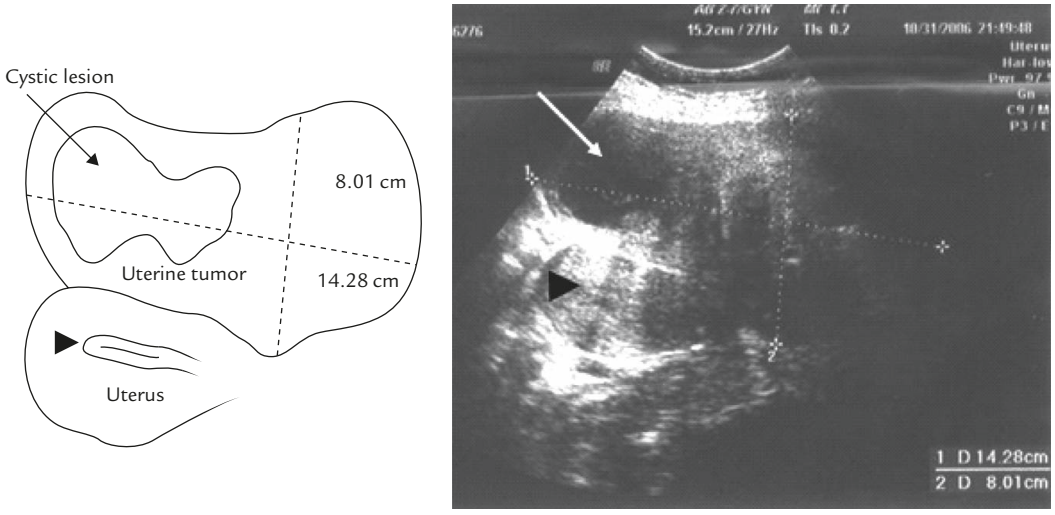

Figure 1. Abdominal sonography shows a midline sagittal section of the myoma and uterus (arrowhead). Note the significant hypoechogenicity in the upper portion of the myoma (arrow) compared with the cystic lesion of the gross specimen.

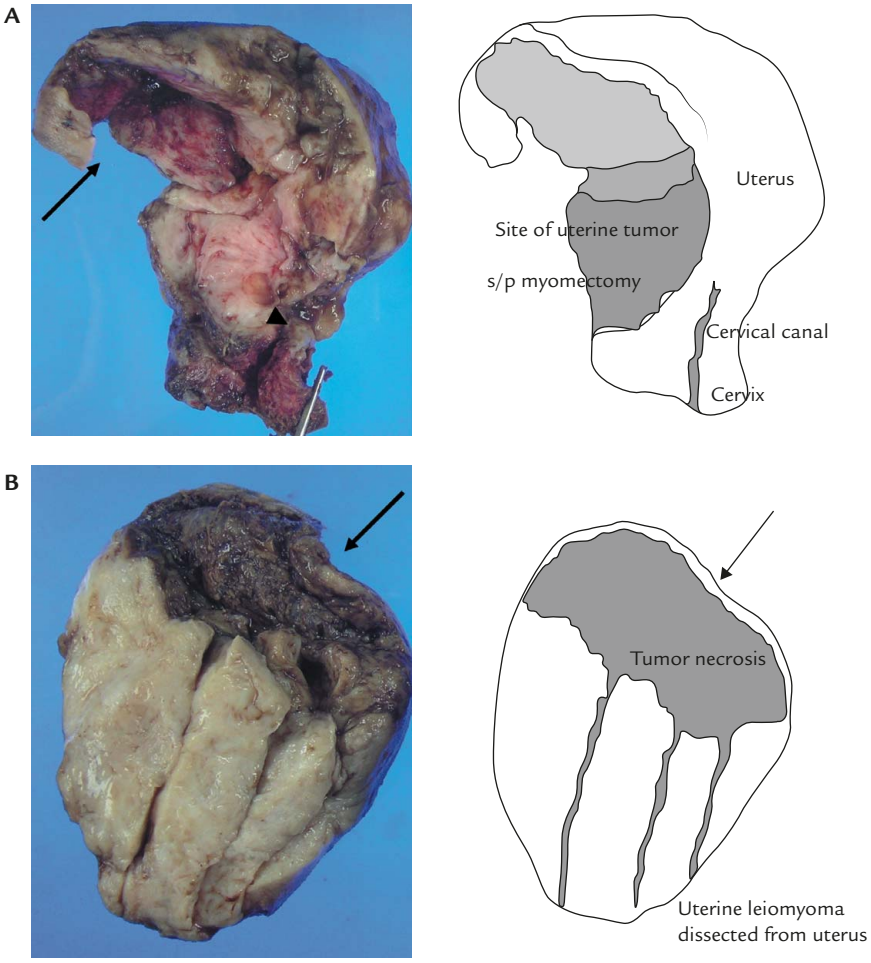

Figure 2. (A) Uterus fixed in formalin seen laterally, revealing the remaining capsule (arrow) of the tumor after a myomectomy. There is a well-circumscribed myoma located over the anterior wall of the uterus. No obvious communication between tumor and endometrial cavity was evident. (B) The myoma dissected from the anterior wall of the uterus. The cystic lesion with tissue necrosis was localized in the upper part of the myoma (arrow). The cut surface of the tumor was whitish to yellowish in color, with a whorled trabecular pattern.

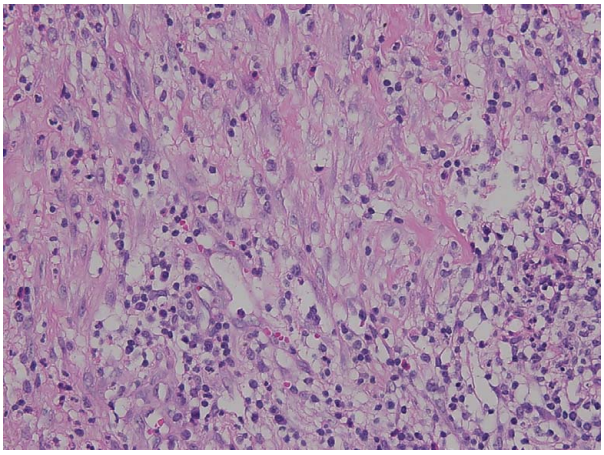

Figure 3. Hematoxylin and eosin-stained section of the leiomyoma shows massive inflammatory cell infiltration in the leiomyoma without malignant change (original magnification, ×400).

An ordinary leiomyoma of the uterus is the most common neoplasm, and pelvic infection is not rare in women of a reproductive age. However, the appearance of massive inflammatory cell infiltration in a leiomyoma is a rare clinical event. A pyomyoma is rarely reported and can be a lethal complication of a uterine leiomyoma. We reviewed the literature and found that only 22 cases have been reported since 1945. We list and briefly describe the reports chronologically (Table) [1–22]. Although there are several hypotheses, the definitive cause of a pyomyoma remains unclear. All cases can be explained by the hypothesis of infection following tumor necrosis/ischemia [3]. According to the literature, the offending organisms are diverse (including *Clostridium* spp., *Staphylococcus aureus, Streptococcus milleri, Str. hemolyticus, Str. agalactiae, Proteus* spp., *Serratia marcescens, Actinomyces meyeri, Enterococcus faecalis, Edwardsiella tarda, Klebsiella pneumonia,* and *Candida parapsilosis*), but infection routes can be ascending, by contiguous spread, or by seeding from distant sites with lymphatic or hematogenous spread [2]. Regardless of the size or location, the tumor can potentially become infected, and an early diagnosis of a pyomyoma is difficult due to the lack of specific findings from imaging studies. Upon analyzing the characteristics of all reported cases according to age and etiology, three categories were determined: postmenopausal; pregnancy-related; and pre-menopausal pyomyomas [3].

Necrosis of a leiomyoma occurs in postmenopausal women, especially in those with diabetes mellitus, hypertension, atherosclerosis, or vascular insufficiency (1 patient had third-stage uterine prolapse), and even though this event is not rare, only eight cases of pyomyoma have been reported. One patient received conservative antibiotics for 10 days and then underwent surgery but died 8 hours after surgery [4]. Another patient with diabetes mellitus and hypertension presented with septic shock and died 45 minutes after admission [5].

Ten case reports indicated that pregnancy might increase the risk of a pyomyoma, but a cesarean section might not increase the risk of infection. The conditions of uterine instrumentation, such as abortions [6–8], cesarean sections [9], and intravenous drug abuse [10] are additional factors or possible routes of infection. Caution must be taken if a patient with a uterine tumor experiences severe abdominal pain, signs of infection, and preterm labor. Immediate intervention and intensive care are crucial. A pyomyoma occurring in all four premenopausal women, not including our patient, can be explained as secondary infections of the tumor following a medical procedure such as cystoscopy [11] or intrauterine device insertion [12], or in the presence of severe systemic diseases [13] like choledocholithiasis and pancreatitis. In a case reported by Greenspoon et al [1], a 49-year-old woman with a uterine leiomyoma used a folk prescription of apricot juice injected by herself and refused surgical intervention. The infection route was similar to that seen with intravenous drug use. This patient eventually died due to the delay in initiating medication and surgery. Compared to those cases, our patient was relatively healthy. The only risk factor for impaired immunity was anemia, which is commonly seen in women with leiomyoma and hypermenorrhea. To our knowledge, this is the only case without clear etiology of a pyomyoma at a premenopausal age.

The mortality rate associated with pyomyomas is high (3 of the 22 cases died), the clinical course is rapid, and conservative treatments yield unsatisfactory results. The decision to proceed with a hysterectomy or myomectomy should take into consideration the desire for fertility and clinical severity [14]. Compared with the etiology mentioned in previous reports, our patient was relatively healthy and did not have a previous surgical history within the last 2 years. The operative findings and pathological studies neither showed a sinus tract between the endometrial cavity nor provided evidence of a source of infection. The nonspecific clinical symptoms and signs increase the difficulty of early detection for this dangerous complication. Young or premenopausal patients who undergo surgery without delay plus intensive care and broad-spectrum antibiotic treatment can result in a significantly improved outcome.

In conclusion, a pyomyoma is a life-threatening medical condition. Whether asymptomatic uterine tumors with degenerative changes require immediate surgery or not is controversial. Our case presented with a uterine tumor and sepsis, but no obvious source of infection [1], therefore surgical treatment was expeditiously introduced

Table. Brief summary of cases reported in the literature since 1945

Author	Year	Age (yr)	Underlying condition	Presentation	Treatment	Pathogen	Complication/Outcome
Miller [4]	1945	51	Congestive heart failure, uterine prolapse, menopause	Fever, sepsis	Sulfadiazine. Subtotal hysterectomy + BSO	*Streptococcus hemolyticus*	Ruptured pyomyoma caused peritonitis/died after the operation
Bedrosin et al [11]	1956	50	Nephrolithiasis, cystoscopy, premenopausal	Sepsis, acute abdominal pain	Tetracycline. TAH + BSO	Coliform group	Peritonitis/cured
Dubois & Neumman [15]	1957	29	3 wk postpartum	Fever, abdominal pain	Penicillin + streptomycin + oxytetracycline. Vaginal myomectomy	Not mentioned	Cured
Ruch [16]	1963	34	Leiomyoma, preterm labor	Persistent fever, abdominal pain	Penicillin + septomycin. TAH + BSO	Gram-positive cocci	Preterm delivery, peritonitis/cured
Kaufmann et al [5]	1974	58	Leiomyoma, hypertension, diabetes mellitus, menopause	Hemolysis, septic shock	Penicillin	*Clostridium perfringens*	Died (due to delayed treatment)
Weiss et al [2]	1976	59	Diabetes, menopause	Acute abdomen	Penicillin + streptomycin. TAH + BSO	*Clostridium* sp., Enterobacteriaceae, *Streptococcus* sp.	Abdominal wall fistulization/cured
Fuller & Lawrence [17]	1986	68	No systemic disease, huge uterine tumor, menopause	Lower extremity edema for 2 mo	TAH + BSO	*Streptococcus* sp.	Cured
Prichard et al [6]	1986	37	Leiomyoma, spontaneous abortion at 20 wk	9 wk post-abortion, fever, pelvic tenderness	Penicillin + streptomycin. TAH + BSO	*Streptococcus milleri*	Endocarditis/cured
Wong et al [7]	1986	29	Intrauterine device, spontaneous abortion	Post-abortion sepsis	Ampicillin + gentamycin. TAH + BSO	*Staphylococcus aureus, Serratia marcescens*	Right ovarian vein thrombosis, hematoperitoneum/cured
Greenspoon et al [1]	1990	49	Leiomyoma, premenopausal, apricot juice self-injection	Intermittent fever for 3 wk, chills, abdominal pain	Gentamycin + clindamycin + ampicillin	*Staphylococcus aureus, Actinomyces, Enterococcus* sp.	Septic shock, endocarditis/ died (patient was against surgery)
Yang & Wang [13]	1999	46	Choledocholithiasis	Pancreatitis	Amikacin switched to IV cefoxitin, TAH + BSO	*Edwardsiella tarda*	Cured

Tobias et al [8]	1996	32	Elective abortion	Post-abortion fever, abdominal pain	Cefoxitin, ampicillin + gentamycin + metronidazole. TAH + BSO	*Enterococcus faecalis*	Peritonitis/cured
Prahlow et al [10]	1996	31	Pregnancy, IV drug abuse	Abdominal pain	Ampicillin + clindamycin + aztreonam. TAH + BSO	*Staphylococcus aureus*	Peritonitis/cured
Gupta et al [18]	1999	75	Not mentioned	Fever, abdominal pain	Antibiotics. TAH + BSO	*Streptococcus* spp.	Cured
Genta et al [3]	2001	60	Leiomyoma, diabetes, menopause	Abdominal mass, anorexia, fever	Penicillin + amikacin. TAH + BSO	*Streptococcus agalactiae*	Endocarditis, deep vein thrombosis/cured
Grune et al [14]	2001	44	Leiomyoma, pregnancy	Fever	Antibiotics. C/S + myomectomy	*Klebsiella pneumoniae*	Sepsis/cured
Lin et al [9]	2002	33	Leiomyoma, pregnancy	Preterm labor, postpartum wound infection, abdominal pain	Antibiotics. TAH	Blood culture: *Escherichia coli*, pus culture: candida	Peritonitis, septic shock, wound infection/cured
Karcaaltincaba & Sudakoff [19]	2003	36	Leiomyoma, pregnancy	Fever, abdominal pain	Antibiotics. Myomectomy	*Peptostreptococcus tetradrus*	Pyomyoma rupture, peritonitis/cured
Manson et al [20]	2005	29	Leiomyoma. 3 wk after vaginal delivery	Fever, abdominal pain, dysuria	Ampicillin + gentamicin + metronidazole. Myomectomy	No growth	Cured
Yeat et al [21]	2005	53	Leiomyoma, menopause	Fever, abdominal pain	Cefmetazole to ceftizoxime + metronidazole. TAH + BSO	*Proteus mirabilis*	Pyomyoma rupture, peritonitis/cured
Sah et al [22]	2005	64	Huge uterine leiomyoma, menopause	Fever, abdominal pain	Antibiotics. TAH + BSO	*Staphylococcus aureus*	Cured
Manchana et al [12]	2007	42	Leiomyoma, IUD, premenopausal	Fever, abdominal pain	Antibiotics. TAH + BSO	No growth	Cured
Present case		43	Leiomyoma, premenopausal	Fever, hypermenorrhea	Cefazoline + gentamycin + clindamycin. TAH	*Escherichia coli*	Cured

TAH = total abdominal hysterectomy; BSO = bilateral salpingo-oophorectomy; C/S = cesarean section; IUD = intrauterine device.

before severe sepsis or death occurred. We herein report a relatively healthy premenopausal woman with an idiopathic pyomyoma to remind clinicians of this rare disease and the need for rapid intervention.

References

1. Greenspoon JS, Ault M, James BA, Kaplan L. Pyomyoma associated with polymicrobial bacteremia and fatal septic shock: case report and review of the literature. *Obstet Gynecol Surv* 1990;45:563–9.
2. Weiss G, Shenker L, Gorstein F. Suppurating myoma with spontaneous drainage through abdominal wall. *NY State J Med* 1976;76:572–3.
3. Genta PR, Dias MLN, Janiszewski TA, Carvalho JP, Arai MH, Meireles LP. *Streptococcus agalactiae* endocarditis and giant pyomyoma stimulating ovarian cancer. *South Med J* 2001; 94:508–11.
4. Miller I. Suppurating fibromyomas. Report of a case with a review of the literature. *Am J Obstet Gynecol* 1945;50:522–6.
5. Kaufmann BN, Cooper VM, Cookson P. *Clostridium perfringens* septicemia complicating degenerating uterine leiomyomas. *Am J Obstet Gynecol* 1974;118:877–8.
6. Prichard JG, Lowenstein MH, Silverman IJ, Brennan JC. *Streptococcus milleri* pyomyoma simulating infective endocarditis. *Obstet Gynecol* 1986;68(Suppl 3):46S–9S.
7. Wong TC, Bard DS, Pearce LW. Unusual case of IUD associated postabortal sepsis complicated by an infected necrotic leiomyoma, suppurative pelvic thrombophlebitis, ovarian vein thrombosis, hematoperitoneum and drug fever. *J Ark Med Soc* 1986;83:138–47.
8. Tobias DH, Koenigsberg M, Kogan M, Edelman M, LevGur M. Pyomyoma after uterine instrumentation: a case report. *J Reprod Med* 1996;41:375–8.
9. Lin YH, Hwang JL, Huang LW, Chen HJ. Pyomyoma after a cesarean section. *Acta Obstet Gynecol Scand* 2002;81:571–2.
10. Prahlow JA, Cappellari JO, Washburn AS. Uterine pyomyoma as a complication of pregnancy in an intravenous drug abuser. *South Med J* 1996;89:892–5.
11. Bedrosian L, Gabriels AG Jr, Hengerer AD. Ruptured suppurating myoma. *Am J Obstet Gynecol* 1956;71:1145–7.
12. Manchana T, Sirisabya N, Triratanachat S, Niruthisard S, Tannirandorn Y. Pyomyoma in a perimenopausal woman with intrauterine device. *Gynecol Obstet Invest* 2007;63: 170–2.
13. Yang CH, Wang CK. *Edwardsiella tarda* bacteraemia complicated by acute pancreatitis and pyomyoma. *J Infect* 1999; 38:124–31.
14. Grüne B, Zikulnig E, Gembruch U. Sepsis in second trimester of pregnancy due to an infected myoma: a case report and a review of the literature. *Fetal Diagn Ther* 2001; 16:245–7.
15. Dubois J, Neumann E. Necrobiosis of a voluminous fibroma 3 weeks after a normal labor. *Bull Fed Soc Gynecol Obstet Lang Fr* 1957;9:160–1.
16. Ruch WA. Intrapartum rupture of suppurative myoma with purulent peritonitis. Report of a case. *Obstet Gynecol* 1963; 21:593–6.
17. Fuller AF Jr, Lawrence WD. Case records of the Massachusetts General Hospital. Case 23-1985. *N Engl J Med* 1985;312: 1505–11.
18. Gupta B, Sehgal A, Kaur R, Malhotra S. Pyomyoma: a case report. *Aust NZ J Obstet Gynecol* 1999;39:520–1.
19. Karcaaltincaba M, Sudakoff GS. CT of a ruptured pyomyoma. *AJR Am J Roentgenol* 2003;181:1375–7.
20. Mason TC, Adair J, Lee YC. Postpartum pyomyoma. *J Natl Med Assoc* 2005;97:826–8. [Review]
21. Yeat SK, Chong KM, Pan HS, Cheng WC, Hwang JL, Lee CC. Impending sepsis due to a ruptured pyomyoma with purulent peritonitis: a case report and literature review. *Taiwan J Obstet Gynecol* 2005;44:75–9.
22. Sah SP, Rayamajhi AK, Bhadani PP. Pyomyoma in a postmenopausal woman: a case report. *Southeast Asian J Trop Med Public Health* 2005;36:979–81.