SPANDIDOS PUBLICATIONS

MOLECULAR AND CLINICAL ONCOLOGY 5: 527-531, 2016

Large uterine pyomyoma in a perimenopausal female: A case report and review of 50 reported cases in the literature

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Received February 15, 2016; Accepted April 7, 2016

DOI: 10.3892/mco.2016.1005

Abstract. Pyomyoma is a rare complication, which withoug antibiotics or surgical intervention, may cause sepsis and mortality. The present study reported a case of large uterine pyomyoma in a perimenopausal female. A 53-year-old multigravida woman was referred to the Department of Obstetrics and Gynecology (Wakayama Medical University, Wakayama, Japan) due to progressive abdominal distension. The patient presented with anemia gravis, severe inflammatory reaction and cachexia. Computed tomography revealed a large unilocular mass, 50 cm in size, with an irregular surface and thickened wall, occupying the entire abdomen. Following antibiotic medication, the patient underwent a total abdominal hysterectomy and bilateral salpingo-oophorectomy. Intraoperative findings demonstrated a solid tumor arising from the back of the uterine body. A total of 12 liters of purulent, malodorous fluid was drained from the tumor. The resected mass was 50 cm in size and 13.5 kg in weight. Cultures of the pus revealed the presence of *Streptococcus agalactiae*. Pathological findings revealed suppurative leiomyoma with no malignancy. Large pyomyoma is difficult to distinguish from a gynecological malignant tumor types, particularly in perimenopausal women with non-specific clinical presentation. Although pyomyoma is a benign tumor, care must be taken to discriminate these from large abdominal tumors.

Introduction

Pyomyoma (suppurative leiomyoma) is a rare, but life-threatening, condition resulting from infarction and infection of uterine leiomyoma (1,2). Incidence of pyomyoma has decreased due to the development of antibiotics. Since 1945, only 50 pyomyoma cases have been documented in the literature, with a mortality rate of 6% (3/50) (3-5). The most likely cause of mortality was delayed and difficult diagnosis. Although the triad of pyomyoma are sepsis, leiomyoma, and no other source of infection (5), it may present with silent or non-specific symptoms, which results in delayed diagnosis and treatment. Visualization of intratumoral gas formation may be suggestive of pyomyoma, but has not been consistently reported in all cases. Furthermore, large abdominal complex masses are likely to be first suspected as pelvic malignancies if found incidentally in perimenopausal women.

The present study experienced a rare case of large uterine pyomyoma in a perimenopausal woman who presented with anemia gravis, severe inflammatory reaction and cachexia. A total of 50 reported cases of pyomyoma in the literature since 1945 were also studied.

Case report

A 53-year-old multigravida woman with 7 months of amenorrhea was referred to the Department of Obstetrics and Gynecology (Wakayama Medical University, Wakayama, Japan) due to gradual abdominal distension starting 2 years previously. The patient exhibited muscle weakness and walking difficulty, but no fever, abdominal pain or metrorrhagia. Her abdomen was swollen to the size of a beach ball, with a 126 cm abdominal circumference and body weight of 84.1 kg. An intrauterine device (IUD) had been inserted following her third birth, and no history of any lower abdominal or pelvic discomfort, leiomyoma, pelvic surgery, or other predisposing factors were known. The present study was unable to determine the uterine cervix with pelvic examination due to its deviation, and was unable to acquire cytopathological findings of the cervix and endometrium. Transvaginal and transabdominal ultrasound examinations revealed a large abdominal mass with heterogeneous echogenicity. Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a large unilocular mass with an irregular surface and thickened wall, occupying the abdominal cavity without gas formation (Fig. 1). Contrast CT revealed an expanded branch of the left internal iliac artery, which was suspected to be the uterine artery, surrounding the mass (Fig. 1B). Hemoglobin levels were 5.7 g/dl, white blood cell count was 57,300/μl and C-reactive protein was elevated to 20.24 mg/dl. Cancer antigen (CA)125 was also elevated to 200 U/ml. Blood and

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Key words: pyomyoma, gynecological tumor, severe inflammatory reaction, cachexia, perimenopause

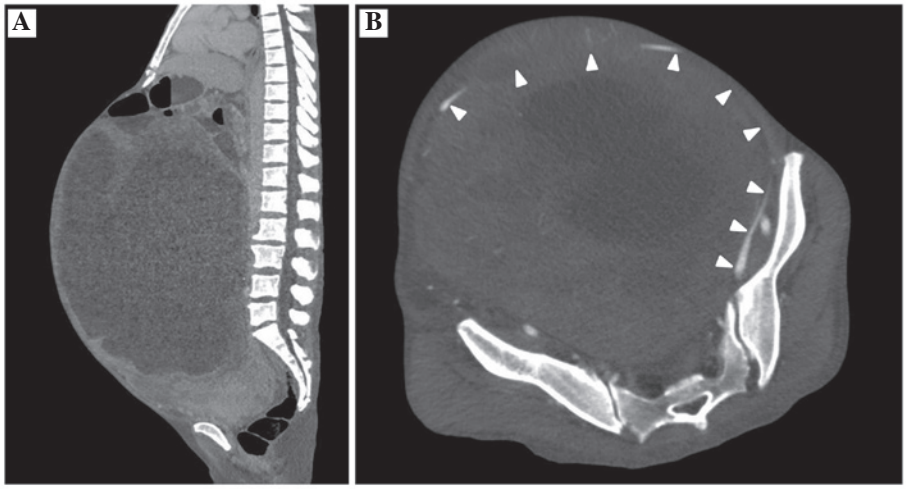


Figure 1. CT findings of pyomyoma. (A) Non-contrast CT and (B) contrast CT revealed the left uterine artery (arrow head) running over a large mass. CT, computed tomography.

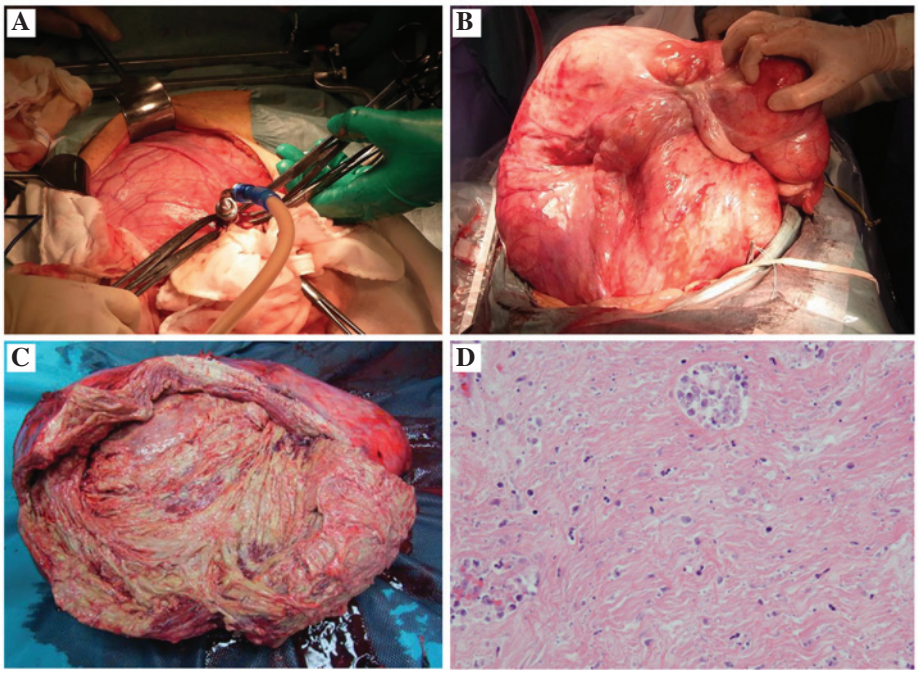


Figure 2. Macroscopic and microscopic findings of pyomyoma. (A) Open drainage from the mass. (B and C) Macroscopic finding of the mass. (D) Microscopic findings following staining with hematoxylin and eosin (magnification, x200).

vaginal cultures were negative. Possible diagnoses of the large abdominal mass included gynecologic tumor types (benign or malignant ovarian tumor, uterine sarcoma or pyometra) and gastrointestinal stromal tumor or mucocele of the appendix. Gastroscopy revealed no specific findings and colonoscopy revealed difficulty of insertion above the sigmoid colon due to pressure from the mass. Due to the finding of the uterine artery by contrast CT, the origin of the mass was suspected to be the uterus. Following antibiotic medication and blood transfusion, the patient underwent a total abdominal hysterectomy and bilateral salpingo-oophorectomy. Ureteral stents were indwelled in each side pre-operatively. Intraoperative findings demonstrated a solid tumor arising from the back of the uterine body with normal bilateral adnexa. Prior to removal, 12.4 liters of purulent, malodorous fluid was drained from the tumor (Fig. 2A and B). The resected mass was 50x37x20 cm in size and 13.5 kg in weight. The surrounding myometrium appeared normal and measured 3 cm in thickness, and the cut surface of the mass was purple-yellowish (Fig. 2C). Cultures of the pus in the tumor revealed the presence of *Streptococcus agalactiae*. An IUD was identified near the cervix, however culture was negative. The pathological diagnosis was leiomyoma with marked necrosis and chronic inflammation, with no evidence of malignancy (Fig. 2D). Following resection of the tumor, the patient's weight decreased to 55 kg. The post-operative course was uneventful and the patient was discharged from the hospital on post-operative day 14. At 4 months follow-up, the patient's weight had increased to 62 kg due to a good appetite.

Table I. Previously reported cases of pyomyoma without pregnancy, since 1945.

Author, year	Age	Key points	Laboratory data	Size	Treatment[a]	Refs.
Miller *et al*, 1945	51	STM	WBC 38,700/μl	35x25 cm	Subtotal hysterectomy + BSO	(3)
Kaufmann *et al*, 1974	58	STM, HT, DM	WBC 28,800/μl, Hb 7.3 g/dl	ns	No treatment	(4)
Greenspoon *et al*, 1990	49	STM	WBC 21,200/μl, Hb 7.4 g/dl	11.5x9x11 cm, 2.5 kg	No treatment	(5)
Chen *et al*, 2014	67	Gas production	WBC 12,300/μl, CA125 29.98 U/ml	25x20x15 cm	TAH + BSO	(6)
Manchana *et al*, 2007	42	IUD	WBC 29,380/μl, Hb 8.7 g/dl, CA125 65.2 U/ml	15x15 cm	TAH + BSO	(8)
Kitamura *et al*, 2005	ns	UAE, gas production	ns	ns	TAH	(9)
Abulafia *et al*, 2010	48	UAE, gas production	WBC 22,600/μl, Hb 8.1 g/dl	11x10x6 cm	TAH	(10)
Shukla *et al*, 2012	65	UAE, gas production	WBC 7,900/μl	12x10 cm	TAH + BSO	(11)
Pinto *et al*, 2012	36	UAE	WBC normal, Hb 9.5 g/dl	6.8x5.6x5.5 cm	Laparoscopic drainage	(12)
Rosen *et al*, 2013	47	UAE, gas production	WBC 15,900/μl	ns	Supracervical hysterectomy + RSO	(13)
Weiss *et al*, 1976	59	DM	ns	15 cm	TAH + BSO	(14)
Genta *et al*, 2001	60	DM, DVT	WBC 14,100/μl, Hb 7.7 g/dl, CA125 109.7 U/ml	25x20 cm	TAH + BSO + omentectomy	(15)
Fletcher *et al*, 2009	44	DM	WBC 22,500/μl, Hb 7.8 g/dl, CA125 17.5 U/ml	15.5x16x9 cm	TAH + BSO	(16)
Ono *et al*, 2014	69	DM	WBC 10,710/μl, CRP 2.71 mg/dl, Hb 7.6 g/dl	ns	TAH	(17)
Goyal *et al*, 2015	42	DM	WBC 10,200/μl, Hb 9.5 g/dl	6 cm	Subtotal hysterectomy +	(18)
Lee *et al*, 2010	46	FDG-PET	WBC 10,100/μl, Hb 8.8 g/dl, CA125 59.2 U/ml	38x30x10 cm, 3 kg	LSO +TAH	(22)
Bedrosin *et al*, 1956	50	N/A	WBC 12,800/μl, Hb 11.0 g/dl	7 cm	TAH + BSO	(23)
Fuller *et al*, 1985	68	N/A	WBC 24,000/μl	10 cm	TAH + BSO	(24)
Yang and Wang, 1999	46	N/A	WBC 45,400/μl, Hb 7.0 g/dl	13x12 cm	TAH + BSO	(25)
Gupta *et al*, 1999	60	N/A	WBC 14,000/μl	30x25 cm, 4.3 kg	TAH + BSO	(26)
Sah *et al*, 2005	64	N/A	WBC 15,000/μl, Hb 8.0 g/dl	22x23x10 cm, 3.5 kg	TAH + BSO	(27)
Yeat *et al*, 2005	53	N/A	WBC 52,600/μl, CRP 42.4 mg/dl, Hb 8.6 g/dl	12x12x10 cm, 1,020 g	TAH + BSO	(28)

Table I. Continued.

Author, year	Age	Key points	Laboratory data	Size	Treatment[a]	Refs.
Patwardhan and Bulmer, 2007	38	N/A	WBC 18,500/μl, CRP 22.5 mg/dl	ns	Myomectomy	(29)
Chen *et al*, 2010	46	N/A	WBC 13,000/μl, Hb 7.9 g/dl	14.3x12x8 cm	TAH	(30)
Kuriyama *et al*, 2010	51	N/A	WBC 15,900/μl, CRP 13.1 mg/dl	ns	TAH	(31)
Zangeneh *et al*, 2010	47	N/A	WBC normal, Hb 10.3 g/dl	3x5 cm	TAH + BSO	(32)
Liu and Chen, 2011	42	N/A	WBC 42,880/μl	9.0x8.0x6.5 cm	Open drainage	(33)
Present report	53	IUD	WBC 57,300/μl, CRP 20.24 mg/dl, Hb 5.7g/dl, CA125 200.2 U/ml	50x37x20 cm, 13.5 kg	TAH + BSO	-

[a]Treatment not including any antibiotics. ns, not specified; TAH, total abdominal hysterectomy; BSO, bilateral salpingo-oophorectomy; RSO, right salpingo-oophorectomy; CS, cesarean section; IUD, intrauterine device; DM, diabetes mellitus; UAE, uterine artery embolization; STM, succumbed to mortality; WBC, white blood cell count; Hb, hemoglobin; CA, cancer antigen; CRP, C-reactive protein; N/A, not applicable; HT, hypertension; DVT, deep vein thrombosis; FEG-PED, fluorodeoxyglucose-positron emission tomography; LSO, left salpingo-oophorectomy.

Discussion

Pyomyoma occurs in both post-and pre-menopausal women, however, the risk of suppurative myoma is increased by pregnancy (1,2). For post-menopausal patients, systemic vascular changes have been suggested to be the likely underlying cause of pyomyoma (6). Necrosis of the leiomyoma caused by vascular flow insufficiency in the uterus following menopause is also a possible cause. A history of uterine leiomyoma, pregnancy, abortion, menopause, uterine artery embolization (UAE), IUD, vascular insufficiency (diabetes, hypertension and atherosclerosis) and systemic disease or infection may be predisposing factors for pyomyoma; however, definitive diagnoses remain difficult. Although rapid clinical diagnosis for pyomyoma is often difficult due to its low incidence and the requirement to rule out the possibility of malignancy, mortality has decreased due to the improvement in surgical treatments, including myomectomy and hysterectomy, and broad-spectrum antibiotics.

A MEDLINE search since 1945 revealed only 50 reported cases of pyomyoma: 27 were non-pregnant woman (mean age, 51.8 years; range, 36-69-years-old; Table I) (3-29) and 23 cases were associated with pregnancy or abortion (mean age, 33.6 years; range, 28-44-years-old). The mean pyomyoma size in non-pregnancy and pregnancy-associated cases were 16.2 cm (3-38 cm) and 13.5 cm (5-58 cm), respectively. Non-pregnancy pyomyoma tended to be larger compared with pregnancy-associated pyomyoma, and the present case was the largest among all reported cases of non-pregnancy-associated pyomyoma. Severe anemia gravis and inflammatory reaction were described in numerous pyomyoma cases, including the present study. The presentation and complications of pyomyoma vary. It has been shown that 2/50 cases had a history of IUD usage (7,30), 5/50 cases had UAE (8-12), 6/50 cases had vascular insufficiency (4,13-17) and 8/50 cases demonstrated gas production (6,8-10,12,31-33). Notably, gas production was observed in 4/5 UAE cases (8-10,12). Although pyomyoma arises spontaneously, post-partum, post-instrumentation or post-surgery have been reported in the literature, only two cases include a IUD, and only one case has been previously reported in a non-pregnant woman (7). Knowledge of IUD history may be helpful in the diagnosis of pyomyoma.

Pyomyoma was often associated with polymicrobial infection. Among the 50 reported cases, infection by *Staphylococcus* species was reported in 8 cases, *Streptococcus* species in 7 cases, *Escherichia coli* in 6 cases, *Enterococcus faecalis* in 5 cases, *Clostridium* species in 3 cases, *Proteus* species in 2 cases and *Candida* species in 2 cases. In the present case, *Streptococcus agalactiae* was cultured from the pus and tumor, however, not from the blood and vagina, suggesting the infection existed focally within the tumor. Therefore, conservative management with pre-operative broad-spectrum antibiotics and appropriate surgery was performed successfully. For surgical treatment, hysterectomy was performed in 32 cases, myomectomy in 10 cases and drainage in 6 cases.

Diagnosis of large pyomyoma is difficult since surgery is required for a definitive diagnosis. A malignant tumor, in particular ovarian cancer, was initially suspected due to the findings of a large abdominal mass with signs of necrosis in a cachexic perimenopausal woman with an elevated CA125 level. In a previous report, imaging analysis was shown to identify only non-specific results (6). MRI and positron emission tomography did not improve the specificity of pyomyoma diagnosis. Although intratumoral gas formation is markedly suggestive of pyomyoma on ultrasound and CT imaging, gas formation is not consistently observed, as with the present case. In the present case, contrast CT contributed

to the diagnosis of pyomyoma due to the identification of the uterine artery location.

CA125 has been observed to be increased in other gynecologic and non-gynecologic malignancies, as well as in a variety of benign disorders, including leiomyoma. Among the previously reported cases of pyomyoma, 5 cases reported CA125 levels (6,7,14,15,18), with a mean level of 56.3 U/ml (29.98-109.7 U/ml). CA125 was measured only in non-pregnancy-associated cases, possibly due to suspicion of gynecological malignancy based on the age of the patient and size of tumor. Furthermore, in pregnancy-associated cases, diagnosis of myoma was likely during pre-natal examination prior to the onset of the symptom, which may have contributed to the identification of the tumor origin. In the present case, CA125 level was the highest compared with the previously reported cases.

To the best of our knowledge, this is the third IUD-associated case of pyomyoma, with the largest tumor size and highest CA125 level compared with previously reported non-pregnancy cases. A large pyomyoma is difficult to distinguish from a gynecological malignant tumor, particularly in perimenopausal, cachectic women with non-specific clinical presentation and without a history of leiomyoma. A history of IUD usage and contrast CT may be helpful in the diagnosis of pyomyoma. Gynecologists and general surgeons must be aware of the possibility of pyomyoma when presented with a large abdominal tumor, and in certain cases, surgery in combination with broad spectrum antibiotics may result in a good outcome.

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