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Pyomyoma mimicking tubo-ovarian abscess: Two case reports

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ABSTRACT

Pyomyoma is an extremely rare complication, defined as an infection of a uterine leiomyoma. We describe two cases of pyomyoma that were initially considered to be tubo-ovarian abscesses but were later diagnosed as pyomyomas and managed with laparoscopic surgery. Case 1 was a 26-year-old nulliparous woman who was previously diagnosed with bilateral endometriomas and presented to the hospital with lower abdominal pain. Magnetic resonance imaging revealed bilateral endometrial cysts and a 4-cm mass consistent with a tubo-ovarian abscess. The patient experienced continuous pain, and the cyst in the left adnexa enlarged; thus, laparoscopic surgery was performed. The cystic tumor in her uterus contained purulent fluid. Therefore, an abscess in the degenerative subserous myoma was diagnosed. Case 2 was a 47-year-old nulliparous woman who had undergone total mastectomy and postoperative radiotherapy for breast cancer. She was undergoing hormone therapy when she presented to the hospital with lower abdominal pain, fever, and increased inflammatory markers. Computed tomography revealed a 7-cm tumor with rim enhancement in her left adnexa; therefore, a tubo-ovarian abscess was suspected. After admission, drainage was performed under transvaginal ultrasound guidance, and antibiotics were administered. However, these treatments did not relieve her abdominal pain. Emergency laparoscopic surgery was performed, and intraoperative findings demonstrated an abscess in the degenerative subserous myoma of the uterus with normal adnexa. Laparoscopic hysterectomy and bilateral salpingectomy were performed. Laparoscopic surgery was effective for both patients. Delayed diagnosis of pyomyoma can result in serious complications. Timely surgery with concomitant antibiotic treatment may facilitate good outcomes.

1. Introduction

Pyomyoma is an extremely rare complication resulting from an infected leiomyoma or a cystic lesion that develops in the myometrium [1]. Although the criteria for the diagnosis of pyomyoma include the presence of a leiomyoma, sepsis, and the absence of any other infected tissues [2], the symptoms of pyomyoma are not always specific. It is difficult to differentiate between pyomyoma and tubo-ovarian abscess, especially when the patient is premenopausal and has endometriomas or hematosalpinx., and diagnostic delays may lead to serious complications. Surgical interventions, such as hysterectomy or myomectomy, are important treatment options. We describe two cases of pyomyoma successfully treated with laparoscopic surgery.

2. Case Presentations

2.1. Case 1

A 26-year-old nulliparous woman presented with abdominal pain after sexual intercourse and was referred to the hospital for surgery. She had previously been prescribed oral contraceptive pills for endometriosis. Laboratory tests revealed a white blood cell count of 6900/μL (reference range, 3300–8600/μL), C-reactive protein level of 3.0 mg/dL (reference range, 0–0.3 mg/dL), and cancer antigen-125 level of 44 U/mL (reference range, 0–35 U/mL). Transvaginal ultrasonography showed a 4-cm mass, and magnetic resonance imaging (MRI) revealed a 4-cm mass in the area of the left adnexa. Endometrioma was confirmed in both ovaries; thus, the left mass was suspected to be a tubo-ovarian abscess, and conservative treatment was initiated with antibiotics. The patient was initially treated with a 5-day course of cefdinir 300 mg per day, but her condition did not improve. The culture from her vaginal

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secretions was positive for *Gardnerella*; thus, the antibiotic therapy was changed to metronidazole 750 mg per day for 2 weeks. The patient's C-reactive protein level decreased, but her lower abdominal pain did not resolve. Repeat transvaginal ultrasonography revealed an enlargement of the mass despite antibiotic treatment. Repeat MRI showed a 10-cm mass that appeared hypointense on T1-weighted images and hyperintense on T2-weighted images (Fig. 1). Considering the enlargement of the mass and no clinical improvement, we planned surgical intervention.

The patient underwent laparoscopic surgery to confirm the diagnosis and determine the appropriate treatment. Intraoperative findings were those of a large mass encompassed by the omentum, which was adherent to the abdominal wall. Dark-brown fluid (pus) leaked through the ruptured wall of the mass during adhesiolysis. After dissection of the adhesions, we observed that the mass was connected to the uterus and it was subsequently resected. Both fallopian tubes were normal. As the right ovary contained a cyst, cystectomy was also performed. Finally, the abdominal cavity was irrigated. The operation lasted 150 min, and the measured blood loss was 50 mL.

Postoperatively, the patient recovered rapidly, with immediate relief of the abdominal pain and further improvement in her C-reactive protein level. She was discharged on the 5th postoperative day. The final pathology report indicated a leiomyoma with cystic degeneration and partially infiltrated inflammatory cells (Fig. 2), and an endometrial cyst on the right ovary. At a 2-year follow-up using ultrasound, there was no recurrence of the myoma.

2.2. Case 2

A 47-year-old nulliparous woman presented with lower abdominal pain and fever. She had a history of abdominal myomectomy and hysteroscopic myomectomy at the ages of 34 and 41 years, respectively, and infection of a left endometrioma, which was treated with antibiotics 3 years before the current admission. The patient had also been diagnosed with breast cancer a year previously and had undergone total mastectomy and postoperative radiotherapy. Upon visiting the hospital after hormone therapy, she underwent an endometrium smear test. She developed lower abdominal pain and high fever after the smear test, and transvaginal ultrasonography revealed a non-uniform internal mass in the area of the left ventral uterus. Contrast-enhanced computed tomography (CT) showed a 7-cm mass with rim enhancement in the left uterine adnexa of the anterior uterine body; a tubo-ovarian abscess was suspected, and the patient was admitted to the hospital for treatment. Aside from a white blood cell count of 11,600/μL and a C-reactive protein level of 0.4 mg/dL, all other laboratory tests, including tumor markers, were normal. Contrast-enhanced MRI revealed a 5-cm mass in the left ventral uterus that appeared hypointense on T1-weighted images and hyperintense on T2-weighted images (Fig. 3). The ovaries could not be visualized; however, based on the patient's medical history, infection

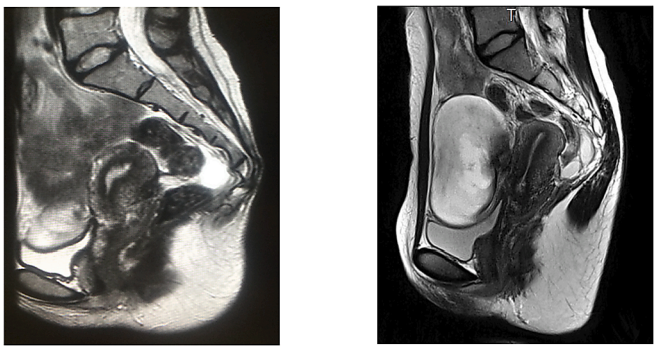

Fig. 1. T2-weighted magnetic resonance images of the mass in case 1.
Left: 4 cm, right: 1 month later, 10 cm.

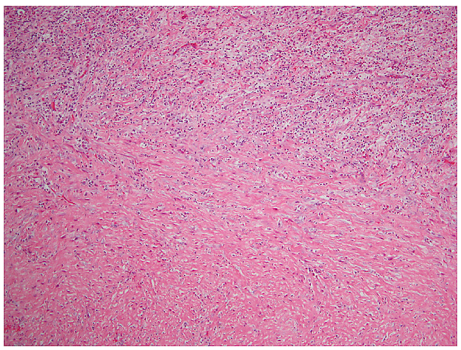

Fig. 2. Pathological findings of case 1: Leiomyoma with cystic degeneration and partially infiltrated inflammatory cells within the wall (HE, ×200).

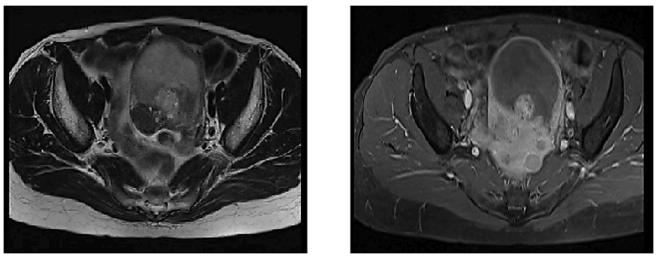

Fig. 3. Preoperative contrast-enhanced magnetic resonance imaging scans in case 2.
Left: T1-weighted image showing a 5-cm mass with hypointense ring enhancement, right: T2-weighted image showing hyperintensity.

of the left endometrioma was suspected. Treatment included intravenous ampicillin/sulbactam 12 g per day and transvaginal aspiration and drainage with Douglas pouch puncture. The culture from the abscess was positive for *Escherichia coli*, and the antibiotic therapy was switched to intravenous treatment with cefotaxime 4 g and clindamycin 1.2 g per day. Although the mass temporarily decreased in size because of antibiotic therapy and transvaginal aspiration, there was no significant improvement in symptoms, and high levels of inflammatory markers persisted. Therefore, the patient underwent laparoscopic surgery to confirm the diagnosis and determine the appropriate treatment. Intraoperative findings showed a mass continuous with the uterus, with multiple uterine leiomyomas. Adhesion of the vesicouterine pouch was removed. Since the patient did not desire future pregnancy, a hysterectomy was performed considering her hormone treatment for breast cancer. The operation lasted 100 min, and the measured blood loss was 30 mL.

The patient's postoperative recovery was uneventful, and her inflammatory parameters improved. She was discharged on the 3rd postoperative day. The final pathology report indicated that the largest

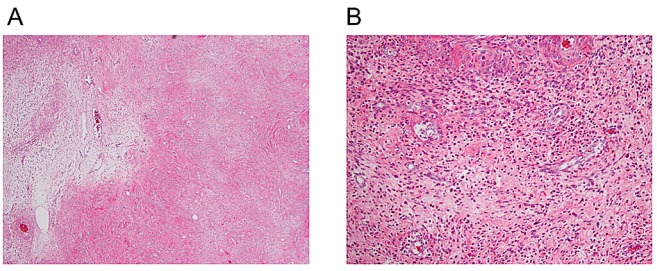

Fig. 4. Pathological findings of case 2.
A: Leiomyoma with active inflammation and infiltrating multinucleated giant cells (HE, ×20).
B: The same image as in Fig. 4a at increased magnification (HE, ×200).

mass was a leiomyoma that had active inflammation and infiltrating multinucleated giant cells in the endometrial tissue of the uterine body (Fig. 4 A, B). Follow-up care has included an annual ultrasound examination, which has shown no abnormality thus far, three years after surgery.

3. Discussion

Pyomyoma is extremely rare. Although the condition was first described in 1871, only 75 cases were reported between 1871 and 1945 [3]. Since 1945, only 50 cases have been reported [4]. As most reported cases are related to pregnancy, the postmenopausal period, and gynecologic procedures such as uterine artery embolization, it is thought that the pathophysiology of pyomyoma is associated with the change in blood flow to a leiomyoma [5]. Consequently, ischemia of the leiomyoma occurs, and pyomyoma is thought to be caused by bacterial dissemination. Many postmenopausal cases were caused by ischemia resulting from hypertension, diabetes, or atherosclerosis [3,6]. Immunocompromise is also associated with the development of pyomyoma. When the route of infection includes direct spread or lymphatic spread from the abdominal cavity to a leiomyoma, the risk of developing a pyomyoma is increased significantly [7]. The second case was that of a patient who was immunocompromised because of breast cancer treatment, such that a pyomyoma developed because of suppurative infection; this was confirmed by a smear test of the endometrium. Similar to previous cases [3,8], pyomyoma in the second case was caused by a change in blood flow to the myoma. In contrast, the first case was extremely rare. Considering that this case may have been caused by suppurative spread after sexual intercourse, this is a mechanism by which pyomyoma may occur in young women.

Pyomyoma has nonspecific presentations, thus making its diagnosis difficult. Clinical symptoms such as fever and abdominal pain are similar to those of pelvic inflammatory disease or a tubo-ovarian abscess; therefore, it is difficult to differentiate pyomyoma from these diseases. CT shows an increase in fluid level in the mass and hypertrophy of the wall of the mass, which is demonstrated as rim enhancement. However, a tubo-ovarian abscess and degenerated leiomyoma may also show an increase in the fluid level and changes to the wall of the mass [7,9–11]. A previous report demonstrated that a contrast-enhanced CT scan showing gas and debris in the mass supports the diagnosis of pyomyoma. However, the cystic component of pyomyoma (such as the fibrous capsule) appears hypointense to isointense on T1-weighted MRI, and hypointense to hyperintense on T2-weighted MRI [12]. The peripheral rim appears hyperintense on T1-weighted MRI and hypointense on T2-weighted MRI, and the connection between the uterus and mass helps make the diagnosis. However, the peripheral rim corresponds with red degeneration of leiomyoma, and this finding does not indicate pyomyoma. Unless pyomyoma is suspected in advance, specific findings might be missed.

Pyomyoma is associated with fatal complications, such as sepsis, peritonitis, and respiratory insufficiency [13,14]. The mortality rate is around 6% (3/50) [15,16]. Of the fatalities reported, two patients were treated with antibiotics only, and surgical interventions were delayed. The standard treatment for pyomyoma is broad-spectrum antibiotics and surgery. As the diagnosis of pyomyoma is difficult, pyomyoma is confirmed mostly during surgical intervention. In contrast, most patients with a tubo-ovarian abscess are managed conservatively with antibiotics and drainage; surgical intervention is not always required [17]. Laubach et al. used CT-guided drainage and lavage of pyomyoma in pregnant women [18]. In our case, transvaginal drainage was performed; however, the drainage alone did not improve the symptoms significantly; hence, laparoscopic surgery was eventually performed. Although image-guided drainage is safe, aspiration of the fluid is difficult because of its viscosity. Therefore, when drainage for pyomyoma is performed, surgery must be considered. Furthermore, one report mentioned that pyomyoma develops slowly, over weeks [7]. Compared with other infections, such as a tubo-ovarian abscess, our cases of pyomyoma developed slowly. Considering the risk of a life-threatening event when inflammation becomes chronic, it would be best to decide on the diagnostic and therapeutic surgical intervention early.

In our two cases, a tubo-ovarian abscess was suspected, and conservative treatments were initiated. As there was no significant improvement, surgical treatment was performed, and pyomyoma improved shortly thereafter. Advancements in minimally invasive treatments have enabled patients to recover quickly following surgery.

4. Conclusion

Pyomyoma should be considered in cases of large lower abdominal masses with continuous fever and abdominal pain, even in the absence of recent intrauterine intervention. Radiography can help rule out common differential diagnoses. Furthermore, when there is a high probability of pyomyoma, it is important to consider the need for surgical management in addition to antibiotic therapy.

Patients' perspectives

The patients were satisfied with the treatment outcomes as they recovered quickly following laparoscopic surgery.

Contributors

Kyoko Oshina was involved in patient care and surgery in the two reported cases, and contributed to writing and editing the manuscript.

Rie Ozaki was involved in patient care and surgery in the two reported cases, and contributed to writing and editing the manuscript.

Jun Kumakiri was involved in patient care and surgery in the two reported cases, and contributed to writing and editing the manuscript.

Keisuke Murakami was involved in patient care and surgery in the two reported cases, and contributed to writing and editing the manuscript.

Yu Kawasaki contributed to writing and editing the manuscript.

Mari Kitade was involved in patient care and surgery in the two reported cases, and contributed to writing and editing the manuscript.

Atsuo Itakura contributed to writing and editing the manuscript.

All authors read and approved the final manuscript.

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Patient consent

Written informed consent was obtained from the patients for publication of this case report and the use of accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

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Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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