Ruptured pyomyoma – Rare complication of post-uterine artery embolization: A case report

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ABSTRACT

Leiomyomas, also termed as fibroids, are benign smooth, muscle neoplasms seen in 70–80% of women by the age of 50 years. Uterine artery embolization (UAE) is a minimally invasive procedure that involves cessation of vascular supply to the fibroids, by infusion of gelatinous microspheres into the uterine arteries. Pyomyoma is a supplicative leiomyoma, secondary to infection of necrotic tissue. It is an infrequent complication of uterine artery embolization (UAE). Pyomyoma can lead to sepsis, peritonitis, and respiratory distress syndrome resulting in high morbidity and mortality. Due to its rarity, high suspicion is crucial in the diagnosis, and prompt treatment is recommended to reduce mortality. Ultrasound, computed tomography, and magnetic resonance imaging assist in diagnosis. We present a case of a 44-year-old woman with ruptured pyomyoma, following an UAE intervention. The patient was treated with total abdominal hysterectomy and salpingo-oophorectomy along with peritoneal irrigation and drainage.

Keywords: Pyomyoma, Uterine artery embolization, Uterine artery embolization complications, Ruptured pyomyoma, Peritonitis

INTRODUCTION

Pyomyoma is an infected leiomyoma that can develop spontaneously, or be observed in postpartum, post-surgical, and postmenopausal phases.[1] It forms secondary to infarction, necrosis, and infection of uterine leiomyoma.[1] Pyomyoma, secondary to uterine artery embolization (UAE) is a rare complication with a higher mortality rate ranging from 20 to 30%.[2] UAE induces ischemia and infarction of leiomyoma by occluding uterine arteries.[3] The infarcted tissue serves as a nidus for microbes, leading to the formation of pyomyoma. Pyomyoma in post-UAE patients is rare, and to the best of our knowledge, only 11 cases were reported in the literature.[4] The diagnosis is arduous due to non-specific clinical and radiological findings. Strong clinical suspicion is imperative in correlating the findings for prompt diagnosis. In this manuscript, we have described a case of ruptured pyomyoma which ensued after the UAE procedure.

CASE REPORT

A 44-year-old woman was presented to the emergency room, with complaints of vaginal bleeding and fatigue. She had a prior history of fibroids and underwent UAE, 30 days before the current presentation.
During the current visit, her hemoglobin was noted to be 5.1 g/dl, and she received packed red blood cell transfusions. She was noted to have a persistent lower abdominal pain and fever. Pertinent laboratory investigations, including COVID swab, respiratory panel, blood culture, and urine culture, were negative. Diagnostic imaging with computed tomography (CT) scan was performed, which revealed an enlarged uterus with necrotic fibroids containing a large pocket of gas/fluid within the endometrial cavity, uterine perforation along with the fundal fibroid, dilated right fallopian tube, and inflamed right ovary. CT findings in conjunction with a clinical picture raised a suspicion for a ruptured pyomyoma with ovarian adhesions, ensued post-UAE. The patient was initiated on intravenous fluids and antibiotics. She was taken to the operating room, and a total abdominal hysterectomy with bilateral salpingectomy and right oophorectomy was performed. The intra-operative findings include 6 cm fundal fibroid with two necrotic foci of 1 cm each, draining pus into the peritoneal cavity, 5 cm anterior exophytic fibroid, 3 cm posterior exophytic fibroid, and right ovarian adhesions to the uterus. The gross examination of the resected specimen revealed multiple fibroids, one of which had evidence of necrosis, as shown in [Figure 1a and b]. [Figures 2 and 3] describe the histopathological findings of acute inflammation. Post-operatively, the patient was continued on antibiotics and was discharged from the hospital after a favorable recovery.

**DISCUSSION**

Uterine leiomyomas are the most prevalent benign neoplasms, comprising of smooth muscle and fibrous connective tissue. Its prevalence ranges from 70 to 80% in women of aged 50 years.[3] Abnormal uterine bleeding and pelvic pressure are the most common symptoms of leiomyoma.[3] Conservative therapy of leiomyomas includes oral contraceptive pills and gonadotropin-releasing hormone agonists. Surgically, fibroids can be managed through myomectomy, magnetic resonance (MR)-guided high-intensity focused ultrasound, UAE, and hysterectomy.[3] UAE is a minimally invasive procedure performed as an alternative to myomectomy and hysterectomy. It
involves the injection of gelatinous microspheres into the uterine artery, thereby halting the blood supply to leiomyoma.\cite{3} The employment of embolization, helped avoid avoided hysterectomy in 96% and improved symptoms in 74% of the patients, in a study by Yuan et al.\cite{5} Besides, polyvinyl alcohol particles (PVA), tris-acryl gelatin microspheres, and gelatin sponge particles are being used as embolic agents in the UAE. Various complications associated with these agents are listed in [Table 1]. The resulting ischemia, due to UAE, leads to infarction and necrosis of target leiomyoma. Post-interventional complications include fibroid expulsion, infection (endometritis, tubo-ovarian abscess, and pyomyoma), pulmonary embolism, and fibroid regrowth.

Pyomyoma is an infrequent complication characterized by the infected necrotic tissue of embolized leiomyoma.\cite{4} Often, it is due to polymicrobial infection, including anaerobic, Gram-positive, and Gram-negative bacteria, submucosal leiomyomas, elderly old age, pre-existing infection, intravenous drug users, presence of intra-uterine devices, diabetes, hypertension, post-partum, and immunocompromised states are the known risk factors predisposing patients to pyomyoma.\cite{6} Employing UAE in patients with leiomyoma of size <8 cm reduces the risk of pyomyoma. [Figure 4a-d] shows embolization of the right and left uterine arterial branches with PVA; post-embolization, there is a significant reduction in the vascularity of the fibroids.

After embolization, the route of infection is often a direct spread from the endometrial cavity, adnexa, bowel, or blood.\cite{1} Pyomyoma presents with a classic triad of pelvic pain, sepsis, and no an unknown source of infection.\cite{8} However, diagnosis based on clinical findings is challenging, as its presentation mimics post-embolization syndrome (PES), which may also present with pelvic pain, elevated white cell count, and fever within 1 week of UAE.\cite{4,7} PES can be seen in 21% of patients undergoing UAE, and is a self-limiting condition managed with hydration, pain control, and anti-inflammatory medications.\cite{4} The persistence of symptoms beyond 1 week should raise suspicion of pyomyoma.\cite{7} Table 2 describes the differentiating features of pyomyoma and PES. Alternate differential diagnoses of pyomyoma include tubo-ovarian abscess, infected ectopic pregnancy, degenerated leiomyoma, or malignancy [Table 3].\cite{8} Pyomyoma might traverse the thickness of the myometrium and rupture into the peritoneal cavity or the

| Embolic agent                  | Complications                                                |
|-------------------------------|--------------------------------------------------------------|
| Polyvinyl alcohol             | Uterine ischemia, endometrial infection, ovarian dysfunction, |
|                               | tubal dysfunction, permanent amenorrhea, and pulmonary       |
|                               | embolism                                                     |
| Tris-acryl gelatin microspheres | Permanent amenorrhea                                          |
| Gelatin sponge                | Acute inflammatory reaction; abscess formation; and vascular |
|                               | recanalization                                              |

**Table 2:** A 44-year-old woman with uterine fibroids status post-UAE presented with vaginal bleeding and fatigue. Table described the differentiating features of pyomyoma and post-embolization syndrome.

|                        | Pyomyoma | Post-embolization syndrome |
|------------------------|----------|----------------------------|
| Foul smelling vaginal discharge | Present  | Absent                     |
| Course of symptoms     | Over days to weeks after UAE | Within a few days of UAE   |
| Leukocytosis           | High-grade leukocytosis    | Reactive leukocytosis      |
| Response to therapy    | Responds to antibiotic therapy | Responds to anti-inflammatory medication |
| Prognosis              | Worse     | Good                       |

UAE: Uterine artery embolization.
endometrial cavity resulting in peritonitis or pyometra, respectively, as shown in [Figures 1 and 5].

Imaging findings of pyomyoma on ultrasonography demonstrate a heterogeneous mass with solid and cystic echogenic foci. The reverberation artifacts and echogenic foci within the lesion indicate suppurative (gas forming) pyomyoma. In the presence of concomitant endometritis, there are abnormal echoes in the uterine cavity, as shown in [Figure 6]. CT images displaying gas-filled uni- or multilocular mass suggest contained pyomyoma [Figure 5]. Gas within leiomyoma can also be considered a normal post-UAE finding in patients with no signs of infection. The ruptured pyomyoma is evident by visualizing the disrupted wall of the leiomyoma and the presence of gas and fluid in the peritoneal cavity.

Pre-procedural MR imaging assists in a comprehensive evaluation of potential candidates for UAE, thereby reducing the post-procedural complications. Fibroids with high signal intensity on T2-weighted (T2W) MR imaging show greater size reduction in size following UAE. Furthermore, fibroids with strong perfusion on T1 perfusion imaging adapt to UAE better than those with weak perfusion. Demonstration of infarcted fibroid and pedunculated fibroid on MR imaging is considered a contraindication to UAE. Hemorrhagic infarction (red degeneration) displays high signal intensity on T1-weighted (T1W) imaging and variable intensity on T2W imaging. In addition, MR imaging assesses the post-UAE response of leiomyomas and identifies the complications such as pyomyoma.

MR imaging of pyomyoma shows a cystic mass that is hypointense to iso-intense on T1W imaging, and hyperintense on T2W imaging. Besides, similar findings on MR imaging can be elicited in patients with degenerated leiomyoma. The demonstration of the fibrous capsule surrounding the mass is the distinguishing feature that is indicative of pyomyoma.

### Table 3: A 44-year-old woman with uterine fibroids status post-uterine artery embolization presented with vaginal bleeding and fatigue. The table describes the magnetic resonance imaging features of the differential diagnosis of pyomyoma.

| Differential diagnosis       | MRI findings                                                                 |
|------------------------------|------------------------------------------------------------------------------|
| Tubo-ovarian abscess        | Lateral adnexal location  
Cystic component: Hypo-intense on T1W imaging and hyper-intense on T2W imaging  
Post-contrast: Thin enhancing rim |
| Red degenerated leiomyoma   | Diffuse or peripheral hyperintensity on T1W imaging and variable intensity on T2W imaging  
Rim: Hyperintense on T1W imaging and hypointense on T2W imaging; ill-demarcated border  
Post-contrast imaging: No enhancement |
| Endometritis                 | Enlarged uterus with hyperintense hematoma in the endometrial cavity on T1W imaging; signal voids due to gas on both T1W and T2W imaging  
No fibrous capsule |
| Malignant leiomyosarcoma     | >50% of fibroid appearing hyperintense on T2W imaging; Multiple hyperintense areas on T1W imaging indicating indicate necrosis and hemorrhage  
Post-contrast imaging: Avascular pocket-like necrotic areas |

 ![Figure 5](image_url)  
A 44-year-old woman with uterine fibroids status post-uterine artery embolization presented with vaginal bleeding and fatigue. (a) Sagittal and (b) coronal CT demonstrates the fundal necrotic fibroid with perforation (arrow), notice multiple non-enhancing fibroids (star).

![Figure 6](image_url)  
A 44-year-old woman with uterine fibroids status post-uterine artery embolization presented with vaginal bleeding and fatigue. Gray and color Doppler images of the uterus post-day 30 after embolization demonstrate thickened and heterogeneous content in the cavity with diminished vascularity indicating necrotic area (arrow).
The fibrous capsule appears hyper-intense on T1W imaging and hypo-intense on T2W imaging, and the intensity of the signal from the fibrous capsule is influenced by the free radicals generated by the macrophages. The low apparent diffusion co-efficient value and hyper-intensity on diffusion-weighted MR imaging imply pus in the pyomyoma.

The definitive therapy of pyomyoma includes intravenous antibiotics, and myomectomy or hysterectomy, depending on the patient's desire for future pregnancy and the size and number of pyomyoma. In a few patients with ruptured pyomyoma, CT- or laparoscopic-guided peritoneal drainage and lavage can be considered with antibiotic therapy.

CONCLUSION

High index of clinical suspicion is imperative in diagnosing pyomyoma, given its rare occurrence and non-specific presentation. A clinical triad of sepsis, leiomyoma, and no known source of infection is suggestive of pyomyoma. Correlation of clinical and multimodality radiological findings aids in accurate diagnosis. On CT imaging, specific signs of discontinuity in the wall and intraperitoneal gas/fluid indicate a rupture of pyomyoma and impending peritonitis. Hysterectomy or myomectomy, along with IV antibiotic therapy, is the definitive treatment of pyomyoma. Peritoneal lavage and irrigation are required in patients with pyomyoma ruptured into the peritoneal cavity.

ACKNOWLEDGMENT

None.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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