Radiology Case Reports 14 (2019) 1031–1035

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Case Report

Uterine-sparing management of pyomyoma after uterine fibroid embolization

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ARTICLE INFO

Article history:
Received 20 April 2019
Revised 7 May 2019
Accepted 8 May 2019
Available online 12 June 2019

Keywords:
Uterine artery embolization
Pyomyoma
Uterine-sparing

ABSTRACT

Uterine fibroid embolization (UFE) is an increasingly popular treatment for uterine fibroids. One extremely rare complication after fibroid embolization is pyomyoma, which is the localized infection of the leiomyoma after embolization. Only 10 cases of pyomyoma after UFE have been reported in the literature. We present a case of delayed submucosal pyomyoma identified on computed tomography after 42 days post-UFE. While the majority of previously reported cases were managed by hysterectomy, our patient was treated with a uterine-sparing hysteroscopic transcervical approach. A high level of clinical suspicion is necessary to diagnose this complication after UFE to avoid major morbidity. Submucosal pyomyomas offer a favorable anatomical location easily accessible by hysteroscopy and a conservative approach may be sufficient to manage this complication.

Introduction

Uterine fibroids are the most common benign gynecological neoplasm occurring in up to 70% of the women by the time they reach 50 years old [1]. Common presentations include abnormal uterine bleeding, fertility, or obstetric complications and bulk symptoms, such as constipation and urinary incontinence. While surgical removal of the uterus can effectively treat these fibroid-related symptoms, uterine fibroid embolization (UFE) has also gained popularity as an alternative to hysterectomy given its shorter hospitalization time, faster recovery, and lack of general anesthesia risk among other reasons [2,3]. Compared to surgery, embolization carries a lower risk of infection and deep venous thrombosis. One extremely rare complication of fibroid embolization is pyomyoma, which is the localized infection of the embolized leiomyoma after developing necrosis. Only 10 cases of pyomyoma after UFE have been reported in the literature and 9 out 10 were treated with hysterectomy [4–12] The most common presentation of pyomyoma after UFE can be rather nonspecific and mimic postembolization syndrome, with overlapping

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https://doi.org/10.1016/j.radcr.2019.05.009

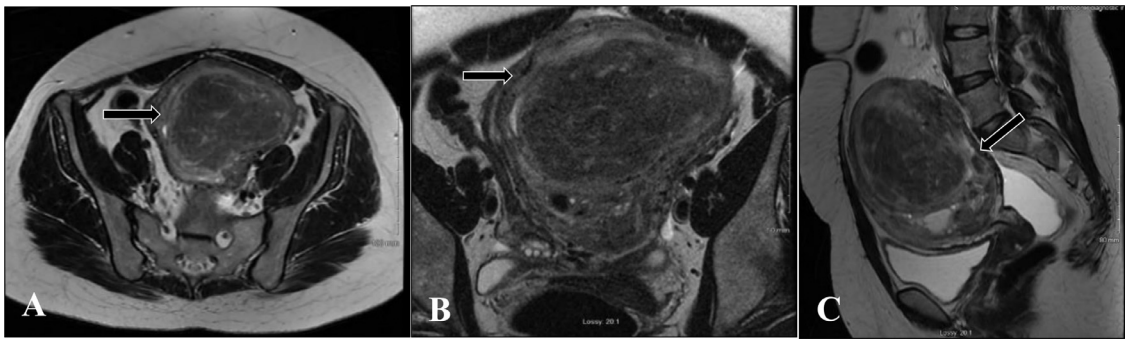

Fig. 1 – T2 weighted MRI of abdomen and pelvis demonstrating large submucosal uterine fibroid (black arrow) prior to embolization: axial (A), oblique coronal (B), and sagittal views (C).

symptoms of fever, abdominal pain, vaginal discharge, and elevated white blood cells [4–12]. Therefore, pyomyomas must be diagnosed in a timely fashion to avoid major morbidity.

In this article, we describe a case of delayed pyomyoma after UFE managed with a uterine-sparing transcervical approach. A survey of the literature was also performed to identify potential common anatomical, prognostic, and histological characteristics leading to this complication.

Case Report

A 28-year-old nulliparous woman with past medical history of successfully treated tibial chondrosarcoma and known uterine fibroids presented with menorrhagia. She initially underwent an abdominal myomectomy for a large 10 cm fundal fibroid at the age of 21. Two years later, she was found to have another 12.5 × 9.2 × 15 cm fibroid within the posterior myometrium and again elected to undergo abdominal myomectomy. During the postoperative period, she developed deep venous thrombosis and pulmonary embolism requiring anticoagulation. Three years later, she presented with another large fundal uterine fibroid measuring 3.9 × 3.5 × 4.6 cm and insisted on avoiding surgery this time. Her bleeding could last up to 1 month, requiring her to change tampons every 30 minutes during menstruation; she initially elected conservative management with levonorgestrel-releasing intrauterine device. Her symptoms improved and remained well-controlled for 1 year. When her bleeding symptoms returned yet again, at this current visit, she wanted to receive a more definitive treatment. Contrast enhanced magnetic resonance imaging demonstrated a dominant 10.6 × 8.2 × 8.0 cm fundal submucosal uterine fibroid with multiple smaller fibroids abutting the endometrial cavity with associated cystic and hemorrhagic changes (Fig. 1). Patient agreed to proceed with uterine artery embolization.

A standard UFE was performed using Embospheres microspheres (Embospheres, Meritt Medical Systems, South Jordan, UT) 500–700 microns followed by 700–900 microns until near stasis was achieved (Fig. 2). The procedure was technically successful with no periprocedural complications. The patient was observed overnight receiving patient controlled epidural analgesia and was discharged home with ciprofloxacin (500 mg PO Q 12 hour) and metronidazole (500 mg Q 8 hour) for 7 days, in addition to pain medications and antiemetics.

On postprocedure day (PPD) 42, the patient presented to the emergency department with 1-day history of fever, abdominal pain, vomiting, diarrhea, and gray-red vaginal discharge with clots. She had a documented temperature of 102.8 F at home. In the emergency room, her blood work revealed elevated white blood cell count of 31.4 k/uL. Urinalysis was unremarkable. Contrast enhanced abdominopelvic CT demonstrated fluid filled distended uterus with foci of gas (Fig. 3). Based on these findings, pyomyoma with endomyometritis was suspected. She was taken to the operating room for transcervical myomectomy and drainage.

A large amount of prolapsed tissue was observed protruding through the external cervical os. Tissue was grasped and removed with ring forceps, and transabdominal ultrasound was performed intraoperatively to minimize the risk of uterine perforation. Postoperative histological examination of the prolapsed fibroid was consistent with ischemic necrosis and acute inflammation with micro-abscess formation. Embolic material and nonviable leiomyoma tissue were also observed. (Fig. 4). The specimen was not cultured, and the blood cultures obtained preoperatively did not show any bacterial growth.

After transcervical evacuation and drainage, the cervix was hemostatic. No periprocedural complications were encountered. Postoperatively, she was started on IV Vancomycin and Piperacillin/Tazobactam and then converted to oral antibiotics after 48 hours. Her leukocytosis continued to improve and resolved on postoperative day 4 when she was discharged home in stable condition.

The patient followed up in clinic 2-weeks and at 6 months respectively without issues. She continued to pass tissue fragments occasionally per vagina without heavy menses, pelvic pain, fever, or chills. MRI indicated interval resolution of pyomyoma with a remnant of the submucosal fibroid measuring 3 cm x 3 cm without any contrast enhancement (Fig. 5).

Discussion

Though 50 pyomyoma cases were identified since 1945 through a Medline search of the English-language literature [13], only 10 were cases of pyomyoma after uterine emboliza-

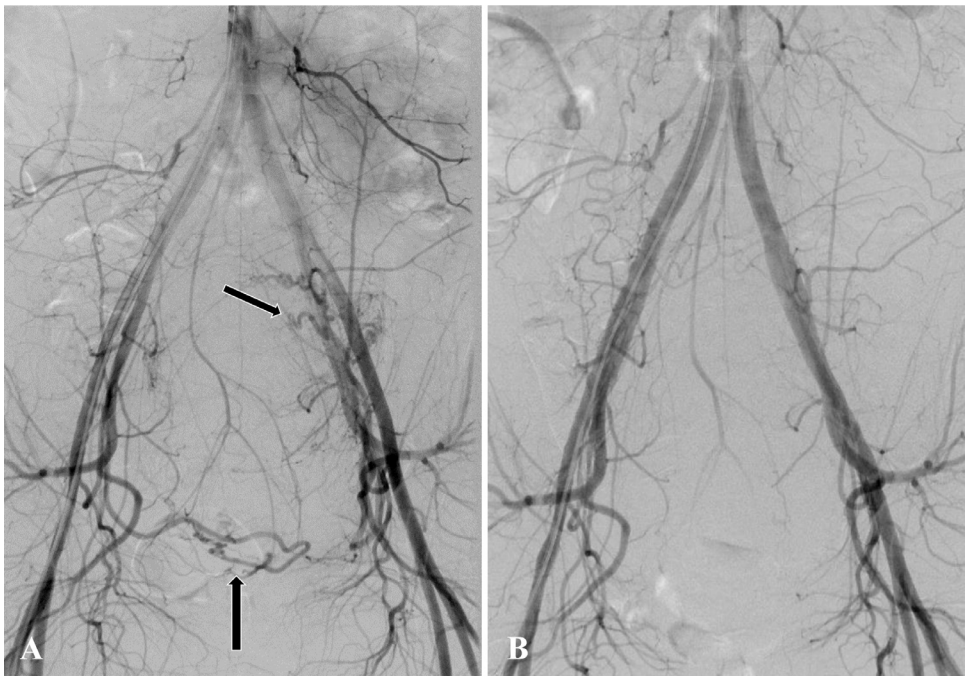

Fig. 2 – Digital subtraction angiography (DSA) images during uterine artery embolization (UAE) procedure. (A) Pelvic aortic angiogram demonstrating prominent uterine arteries (black arrows) prior to uterine artery embolization. (B) DSA post embolization showing effective embolization of the uterine arteries.

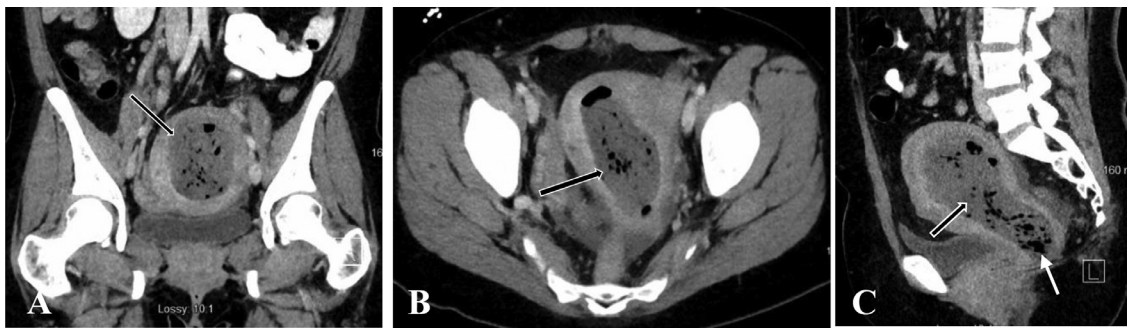

Fig. 3 – Pyomyoma diagnosis: CT of abdomen and pelvis demonstrating pyomoma (black arrows) complex fluid collection with foci of gas within the endometrial cavity in the coronal (A), axial (B), and sagittal (C) views. Pyomyoma is seen prolapsing into the endocervical canal with a distended and thinned cervix (white arrow).

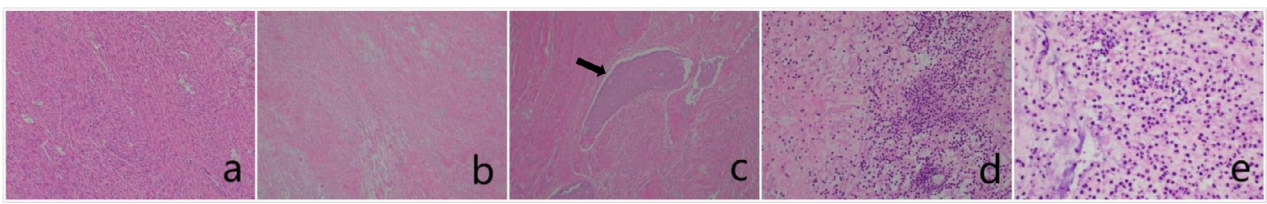

Fig. 4 – Histology of the prolapsed fibroid evacuated via D&C. (A) Normal leiomyoma tissue with spindle cells (10×); (B) nonviable leiomyoma tissue (10×); (C) embolization material within vessels (black arrow) surrounded by nonviable leiomyoma tissue; (D) hematoxylin and eosin (H&E) staining showing groups of neutrophils (left side) near "dead" fibroma tissues (right side) suggesting abscess formation (20×); (E) H&E staining showing groups of neutrophils near "dead" fibroma tissues suggesting abscess formation (40×).

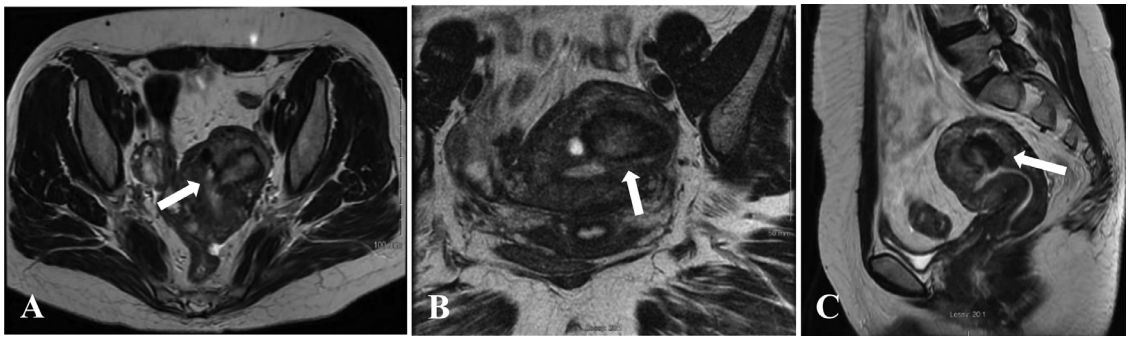

Fig. 5 – T2 MRI of abdomen and pelvis demonstrating approximately 6 months postuterine fibroid embolization. The fibroid remnant (white arrow) is seen in a transmural location projecting into the endometrial cavity: coronal (A), axial (B), and sagittal (C) views.

tion [4–12]. Patient age in the studies ranged from 34 to 65 years. The patient in our case was only 28 years old, representing the youngest reported to date. Among the reported cases in which fibroid locations were described, all 3 pyomyomas arose from intramural leiomyomas with diameters greater than or equal to 7 cm [4,5,7]. Our patient's fibroid measured 10 cm, the largest reported thus far. In addition to fibroid size, we also attempted to identify other potential risk factors for pyomyoma. Given limited information provided in these case reports and case series, it was not possible to make any association between fibroid size or other medical comorbidities with the risk of developing pyomyoma after UFE.

Of the 10 cases identified, only 2 presented with malodorous vaginal discharge [10,12]. Most patients presented with fever, pelvic pain, and leukocytosis [4–12]. These findings were also consistent with postembolization syndrome, which can occur in up to 21% patients [14]. Furthermore, whereas postembolization syndrome symptoms typically peak at 48 hours postprocedure and abate within a week, the majority of patients reported in the literature were still symptomatic between the first and second weeks post-UFE. In terms of imaging findings, patients with pyomyoma usually showed an enlarged, irregular uterus, heterogeneous tissue appearance, free pelvic fluid, and foci of gas on US, CT or MRI [4–12]. Nevertheless, these findings can be observed in cases after UFE and are considered a normal process following tissue infarction-desiccation [15]. Though this necrosis-related cavitation can appear up to 1-month postembolization, it could occur as early as 10 days in cases diagnosed with pyomyoma [16]. For those diagnosed with pyomyoma after 1 month, however, additional constitutional symptoms such as fever and abdominal pain are present along with leukocytosis. The diagnosis of a pyomyoma should be made on the basis of clinical symptoms, laboratory values, and imaging studies combined [11].

Once a pyomyoma is suspected, hysterectomy is currently the standard management. Purulent peritoneal fluid[11], myometrial abscess formation[5], endometrial ulceration[5], inflammation and adhesions involving adjacent structures such as the fallopian tubes [5–7], ovaries[6], appendix[4], bladder[4], and omentum[4] may be present during surgical exploration depending on the severity of the infectious process. In 1 case in which a pyomyoma was perforated, laparoscopic drainage and irrigation were performed without performing a hysterectomy [7]. Despite an uneventful recovery, the patient underwent surgery for adhesion removal 3 months later [7]. In comparison, we adopted a hysteroscopic transcervical approach with ultrasound guidance. This is the second reported case in which a pyomyoma is managed via transcervical approach after UFE. In the other reported case, a patient underwent emergent uterine artery embolization in the management of postpartum hemorrhage; she was only diagnosed with uterine fibroids after the procedure [12]. Patient was readmitted later with fever, leukocytosis, and foul-vaginal discharge. Dilation and curettage with rectoscope were performed for evacuation of fluid collection and final pathology report suggested diagnosis of pyomyoma. Unfortunately, though the authors mentioned that the original leiomyomas were endometrial and myometrial, the exact location of the pyomyoma was not reported in their manuscript. Furthermore, our patient did not undergo any instrumentation prior to or after UFE, while this reported case took place after a complicated vaginal delivery, which could serve as a potential infectious source. Nevertheless, both patients recovered uneventfully and were discharged within a week. For submucosal pyomyomas a uterine-sparing transcervical approach may be sufficient to manage this complication and therefore avoid major surgery.

Conclusion

Overall, all reported cases in the literature recovered well after pyomyoma treatment regardless of type of therapy and were discharged within a week without further complications [4–12]. No deaths were reported following surgical and medical management. Seven out of nine reported patients underwent hysterectomy. We were able to perform a transcervical myomectomy and drainage in our case taking advantage of its favorable submucosal location. Whereas intramural and subserosal pyomyomas might require a more invasive surgical approach such as hysterectomy. It is important for operators to be aware of the clinical, imaging, and anatomical features of pyomyoma in order to diagnose, treat, and adopt the most

minimally invasive intervention to treat this uncommon complication after UFE.

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