

Uterus-preserving management of pyomyoma: Case report of a rare complication after uterine artery embolization

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ABSTRACT

Background: Pyomyoma of the uterus is a rare but severe complication of uterine artery embolization (UAE). This report describes the uterus-preserving management of a case of fast-developing pyomyoma. Screening methods to minimize the risk of this condition are discussed.

Case: A 46-year-old woman presented with fever, abdominal pain and blood stream infection on the third day after UAE, which had been performed to treat symptomatic uterine myomatosis. Broad-spectrum antibiotics proved inadequate. Magnetic resonance imaging (MRI) showed a possible superinfection of the necrotic myoma. Vaginal smear showed the same organism, a resistant *Escherichia coli*, as in the blood culture. Because the patient declined hysterectomy, multiple hysteroscopies with removal of necrotic pyomyoma were performed. In addition, an intrauterine gentamicin chain was placed. To our knowledge, this is the first case of pyomyoma immediately after UAE and the first report of successful hysteroscopic treatment in a septic patient.

Conclusion: In order to reduce the risk of pyomyoma, pre- and postinterventional algorithms should be used.

1. Introduction

Leiomyoma is the most common benign tumor of the uterus and often results in hypermenorrhea, dysmenorrhea, lower abdominal pain in general, or a reduction in the chance of a successful pregnancy [1].

High-intensity focused ultrasound (HIFU) and uterine artery embolization (UAE) are conservative alternative treatments to surgery. In UAE, a hemorrhagic infarction of fibroids is induced while maintaining endometrial and myometrial perfusion [2]. Although UAE is usually safe and effective, pyomyoma is a very rare but severe complication [3]. A pyomyoma presents itself as an intramyometrial abscess which can lead to blood stream infection, sepsis and death unless it is immediately treated with antibiotics and surgical intervention [4].

2. Case Presentation

A 46-year-old woman presented with recurrent uterine myomatosis and anemic hypermenorrhea. T2-weighted magnetic resonance imaging (MRI) showed two non-homogeneous and signal-intense myomas with a total area of 12 × 8 cm, in the posterior uterine wall.

The patient had undergone multiple abdominal operations to treat

her Crohn's disease. Therefore, the risk of surgical intervention appeared to be inadmissibly high. High-intensity focused ultrasound was not a treatment option because of the localization of the myomas in the posterior uterine wall and their hyperintensity on T2-weighted MRI (Funaki 3). Conservative treatment with oral medication was not considered because it would have to be continued long term, through to menopause. Uterus artery embolization (UAE) was therefore recommended, though it can be associated with complications in future pregnancies [5].

UAE was performed successfully and without periprocedural complications. The patient complained of pain afterwards, but UAE is known to be painful; in addition, because the patient was allergic to PCIA she was on a low level of analgesia. Her report of pain was therefore not unexpected.

Three days later, the patient presented with fever up to 39.4 °C and high infection parameters (C-reactive protein 290 mg/L (R < 5), leukocytosis 17 10⁹/L (R 4.4–11.3), PCT 2.1 µg/L (R < 0.1)). In the blood culture a multi-resistant *Escherichia coli* (3MRGN, afterwards corrected to 4MRGN with carbapenemase production) was detected. Piperacillin/tazobactam antibiotic treatment was started on same day. Due to an allergic reaction, it was changed to meropenem. At first this was suspected to be a new episode of the patient's preexisting Crohn's

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disease.

Despite antibiotic treatment, the fever, high infection parameters and lower abdominal pain persisted. Computerized tomography (CT) showed a necrotic lesion in the position of the former myoma, as well as a possible craniolateral abscess formation. Since gas inclusions are a regular radiological finding after UAE, interpretation of the imaging was difficult [6]. A complementary MRI scan showed a possible superinfection of necrotic myoma with gas inclusions (see Figs. 1 and 2).

Vaginal smear showed the same organism as in the blood culture (4MRGN). A proposal for hysterectomy to remove the focus of infection, to avoid a severe septic course, was refused by the patient several times, so it was decided to perform a transcervical tissue removal eight days after UAE. Hysteroscopy was the only surgical treatment option the patient agreed to. The risk of aggravating the inflammation by dissemination of the infective material via the fallopian tubes into the abdomen seemed to be acceptably low.

In total, 320 g of tissue (Fig. 3) was resected with a combination of hysteroscopy and the use of a grasping forceps.

Because of persistent elevated infection parameters and abdominal pain, as well as episodic fever, three additional similar tissue-removing operations followed, without complication. In an in-vitro microbiological test gentamicin proved to be effective against the multi-resistant *E. coli*. During the third intervention, a gentamicin chain (containing 17 mg of gentamicin in 10 balls) was placed inside of the uterine cavity to promote healing. These chains are most commonly used in orthopedic infected osteomyelitic cavities and have a highly localized effect, which reduces the risk of gentamicin side-effects [7]. The chain was removed before discharge two weeks later.

In intrauterine swaps, the same organism as in the blood culture and vaginal smear (4MRGN *E.coli*) was found. When initial 3MRGN was corrected to 4MRGN, antibiotic treatment was switched to ceftazidim + avibactam and metronidazol for a total of 14 days. The patient showed a good clinical and laboratory response to therapy. The patient remained stable, without fever; her pain was well controlled and her inflammatory parameters returned to normal. She was discharged from hospital four weeks after the UAE.

Multiple follow-up examinations were performed. After discharge the patient presented with sonographic intrauterine fluid accumulation of 34x31x47mm with multiple gas inclusions. A transcervical drainage tube was inserted and after the fluid had drained off, it was removed after five weeks.

The patient remained well, without any myoma-related complaints.

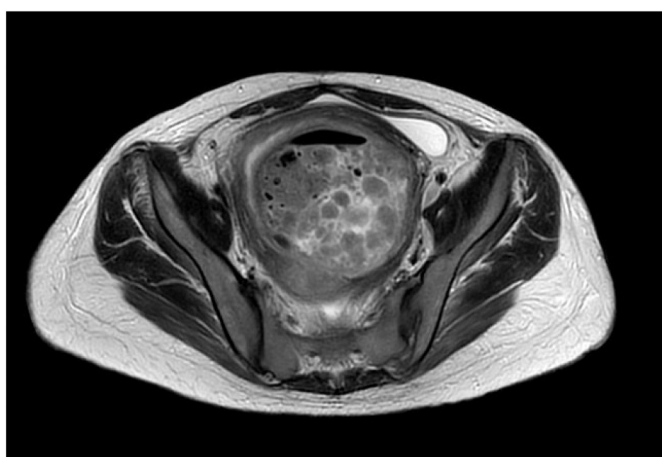


Fig. 1. Preoperative T2-weighted axial MRI scan showing necrotic myoma with hyperintense signals. The myoma is displacing the uterine cavity to the right. Multiple point-shaped regions with signal loss represent diffuse gas distribution inside the myoma. At the anterior border of the myoma, a larger collection of gas can be seen.



Fig. 2. Transvaginal sagittal ultrasound with hypoechoic non-homogeneous regions inside the myoma, representing necrosis. Multiple hyperechoic foci were compatible with gas bubbles.

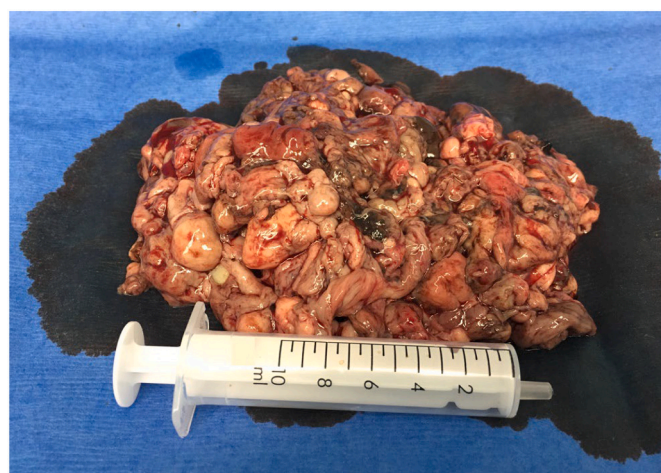


Fig. 3. Tissue (320 g) removed intraoperatively, comprising necrotic components of the myoma.

3. Discussion

Since its first description in the year 1871, fewer than 100 cases of pyomyoma have been reported, and only 10 after UAE [8].

Initial symptoms can easily be confounded with post-embolization syndrome after UAE. Pain, moderate fever and elevated infection parameters caused by necrosis of leiomyomas can be expected.

Without adequate and immediate treatment, mortality of pyomyoma reaches up to 20% [8,9]. Therefore, the timely identification and treatment of pyomyoma are essential.

Gas inclusion seen on CT or MRI is considered a normal finding after UAE [6] in the absence of any clinical or laboratory signs of infection. Spaces left by tissue infarction may be an explanation for this. Localized gas collection of a necrotic myoma combined with signs of infection can be an indication of pyomyoma [10]. It remains difficult to diagnose pyomyoma on all current imaging modalities [11].

Hysterectomy, besides broad-spectrum antibiotic treatment, is the therapy of choice for pyomyoma [8,12,13]. Consequently, only a few cases of uterus-preserving management have been reported. Magro and Gafson presented a case of post-partum pyomyoma, where four liters of pus was drained transabdominally, and no hysterectomy was performed [14]. Two cases of spontaneous pyomyoma after abortion were reported by Pinton and Bagga, where laparotomy with drainage and

myomectomy were performed [15,16]. Pinto reported a patient who underwent laparoscopic drainage and lavage 8 weeks after UAE [12]. Yu et al. were the first to attempt a hysteroscopic treatment in a non-septic patient with pyomyoma one day after UAE [17].

In general, uterus-preserving management in cases of pyomyoma should be preferred only if there is a wish to preserve fertility.

To our knowledge, this is the first successful transcervical treatment approach in a critically ill patient with sepsis.

Hysteroscopically assisted transcervical resection is an alternative to laparoscopy and laparotomy, especially in patients with a high operative risk in abdominal surgery. Chronic diseases of the gastrointestinal tract, other risk factors like diabetes and immunocompromised status or an existing uterine or cervical infection should be identified before performing UAE [12].

4. Conclusion

As UAE is a well-established procedure performed in patients suffering from symptomatic myoma, physicians involved in the treatment need to be familiar with potential complications and imaging in order to ensure appropriate and fast diagnosis and treatment.

For a patient with fever, high inflammatory parameters and abdominal pain after UAE, a pyomyoma should be considered. The period of occurrence can extend from immediately after intervention up to weeks afterwards.

Aside from broad-spectrum antibiotics, surgical treatment is the therapy of choice to remove infected necrotic tissue. Transcervical intervention is a possible uterus-preserving alternative to abdominal surgery. An intrauterine gentamicin chain was used to promote healing and this needs further evaluation.

A detailed medical history and vaginal smear can indicate dysbiosis or infection and so should be considered before any intervention. If infection is detected, a pre-interventional antibiotic treatment could be considered to avoid the occurrence of pyomyoma. The detection of infection indicates that the patient is at high risk of developing a pyomyoma after UAE.

Contributors

Jana Busshoff was involved in clinical treatment and patient care and wrote the case report.

Fabinshy Thangarajah was involved in clinical treatment and patient care and wrote the case report.

Bernd Morgenstern was involved in clinical treatment and patient care, performed the hysteroscopic resection and wrote the case report.

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Patient consent

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

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