Case Reports in Women's Health 26 (2020) e00186

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

Pyomyoma after uterine artery embolization: A case report

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ARTICLE INFO

Article history:
Received 22 January 2020
Received in revised form 25 February 2020
Accepted 28 February 2020

Keywords:
Pyomyoma
Uterine artery embolization

ABSTRACT

Background: Uterine pyomyoma is a rare complication of uterine artery embolization (UAE), and causes significant morbidity and mortality. This report describes the diagnosis and prompt management of this condition.
Case: A 48-year-old woman presented with fever, chills, and diffuse abdominal pain 15 days after undergoing UAE for symptomatic fibroids. Computed tomography showed the uterus to be significantly distended, with multiple intra-cavitary masses containing a large amount of gas and air–fluid level. Sepsis secondary to post-UAE pyomyoma was suspected. Hemodynamic resuscitation and broad-spectrum antibiotics were immediately started. The patient underwent emergency exploratory laparotomy with total hysterectomy.
Conclusion: In order to ensure appropriate and timely intervention, the diagnosis of uterine pyomyoma should be considered in patients presenting with signs of infection and abdominal pain after UAE.

1. Introduction

Uterine leiomyoma is the most common benign tumor of the uterus and a common indication for uterine artery embolization (UAE) for patients desiring conservative, non-surgical management. One of the very rare yet serious complications of UAE is pyomyoma [1]. Pyomyoma, also known as suppurative leiomyoma, is intra-myometrial abscess formation leading to sepsis. Without prompt diagnosis and treatment, pyomyoma carries a significant risk of morbidity and mortality [2].

2. Case Report

A 48-year-old woman, G6P2042, presented to the emergency department with fever, chills and diffuse abdominal pain 15 days after undergoing UAE for symptomatic uterine fibroids at another facility. The patient initially presented with abnormal uterine bleeding and dysmenorrhea secondary to large uterine fibroids. After counseling on her treatment options, she elected for UAE. The patient's obstetrical history was significant for two term uncomplicated vaginal deliveries and four spontaneous abortions. Her past medical history was significant for hypertension and obesity class III. Her past surgical history was significant for cholecystectomy, thyroidectomy and left knee meniscal repair.

The patient's initial vital signs were temperature of 101.5F, heart rate of 110 beats per minute and respiratory rate of 24–37 breaths per minute with a normal blood pressure. Her abdomen was distended and diffusely tender. Her pelvic exam was remarkable for scant blond-tinged vaginal discharge. Her white blood cell count was 26,300 with 91% neutrophils. Computed tomography (CT) scans of the abdomen and pelvis showed a distended uterus with multiple intra-cavitary uterine masses spanning 13 cm × 13 cm × 12 cm, containing a large amount of gas and air–fluid level (see Figs. 1 and 2). The CT findings and clinical picture raised suspicion for sepsis secondary to post-UAE pyomyoma. The patient was immediately started on broad-spectrum intravenous antibiotics (Cefepime, flagyl and amikacin) and fluid resuscitation with lactated ringers. We decided to proceed with an exploratory laparotomy and possible hysterectomy versus abscess drainage. Intraoperative findings were consistent with an extensive pelvic infection: purulent material in the pelvis, and an enlarged uterus with an inflamed, necrotic uterine fundus adherent to severely inflamed small bowel. In addition to the total hysterectomy and bilateral salpingectomy, the patient underwent a small bowel resection with primary end-to-end anastomosis. Frozen section of the uterus was negative for malignancy and, given the patient's age and grossly normally appearing adnexa, both ovaries were spared. Post-operatively, the patient was transferred to the surgical intensive care unit for close monitoring. She continued on a prolonged course of antibiotics and was discharged from the hospital three weeks later.

An incidental finding of a poorly differentiated serous carcinoma of the fallopian tube was diagnosed on final pathology. One month later, the patient underwent a staging procedure revealing stage 1a fallopian tube carcinoma. The patient is currently doing well and is following up with the oncology service.

3. Discussion

Since its first description in 1871, fewer than 100 cases of pyomyomas have been reported. They have been described in postpartum patients,

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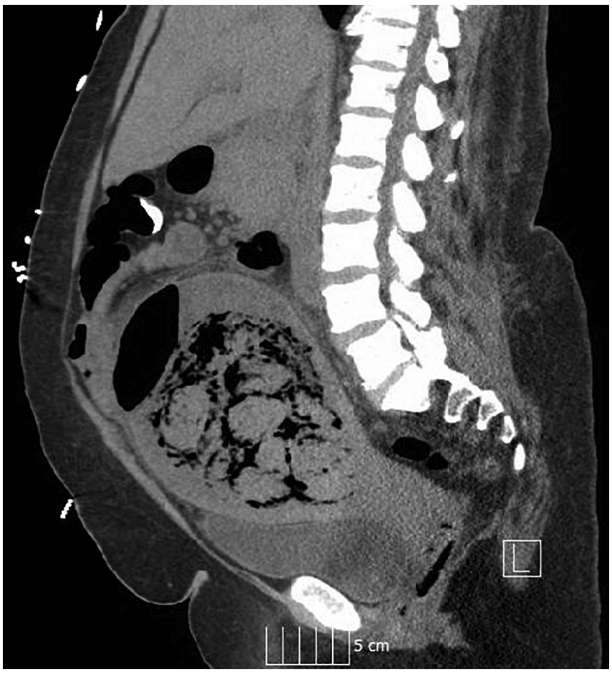

Fig. 1. Sagittal computed tomography image with intra-venous contrast showing a distended uterus with multiple intra-cavitary masses spanning containing a large amount of gas with air-fluid level.

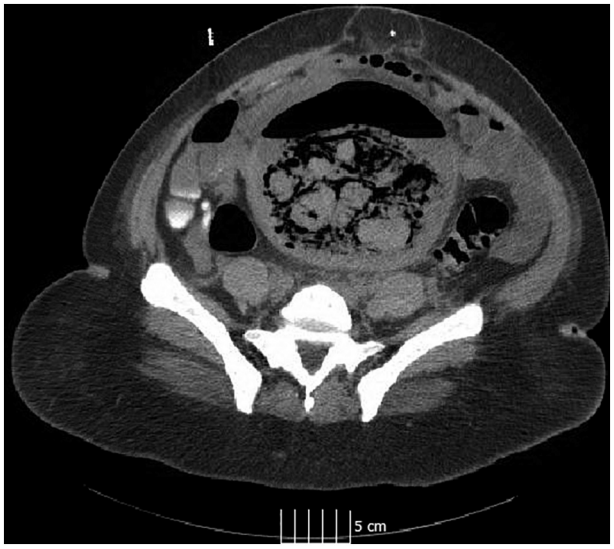

Fig. 2. Transverse computed tomography image with intra-venous contrast showing a distended uterus with multiple intra-cavitary masses spanning containing a large amount of gas with air-fluid level.

after gynecological procedures and following UAE, and can also occur without an inciting factor [2–4]. The occurrence of pyomyoma after UAE poses a diagnostic challenge since its presentation resembles that of post-embolization syndrome (PES), which consists of fever, pelvic pain and leukocytosis within 1 week of UAE. PES is a self-limited complication occurring in about one-third of UAE cases and is typically managed conservatively with hydration, anti-inflammatory medications and pain control [5]. However, the development of fever, pelvic pain, and leukocytosis beyond one week following UAE should raise clinical concern for pyomyoma [5,6]. The route of infection leading to a pyomyoma often includes direct spread from the endometrial cavity, spread from adjacent structures such as the adnexa or the bowel, and hematogenous or lymphatic spread [2]. Pyomyoma is often polymicrobial. Prompt diagnosis is essential as pyomyoma may lead to serious complications such as myometrial rupture with peritonitis, renal cortical necrosis, deep vein thrombosis, endocarditis, pancreatitis and death [3]. Without appropriate treatment, the mortality rate reaches 20% [7]. Nonetheless, pyomyomas are difficult to definitively diagnose in the post-UAE patient because necrosis of leiomyomas is expected. On imaging, visualization of gas in the uterus is often expected following UAE. This is thought to be the result of gas filling potential spaces left by tissue infarction and desiccation. It is considered a normal finding, but only in the absence of signs of infection. In the reported cases of pyomyoma after UAE, the most common radiologic finding was the presence of an abundance of intrauterine air [2]. Thus, clinical acumen and a high index of suspicion are imperative. Hysterectomy with antimicrobial coverage is the treatment of choice for pyomyoma [4,8,9]. However, there have been reports of conservative therapies primarily for fertility preservation. Magro and Gafson [4] reported a case of a postpartum pyomyoma in which four liters of purulent material was drained from an anterior uterine myoma. They did not perform myomectomy or hysterectomy. Moreover, Pinto and his colleagues [8] reported a case of a perforated pyomyoma eight weeks after UAE. The patient underwent laparoscopic drainage and lavage of the leiomyoma, with no attempt at resection. Also, Pinton and his colleagues [9] reported a case of pyomyoma two weeks following a spontaneous abortion in a 28-year-old patient desiring future fertility. After 10 days of antibiotics with clinical deterioration, the patient underwent a laparotomy with drainage of purulent material and a myomectomy. Since the present patient, at age 48, had no desire for future childbearing, we proceeded with hysterectomy.

4. Conclusion

This case serves as an example of the importance of maintaining a high level of suspicion for uterine pyomyoma and taking quick action to avoid morbidity and mortality. Fever, pelvic pain and leukocytosis occurring over 1 week after UAE should raise suspicion for pyomyoma. It is important for all physicians, including radiologists, interventional radiologists, emergency physicians, and gynecologists, to be aware of this entity and its serious consequences.

Contributors

Dima Ezzedine is the first author and was responsible for manuscript writing and the review of the literature.

Chima Ndubizu contributed to the review of the literature and editing of the manuscript.

Sohaib Kayani provided the figures and contributed to the case write-up.

Allison David contributed to revision of the manuscript and the review of the literature, and was responsible for patient contact.

Conflict of Interest

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

Funding

No funding from an external source supported the publication of this case report.

Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

References

[1] P.A. Shukla, A. Kumar, D. Klyde, S. Contractor, Pyomyoma after uterine artery embolization, J. Vasc. Interv. Radiol. 23 (2012) 423–424.
[2] M.L. Rosen, M.L. Anderson, S.M. Hawkins, Pyomyoma after uterine artery embolization, Obstet. Gynecol. 121 (2013) 431–433.
[3] J.S. Greenspoon, M. Ault, B.A. James, L. Kaplan, Pyomyoma associated with polymicrobial bacteremia and fatal septic shock: case report and review of the literature, Obstet. Gynecol. Surv. 45 (1990) 563–569.
[4] M. Magro, I. Gafson, Postpartum pyomyoma: a rare complication of leiomyoma, J. Obstet. Gynaecol. 34 (2014) 202–203.
[5] H. Leonardt, A. Aziz, L. Lonn, Post-embolization syndrome and complete expulsion of a leiomyoma after uterine artery embolization, Acta Obstet. Gynecol. Scand. 84 (2005) 303–305.
[6] C.C. Obele, S. Dunham, G. Bennett, J. Pagan, L.Y. Sung, H.W. Charles, A case of pyomyoma following uterine fibroid embolization and a review of literature, Case Rep. Obstet. Gynecol. (2016) https://doi.org/10.1155/2016/9835412.
[7] Z.H. Chen, H.D. Tsai, M.J. Sun, Pyomyoma a rare and life-threatening complication of uterine leiomyoma, Taiwan J. Obstet. Gynecol. 49 (2010) 351–356.
[8] E. Pinto, A. Trovão, S. Leitão, C. Pina, F.K. Mak, A. Lanhoso, Conservative laparoscopic approach to a perforated pyomyoma after uterine artery embolization, J. Minim. Invasive Gynecol. 19 (2012) 775–779.
[9] A. Pinton, G. Aubry, V. Thoma, I. Nisand, C.Y. Akladios, Pyomyoma after abortion: uterine conserving surgery is possible to maintain fertility. Case report, Int. J. Surg. Case Rep. 24 (2016) 179–181.