

CASE REPORT

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Erosion of calcified uterine leiomyoma into the urinary bladder: A case report

Laura Ramirez-Caban, Maral Malekzadeh, Fernando Cabrera, Adria Suarez-Mora

ABSTRACT

Introduction: Uterine leiomyomas are benign smooth muscle tumors that affect approximately 70–80% of women worldwide. Erosion of leiomyomas into surrounding tissues is a rare complication. We present a case of a postmenopausal female with uterine leiomyoma eroding into the urinary bladder.

Case Report: A 55-year-old, postmenopausal female with history of uterine fibroids previously treated with uterine artery embolization presented with recurrent bladder stones, hematuria, and pelvic pain. Pelvic magnetic resonance imaging described a 4.3 cm heterogenous mass involving the ventral uterine fundus and bladder dome concerning for malignancy. She underwent robotic-assisted total laparoscopic hysterectomy, right salpingo-oophorectomy, left salpingectomy, and partial bladder cystectomy. Pathology was significant for benign calcified leiomyoma and benign calcifications within the bladder mucosa.

Conclusion: Erosion of leiomyoma through the bladder mucosa is rare complication but should be considered in the differential diagnosis for patients presenting with hypercalciuria and recurrent bladder stones in the setting of uterine fibroids previously treated with uterine artery embolization.

Keywords: Bladder erosion, Hypercalciuria, Uterine artery embolization, Uterine fibroids

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INTRODUCTION

Uterine leiomyomas are benign smooth muscle tumors estimated to occur in 70–80% of women by the onset of menopause [1, 2]. Approximately 25% of patients experience symptoms from uterine fibroids requiring intervention [1]. Management of fibroids includes medical, procedural, and surgical treatment options. Uterine artery embolization (UAE) is a procedure used in the treatment of symptomatic fibroids. An embolic agent is delivered through bilateral uterine arteries causing devascularization and involution of uterine leiomyomas [1]. Major complications from this type of procedure range from 1% to 12% and may include unplanned hysterectomy, sepsis, ovarian failure, and pulmonary embolism [1, 3].

Infarction of leiomyoma is an expected phenomenon following UAE. Infarction leads to pain and inflammation typically seen after UAE, but is rarely associated with long-term complications. However, rare reports of neovascularization and fistulization to surrounding organs following UAE have been described in the literature [4–7]. Here we present a case of a benign calcified fibroid eroding into the urinary bladder in a postmenopausal woman with history of fibroids previously treated with uterine artery embolization.

CASE REPORT

A 55-year-old postmenopausal female with history of uterine fibroids presented for evaluation of pelvic pain, hematuria, and recurrent bladder stones.

The patient's history was notable for uterine fibroids treated with uterine artery embolization 12 years prior to presentation during her premenopausal years. She was referred to urology due to a four-year history of recurrent bladder stones treated with extraction. Initial urine studies revealed hematuria and calcium carbonate apatite stones. She underwent computed tomography (CT) imaging and results were notable for a calcified uterine mass with invasion into the urinary bladder. A subsequent pelvic magnetic resonance imaging (MRI) described a 4.3 cm heterogenous mass involving the ventral uterine fundus and bladder dome. Figure 1 shows an MRI sagittal section of the uterine leiomyoma invading into the bladder mucosa. Figure 2 shows an MRI transverse section of the pathology. The patient underwent office cystoscopy with findings notable for a 2 cm mass arising from the posterior bladder. Biopsy of the mass revealed granulation tissue.

She was referred to gynecology oncology due to concerns for possible uterine leiomyosarcoma with invasion into the urinary bladder. On pelvic exam, the cervix was normal and uterus was slightly enlarged. The patient underwent endometrial biopsy and results were significant for benign atrophic endometrium. After counseling, the patient underwent robotic-assisted total laparoscopic hysterectomy, right salpingo-oophorectomy, left salpingectomy, and partial bladder cystectomy. Figure 3 shows an intraoperative view of anterior leiomyoma with invasion into the urinary bladder. Final pathology described a benign uterine leiomyoma with focal degeneration, hyalinization, and calcification. The leiomyoma was 5 cm in greatest dimension with changes consistent with prior embolization. The final pathology for the bladder mucosa was consistent with inflamed granulation tissue focally lined by urothelium with calcification. The surgery was uncomplicated, and she went home with a foley catheter in place on postoperative day 1.

She underwent voiding cystogram 7 days postoperatively and the foley catheter was removed without incidence. The patient's postoperative course and recovery were unremarkable. At her 6-week visit, her postoperative urinalysis was normal and urine culture was sterile. The patient expressed symptoms of urinary frequency and urgency which were treated with a trial of anticholinergics. Six months postoperatively, the patient had resolution of initial presenting symptoms and she did not require further work up for bladder stones.

DISCUSSION

Benign leiomyomas are commonly located within the uterine body without involvement of surrounding tissues. Variations of leiomyoma location have been described in

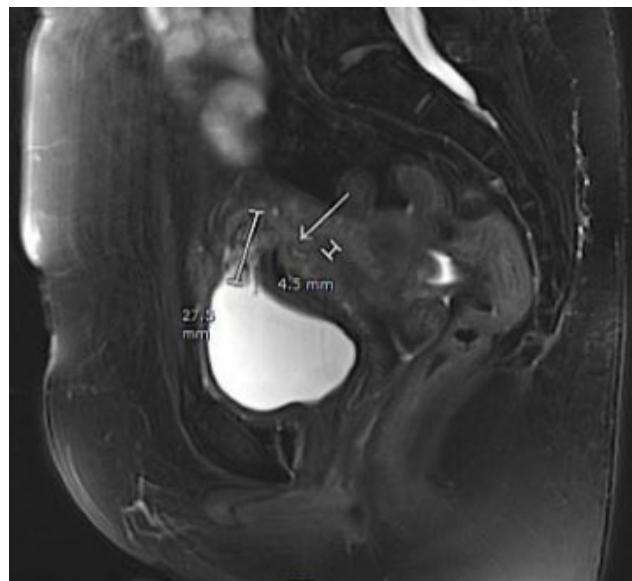


Figure 1: MRI T2-weighted sagittal section of uterine leiomyoma invading into the bladder mucosa.

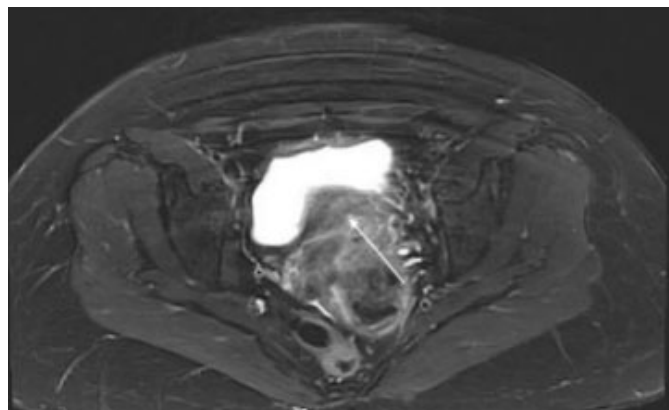


Figure 2: MRI T2-weighted transverse section of uterine leiomyoma invading into the bladder mucosa.

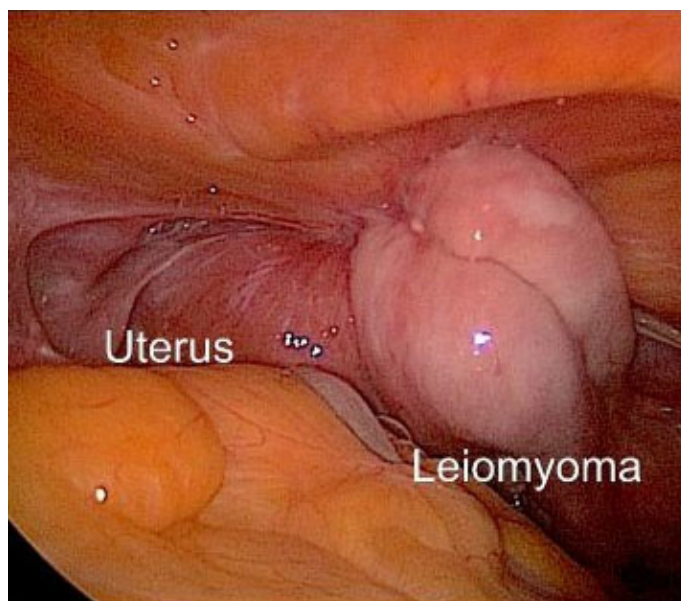


Figure 3: Intraoperative view of anterior leiomyoma with invasion into the urinary bladder.

the literature with rare occurrence. Conditions and events described include: parasitic leiomyomas, leiomyomatosis peritonealis disseminata, bladder leiomyomas, and leiomyoma fistulization following UAE [5–11]. In this report, we describe a rare complication of postembolization vesicouterine fistula formation.

Uterine artery embolization is a minimally invasive procedure used for the treatment of symptomatic uterine fibroids. Uterine artery embolization can be performed for multiple indications including treatment for acute hemorrhage or bleeding, definitive treatment for symptomatic adenomyosis and fibroids, or as a temporizing measure for symptomatic bleeding in anticipation of myomectomy or hysterectomy [3, 12]. During UAE, embolic agents such as polyvinyl alcohol (PVA) and gelatin microspheres of varying sizes are injected into bilateral uterine arteries [12, 13]. The embolic material is carried by the arterial blood flow to vessels feeding the fibroids causing occlusion of fibroids' blood supply. This occlusion ultimately leads to fibroid ischemia while maintaining perfusion to the uterine myometrium [12]. Pelage et al. showed rates of complete fibroid infarction were 70.6% at three months post-procedure, and of those incompletely infarcted fibroids, 30% progressed to complete infarction by three years [3, 14].

Following UAE, fibroids are expected to undergo ischemia and necrosis. Most leiomyomas typically undergo hyaline type necrosis, with about 5% undergoing cystic degeneration and calcification [15, 16]. Following necrosis, fibroids may expulse through the uterine cavity or into the pelvis [17]. Fistulization with other structures or organs is a rare event but may occur after UAE. In our search of the English literature only six case reports were identified which reported vesicouterine fistula from uterine fibroids with three of these occurring following uterine artery embolization [5–7, 16, 18, 19]. Reports of uteroenteric fistulization have also been cited in the literature with equal rare event [8]. Postulated theories surrounding fistulization after UAE have been proposed. One theory proposes that inflammation and necrosis of the uterine tissue after UAE promotes vascularization and fistulization to surrounding structures. Another theory proposes that incomplete fibroid ischemia promotes neovascularization to adjacent tissues and subsequent fistulization [8].

Other phenomenon of leiomyoma neovascularization has been reported. Parasitic leiomyomas are rare variants thought to arise from detached subserosal or pedunculated uterine fibroids that gain new blood supply from reattachment to other tissues [9, 10]. Accounts of parasitic fibroids with neovascularization to bowel and omentum have been cited in the literature [8, 11]. A more extensive condition, leiomyomatosis peritonealis disseminata (LPD), involves proliferation of peritoneal and subperitoneal benign smooth muscle nodules within the peritoneum. Two theories regarding LPD etiology have been proposed. One theory attributes the use of uncontained power morcellation at the time of hysterectomy or myomectomy which allows remnants

of benign fibroids to be disseminated throughout the abdomen forming attachments to multiple organs and structures [9]. A second theory focuses on LPD as a process where peritoneal mesenchymal cells transform into smooth muscle cells in the setting of estrogen and patient predisposition [20, 21]. Parasitic leiomyomas and LPD are important conditions to highlight for understanding neovascularization of fibroids and for consideration when developing a differential diagnosis in similar clinical scenarios.

Rare reports of bladder leiomyomas have also been documented. Leiomyomas within the bladder occur in less than 1% of all urinary bladder tumors [22]. These are rare tumors of whirling smooth muscle cells with similar histology to uterine fibroids. Their etiology is uncertain but estrogen is thought to contribute to bladder leiomyoma growth [22]. Review of the literature showed majority of patients with bladder leiomyomas presented with obstructive symptoms (49%) followed by irritative voiding symptoms (38%) [23]. In our case, the fibroid likely originated from the uterus with erosion into the urinary bladder given the history of prior uterine fibroids with UAE procedure.

In review of the literature, a total of six case reports were identified pertaining to vesicouterine fistulization with fibroid involvement. Three of these cases occurred following UAE. In these reports, fibroids were described as either calcified or degenerating. It is possible that fibroids undergoing necrosis, either spontaneously or as postembolization effect, may have increased predisposition to fistula formation through postulated theories previously described. Treatment in all of these cases included hysterectomy with partial cystectomy or hysterectomy with debridement of bladder mucosa and suture repair. Our case report is the first to identify vesicouterine fistulization as a long-term consequence of UAE. Prior reports described this event occurring within one year of UAE, and in our case, fistulization occurred 12 years postembolization. Additionally, our case report is the first to describe recurrent bladder stones and hypercalciuria as presenting symptoms. Other reports described pelvic pain, dysuria, urinary retention, and vaginal leakage of urine as presenting symptoms. These presenting symptoms and prolonged timing of presentation of vesicouterine fistulization following UAE further add to the emerging body of literature of this rare event.

CONCLUSION

Erosion and fistulization of uterine fibroids to surrounding tissues are rare events. Review of the literature confirms only three prior case reports following UAE, none of which presented with calcified bladder stones. While malignancy should remain in the differential diagnosis for patients diagnosed with organ invasion, other phenomenon such as benign neovascularization and invasion should be considered specifically in those

with history of prior uterine artery embolization. In addition, patients with this history and persistent bladder symptoms should have prompt imaging to prevent delay in accurate treatment.

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Author Contributions

Laura Ramirez-Caban – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Maral Malekzadeh – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Fernando Cabrera – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Adria Suarez-Mora – Conception of the work, Design of the work, Revising the work critically for important

intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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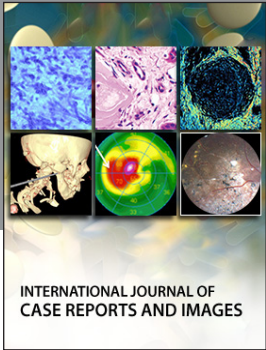
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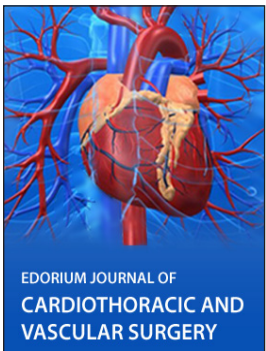
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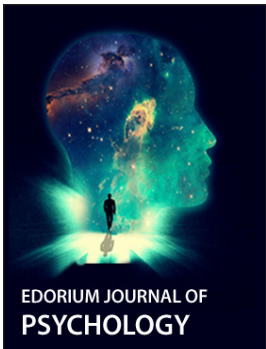
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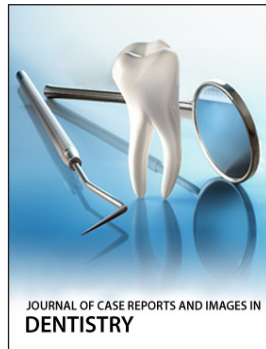
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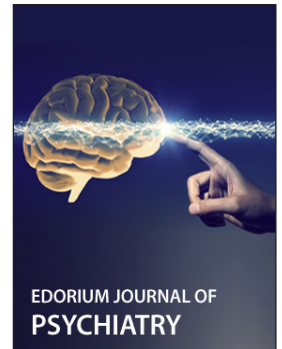
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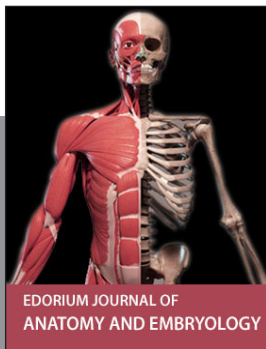
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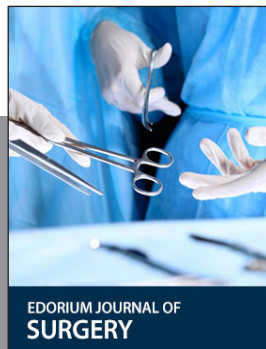
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