

Case series — Prioritizing bladder-sparing treatments in patients with urinary tract leiomyoma: A report of three cases and updated literature review

Shannon McNall¹, Terry Park¹, Nikhil Gopal², Evan Spencer³, Lakshmisree Akhila Vemulakonda⁴, Muhammad Choudhury³, Nathan Colin Wong³

¹New York Medical College, Valhalla, NY, United States; ²Urologic Oncology Branch, National Cancer Institute, Bethesda, MD, United States; ³Department of Urology, Westchester Medical Center, Valhalla, NY, United States; ⁴Department of Pathology, Westchester Medical Center, Valhalla, NY, United States

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Abstract

Urinary tract leiomyomas are rare, benign mesenchymal tumors that are traditionally treated with complete surgical excision. We present three cases of urinary tract leiomyoma — two located in the bladder and one in the ureter. Both bladder leiomyomas were treated with transurethral resection of bladder tumor (TURBT) with no evidence of recurrent disease, while the upper tract leiomyoma was managed with surveillance by patient preference, which to our knowledge, has not been previously described. The ureteral leiomyoma has remained stable at five years followup from initial diagnosis. Given the benign nature of leiomyomas and the overall low recurrence rate reported in the literature, bladder-sparing options should be emphasized when feasible, with active surveillance potentially applicable for highly select patients.

Introduction

While leiomyomas are commonly encountered by gynecologists, it is a rare entity for the practicing urologist, accounting for only 0.43% of all bladder tumors.¹ Upper tract involvement is even rarer, with only 15 cases reported in the literature since 1955.² Given the rarity of this tumor, there are no standardized guidelines for management; however, surgical extirpation, whether through transurethral resection of bladder tumor (TURBT) or partial/radical cystectomy, has been generally advocated. Herein, we describe two cases of bladder leiomyoma and one case of ureteral leiomyoma, with the latter successfully managed with observation, illustrating a novel approach to these benign tumors.

KEY MESSAGES

- Leiomyomas are benign mesenchymal tumors that rarely affect the bladder and are even less frequently seen in the upper urinary tract.
- Surgical resection is the treatment of choice, with minimally invasive options, such as transurethral resection of bladder tumor, being the preferred method.
- Conservative management, specifically surveillance, could be a valid option for a select group of asymptomatic patients with urinary tract leiomyoma.

Case reports

Case 1

A 37-year-old male presented with left lower quadrant abdominal pain. Non-contrast computerized tomography (CT) scan revealed a bladder mass, and a subsequent magnetic resonance urography (MRU) (Figure 1) demonstrated a well-circumscribed 2.4x2.3 cm T1-isodense, T2-hypodense lesion with mild enhancement in the right posterolateral bladder wall near the right ureterovesical junction without hydronephrosis. Cystoscopy demonstrated a well-circumscribed flat lesion without erythema that was subsequently resected endoscopically. Histological analysis of the TURBT specimen (Figure 2) revealed numerous spindle cells with pink cytoplasm and elongated nuclei with a low nucleus-to-cytoplasm ratio. There was no atypia, increased cellularity, necrosis, or mitotic figures present. Immunohistochemistry was positive for smooth muscle actin (SMA) and negative for proto-oncogene c-kit (CD117). These features were consistent with leiomyoma of the bladder. This patient was fol-

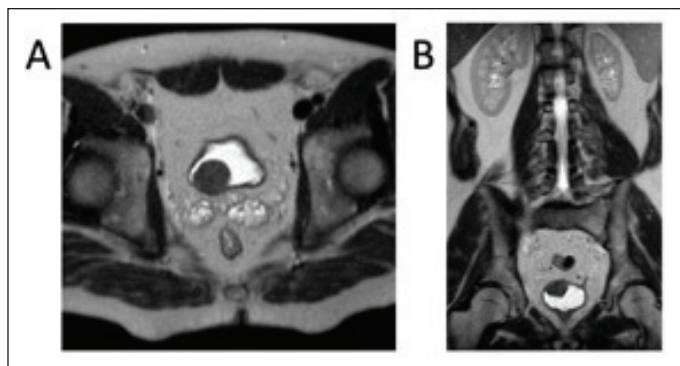


Figure 1. Case 1: Magnetic resonance urogram showing circumscribed lesion along the right posterolateral bladder wall (2.4x2.3 cm) without hydronephrosis. (A) Axial view; (B) coronal view.

lowed with cystoscopy and MRU annually without evidence of recurrence at three years.

Case 2

A 34-year-old healthy female presented with lower pelvic pain. Abdominal ultrasound was suspicious for a bladder mass and CT urogram demonstrated a mural mass seen in the right anterolateral aspect of the bladder measuring 4.3x2.8x2.7 cm. Cystoscopy demonstrated a submucosal protrusion in the area corresponding to the CT (Figure 3). The patient subsequently underwent a TURBT that demonstrated chronic cystitis and fragments of smooth muscle consistent with leiomyoma. Residual bladder mass was seen on interval imaging six weeks later, with CT-guided percutaneous biopsy confirming persistent leiomyoma. The patient underwent complete resection with a repeat TURBT with grossly negative margins. Repeat imaging at two months' followup demonstrated no evidence of disease.

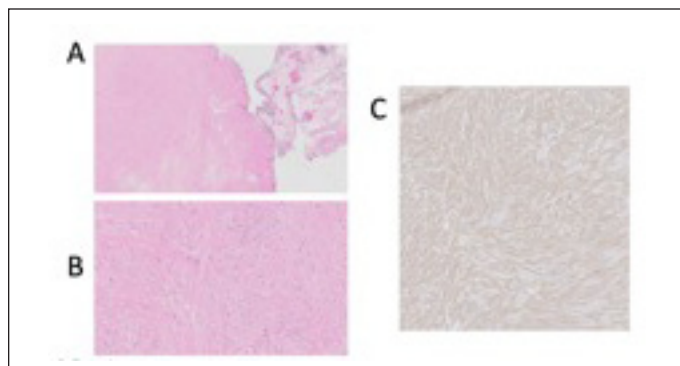


Figure 2. Case 1: (A) H&E (20x magnification) demonstrating typical intersecting smooth muscle fibers and normal bladder mucosa consistent with bladder leiomyoma. (B) H&E (100x magnification) showing proliferation of spindle cells with eosinophilic cytoplasm, elongated nuclei, and low nuclear to cytoplasmic ratio. There are no mitoses, atypical features, or necrosis present. (C) Immunohistochemistry demonstrating smooth muscle actin (SMA) diffusely positive, confirming the smooth muscle origin of the tumor.



Figure 3. Case 2: Cystoscopy showing a submucosal protrusion of the bladder wall, which is a characteristic presentation of bladder leiomyomas.

Case 3

A 63-year-old female presented with intermittent mild abdominal pain. CT demonstrated a 1.5 cm soft tissue mass located eccentrically in the wall of the left distal ureter without hydronephrosis (Figure 4). Left ureteroscopy and biopsy were performed, and pathology was consistent with leiomyoma of the ureter. Surgical excision with distal ureterectomy and reimplantation was discussed but the patient deferred definitive surgical management, as her symptoms were mild, and the pathology was benign. She elected for active surveillance, including routine imaging. At five years' followup, she has been clinically well with normal renal function, and the most recent CT scan demonstrated no radiological progression of the leiomyoma.

Discussion

Herein, we described two cases of bladder and one case of ureteral leiomyoma, reinforcing the benign clinical and histological nature of these smooth muscle neoplasms and, as the last case demonstrated, the potential role of active surveillance in select patients who are unable or unwilling to undergo surgical resection.

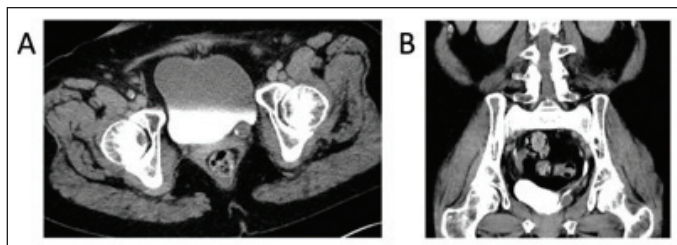


Figure 4. Case 3: Computed tomography urogram showing a soft tissue mass centered on wall of distal left ureter as evidenced by the filling defect on the excretory phase. (A) Axial view, delayed phase; (B) coronal view, delayed phase.

Table 1. Bladder leiomyoma case data (2018–2021)

Author (ref.)	Country	Age (yrs)/sex	Presenting symptoms	Tumor location	Tumor size (cm)	Treatment	Followup (months)	Recurrence
Yucel et al, 2018 ¹⁸	Turkey	44/M	Suprapubic pain	Inferior wall	2.2	TURBT	3	Unknown
He et al, 2018 ⁶	China	47/F	LUTS	Right posterior wall	4	Open partial cystectomy	8	None
Sharma et al, 2018 ¹⁹	India	65/M	LUTS	Left ureterovesical junction	3.8	TURBT	Unknown	Unknown
Li et al, 2019 ⁸	China	34/F	Incidental finding	Left lateral wall	4	Transurethral enucleation	30	None
		55/F	LUTS	Trigone	3.3	Transurethral enucleation	21	None
		54/F	Hematuria	Trigone	5.4	Transurethral enucleation	13	None
		46/F	LUTS	Left lateral wall	6.7	Transurethral enucleation	12	None
		45/F	Incidental finding	Bladder neck	2.4	Transurethral enucleation	11	None
		67/M	Incidental finding	Right lateral wall	1.8	Transurethral enucleation	2	None
Sodo et al, 2019 ²⁰	Italy	33/M	Pelvic pain and LUTS	Urachus	6	Laparoscopic partial cystectomy	9	None
Armas-Alvarez et al, 2019 ²¹	Spain	27/F	LUTS	Dome	6	Partial cystectomy	24	None
		29/F	LUTS	Posterior wall	Unknown	Partial cystectomy	48	None
		42/M	Incidental finding	Right anterosuperior wall	4	Partial cystectomy	48	None
Mitchell et al, 2019 ¹¹	USA	64/M	Hematuria	Left ureterovesical junction	6	Refractory to TURBT x 4, ultimately open radical cysto-prostatectomy with ileal conduit urinary diversion	16	Yes
Yoshioka et al, 2019 ²²	Japan	46/F	Incidental finding	Left wall	7.6	Laparoscopic enucleation of bladder tumor	4	None
Mavridis et al, 2020 ²³	Greece	28/M	Incidental finding	Left wall	3.3	Open excision of bladder tumor	Unknown	Unknown
Pramod et al, 2020 ²⁴	Indonesia	42/F	Hematuria and LUTS	Trigone	7	Open excision of bladder tumor	Unknown	Unknown
Tobias-Machado et al, 2020 ⁵	Brazil	25/M	LUTS	Posterolateral wall	3	Robot-assisted partial cystectomy	Unknown	Unknown
Zachariou et al, 2020 ²⁵	Greece	52/M	Hematuria and LUTS	Trigone	4.1	TURBT	12	None
Moawad et al, 2020 ²⁶	United Arab Emirates	33/F	Abdominal pain and Hematuria	Bladder neck	7.6	Open enucleation of bladder tumor	Unknown	Unknown
Rey Valzacchi et al, 2020 ²⁷	Argentina	45/F	LUTS	Trigone	4.0	Laparoscopic excision of bladder tumor	60	None
Delara et al, 2021 ²⁸	USA	33/F	Abdominal pain, LUTS, and menorrhagia	Left posterior wall	2	TURBT	Unknown	Unknown
Al Solumany et al, 2021 ¹⁴	Saudi Arabia	50/F	Hematuria	Left anterolateral wall	3	Open partial cystectomy	18	None

LUTS: lower urinary tract symptoms; TURBT: transurethral resection of the bladder tumor.

Table 1 (cont'd). Bladder leiomyoma case data (2018–2021)

Author (ref.)	Country	Age (yrs)/sex	Presenting symptoms	Tumor location	Tumor size (cm)	Treatment	Followup (months)	Recurrence
McNall et al, 2021	USA	37/M	Left lower quadrant abdominal pain	Right ureterovesical junction	2.4	TURBT	36	None
		34/F	Pelvic pain	Right anterolateral and inferior wall	4.3	TURBT	2	None
Summary		Mean age (yrs)	Female, n (%)	Male, n (%)	Median size (cm)	Treatment, n (%)	Median followup (months)	Recurrence, n (%)
2018–2021 n=25		44	15 (60%)	10 (40%)	4	TURBT and transurethral enucleation=12 (48%), laparoscopic=3 (12%), robotic=1 (4%), open=6 (24%), unknown=3 (12%)	13	1 (4%)
2012–2017 n=21 ⁶		48	14 (67%)	7 (33%)	4.3	TURBT and TUR=9 (43%), laparoscopic=3 (14%), robotic=1 (5%), open=5 (24%), vaginal resection=2 (10%), unknown=1 (5%)	Unknown	0 (0%)

LUTS: lower urinary tract symptoms; TURBT: transurethral resection of the bladder tumor.

Surgical resection has traditionally been the mainstay of management for urinary tract leiomyomas. While bladder leiomyomas have traditionally been treated with open partial cystectomy,³ contemporary series have advocated minimally invasive approaches, including endoscopic management with complete TURBT or enucleation, as well as laparoscopic or robotic partial cystectomy.^{4,5}

We have summarized all reported cases of urinary tract leiomyomas in the literature (n=25 patients) from 2018–2021 in Table 1. In our literature review, we found that over the past three years, 60% of reported bladder leiomyomas have occurred in females, with a mean age of 44 years. Of the 25 cases reported, six were found incidentally and only six patients (24%) were treated with an open surgical approach; the rest were managed with minimally invasive surgery. This extends on the literature review conducted by He et al, which summarized reported bladder leiomyomas from 2012–2017.⁶ In all cases reported since 2012, over 65% of patients were treated using minimally invasive approaches,⁶ with more than two-thirds of those patients managed transurethrally. Similar to present day, prior to 2012 bladder leiomyomas were primarily treated with TURBT or open resection depending on size and anatomical location of the tumor;⁷ however, since 2012, treatment options have expanded to include methods such as vaginal resection and transurethral enucleation.^{6,8}

Endoscopic management maximizes bladder preservation and should be the recommended surgical approach if feasible for bladder leiomyomas. Li et al described transurethral enucleation of these benign tumors in which, after initial circumferential incision around the tumor margin, the resectoscope loop is used to bluntly separate the tumor from the surrounding urothelium prior to segmental tumor resection. This technique was performed successfully in six patients, without evidence of recurrence at median followup of 14 months.⁸ Given the benign nature of these neoplasms, minimizing resection of normal bladder tissue should be a priority, making enucleation an appealing approach.

As with their lower tract counterparts, all previously reported cases of ureteral leiomyoma were treated surgically with either segmental ureterectomy or nephroureterectomy.^{9,10} Unlike the bladder, complete endoscopic management of tumors in the ureter is limited given the smaller working channel of the ureter and the relatively thin muscle layer, the latter of which increases risk for perforation. These challenges likely account for the absence of endoscopic approaches to ureteral leiomyoma in the literature.

While the majority of uterine leiomyomas are managed conservatively, the same principle has not been applied to urological manifestations, even when patients are relatively asymptomatic. Nevertheless, we have reported, to our knowledge, the first case of conservative management for

ureteral leiomyoma. The absence of tumor growth in this patient is in line with the underlying benign pathology of leiomyoma and the overall absence of reported tumor recurrence. There has been only one published case of urinary leiomyoma recurrence reported since 2012.¹¹ Provided that a sufficient tissue sample has been obtained for pathological analysis, highly selective patients diagnosed with leiomyomas who are asymptomatic or minimally symptomatic and who are poor candidates or refuse surgery may be safely monitored with active surveillance.

Serial imaging should be performed, with magnetic resonance imaging (MRI) potentially preferred due to its superior resolution of soft tissue masses and absence of radiation. With regard to timing, we may draw upon the surveillance regimen for low-risk bladder and upper tract urothelial cancer by recommending repeat cross-sectional imaging (CT/MRI urogram) at three months after initial evaluation, with repeat imaging obtained at 12 months and annually thereafter for at least five years.^{12,13} In the event of tumor growth, patients should be offered surgical resection, with endoscopic treatment being first-line when feasible. Following tumor removal, surveillance imaging for 2–3 years may be considered to exclude recurrence (the only recent case of leiomyoma recurrence occurred 12 months after initial TURBT).

In cases where partial cystectomy was elected over lesser aggressive approaches, rationale cited include cases of large tumors, tumors with extravesical growth, or tumors with unfavorable position.¹⁴ In the singular case of recurrence mentioned above, a cystoprostatectomy was elected following four unsuccessful TURBTs, as well as invasion into the prostatic urethra. The authors believed that the unique tumor location made complete resection difficult, ultimately resulting in multiple recurrences and the need for more aggressive surgical management. While local invasion is not common with bladder leiomyoma, it can occur, as in the case that required cystoprostatectomy, and may be an indication for a more invasive approach.¹¹ Overall, we would recommend a majority of cases be managed via TURBT or with surveillance; however, if a symptomatic tumor proves to be too large for TUR, is in a location preventing a successful TURBT, or shows local invasion, one may have to consider a more aggressive treatment option.

In addition to active surveillance, future directions for management of urinary tract leiomyomas may include medical therapy. Randomized controlled trials have demonstrated significant decrease of uterine leiomyoma size with use of selective progesterone receptor modulators and gonadotropin-releasing hormone agonists.^{15,16} Given that a majority of bladder leiomyomas also occur in females,¹⁷ hormonal treatment may represent another potential urinary tract-preserving treatment option.

Conclusions

We present three cases of urinary leiomyoma, one of ureteral and two of bladder origin successfully managed with observation and TURBT, respectively. Surveillance of urinary tract leiomyoma has not previously been described and may be a viable option — once the diagnosis is confirmed pathologically — in select patients due to the generally benign nature of this entity. In cases dictating surgical management, endoscopic resection should be prioritized whenever possible.

Competing interests: The authors do not report any competing personal or financial interests related to this work.

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Correspondence: Ms. Shannon McNall, New York Medical College, Valhalla, NY, United States; smcnall@student.nymc.edu