

GIANT SIZED EXTRAVESICAL BLADDER LEIOMYOMA MIMICKING UTERINE CERVICAL LEIOMYOMA: A CASE REPORT WITH 8 YEARS' POSTOPERATIVE FOLLOW-UP AND A REVIEW OF THE LITERATURE

UTERİN SERVİKAL LEİOMYOMU TAKLİT EDEN DEV EKSTRAVEZİKAL MESANE LEİOMYOMU: 8 YILLIK POSTOPERATİF TAKİBİ VE LİTERATÜRÜN GÖZDEN GEÇİRİLMESİ

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ABSTRACT

The incidence of bladder leiomyoma is estimated at only 0.43% because most bladder tumors arise from the urothelium. Moreover, 30% of them are the extra-vesical type. The symptoms include urinary tract obstruction, bladder irritation, and dysuria, with the possibility of dyspareunia or hematuria depending on the size and location of the tumor. Bladder leiomyomas cannot be diagnosed solely by combining symptoms and imaging techniques. We present a rare case of giant sized extravescical-type bladder leiomyoma with a diameter of 15.5x14.5x14 cm, which was misdiagnosed as a uterine cervical leiomyoma. It caused unilateral ureteral distension without related symptoms. During the 8-year postoperative follow-up, the patient had two healthy full-term pregnancies and no recurrence was observed. We reviewed the literature of the most recent five years and discussed the characteristics of 22 bladder leiomyoma cases. Unlike the literature, the present case is unique due to the giant size of the bladder leiomyoma, its ureteral complications, and long follow-up period. Bladder leiomyomas are rare bladder tumors which should be borne in mind during the differential diagnosis of a pelvic mass. Surgical excision is the most useful procedure for diagnosis and treatment.

Keywords: Bladder leiomyoma, bladder tumor, cervical leiomyoma, cervical tumor, differential diagnosis

ÖZET

Mesane leiomyomunun insidansı sadece %0,43 olarak tahmin edilmektedir; çünkü mesane tümörlerinin çoğu üroepitelyumdan kaynaklanmaktadır. Bunun yanı sıra, mesane leiomyomlarının sadece %30'u ekstra-vezikal tiptedir. Üriner sistem obstrüksiyonu, mesane irritasyonu, dizüri, disparoni veya hematüri gibi semptomlar kitlenin boyut ve yerine göre değişebilir. Yalnızca semptom ve görüntüleme tekniklerine dayanarak mesane leiomyomu tanısı konulamamaktadır. Preoperatif uterin servikal leiomyom ön tanısı düşünülen, 15,5x14,5x14 cm çapında tek taraflı üreteral distansiyona yol açan, nadir görülen bir ekstrevezikal tip mesane leiomyomu olgusunu sunuyoruz. Sekiz yıllık postoperatif takip boyunca, hastanın iki sağlıklı term gebeliği mevcuttu ve nüks izlenmedi. Son beş yılın literatürü gözden geçirilerek 22 mesane leiomyomunun özellikleri sunuldu. Literatürden farklı olarak, mevcut olgu mesane leiomyomunun büyüklüğü, üreteral komplikasyonları ve uzun takip süresi ile benzersizdir. Mesane leiomyomları, pelvik kitlenin ayırıcı tanısında göz önünde tutulması gereken, nadir görülen mesane tümörleridir. Tanı ve tedavi için en faydalı prosedür cerrahi eksizyondur.

Anahtar Kelimeler: Mesane leiomyomu, mesane tümörü, servikal leiomyom, servikal tümör, ayırıcı tanı

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INTRODUCTION

Bladder leiomyoma is a rarely seen mesenchymal neoplasm of the urinary bladder, and it is only estimated to occur in 0.43% of adults. Incidence of this condition is higher in female patients in their fifth decade (1). The etiology is uncertain; effects of hormones, inflammation, localized infection, and chromosomal abnormalities may play a role. The symptoms depend on the location and size of the tumor, which is categorized as endovesical, intramural or extravescical with frequencies of 63%, 7% and 30%, respectively (2). Voiding symptoms such as hematuria, dysuria and/or obstructive symptoms are generally seen in endovesical-type tumors, whereas bladder neck obstruction due to compression of the ureter may lead to upper urinary tract obstruction and hydronephrosis in extravescical-type tumors (3). Like uterine leiomyoma, radiologic differentiation of bladder leiomyoma from leiomyosarcoma is not clearly defined.

We present a rare case of a giant extra-vesical type bladder leiomyoma, leading to unilateral ureteral distension, which was misdiagnosed as a cervical leiomyoma. We also report the 8-year follow-up and review the related literature on the last five years between 2016 and 2021.

CASE REPORT

A 23-year-old nulliparous woman was referred due to a pelvic mass with symptoms of dysuria and chronic pelvic pain. Her urologic and gynecologic histories were unremarkable. On gynecologic examination, the mass occupied the entire pelvis till 2 cm above the umbilicus, bulging the right vaginal wall and obliterating the vaginal lumen; the uterine cervix was unremarkable. Laboratory tests revealed normal tumor markers and urinalysis. Magnetic resonance imaging (MRI) showed a 15x10x10-cm mass of low signal intensity on T2-weighted images, which appeared adjacent to the right cervical wall, compressing the right ureter and the bladder (Figure 1A, 1B).

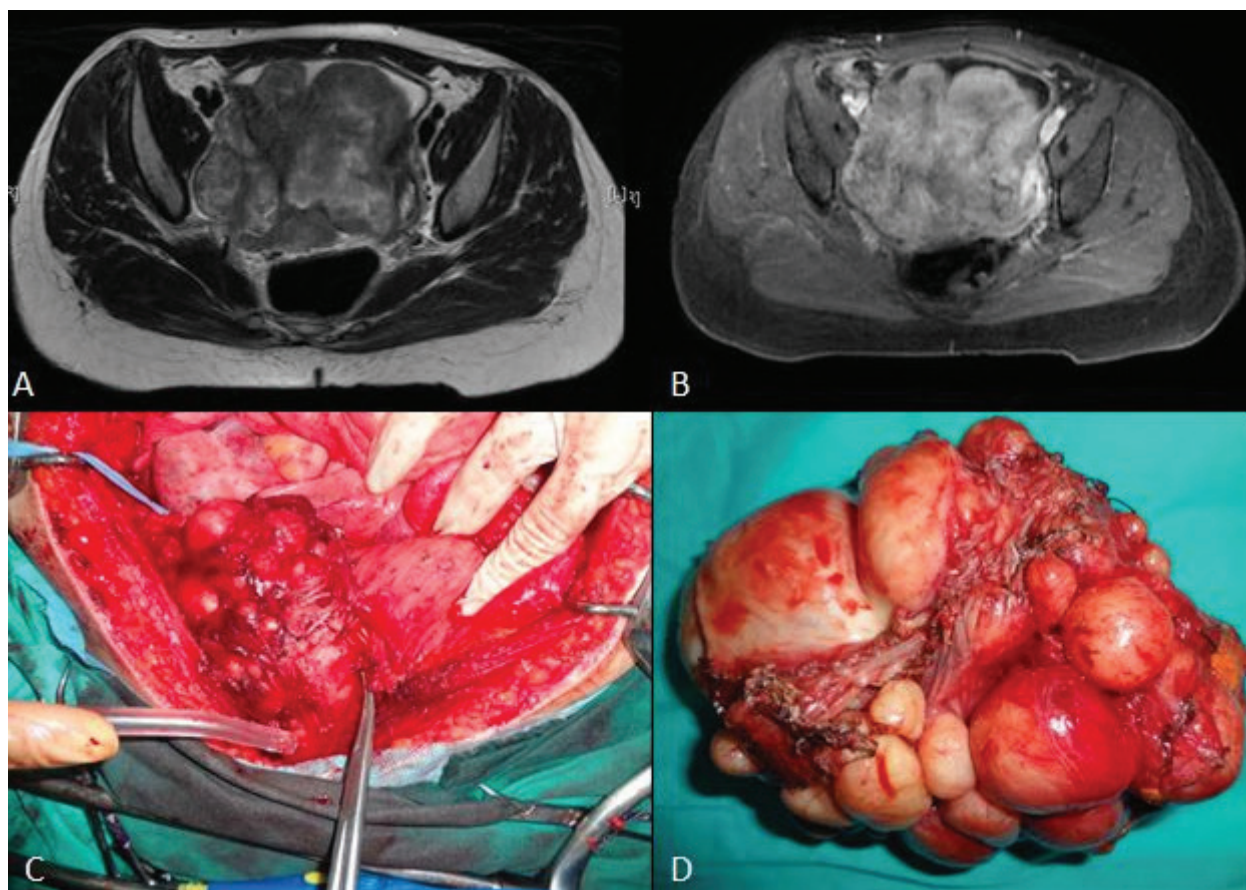


Figure 1: (A). Fat-suppressed T1-weighted sequence after intravenous gadolinium administration demonstrating a giant mass with heterogeneous contrast agent fixation that occupies the entire pelvis. (B). T2-weighted sequence showing a solid lesion with partially high signal areas and no clear border separation visible in the adjacent bladder. (C). The bladder leiomyoma reached the bladder mucosa and compressed the uterine cervix from the antero-lateral part. (D). The bladder leiomyoma was approximately 10x15 cm in size.

The signal intensity was similar to leiomyoma and leiomyosarcoma of the uterus on MRI. Based on the clinical and radiologic findings, the mass was estimated to be leiomyoma or leiomyosarcoma of the cervix.

Surgery was performed with a low midline incision; it was observed that the mass originated from the right side of the bladder wall and extended as far as the right parametrium (Figure 1C). Perioperatively, it was considered as 15.5x14.5x14 cm extravesical bladder leiomyoma that reached the bladder mucosa (Figure 1D). The distal part of the right ureter was passing through the mass. Pararectal, paravesical and Retzius spaces were developed. The distended right midureter was dissected and a full cut incision was performed to the bladder wall for total excision. The right distal ureter was excised and ureteroneocystostomy was performed. The bladder wall was repaired in two layers, the first consisting of mucosa using 3-0 vicryl, and the second consisting of detrusor and serosa using 2-0 vicryl sutures and a double-J catheter which was inserted through the lumen. The histopathologic investigation reported a bladder leiomyoma without cytologic atypia or necrosis. It stained positively for smooth muscle actin and vimentin, and estrogen and

progesterone receptors were positive. The postoperative period was uneventful and the patient was discharged on the 6th day after the operation without complications.

During follow-up, spontaneous pregnancy occurred two years later. During the 8-year postoperative follow-up, the patient had two healthy full-term pregnancies and delivered via an uncomplicated cesarean section. The indication for the first cesarean section was fetal macrosomia. During follow up no recurrence was observed.

DISCUSSION

Bladder leiomyoma was first described by Virchow in 1931 and there are about 250 cases in the literature (4). As recent studies have shown promise in establishing best practices for evaluation and management, we reviewed the related studies on the last five years between 2016 and 2021. To review the features of bladder leiomyoma such as symptoms, diagnosis tools, treatment and prognosis, a literature search of PubMed and the Web of Science was conducted, which included 22 cases of bladder leiomyomas in 17 publications as shown in Table 1 (1, 3-18). The symptoms vary according to the location of the

Table 1: A review of case reports reported as bladder leiomyomas in histopathologic examinations between 2016 and 2021

Study	Age/sex	Symptom/history	Size	Location	Diagnosis tool	Initial diagnosis before surgery	Treatment	Follow-up
Xin et al. 2016	44/F	dyspareunia	7.3x6.1x5.8 cm	Trigone of urinary bladder	USG, CT	Soft tissue mass	Open surgical excision	8 months without recurrence
Haddad et al. 2016	37/M	Severe dysuria, urgency, frequency, fever and chills	5.5x4.3 cm	Right anterolateral wall of urinary bladder	USG, CT	Febrile urinary tract infection	TUR-B	1 year without recurrence
Mendes et al. 2017	22/F	Pelvic discomfort, dysuria, and dyspareunia	1.6x1.1x0.6 cm	Anterior bladder wall	USG, Cystoscopy and biopsy	Cystitis	TUR-B	N/A
Jamshaid et al. 2017	38/F	lower urinary tract symptoms (straining, intermittency, sense of incomplete bladder emptying and increased frequency of urination)	4.4x4.5 cm	Posterior inferior surface of the bladder near the bladder neck	CT, Cystoscopy	Polypoidal mass	TUR-B	After 6 months, urinary retention occurred. She underwent an open local excision as second operation. After 1 year follow up, no recurrence was observed.
Dodia et al. 2017	35/F	Hematuria, dysuria	3.3x4.3 cm	Right posterolateral bladder wall	USG, MRI, Cystoscopy and biopsy	N/A	Open transvesical enucleation	N/A
He et al. 2018	47/F	Dysuria	4 cm	Right posterior bladder wall	Pelvic CT	N/A	Open partial cystectomy	8 months without recurrence

Table 1: Continued

Sharma et al. 2018	65/M	Lower urinary tract symptoms	3.8×3.3×2.4 cm	Left posterolateral wall and left vesico-ureteral junction of the bladder	USG, Contrast-enhanced CT, FDG PET/CT	Favor of low-grade urinary bladder neoplasm	TUR-B	N/A
Yucel et al. 2018	44/M	Suprapubic pain and history of neurofibromatosis	1.8×2.2 cm	Inferior bladder wall	USG	N/A	TUR-B	After 3 months, grade 3 hydronephrosis on left side and 6×3.5 cm mesenchymal tumor from the left anterolateral wall of the bladder.
Pramod et al. 2020	42/F	Painless hematuria and irritative symptoms	7×6.5×4 cm	Posteroinferior wall of the bladder	USG, Doppler, CT, Cystoscopy	Soft tissue mass	Open transvesical excision of the bladder mass	N/A
Sodo et al. 2019	33/M	Pelvic pain, urgency, dysuria	6×5.8×4.7 cm	Upper side of the bladder located along the urachus	CT, MRI	Bladder tumor without distant metastasis	Laparoscopic partial cystectomy and urachus removal	9 months without recurrence
Mitchell et al. 2019	64 /M	Recurrence of bladder leiomyoma after TUR-B, History of testicular teratoma	6 cm	Postero-lateral wall of the bladder causing obstruction of the left ureterovesical junction. Severe hydronephrosis and hydro-ureter	CT	Metastasis of testicular teratoma	Open radical cystoprostatectomy	6 months without recurrence after radical surgery
Fridman et al. 2018	51/F	Heavy vaginal bleeding, pelvic pressure, bladder incontinence, dyspareunia	5.5 cm	Inferior of bladder & superior anterior wall of urethra	MRI	Paraurethral mass	Open paraurethral resection	N/A
Mavridis et al. 2020	28/M	Asymptomatic, History of solitary fibrous tumor of pleura	3.7 cm	Left bladder wall	CT	Metastasis of pleural tumor	Open retropubic resection	N/A
Tobias-Machado et al. 2020	25/M	Urinary frequency, urgency	3×2 cm	Postero-lateral bladder wall	USG, MRI	Bladder leiomyoma	Robot-assisted transvesical partial cystectomy & placement of right urethral stent	N/A
Li et al. 2019	50.2 (n=6) F (n=5) M (n=1)	Asymptomatic (n=3) Irritative (n=2) Hematuria (n=1)	Mean: 3.9 cm	Endo-vesical (n=3) Intramural (n=3)	USG, CT, Cystoscopy	Bladder tumor	Transurethral enucleation	Mean 14.8 months without recurrence
Zachoval et al. 2018	32/F	Asymptomatic pregnant woman	5 cm	Anterior wall of the bladder	USG, MRI	Bladder leiomyoma	Open resection	1 year without recurrence
Delara et al. 2021	33/F	Abdominal pain, urinary urgency, frequency, heavy menses	2.0×1.3×1.7 cm	The left posterior bladder wall adjacent to the left ureterovesical junction	MRI, cystourethroscopy	Bladder mass	TUR-B	N/A

TUR-B: Transurethral resection of the bladder; USG: Ultrasound; CT: Computed Tomography; MRI: Magnetic Resonance Imaging

tumor, and may even be asymptomatic (Table 1). The data analysis in Table 2 shows that computed tomography (CT) is mostly used for diagnosis and transurethral resection of the bladder (TUR-B) is the common treatment method for endovesical-type bladder leiomyomas. Additionally, the mean size is 4.16 (range, 1.8-6.7) cm (n=16), and the mean follow up period is 11.5 (range, 2-30) months (n=11) (Table 2). Here, we report a rare, giant, extravesical-type bladder leiomyoma. Our case is unique with its giant size (15.5x14.5x14 cm), its extension to the parametrium, which created confusion in diagnosis by mimicking a cervical tumor, and its ureter complications. Although recurrence had been reported in two cases previously, the present case showed no recurrence during cesarean sections.

Table 2: Analysis of case reports reported as bladder leiomyomas between 2016 and 2021

Mean age	42.81
Gender (n, %)	F: 14, 63.6% M:8, 36.4%
Mean size (cm)	4.04 cm (1.1 to 6.7 cm)
Diagnostic tool	CT (n=15, 68.2%), USG (n=15, 68.2%); MRI (n=6, 30%); Cystoscopy (n=11, 5%); Doppler USG (n=1, 4.6 %); PET-CT (n=1, 4.6%)
Treatment	Open abdominal resection (n=7, 31.8%); TUR-B (n=13, 59.1%); Robotic surgery (n=1, 4.6%); Laparoscopic surgery (n=1, 4.6%)
Mean follow up period (months)	11.5 months (2 to 30 months) (n=14) N/A (n=8) In three of them, recurrence was reported.

Differential diagnosis between cervical pathologies and extravesical bladder tumor is difficult, even with an examination and radiologic investigation. Also, benign conditions such as lipomas, hamartomas or nephrogenic adenoma and malignant conditions such as urothelial carcinoma, adenocarcinoma or metastatic cancer of the urinary tract should be considered. Ultrasound is the first tool for suspected bladder leiomyoma due to the characteristic image of the mass protruding into the bladder cavity, especially in endovesical-type tumors (3). A smooth, homogenous, and hypoechoic mass in the bladder wall is observed in an ultrasound. Blood flow of the mass can be evaluated using Doppler flow to assess the possibility of malignancy (6). CT is a good option to show hypoattenuating mass, to detect the location and relation of mass with surrounding structures, and to identify cystic and solid components. Contrast-enhanced CT images improve the differential diagnosis (5). MRI is superior to CT in distinguishing the mesenchymal component of leiomyomas and preservation of the muscle layer (19). On MRI, blad-

der leiomyoma is similar to uterine leiomyoma imaging features, which visualize at intermediate signal intensity on T1-weighted images and low signal on T2-weighted images. Multiphase dynamic MRI studies with gadolinium can demonstrate the enhancement pattern of the solid component. Cystoscopy can be used to take a biopsy and successfully diagnose endovesical bladder leiomyoma and other types that have intravesical protrusion (3). Ultrasound and cystoscopy are sufficient tools for diagnosing intravesical bladder leiomyoma, whereas MRI is generally used for extravesical-type tumors. Pathologic evaluation is the golden standard for diagnosis (3).

Since we could not completely exclude malignancy histopathologically, we chose laparotomic surgical excision to prevent possible spread and seeding of tumors. For endovesical-type bladder leiomyomas, transurethral resection is one of the primary options (3). Minimally invasive surgery, such as laparoscopic or robotic myomectomy, are suitable options for extravesical and intramural-type tumors (13). However, surgeons must keep in mind morcellator restrictions and patients should be informed about the risk of intraperitoneal myometrial cell spread (20). Injury of the ureter and bladder, and severe hemorrhage of pelvic organs can be seen as complications.

Histopathologic diagnosis is needed to rule out malignancy. Microscopically, bladder leiomyoma consists of smooth muscle cells with eosinophilic cytoplasm and cigar-shaped nuclei. The feature that distinguishes uterus and bladder leiomyoma from leiomyosarcoma is the absence of mitotic activity, cytologic atypia or necrosis (21). Immunohistochemically, bladder leiomyoma stains diffusely with smooth muscle actin, desmin, and vimentin, but not with cytokeratin and S100 protein (2).

CONCLUSION

We present a unique case of giant sized extravesical bladder leiomyoma with ureteral complications, and a long follow-up period. Despite the best clinical examination and radiologic imaging, it may be difficult to distinguish extravesical bladder leiomyoma. Although bladder leiomyomas are rarely seen tumors, surgeons should keep them in mind during the differential diagnosis of a pelvic mass. Appropriate surgery should be performed for the diagnosis and treatment.

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