

Urethral leiomyoma: A rare neoplasm

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Abstract

Extrauterine leiomyoma is a rare benign tumor, originating from smooth muscle cells, most often localized in the uterus. Extrauterine localization of leiomyoma is extremely rare. The presence of a neoplasm near the urethra always poses a challenge for differential diagnosis. Variety of pathologies sharing similar clinical findings requires further diagnostic testing and doctor alertness. We present the clinical case of urethral leiomyoma in a female patient aged 42 years complaining of frequent urination and vulval mass. The patient underwent neoplasm removal with further histologic examination revealing urethral leiomyoma. The article features the diagnosis and treatment of this rare pathology.

Keywords: Female urethra, neoplasm, urethral leiomyoma

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Received: 09.06.2020, **Accepted:** 05.01.2021, **Published:** 04.03.2021

INTRODUCTION

Leiomyoma is a benign neoplasm originating from smooth muscle cells, most often localized in the genitals, especially the uterus. Urethral leiomyoma is an extremely rare tumor type. Presence of a neoplasm near the urethra always poses a challenge for differential diagnosis. Abundance of pathologies from a urethral polyp to squamous carcinoma sharing similar clinical findings requires further diagnostic testing and doctor alertness.

CASE REPORT

The female patient received an outpatient urologist consultation about lower urinary tract symptoms-frequent urination and urgency. Overactive bladder was diagnosed, although examination on the gynecological chair revealed a urethral neoplasm. For further investigation, the patient was referred to a hospital with the primary diagnosis of

paraurethral cyst. The patient did not complain of dysuria, straining when urinating or feeling of incomplete emptying of the bladder. Detailed examination revealed a paraurethral mass at 7–10 o'clock, sized at ~ 3 cm-oval, dense, nondisplaceable, painless, with even clear boundaries [Figure 1].


The examination revealed no gynecological diseases. The patient had two uncomplicated vaginal deliveries in the past, and 2 years earlier she underwent hysteroscopic endometrial polyp resection.

Transperineal ultrasound (US) examination of the neoplasm with a linear 7 Hz transducer was conducted. The present study revealed a uniform hypoechoic region with a clear capsule all along urethral tissues without the signs of fluid or calcified inclusions [Figure 2]. No flow was detected in the region with the Doppler scanning. Those findings were evaluated as a urethral tumor, and incisional

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How to cite this article: Popov SV, Orlov IN, Chernysheva DY, Grin' EA. Urethral leiomyoma: A rare neoplasm. *Urol Ann* 2021;13:194-7.

Access this article online	
Quick Response Code:	Website: www.urologyannals.com
	DOI: 10.4103/UA.UA_90_20

biopsy was planned. The patient has a history of a high-energy traffic accident and underwent an intramedullary osteosynthesis of the right thigh, which was a relative contraindication to MRI.

Under combined anesthesia (30 ml of 0.5% novocaine + intravenous infusion) after insertion of a Foley 16F catheter, the front vaginal wall was dissected in the layers for 3 cm, visualizing a wall of the mass. When attempting to isolate the neoplasm's left edge, bleeding started, requiring complete isolation of the neoplasm for achieving adequate hemostasis. The tissue defect was sutured with 5.0 dissolvable running suture.

Macroscopic examination revealed a rose-red firm mass with even surface sized 28 mm × 20 mm × 25 mm. Dissection under the dense capsule visualized a firm yellowish-gray solid neoplasm with no necrosis or hemorrhages [Figure 3].

Microscopically, it presented the bundles of smooth muscle cells with fusiform nuclei. No foci of mitotic

division or atypia were detected. The intact neoplasm capsule presented cell bundles [Figures 4 and 5]. These characteristics indicate urethral leiomyoma diagnosis.

The postoperative period was uneventful. The Foley catheter was removed 10 days after the surgery. For about 6 weeks, the patient suffered from mild stress urinary incontinence which subsequently stopped.

DISCUSSION

Urethral leiomyoma is an extremely rare tumor, with <45 cases described in literature.^[1,2] Urethral leiomyomas are usually small neoplasms ~1–2 cm in diameter growing mainly in the distal part of the urethra and from its posterior surface. About 25% of cases of urethral leiomyoma are asymptomatic, otherwise patients complained of urethral mass, hematuria, acute urinary retention, recurrent lower urinary tract infections, and vaginal bleeding.^[1,3,4] Migliari *et al.* noted that neoplasms located at 12



Figure 1: Photo of the paraurethral mass and its relationship with urethral meatus

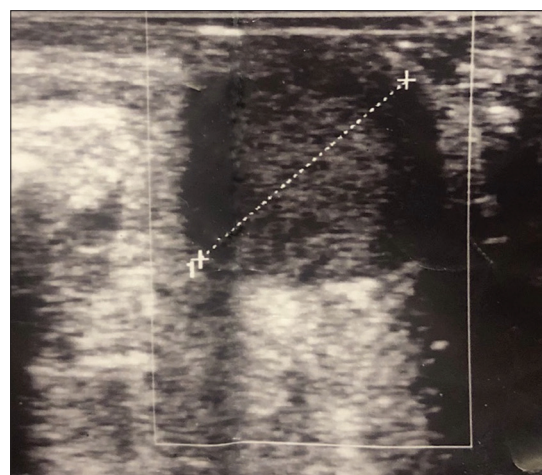


Figure 2: Sonographic picture of the paraurethral mass – 22 mm × 24 mm × 18 mm, no flow in Doppler



Figure 3: Macroscopic appearance following the surgical removal

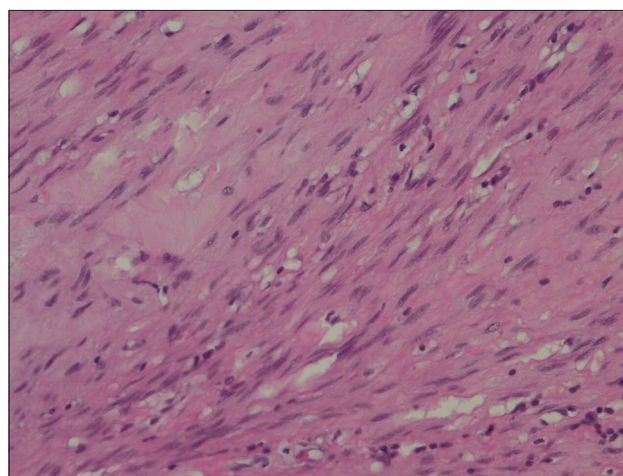


Figure 4: Microscopic appearance of the neoplasm with hematoxylin and eosin staining, ×100

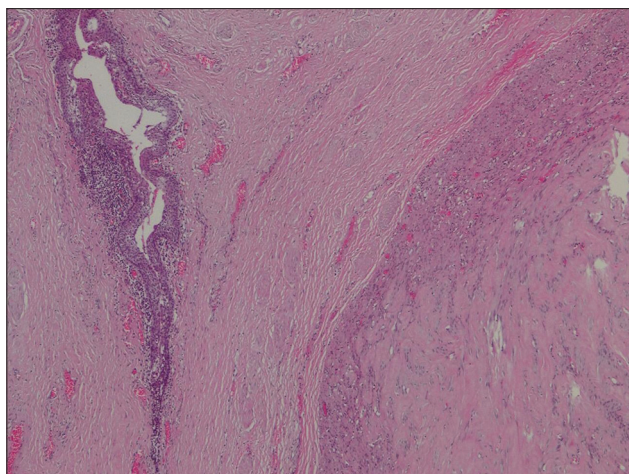


Figure 5: Microscopic appearance of the paraurethral tissues with hematoxylin and eosin staining, x40

or 6 o'clock lead to obstructive urination symptoms, whereas laterally dislocated leiomyomas are more likely to manifest with irritative complaints.^[5]

Urethral leiomyoma often found in women usually aged 30–40, with an average age of 41 years.^[1] According to some reports, tumor size may decrease with the onset of menopause or after childbirth, thus suggesting hormone-dependent nature of the disease.^[6]

There is still no agreement or single algorithm in urethral leiomyoma diagnosis. Most often, the diagnosis can be made with examination. Sometimes, the use of visualization techniques (US, computed tomography, and magnetic resonance imaging [MRI]) is required. Upon the results of these examinations, the doctor can assess tumor size and depth, its vascularization degree, and presence of a capsule for adequate planning of surgical intervention. Differential diagnosis includes urethral mucosa prolapse, urethral caruncula, female urethral gland cyst, urethral diverticula, ectopic ureterocele, and urethral carcinoma.^[7]

Özel *et al.* and Ballard suggest differentiating between urethral leiomyoma and paraurethral located leiomyoma. Urethral leiomyoma features fixed position, protrusion through the urethral meatus, and need for urethroplasty after neoplasm removal.^[8] Migliari *et al.* insist that urethral leiomyoma is a misleading term. According to the authors' team, leiomyoma develops not from urethral muscle layer tissues, but from the smooth muscle cell cluster around the urethra, thus suggesting a more correct term of paraurethral leiomyoma.^[5]

We have not seen a single described case of urethral leiomyoma malignization. However, there is evidence of possible local recurrence.^[9]

Tumor excision is the most common way to treat patients with urethral leiomyoma. Vaginal (U-shaped) access along the front vaginal wall is predominant, although transurethral dissection can also be used. Importantly, after urethral leiomyoma ectomy with any technique, stress urinary incontinence, urethrovaginal fistula, or urethral stenosis can be observed.^[5] Urinary incontinence is successfully corrected using mid-urethral slings. However, according to some authors, such interventions should be conducted no earlier than 6 months after original operation and after MRI confirming absence of neoplastic process recurrence.^[1]

CONCLUSION

Urethral leiomyoma is a rare benign tumor with difficult diagnosis by a primary care physician or the one not specializing in urogynecology. Urethral leiomyoma can be successfully removed with good oncological and functional outcomes.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgment

We would like to thank Dr. Peremyshlenko A.S for performing the morphology studies and Dr. Neradovskiy V.A for the technical help.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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