Paraurethral leiomyoma in a 52 year-old woman: Clinical and diagnostic features and surgical treatment

Nicolò Fiorello,1 Andrea Di Benedetto,2 Andrea Mogorovich,2 Daniele Summonti,2 Sandro Benvenuti,2 Carlo Alberto Sepich1
1UOC Urologia Ospedale San Luca, Lucca; 2UOC Urologia Ospedale Versilia, Camaiano, Italy

Abstract
Leiomyomas are benign tumors of smooth muscle origin occurring throughout the genitourinary system. Sometimes paraurethral leiomyoma can cause a bulking effect on the bladder without gross involvement of the urethra. In our work we describe the case of a woman with a vulvar mass found after self-examination. She was investigated pelvic Magnetic Resonance Imaging (MRI) which showed a well encapsulated solid mass, about 24mm (maximum diameter) located at paraurethral level, on right anterolateral side. It showed uptake of contrast, with a compressive-dislocative effect on the urethra, without involvement of the sphincters. Surgery was performed with excision “en-block” of the mass. The procedure was well tolerated under spinal anesthesia and the patient was discharged 24h later, after removing the bladder catheter. No signs of haematuria or symptoms of incontinence were reported in the following weeks. Pathology of the excised specimen revealed complete resection of a benign, submucous leiomyoma. Although leiomyomas are very common in organs such as the uterus, the presentation of a urethral leiomyoma, as found in the present case, is very rare. The description of these lesions is however important to create a scientific path that can clarify the cause and the evolution. It can also represent an evolution for imaging through MRI, improving its framework and distinguishing it from malignant tumors that require another surgical treatment.

Introduction
Benign tumors of the urethra are rare and generally only described in small case reports. Leiomyoma, hemangioma, and fibroepithelial polyp are most frequently reported. Female urethral prolapse and urethral caruncles are benign lesions that can mimic tumors.1 Leiomyomas are benign tumors of smooth muscle origin occurring throughout the genitourinary system.2 While leiomyomas in the uterus are frequently seen, urethral and paraurethral leiomyomas are extremely rare with a hand full of cases in the literature.1,3 Sometimes paraurethral leiomyoma can cause a bulking effect on the bladder without gross involvement of the urethra.4

Case Report
A 52-year-old woman was referred to our observation due to the presence of a vulvar mass found after self-examination. She had no Lower Urinary Tract Symptoms (LUTS), or dyspareunia and metrorrhagia. Pelvic examination showed no visible masses on the vulva and external urethral meatus, but a small palpable mass, in accordance with the patient’s self-examination A transvaginal pelvic Ultrasound (US) was performed, which revealed as chestnut-shaped mass, under the bladder, probably at the right paraurethral level, well circumscribed and vascularized. She denied pelvic pain or pressure. A pelvic Magnetic Resonance Imaging (MRI) was performed, which showed a well encapsulated solid mass, about 24mm (maximum diameter), located at paraurethral level, on right anterolateral side. This mass showed uptake of contrast, with a compressive-dislocative effect on the urethra, without involvement of the sphincters (Figure 1). There were no signs of lymphadenopathy.

The patient was subsequently referred to the definitive resection. The surgery was performed in a lithotomy position, after the insertion of a bladder catheter for safety and as a reference for the urethra. A right paraurethral incision was performed to reach the mass, careful to injure the urethra, labia majora or the underlying vaginal wall. Then an en-block excision was performed (Figure 2).

The procedure was well tolerated under spinal anesthesia and the patient was discharged 24 hours later, after removing the bladder catheter. No signs of haematuria nor symptoms of incontinence were reported in the following weeks. Pathology of the excised specimen revealed complete resection of a benign, submucous leiomyoma (Figure 3).

Discussion
From among all the leiomyoma cases reported to date, the most common site of occurrence has been the genital tract (95%), while the remainder are scattered over various sites, including the skin (230 cases), gastrointestinal tract (67 cases), and bladder (5 cases).1 Although leiomyomas are very common in organs such as the uterus, the presentation of a urethral leiomyoma, as found in the present case, is very rare. Categorized as deep tissue leiomyomas, urethral leiomyomas are much larger than their superficial counterparts, and usually display a greater spectrum of histological changes; therefore, it is important to clearly distinguish them from leiomyosarcomas, which are statistically more common in deep soft tissue.5 Differential diagnosis typically comprises urethral diverticulum (84%), leiomyoma (7%), vaginal wall inclusion cyst (6%), Skene’s gland cyst or abscess, urethral prolapse, urethral caruncle, and primary neoplasm (squamous cell carcinoma, urothelial carcinoma, and adenocarcinoma). Presentation may resemble

Correspondence: Nicolò Fiorello, UOC Urologia Ospedale San Luca, Via Guglielmo Lippi Francesconi 556 - 55100 Lucca (LU), Lucca, Italy.
E-mail: fiorellourologia@gmail.com

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other periurethral masses, and, as a result, a careful history and physical examination is crucial for the proper diagnosis.\(^8\)

These masses may develop along any part of the urethra; however, the majority of paraurethral leiomyomas present in the proximal segment. As a result, diagnosis via physical examination is typically standard protocol and is based on palpation of the vaginal mass. In addition, confirmation of diagnosis is encouraged and obtained through imaging, such as ultrasound or MRI.\(^9\)

The surgical treatment is interesting because, although it was a small mass and benign in nature, it is a rare lesion for which there is no standardized technique. There is no risk of metastases, but local recurrence can occur in the case of incomplete resection.

Management usually involves local resection, which can be achieved both vaginally or abdominally depending on size, location, and comfort level of operating surgeon with the aim of minimizing recurrence and relieving symptoms. Due to the intimate relationships with the urethra or the bladder, care must be exercised so that injury to nearby organs and complications such as stress urinary incontinence and urethral stenosis are curtailed.\(^10\) Consequently, being in an area very near to the sphincters, the probability of causing injury and subsequent incontinence is not irrelevant. If the patient develops stress urinary incontinence, the correction can be performed by a tension-free vaginal tape-obturator (TVT-O) placement.\(^11\)

**Conclusions**

Urethral leiomyomas are very rare diseases whose cause is ill unknown. However, it does not seem to originate from intravaginal smooth-muscle component.\(^12\) The description of these lesions is however important to create a scientific path that can clarify cause and the evolution. It can also represent an evolution for imaging through MRI, improving its framework and distinguishing it from malignant tumors that require another surgical strategy.

Finally, it is an important challenge for the urologist or gynecologist, because the surgical performance requires anatomical and functional knowledge being an anatomical area of multidisciplinary interest.

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