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Case report

Primary urethral leiomyoma in a female patient: A case report and review of the literature



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KEYWORDS

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Abstract

Female urethral leiomyoma is a rare clinical entity. Cystourethroscopy, magnetic resonance imaging (MRI) and trucut biopsy are helpful tools for establishing a prompt clinical diagnosis. Malignant transformation has not been reported, and recurrence is rare. We present a case of female urethral leiomyoma presenting as a urethral mass which could be completely excised surgically with a good outcome.

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Introduction

Primary urethral neoplasm in women is a rare condition with less than 45 cases reported in the literature worldwide [1–14]. Primary urethral leiomyoma is an even rarer clinical entity. Most patients present with a mass protruding from the urethra, associated with obstructive voiding in some rare cases. Prompt diagnosis, especially exclusion of a malignant tumor, and proper treatment are necessary to prevent complications. Herein we report on a new case of female urethral leiomyoma. Our case was different in that the mass was located in the anterior wall of the distal urethra, which is not the common site of presentation of leiomyoma in females.

Case report

A 31-year-old female patient presented with swelling in the region of the external urethral orifice, gradually increasing over the past 6 months. She had no history of hematuria, dysuria or voiding problems. Physical examination revealed a firm, mobile, non-tender mass sized 2.5 cm × 2 cm, located just proximal to the urethral meatus anteriorly. The urethral meatus was stretched, located at the bottom of the swelling and not easily visible. The patient's results on routine investigations were within normal limits. Ultrasonography revealed a normal urinary system. Uroflowmetry was normal. Clinical examination and cystourethroscopy showed a mass arising from the anterior wall of the urethra, extending 2.5 cm proximally from the meatus with mild congestion of the overlying mucosa. The remaining bladder and urethra were normal (Figs. 1 and 2). A trucut biopsy was taken during preoperative cystoscopy, and histopathology showed a benign leiomyoma.

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Figure 1 Anterior urethral mass with cystoscope sheath placed inferiorly.

The mass was completely excised under spinal anesthesia. Histopathology revealed an encapsulated tumor containing spindle to plump oval nuclei-bearing cells with eosinophilic cytoplasm in collagenous tissue, suggesting a benign leiomyoma (Fig. 3). Immunohistochemistry for smooth muscle actin (SMA) was performed using the avidin-biotin-peroxidase complex method (Fig. 4). On follow-up, the patient had mild stress urinary incontinence without any evidence of tumor recurrence.

Discussion

Urethral tumors are rare and may arise from the lining or glandular epithelium, the smooth muscle fibers or the striated muscle fibers. Polyps and papillomas are the most common and leiomyomas the least common clinical entities [1]. Primary urethral leiomyoma is seen more frequently in females than in males, and it usually develops at the posterior wall of the proximal urethra [2,3]. Our case was different in that the mass was located at the anterior wall of the distal urethra. Nevertheless, the patient did not complain of voiding problems. The most common symptom found in 50% of the cases is swelling near the periurethral region [4–6]. Other symptoms include hematuria, dysuria, repeated urinary tract infection and

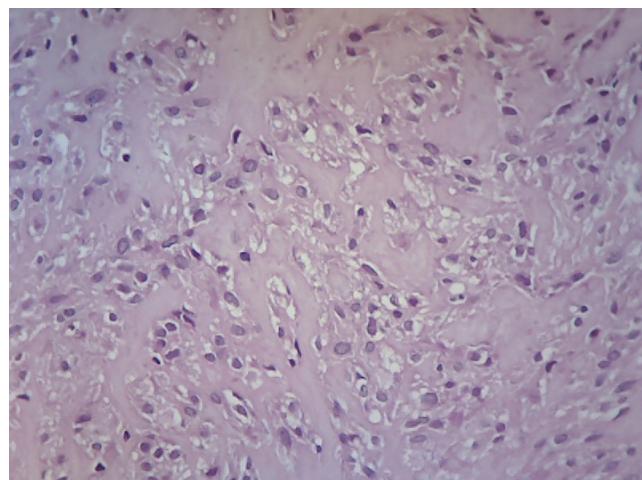


Figure 3 Histopathology showing benign urethral leiomyoma (H&E, 100 \times)

dyspareunia [3,7]. Obstructive voiding symptoms are rare due to the paraurethral rather than periurethral position of the neoplasm [8]. However, in extremely rare cases, a patient may present with acute or chronic renal failure [9], while other patients may be completely asymptomatic [10]. It has been noted that the tumor enlarges during pregnancy and shrinks after delivery, suggesting an underlying hormonal etiology [1,7]. In some cases, estrogen and progesterone receptors have also been identified in this kind of tumor.

Differential diagnosis of this tumor mainly includes urethral diverticulum, urethral mucosal prolapse, urethral caruncle, Bartholin gland cyst, Gartner duct cyst, urethral carcinoma and vaginal wall cysts [3,7,11]. Proper clinical examination, cystourethroscopy and radiographic imaging studies such as intravenous pyelography, voiding cystourethrography, urethrography, vaginal ultrasonography and pelvic magnetic resonance imaging (MRI) may be helpful in establishing a definitive diagnosis [5,7]. However, only histopathological examination can distinguish a leiomyoma from a malignant tumor. Immunohistochemistry using monoclonal anti-smooth muscle antibodies confirms the smooth muscle origin of this tumor.



Figure 2 Closed bladder neck with mass located anteriorly as seen on urethroscopy.

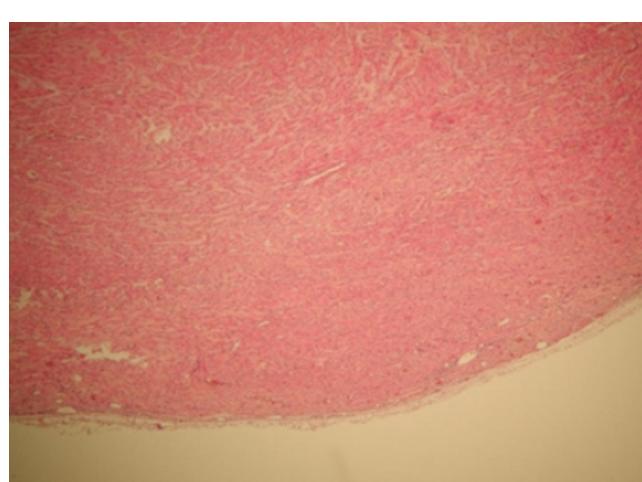


Figure 4 Immunohistochemical staining for smooth muscle actin (SMA) (avidin biotin-peroxidase complex method, (40 \times)).

Simple surgical excision is the treatment of choice [9]. Other treatment options include transurethral resection [12] and gonadotropin-releasing hormone (GnRH) therapy [13], but these have not been well established as yet. The prognosis is excellent as, so far, malignant transformation has not been reported and tumor recurrence is rare [14].

Conflict of interest

The authors have no conflict of interest to declare.

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