

Leiomyoma of the Female Urethra with Intermittent Urinary Retention: A Case Report

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Abstract

Urethral leiomyomas are rare benign mesenchymal tumors that arise from the smooth muscle of the urethra. Most reported cases in the literature have occurred in women of child bearing age. These tumors present with a variety of symptoms. Excision is curative however occasional cases of relapse have been described. Here we describe a case of a female teenager who presented with intermittent urinary retention and final histopathological diagnosis of leiomyoma of the distal urethra following excision. She had an uneventful recovery and has remained completely symptom-free. To the best of our knowledge, this is first case of urethral leiomyoma in Pakistan that has presented with a history of recurrent urinary retention.

Keywords: Benign, Leiomyoma, Smooth muscle, Urethra, Urinary retention.

Introduction

Urethral leiomyomas are rare benign mesenchymal tumors that originate from the smooth muscle of the urethra.¹⁻⁵ This tumor was first described in this location by Buttner in 1984. Although mostly reported in women of childbearing age, however no age or gender is exempt.⁶ These tumors present with a variety of symptoms including recurrent urinary tract infections (UTIs), urinary retention and dysuria. A protruding mass from the external urethral

meatus is frequently reported when the tumor is located in the distal part of the urethra.⁷⁻¹⁰ Simple excision is curative. Occasional cases of recurrence reflect incomplete removal. No malignant transformation has been reported.^{2,5,7,10} Herein, we describe a case of distal urethral leiomyoma in a teenage girl who presented with recurrent urinary retention. The diagnosis was confirmed on histopathology and immunohistochemistry of the excised polyp. To the

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Case Report

An 18-year-old unmarried girl presented with a history of recurrent urinary retention and tissue protrusion from the urethra to the Urology Outpatient Department (OPD) of Sindh Institute of Urology and Transplantation (SIUT), Karachi, Pakistan. According to the patient, she developed intermittent acute urinary retention and burning micturition six months prior, for which she took homeopathic treatment with no resolution of symptoms. Later, she went to a local hospital where suprapubic cystostomy was performed. In between, she had also been treated by antibiotics for recurrent UTIs.

At SIUT, an abdominal ultrasound that included the kidneys, ureter and bladder (KUB) was performed, the results of which were unremarkable. She underwent an examination

under anesthesia (EUA) and cystoscopy. On examination, she had a fleshy polypoid mass that measured 2.5 cm in size which was pedunculated, pinkish white in color, round, firm in consistency and located at the 5 o'clock position. The mass was protruding from inside the distal urethra. Cystoscopy was performed, which was normal. A simple excision of the lesion was done. The urethra could be calibrated upto 18 Fr. She underwent a suprapubic cystostomy and was subsequently catheterized for 48 hours. She voided well following removal of the catheter after two days. Postoperative recovery was uneventful. She was symptom-free at the time of her last follow up.

Histopathological examination of the excised mass revealed a solid, ovoid, encapsulated mass which, on slicing, showed a homogenous, gray white, whirling surface. Microscopy revealed a benign cellular lesion composed of interlacing fascicles of smooth muscle cells (Figure 1A). The cells possessed elongated, cigar shaped nuclei and scanty eosinophilic cytoplasm. Cell out lines

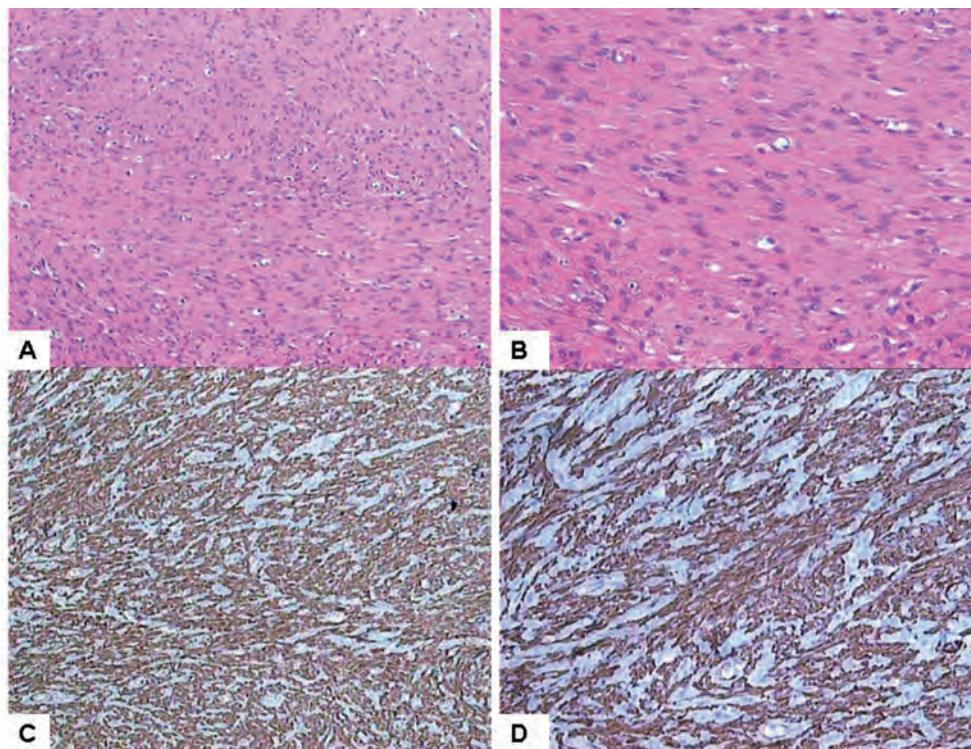


Figure 1. A. Hematoxylin and eosin (H&E) staining shows interlacing bundles of monomorphic spindle cells with eosinophilic cytoplasm. (magnification: 100 \times). B. Similar area at medium power shows spindle cells with eosinophilic cytoplasm and bland cigar shaped nuclei (H&E, magnification: 200 \times). C. Immunohistochemical (IHC) staining shows diffuse positivity of smooth muscle actin in the tumor cells (magnification: 100 \times). D. IHC staining shows diffuse positivity of myosin in the tumor cells (magnification: 100 \times).

were indistinct. No nuclear atypia or atypical mitoses were noted (Figure 1B). There was no necrosis. Immunohistochemical (IHC) markers were performed which revealed diffuse positivity of caldesmon, smooth muscle actin, myosin and vimentin, while cytokeratin and S-100 were negative (Figures 1 C, D). The morphological and IHC features were consistent with a diagnosis of leiomyoma.

When histopathology revealed leiomyoma, ultrasound pelvis was performed to rule out uterine leiomyoma which was negative. Therefore, this case represented an isolated primary leiomyoma of the distal urethra.

Discussion

Leiomyoma of the urethra is a rare benign mesenchymal tumor that affects women more often than men.¹⁻⁵ Numerous individual case reports and small series of this lesion have been reported in the English literature.⁶⁻¹⁰ The tumor can occur at any age but is most common in the third and fourth decades of life; the mean age in most cases is around 41 years.^{2,3,7-10} Our patient was from the adolescent age group. The tumor typically affects the bulbous part of urethra (80% of cases), but the distal segment can also be affected, as in the current case. The mean diameter reported in literature is 3.7 cm, but larger tumors have also been reported.^{1,4,6,10}

The tumor can present as an asymptomatic mass or be a part of an acute urinary retention syndrome and cause symptoms such as dyspareunia, hematuria, recurrent UTIs, urinary retention and irritative symptoms.⁶⁻¹⁰ Different studies have found different predominant presenting features.⁶⁻¹⁰

Leiomyomas can originate from any part of the urinary tract including the kidney, which is the most common site. Other sites of origin include the urinary bladder, prostate, scrotum, penis, spermatic cord, epididymis, and seminal vesicles. The site of the lesion determines the clinical features and therapeutic approach.

Diagnosis is based on clinical history, physical examination and imaging techniques. In any case,

the final diagnosis is made by histopathological examination. The clinical differential diagnosis of the urethral leiomyoma is broad and includes urethral caruncle, extravesical bladder leiomyoma, plasmacytoma and lymphoma. The tumor must be differentiated from its malignant counterpart, the leiomyosarcoma, which shows marked pleomorphism, increased cellularity and frequent mitoses. In females, the origin of the tumor from the female genital tract, especially the uterus must be excluded.

It is difficult to ascertain the definite cause of the tumors in an individual patient. The enlargement of the tumors during pregnancy and their frequent concurrence with uterine leiomyomas has led to speculations about the hormonal etiology, especially estrogens, for these tumors. However, the tumors also occur in males and in postmenopausal women, suggesting a multifactorial etiology.

The most appropriate treatment for this condition is local excision or transurethral resection.⁶⁻¹⁰ There is usually no recurrence or malignant transformation after treatment. However, occasional recurrences reported reflect incomplete removal of the tumor.

Our case was located in the distal urethra, an uncommon site for leiomyomas in females. This patient also presented with repeated urinary retention and recurrent UTIs which were not well documented in the literature. She was unusually young compared with the majority of reported cases. To the best of our knowledge, this was the first case of urethral leiomyoma in Pakistan that presented with a history of recurrent urinary retention.

In conclusion, urethral leiomyomas are rare benign tumors that are clinically difficult to diagnose. Histopathology and immunohistochemistry provide the definitive diagnosis. A high index of suspicion is needed to accurately diagnose and appropriately manage this tumor.

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