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Leiomyoma of the Ureter: A Conservative Approach

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ABSTRACT

We present a case of bladder leiomyoma and concurrent left lower ureteric leiomyoma in a sixty-two year old Bahraini male without any other comorbid conditions. He had infrequent hematuria for the last two years; it was associated with burning and discomfort during urination, frequency and urgency.

Urine culture did not reveal any growth. Contrast CT abdomen revealed a mass in the bladder and lower ureteral narrowing with hydronephrosis. Histopathology of both lesions revealed leiomyoma. Limited resection with subsequent stenting saved patient from having major surgery. The patient was followed up after removal of left ureteric stent. He had MAG3 (Mercapto Acetyl Triglycine) scan, which was satisfactory with no restriction to drainage.

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INTRODUCTION

Leiomyoma of the genitourinary tract is an uncommon tumor. Leiomyoma is a benign tumor developed from smooth muscle cells[1]. The common site for leiomyoma in genitourinary tract is the uterus. Ureteric leiomyoma comprises less than 3% of ureteral tumors[2]. A total of ten cases of primary ureteric leiomyoma have been reported in the literature till 2006[3,4].

Ureteric leiomyoma could present with hematuria, or flank pain. Lower ureteric lesion could cause irritative bladder symptoms or urinary tract infection[5]. Bladder leiomyoma is rare; it constitutes 0.43% of all bladder neoplasms[6]. It arises from the mesenchymal tissue[6]. Bladder leiomyoma is not as rare as ureteric leiomyoma. In the English literature, around 170 cases of bladder leiomyoma have been reported till 2010[6].

In the past, the treatment of ureteric tumors used to be nephroureterectomy, but the advances in imaging and endourology has helped to preserve the kidney and the ureter[7]. Concurrent bladder leiomyoma along with lower ureteric leiomyoma is rare entity for which I was unable to find a single citation in the literature.

The aim of this report is to highlight the rarity of concomitant bladder and lower ureteric leiomyoma, which was managed conservatively.

THE CASE

A 62-year-old male presented with history of burning micturition, sense of incomplete emptying of bladder, hematuria, urgency, with urge incontinence and nocturia of 4-5 times per night. Past history was significant for surgically treated bladder stones.

Physical examination revealed soft non-tender abdomen; external genitalia were normal. Digital rectal examination revealed a benign feeling prostate around 70-80 cubic centimeter (cc) in volume. Urinalysis and urine culture revealed microscopic hematuria of 6-10 RBC/hpf and WBC of 45-50 WBC/hpf, no growth on culture was seen. Complete blood count and renal function tests were unremarkable.

Abdominal CT with contrast revealed thickened urinary bladder trigonal area and left ureterovesical junction (UV junction) and left hydroureteronephrosis, see figure 1.

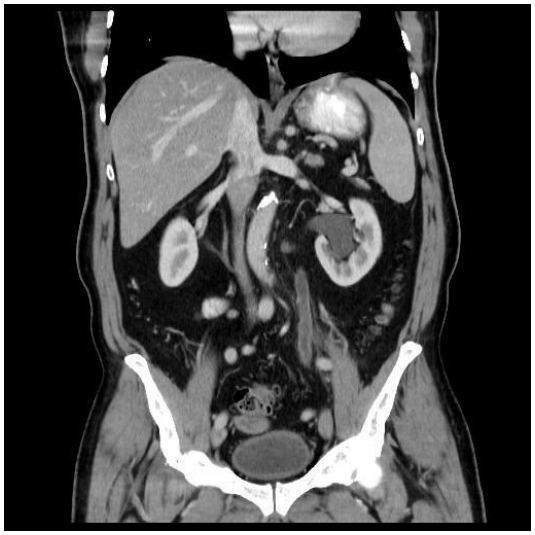

Figure 1: A Contrast CT Showing Left Dilated Renal Pelvis and Ureter

Cystoscopy and left ureteroscopy was performed which revealed an obstructing prostate with narrow left ureteral opening and a bulge noted in left lateral wall of bladder, next to the ureterovesical junction and trigonal area involving the left ureteral orifice. No obvious mucosal abnormality was seen in the bladder. Lower ureteral mucosal thickening and irregularity was seen on ureteroscopy. Excision biopsies were taken from the abnormal bladder and the lower ureter. Ureteral washing from the lower and upper ureter was performed separately. Open-ended ureteral catheter was left in the left ureter and incorporated into 3-way catheter.

Histopathological examination revealed solid nodules of tissue composed of closely compacted vascular and smooth muscle elements, see figure 2.

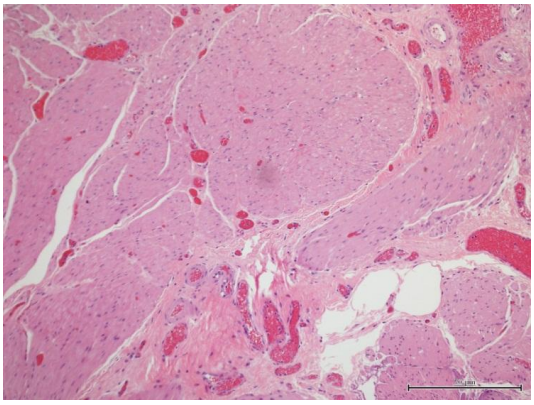

Figure 2: Low Power Showing Vascular and Smooth Muscle Elements (H&E)

The muscle fibers were mature and showed spindle nuclei. There was no necrosis, mitoses or pleomorphism. The vascular component consisted of large number and closely compacted blood vessels, some of which were dilated and the walls revealed little muscular thickening. The appearances suggested a cavernous type of angioleiomyoma. Immunohistochemical stains for smooth muscle actin and desmin (smooth muscle markers) were positive, see figure 3. The marker for blood vessels known as Cd31 was also positive.

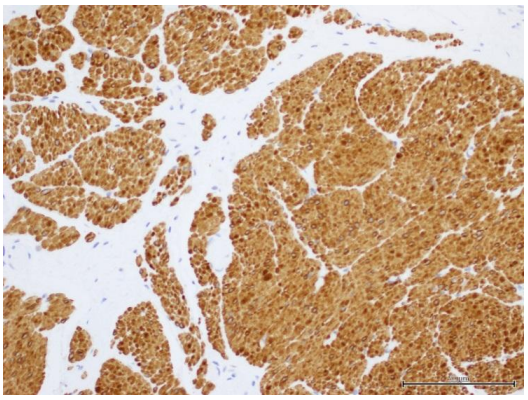

Figure 3: Immunohistochemical Stains for Smooth Muscle Actin and Desmin

After confirming the benign nature of the disease, the patient was retaken to the operation theatre a few days later and a left Ureteric JJ stent was placed to avoid ureteral orifice stenosis. The stent was removed after 6 weeks and subsequent Mercapto Acetyl Triglycine (MAG3) renogram revealed no hold up of radioisotope. The patient had no hematuria, frequency and urgency. He was asked for follow up for his bladder outflow symptoms.

DISCUSSION

Non-epithelial benign ureteral tumor of mesodermal origin is a rare disease and leiomyoma is even rarer[8]. The presentation is delayed because of the slow growing nature of tumor or it is discovered incidentally while investigating a non-related symptoms[9]. The common symptoms associated with bladder leiomyoma are irritation (38%), obstruction (31%), hematuria (25%), abdominal pain or mass (15%) and twenty-eight percent of patients were asymptomatic[4].

The etiology of leiomyoma is unclear. Many theories have been proposed including inflammation, trauma, urolithiasis and multiple endocrine neoplasia type 1[7,9]. The age

incidence of this condition could be encountered usually at the age of 30-40 years. There is no obvious predisposition to left or right side, specific site in ureter or patient's sex[9]. Two reports have suggested that leiomyomas are more common in females; the age incidence is in the 4th decade[6,7]. The use of pelvic ultrasound in females has improved the detection rate of leiomyomas[10]. Females' sex hormones were implicated in the causation of these benign tumors. Hormonal activity in the 4th decade is suspected to be the reason for the formation of leiomyoma[11,12]. It is difficult to differentiate lower ureteral tumors from the bladder leiomyomas. The only feature which might suggest that it is ureteral growth is the abnormality and narrowing of the ureteral lumen which was encountered in our case. Nephroureterectomy is usually done for the lack or difficulty in diagnosis[9,13,14].

Concurrent bladder leiomyoma and uterine leiomyoma was reported in literature, but no ureteric tumor with bladder leiomyoma is reported[15]. Uterine findings were incidental on imaging while investigating the lower urinary tract symptoms. In our case, the lower ureteric lumen was narrowed and bladder wall was thickened as seen on the CT.

This case is unique; both kidney and ureter were preserved. There was no compromise of renal function or ureteral stenosis as shown by MAG3 renogram. Histopathology report was of great value in modifying the plan and insertion of ureteral stent; this step, in the authors' view, was a decisive factor in the maintenance of ureteral lumen and renal function. The authors also believe that leaving an open-ended ureteral catheter helped in identifying the ureteral orifice when the ureteral stent was inserted. Concurrent bladder and lower ureteric leiomyoma is a rare entity. To the author's knowledge, it has not been reported in the published literature.

CONCLUSION

A case of rare entity of concomitant bladder and lower ureteric leiomyoma was presented. The symptoms were hematuria, irritation, urgency and frequency of micturition. No bacterial growth on urine culture was seen. Staged conservative operative approach, with transurethral resection of the growth avoided major surgery. Follow up with MAG3 revealed a satisfactory drainage from the kidney.

Bladder tumors close to ureteric opening and ureteral tumors with unusual cystoscopic or ureteroscopic appearance need to be evaluated fully before definitive treatment is instituted. Histological diagnosis of the lesion should be at hand before embarking on any extensive mutilating surgery.

It is advisable to perform ureteroscopy in cases of bladder leiomyoma if contrast CT abdomen shows abnormality in the lower ureter.

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