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Leiomyoma Of Urinary Bladder a Rare Entity: Series Of 3 Cases And Review Of Literature

Nitesh Kumar*  Karthik M  Samyuktha K  Sunil Palve  Tushar Agrawal

MS (General Surgery), MCH trainee Urology, Osmania Medical College, India

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ABSTRACT

Introduction: Leiomyoma of urinary bladder is a rare entity and comprises 0.43% of all bladder tumors. Here we present our series of 3 cases and the related review of literature. Methods: 3 cases of bladder leiomyoma presented over a period of 16 months in Osmania Medical College and Hospital. Detailed history was taken, physical examination, routine blood, urine and radiological investigations were done. Patients were treated by Tresurethral Resection (TUR) of the mass and histopathological analysis with Immunohistochemistry was done for all cases. Results: All 3 cases were females with mean age of 31.6 years. All cases were endovesical type, mass near bladder neck and presented with obstructive symptoms. Radiological investigations in all patients suggested a possibility of leiomyoma and all cases had reduced urinary flow rates. Histopathology confirmed the diagnosis in all cases. No recurrence was found at one year of follow up. Conclusion: Leiomyoma of urinary bladder is a rare disorder which frequently occurs in middle aged females. Symptoms are related to its size and location, diagnosis is confirmed by histopathological analysis. Treatment is by surgery (mainly TUR). Prognosis of the disease is excellent.

1. Introduction

Benign mesenchymal tumours of urinary bladder are rare and represent about 1-5% of all neoplasms [1]. Leiomyoma is the most common benign neoplasm among them accounting for 35% of mesenchymal bladder tumours and 0.43% of all bladder tumours [2]. They are frequently encountered in middle aged females [3]. Endovesical variety with obstructive symptoms is the most common presentation [3, 4]. Around 300 cases have been reported till date and among them very few have reported more than 2 cases [2, 3]. We present our series of three cases and the related review of literature.

2. Case Report

We report three cases which presented to urology department of Osmania Medical College and Hospital during October 2017 to January 2019. Written and informed consent was taken from all three cases for treatment, use of the data and pictures for the purpose of publication.

Case 1:
27 year female presented with intermittent stream of urine, straining during micturition and dysuria since 3 months...
which increased with passage of time. There was no other urinary or general complaint. Patient had no other co-morbidities and the physical examination was unremarkable. All laboratory values were in normal limit and routine urine examination showed pus cells (4-5) and plenty of Red Blood Cells (RBC) and the urine culture was sterile.

Uroflowmetry recorded max urinary flow rate ($Q_{\text{max}}$) of 5.6 ml/sec and average urinary flow rate ($Q_{\text{avg}}$) of 2.5 ml/sec. Ultrasonography (USG) of abdomen showed $2 \times 2 \times 3$ cm homogeneous mass at the bladder neck with mild increase in vascularity. Magnetic Resonance Imaging (MRI) abdomen and pelvis reported $29 \times 25 \times 24$ mm$^3$ polypoidal lesion arising from the urethra and base of the bladder, protruding into the bladder lumen, iso-to-hyper intense in T1 and hypointense in T2 sequence (figure 1). Cystoscopy showed a pedunculated mass hanging down from the bladder neck at 12 o’clock position with prominent vessels on its surface (figure 2). Mild bladder trabeculations were present.

**Case 2:**
30 year female presented with obstructive lower urinary tract symptoms for 6 months and acute retention of urine since 2 days. There was no history of hematuria, fever, increased frequency of urine or pain abdomen. Patient had no other co-morbidities and the physical examination was unremarkable. All laboratory values were in normal limit and routine urine examination showed pus cells (4-5), no RBCs and the urine culture was sterile. She was catheterised on presentation and catheter was removed after 2 days after which she was able to void but with difficulty.

Uroflowmetry recorded $Q_{\text{max}}$ of 12 ml/sec and $Q_{\text{avg}}$ of 5 ml/sec. USG abdomen showed $2.2 \times 1.5 \times 1.6$ cm$^3$ homogeneous mass at the bladder neck with mild increase in vascularity (figure 3). Contrast Enhanced Computed Tomography (CECT) abdomen and pelvis reported $27 \times 16 \times 17$ mm$^3$ mass arising from the base of the bladder near the bladder neck, protruding into the bladder lumen. The bladder wall was thickened and she had right sided mild hydronephrosis (figure 4). Cystoscopy showed a broad base mass near the bladder neck extending from 10 o’clock to 2 o’clock position and abutting the posterior wall of the bladder neck with prominent vessels on its surface (figure 5). Mild bladder trabeculations were present.
Case 3:
35 year female presented with difficulty in voiding since 2 months and acute urinary retention since 1 day. She had no other urinary or general symptoms, no co-morbidities and an unremarkable physical examination. She was catheterised on presentation and later given a catheter free trial but was not able to void. All other laboratory parameters were normal. Urine routine showed few pus cells, plenty of RBCs and urine culture was positive for Escherichia coli >10^5 colony forming units.

USG showed 3.5×2.5×3 cm homogeneous mass at the bladder neck with mild increase in vascularity (figure 6). MRI abdomen and pelvis reported 37×26×31 mm^3 mass arising from the base of the bladder near the bladder neck, protruding into the bladder lumen (figure 7). Cystoscopy showed a broad base mass near the bladder neck extending from 9 o’clock to 3 o’clock position, abutting the posterior wall of the bladder neck and completely closing it, with prominent vessels on its surface (figure 8). Moderate bladder trabeculations were present.

Figure 4. CECT showing mass at the bladder neck (A) and thickened bladder wall (B)

Figure 5. Cystoscopic picture showing mass which is broad base arising from the anterior wall of bladder near the bladder neck

Figure 6. USG showing large homogeneous mass at the bladder neck

Figure 7. T2 images of MRI showing mass near the bladder neck
None of the three cases had present incidental finding or past history of uterine leiomyoma. All patients underwent Trans-Urethral Resection (TUR) with 26 French Resectoscope using bipolar loop working element with normal saline irrigation. Complete clearance of mass was done in all cases. Tumour in the third case bled profusely during resection but was controlled. None of the cases required any blood transfusion. Foley's catheter was removed on day 3 in all the cases and they were able to void. Follow up was done on day 14 with USG abdomen and Uroflowmetry, all cases showed marked improvement in flow rates reaching near normal values.

Histopathology (HPE) was done in all three cases. Grossly they were grey white to grey brown and soft. Microscopic examination showed interlacing fascicles & bundles of uniform spindle cells with elongated nuclei, minimal or absent cytological atypia (figure 9). The epithelium over the tumour was maintained and consisted of mostly transitional cell and in few places stratified squamous epithelium. This might indicate possibility of urethral origin of these tumours which have grown into the bladder lumen. Immunohistochemistry was done for desmin (positive) (figure 10A), Smooth Muscle antigen (SMA) (strong positive) (figure 10B), Ki67 (low) (figure 10C), Anaplastic Lymphoma Kinase (ALK) (negative), P63 (negative), CD34 (negative), Vimentin (negative). These confirmed the tumour as benign leiomyoma.

All cases were followed up for a minimum duration of 1 year. 3 monthly USG abdomen, Uroflowmetry and Cystoscopy was done. There were no signs of recurrence.

3. Discussion

Leiomyomas can occur at any site in the genitourinary system and most common site of occurrence is at the Renal capsule. Around 95% of the bladder tumours are of epithelial origin and only around 5% constitute the mesenchymal tumours. Leiomyoma of bladder is the most common mesenchymal tumour comprising one third of them and accounts for only 0.43% of all bladder tumours.

The occurrence of leiomyoma was initially thought to be equal among males and females. But Goluboff et al in their review of 37 patients, reported female preponderance and occurrence of most of the cases in the third to sixth decades with mean age of 44 years. In our series all patients were females with mean age of 31.6 years.

Bladder leiomyoma can occur at different locations with endovesical (63-86%) being the most common one followed by extravesical (11-30%) and intramural (3-7%) locations. Symptomatology ranges from being totally asymptomatic to a variety of other symptoms like, obstructive (49%), irritative (38%), hematuria (11%) and flank pain (13%), which depend mainly on their location and the size they attain. The endovesical type
most commonly produces obstructive or irritative symptoms depending on the size \cite{6}. Very few case reports of leiomyoma presenting with acute retention of urine have been reported \cite{6,7}. Hydronephrosis can also result from the increased intravesical pressure created by the ball valve effect of the endovesical leiomyomas \cite{7}. Matsusmiha et al \cite{6} reported the urodynamic changes which occur due to a leiomyoma causing bladder outlet obstruction. In our series all three patients had obstructive symptoms, one had irritative symptoms, microscopic hematuria was present in one case, two cases presented with acute retention of urine and mild degree of hydronephrosis was present in one case.

Teran and Gambrell \cite{8} proposed 4 theories for the pathophysiology of these lesions: (1) hormonal influences, (2) embryonic rests of the tissues residing in the bladder, (3) perivascular inflammation and metaplastic transformation, (4) infection and inflammation of the bladder musculature. But the exact pathophysiology is still unknown.

The female predominance of these leiomyomas is also a matter of debate and theories of association with female hormones have been advocated \cite{8,9,10}. These tumours are most commonly encountered during fourth to fifth decades when the female hormones are released abundantly. The increased use of the ultrasonography in females could account for the pickup of asymptomatic cases \cite{7}. The occurrence is rare before puberty and decrease in size after menopause have been reported. So the influence of female hormones in the development of leiomyoma can be considered a possibility \cite{11}. In our series all cases were females of child bearing age.

Ultrasoundography is the most common and initial tool for the diagnosis of leiomyomas of bladder. They appear as a homogeneous mass with smooth outline which is usually solid but few cystic appearing lesions have been also reported \cite{12,13}. MRI is a very useful tool in the current era for the accurate depiction of the anatomic location, morphology and the extent of leiomyomas. MRI will show an intermediate signal intensity on both T1 weighted images and intermediate to low signal intensity on T2 weighted images with smooth surface. Few degenerate leiomyomas may have heterogeneous signal intensity. After injection of gadolinium contrast, a variable pattern of enhancement is observed, some enhance heterogeneously and some do not \cite{6,14}. In our series MRI was done in two cases which clearly delineated the mass and the relationship to bladder and surrounding structures. The signal intensity and enhancement pattern was consistent with the classical descriptions. Imaging modalities cannot confirm the exact diagnosis and cannot differentiate a benign leiomyoma from a malignant leiomyosarcoma, so histopathological studies are needed to confirm the diagnosis \cite{15}.

Treatment methods depend on the size, extent and location the tumour and involvement of other structures. Small endovesical tumours are best managed by TUR but the larger endovesical, intramural and the extravesical will require segmental resection \cite{7}. Other approaches of resection like transvaginal, laparoscopic, cystoscopic assisted laparoscopic and robot assisted laparoscopic techniques have been also reported \cite{5,16,17}. Few surgeons advocate reserving surgical treatment only for symptomatic tumours based on the good diagnostic accuracy of imaging and the benign nature of the tumour \cite{5,7}. But the bladder leiomyomas often mimic malignant lesions and can be definitely diagnosed by histopathological analysis only; the ease of TUR resection for diagnosis and treatment favours surgical removal \cite{2}.

Histopathological analysis is the definite diagnosis, grossly the tumour are well circumscribed, white to grey and fleshy. There have been descriptions of few millimeters to 30 cm and few grams to 9 kg in literature \cite{7}. Microscopically they are composed of interlacing fascicles and bundles of uniform spindle cells containing moderate to abundant eosinophilic cytoplasm. The nuclei are elongated, minimal to absent cytological atypia and mitotic figures are absent \cite{15}. Immunohistochemistry is being used routinely to distinguish from the other differential diagnosis like leiomyosarcoma, inflammatory mayofibroblastic tumour, solitary fibrous tumour, perivascular epithelial tumours. Leiomyomas are desmin positive, SMA positive, Ki67 is low, ALK negative, P63 negative, CD34 negative and Vimentin negative \cite{18}.

Prolonged follow up of these patients is not mandatory because of the excellent prognosis and no reports of malignant transformation till date. Few cases of recurrences have been reported, probably because of incomplete excision \cite{2,9}. The recurrence rates may increase after widespread use of TUR which has more possibility of incomplete resection. We followed all the patients by three monthly USG, Uroflowmetry and Cystoscopy for the first year to detect any recurrence. We propose some duration of follow up after TUR, preferably a year. Further studies are needed to find the causes and patterns of recurrences and define the treatment and follow up protocol.

4. Conclusion

Leiomyoma of urinary bladder is a rare disorder which frequently occurs in middle aged females. Symptoms are related to its size and location, the endovesical location and obstructive symptoms are the most common finding. Imaging modalities can diagnose the condition but histopathological analysis is the confirmatory. A number
of surgical modalities of treatment have been described, TUR being the most common modality now. Prognosis of the disease is excellent.

References