Inflammatory Pseudotumor of Urinary Bladder: A Masquerader of Bladder Malignancy

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Abstract

Inflammatory pseudotumor of urinary bladder is a rare benign disease of unknown etiology, characterized by nonneoplastic proliferation of fibroblastic spindle cells, in a background of myxoid and granulation tissue. We report a case of an inflammatory pseudotumor of urinary bladder and discuss the important differentials. A 47-year-old woman presented with longstanding frequency and urgency of micturition. Ultrasound pelvis, color Doppler, computed tomography, and magnetic resonance imaging were performed. Focal bladder wall thickening at the dome was observed. In addition, there was a heterogeneous solid cystic intramural lesion within this bladder wall thickening showing peripheral rim enhancement with nonenhancing central component. Repeated urine cultures were sterile and urine cytology was found to be negative for malignancy. Finally, histopathology and immunohistochemistry confirmed this lesion to be inflammatory pseudotumor.

Keywords: *Histopathology, immunohistochemistry, inflammatory pseudotumor, magnetic resonance imaging, urinary bladder, urine cytology*

Introduction

Inflammatory pseudotumor of urinary bladder is a rare benign entity of unknown etiology.[1] Inflammatory pseudotumor of the bladder was first reported by Roth in 1980.[1] It is a nonneoplastic proliferation of loosely packed fibroblastic spindle cells in a myxoid background with granulomatous reparative response. Inflammatory pseudotumor most commonly occurs in lung and orbit but has been reported to involve almost all organs in the body, with urinary bladder being one of the uncommon sites.[2]

Inflammatory pseudotumor was described for the first time by Brunn in 1939, in the lung. The term "pseudotumor" was coined by Umiker and Iverson in 1954, owing to its tendency to simulate malignancy both clinically and radiologically.[3] Due to various histological appearances, this entity is known by several names such as myofibroblastic tumor, myofibroblastoma, xanthomatous pseudotumor, pseudosarcomatous fibromyxoid tumor, nodular fasciitis of bladder, and atypical fibromyxoid tumor.[4]

Although majority of these lesions are benign, a few of them are locally aggressive. The clinical presentation and nonspecific imaging features pose a great challenge to both clinicians and radiologists as they mimic bladder malignancies, particularly the locally aggressive lesions.[2] Pseudotumor should be considered as an important differential in patients presenting with urinary symptoms with repeatedly sterile urine cultures and negative urine cytology for malignancy. Appropriate knowledge about this benign disease can help obviating the number of unnecessary radical cystectomies performed in cases misdiagnosed as malignancy.

Case Report

A 47-year-old woman presented to the Urology Department with urgency and frequency of micturition for 3 years, with no history of dysuria or hematuria or constitutional symptoms. No significant history of tuberculosis, urolithiasis, trauma, instrumentation or any urinary tract surgery was obtained. Informed consent was taken from the patient. On physical examination, the patient was afebrile and hemodynamically stable. On local examination, mild suprapubic tenderness

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was observed. Urine analysis revealed few pus cells (3–5 cells/HPF) on routine microscopy. Urine cultures were repeatedly sterile with negative acid-fast bacilli cultures. Urine cytology was reported to be negative for malignancy.

The patient was referred to the radiology department for imaging, with a provisional diagnosis of urinary bladder malignancy.

Ultrasound pelvis revealed thickening of the apico-anterior wall of the bladder. A relatively well-defined heterogeneous lesion with a central oval-shaped cystic component was observed within the focally thickened bladder wall [Figure 1a]. On color Doppler, color flow was noted within the mass lesion. However, no flow was noted in the cystic component [Figure 1b].

Further, on noncontrast computed tomography (CT), an intramural solid cystic heterogeneous soft-tissue density mass in the region of dome of bladder was observed [Figure 2]. On postcontrast scans, lesion showed peripheral rim enhancement, with a central oval-shaped nonenhancing hypodense area. Mild perivesical fat stranding was noted anterior to the lesion, abutting the left rectus abdominis muscle and left lateral pelvic wall. No pelvic lymphadenopathy was observed. Bilateral kidneys, ureters, and vesicoureteral junctions were found to be normal.

Subsequently, magnetic resonance imaging (MRI) was performed for better characterization of this mass lesion. An intramural solid cystic lesion with focal wall thickening was observed at the apico anterior wall of the bladder. The intramural lesion depicted predominantly low-signal intensity on both T1- and T2-weighted images. However, there was a central T2 hyperintense oval-shaped area, within this lesion. This central T2 hyperintense component showed peripheral rim enhancement and a hypoenhancing core [Figure 3].

Based on the imaging features observed, the top three imaging differential diagnoses considered were: first, inflammatory pseudotumor of bladder, owing to the typical T2 hypointense signal on MRI with peripherally enhancing central cystic component; second, malakoplakia, owing to focal bladder wall thickening and presence of inflammatory response; and third, eosinophilic cystitis due to the presence of central cystic component within a solid lesion.

Further, cystoscopy and biopsy were performed to confirm the etiology. Histopathology and immunohistochemistry (IHC) was done on biopsy sample. Cystoscopy showed a bulge with reddish velvety thickened patches at the dome of the bladder. Bilateral ureteral orifices and posterior and bilateral lateral walls of bladder were observed to be normal.

Histopathology revealed spindled myocytes and inflammatory cells (predominantly lymphocytes), in a background of granulation tissue [Figure 4]. On IHC, the myocytes stained positive for vimentin and desmin and negative for cytokeratin, actin, and epithelial membrane antigen. These IHC features together with histopathology findings confirmed the lesion to be inflammatory pseudotumor of the bladder and also aided in distinguishing this entity from other varieties of spindle cell carcinomas.

Moreover, lack of any eosinophils or Michaelis–Gutmann bodies, in the tissue sample, helped in excluding the diagnosis of eosinophilic cystitis and malakoplakia, respectively.

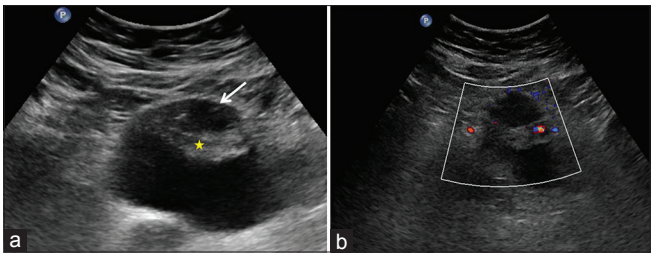

Figure 1: Ultrasound pelvis shows a heterogeneous lesion with a central oval-shaped cystic component (thick white arrow) within focally thickened apico-anterior bladder wall (yellow star) (a). Color Doppler reveals color flow within the mass and no flow in the central cystic component (b)

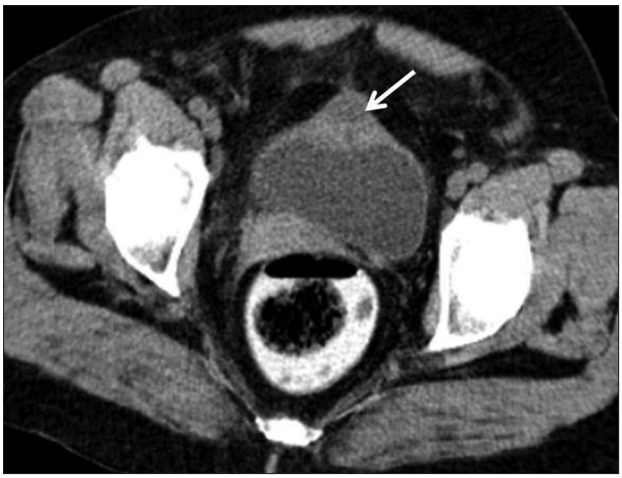

Figure 2: Axial noncontrast computed tomography images showing an intramural solid cystic soft-tissue density mass, in the dome of the bladder (thick white arrow)

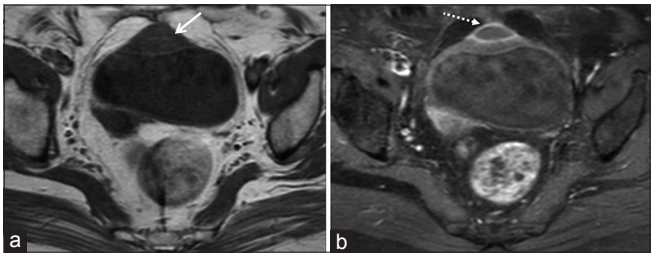

Figure 3: Axial T1-weighted magnetic resonance image showing a low-signal intensity intramural solid cystic lesion (thick white arrow), with focal apico-anterior bladder wall thickening (a). Axial contrast-enhanced magnetic resonance image showing peripheral rim enhancement in the central T2 hyperintense cystic component (dotted white arrow), with a hypoenhancing central core (b)

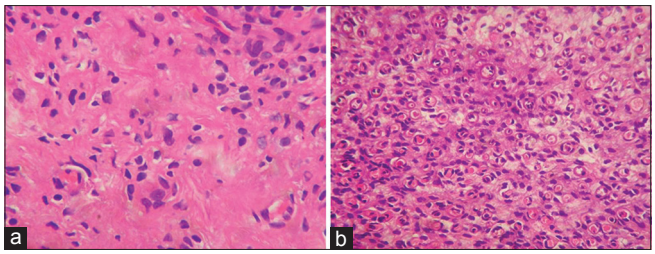

Figure 4: Histopathology image shows spindled myoepithelial cell proliferation with inflammatory infiltrate (a) and spindle cells in a background of granulation tissue (b) (H and E, ×400)

Discussion

Inflammatory pseudotumor of the bladder occurs commonly in adults between 15 and 74 years, with mean age being 38 years.[4,5] Clinically, painless hematuria is reported to be the most common initial presentation, followed by frequency, urgency, and dysuria.[4-6]

Few lesions have been seen to develop in association with prior infection, inflammation, trauma, or surgery. However, majority of them are known to be idiopathic. Unlike transitional cell carcinoma, inflammatory pseudotumor has no reported association with cigarette smoking.[4,6]

Macroscopically, inflammatory pseudotumor of the bladder appears as a solitary exophytic or polypoidal mass lesion. The lesion can be seen at any location within the bladder, with relative sparing of trigone. However, secondary infiltration of trigone has been reported from posterior bladder wall masses.[4]

On imaging, inflammatory pseudotumor of the bladder usually appears as a single polypoidal mass projecting into the bladder lumen or as a submucosal/intramural lesion of solid cystic nature. Although most of these lesions are benign, some lesions are locally aggressive, which depict extension into the perivesical fat and involve rectus abdominis muscles. The aggressive behavior of this variety may masquerade as bladder malignancy.[7] Ultrasound features are nonspecific and may present as asymmetric bladder wall thickening with heterogeneously hypoechoic lesions. Color Doppler reveals increased vascularity within the lesion. CT scan also shows variable appearances with hypodense or hyperdense components. Thickening of the bladder wall with perivesical extension may be observed. The peripheral rim enhancement seen in few lesions is typically produced by the characteristic histological pattern of peripherally arranged spindle and inflammatory cells and central myxoid component.[4,6,7] On MRI, these masses are usually hypointense on both T1- and T2-weighted sequences. On T2-weighted images, however, the lesion may depict heterogeneous nature with a central hyperintense area and a peripheral low signal intensity rim. The T2 hyperintensity of the central component is attributed to necrosis while hypointense periphery is formed by spindle cells, inflammatory cells and fibrotic tissue. On postcontrast MRI, this peripheral component shows rim enhancement, while central necrotic area remains hypoenhancing.[8]

Owing to the nonspecific and varied imaging appearances, histopathological confirmation becomes essential. Invariably, routine light microscopy may prove inefficient in reaching a diagnosis as these lesions may mimic various bladder sarcomas, transitional cell carcinoma, or adenocarcinoma. However, IHC can aid in differentiating spindle cell carcinomas from benign inflammatory pseudotumors. On IHC, the spindle cells of benign inflammatory pseudotumors are negative for cytokeratin, usually stain positive for vimentin, and variably positive for desmin and actin, whereas the spindle cell carcinomas are immunopositive for cytokeratin and epithelial membrane antigen.[9]

Other important differentials to be considered are urothelial malignancies, especially transitional cell carcinoma, bladder endometriosis, malakoplakia, eosinophilic cystitis, bladder tuberculosis, and schistosomiasis.[4] The important differentiating features of urothelial carcinoma are T2 hyperintensity and early avid enhancement, in comparison to T2 hypointensity and peripheral ring-like enhancement with a hypoenhancing central area in inflammatory pseudotumor. Bladder schistosomiasis shows nodular bladder wall thickening on ultrasound, CT, and MRI in acute phase, while in chronic phase, bladder appears contracted and thick-walled with peripheral calcifications. Bladder endometriosis shows single or multiple implants typically situated in the uterovesical pouch. The characteristic MRI features consist of hemorrhagic lesions which are T1 hyperintense and show blooming on gradient sequences. Eosinophilic cystitis is characterized by eosinophilic infiltration of the bladder wall and on imaging shows normal or thickened bladder wall. A cystic variant depicts enhancing walls mimicking inflammatory pseudotumor. Malakoplakia is an uncommon chronic granulomatous condition, most commonly affecting the urinary tract. Imaging reveals either multiple enhancing solid masses or circumferential wall thickening. At times, malakoplakia may be highly aggressive, showing perivesical region infiltration and bone destruction. Vesicoureteric reflux and ring-shaped bladder calcification may also be seen. Histological confirmation is ultimately required to distinguish inflammatory pseudotumor from these similar appearing bladder lesions.[4]

There is no consensus on definite treatment of inflammatory pseudotumor of the bladder. However, as these lesions have a benign course, partial cystectomy with total resection of tumor and preservation of normal tissues at surgical margins remains the best curative treatment.[1-9] Reviewing the literature, a few cases of spontaneous resolution have been documented, with no reports of either the local recurrence or distant metastasis.[5-12]

Conclusion

Inflammatory pseudotumor should be considered as a differential diagnosis in urinary bladder masses presenting with hematuria or other urinary complaints, especially in patients with negative urine cytology and sterile urine cultures. The few characteristic imaging features, especially the T2 hypointensity on MRI, coupled with histopathology and IHC, play an essential role in reaching an accurate diagnosis of a benign process, thus reducing the number of false positive radical bladder surgeries, being performed on benign masses.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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