Case Report

Inflammatory Pseudotumor of Bladder Report of 2 Cases and Review of Literature

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Keywords: bladder, pseudosarcoma, inflammatory pseudotumor

Urol J. 2008;5:62-5. www.uj.unrc.ir

INTRODUCTION

Inflammatory pseudotumor of the bladder is a rare benign proliferative lesion that resembles a neoplastic tumor. (1-5) Most of the tumors of the bladder are malignant, and transitional cell carcinoma is the most common malignant tumor; however, other tumors such as adenocarcinoma, squamous cell carcinoma, sarcoma, and teratoma have to be included in the differential diagnosis. In fact, their clinical manifestations are similar and patients usually present with hematuria (painless and microscopic or gross). Pelvic pain and urinary bladder irritation may be seen uncommonly. These symptoms may also be caused by nonmalignant conditions such as clustering polypoid cystitis or granulomatous cystitis. These lesions may mimic malignant tumor. (5,6) We report 2 cases of inflammatory pseudotumors of the bladder presented with gross hematuria and urinary symptoms. We reviewed the literature and found few cases of pseudotumor, and our cases were unique in terms of their etiologies. (7-9)

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> Received May 2006 Accepted February 2007

CASE REPORT

Case 1

A 58-year-old woman was referred to our hospital with long-standing

dysuria (more than 2 years) and painless hematuria. At the time of admission the patient was anemic. She had no prior history of trauma, instrumentation, or surgical operation on the pelvis or abdomen. She seemed mentally normal with no history of spurious self-trust. On physical examination, tenderness on deep palpation of the suprapubic area was found. Urinalysis showed persistent microscopic hematuria, whereas urine culture remained negative for microorganisms for past 2 years. Cystoscopy revealed blister edema in the bladder. Evaluation for tuberculosis and other organisms were negative. Intravenous urography revealed a normal pyelocaliceal system and a filling defect within the urinary bladder. Contrast-enhanced computed tomography (CT) showed an oval polypoid mass in the left side of the urinary bladder with a linear shape high-density foreign body within the mass (Figure 1). The lesion also exhibited an infiltrative feature and extension to the perivesicular fat and the lower abdominal and pelvic walls (Figure 1). The patient underwent transurethral resection of the lesion. It was a 60×65 -mm pinktan rubbery tissue. Microscopic examination of lesion revealed severe lymphoplasmocytic

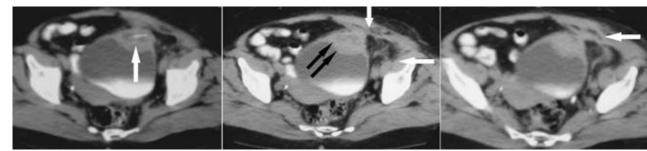


Figure 1. Left, Contrast-enhanced pelvic CT scan shows an oval shape polypoid mass protruded into the bladder in the left anterior aspect (white arrow). Note a linear high-density foreign body within the mass. **Middle,** A slightly upper slide of the pelvic CT scan demonstrates an infiltrative feature as extension of the lesion out of the bladder with involvement of the perivesicular fat (long white arrow) and the pelvic wall (short white arrow). Some low-attenuation area (necrotic) within the lesion is noted (black arrows). **Right,** The lesion shows extension to the abdominal wall adjacent to the pelvic cavity with subcutaneous edema (white arrow).

infiltration with lymphoid follicles in a fibrotic foreign-body type granulomatous inflammatory background. Two irregular pieces of foreign bodies (wood) were also seen. The patient denied any possible cause of the appearance of a foreign body in the bladder. No malignancy or recurrent lesions were detected 1 year after the surgery.

Case 2

A 23-year-old woman was admitted to the rheumatology clinic for acute pelvic pain and painful gross hematuria following 2 months of oral cyclophosphamide intake (100 mg/d) for treatment of lupus erythematous which was resistant to corticosteroid therapy.

Physical examination was unremarkable. Urinalysis confirmed gross hematuria and a urine culture was negative for microorganisms. Ultrasonography revealed a round-shape hypoechoic mass in the right side of the bladder and irregularity and thickness of its anterior wall. Contrast-enhanced CT showed a filling defect in the wall of the bladder (Figure 2). On cystoscopy, the floor and the lateral lower third of the bladder were normal, but a papillary-type lesion had occupied the remaining portion of the walls and entire dome of the bladder, which was more prominent on right side. The patient underwent transurethral resection. The surgical specimen was a firm polypoid mass with surface

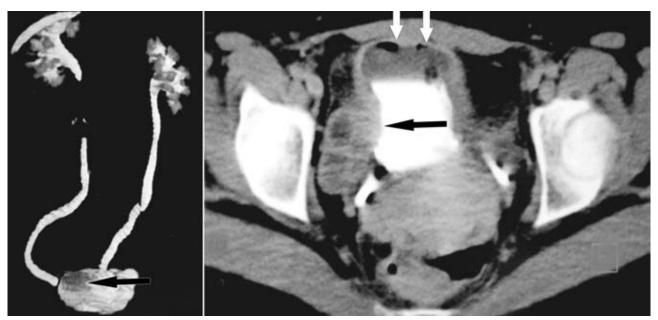


Figure 2. Left, Contrast-enhanced pelvic CT scan shows filling defects with an exophytic growth pattern of the lesion in the right side of the bladder (black arrow). Air bubbles in the bladder are related to previous catheterization (white arrows). Right, Spiral CT intravenous urography shows an irregularly outlined mass within the right side of bladder (black arrow). Note mild dilatation of the right distal ureter.

irregularity and hemorrhage. On physical examination, transitional mucosa with subjacent muscle layer was seen. The mucosa showed edema, infiltration of mononuclear leukocytes, and formation lymphoid follicles. No evidence of malignancy was noted. The final diagnosis was polypoid cluster-type cystitis. During the follow-up, cyclophosphamide was discontinued and the patient was free of symptom.

DISCUSSION

Inflammatory pseudotumor of the urinary bladder was first reported in 1980 by Roth. (10) This lesion has been referred to by various terms including nodular fasciitis, pseudosarcomatous fasciitis, inflammatory pseudotumor, and reactive pseudotumor. (7,8) There is no sex or age predilection. (4) Although some of these lesions are considered to represent an unusual inflammatory response to infection, trauma, or surgery, most patients show no predisposing factors. (4) It is assumed that multiple etiologic factors may play a role in the pathogenesis. In our patients, we assumed these factors may be related to cyclophosphamide administration and foreign body (wood). Diagnosis of the lesion may remain as a dilemma for the urologist, radiologist and pathologist.

Because of an aggressive appearance of lesion, inflammatory pseudotumor may be confused with transitional cell carcinoma, adenocarcinoma, or other malignant tumors. In children, this lesion may clinically mimic and may even pathologically resemble rhabdomyosarcoma. (2,7,9) Immunohistochemical study can be misleading since pseudotumor of the bladder shares in common with those malignant conditions, positivity with some markers, such as desmin with rhabdomyosarcoma and cytokeratin with sarcomatoid carcinoma (11); however, immunohistochemistry and electron microscopy can be confirmatory when diagnosis by routine light microscopy is deficient.

The radiological appearance of inflammatory pseudotumor is nonspecific and cannot be differentiated from a malignant neoplasm. (2,7,12) A broad-based enhancing centrally necrotic mass may be seen on CT scan involving the bladder

wall with extension to the perivesical soft tissues and the rectus abdominus muscle. Our case 1 showed an oval polypoid mass on the CT scan. The lesion protruded into the left and anterior walls of the bladder and contained cystic area with a linear-shape density which was proved a foreign body (wood). The perivesical soft tissue and abdominal muscles were also involved.

In the case 2, the patient showed a slightly irregularly outlined defect on CT scan. The lesion showed enhancement with an exophytic growth pattern. In the recent studies, magnetic resonance imaging of inflammatory pseudotumor of the bladder and polypoid and papillary cystitis showed a large intraluminal mass with a narrow stalk, mimicking a welldifferentiated papillary tumor that was accompanied by massive hematuria. (4,6) Kim and colleagues reported an isosignal intensity with the bladder wall on T1-weighted images with central necrosis. On T2-weighted image, the lesion had a low-signal intensity with central necrosis and mild enhancement after contrast media administration. (6)

There is no consensus on the best treatment method due to the rarity of the disease. Partial or total cystectomy has been performed, and no recurrence has been reported after mass excision or transurethral resection. Our patients showed no evidence of recurrent lesion, either.

CONFLICT OF INTEREST

None declared.

ACKNOWLEDGMENT

We would like to thank Dr M Kashi for his invaluable assistance.

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