

Inflammatory pseudotumor of the urinary bladder: A case series among more than 2,000 urinary bladder tumor cases

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Abstract

“Inflammatory pseudotumor” (IPT) has infrequently been reported in the medical journals. A retrospective analysis was conducted among more than 2,000 bladder tumor cases from January 1999 to December 2012 looking for patients with IPT in the final diagnosis. Six patients were found with median tumor size of 3.5 cm (range: 3–8 cm); computed tomography and/or magnetic resonance imaging was used to diagnose the tumor. All patients had complete resection of the tumors. On a median follow-up of 6 years (range: 2–10 years), no recurrences for IPT have been observed in all patients. We concluded that IPT is a rare disease of the urinary bladder and should be regarded with a high degree of suspicion. Although an extensive workup may be needed for definite diagnosis, it is worth to avoid unnecessary chemoradiotherapy or radical surgeries.

Key Words: Benign tumors, inflammatory pseudotumor, postoperative spindle cell nodule, urinary bladder

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INTRODUCTION

Benign tumors of the urinary bladder are rare, but there are special concerns as they may mimic cancer during a clinical diagnosis, with radiologic and cystoscopic findings. Once mistaken for cancer, the patient may be subjected to unnecessary cancer management and its deleterious consequences.

“Inflammatory pseudotumor” (IPT) is a broad term that refers to benign proliferative or reactive lesions of the submucosal stroma.^[1] Postoperative spindle cell nodule (PSCN) is a sub-entity of these tumors when it is diagnosed within the few months of previous surgical procedures at the surgical site.^[2]

As a rare condition, case reports and series have been mentioned in the literature^[3] with little being known regarding the long-term follow-up. Urologists need to report their findings from more than 2,000 bladder tumor cases diagnosed at larger urology centers in the region with long-term follow-up.

METHODS AND RESULTS

From January 1999 to December 2012, a retrospective analysis was conducted on 2,050 patients found in the electronic database. These patients had been previously

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diagnosed with bladder tumors. All pathological reports were reviewed to identify any patients with ISPT (ISP) or PSCNs.

Data, including demographics, history, presentation at diagnosis, and radiologic and cystoscopic findings, are summarized in Table 1.

All lesions were diagnosed by computed tomography (CT) and/or magnetic resonance imaging demonstrating a median tumor size of 4.5 (range: 3–8) cm in maximum diameter. In one patient, CT revealed mild right hydronephrosis extending into the lower ureteric end [Figure 1a], as well as a large exophytic enhancing soft tissue mass at the right and posterior bladder walls [Figure 1b]. Four months after resection, symptoms and signs were markedly improved. Control CT demonstrated resolution of the right hydronephrosis [Figure 2a] and a marked reduction in the pelvic mass size [Figure 2b].

In all patients, the final diagnosis was confirmed by histopathological examination revealing IPT/PSCN associated with hyperplastic urothelial changes and polypoid cystitis with no evidence of malignancy [Figure 3]. At a median follow-up of 6 years (2–10), no recurrences for PSCN were observed in all patients. However, one patient presented with hematuria and recurrent bladder tumor after 10 years of follow-up who was managed with radical cystectomy with GIII T2a transitional cell carcinoma of the urinary bladder at the final diagnosis.

COMMENT

This study describes six patients of ISPT/PSCN diagnosed over a 10 years period at a referral tertiary center. Our main objective in this report is to alert urologist to this rare type of bladder tumor and to advise them to keep in mind the possibility of developing such tumor after bladder instrumentation. Furthermore, repeat negative biopsies in the presence of solid bladder lesion warrants careful attention. After resection, close monitoring and follow-up are mandatory, as there is a possibility of recurrence. In our series, painless hematuria and obstructive/irritative voiding symptoms are the most common symptoms.

These tumors are characterized grossly by their circumscribed or multinodular firm, white, or tan mass with a whorled fleshy or myxoid cut surface. Focal hemorrhage, necrosis, and calcification may be seen in a minority of cases. Microscopically, 83% of the lesions were noted to have a myxoid or vascular pattern, 55% had a compact spindle cell pattern, and up to 41% of these patients had mixed histologic patterns. Significant numbers of inflammatory cells (plasma cells, lymphocytes, eosinophils, and neutrophils) were reported.^[4]

Harik *et al.* reported development of PSCN in 9 patients after bladder instrumentation among 42 diagnosed with inflammatory myofibroblastic tumors.^[5] In another study, Montgomery *et al.* reported 8 patients who developed PSCN from 47 benign bladder lesions. PSCN could develop after transurethral resection, open prostatectomy, radical

Table 1: Patient demographics and tumor criteria

Cases	Age	Sex	History	Period (months)*	Presentation	Diagnostic modalities	Tumor site	Tumor size (cm)	Tumor shape	Management	Follow-up (years)	Condition at last follow-up
1	19	Male	Cystoscopic biopsy	1	Painless hematuria, suprapubic pain, right flank pain	CT, cystoscopic biopsy	Posterior and right lateral walls	8	Fungating polypoidal	TUR	6	No right hydronephrosis and no recurrence for PSCN
2	58	Female	TURBT (grade II TCC pT1a)	3	Irritative lower urinary tract symptoms	CT	Posterior wall	3	Nodular	TUR	10	Recurrent bladder tumor, radical cystectomy, GIII T2a TCC of the UB
3	28	Male	Irrelevant	-	Hematuria	MRI-pelvis	Anterior wall to the right	4	Nodular	TUR	5	No recurrence of PSCN
4	34	Female	Complicated IUD, cystoscopic guided biopsies	3	Irritative lower urinary tract symptoms	MRI-pelvis, diagnostic laparoscopy	Dome and right lateral wall	5	Nodular	Biopsy by diagnostic laparoscopy and TUR	7	No recurrence of PSCN
5	34	Female	Cystoscopic guided biopsies	3	Hematuria	MRI-pelvis	Trigone	5	Fungating polypoidal	TUR	3	No recurrence of PSCN
6	18	Male	Cystoscopic guided biopsies	4	Hematuria	MRI-pelvis	Trigone and posterior wall	4	Nodular	TUR	2	No recurrence of PSCN

*Period between instrumentation and diagnosis of PSCN. CT: Computed tomography, IUD: Intrauterine device, MRI: Magnetic resonance imaging, PSCN: Postoperative spindle cell nodules, UB: Urinary bladder, TUR: Transurethral resection, TURBT: Transurethral resection of bladder tumor, TCC: Transitional cell carcinoma

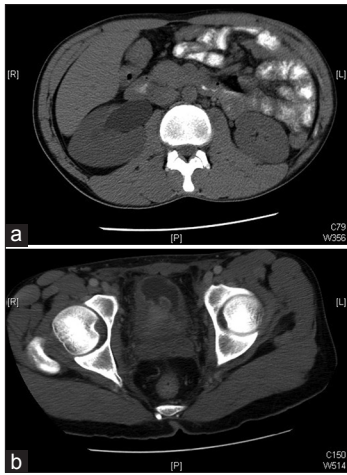


Figure 1: (a) Preoperative computed tomography showing right hydronephrosis. (b) Preoperative postcontrast computed tomography showing large exophytic enhancing soft tissue mass at the right and posterior bladder walls



Figure 2: (a) Postoperative computed tomography showing resolution of right hydronephrosis. (b) Postoperative computed tomography showing reduction of mass size

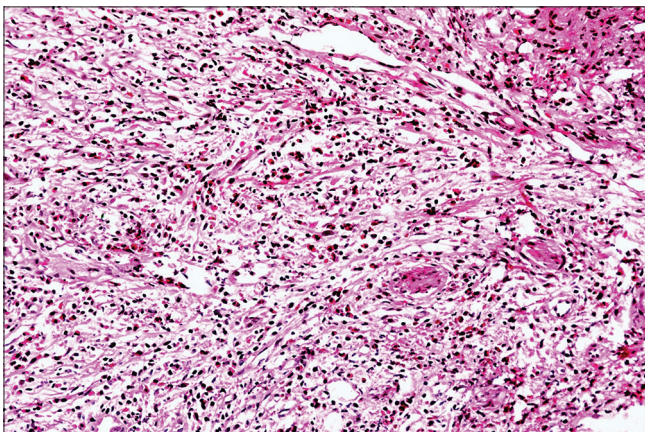


Figure 3: H and E stain showing postoperative spindle cell nodules associated with hyperplastic urothelial changes and polypoid cystitis

prostatectomy, and indwelling stent for ureteropelvic junction obstruction.^[1]

Different terms have been used in the literature to describe the lesion, for example, “nodular fasciitis,” “pseudomalignant spindle cell proliferation,” “pseudosarcomatous fibromyxoid tumor,” “reactive pseudosarcomatous response,” and “pseudosarcomatous myofibroblastic proliferation.”^[6-9] More recently, the term “inflammatory myofibroblastic tumor” (IMT) has come to be commonly used based on electron microscopic and immunohistochemical findings,^[6,8] These lesions showed strong diffuse cytoplasmic positivity for vimentin. Smooth muscle actin and muscle specific actin vary from focal to diffuse pattern.

A genetic cause has been identified involving the human anaplastic lymphoma kinase (ALK) gene, present on short arm of “chromosome 2.” On reviewing 182 patients who had IMTs, 65% were ALK-positive.^[4] Such genetic abnormalities could be detected by fluorescence *in situ* hybridization technology and might play a role in the future for identifying the nature and course of these lesions.^[2]

After complete resection of the tumor and median follow-up of 6 years (range: 5–10 years), no recurrences have been observed. However, the possibility of recurrence should be kept in mind. Harik *et al.* reported recurrences in 3 patients (out of 9) with no metastasis.^[5] Proppe *et al.* reported 2 recurrences after resection; nevertheless, there were no subsequent recurrences after re-excision.^[10] On the other hand, with median follow-up of 3.1 years (2 months to 5.6 years), no recurrence has been reported.

CONCLUSION

IP/PSCN is rare diseases of the UB and should be regarded with a high degree of suspicion. This is especially the case if the tumor appears after recent bladder instrumentation. Although a definite diagnosis may need an extensive workup, repeated biopsies, and special staining, it is worth to avoid unnecessary chemoradiotherapy or radical surgeries with their deleterious consequences on patient’s quality of life.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Montgomery EA, Shuster DD, Burkart AL, Esteban JM, Sgrignoli A, Elwood L, *et al.* Inflammatory myofibroblastic tumors of the urinary tract:

- A clinicopathologic study of 46 cases, including a malignant example inflammatory fibrosarcoma and a subset associated with high-grade urothelial carcinoma. *Am J Surg Pathol* 2006;30:1502-12.
2. Shanks JH, Iczkowski KA. Spindle cell lesions of the bladder and urinary tract. *Histopathology* 2009;55:491-504.
 3. Angulo JC, Lopez JI, Flores N. Pseudosarcomatous myofibroblastic proliferation of the bladder: Report of 2 cases and literature review. *J Urol* 1994;151:1008-12.
 4. Teoh JY, Chan NH, Cheung HY, Hou SS, Ng CF. Inflammatory myofibroblastic tumors of the urinary bladder: A systematic review. *Urology* 2014;84:503-8.
 5. Harik LR, Merino C, Coindre JM, Amin MB, Pedeutour F, Weiss SW. Pseudosarcomatous myofibroblastic proliferations of the bladder: A clinicopathologic study of 42 cases. *Am J Surg Pathol* 2006;30:787-94.
 6. Das S, Upton JD, Amar AD. Nodular fasciitis of the bladder. *J Urol* 1988;140:1532-3.
 7. Albores-Saavedra J, Manivel JC, Essendorf H, Dehner LP, Drut R, Gould E, *et al.* Pseudosarcomatous myofibroblastic proliferations in the urinary bladder of children. *Cancer* 1990;66:1234-41.
 8. Lundgren L, Aldenborg F, Angervall L, Kindblom LG. Pseudomalignant spindle cell proliferations of the urinary bladder. *Hum Pathol* 1994;25:181-91.
 9. Coyne JD, Wilson G, Sandhu D, Young RH. Inflammatory pseudotumour of the urinary bladder. *Histopathology* 1991;18:261-4.
 10. Proppe KH, Scully RE, Rosai J. Postoperative spindle cell nodules of genitourinary tract resembling sarcomas. A report of eight cases. *Am J Surg Pathol* 1984;8:101-8.