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

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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 diagnosed 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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**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.

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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 hydroureteronephrosis 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 hydroureteronephrosis [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 (years) | 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 polyoidal | TUR | 6 | No right hydronephrosis and no recurrence for PSCN |
| 2     | 58          | Female | TURBT (grade II TCC pT1a) | 3 | Irritative lower urinary tract symptoms | Hematuria | Posterior wall | 3 | Nodular | TUR | 10 | Recurrent bladder tumor, radical cystectomy, GIII T2a TCC of the UB |
| 3     | 28          | Male  | Irrelevant | - | - | MRI-pelvis | Anterior wall to the right Dome and right lateral wall | 4 | Nodular | Biopsy by diagnostic laparoscopy and TUR | 7 | No recurrence of PSCN |
| 4     | 34          | Female | Complicated IUD, cystoscopic guided biopsies | 3 | Irritative lower urinary tract symptoms | Hematuria | MRI-pelvis, diagnostic laparoscopy | 5 | Nodular and bullous edema | Biopsy by diagnostic laparoscopy and TUR | 7 | No recurrence of PSCN |
| 5     | 34          | Female | Cystoscopic guided biopsies | 3 | Hematuria | MRI-pelvis | Trigone and posterior wall | 5 | Fungating polyoidal Nodular | 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
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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.

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