A Rare Case in Urology: Inflammatory Myofibroblastic Tumour

Murat Akgul[1], Mehmet Fatih Sahin[1], Aysegul Arslan[2], Meltem Oznur[2] and Cenk Yazici[1]

[1]*Department of Urology, Faculty of Medicine, Tekirdag Namik Kemal University, Tekirdag, Turkey*
[2]*Department of Pathology, Faculty of Medicine, Tekirdag Namik Kemal University, Tekirdag, Turkey*

ABSTRACT

Inflammatory myofibroblastic tumour (IMT) is a soft tissue malignancy with a mixture of myofibroblastic spindle cells with hyalinised stroma and inflammatory infiltrates. We report a case of a 35-year male patient with a 100×90 mm mass located at the posterior wall of the urinary bladder just adjacent to the prostate and rectum. The mass caused grade 3 hydronephrosis in the left kidney and grade 1 hydronephrosis in the right kidney. The patient was operated and the mass lesion was excised completely. Because the left ureter was adherent to the mass, it was excised at the most distal part and uretero-neo-cystostomy was performed as the bladder and the prostate were preserved. Morphology and immunohistochemistry were consistent with IMT. With surgical excision of the tumour, no recurrence or metastases were seen up to 3 years postoperatively.

Key Words: *Inflammatory myofibroblastic tumour, Bladder, Urinary system, Hydronephrosis.*

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INTRODUCTION

Inflammatory myofibroblastic tumour (IMT) is a type of soft tissue malignancy with myofibroblastic spindle cells, hyalinized stroma and inflammatory infiltrate. There are limited numbers of case reports on IMT in the literature. The pathophysiology of IMT is unknown and it has mostly been reported in the lungs and liver.[1] There are very few case reports documenting the presence of IMT in and around the urinary tract. In this case report, we present an IMT case in which the tumour was located in serosa of the bladder adjacent to the prostate and rectum, which is a very rare location for IMT.

CASE REPORT

A 35-year male patient was referred with left lumbar pain. He had no comorbidity and no family history related to IMT. Urinary ultrasound scan revealed grade 3 hydronephrosis in the left kidney and grade 1 hydronephrosis in the right kidney. The ultrasonography also reported a 100×90 mm mass at the posterior wall of the bladder just adjacent to the prostate. The patient's serum creatinine level was 1.44 mg/dL. Magnetic Resonance Imaging (MRI) scan revealed an 82×89×96 mm mass at the posterior wall of the bladder, which significantly pushed the bladder anteriorly (Figure 1). The demarcation line was very close to the prostate but with careful evaluation, it was observed that the tumour was not originating from the prostate.

In order to relieve hydronephrosis and stabilise the renal function, left percutaneous nephrostomy was performed. In antegrade pyelography, a complete obstruction of the left distal ureter at the level of pelvic mass was observed. The surgery was preferred as an initial treatment. The operation was started with 10 cm infra-umbilical midline incision; the mass was seen in the retroperitoneal area. The anterior side of the mass lesion was dissected and separated from the detrusor muscle of the bladder, and the posterior side was separated from the recto-sigmoid colon. As the tumour involved the distal left ureter completely, the distal part of the left ureter could not be separated from the tumour. Therefore, the left ureter was excised at the most distal point and extravesical uretero-neo--cystostomy was performed after excision of the mass lesion. No complications were reported peroperatively, and operation time was 5 hours with total blood loss of 400 cc.

In pathological examination, it was a nodular mass with regular borders macroscopically. In microscopic examination, a spindle cell lesion composed of plump spindle cells with abundant eosinophilic cytoplasm and large vesicular nuclei with occasional prominent nucleoli was noted. The spindle cells were disposed in varying dimensions in edematous and fibromyxoid stroma (Figure 2). A small number of mitotic figures were detected. Lymphocytes, plasma cells, histiocytes and rare eosinophils and neutrophils formed inflammatory cell filtrates. On light microscopic evaluation, gastrointestinal stromal tumour (GIST), smooth muscle and neural tumours were in the

Correspondence to: Dr. Murat Akgul, Department of Urology, Faculty of Medicine, Tekirdag Namik Kemal University, Tekirdag, Turkey
E-mail: drmuratakgul@gmail.com

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differential diagnosis. On immunohistochemistry (IHC) staining, spindle cells were ALK (+), Vimentin (+), SMA (+), Desmin (+), C-Kit (-), CD68 (-), Ki-67 (-), S-100 (-), while inflammatory cells were CD45 (+) and CD138 (+). Histopathological and IHC staining results confirmed it as IMT. Patient follow-up showed no sign of recurrence or metastasis in postoperative 36 months (Figure 3). Thoraco-abdomino-pelvic CT scans were performed with a three-month period initially and with a six--month period afterwards.

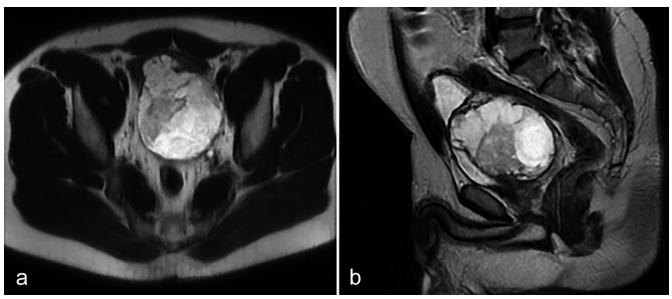

Figure 1: Preoperative magnetic resonance imaging of the pelvis. (a) Transverse plane (b) Sagittal plane.

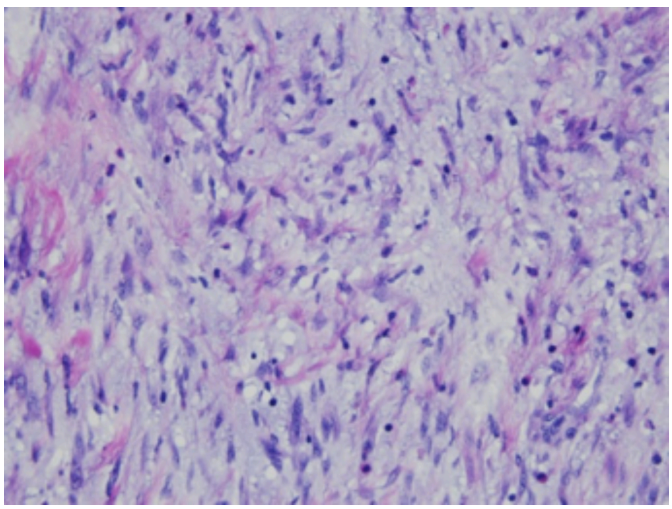

Figure 2: Histopathology of inflammatory myofibroblastic tumour. Eosinophilic spindle cells and scattered inflammatory cells are seen embedded in fibromyxoid stroma (Hematoxylin and Eosin stain, ×200).

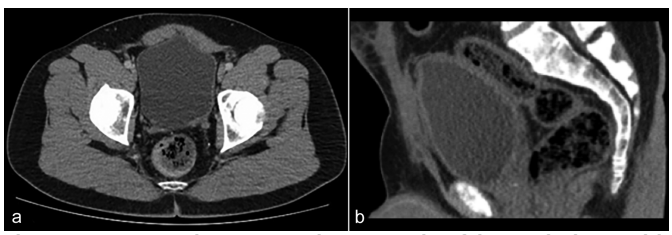

Figure 3: Postoperative Computed Tomography of the surgical area with no recurrence. (a) Transverse plane (b) Sagittal plane.

DISCUSSION

IMT is a spindle cell proliferation that can mimic leiomyosarcoma and may appear in different organ systems at any age. Histopathological examination and IHC evaluation are essential for an exact diagnosis. It has a unique histopathology with spindle cell proliferation associated with acute/chronic inflammatory cells (*e.g*. lymphocytes, eosinophils and macrophages) that rarely shows mitotic activity. Spindle cells are atypical with mild nuclear pleomorphism. In present case, typical histopathological and IHC features were present.

Roth *et al*. first described IMT in 1980. It can be seen in different parts of the body and different organ systems. It is most commonly seen in liver and biliary tract (32%), lungs (27%) and gastrointestinal system (10%). IMT could be seen at any age (3-89 years). There is no data in the literature about the most prevalent age group.[2] In this case, the patient was 35 years old. In the urinary tract, it can be seen in the renal parenchyma, renal pelvis and more frequently in the bladder. There are even some case reports documenting the tumour as mesh reactions after mid-urethral sling surgery.[3]

It is clinically and pathologically difficult to differentiate IMT localised in the bladder from rhabdomyosarcoma, leiomyosarcoma, and sarcomatoid urothelial carcinoma.[4] GIST should be considered in differential diagnosis with intra-abdominal IMT. In majority of cases, submucosa of the bladder is affected. However, in the present case, the tumour originated from the serosa of the bladder. If submucosa of bladder is affected, cases usually present with gross hematuria.[5] In this case, the presentation was with lumbar pain due to obstruction of the ureter. Cases localised in the bladder can be treated with surgery such as transurethral resection or partial cystectomy.[6,7] In present case, we also preferred surgery as an initial treatment. In literature, patients with ALK gene rearrangements and positive Rain Binding Protein (RANBP2) are more prone to local recurrence and metastasis and there are also cases treated with Celcoxib, without any surgical intervention.[8]

In this case, the demarcation line was very close to the prostate. There are also limited IMT cases originating from the prostate in the literature.[9] It can appear either after a transurethral resection of the prostate (TUR-P) or without any TUR surgery. A five-year survival rate is approximately 72%.[10] Complete resection is recommended in the management of the disease and radiation therapy might be the other option. It is also reported in the literature that corticosteroid therapy can be added to anti-inflammatory therapy. It generally has low malignant potential and low local recurrence rates (2-25%); however, there are cases of IMT originating from prostate and transforming into leiomyosarcoma.[2,8]

In summary, we presented an IMT case that was localised to the posterior wall of bladder that caused obstructive renal failure. According to our knowledge, there is no such IMT case with the same location and similar clinical presentation reported in the literature. We preferred surgical treatment because of obstructive renal failure due to hydronephrosis. No sign of recurrence or metastasis was detected 36 months postoperatively. As IMT is a very rare pathology, there is no guideline recommendation about treatment and follow-up protocols.

PATIENT'S CONSENT:

Informed consent was obtained from the patient.

COMPETING INTEREST:
The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:
MA: Involved in writing the case report section, reviewed the literature section, and performed proofreading.
MFS: Wrote introduction and discussion.
AA, MO: Involved in writing case report section.
CY: Reviewed the literature section and performed proof-reading.

REFERENCES

1. Liang W, Zhou X, Xu S, Lin S. CT manifestations of inflammatory myofibroblastic tumours (inflammatory pseudotumours) of the urinary system. *American J Roentgenology* 2016; **206(6)**:1149-55. doi:10.2214/AJR.15.14494.
2. Lecuona AT, Van Wyk AC, Smit SG, Zarrabi AD, Heyns CF. Inflammatory myofibroblastic tumour of the bladder in a 3-year-old boy. *Urology* 2012; **79(1)**:215-18. doi: 10.1016/j.urology.2011.04.052.
3. Kwon SY, Latchamsetty KC, Benson J, Carreno M. Inflammatory myofibroblastic tumour of the urinary tract following a TVT. *Female Pelvic Medicine Reconstructive Surgery* 2012; **18(4)**:249-51. doi: 10.1097/SPV.0b013e318262d994.
4. Lott S, Lopez-Beltran A, MacLennan GT, Montironi R, Cheng L. Soft tissue tumours of the urinary bladder, part I: Myofibroblastic proliferations, benign neoplasms, and tumours of uncertain malignant potential. *Human Pathology* 2007; **38(6)**:807-23. doi: 10.1016/j.humpath.2007.03.017.
5. Higazy A, Kandel M. Prolapsing inflammatory myofibroblastic tumour of the urinary bladder: A case report and a review of literature. *Urol Case Rep* 2020; **31(4):**101191. doi: 10.1016/j.eucr.2020.101191.
6. Santos Lopes S, Furtado A, Oliveira R, Cebola A, Graça B, Ferreira Coelho M, *et al*. Inflammatory myofibroblastic tumour of the bladder: 2 rare cases managed with laparoscopic partial cystectomy. *Case Reports Urology* 2016; doi: 10.1155/2016/4976150.
7. Xu H, He B, Tu X, Bao Y, Yang L, Zhuo H, *et al*. Minimally invasive surgery for inflammatory myofibroblastic tumour of the urinary bladder: Three case reports. *Medicine* 2018; **97(49)**. doi: 10.1097/MD.0000000000013474.
8. Wein A, Kavoussi L, Partin A, Peters C. Campbell-Walsh Urology. 11th ed. 2015; p.2599.
9. Kuramoto T, Kohjimoto Y, Mori T, Kikkawa K, Senzaki H, Hagino K, *et al*. Inflammatory pseudotumour of the prostate: A case report. *Hinyokika kiyo Acta Urologica Japonica* 2005; **51(11)**:767-70.
10. Montgomery EA, Shuster DD, Burkart AL, Esteban JM, Sgrignoli A, Elwood L, *et al*. Inflammatory myofibroblastic tumours 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. *American J Surgical Pathology* 2006; **30(12)**:1502-12. doi: 10.1097/01.pas.0000213280.35413.1b.