ORIGINAL RESEARCH PAPER
VOLUME-6 | ISSUE-8 | AUGUST - 2017 | ISSN No 2277 - 8179 | IF : 4.176 | IC Value : 78.46
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

INFLAMMATORY MYOFIBROBLASTIC TUMOUR OF URINARY BLADDER- A CASE REPORT

Pathology

Jamal Arshad — Associate Prof & HOD, Department of Urology, RIMS, Ranchi, Jharkhand, India,

Chakrabarty Barun K — Graded Specialist (Pathology), Department of Pathology;151 Base Hospital, Guwahati, Assam, India- Corresponding Author

Deb Prabal — Senior Advisor and Prof (Path & Neuropath); Command Hospital (EC), Kolkata,West Bengal; India;

Rai Manoj K — Ex Prof and HOD, Department of Pathology, RIMS, Ranchi, Jharkhand, India,

ABSTRACT

We illustrate a case of an inflammatory myofibroblastic tumor (IMT) involving the urinary bladder in a 17-year-old woman with no previous surgical history and review the literature and differential diagnosis. Genitourinary inflammatory myofibroblastic tumor is a rare lesion and is characterized by fascicular arrangement of myofibroblasts admixed with inflammatory cells and characteristic slit-like vessels. Urinary bladder IMT can be a diagnostic pitfall because of its overlapping histologic features that can mimic malignancy. Proper diagnosis requires histopathological examination in combination with an exhaustive immunohistochemical panel, alongwith demonstration of anaplastic lymphoma kinase overexpression (by immunohistochemistry or gene rearrangement studies), to distinguish IMT from its malignant mimics. We have provided economical and modified panel for diagnosis of IMT suitable for resource constraints countries.

KEYWORDS:

Inflammatory myofibroblastic tumor, malignant potential, urinary bladder

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) of the urinary bladder is a very rare spindle cell tumor with undetermined malignant potential. We report a case of urinary bladder IMT in a young adult female and discuss its clinico-radiological findings, histopathological and immunohistochemical diagnostic criterias, differential diagnosis, management and follow up.

CASE STUDY

A 17-years-old female with no previous surgical history presented with painless hematuria, urinary blood clots, burning micturition, gross pallor, edema, pain lower abdomen and weakness of 03 weeks duration. Since birth, she had alopecia with sparse eyebrow. There was no significant past history or similar family history. Routine laboratory examinations showed anemia with hemoglobin 5.6 gm/dl along with raised serum creatinine of 4.2 mg/dl. On ultrasound abdomen a heteroechoic mass was noticed, while computerized tomography scan revealed a large 9.4 x 7.5 x 6.2 cm polypoidal mass that occupied the entire urinary bladder. Right and left kidneys showed mild hydronephrosis. Subsequent cystoscopic examination confirmed the same. The patient was offered bilateral percutaneous nephrostomy followed by partial cystectomy with 1 cm clearance around the mass.. Peroperative examination also showed a large spherical mass of 10 x 6 x 6 cm arising from the left lateral wall of the urinary bladder about 2cm from the left vesicoureteric junction with a base of about 4cm.

On gross examination, the polypoidal bladder lump specimen measured 10 x 6 x 6 cm, and was firm in consistency, pinkish white in colour (Fig 1). It was gritty to cut and cut surfaces appeared homogenous grayish white. The base or stalk of the mass was received as four pinkish white to light brown tissue masses ranging from 6x3x2 cm to 1.5x1x1cm along with haemorrhagic clot.

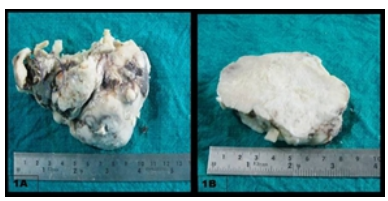

Figure 1. A. Partial cystectomy specimen with a polypoidal growth in the bladder lumen B. Cut section of bladder mass.

Histopathological examination from the bladder mass revealed a highly cellular and irregular spindle cell tumor with diffuse inflammatory mononuclear cell infiltrates comprising of lymphocytes and plasma cells infiltrating the muscular layer with a focal myxoid stroma. The spindle shaped cells displayed a high N:C ratio, oval to elongated mildly pleomorphic hyperchromatic nuclei, prominent nucleoli, and moderate amount of eosinophilic cytoplasm. No increased mitosis or necrosis was seen. The sections from the base of the mass showed similar histological picture indicating deep penetration of bladder wall. Morphologic diagnosis of low grade spindle cell neoplasm was offered. Further immunohistochemical examination displayed these tumors cells to be immunopositive for vimentin, smooth muscle actin (SMA) and muscle specific actin (MSA). Ki 67 index was very low. A few small, slit like vessels showed CD34 immunopositivity. A final confirmatory diagnosis of inflammatory myofibroblastic tumour was made (Fig 2). During subsequent 3 monthly followup, the patient was observed to have recurrence on CT urography.

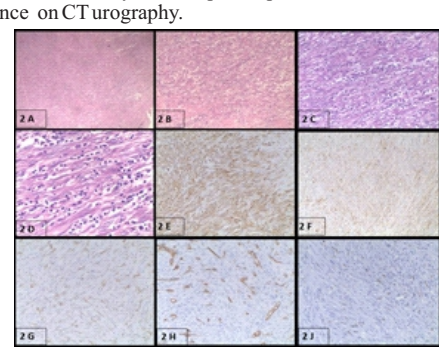

Figure 2. Inflammatory myofibroblastic tumour (A:H & Ex4; B:H&Ex10; C:H&Ex40; D:H&Ex100); Tumour diffusely positive for vimentin, MSA and SMA(E:Vimentinx40; F: MSAx40; G:SMAx40) while CD34 highlighted the slit-like vessels (G:CD34x40), and proliferation by Ki67 was low (J :Ki67x40) vessels showed slit.

DISCUSSION

We IMT is an unusual spindle cell neoplasm of the urinary bladder characterized by atypical spindle cell proliferation along with mononuclear inflammatory cell infiltrate primarily composed of lymphocytes and plasma cells and slit like vessels. Roth reported the first case in 1980 [1]. IMT has been variably described as pseudosarcomatous myofibroblastic proliferation, inflammatory pseudotumor, pseudosarcomatous fibromyxoid tumor and nodular fascitis. However, recently all these myofibroblastic group of lesions have collectively been redesignated as low grade myofibroblastic proliferation [2].

Though IMT may affect any age group, but genitourinary IMT is more common in the fourth to fifth decades of life with a mild male predilection, unlike the index case who was a female in her second decade (M:F ratio 1.33:1) [3]. IMT has been described in a wide variety of anatomical sites, including lung, soft tissues, retro peritoneum, and bladder. In genitourinary tract the most common site is urinary bladder with hematuria being the most common clinical manifestation, similar to the current case [4].

The origin and nature of IMT is controversial. A recent report favoured neoplastic nature owing to its congenital clonality, aggressive behavior and involvement of chromosome 2p23 [5].

IMT is notorious for exhibiting various morphologic and immunohistochemical similarities with other malignant spindle cell tumors of the urinary bladder thus creating a diagnostic dilemma. Morphologic similarities may mimic sarcomatoid carcinoma, rhabdomyosarcoma and leiomyosarcoma [6]-[7]. Histological features strongly favouring IMT include presence of inflammatory cell infiltrate, myxoid stroma, spindle cell proliferation, presence of stellate cells, lymphoplasmacytic infiltrates, scattered mitoses in haphazardly distributed myxoid stroma, absence of atypical mitoses and invasion into muscularis propria. Although necrosis is described in 30% or more of IMTs, the presence of tumor–detrusor muscle interface necrosis in muscle invasive cases was one criterion of sarcoma that distinguished it from IMT [8]. Immunohistochemical staining may demonstrate positivity for anaplastic lymphoma kinase, vimentin, SMA, and cytokeratin [3],[5]-[8]. Anaplastic lymphoma kinase has been described as a good marker for IMT [9]. Myogenin is a potent marker for rhabdomyosarcoma which helps in exclusion of this tumors [10]. Because of its highly cellular nature and aggressive behaviour , it can be confused with malignancy.[5] Initial biopsy and complete histopathologic examination is recommended where complete resections are problematic. Whole surgical resection is performed to avoid local recurrence [3].

To conclude, the aim of this case report was to document an unusual presentation in the form of deep bladder muscle invasion of urinary bladder IMT, which is a rare tumour in an uncommon site having a distinctive presentation with intermediate malignant potential. The report also highlights an economical IHC panel for resource constraint setups, which will be helpful in distinguishing IMT from its mimics in the urinary bladder.

REFERENCES:

[1] Roth, J. (1980). Reactive pseudosarcomatous response in urinary bladder. Urology, 16(6), 635-637.

[2] Alquati, S., Gira, F., Bartoli, V., Contini, S., & Corradi, D. (2013). Low-Grade Myofibroblastic Proliferations of the Urinary Bladder. Archives Of Pathology & Laboratory Medicine, 137(8), 1117-1128.

[3] Alderman, M., & Kunju, L. (2014). Inflammatory Myofibroblastic Tumor of the Bladder. Archives Of Pathology & Laboratory Medicine, 138(10), 1272-1277.

[4] Lott, S., Lopez-Beltran, A., MacLennan, G., Montironi, R., & Cheng, L. (2007). Soft tissue tumors of the urinary bladder, part I: myofibroblastic proliferations, benign neoplasms, and tumors of uncertain malignant potential. Human Pathology, 38(6), 807-823.

[5] Harik, L., Merino, C., Coindre, J., Amin, M., Pedeutour, F., & Weiss, S. (2006). Pseudosarcomatous Myofibroblastic Proliferations of the Bladder. The American Journal Of Surgical Pathology, 30(7), 787-794.

[6] Montgomery, E., Shuster, D., Burkart, A., Esteban, J., Sgrignoli, A., & Elwood, L. et al. (2006). 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. The American Journal Of Surgical Pathology, 30(12), 1502-1512.

[7] Weiss, S. (2007). Pseudosarcomatous Myofibroblastic Proliferations of the Bladder: A Clinicopathologic Study of 42Cases. The American Journal Of Surgical Pathology, 31(4), 642.

[8] Iczkowski, K., Shanks, J., Gadaleanu, V., Cheng, L., Jones, E., & Neumann, R. et al. (2001). Inflammatory Pseudotumor and Sarcoma of Urinary Bladder: Differential Diagnosis and Outcome in Thirty-Eight Spindle Cell Neoplasms. Modern Pathology, 14(10), 1043-1051.

[9] Sukov, W., Cheville, J., Carlson, A., Shearer, B., Piatigorsky, E., & Grogg, K. et al. (2007). Utility of ALK-1 protein expression and ALK rearrangements in distinguishing inflammatory myofibroblastic tumor from malignant spindle cell lesions of the urinary bladder. Modern Pathology, 20(5), 592-603.

[10] Cessna MH, Zhou H, Perkins SL, Tripp SR, Layfield L, Daines C, et al. (2001). Are Myogenin and MyoD1 expression specific for Rhabdomyosarcoma? Am J Surg Pathol, 25,1150–1157.