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Radical cystectomy for management of inflammatory myofibroblastic tumor of the urinary bladder: A rare case report

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ABSTRACT

Inflammatory myofibroblastic tumor (IMT) of the bladder also known as pseudosarcomatous myofibroblastic proliferation is a rare lesion in the genitourinary tract was first described in 1980 by Roth. We herein report a case of urinary bladder IMT in a 42-year-old female patient presented with gross hematuria. The hematuria was difficult to control and radical treatment has been done. Monitoring for recurrence is strongly advisable.

Introduction

Inflammatory myofibroblastic tumor (IMT) of the bladder is a rare lesion in the genitourinary tract and consists of several inflammatory cells and myofibroblastic spindle cells.[1] Other malignant spindle cell tumors including sarcomatoid carcinoma and leiomyosarcoma has to be considered in the differential diagnosis of IMT. IMT etiology remains uncertain. Several parts of the body are involved, including lung, liver, oral cavity and soft tissue and symptoms are dependent on tumor location. Most of patients with IMT of the bladder can develop several symptoms, including hematuria, irritative voiding, and lower abdominal pain.[2] The reported cases are young with both male and female predominance. Herein, we report on a patient with IMT, who was treated with radical cystectomy.

Case presentation

A 42-year-old female presented with a 6-month history of hospitalization and medical treatment due to refractory gross hematuria with occasional presence of clotted blood in the urine. At the time of admission, she had severe hematuria, clot retention and over-distended bladder (Hb:7 gr/dl, Cr: 3 mg/dl). It was impossible to remove the blood clots within the bladder through a Foley catheter. The patient underwent cystoscopy for two times to evacuate the blood clots. Due to severe hematuria and the presence of large, organized blood clots within the bladder the bladder wall was unremarkable.

Due to the inability to remove blood clots and bleeding control, the patient underwent open surgery and the clots were removed from the bladder. There were 3 large hemorrhagic masses located at the dome of the urinary bladder, at the left lateral wall, and at the interior wall which were resected (Fig. 1). The partial cystectomy was performed and patient was discharged after 3 days in a good condition with follow up recommendation.

Post-surgery pathology results reported IMT. However, six month after the surgery, the patient was readmitted for hematuria recurrence, blood clot excretion, and a tangible mass in the pelvic area. In the CT scan, involvement of the bladder wall and adjacent tissues (muscle, abdominal rectus fascia), and bilateral lymphadenopathy were observed. The patient underwent open radical cystectomy (ORC) with pelvic lymph node dissection (PLND) and ileal conduit. Microscopic examination of surgical specimens has shown infiltration of a neoplasm composed of mainly spindle cells some with bland nuclear morphology and some with nuclear atypia arranged in fascicles in vascular myxoid edematous background. Mixed population of inflammatory cells composed of lymphocytes, plasmacells, eosinophils and neutrophils was present between neoplastic cells. In some foci tumoral cells were large and epithelioid and mitotic activity was increased. Foci of tumor necrosis were also noted. On Immunohistochemistry study tumoral cells were positive for Vimentin, smooth muscle actin and ALK. Desmin and H. caldesmon were negative. Considering foci of sarcomatoid features and for excluding undifferentiated/sarcomatoid carcinoma, CK AE1/AE3, EMA,HMWK and P63 were requested in several blocks and all showed negative results.KI67 proliferative activity was about 30% in hotspot areas. Based on overall findings inflammatory myofibroblastic

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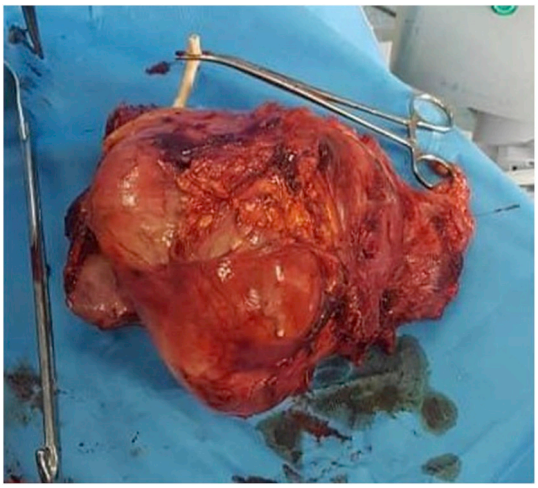

Fig. 1. The tumor which was removed from the patient's bladder.

tumor/sarcoma should be considered as the main differential diagnosis (Figs. 2 and 3).

Six month has passed since the surgery, and currently, no recurrence has been noted.

Discussion

IMT is characterized by spindle cell proliferation with inflammatory cell infiltration which can occur in any organ and equally in genders. The most common symptoms of IMT include gross hematuria and dysuria. It was presumed that multiple causes including urinary tract infection (UTIs), prior history of surgery or instrumentation, diabetes mellitus, trauma, steroid, and immune disorders can lead to this tumor.[1,2] However, our case had any underlying illness.

According to the World Health Organization (WHO) classification, IMTs are designated as tumors of intermediate biological potential due to the low risk of distant metastasis.[3] IMTs tend to have benign morphological appearances, whereas they can be aggressive. Moreover, metastatic and recurrent cases have been reported as well.[2] IMTs are occasionally hemorrhagic and are composed of spindle cells. During initial phases of the disease predominance of inflammatory components is seen. However, the mature lesions contain collagen bundles. Urinary bladder IMT may sometimes represent a diagnostic drawback. Therefore, its differential diagnosis from other malignant spindle cell tumors including sarcomatoid carcinoma and leiomyosarcoma is essential in order to provide better treatment strategies and enhanced outcomes. It seems that IMTs are reactive condition and their recurrence after complete surgical resection is unusual.[3]

Cystectomy is not routine treatment for IMT. In general, it has been recommended to resect the tumor completely to avoid local recurrence. However, there are some reports on the tumors treated by anti-inflammatory drugs without radical treatment.[3] Our case was not responded to partial cystectomy. Consequently radical cystectomy was done. Regarding the probability of recurrence, and dissimilar clinical behavior of IMT in different patients it is our opinion that total removal of the lesion and continues monitoring of patient is necessary to make a prompt and suitable decision, as presented in this paper. There is no standardized schedule concerning follow-up of these patients. However, with regard to the possibility of recurrence in 25% of cases[4] every four to six months is the recommended interval for having follow-up.

In summary, with regard to these findings, complete surgical resection and regular monitoring of clinical outcomes with routine cystoscopy and CT scans every 3–6 months are needed to ascertain the optimal consequences and to monitor the local recurrences.

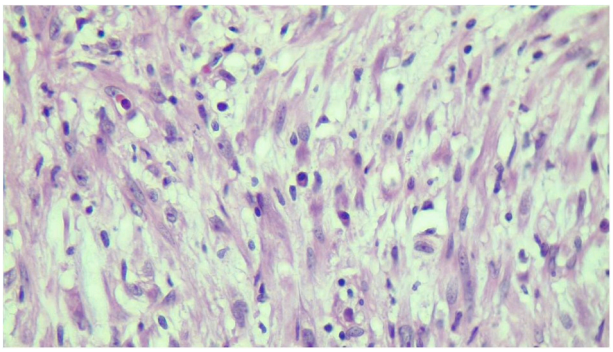

Fig. 2. Histological examination (hematoxylin-eosin staining; magnification 10× 40), showing neoplasm composed of mainly spindle cells some with bland nuclear morphology and some with nuclear atypia arranged in fascicles in vascular myxoid edematous background.

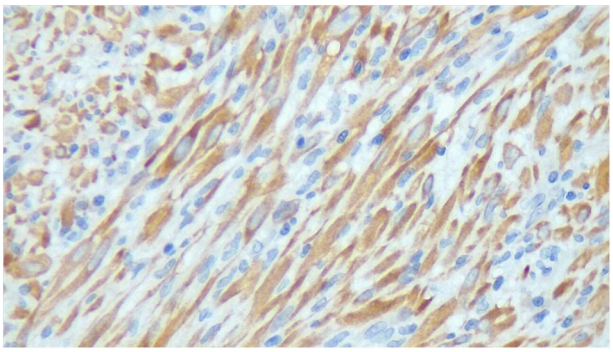

Fig. 3. Histological examination (magnification 10× 20), positive ALK-1 staining.

Conclusion

To conclude, IMTs are very rare tumors often present with unpredictable clinical behavior, requiring complete surgical resection and regular monitoring of clinical outcomes. Clinical and radiological follow-up for recurrence and metastasis monitoring is our recommendation in IMT cases. To our knowledge, this was the first report of IMT in Iran.

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None.

Conflicts of interest

There is no conflict of interest.

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