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CASE REPORT

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A rare noncancerous but life-threatening tumor in urinary bladder

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Abstract

IMT is a rare but sometimes life-threatening tumor. Although presenting with muscle invasion, local surgical resection with TURBT and close follow-up are adequate with bladder function and life quality preservation compared to partial cystectomy.

KEYWORDS

inflammatory myofibroblastic tumor, laparoscopic surgery, transurethral resection of the bladder tumor

1 | INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare benign tumor with no established treatment. Even though the presentation of massive hematuria with a critical life status, IMT can be treated safely by transurethral resection of the bladder tumor (TURBT).

An inflammatory myofibroblastic tumor is a rare benign myofibroblastic proliferation with no established treatment. It uncommonly occurs in the urinary bladder, comprising <1% of all bladder tumors and was first described by Dr JA Roth in 1980.[1] Patients with IMT often experience hematuria, lower abdomen pain, or voiding symptoms involving irritation or obstruction.[2] Although the etiology is still unknown, it is believed to be associated with trauma, previous surgery history, prior instrumentation, and steroid use.[2]

The mainstream recommended therapy for IMT originating in the bladder is local surgical resection including transurethral resection of the bladder tumor (TURBT) and partial cystectomy, although there has been limited clinical experience due to its rarity. So far, no metastases have been reported, except for recurrence in 10%-25% of the population because of incomplete tumor resection. It is important to differentiate the diagnosis from other malignant spindle cell neoplasms such as rhabdomyosarcoma or leiomyosarcoma with expression of anaplastic lymphoma kinase (ALK) detected by fluorescence in situ hybridization (FISH). Here, we describe a case of IMT of the bladder in a 74-year-old man

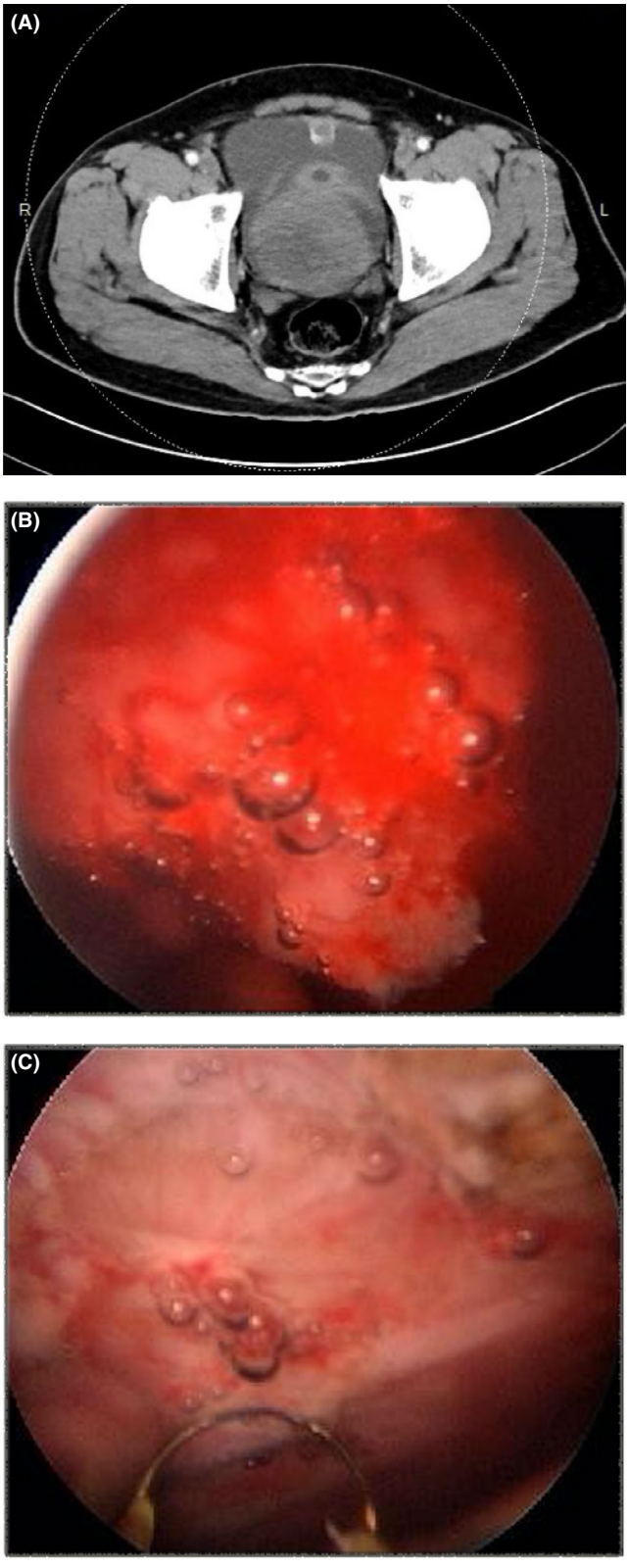

FIGURE 1 A, Enhanced computed tomography (CT) of the abdomen. CT of the abdomen showed an early enhancing bladder tumor at anterior wall with asymmetrical thickening. B and C, Intraoperative view of transurethral resection. Well-defined solid, wide base mass measuring 3 cm in size over dome to the anterior wall of the bladder

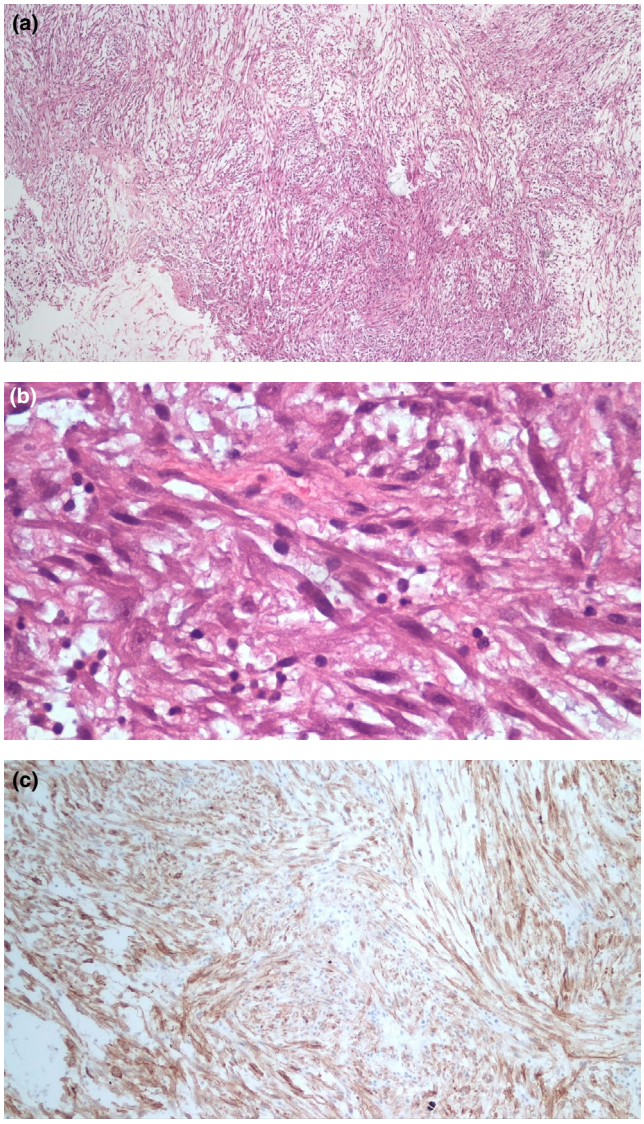

FIGURE 2 A and B, Histological examination. Tumor showing stellate cells in a myxoid background, spindle cells with a fascicular pattern, and focal cellular collagenized areas. Higher magnification revealed mild atypia without increased mitotic activity, yet it infiltrated the muscularis propria. C, The immunohistochemical staining. The immunohistochemistry study was positive for ALK-1

with critical status on arrival diagnosis treated by transurethral resection of bladder tumor (TURBT).

2 | CASE REPORT

A 74-year-old man presented to our emergency room with burning micturition and massive hematuria with blood clot for 5 days. His medical history revealed a diagnosis of depression for 10 years and regular taking of antidepressants. Blood pressure on arrival was only 72/53 mmHg, and blood profile showed anemia, with hemoglobin as low as 7.1 g/dL.

Based on enhanced computed tomography (CT) of the abdomen and pelvis, an early enhancing bladder tumor with asymmetrical thickening about 1.5 cm at the anterior bladder wall was revealed (Figure 1A). There was no lymphadenopathy or metastases of other organs.

Even though aggressive resuscitation and blood transfusion, the patient still presented with hypotension and shock status. With regard to his critical condition, urgent transurethral coagulation/resection was subsequently performed to stop tumor bleeding and remove bladder clots. Intraoperative findings showed a well-defined solid, wide base mass measuring 3 cm in size with engorged vessel located from the dome to the anterior wall of the bladder (Figure 1B). Its smooth surface was not typical of urothelial carcinoma. We resected the tumor to the depth of the muscle layer where normal muscle tissue was seen (Figure 1C). Electrocauterization was performed to seal off bleeding vessels. This surgery dramatically improved the patient's vital signs. The patient had an uneventful postoperative course and was discharged on postoperative day 3.

Final pathology of the specimen acquired via transurethral resection revealed negative surgical margins with loose stellate cells in a myxoid background, spindle cells with a fascicular pattern, and focal cellular collagenized areas. The tumor cells only showed mild cytologic atypia without increased mitotic activity, yet had infiltrated the muscularis propria. Focal areas of necrosis, which are often increased in IMTs, were also recognized in this specimen. The immunohistochemistry study was positive for ALK-1 and cytokeratins (CK) and negative for desmin, S100, and CD34 (Figure 2). The final diagnosis of IMT was supported by the immunohistochemical staining positivity of ALK-1 and overall morphology. No adjuvant was deemed necessary since IMT generally demonstrates a good clinical course even after limited resection. At 42 months (3.5 years) of follow-up, the surveillance imaging studies of pelvic CT and cystoscopy annually were free of local recurrence signs, and the patient was asymptomatic, with no voiding or storage symptoms.

3 | DISCUSSION

As depicted by spindle cell proliferation and infiltration, IMTs are categorized as intermediate tumors based on World Health Organization (WHO) soft tissue tumor classifications. It mainly occurs in the retroperitoneum, omentum, and mesentery in nearly three-quarters of cases. Nevertheless, the occurrence of IMTs of the bladder is very rare with the mean age of patients being in their twenties and gender discrepancy of being female.[3] Reviewing the literature, the trigone tends to be spared compared to the most frequent tumor in the bladder, which was urothelial carcinoma. Symptoms of IMTs of the bladder include dysuria, macrohematuria, pelvic pain, and even weight loss. Pre-operative ultrasound examinations often result in unspecific findings. CT with contrast is a widely used diagnostic imaging technique, which can present submucosal polypoid or intramural mass with sometimes peri-vesical fat being involved. To obtain a precise diagnosis, immunohistochemistry study of anaplastic lymphoma kinase (ALK) is demonstrated in a significant percentage of IMTs.[4] A positive immunostaining for ALK has highlighted the differentiation from other spindle cell proliferation tumors in up to 89% cases of IMTs.[5]

Despite being a benign lesion, IMTs may present with locally invasion and the convincing therapy of IMTs is absolute tumor surgical resection with the goal of attaining negative microscopic margins.[4] In the consideration of complete local resection, the treatment option includes TURBT and partial cystectomy. Concerning the possibility of local recurrence, partial cystectomy is often the treatment option to ensure total resection of the tumor.[2,6,7] Sofia et al have reported 2 cases treated with laparoscopic partial cystectomy to achieve total resection, and no recurrence was noted after 58 months of follow-up.[6]

However, there are no studies regarding a safety margin during partial cystectomy in cases of IMTs, and although it is required to achieve free resection margins, the benefit of negative margin resection must be weighed against the bladder capacity and function preservation in patients whose tumor invades only superficial layer and are of relatively old age. Teoh et al's have conducted a systemic review, and among the 182 patients, 60.8% were treated with TURBT and they indicated that most cases can be adequately treated with transurethral resection.[2] Gass et al, Fadaak et al, and Song et al have also reported successful treatment with TURBT with no recurrence during close follow-up. Even with incomplete resection, EA Montgomery et al have found that IMTs tend to remain stable or even spontaneously regress in nature.[4] In Table 1, we summarize some similar cases of IMTs published in the literature.

In this case, we opted not to carry out partial cystectomy due to the patient's old age and to preserve the function of the bladder. Although the histopathological report showed the tumor had infiltrated the muscularis propria, the surgical margin was tumor-free. Regarding follow-up, CT and cystoscopy were adopted annually for 3.5 years and no tumor recurrence nor lower urinary tract symptoms were noted during follow-up. Although no standardized schemes are established for follow-up, it is still important since Rosado et al have reported a recurrence rate of the 25%.[3] Accordingly, this study demonstrates a good clinical course of IMTs with infiltrating muscularis propria treated with local resection by TURBT.

TABLE 1 Clinical summary of inflammatory myofibroblastic tumor (IMT) cases

Age (years)	Sex	Lesion location	Greatest diameter of lesion (cm)	Treatment	Reference
74	M	The dome to the anterior bladder wall	3	TURBT	Our case
38	M	Right anterolateral bladder wall	3.2	Laparoscopic partial cystectomy	6
56	M	Right anterolateral bladder wall	6	TURBT + Laparoscopic partial cystectomy	6
52	M	The dome	3	TURBT + subsequent partial cystectomy	7
29	M	Left lateral bladder wall	4	TURBT + subsequent robot-assisted partial cystectomy	8
17	F	Right anterolateral bladder wall	10	TURBT + subsequent partial cystectomy	9
51	M	Right-side bladder wall	4	TURBT	10
14	F	Right posterolateral bladder wall	2	TURBT	11
28	F	Near bladder neck	4	TURBT	12

Abbreviations: F, female; M, male; TURBT, transurethral resection of the bladder tumor.

4 | CONCLUSION

In conclusion, inflammatory myofibroblastic tumor (IMT) is a relatively rare tumor that might represent a critical life status. Although presenting with muscle invasion, local surgical resection with TURBT and close follow-up are treatable options with better bladder function preservation and life quality compared to partial cystectomy.

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CONFLICT OF INTEREST

The authors declare no conflicts of interest.

AUTHORS' CONTRIBUTIONS

J-HJ: performed the surgery and reviewed the related articles. S-JG: reviewed the related articles and wrote the manuscript. C-JW: performed the surgery. S-FY: examined and interpreted the pathology. H-LK: analyzed and interpreted the patient's image of CT. Y-CL: analyzed and interpreted the patient's image of CT. C-SW: reviewed the related articles and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

ETHICAL APPROVAL

Approval for the study was obtained from the Institutional Review Board of Kaohsiung Medical University Hospital.

CONSENT FOR PUBLICATION

Informed consent was obtained from the patient for the publication of this case report.

DATA AVAILABILITY STATEMENT

The authors do not wish to share the patient's data. The privacy of this participant should be protected.

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REFERENCES

1. Roth JA. Reactive pseudosarcomatous response in urinary bladder. *Urology*. 1980;16(6):635e637.
2. Teoh JYC, Chan N-H, Cheung H-Y, Hou SSM, Ng C-F. Inflammatory myofibroblastic tumors of the urinary bladder: a systematic review. *Urology*. 2014;84(3):503-508.
3. Rosado E, Pereira J, Corbusier F, et al. Inflammatory pseudotumor of the urinary bladder. *Radiol Case J Radiol Case Rep*. 2015;9(1):36-42.
4. Montgomery EA, Shuster DD, Burkart AL, 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(12):1502-1512.
5. Lott S, Lopez-Beltran A, MacLennan GT, Montironi R, Cheng L. Soft tissue tumors of the urinary bladder, part I: myofibroblastic proliferations, benign neoplasms, and tumors of uncertain malignant potential. *Hum Pathol*. 2007;38(6):807-823.
6. Santos Lopes S, Furtado A, Oliveira R, et al. Inflammatory myofibroblastic tumor of the bladder: 2 rare cases managed with laparoscopic partial cystectomy. *Case Rep Urol*. 2016;2016:4976150. https://doi.org/10.1155/2016/4976150

7. Etani T, Naiki T, Nagai T, et al. Inflammatory myofibroblastic tumor of the urinary bladder: a case report. *Case Rep Oncol*. 2016;9(2):464-469. https://doi.org/10.1159/000448550
8. Rotenberry C, Dowd K, Russell D, DeRiese W, Teeple S, Cammack T. Robot-assisted partial cystectomy for treatment of inflammatory myofibroblastic tumor of the bladder. *Urol Case Rep*. 2017;11:25-27. https://doi.org/10.1016/j.eucr.2016.11.014
9. Kato M, Masui S, Kanda H, Arima K, Shiraishi T, Sugimura Y. Successful preservation of the bladder in a case of inflammatory myofibroblastic tumor with the diagnostic efficacy of ALK/p80 immunohistochemistry and fish analysis: case report and review of the literature. *Urol Case Rep*. 2017;11:19-21. https://doi.org/10.1016/j.eucr.2016.11.018
10. Gass J, Beaghler M, Kwon M. Inflammatory myofibroblastic tumor of the urinary bladder: a case report. *J Endourol Case Rep*. 2019;5(2):31-33. https://doi.org/10.1089/cren.2018.0096
11. Fadaak K, Al-Otaibi A, Al-Zahrani A, et al. Transurethral resection for the treatment of an inflammatory myofibroblastic tumor of the urinary bladder: a case report. *Case Rep Oncol*. 2019;12(2):344-353. https://doi.org/10.1159/000500503
12. Song D, Jiao W, Gao Z, et al. Inflammatory myofibroblastic tumor of urinary bladder with severe hematuria: a Case report and literature review. *Medicine (Baltimore)*. 2019;98(1):e13987. https://doi.org/10.1097/MD.0000000000013987

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