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Metastatic inflammatory myofibroblastic tumor of the bladder

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ABSTRACT

A 61-year-old male presented with gross hematuria and transurethral resection of bladder tumor revealed inflammatory myofibroblastic tumor (IMT). Due to extent of disease leading to ureteral obstruction and hydronephrosis, radical cystectomy (RC) with ileal conduit urinary diversion was performed. Five months after RC, the patient presented with decreased urine output. Exploratory laparotomy revealed mass in right colon and right hemicolectomy revealed metastatic IMT to the bowel and pericolonic fat. To our knowledge, this is the first report of primary IMT of the bladder metastasizing to other organs.

Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare spindle-cell tumor that has been identified in various organ systems.[1–4] In the genitourinary (GU) tract, it can involve the kidneys, ureters, urethra, and rete testis, but most commonly involves the bladder.[1,2] The pathogenesis of IMT is unclear.[2,4] A 2–3:1 male predominance exists.[1,2] Tumors have been found in all ages and have been reported from 2.5 months to 89 years.[1,2]

Clinically, IMT causes site-specific and systemic symptoms including fever, weight loss, and pain.[1,2,4] However, these systemic symptoms are characteristically absent for tumors in the GU tract.[2] The most common presenting symptom for IMT in the bladder is painless hematuria; however, presentation may include dysuria, pelvic pain, or urinary tract infection.[1,2,4] IMT can become very large, leading to bladder outlet obstruction and hydronephrosis.[1,2,4] While these tumors are locally aggressive, to our knowledge, no case series has identified metastasis of typical IMT.[1–4] We present a case of bladder IMT with tumor recurrence and metastasis following presumed complete resection.

Case presentation

A 61-year-old man with no significant past medical history presented to the urology clinic with 6-month history of gross hematuria. Cystoscopy revealed a large, nodular mass arising from the bladder floor and involving the left ureteral orifice. Staging computed tomography (CT) revealed left-sided hydronephrosis, atrophic left kidney and tumor in the bladder. Transurethral resection of bladder tumor (TURBT) was performed. Due to tumor burden, the patient underwent radical cystectomy (RC) with ileal conduit urinary diversion. The patient had an uneventful hospital course and was discharged to home on day eight after surgery.

Histologic examination of the cystoprostatectomy specimen demonstrated whorls of spindle cells as shown in Fig. 1A–B. There was mild nuclear pleomorphism with less than one mitosis per ten high-power fields. Infiltration of tumor cells through the muscularis propria and into the perivesical fat, prostate and seminal vesicles was identified. Immunohistochemistry (IHC) revealed expression of CD68, CAM5.2 and smooth muscle actin by the tumor cells. The tumor cells were negative for DOG-1, CD117, A103, Alk-1 and S-100 protein (Fig. 2). The tumor was diagnosed as inflammatory myofibroblastic tumor (IMT) of the bladder.

After discharge, the patient was admitted to the hospital for acute renal insufficiency. He developed prolonged ileus and exploratory laparotomy was performed. Tumor was identified in the pelvis and studding the peritoneal cavity. Obstruction of the rectum by tumor resulted in requirement of a right hemicolectomy and diverting

Abbreviations: IMT, inflammatory myofibroblastic tumor; GU, genitourinary; TURBT, Transurethral Resection of Bladder Tumor; RC, Radical Cystectomy; IHC, immunohistochemistry; CT, computed Tomography

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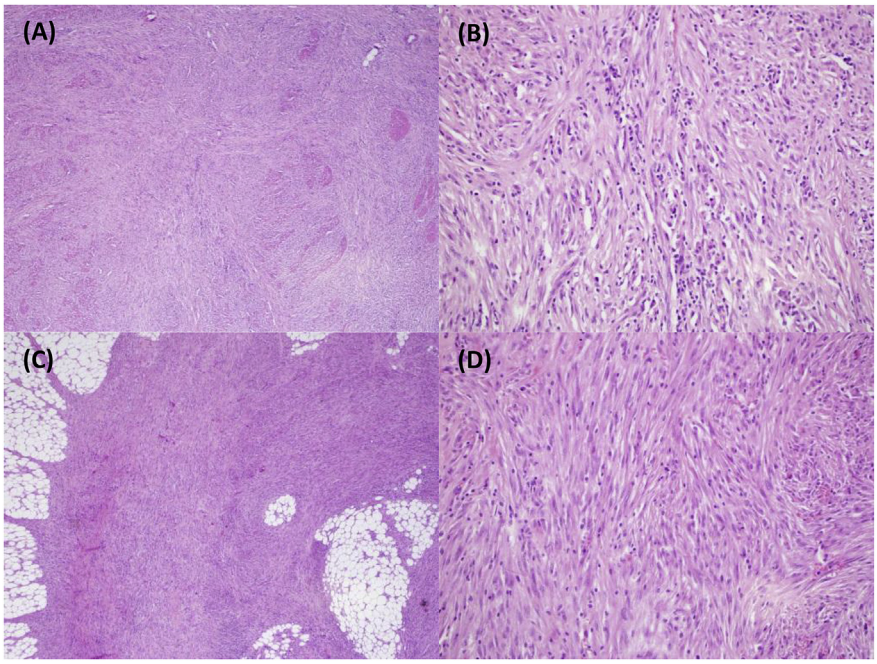

Fig. 1. Light microscopy **(A) Tumor of bladder.** Hematoxylin and eosin (H&E) stain, × 4 magnification, shows whorls of spindle cells **(B) Tumor of bladder.** H&E stain, × 20 magnification, shows cells have eosinophilic cytoplasm, indistinct nuclear borders and mild nuclear pleomorphism **(C) Tumor of ileum.** H&E stain, × 4 magnification, shows similar architecture to (A) **(D) Tumor of ileum.** H&E stain, × 20 magnification, shows similar cytologic features to (B).

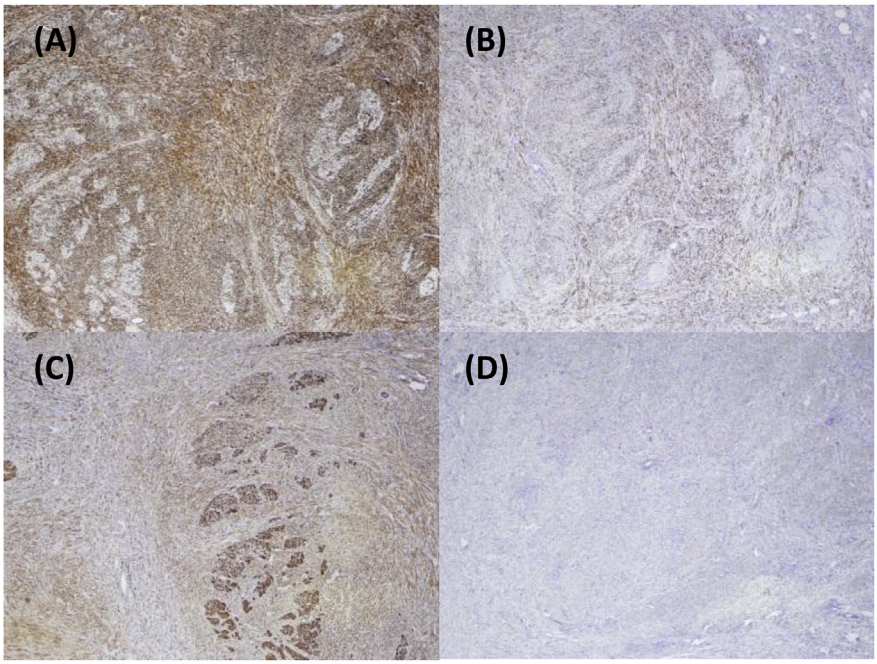

Fig. 2. Tumor of bladder, light microscopy **(A)** Vimentin cytoplasmic marker, × 4 magnification, expressed by tumor cells **(B)** Cytokeratin CAM5.2 cytoplasmic marker, × 4 magnification, expressed by tumor cells **(C)** Smooth muscle actin cytoplasmic marker, × 4 magnification, expressed by tumor cells **(D)** S-100 cytoplasmic and nuclear marker, × 4 magnification, not expressed by tumor cells.

ileostomy.

Pathologic examination of the bowel demonstrated similar gross and histologic features (Fig. 1C–D) to the bladder tumor involving the bowel wall and invading the pericolonic fat. The patient was diagnosed with IMT metastatic to the peritoneum and large intestines. He ultimately succumbed to his disease 3 weeks after this operation.

Discussion

The clinical presentation of IMT is similar to leiomyosarcoma, low-grade sarcomas, and rhabdomyosarcoma.[1,4] IHC helps differentiate IMT from these malignant neoplasms. IMT expresses vimentin, smooth muscle actin and CK18 in up to 100% of cases.[1,2,4,5] FISH can be performed to confirm IHC findings but was not done in this particular case. Additionally it commonly displays muscle-specific actin, desmin, CAM5.2, and cytokeritins.[1,2,4,5] Although ALK-1 expression is correlated with ALK rearrangement in these tumors and is expressed by 50–60% of IMT, it is rare to have an ALK rearrangement in IMT diagnosed in adults older than 40 years of age.[5] IMT does not express S100 or MyoD1, which are characteristically found in rhabdomyosarcomas.[2] Histologically, IMT has less nuclear and cellular atypia and fewer mitotic figures when compared to these other malignant lesions.[2,3,5] The pathologic findings in this case were consistent with IMT rather than sarcoma.

There is some controversy regarding the metastatic potential of IMT of the bladder. Some describe it as a distinct, benign neoplasm while others argue that it exists on a continuum of neoplasms ranging from benign to malignant.[1–4] Only one prior case has described metastasis of bladder IMT. However, this occurred in a patient who had received prior pelvic radiation making the distinction between post-irradiation

sarcoma versus primary metastatic IMT unclear.[1] Other case series reviewed identified no cases of metastasis with typical IMT.[1–4]

Regardless of malignant potential, IMT are often locally aggressive with invasion of the muscularis propria in up to 77% of cases. Tumors have been found to invade surrounding structures in the pelvis, including the prostate.[1–3] The risk for local recurrence is as high as 10–25%; however previously it was thought to be uncommon following complete resection.[1–4]

Due to the local aggressive nature and uncertain malignant potential, clinical judgement must be utilized to determine the proper course of treatment. IMT of the bladder can be managed conservatively with local resection or more aggressively with partial or complete cystectomy.[1–3] There is little evidence supporting the use of chemotherapy and radiation in the treatment of these tumors.[3] This case further demonstrates the invasive nature of the tumor, and highlights that local recurrence, multifocality or even distant metastases is a possibility despite complete resection.

Conclusion

IMT is a rare spindle cell tumor. While historically considered benign, this case demonstrates that IMT does have malignant potential and can have a poor prognosis. As such, consideration of more aggressive surgical approaches and close follow up is warranted.

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Appendix A. Supplementary data

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