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Oncology

A rare case of inflammatory myofibroblastic tumor of the bladder with local invasion of the cervix and right ovary

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ABSTRACT

Inflammatory myofibroblastic tumors (IMT) are rare spindle cell neoplasms derived from mesenchymal cells. Primary genitourinary IMTs share morphological and molecular features with various malignant spindle cell sarcomas, which introduces a diagnostic challenge. We present the case of a 50-year-old female who was referred for evaluation of hematuria and nonspecific urinary symptoms and was found to have a mass originating from the urinary bladder that involved the cervix and right ovary. Transurethral resection of bladder tumor (TURBT) and immunohistochemical analysis revealed an IMT. To our knowledge, this is the first documented case of primary genitourinary IMT with cervical and ovarian involvement.

1. Introduction

Inflammatory myofibroblastic tumors (IMT) are rare spindle cell neoplasms of mesenchymal origin.[1] They are generally considered benign tumors with a variable rate of local recurrence depending on the anatomical site.[2] IMTs are most frequently found in the lungs, abdominopelvic cavity, and retroperitoneum.[1–3] Involvement of the genitourinary system is uncommon. While the urinary bladder has been reported to be the most common site of origin within the genitourinary tract, extravesical invasion of surrounding structures is rare.[4] Furthermore, to our knowledge, no reports have documented cases of primary IMTs of the urinary bladder with invasion of the cervix and ovaries. We present a case of a 50-year-old female with a large IMT of the urinary bladder with both cervical and ovarian involvement.

2. Case presentation

A 50-year-old female with a history of hematuria, abdominal pain, urinary retention, and recurrent urinary tract infections presented for evaluation of a right adnexal mass discovered on ultrasound. A physical exam of her pelvis revealed fullness in the right adnexa. Initial laboratory studies for CEA, CA 19–9, and CA-125 were within normal limits. A CT abdomen/pelvis showed a 10.4 × 10.3 × 6.8 cm cystic mass involving the urinary bladder, anterior aspect of the cervix, and possibly the right ovary. Cystoscopy revealed a highly vascular, large, nodular mass emanating from the posterior bladder wall without involvement of the ureteral orifices. Preoperative MRI confirmed the CT findings (Fig. 1). A limited transurethral resection of bladder tumor (TURBT) was performed for tissue sampling. Grossly, the tumor was tan-brown in color with multiple unoriented, friable portions of soft tissue and no discrete areas of variegation or discoloration. Histologic examination revealed edematous spindle cell stroma changes and ulceration associated with prominent vascular proliferation (Fig. 2). Immunohistochemical analysis for anaplastic lymphoma kinase (ALK) rearrangement was positive. Together, these findings support the diagnosis of an inflammatory myofibroblastic tumor. Preoperative IR embolization was performed with the goal of debulking the tumor and improving the odds of organ preservation at the time of extirpation. Unfortunately, the size of the mass did not significantly decrease after embolization. A robot-assisted laparoscopic partial cystectomy, right pelvic node dissection, total abdominal hysterectomy, and bilateral salpingo-oophorectomy (TAH-BSO) was performed with grossly negative margins. No intraoperative and postoperative complications were noted. Surveillance cystoscopy and imaging with serial MRI are negative for recurrence with one year of postoperative follow-up.

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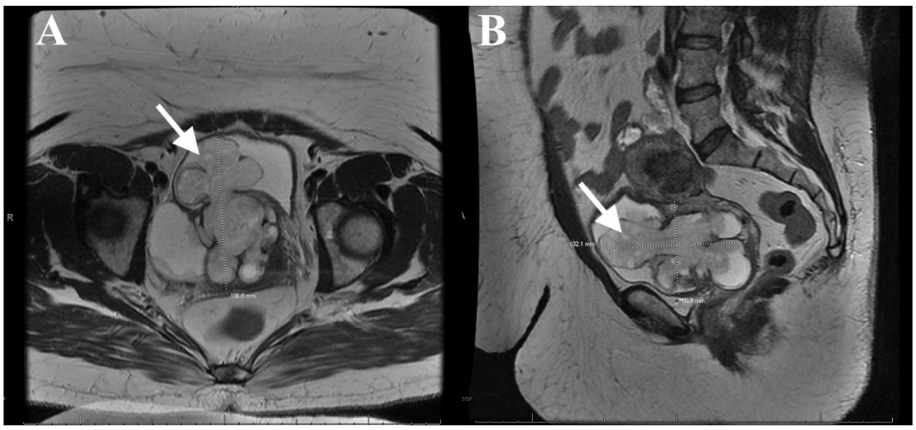

Fig. 1. (A) Transverse and (B) sagittal T2-weighted MRI images demonstrating inflammatory myofibroblastic tumor of the urinary bladder (solid white arrow).

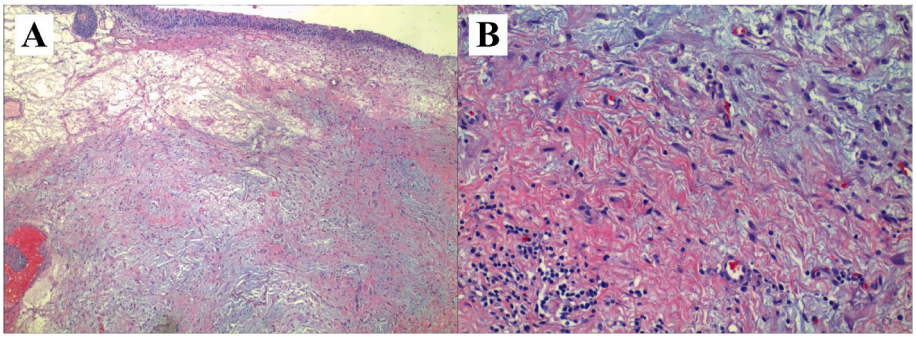

Fig. 2. (A) Low power hematoxylin and eosin stain (H&E, x4) of an inflammatory myofibroblastic tumor within lamina propria, with overlying benign urothelium. (B) High power hematoxylin and eosin stain (H&E, x20) showing myofibroblasts and inflammatory infiltrate.

3. Discussion

Inflammatory myofibroblastic tumors have the potential to arise anywhere in the body, with a preference for the visceral organs and somatic soft tissue structures.[2] IMTs frequently originate in the lungs, and less commonly in the retroperitoneal and abdominopelvic cavities. Inflammatory myofibroblastic tumors of the urinary bladder (IMTUB) are rare, and most frequently occur in young adults.[5] While localized involvement of adjacent structures including the prostate and seminal vesicles has been reported, extravesical invasion of the cervix and ovaries has not previously been documented.[4]

The first case of IMTUB was reported in 1980.[5] In the most recent systemic review by Toeh et al., 182 cases of IMTUB were identified with a median age of diagnosis of 38.9 years. IMTUBs present with a variety of non-specific symptoms with hematuria being the most common, followed by dysuria, lower abdominal pain, and flank pain. In rare cases, hemodynamic instability, anemia, and renal insufficiency have been identified on laboratory analysis. Within the bladder, IMTs primarily originate from the posterior wall, dome, and right lateral wall.[5] While IMTUBs are generally benign, as many as 60% exhibit local spread to the muscularis propria.[2,5] However, there have been few cases of extravesical invasion, and only one documented case of distant metastasis.[4] Of these cases, to our knowledge none have reported invasion to the cervix or ovaries.

IMTs appear grossly as a white or tan mass with a fleshy, firm, or gelatinous composition.[2] On histologic examination, IMTs consist of spindle cells with varying densities of inflammatory cell infiltrates arranged in myxoid, compact, or hyalinized patterns.[1,3] Myxoid and compact refer to loosely and densely arranged spindle cell formations, respectively. This is in contrast to a hyalinized organization, which contains elongated spindle cells in addition to sparse cellular collagen.[1,2] IMTs generally express smooth muscle actin, calponin, and desmin.[2–4] This presents a diagnostic dilemma due to lack of specificity of these markers in differentiating IMTs from other malignant spindle cell neoplasms such as leiomyosarcoma and rhabdomyosarcoma.[2–4] One biomarker which can be used to distinguish IMTUB from other spindle cell neoplasms is the anaplastic lymphoma kinase (ALK) locus translocation on 2p23. ALK translocations have been found in 50–60% of IMT cases.[3] This marker is generally not seen in other spindle cell neoplasms.

Given that IMTUBs are generally benign with minimal invasion of surrounding tissue, surgical resection is the preferred method of treatment.[3] The most common surgical approaches include TURBT (60.8%) followed by partial (29.2%) and radical cystectomy (9.2%).[5] Prognosis following treatment is generally very good, with a recurrence rate of 4%.[5] Most patients remain disease free on subsequent surveillance.

4. Conclusion

IMTUBs are rare, benign spindle cell neoplasms with a low metastatic potential. Local invasion, while rare, can pose a substantial risk to surrounding structures. To our knowledge, we present here the first documented case of IMTUB with extravesical involvement of the cervix and ovaries.

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Consent

Written informed consent was obtained from the patient.

Declaration of competing interest

The authors declare no conflict of interest.

References

1. Bennett JA, Nardi V, Rouzbahman M, Morales-Oyarvide V, Nielsen GP, Oliva E. Inflammatory myofibroblastic tumor of the uterus: a clinicopathological, immunohistochemical, and molecular analysis of 13 cases highlighting their broad morphologic spectrum. *Mod Pathol.* 2017;30(10):1489–1503. https://doi.org/10.1038/modpathol.2017.69.
2. Gleason BC, Hornick JL. Inflammatory myofibroblastic tumours: where are we now? *J Clin Pathol.* 2008;61(4):428–437. https://doi.org/10.1136/jcp.2007.049387.
3. Laylo JCV, Lim NL, Remo JJV. Inflammatory myofibroblastic tumor of the urinary bladder: a prognostically favorable spindle cell neoplasm. *Urology Case Rep*. 2021;34, 101474. https://doi.org/10.1016/j.eucr.2020.101474.
4. Libby EK, Ellis LT, Weinstein S, Hammer RD, Murray KS. Metastatic inflammatory myofibroblastic tumor of the bladder. *Urology Case Reports*. 2019;23:10. https://doi.org/10.1016/j.eucr.2018.11.007.
5. Teoh JYC, Chan NH, Cheung HY, Hou SSM, Ng CF. Inflammatory myofibroblastic tumors of the urinary bladder: a Systematic review. *Urology*. 2014;84(3):503–508. https://doi.org/10.1016/j.urology.2014.05.039.

Abbreviations

IMTUB: Inflammatory myofibroblastic tumor of the urinary bladder
IMT: Inflammatory myofibroblastic tumor
ALK: Anaplastic lymphoma kinase
TURBT: Transurethral resection of bladder tumor
TAH-BSO: Total abdominal hysterectomy with bilateral salpingo-oophorectomy