Immunopathologia Persa
http www.immunopathol.com
DOI:10.34172/ipp.2022.148

Inflammatory myofibroblastic tumor of the urinary bladder; a case report

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Received 4 Aug. 2019
Accepted 10 Nov. 2019
Published online 29 Aug. 2020

Keywords: Myofibroblastic tumor, Urinary bladder, Neoplasm

Abstract

Inflammatory myofibroblastic tumor (IMT) is a rare neoplasm exhibiting local aggressive behavior and needs to be differentiated from sarcoma and sarcomatoid carcinoma in particular. Its exact etiology is uncertain. IMT is more common among children and young adults. Pulmonary, gastrointestinal, genitourinary, retroperitoneal sites are frequently affected. History, physical examination, laboratory workup and imaging modalities have limited diagnostic value. IMT has a tendency for local recurrence. Here, we report a case of an IMT of the bladder in a female patient with difficulty urinating, dysuria and pelvic pain.

Citation: Benlier N, Solakhan M, Sever ÖN, Gürbüz H, Yıldırım M. Inflammatory myofibroblastic tumor of the urinary bladder; a case report. Immunopathol Persa. 2022;x(x):e148. DOI:10.34172/ipp.2022.148.

Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare neoplasm. Currently, this tumor is considered as a locally aggressive neoplasm associated with certain genetic features; autoimmune disorders and infectious agents have also been implicated (1-5). IMT of the urinary bladder is a proliferative mesenchymal lesion. Histopathologically, it was first described by Roth in 1980 as an unusual pseudosarcomatous entity in the bladder of a female (6). Apart from the urinary bladder, IMT has been reported in vagina, prostate and urethra. Epithelial neoplasms with marked spindle cell proliferation have been reported in the skin, nasopharynx, larynx, salivary glands, oral cavity, thyroid, thymus, lungs, gastrointestinal tract, liver, gallbladder, pancreas, kidneys, breast, uterus and ovaries (7,8). Although IMT is benign clinically, it resembles sarcoma histopathologically. Since it can mimic sarcoma, clinical findings as well as histopathological, histochemical, immunohistochemical and ultrastructural studies are critical for differential diagnosis (8,9). Immunohistochemically, epithelial membrane antigen, cytokeratin, vimentin, actin and desmin are particularly important (7,10-12).

While IMT is common in childhood, adults less than 40 years of age are mostly affected and its prevalence is equal in men and women. It may be asymptomatic but depending on the anatomic location of the mass, symptoms may occur including cough, weight loss, urinary tract infection, fever and fatigue (9). On physical examination, the mass may be palpable. IMT is often detected incidentally during imaging studies. Ultrasonography (USG), computed tomography (BT) and magnetic resonance imaging (MRI) may be used to detect the mass and differentiate it from other mass lesions but may not show typical findings in imaging (2-5,13). Treatment requires complete tumor resection with tumor-free surgical margins. Close postoperative follow-up is needed for IMT cases with a risk of recurrence (4,5,14). A multidisciplinary approach for diagnosis, management and follow-up of IMT involving specialists from surgery, radiology and pathology clinics is crucial not only to reduce unnecessary radical surgical interventions

Key point

- We hereby report a case of an inflammatory myofibroblastic tumor of the bladder with difficulty urinating, dysuria and pelvic pain.
- Inflammatory myofibroblastic tumor is an uncommon lesion of unknown cause.
- Differential diagnosis should include sarcomas.

but also to prevent tumor recurrence (2-5). In this case report, we aimed to present a case of an IMT of the bladder in a female patient with pelvic pain, dysuria and straining to urinate along with relevant literature data.

Case Presentation

A 27-year-old woman admitted to our hospital with complaints of difficulty urinating, straining to urinate, and burning sensation with urination and occasional hematuria. Her physical examination and personal history were unremarkable. Complete blood count, routine biochemical investigations and urinalysis were normal. Computed tomography (CT) imaging showed a solid mass extending from the bladder neck to the dome (Figure 1A and 1B). The tumor was diagnosed as an IMT based on immunohistochemical and histological findings (Figures 2A-2E). Immunohistochemical staining showed negative immune reactions to S-100, CD68, p63 (in 2 separate blocks), pan-cytokeratin (in 2 separate blocks), myogenin and CD117, positive immune reactions to vimentin, actin and desmin and weak positive immune reactions to CD56. Ki-67 proliferative index was approximately 2%. The patient underwent endoscopic tumor resection (Figure 1C) and was discharged on first postoperative day.

Discussion

IMT is a benign rare lesion which usually develops in children and young adults (3-5). Clinical manifestations mostly include hematuria, pain and dysuria and some patients may experience systemic symptoms such as fever, weight loss and asthenia. Radiographic findings are variable and non-specific; therefore, differential diagnosis is needed to exclude malignant lesions (3).

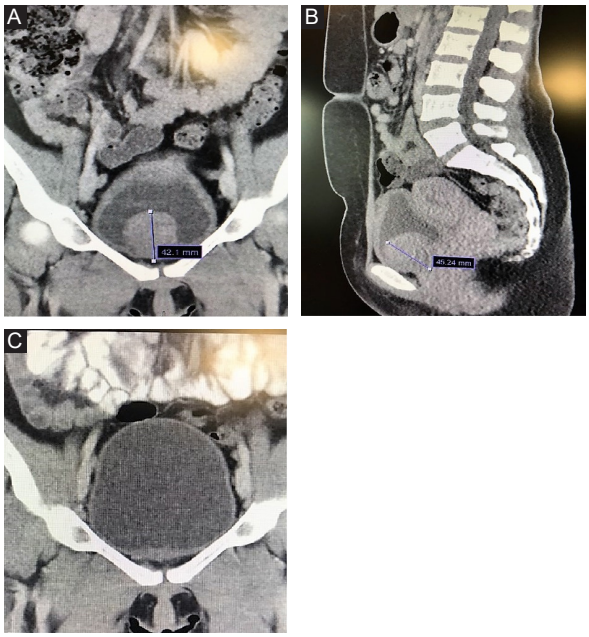

Figure 1. (A) Computed tomography image of the mass lesion in coronal plane. (B) Computed tomography image of the mass lesion in sagittal plane. (C) Postoperative computed tomography image of the mass lesion (coronal plane).

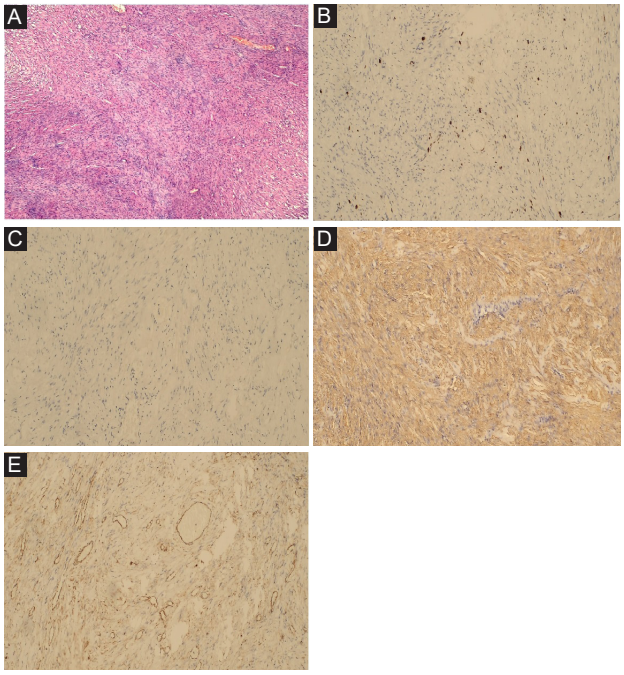

Figure 2. (A) Fascicles of spindle cells embedded in fibrous stroma (×40). (B) Ki-67 proliferative index 2% (×100). (C) Pancytokeratin-negative immune reaction (×100). (D) SMA-positive immune reaction (×100). (E) Vimentin-positive immune reaction (×100).

IMT cases may be confused with sarcoma or sarcomatoid carcinoma, especially on initial histopathological examination. Differential diagnosis is important particularly in the case of small biopsy samples. More than half of the cases are considered as malignant tumors and specifically, myxoid leiomyosarcoma. However, with regard to differential diagnosis, IMTs are more cellular, more vascular and have a more prominent inflammatory component. Additionally, they lack longitudinal striation and smooth muscle actin and desmin immune reactivity. The absence of true atypical mitoses and the presence of typical vascular proliferation mimicking granulation tissue differentiate IMTs from myxoid leiomyosarcomas (10-12). In our case, immunohistochemical staining was negative for S-100, CD68, p63, pan-cytokeratin, myogenin and CD117, positive for vimentin, actin and desmin and slightly positive for CD56 (Figures 2A-2E). The lesion resembled myosarcoma in small cystoscopy biopsy samples. Microscopically, the lesion consisted of elongated spindle cells with eosinophilic cytoplasm with an edematous, myxoid background containing plenty of blood vessels and inflammatory cells. Nuclei of the spindle cells were enlarged with no hyperchromasia (10-12). Although IMT is now considered as a benign tumor, very rarely it may show a malignant pattern including local invasion, recurrence, distant metastasis and malignant transformation (3). Preoperative diagnosis of the lesion poses challenges and complete surgical excision of the tumor and histological and immunohistochemical studies

are required for definite diagnosis (15).

IMT of the urinary bladder has a female predominance with a female to male ratio of 3:1. It may occur at any age from 2 to 73 years with a mean age of 25 years. It is worth noting that 50% of the cases occur in the first two decades of life. Tumor diameter ranges between 1 and 8 cm. Bladder fundus is involved in the majority of the cases but right wall, left wall, anterior wall, posterior wall and vesical trigone of the bladder may also be affected with decreasing order of frequency (11, 12). Consistently, our patient was a 27-year-old female with a 4.5 cm lesion in the bladder neck. Patients usually present with nonspecific symptoms including hematuria (the most common symptom), dysuria, hypogastric pain, urinary frequency, urinary retention and incontinence. Our patient had a primary complaint of excessive difficulty urinating. Clinical course and symptoms are completely benign (11).

Rhabdomyosarcoma, sarcomatoid carcinoma, and inflammatory fibrosarcoma of the mesentery and retroperitoneum should also be considered for differential diagnosis. Immunohistochemical and ultrastructural findings are helpful for the diagnosis (10-12). Lack of rhabdomyoblasts and cambium layer is essential for differentiation of the lesion from rhabdomyosarcoma. Histopathological findings as well as ultrastructural characteristics are useful for differential diagnosis of sarcomatoid carcinoma. Sarcomatoid carcinomas do not show myofibroblastic differentiation. Additionally, clinical manifestations typically occur in elderly patients and this helps distinguishing sarcomatoid carcinoma from IMT which generally occurs in the first two decades of life. Sarcomatoid carcinoma more commonly develops among males (8,10-12,16).

Conclusion

In summary, IMT is a distinctive morphological entity and an uncommon non-neoplastic, proliferative, reparative lesion of the urinary bladder with characteristic histopathological, histochemical, immunohistochemical features. Differential diagnosis should include sarcomas.

Authors' contribution

NB performed the literature search, planned the study. MS, ÖNS and MY evaluated the volunteer patient. NB and HG wrote the article. All authors read and approved the final manuscript.

Conflicts of interest

The authors declare no conflict of interest.

Ethical considerations

Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors. Informed consent was obtained from the patient for publication as a case report.

Funding/Support

None.

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