Urology Case Reports 40 (2022) 101931

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

A brief review and case report of urothelial carcinoma and metachronous leiomyosarcoma of the bladder at the same anatomic region

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ARTICLE INFO

Keywords:
Urothelial carcinoma
Leiomyosarcoma
Urinary bladder
Partial cystectomy
Radical cystectomy

ABSTRACT

One patient with bladder leiomyosarcoma and urothelial carcinoma is very rare. Only 10 cases have been reported in the literature. A 70-year-old patient was admitted due to bladder tumor. Two TURBTs were performed confirming the patient was free of tumor, and pathology reported low-grade urothelial carcinoma. Three years later, a tumor was also found on the right anterolateral wall of urinary bladder and was diagnosed as leiomyosarcoma by pathological examination. Radical cystectomy was performed. With 45 months follow-up, the patient has no recurrence. Two malignancies in the same anatomic region at different time has never been reported to date.

1. Introduction

Bladder cancer is one of most common malignant neoplasm in the world,[1] but the coexistence of two primary urinary bladder tumors of different histogenesis is very rare.[2] According to the database of PubMed, there are 10 cases of coexisting urothelial carcinoma and leiomyosarcoma (LMS) have been reported in the literature. To our knowledge, the metachronous occurrence of those two tumors at same focus has not yet been reported.

2. Case presentation

A 70-year-old male patient was admitted by macroscopic hematuria for 1 month in July 2014. The patient presented a 30-years pathologic antecedent of Diabetes Mellitus type II with a well-controlled glycemia by subcutaneous insulin injection. The patient denied any exposure to chemical or radiation substances or family history of malignant neoplasm. The Computed Tomography Urography (CTU) and cystoscopy examination showed an approximately 3.0 cm × 2.0 cm blur margin mass located at the right anterior-lateral wall of the urinary bladder (Fig. 1). Transurethral resection of the bladder tumor (TURBT) was performed under combined spinal-epidural anesthesia. The resection was deeply into the muscle layer. The pathological diagnosis was low-grade urothelial carcinoma of the bladder without cancer cells in the base (Fig. 2A). The patient received 4 times weekly intravesical chemotherapy of 30 mg pirarubicin. One month later, the patient was performed a second TURBT. There was only cicatrize tissue was observed in the surgery. The pathology report showed a small amount of mucous tissue and necrosis tissue (Fig. 2B). Postoperatively the patient received intravesical chemotherapy of 30 mg pirarubicin during a year period, and abdominal ultrasonography, thorax CT and cystoscopic examination were done regularly.

Three years later, the patient was admitted in our hospital for the third time by macroscopic hematuria accompanied with dysuria. The CTU showed a mass occupying the same location of the anterior carcinoma (Fig. 1). The cystoscopy biopsy suggested the tumor was malignant. Laparoscopic radical cystectomy and bricker operation were performed under general anesthesia, and a bilateral pelvic lymph node dissection was performed simultaneously. Spindle cells and pleomorphism cells were presented microscopically. The cell had large nucleus with obvious heteromorphism, and giant tumor cells could be observed (Fig. 2C). The immunohistochemistry (IHC) demonstrated (Fig. 2D, E,

Abbreviations: LMS, Leiomyosarcoma; CTU, Computed Tomography Urography; TURBT, Transurethral resection of the bladder tumor; IHC, Immunohistochemistry; NCCN, National Comprehensive Cancer Network.

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https://doi.org/10.1016/j.eucr.2021.101931
Received 11 October 2021; Accepted 6 November 2021
Available online 8 November 2021

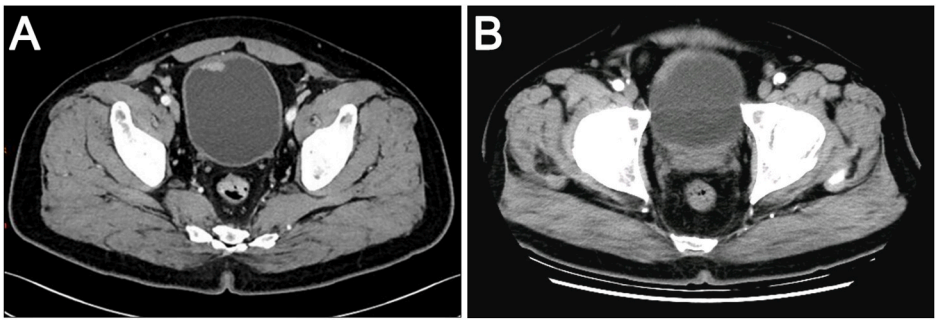

Fig. 1. Computed tomographg (CT) scan shows the calcified right anterolateral mass lesion. A: First bladder malignancy (urothelial carcinoma); B: Second bladder malignancy (leiomyosarcoma).

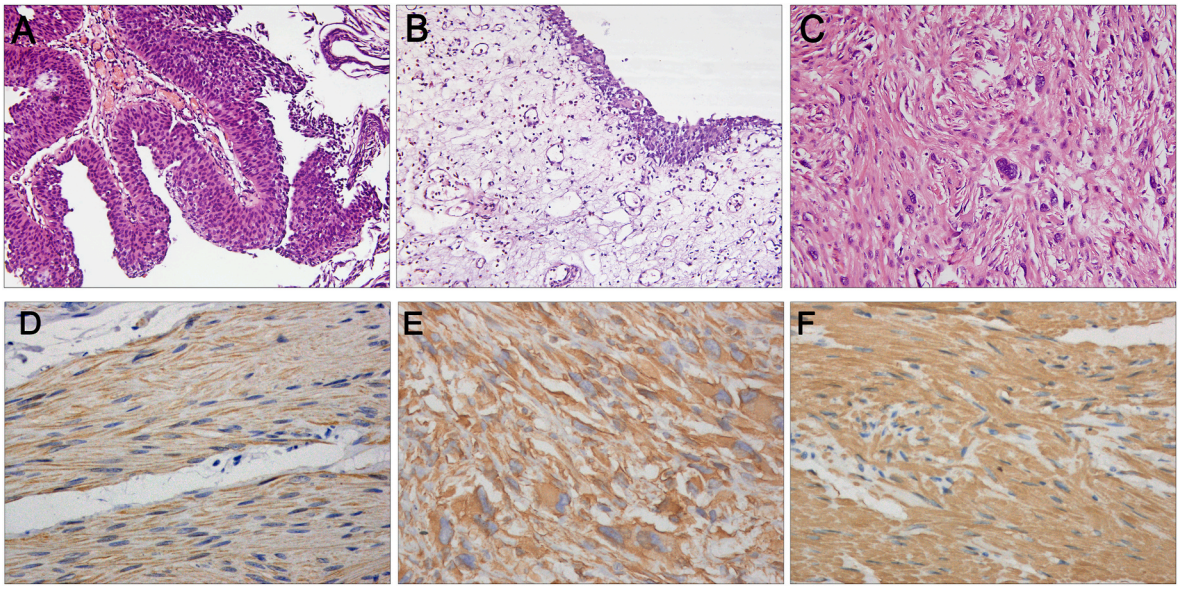

Fig. 2. Microscopic findings of the vesical tumor (× 200). A: urothelial carcinoma (Low-grade urothelial carcinoma of the bladder without cancer cells in the base and glandular cystitis), B: cicatrize tissue (mucous tissue and necrosis tissue), C: leiomyosarcoma (High-grade pleomorphic leiomyosarcoma with spindle cells and pleomorphism cells). Immunohistochemical stainings of bladder leiomyosarcoma cells (× 400). D: MSA (+), E: Vim (+), F: desmin (+).

Table 1
Summary of articles of coexisting urothelial carcinoma and LMS published over the last sixty years.

Author	Gender	Age (year)	Location	Procedure	Adjuvant treatment	Follow up
Bakaris[2]	female	44	LMS of left anterolateral wall; carcinoma of anterolateral wall	Radical cystectomy	Radiation therapy	Died of metastasis 1 year later
Chen[3]	male	54	Mixture of recurrent carcinoma and primary LMS	TUEV; partial cystectomy; radical cystectomy	Radiation therapy	No recurrence of 3.5 years followup
Uemura[4]	male	74	LMS of anterior wall, carcinoma of left lateral wall	TURBT; radical cystectomy	No referred	6 months died by plumonnary metastasis
Hejtmancik[5]	male	74	Mixture of LMS and carcinoma on the floor of the bladder	Partial cystectomy	No referred	No recurrence of 6 months
Powers [6]	male	79	Mixture of LMS and carcinoma on lateral bladder wall	Radical cystectomy	No referred	No referred
Makeles [7]	male	52	Mixture of LMS and carcinoma of almost all bladder	Radical cystectomy	Radiation therapy	No recurrence of 6 months
Ozteke [8]	female	70	Carcinoma of left-lateral bladder wall; Mixture of LMS and carcinoma of middle lateral bladder wall	Radical cystectomy	No referred	No recurrence of 8 months
Thompson [9]	male	43	Mixture of LMS and carcinoma almost whole bladder	Radical cystectomy	No referred	Died 1 month later
	female	71	Mixture of LMS and carcinoma on left lateral bladder wall	Partial cystectomy	No referred	No recurrence of 18 months followup
	male	56	Mixture of LMS and carcinoma on trigone of bladder	Radical cystectomy	No referred	No recurrence of 14 months followup

2F): MSA (+), desmin (+), Vim (+). The pathologist diagnosed a urinary bladder high-grade pleomorphic LMS infiltrating deep muscle and ranked FNCLCC Grade III. The patient declined chemotherapy and opted for active surveillance. During this surveillance period he had clinical and imaging follow-up every 3 months. With 45 months follow-up, we have not found any sign of recurrence up to date.

3. Discussion

The coexistence of two or more primary urinary bladder tumors of different histologic types is very unusual. 10 cases of coexisting bladder urothelial carcinoma and LMS have been reported (Table 1). We studied all the papers, those cases were two types malignancies appeared simultaneously. The characteristic of this case is the metachronous occurrence of bladder urothelial cancer and LMS at the same anatomic region, and this case has not yet been reported.

LMS is a rare malignancy, so it makes us hard to analysis the statistic data to surmise its etiology. One of them is the relationship between long-term chemotherapy of cyclophosphamide and bladder sarcoma, and a minority reports have revealed this point. Apart of above mentioned, the presence of urinary bladder diverticulum, abuse use of ketamine,[3] and radiation exposure,[4] could be a contribution of risk factor to this rarity. We could not discern any those factors, while our patient had a history of smoking and type II diabetes mellitus.

The clinical features of bladder LMS are similar to those of urothelial carcinoma: hematuria, dysuria, pollakiuria and without any typical symptom. The supplementary imaging technology including CTU, magnetic resonance imaging and bone scanning can make a contribution to the diagnosis and classification of staging but lacks of significant differential diagnosis value. The pathological examination and IHC can make assurance diagnosis.

The treatment of this patient is strictly performed according to the international guideline of National Comprehensive Cancer Network (NCCN). The NCCN guideline 2016 suggested a complete TURBT is essential for the patient's prognosis, even the pathology reported a well resection specimen. The first pathology report of this patient confirmed the diagnosis of low-grade urothelial carcinoma and the scope of the lesion is relatively large. Therefore, we performed re-TURBT. Second operation's pathology report was free of tumor cells and proved the success of the first resection. The intravesical instillation of chemotherapy was performed for 1 year to prevent recurrence. A regularly follow-up play an important role in the management, and the patient remained disease free of 41 months.

In December 2017, The CTU showed a mass occupying at the same location of the anterior carcinoma, and the cystoscopic biopsy suggested it was malignant. So, the patient underwent laparoscopic radical cystectomy and Bricker operation. The pathologist diagnosed a urinary bladder high-grade pleomorphic LMS. For the treatment of the urinary bladder LMS, there is no consensus protocol by its rarity and lack of cases. Because of its aggressive and poor prognosis, a wider border resection would be a better choice for the high grade LMS.[5] Chemotherapy and/or radiation therapy are recommended if there is any kind of lymph node metastasis, adjacent invasion. Even the valuation of chemotherapy and radiation therapy have not been implemented, the patient rejected it. Bladder LMS survival rates for patients better than the rates with sarcomas of other organs,[5] while a closely follow-up treatment is needed for the patient. As there is no guideline of LMS, all we did is summarized by experience. The patient has no recurrence of the two malignancies up to date. Therefore, our treatment for bladder urothelial carcinoma and LMS was in the right direction.

4. Conclusion

Complete TURBT is good for the patient with low-grade bladder urothelial carcinoma. High-grade bladder LMS is required to perform radical surgery. IHC is valuable for the diagnosis of bladder LMS. Furthermore, cases and statistical analysis should be worked out to provide an effective guideline.

Funding

This study was funded by grants from Ningxia Hui Autonomous Region focus on research and development projects (No.2020BFH03001) and Ningxia Hui Autonomous Region youth top talent training project.

Ethics approval

The study was done after agreement from the local ethics committee and with the patient's informed consent.

Consent to participate

Not required.

Consent for publication

Not required.

Availability of data and material

Not applicable.

Code availability

Not applicable.

Authors' contributions

Ruining Zhao: Project design, drafting and edited of the manuscript; Lihong Nie: Project design, reviewed and edited of the manuscript; Yajie Li: Data Collection and drafting of the manuscript; Zhang Hang: Data Collection and data analysis; Hongbin Shi: Project design the manuscript; Zhenwei Wang: Data Collection and data analysis.

Declaration of competing interest

The authors declare that there is no conflict of interests regarding the publication of this article.

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