


Case Report

Inflammatory myofibroblastic bladder tumor with divergent behavior in a patient with spinal cord injury

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Abbreviations & Acronyms

IMT = inflammatory myofibroblastic tumor
SCC = squamous cell carcinoma
SCI = spinal cord injury

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Received 9 January 2019; accepted 8 May 2019.
Online publication 29 May 2019

Introduction: An inflammatory myofibroblastic tumor of the bladder is rare. Some urothelial carcinoma with sarcomatoid changes may mimic an inflammatory myofibroblastic tumor.

Case presentation: A 76-year-old man with indwelling urinary catheters because of a spinal cord injury presented with gross hematuria. Transurethral electrocoagulation and a resection were performed. A T2-weighted magnetic resonance imaging showed a tumor and hematoma in the bladder diverticulum that pathologically resembled an inflammatory myofibroblastic tumor. This lesion was then removed en bloc by partial cystectomy. Histology showed a squamous cell carcinoma with sarcomatoid changes. After 4 months, a computed tomography showed the lesion had expanded outside the skin adjacent to the bladder.

Conclusion: Some cases of inflammatory myofibroblastic tumors have a malignant course. Urologists therefore need to be aware of the possibility of rare cases of malignant bladder myofibroblasts.

Key words: bladder cancer, inflammatory myofibroblastic tumor, sarcomatoid changes, squamous cell carcinoma.

Keynote message

IMTs of the bladder are rare and generally have indeterminate malignant potential, although some reports have shown a malignant course. Urologists should therefore keep in mind that some urothelial carcinoma with sarcomatoid changes may mimic an IMT.

Introduction

IMTs of the bladder are extremely rare and usually have a low or indeterminate malignant potential, and therefore a good prognosis and a low risk of distant metastases.¹ Potential treatment option for an IMT of the bladder is either a transurethral resection, partial cystectomy, or radical cystectomy.

SCC of the bladder may occur in patients with either long-term indwelling catheters, schistosomiasis, chronic irritation due to vesical calculus, or chronic cystitis.² Some SCCs develop sarcomatoid changes and sometimes exhibit myxoid features that mimic IMT.^{3,4} The present case describes a SCC bladder tumor with sarcomatoid changes in the bladder diverticulum, in which initial pathology was shown as an IMT.

Case presentation

A 76-year-old man, who had an indwelling urinary catheter for 54 years because of a SCI caused by a traffic accident, presented with hematuria and a bladder tamponade. Cystoscopy

showed a large blood clot in the bladder diverticulum. Transurethral electrocoagulation and a resection were then performed. A T2-weighted magnetic resonance imaging showed a tumor and hematoma in the bladder diverticulum that pathologically resembled spindle cells accompanied by intermingled inflammatory cells (Fig. 1a). The results of immunohistochemistry examination showed the following: Desmin (+), smooth muscle actin (+), and anaplastic lymphoma kinase (–). We suspected IMT and therefore removed the tumor en bloc by partial cystectomy. The tumor dimensions were 11 × 9 × 8 cm and weight 716 g. Histological examination showed a SCC with sarcomatoid changes (Fig. 1b), and pathological stage was T3a. The surgical wound festered 4 months after surgery (Fig. 1c), with a computed tomography showing the lesion had expanded outside the skin adjacent to the bladder (Fig. 1d). The patient was advised of the need for a radical cystectomy although he did not agree to this treatment, and therefore, we decided to carry out radiotherapy and chemotherapy. Two cycles of chemotherapy (cisplatin and gemcitabine) and radiotherapy (total 60 Gy: 2 Gy per fraction:30 fraction) over 2 months resulted in a partial response in the lesion. Conservative therapy was introduced after this period of radiotherapy and chemotherapy, with the tumor size remaining stable for 5 months.

Discussion

An IMT should be treated carefully because the most important entity in the differential diagnosis is urothelial carcinoma with sarcomatoid changes.^{3,4} Urothelial carcinomas occasionally develop sarcomatoid changes. Our report in a patient with long-term indwelling catheters necessary because of a spinal cord injury showed a SCC with sarcomatoid changes that mimicked an IMT.

IMT is a rare mesenchymal tumor that has intermediate malignant potential. These lesions develop in various organs, such as the lung, retroperitoneum, and pelvis.^{3,5} IMT of the bladder represents less than 1% of all bladder tumors⁶ and is therefore one of the most difficult diagnostic dilemmas in genitourinary pathology.⁷

IMTs have low or indeterminate malignant potential and non-metastasizing proliferation of myofibroblasts, with the potential for recurrence and persistent local growth.^{3,8} A typical IMT can be locally aggressive and need to be surgically resected using either a transurethral resection, partial cystectomy, or radical cystectomy.^{3,5,8} An IMT may mimic a urothelial carcinoma with sarcomatoid changes, which represents only 0.1–0.3% of bladder cancers.⁷ Rare cases may arise contemporaneously with urothelial carcinoma with sarcomatoid changes, similar to the lesions described in the present case.³

Numerous case series have been reported that show an association between SCI patients and bladder cancer.^{2,9} For example, Gui-Zhong *et al.* observed in a meta-analysis that bladder cancer occurs more often in SCI patients than in the general population. Bladder cancer is the third leading cause of cancer death in SCI patients. In particular, patients with a long-standing SCI or using an indwelling catheter for 16 years may be at the highest risk for bladder cancer. However, the tumor subtype is different from that in the general population, with the estimated percentage of SCC in the SCI population being 36.8%,² a figure considerably higher than the estimated frequency of SCC of 2–7%.^{2,9,10} Our case had indwelling urinary catheters for 54 years and therefore had a very high risk of developing a SCC. We had to distinguish a possibility of sarcomatoid variant, although the histological examination showed IMT in a small amount of the specimen obtained by transurethral resection.

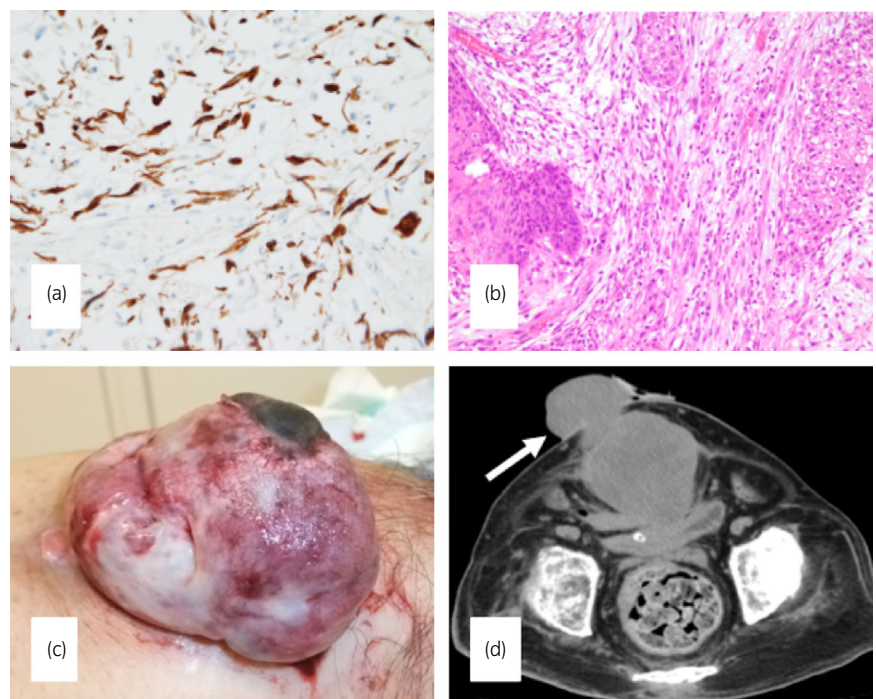


Fig. 1 (a) A spindle cell lesion with an increased size showing partial positivity for AE1/AE3. (b) An atypical squamous cell lesion with hemorrhage, fibrosis, and increasing necrosis that has come in contact with the spindle cell lesion. The lesion has changed partially to a sarcoma. (c) The tumor grew out of the abdominal skin of the surgical wound 4 months after surgery. (d) Computed tomography showing a 12-cm-long tumor extending from the bladder to the abdominal wall and skin (arrow).

Patients with a pure SCC of the bladder have been reported to have a poor prognosis with most patients dying within 1–3 years from diagnosis.¹¹ However, mortality attributable specifically to bladder cancer does not vary significantly between patients with either urothelial carcinoma or a SCC.^{12,13} Patients with a SCC only rarely develop metastatic disease. One report on these tumors which were treated using a variety of regimens including chemotherapy, radiation, and surgery showed the overall 5-year survival rate was 16%, with only 8% of patients developing metastatic disease.¹⁴ These data indicate loco-regional control plays an important role in managing SCC. Radical cystectomy is established as the gold standard for locally advanced bladder cancer, especially SCC.^{12,13,15} In our case, it was desirable to perform a radical cystectomy when pathology of the tissue identified a SCC.

Conclusion

We report an IMT of the bladder which revealed a SCC. This lesion was difficult to diagnose and treat accurately, because IMT has similar pathological features such as sarcomatoid changes. It is important to keep in mind the divergent behavior of IMT, which may result in a malignant course.

Conflict of interest

The authors declare no conflict of interest.

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