

Urothelial carcinoma with sarcomatoid/osteosarcoma variant of the bladder: A case report

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Abstract

A 67-year-old man was diagnosed with non-muscle invasive bladder cancer and underwent transurethral resection of the lesions in August 2017. The pathological findings revealed high-grade urothelial carcinoma. The tumor relapsed as urothelial carcinoma with sarcomatoid/osteosarcoma variant with vascular invasion, and transurethral resection was performed in December 2017. He underwent laparoscopic radical cystoprostatectomy and orthotopic neobladder reconstruction using ileum in March 2018. The patient developed lung metastasis in July 2018. He underwent four courses of chemotherapy with doxorubicin and thoracoscopic left lower lobectomy of the lung in October 2018.

Keywords

Bladder cancer, sarcomatoid/osteosarcoma variant, transurethral resection in one piece

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Introduction

Sarcomatoid carcinoma of the urinary bladder (SCUB) is an unusual malignancy containing both carcinomatous and sarcomatous components. SCUB has worse disease-specific and overall survival than conventional high-grade urothelial carcinoma.¹ The epithelial component in most reported cases of SCUB is urothelial carcinoma. The sarcomatoid variant of the tumor includes spindle cell sarcoma, chondrosarcoma, and rhabdomyosarcoma. The osteosarcoma variant is extremely rare. We report a case of urothelial carcinoma with sarcomatoid/osteosarcoma variant with therapeutic course.

Case report

A 67-year-old man complained of discomfort at urination, urethralgia, and gross hematuria since April 2017 and visited our hospital in July 2017. Cystoscopy revealed rough mucosal lesions on the left side wall and the trigone of the bladder. Computed tomography (CT) revealed no abnormal findings (Figure 1(a)). Urine cytology was Class III. The patient underwent transurethral resection (TUR-BT) of the lesions in August 2017. The pathological findings revealed

high-grade urothelial carcinoma. Cystoscopy revealed a papillary broad-base tumor on the upper left lateral side of the bladder and a non-papillary tumor on the upper lateral side of the left ureteral orifice in December 2017 (Figure 2(a) and (b)). Urine cytology was Class III. CT revealed contrast-enhanced wall thickening near the left ureteral orifice (Figure 1(b)). Magnetic resonance imaging (MRI) did not reveal obvious muscle invasion of the tumor in the vicinity of the left ureteral orifice (Figure 1(c) and (d)). The patient underwent TUR-BT in one piece (TURBO) for two lesions in January 2018 (Figure 2(c)). The pathological findings of the papillary broad-base tumor showed non-invasive urothelial

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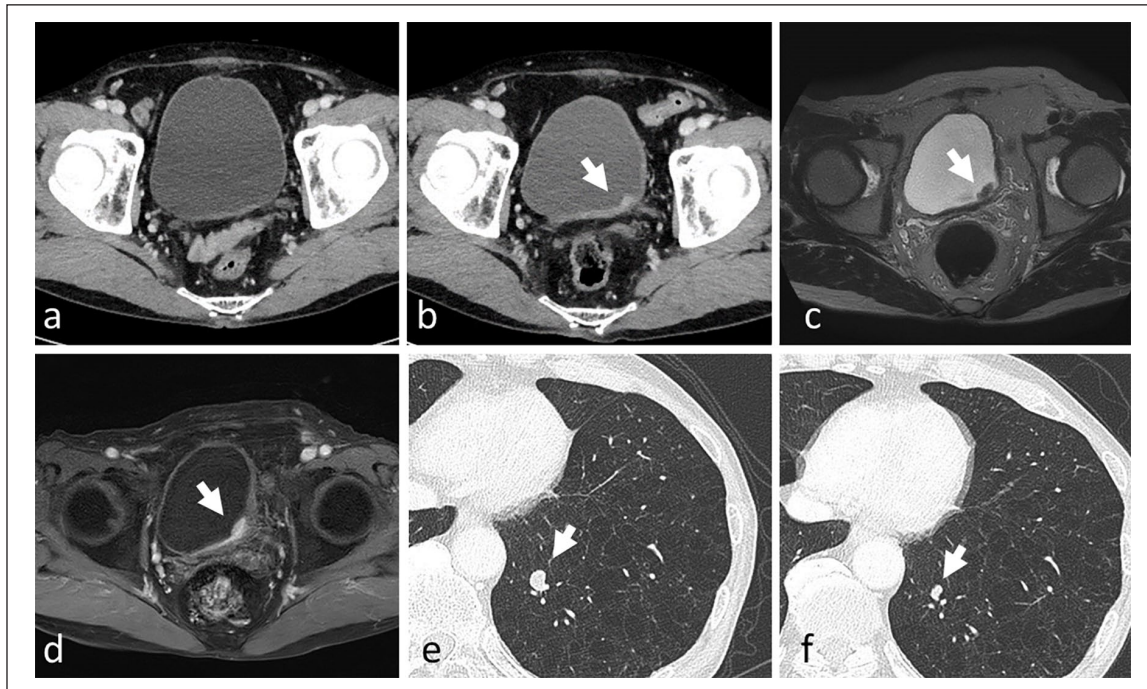


Figure 1. CT and MRI findings. (a) CT reveals no abnormal findings in July 2017. (b) CT reveals the contrast-enhanced wall thickening near the left ureteral orifice in December 2017 (arrow). (c) T2-weighted image of MRI reveals that the tumor in the vicinity of the left ureteral orifice is a low signal lesion without obvious muscle invasion in January 2018 (arrow). (d) On dynamic contrast enhancement imaging, the lesion demonstrates early uptake at the same lesion as the T2-weighted image (arrow). (e) CT findings suspect metastasis in the left lower lobe of the lung in July 2018 (arrow). (f) CT findings reveal a decrease in the size of the nodules in the left lower lobe in September 2018 (arrow).

carcinoma (G2, high-grade, and pTa). The non-papillary tumor showed invasive basal-type urothelial carcinoma with sarcomatous/osteosarcoma variant with vascular invasion (pT2a) (Figure 2(d)–(j)). The patient underwent laparoscopic radical cystoprostatectomy and orthotopic neobladder reconstruction using ileum in March 2018. Pathological findings revealed three lesions: non-invasive papillary urothelial carcinoma (high-grade, G2, and pTis), non-invasive papillary urothelial carcinoma (high-grade, G2, and pTis), and osteosarcomatous/carcinosarcoma (pT3).

Three months after the operation, CT findings suggested metastasis in the left lower lobe of the lungs in July 2018 (Figure 1(e)). The patient underwent four courses of chemotherapy with doxorubicin, and the nodule in the left lower lobe decreased in size (Figure 1(f)). The patient underwent thoracoscopic left lower lobectomy of the lungs in October 2018. The pathological findings revealed metastasis of the sarcomatous tumor. Eighteen months after the lobectomy, he has remained free from recurrence.

Discussion

The current World Health Organization classification recommends the use of the term “sarcomatoid carcinoma” for all biphasic malignant neoplasms of the urinary tract exhibiting morphologic and/or immunohistochemistry evidence

of epithelial and mesenchymal differentiation.² SCUB is a rare and aggressive subtype of bladder cancer.¹ Little information is available regarding its clinical features and appropriate treatments. Only 301 (0.16%) cases of SCUB were reported among the 182,283 patients with a primary bladder tumor in an investigation of the Surveillance, Epidemiology, and End Results database from 1988 to 2001.¹ It most frequently occurred in patients in their sixth to seventh decades and was more common in men than in women.³ The most common symptom at presentation is gross hematuria. The tumors are usually large and contain an admixture of malignant epithelial and mesenchymal elements on microscopic examination. Lymph node metastasis is found in 15%–40% of cases, and the 5-year survival rate is poor at 21%–28%.^{1,3} Recently, the results of molecular genetic studies have provided strong support for a common monoclonal origin of both epithelial and mesenchymal components in SCUB.

Cystoscopy usually shows broad-based, often polypoid masses with ulcerated and hemorrhagic surfaces. The sarcomatoid component of SCUB is often present in the deeper portion of the tumor; therefore, detection of this component in the voided cytologic specimen is low. In this case, urine cytology was Class III. CT scans and MRI can be included in the staging and metastatic workup for invasive bladder cancer. In this case, CT revealed contrast-enhanced wall thickening; however, MRI did not reveal obvious muscle invasion of the tumor.

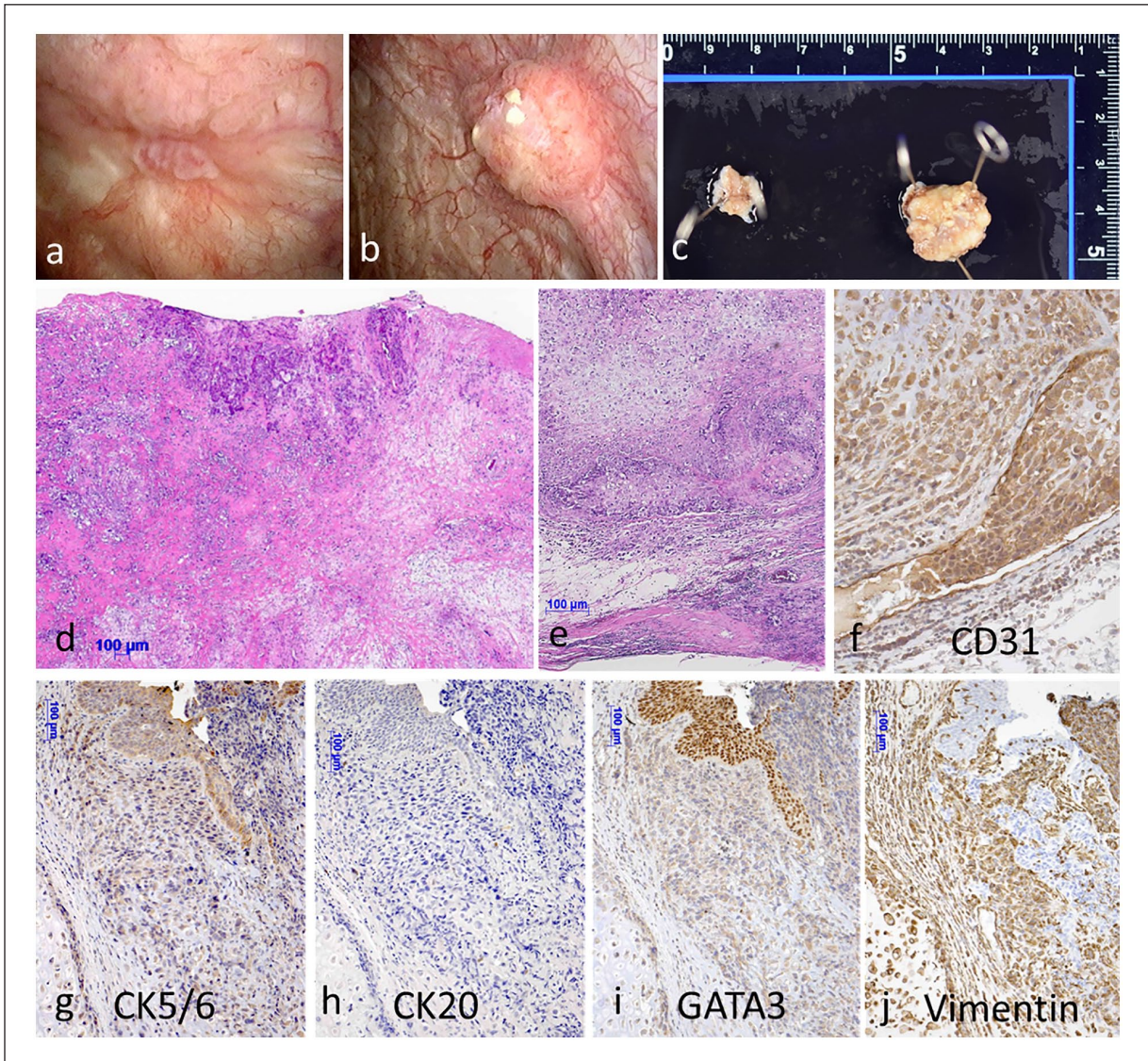


Figure 2. Cystoscopic findings and histopathological findings of excised specimen by TURBO. Cystoscopy revealed a papillary broad-base tumor in the upper left lateral side of the bladder ((a), left side of (c)) and a non-papillary tumor in the upper lateral side of the left ureteral orifice ((b), right side of (c)) in December 2017. (d) and (e) The pathological findings of a non-papillary tumor in the upper lateral side of the left ureteral orifice obtained by TURBO (right side of (c)) reveal invasive urothelial carcinoma with sarcomatous/osteosarcoma variant with vascular invasion (pT2a) in January 2018. (f) The tumor cells located in CD31-positive blood vessels. The immunohistochemical findings of this case are consistent with the sarcomatoid variant of urothelial carcinoma based on both an associated basal-type urothelial carcinoma components: (g) CK5/6-positive, (h) CK20-negative, and (i) GATA3-positive; and a sarcomatoid component: (j) vimentin-positive. The sarcomatoid variant lesion also retains the elements of basal-type urothelial carcinoma: (g) CK5/6 positive, (h) CK20-negative, and (i) GATA3 positive.

Using immunohistochemistry for diagnosis, the expression of epithelial and mesenchymal markers, such as cytokeratin and vimentin, was positive.⁴ Based on immunohistochemical features and hematoxylin and eosin staining, this case was diagnosed as urothelial carcinoma with sarcomatoid/osteosarcoma variant with both an associated urothelial carcinoma component and a sarcomatoid component (Figure 2(g)–(j)). The sarcomatoid variant lesion also retained the elements of basal-type urothelial carcinoma.

TUR-BT is performed for the diagnosis and treatment of SCUB. TURBO or en-bloc TUR-BT is effective for complete tumor resection, accurate staging, and identification of vascular invasion.⁵ In this case, the tissue obtained by TURBO showed the transition from urothelial carcinoma to sarcoma and was also useful in the assessment of resected margins and vascular invasion (Figure 2(e) and (f)).

No standardized treatment protocol is available for this disease because of the rarity of SCUB and the absence of

randomized controlled trials. Various combinations of neoadjuvant or adjuvant chemotherapy, chemotherapy for metastatic disease, and radiation therapy after radical surgery have been advocated due to the high local recurrence and metastasis rates of the tumor after radical cystectomy. We performed radical cystectomy. However, the patient developed lung metastases in a short period. The patient received chemotherapy with doxorubicin according to the chemotherapy regimen for osteosarcoma.⁶ As the patient had only one metastatic site after chemotherapy, we removed the residual site. The patient has been alive without recurrence for 18 months after the lobectomy. SCUB has worse disease-specific and overall survival than conventional high-grade urothelial carcinoma; however, aggressive treatment for SCUB can lead to long-term remission status.

Conclusion

We presented a case of urothelial carcinoma with sarcomatoid/osteosarcoma variant. As most investigators have reported poor outcomes for patients with SCUB regardless of the type of treatment, early diagnosis and treatment are necessary for the cure of the disease.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.


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Informed consent

Written informed consent was obtained from the patient for his anonymized information to be published in this article.

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References

1. Wright JL, Black PC, Brown GA, et al. Differences in survival among patients with sarcomatoid carcinoma, carcinosarcoma and urothelial carcinoma of the bladder. *J Urol* 2007; 178(6): 2302–2306; discussion 2307.
2. Humphrey PA, Moch H, Cubilla AL, et al. The 2016 WHO classification of tumours of the urinary system and male genital organs-part B: prostate and bladder tumours. *Eur Urol* 2016; 70(1): 106–119.
3. Malla M, Wang JF, Trepeta R, et al. Sarcomatoid carcinoma of the urinary bladder. *Clin Genitourin Cancer* 2016; 14(5): 366–372.
4. Sanfrancesco J, McKenney JK, Leivo MZ, et al. Sarcomatoid urothelial carcinoma of the bladder: analysis of 28 cases with emphasis on clinicopathologic features and markers of epithelial-to-mesenchymal transition. *Arch Pathol Lab Med* 2016; 140(6): 543–551.
5. Ukai R, Hashimoto K, Nakayama H, et al. Lymphovascular invasion predicts poor prognosis in high-grade pT1 bladder cancer patients who underwent transurethral resection in one piece. *Jpn J Clin Oncol* 2017; 47(5): 447–452.
6. Mack LA, Crowe PJ, Yang JL, et al. Preoperative chemoradiotherapy (modified Eilber protocol) provides maximum local control and minimal morbidity in patients with soft tissue sarcoma. *Ann Surg Oncol* 2005; 12(8): 646–653.