

Oncology

Penile sarcomatoid urothelial carcinoma: A case report

Shirley Wang, Meredith Metcalf, Jeannie Hoffman-Censits, Trinity Bivalacqua, Sunil H. Patel*

The Brady Urological Institute, The Johns Hopkins School of Medicine, Baltimore, MD, 21287, USA

ARTICLE INFO

Keywords:

Urethral carcinoma
Urologic oncology
Sarcomatoid carcinoma

ABSTRACT

A 74-year-old man with a penile mass was diagnosed with sarcomatoid urothelial carcinoma. Further workup did not show any other lesions or metastases. He was treated with a total penectomy, bilateral inguinal lymph node dissection, and pelvic lymphadenectomy. Following surgery, he received six cycles of cisplatin and gemcitabine. Sarcomatoid carcinoma and carcinosarcoma of the urethra are rare; six prior cases have been reported in the literature, with this being the first urothelial with sarcomatoid component. Survival in patients with sarcomatoid carcinoma or carcinosarcoma of the urinary tract is poor, with the limited data supporting a multimodal approach to improve survival.

Introduction

Sarcomatoid carcinoma of the urethra is rare, with only one prior case report in a female.¹ This is histologically different than carcinosarcoma (CS), however, both act in a similar biological and clinical manner. Here, we present a case of primary urethral sarcomatoid urothelial carcinoma presenting as a slowly enlarging mass and causing lower urinary tract symptoms (LUTS). We review the literature on sarcomatoid carcinoma and CS tumor variants in the urinary tract as well as prognosis and treatment of urethral cancer.

Case presentation

A 74-year-old man presented with a ten-year history of a penile mass, which initially was believed to be Peyronie's disease. The mass had slowly been enlarging in size, prompting further workup. His past history was significant for Dupuytren's contracture, 20-pack-year smoking history, and family history of Hodgkin lymphoma. On physical exam, there was a mass which was hard, non-tender, fixed, and extended towards the glans penis. Inguinal lymphadenopathy was palpable bilaterally.

Computed tomography (CT) scan showed two heterogenous, hypodense masses measuring 3.8×3.4 cm on the left and 2.6×2.6 cm on the right (Fig. 1). Indeterminate iliac and inguinal lymphadenopathy was noted, with the largest node measuring 1.3 cm. Imaging showed no evidence of distant metastases.

A biopsy of the mass demonstrated sarcomatoid urothelial carcinoma, with the epithelial portion consisting of invasive high-grade

urothelial carcinoma and the mesenchymal component consisting of malignant spindle cells (Fig. 2). Flexible cystourethroscopy showed normal mucosa and no bladder abnormalities or masses. Additionally, bladder cytology was negative for urothelial carcinoma. At this time, additional core biopsies of the penile mass were obtained, confirming the diagnosis.

He subsequently underwent a radical penectomy with a perineal urethrostomy, bilateral ilioinguinal lymphadenectomy, and pelvic lymphadenectomy. He recovered well, was discharged from the hospital on postoperative day one, and had no 30-day complications. Final pathology showed a 4.2 cm mass with extension into the corpus cavernosum and perineural and lymphovascular invasion present. Eighty-four lymph nodes were negative for tumor bilaterally. Final staging was pT3N0. He then underwent six cycles of chemotherapy with gemcitabine and cisplatin, with no complications.

Discussion

Male urethral carcinoma is a rare malignancy, with annual incidence reported to be 4.3 cases per million, with predominate histologies being squamous cell and transitional cell.² CS and sarcomatoid carcinoma are distinct entities on a spectrum of epithelial and mesenchymal differentiation. CS involves carcinoma with a true sarcomatous component and sarcomatoid carcinoma involves carcinoma with a spindle cell component which retains epithelial markers. While the utility of distinguishing between the two has been debated, recent data showing worse mortality in CS suggest some differences in clinical behavior. Although they are distinct, CS and sarcomatoid carcinoma are often discussed together in

* Corresponding author. 600 N Wolfe St, Baltimore, MD, 21287, USA.

E-mail address: Spate167@jhmi.edu (S.H. Patel).

<https://doi.org/10.1016/j.eucr.2021.101706>

Received 17 April 2021; Received in revised form 30 April 2021; Accepted 3 May 2021

Available online 7 May 2021

2214-4420/© 2021 The Authors.

Published by Elsevier Inc.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

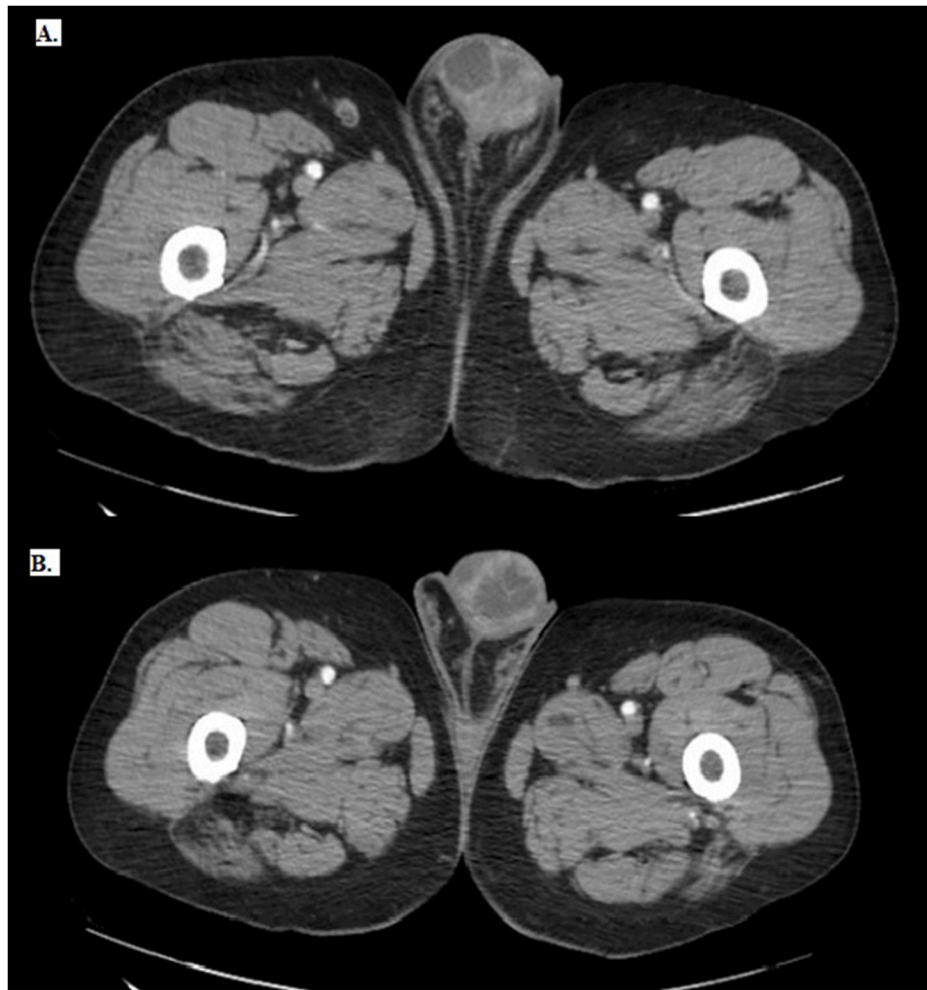


Fig. 1. CT showing hypodense mass in distal penis measuring: a) 2.6×2.6 cm on right side and b) 3.8×3.4 cm on left side.

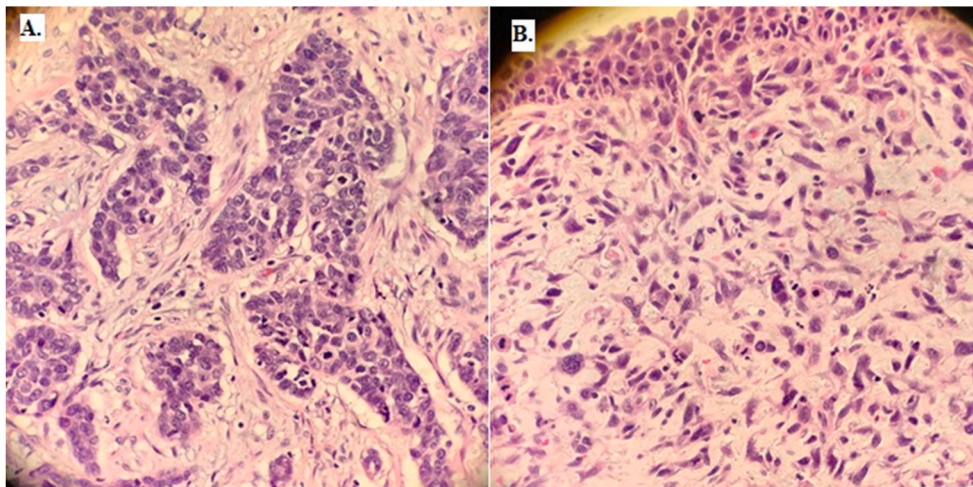


Fig. 2. Histopathologic features of this patient's urothelial sarcomatoid carcinoma (H&E 400x): a) high-grade urothelial carcinoma; b) non-specific malignant spindle cells.

the literature due to their similar presentation, behavior, and treatment. Furthermore, there are few reports of both histologic types in the urethra: four in women and two in men.¹

Sarcomatoid carcinoma and CS of the urethra often present at a late stage, with four of six subjects of previous reports diagnosed with

metastatic disease. Among patients with bladder cancer, those with sarcomatoid and CS histologies presented with more advanced disease as compared to those with urothelial.³ Sarcomatoid carcinoma and CS are aggressive in the urinary tract; even after controlling for stage at presentation in individuals with bladder cancer, sarcomatoid and CS

histologies are associated with poorer survival compared to urothelial.³ Of the four prior subjects of case reports with reported follow-up data, all experienced progression of the disease following treatment.¹

This patient presented with a history of a mass on the dorsum of his penis slowly enlarging over ten years with associated LUTS. Clinical growth rate of sarcomatoid carcinomas has not been correlated with patient outcomes.

Primary urethral carcinoma similarly has poor prognosis, with 5-year survival estimated to be 46%.² Initial evaluation includes cystoscopy to determine presence of bladder lesions as well as imaging for assessment of distant metastases. Nodal stage and proximal location has been found to be predictive of worse OS.⁴

Due to the rarity of primary urethral carcinoma, large studies comparing treatments are limited. Retrospective studies suggest improved survival in those who received surgery with adjuvant radiation compared to surgery alone, use of adjuvant chemotherapy \pm radiation in patients with at least stage T3 disease, and use of neoadjuvant chemotherapy. Overall, data support multimodal treatment with surgery and adjuvant radiation or chemotherapy confers superior survival over monotherapy.⁵

Conclusion

Sarcomatoid carcinoma is an aggressive disease that rarely affects the urinary tract. Prognosis is poor, but available evidence supports improved outcomes for those who receive multimodal treatment with systemic therapy and local resection plus or minus radiation therapy.

References

1. Komai Y, Urushibara M, Saito K, Sakai K, Morimoto S. A case of sarcomatoid carcinoma in the region of the female urethra. *Jpn J Urol.* 2005;96(6):650–653. <https://doi.org/10.5980/jpnjurol1989.96.650>.
2. Swartz MA, Porter MP, Lin DW, Weiss NS. Incidence of primary urethral carcinoma in the United States. *Urology.* 2006;68(6):1164–1168. <https://doi.org/10.1016/j.urology.2006.08.1057>.
3. 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.
4. Janisch F, Abufaraj M, Fajkovic H, et al. Current disease management of primary urethral carcinoma. *Eur. Urol. Focus.* 2019;5(5):722–734. <https://doi.org/10.1016/j.euf.2019.07.001>.
5. Gakis G, Morgan TM, Efstathiou JA, et al. Prognostic factors and outcomes in primary urethral cancer: results from the international collaboration on primary urethral carcinoma. *World J Urol.* 2016;34(1):97–103. <https://doi.org/10.1007/s00345-015-1583-7>.