Urology Case Reports 39 (2021) 101795

Contents lists available at ScienceDirect

Urology Case Reports

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

Malignant priapism as the initial presentation induced by the metastasis of renal clear cell carcinoma after radical nephrectomy

Yun Luo [1], Fei Yang [1], Jiarong Cai, Wenbiao Li, Hailun Zhan [*]

Department of Urology, the Third Affiliated Hospital of Sun Yat-Sen University, Guangzhou, 510630, China

ARTICLE INFO

Keywords:
Priapism
Renal cell carcinoma
Metastasis
Penis

ABSTRACT

The priapism secondary to the tumor metastasis to penis is rare. We described a case of a patient with priapism caused by the metastasis of renal cell carcinoma after radical nephrectomy. The previous reported cases found in the literature were reviewed. The pathophysiology, diagnosis, management and prognosis were discussed. Renal cell carcinoma metastasis to penis usually represents a more advanced stage of disease and carries a poor prognosis. The therapy is only palliative.

1. Introduction

Metastatic tumor to penis is rare. The clinical manifestation includes indurated nodules, mass, priapism, and ulceration.[1] The priapism, which is caused by the metastasis of solid tumors, is more unusual. Renal cell carcinoma (RCC) is characterized by protean manifestations and well known to metastasize to lymph nodes, lung, liver, bone, adrenal, etc. However, the penis is seldom been involved. We are reporting such a case along with a review of literatures and discussing its pathophysiology, diagnosis, management and prognosis.

2. Case presentation

A 60-year-old man presented to the department of urology with persistent non-painful priapism that had started 10 days earlier. The penis was enlarged, tender, and semierect without focal mass. He had no other symptoms. The inguinal lymph nodes were not palpable.

One month ago, he received an open radical nephrectomy and regional lymphadenectomy for giant Bosniak IV cystic lesion at another hospital (Fig. 1A and B). There was no evidence of metastasis before surgery. Microscopic and immunohistochemical examination revealed a mixed clear cell RCC (Fuhrman III-IV) and papillary RCC with perinephric fat infiltration but not through the Gerota fascia (Fig. 1C). Extension to small vessels was also seen. The resection margins and lymph nodes were negative. The patient was diagnosed with pathological stage pT3N0M0.

The results of laboratory evaluation including blood routine test, liver function, renal function, tumor antigen such as CEA, AFP, PSA, CA125, and CA199, blood screening of rheumatoid diseases, HIV and syphilis were all normal.

Color Doppler ultrasonography of phallus showed bilateral corpus cavernosa was significantly swollen, blood flow signal of corpus cavernosa was abundant, blood flow signal of deep dorsal vein and dorsal artery of penis were visible, and the fascia of penis was thickening. It suggested the occurrence of non-ischemic priapism. Magnetic resonance imaging (MRI) revealed that bilateral corpus cavernosa was engorged and heterogeneous enhanced, and the fascia of penis was thickening and focal enhanced (Fig. 2). SPECT/CT coincidence 18F-FDG imaging revealed that bilateral corpus cavernosa was engorged and hypermetabolic, surrounding tissue of previous surgical area of left radical nephrectomy was hypermetabolic, the retroperitoneal lymph node and the lymph nodes along iliac blood vessels were slightly enlarged and hypermetabolic. However, the result of SPECT/CT didn't indicate the tumor metastasis.

Since the priapism is different from the common non-ischemic priapism, the tissue biopsy from corpus cavernosa was performed (Fig. 3A). Microscopic examination revealed that nests of cells with clear cytoplasm, enlarged hyperchromatic nuclei, and indistinct nucleoli infiltrated corpora cavernosa venous spaces and fibromuscular stroma (Fig. 3B). Immunohistochemical examination of tumor cells were positive for CK, Vimt, CD68, Ki-67(30%), CD10, Pax-8, and CA-IX and negative for GATA-3, P63, CK5/6, CK7, P504S. The pathological result

* Corresponding author.Department of Urology, the Third Affiliated Hospital of Sun Yat-sen University, Number 600, Tianhe Road, Guangzhou, 510630, China.
E-mail addresses: luoyun8@mail.sysu.edu.cn (Y. Luo), yangfei30@163.com (F. Yang), jadycai@qq.com (J. Cai), liwbiao@139.com (W. Li), zhanhl@mail.sysu.edu.cn, Hailunzhan@aliyun.com (H. Zhan).

[1] These authors contributed equally to this study.

https://doi.org/10.1016/j.eucr.2021.101795
Received 15 July 2021; Received in revised form 30 July 2021; Accepted 1 August 2021
Available online 3 August 2021

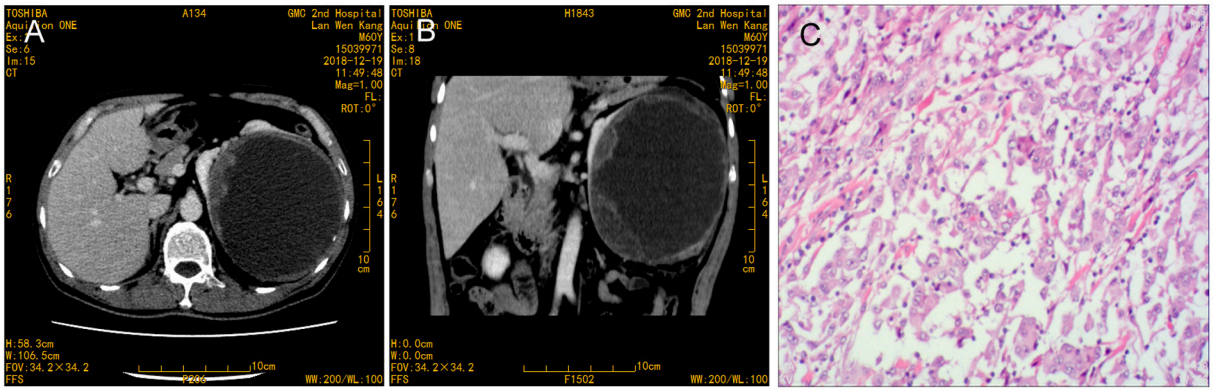

Fig. 1. CT scan of the abdomen (A and B). An axial (A) and coronal (B) image showed a giant Bosniak IV cystic lesion arising from the upper and middle aspect of the left kidney. (C) Histologic slide indicated a mixed clear-cell renal-cell carcinoma (Fuhrman III-IV) and papillary renal-cell carcinoma. (Hematoxylin-eosin stain; original magnification, x200).

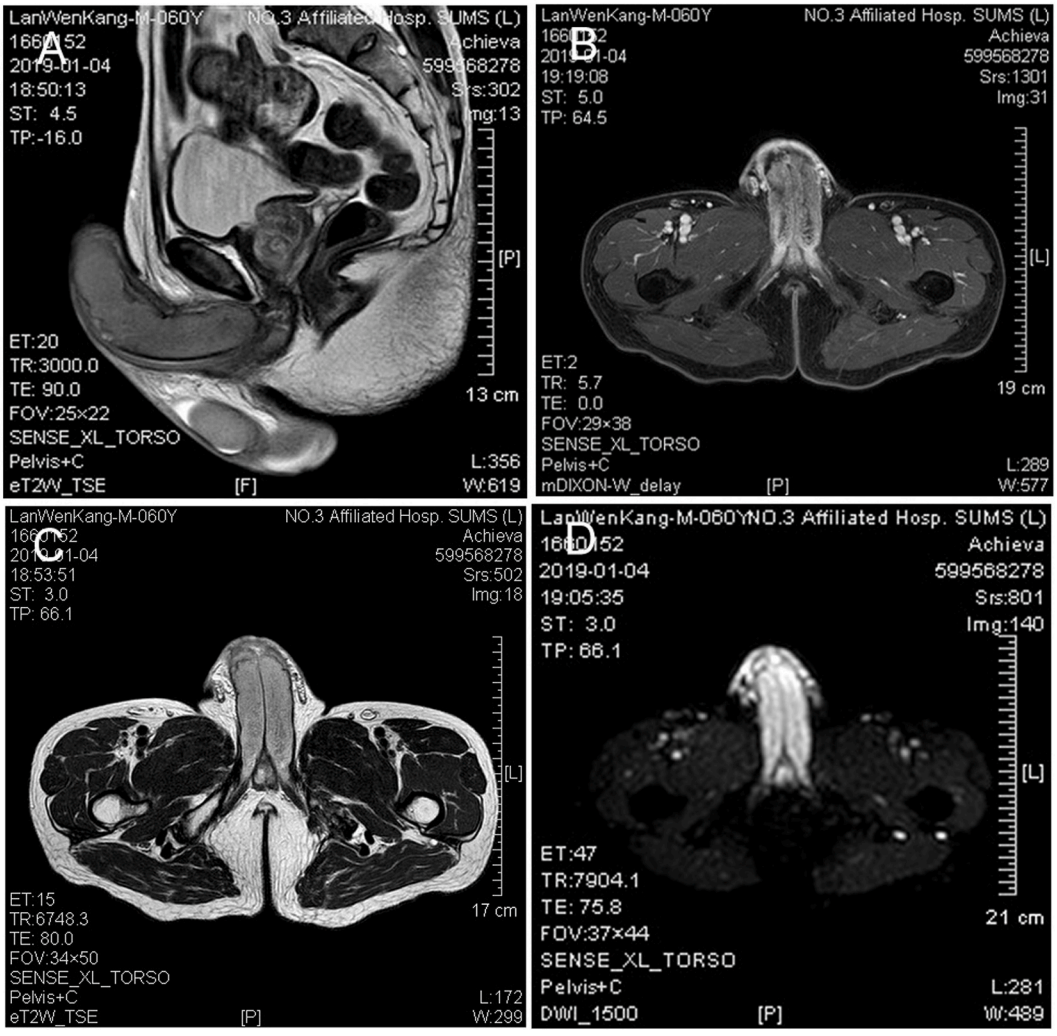

Fig. 2. Contrast enhanced MRI image of pelvis showed that bilateral corpus cavernosa was engorged and heterogeneous enhanced, and the fascia of penis was thickening and focal enhanced.

indicated the metastasis of clear-cell RCC. The tumor profiling (Foundation One CDx) was suggested and the result identified the cancer-associated mutation of MET, TERT and RAC1. The PD-L1 of tumor was negative, and Tumor Mutational Burden (TMB) was low.

Sunitinib and radiation therapy was recommended for the patient. The patient refused radiation therapy. However, sunitinib had limited activity for his priapism and tumor progression. One month later, the local recurrence and the metastatic lesion in liver, lung, retroperitoneal lymph nodes, greater omentum were found by a contrast-enhanced CT. The patient's general condition gradually deteriorated, and he died of

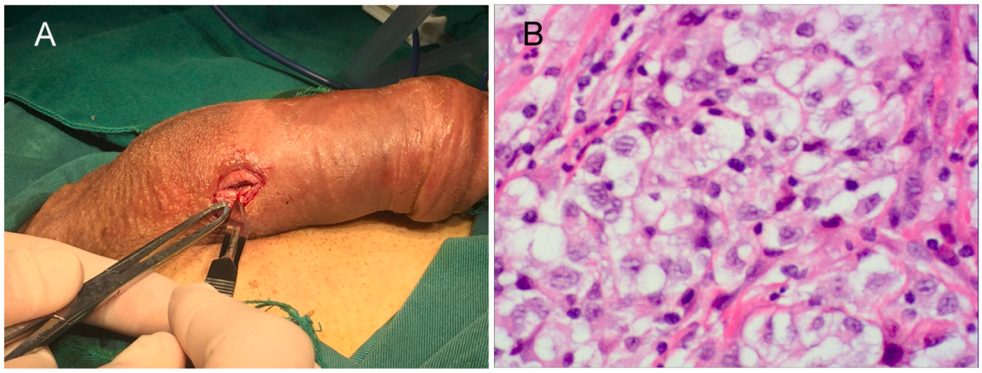

Fig. 3. A The tissue biopsy from corpus cavernosa was performed. The penis was enlarged, tender, and semierect without focal mass. B On hematoxylin and eosin staining, nests of cells with clear cytoplasm, enlarged hyperchromatic nuclei, and indistinct nucleoli infiltrated corpora cavernosa venous spaces and fibromuscular stroma.

the metastatic disease related respiratory and circulatory failure three months after the initial presentation.

3. Discussion

It has been reported that malignant priapism was caused by the metastasis of several solid tumors. However, the priapism, as the first clinical manifestation after the management of the primary tumor, induced by the metastatic RCC to penis is a rare phenomenon. When the patients present with non-ischemic priapism and have no history of penile injury, blood dyscrasias (especially sickle cell disease), chronic myeloid leukemia, use of intracavernous injections, psychotropic drugs or other known etiologies of priapism, the underlying malignant process should be considered, especially the elderly patients or the patients with a history of tumor. Many of the previously reported cases of priapism secondary to RCC were delayed the diagnosis, illustrating the importance of maintaining a high degree of clinical suspicion on malignant priapism.[2] The key point of diagnosis is obtaining the tissue for pathological examination.

Of the reported cases, left kidney was identified as the primary site of carcinoma in more than half cases.[2] It has been explained that RCC metastasizing to penis via retrograde venous flow seems a likely etiology despite the spermatic vein do not normally anastomose with the dorsal venous system.[2,3] In addition, the primary RCC of this Case contained two components, clear cell RCC and papillary RCC. However, the clear cell RCC, not papillary RCC, metastasized to penis. The underlying mechanism needed to be further explored.

Several previous reports indicated that the metastatic lesion of penis secondary to malignant tumor usually involved corpus cavernosa, but the invasion of corpora spongiosum was rare.[4] Moreover, the arterial supply of corpus cavernosa was usually nor affected. However, Nezu FM et al.[3] have reported that a case with malignant priapism as the initial clinical manifestation of metastatic RCC involved both corpora cavernosum and spongiosum.

Most studies suggested that the malignant priapism of RCC, probably represented evidence of more advanced stage of disease and usually indicated a poor prognosis.[1] Despite aggressive surgical treatment combined with radiation therapy, chemotherapy and/or targeted therapy, the reported longest survivor was only sixteen months compared with 5-year survival rate of 10% in metastatic RCC.[1,5] Once the diagnosis is established, the treatment is guided according to the management of metastatic RCC. In our Case, only more than one month later after the occurrence of priapism, the widespread metastatic lesions of RCC were confirmed. The patient died approximately three months after presentation.

3. Conclusions

The development of priapism, although rare, is sometimes a clue for clinicians to be aware of the symptom induced by the metastasis of renal carcinoma. Usually, it represents a more advanced stage of disease and carries a poor prognosis. The therapy is only palliative.

Acknowledgements

We acknowledge Professor Xiangfu Zhou from the department of Urology of the Third Affiliated Hospital of Sun Yat-Sen University.

Abbreviations

CEA	carcinoma embryonic antigen
AFP	alpha-fetoprotein
PSA	prostate-specific antigen
TMB	tumor mutational burden
CT	computed tomography
RCC	renal cell carcinoma

Funding

No funding was obtained for this work.

Authors' contributions

Hailun Zhan conceptualized the analysis. Fei Yang collected data for this analysis. Yun Luo wrote the manuscript. Jiarong Cai and Wenbiao Li provided critical appraisal of the manuscript.

All authors read and approved the final draft of manuscript.

References

1. Escribano G, Allona A, Burgos FJ, Garcia R, Navio S, Escudero A. Cavernosography in diagnosis of metastatic tumors of the penis: 5 newcases and a review of the literature. *J Urol.* 1987;138(5):1174–1177.
2. Sarma DP, Woods 3rd AL, Rodriguez Jr FH, Mendoza J, Weilbaecher TG. Priapism as the presenting feature of renal cell carcinoma. *J Surg Oncol.* 1985;28(2):103–107.
3. Nezu FM, Dhir R, Logan TF, Lavelle J, Becich MJ, Chancellor MB. Malignant priapism as the initial clinical manifestation of metastatic renal cell carcinoma with invasion of both corpora cavernosum and spongiosum. *Int J Impot Res.* 1998;10(2):101.
4. Powell BL, Craig JB, Muss HB. Secondary malignancies of the penis and epididymis: a Case report and review of the literature. *J Clin Oncol.* 1985;3(1):110–116.
5. Pal S, Gong J, Mhatre SK, et al. Real-world treatment patterns and adverse events in metastatic renal cell carcinoma from a large US claims database. *BMC Canc.* 2019;19(1):548.