Case Report

Embryonal rhabdomyosarcoma of prostate combined with prostatic abscess in an adult patient: a case report and literature review

Kaifeng Mao[1#], Peifeng Li[2#], Yuan Yuan[1], Xiang Yi[1], Jiaqi Fang[1], Jiabing Yuan[1], Zhangjun Cao[1], Zhenquan Lu[1], Richard Lo[1], Bingfeng Luo[1]

[1]Division of Urology, Department of Surgery, The University of Hong Kong-Shenzhen Hospital, Shenzhen, China; [2]Division of Urology, Department of Surgery, The Sixth People's Hospital of Huizhou, Huizhou, China

Contributions: (I) Conception and design: K Mao, B Luo; (II) Administrative support: Z Lu; (III) Provision of study materials or patients: R Lo, B Luo; (IV) Collection and assembly of data: P Li, Y Yuan, X Yi, J Fang, J Yuan; (V) Data analysis and interpretation: K Mao, Z Cao, B Luo; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

[#]These authors contributed equally to this work.

Correspondence to: Bingfeng Luo, MD, PhD. Division of Urology, Department of Surgery, The University of Hong Kong-Shenzhen Hospital, Haiyuan 1st Road, Futian District, Shenzhen 518053, China. Email: luobf@hku-szh.org.

Background: Embryonal rhabdomyosarcoma (ERMS) of the prostate is a rare and aggressive malignant tumor in adults with a poor prognosis in general. The main presenting symptoms are dysuria and acute urinary retention. Prostate ERMS is easily misdiagnosed, which leads to delays in treatment.

Case Description: We report the case of a previously healthy 31-year-old man who presented with dysuria for 3 months. Digital rectal examination (DRE), ultrasound, and computed tomography (CT) were suggestive of the possibility of a 2.0 cm × 3.0 cm abscess in the left lobe of the prostate. Prostate-specific antigen (PSA) was at a normal level, and C-reactive protein (CRP) concentration was elevated. The patient underwent surgical drainage by transurethral incision of the prostatic abscess. There was a significant improvement in the maximal urinary flow rate on postoperative day 5. However, the patient presented with acute urinary retention again on the 20th postoperative day. DRE and pelvic magnetic resonance imaging (MRI) showed a recurrence of an enlarged mass in the left lobe of the prostate. PSA level was again normal. Cystoscopy revealed a large neoplasm arising from the prostate obstructing the bladder outlet, and a diagnostic transurethral resection of this prostate tumor was performed. Histopathology revealed the mass to be an embryonic rhabdomyosarcoma. Subsequently, the patient was transferred to the Oncology Department and received chemoradiotherapy. After chemoradiotherapy, the size of the prostate became normal. Unfortunately, the patient deteriorated rapidly and eventually succumbed to multiple organ failure 1 year after the initial presentation.

Conclusions: Due to its non-specific presenting symptoms, prostate ERMS can be easily misdiagnosed, which can lead to treatment delay. Multimodality approaches, including neoadjuvant/adjuvant chemotherapy, radiotherapy, and radical resection, can improve the survival rate and reduce ERMS patient mortality.

Keywords: Case report; embryonal rhabdomyosarcoma (ERMS); prostatic abscess; chemoradiotherapy

Submitted Mar 03, 2024. Accepted for publication Jul 22, 2024. Published online Sep 23, 2024.
doi: 10.21037/tau-24-117
View this article at: https://dx.doi.org/10.21037/tau-24-117

© AME Publishing Company.

Introduction

Embryonal rhabdomyosarcoma (ERMS), the most common tissue subtype of rhabdomyosarcoma in the prostate, is a rare and aggressive malignant tumor in adults (1). In contrast to children with ERMS, the prognosis for adults diagnosed with ERMS is poor (2). ERMS accounts for less than 0.1% of malignant prostate tumors (3,4). Because of the low ERMS incidence, currently, there is no standard treatment for this tumor type. The main presenting symptoms are dysuria and acute retention of urine secondary to bladder outlet obstruction (BOO). It may easily be misdiagnosed as a prostatic abscess, prostatic hyperplasia, or other diseases in clinical practice, leading to delayed treatment due to non-specific symptoms (2,5). Therefore, early diagnosis and timely comprehensive treatment can improve patients' survival rate and quality of life. Herein, we present a 31-year-old man, who was diagnosed with ERMS of the prostate combined with a prostatic abscess and discuss the diagnosis, differential diagnosis, and treatment strategy of the disease. We present this article in accordance with the CARE reporting checklist (available at https://tau.amegroups.com/article/view/10.21037/tau-24-117/rc).

Case presentation

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Declaration of Helsinki (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

A previously healthy 31-year-old man presented to our hospital with a 3 months history of dysuria. After admission, an indwelling urinary catheter was inserted because of acute urine retention. Digital rectal examination (DRE) showed an enlarged prostate with mild tenderness. The ultrasound indicated prostatic enlargement with a hypoechoic lesion measuring 2.0 cm × 3.0 cm in the left lobe, raising the possibility of a prostatic abscess. Computed tomography (CT) confirmed the ultrasound results, and showed that the size of the prostate was 4.5 cm × 5.0 cm × 5.7 cm, of which the size of the abscess in the left lobe of the prostate was about 2.0 cm × 3.0 cm (*Figure 1A*). Prostate-specific antigen (PSA) was at a normal level (0.482 ng/mL, reference range, 0.0–4.0 ng/mL). Complete blood count on admission revealed a total leucocyte count of 7.18×10^9/L (neutrophil count 4.5×10^9/L). Inflammatory biomarkers showed a C-reactive protein (CRP) concentration of 15.7 mg/L (reference range, 0.0–10.0 mg/L) and the procalcitonin (PCT) level was less than 0.05 ng/mL (reference range, 0.05–0.1 ng/mL). A urine culture was negative as well as an acid-fast bacillus stain. Liver and kidney function tests were unremarkable. The patient underwent immediate surgical drainage by transurethral incision of the prostatic abscess. During the operation, purulent fluid was seen coming from the left lobe of the prostate. The Foley catheter was removed on postoperative day 5 and the patient voided with a significant improvement in the peak flow rate. Uroflowmetry showed a maximal flow rate (Qmax) of 17 mL/s after the operation. A follow-up magnetic resonance imaging (MRI) of post-drainage of the prostatic abscess showed interval improvement in the left prostatic abscess (*Figure 1B*). The patient was continued on oral cefuroxime for 2 weeks after discharge.

The patient was admitted again on the 20th postoperative day, due to acute retention of urine. DRE indicated an enlarged, firm prostate. Pelvic MRI showed a recurrence of an enlarged mass in the left lobe of the prostate with no pelvic or para-aortic lymph node enlargement, and the size of the prostate was 4.5 cm × 5.6 cm × 7.2 cm (*Figure 2A*). Cystoscopy showed a large neoplasm arising from the prostate that obstructed the bladder outlet (*Figure 2B*). Diagnostic transurethral resection of this prostate tumor was performed. The histopathology of the tumor findings revealed ERMS (*Figure 3*). The patient was transferred to the oncology department and received systemic therapy that consisted of 5 cycles of chemotherapy with VAC (vincristine, actinomycin D, and cyclophosphamide) and 4

Highlight box

Key findings

- We reported a rare case of prostate embryonal rhabdomyosarcoma (ERMS) combined with prostatic abscess in an adult patient.

What is known and what is new?

- Due to its non-specific presenting symptoms, prostate ERMS can be easily misdiagnosed, which can lead to treatment delay.
- Multimodality approaches, including neoadjuvant/adjuvant chemotherapy, radiotherapy, and radical resection, can improve the survival rate and reduce ERMS patient mortality.

What is the implication, and what should change now?

- It's necessary to distinguish prostate ERMS from prostatic abscess, prostatic hyperplasia, or other diseases to avoid misdiagnosis and provide timely and effective treatment.

© AME Publishing Company.

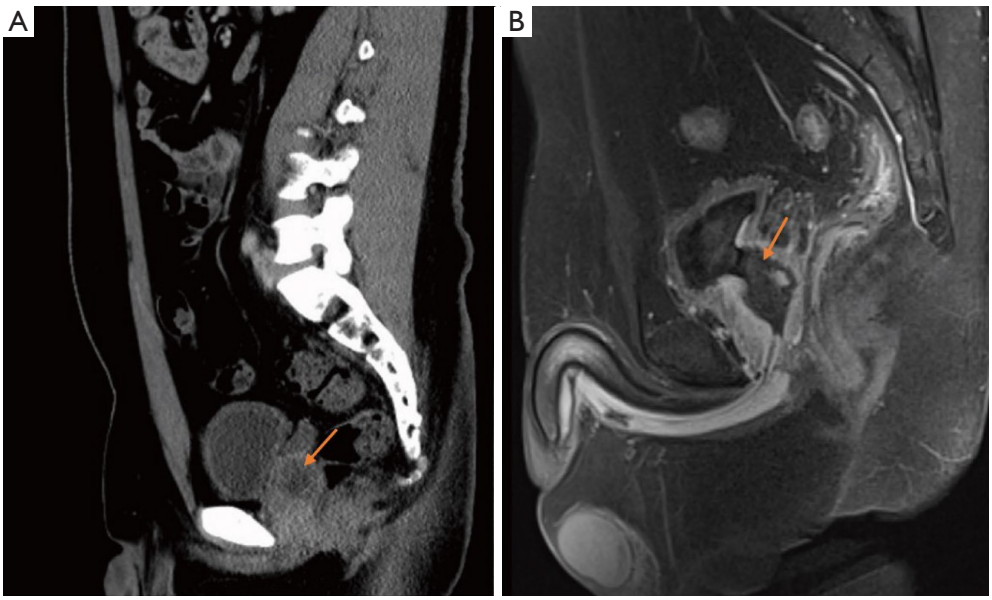

Figure 1 Pelvic CT and MRI examinations. Arrows indicate the prostatic abscess. (A) CT imaging indicated a 2.0 cm × 3.0 cm abscess in the left lobe of the prostate, and the size of the prostate was 4.5 cm × 5.0 cm × 5.7 cm. (B) A follow-up pelvic MRI showed the prostatic abscess disappeared and interval improvement in the prostate after drainage of the prostatic abscess. The size of the prostate was about 3.5 cm × 4.0 cm × 4.5 cm. CT, computed tomography; MRI, magnetic resonance imaging.

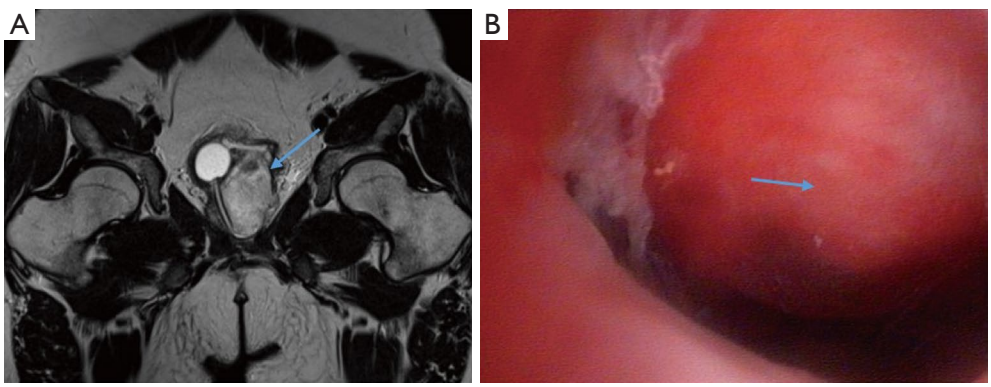

Figure 2 Pelvic MRI and cystoscopy examinations. Arrows indicate the large mass. (A) Pelvic MRI revealed a large mass developed again in the left lobe of the prostate after surgery in 20 days. The size of the prostate was 4.5 cm × 5.6 cm × 7.2 cm. (B) Cystoscopy showed a large neoplasm arising from the prostate that obstructed the bladder outlet. MRI, magnetic resonance imaging.

cycles of IE (ifosfamide and epirubicin) plus radiotherapy. Radiotherapy for prostate ERMS was delivered using 6 MV X-ray intensity-modulated radiation therapy (IMRT). Residual tumor was defined as the gross tumor volume (GTV), and the clinical target volume (CTV) was generated with 1 cm expansion from GTV but no less than the pre-chemotherapy extent, with modification beyond the anatomic barrier. The prescribed dose was 60 Gy in 30 fractions. After chemoradiotherapy, the size of the prostate became normal (4.4 cm × 3.2 cm × 2.1 cm). Unfortunately, the patient had a rapid deterioration and eventually died of distant metastasis and multiple organ

© AME Publishing Company.

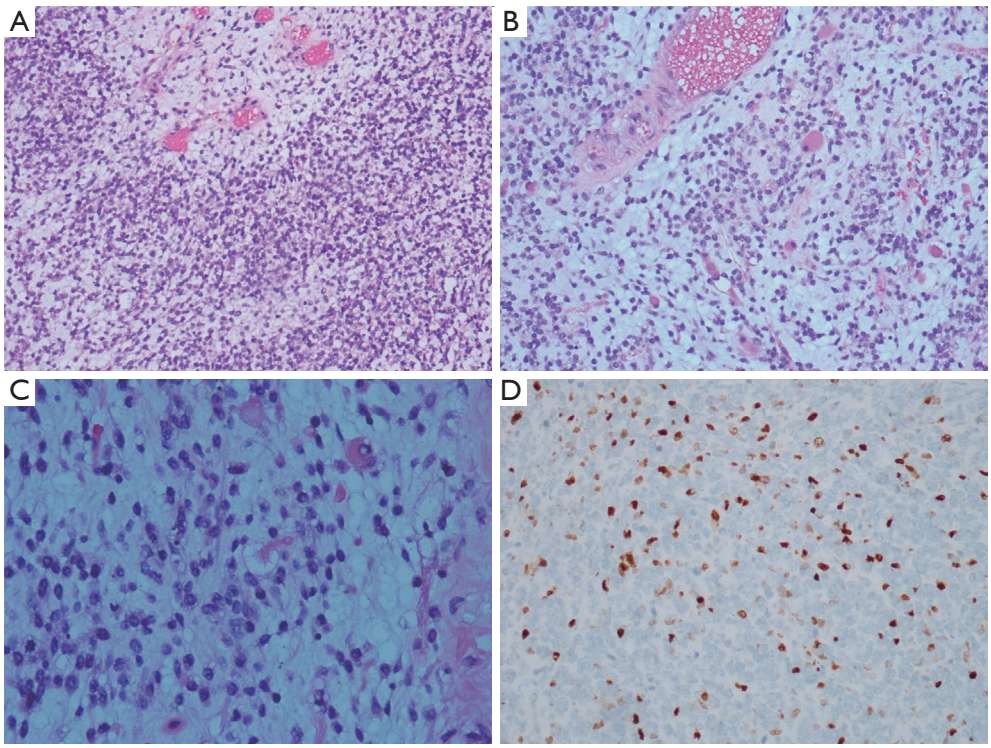

Figure 3 The histopathology of the tumor. (A) Patternless sheets of primitive round to spindled cells show alternating hyper- and hypocellular zones with focal myxoid stromal matrix (H&E stain, ×200). (B,C) Some tumor cells show rhabdomyoblastic differentiation with dense eosinophilic cytoplasm (H&E stain, ×200, ×400). (D) Tumor cells with positive nuclear staining for myogenin (immunohistochemical stain, ×200). H&E, hematoxylin-eosin.

failure 1 year after the initial presentation.

Discussion

ERMS of the prostate is a rare malignant tumor arising from mesenchymal-derived tissue of the prostate with an occurrence rate of less than 1% of all prostate tumors (5,6). Prostate ERMS is a common tumor that accounts for about 40% in infants and children, whereas it is extremely rare in adults, in which it is furthermore associated with more aggressive invasive behavior (2). Studies have demonstrated extensive locoregional involvement of the lung, liver, and bone, as well as lymph node involvement in the early stages (7,8). Musser *et al.* (9) reported the study results involving 38 patients with prostatic sarcomas at the Memorial Sloan Kettering Cancer Center. Of these 38, only 12 were diagnosed with rhabdomyosarcoma. Nineteen patients underwent surgery including cystoprostatectomy (n=9), radical prostatectomy (n=6), and pelvic exenteration (n=4), whereas 2 refused surgery. This study also confirmed that prostatic rhabdomyosarcoma was associated with poor cancer-specific survival. The presence of metastatic disease at the time of diagnosis is a poor prognostic indicator (5). Wang *et al.* (10) published the results of a study comprising 25 patients with prostatic sarcoma at West China Hospital in China, of which 6 were rhabdomyosarcoma cases with a median survival time of 28.6 months. The authors also demonstrated that at an age above 50 years, metastasis at the time of presentation and a lack of surgery with curative intent were independent predictors of an unfavorable outcome. Early diagnosis and multimodality treatment can provide patients with a higher probability of survival (9,11).

For most urologists, however, the experience of diagnosing ERMS is limited (3). Due to the non-specific symptoms in the early stage, the misdiagnosis rate of ERMS is very high, leading to a delay in treatment (5,10). Clinical symptoms of ERMS usually manifest themselves as dysuria, urinary retention, and seldom hematuria. DRE can be suggestive of prostatic enlargement. However, the normal serum PSA will not raise the possibility of taking prostatic

© AME Publishing Company.

ERMS into account (11). The differential diagnosis of ERMS includes prostatic enlargement from hyperplasia, leiomyosarcoma, malignant peripheral nerve sheath tumor, liposarcoma, and prostatic abscess. We reported this case which presented the clinical features of a prostatic abscess that increased the difficulty in the identification of ERMS. Therefore, the initial clinical diagnosis of the patient was considered as a prostatic abscess based on the findings of the transrectal ultrasound (TRUS) and CT scan (2). On transrectal ultrasonography, irregular edges and low echo areas are important features of a prostatic abscess (12,13). In addition, TRUS-guided drainage and transurethral incision of the prostatic abscess can be used to identify the diagnosis and treatment of prostatic ERMS (10).

At present, treatment options of ERMS include chemotherapy, radiotherapy, and radical surgery. Recent studies report that old age, nodal metastasis, and tumor diameter greater than 5 cm are closely associated with poor prognosis (7,14). Radical resection with negative margins is the standard treatment for localized disease in adults. Due to the difficulty of achieving complete resection, the focus in ERMS therapy shifted from radical resection to the use of combined chemotherapy and radiotherapy to achieve oncologic control. Neoadjuvant chemotherapy has been shown to significantly improve surgical outcomes, decrease morbidity, and preserve organ function in children and adolescent patients (15). Treatment intensity and timing should be varied according to the risk classification. VAC together form the standard combination of chemotherapeutic agents for rhabdomyosarcoma treatment in America (16). Recent research shows that the use of irinotecan instead of actinomycin D and cyclophosphamide in combination with vincristine for intermediate-risk rhabdomyosarcoma can reduce the toxicity of VAC regimens while ensuring efficacy (17). Brachytherapy, intensity-modulated proton therapy, and photon therapy have all been reported in small series and were effective components of the therapeutic ERMS strategy (18,19). Longer follow-up is needed to further evaluate the functional outcomes of radiation therapy.

Ball *et al.* (20) reported a multimodality approach with a favorable cancer-specific and recurrence-free survival compared to historic and contemporary series of surgery alone. In 2016, The Rare Cancer Network launched a multicenter study including 6 American and 10 European centers with a total of 61 patients. Those who received curative multimodality therapy (surgery and/or radiation therapy ± chemotherapy), demonstrated an improved 5-year local control (59% *vs.* 13%, P=0.009) and 5-year overall survival (59% *vs.* 28%, P=0.02) compared to those treated palliatively (4). The current treatment of bladder/prostate RMS (BPRMS) in China emphasizes that the bladder should be retained through multimodality treatment such as neoadjuvant/adjuvant chemotherapy and/or radiation, and an early radical organ removal operation should be avoided (21,22). Radical extirpative surgery, however, should be considered in case standard comprehensive treatment fails to control the tumor adequately. Pelvic lymphadenectomy is not routinely performed during the operation. Previous study revealed that negative surgical margins and the absence of metastatic disease at presentation were factors predictive of long-term survival (23).

Conclusions

Due to its non-specific presenting symptoms, prostate ERMS can be easily misdiagnosed, which can lead to treatment delay. Multimodality approaches, including neoadjuvant/adjuvant chemotherapy, radiotherapy, and radical resection, can improve the survival rate and reduce ERMS patient mortality.

Acknowledgments

Firstly, the authors would like to thank the patient for providing his consent to present his case and accompanying images for the publication of this case report. Secondly, the authors would like to express their gratitude to EditSprings (https://www.editsprings.cn/) for the expert linguistic services provided.

Funding: None.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://tau.amegroups.com/article/view/10.21037/tau-24-117/rc

Peer Review File: Available at https://tau.amegroups.com/article/view/10.21037/tau-24-117/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tau.amegroups.com/article/view/10.21037/tau-24-117/coif). The authors have no conflicts of interest to declare.

© AME Publishing Company.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Declaration of Helsinki (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Open Access Statement: This is an Open Access article distributed in accordance with the Creative Commons Attribution-NonCommercial-NoDerivs 4.0 International License (CC BY-NC-ND 4.0), which permits the non-commercial replication and distribution of the article with the strict proviso that no changes or edits are made and the original work is properly cited (including links to both the formal publication through the relevant DOI and the license). See: https://creativecommons.org/licenses/by-nc-nd/4.0/.

References

1. Olivas AD, Antic T. Rhabdomyosarcoma of the Adult Prostate: A Case Report With Complete Molecular Profile. Int J Surg Pathol 2020;28:92-8.
2. Malik SA, Bhat AH, Lone SS. Prostatic rhabdomyosarcoma in the adult, A case report. Urol Case Rep 2022;42:102027.
3. Skapek SX, Ferrari A, Gupta AA, et al. Rhabdomyosarcoma. Nat Rev Dis Primers 2019;5:1.
4. De Bari B, Stish B, Ball MW, et al. Adult prostatic sarcoma: A contemporary multicenter Rare Cancer Network study. Prostate 2017;77:1160-6.
5. Alenezi S, Zahir M, Alenezi J. Embryonal rhabdomyosarcoma of the prostate in a young male: A rare case report. Int J Surg Case Rep 2023;106:108228.
6. Yang B, Yuan J, Yu S, et al. A case report of primary mesenchymal sarcoma of the prostate and the thoughts on its treatment. Asian J Surg 2023;46:4934-5.
7. Luo H, Weng X, Zhong Z, et al. Spindle cell embryonal rhabdomyosarcoma of the prostate in an adult patient: a case report and review of the literature. J Int Med Res 2024;52:3000605241234755.
8. Wang FF, Zhang XN, Wang XJ. Magnetic resonance imaging analysis of embryonal rhabdomyosarcoma of the prostate. Technol Health Care 2023;31:2243-50.
9. Musser JE, Assel M, Mashni JW, et al. Adult prostate sarcoma: the Memorial Sloan Kettering experience. Urology 2014;84:624-8.
10. Wang X, Liu L, Tang H, et al. Twenty-five cases of adult prostate sarcoma treated at a high-volume institution from 1989 to 2009. Urology 2013;82:160-5.
11. Ding B, Zhang Y, Hu W, et al. Adult primary prostate sarcoma: A multi-center cohort study and comparison between Chinese and American cases. Asian J Surg 2021;44:247-53.
12. Shebel HM, Farg HM, Kolokythas O, et al. Cysts of the lower male genitourinary tract: embryologic and anatomic considerations and differential diagnosis. Radiographics 2013;33:1125-43.
13. Arrabal-Polo MA, Jimenez-Pacheco A, Arrabal-Martin M. Percutaneous drainage of prostatic abscess: case report and literature review. Urol Int 2012;88:118-20.
14. Haduong JH, Heske CM, Allen-Rhoades W, et al. An update on rhabdomyosarcoma risk stratification and the rationale for current and future Children's Oncology Group clinical trials. Pediatr Blood Cancer 2022;69:e29511.
15. Ellerkamp V, Schmidt A, Warmann SW, et al. Detailed functional results after bladder-preserving surgery and high-dose-rate brachytherapy in pediatric bladder/prostate rhabdomyosarcoma. J Cancer Res Clin Oncol 2023;149:3161-70.
16. Hosoi H. Current status of treatment for pediatric rhabdomyosarcoma in the USA and Japan. Pediatr Int 2016;58:81-7.
17. Miyachi M, Tsuchiya K, Hosono A, et al. Phase II study of vincristine, actinomycin-D, cyclophosphamide and irinotecan for patients with newly diagnosed low-risk subset B rhabdomyosarcoma: A study protocol. Medicine (Baltimore) 2019;98:e18344.
18. Yao W, Schweitzer N, Biswal N, et al. Impact of bowel and rectum air on target dose with robustly optimized intensity-modulated proton therapy plans. Acta Oncol 2020;59:1186-92.
19. Fowler KG, Mohindra P, Kim A, et al. Foley Catheter as a Vaginal Stent in a Toddler with Vaginal Rhabdomyosarcoma. J Pediatr Adolesc Gynecol 2018;31:315-7.
20. Ball MW, Sundi D, Reese AC, et al. Multimodal Therapy in the Treatment of Prostate Sarcoma: The Johns Hopkins Experience. Clin Genitourin Cancer 2015;13:435-40.
21. Lobo S, Gaze MN, Slater O, et al. Bladder function after conservative surgery and high-dose rate brachytherapy

for bladder-prostate rhabdomyosarcoma. Pediatr Blood Cancer 2022;69:e29574.
22. Martelli H, Borrego P, Guérin F, et al. Quality of life and functional outcome of male patients with bladder-prostate rhabdomyosarcoma treated with conservative surgery and brachytherapy during childhood. Brachytherapy 2016;15:306-11.
23. Agarwal DK, Miest TS, Granberg CF, et al. Pediatric Robotic Prostatectomy and Pelvic Lymphadenectomy for Embryonal Rhabdomyosarcoma. Urology 2018;119:143-5.

Cite this article as: Mao K, Li P, Yuan Y, Yi X, Fang J, Yuan J, Cao Z, Lu Z, Lo R, Luo B. Embryonal rhabdomyosarcoma of prostate combined with prostatic abscess in an adult patient: a case report and literature review. Transl Androl Urol 2024;13(9):2146-2152. doi: 10.21037/tau-24-117

© AME Publishing Company.