

## Malignant Priapism Caused by Metastases to the Penis in a Patient with Prostate Cancer

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### Abstract

Penile squamous cell carcinoma is a rare malignancy and secondary malignancies to the penis are extremely rare and represent a unique challenge for urologists. We report a case of an 80-year-old patient with 3-month painful priapism from locally advanced prostate adenocarcinoma and successfully treated with surgery – total penectomy without emasculation and open radical prostatectomy with suprapubic definitive cystostomy. What becomes exceptional in our clinical case is that the penile metastases were the first disease symptom, since until this moment the primary prostate tumor has been silent. So far, the patient is alive and well after 14 months of follow-up. To the best of our knowledge, this is the first reported Bulgarian case describing malignant priapism in metastatic prostate cancer.

**Keywords:** Malignant priapism; Prostate cancer; Penile metastases; Diagnosis; Surgery.

### Introduction

The penis has a rich and complex vascular and lymphatic supply, but it is surprising that metastases to the penis are such a rare clinical entity. The primary lesion is nearly 75% of pelvic origin and in order of its frequency are the bladder (34.7%), prostate (29.8%), rectosigmoid (15.7%) and kidney (6.5%). Malignant priapism is the main symptom in 40% of patients [1]. The term was originally first used by Peacock in 1938 to describe a condition of painful nonsexual induration and erection of the penis due to metastatic infiltration by a neoplasm [2]. Penile metastatic invasion, regardless of its origin has traditionally been associated with advanced disease and a poor prognosis. Its treatment remains undefined and unclear.

The rarity of this event motivated us to describe this interesting case of malignant priapism as the first sign of locally

advanced prostate adenocarcinoma and discuss diagnosis, treatment, and prognosis of the disease process.

### Case presentation

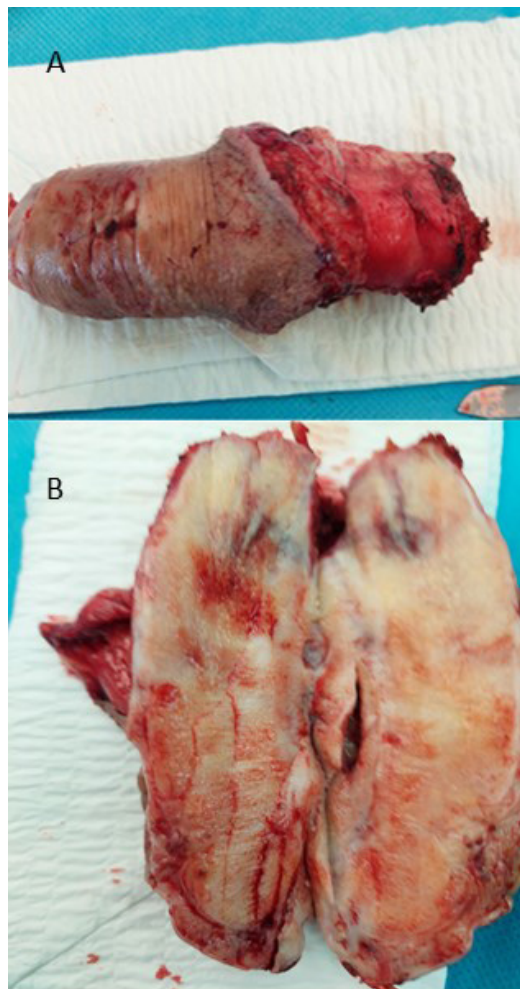
An 80-year-old male presented with painful persistent erection for 3 months prior to the present admission visit in March 2022 (not past medical history for penile cancer). He also complained of dysuria, difficult urination, and hematuria two months earlier. The patient looked well (ECOG performance status 0) and his physical examination revealed rigid penile shaft and glans with no pain on palpation and clinically negative groins – no palpable inguinal lymph nodes (cNO). The glans appeared abnormal with exophytic and ulcerating lesion (Figure 1). Digital rectal examination showed an enlarged prostate but did not raise suspicion of prostate cancer. The indwelling catheter was inserted because of urinary retention. Laboratory

findings showed elevated serum level of prostate specific antigen (PSA) – 8.269 ng/ml. Computed tomography (total body scan) showed no gross abnormalities (01.04.22).

We present sequentially the overview of the diagnostic and treatment process and the patient’s disease outcome for a follow-up period of 19 months after his first visit to doctor (Table 1).



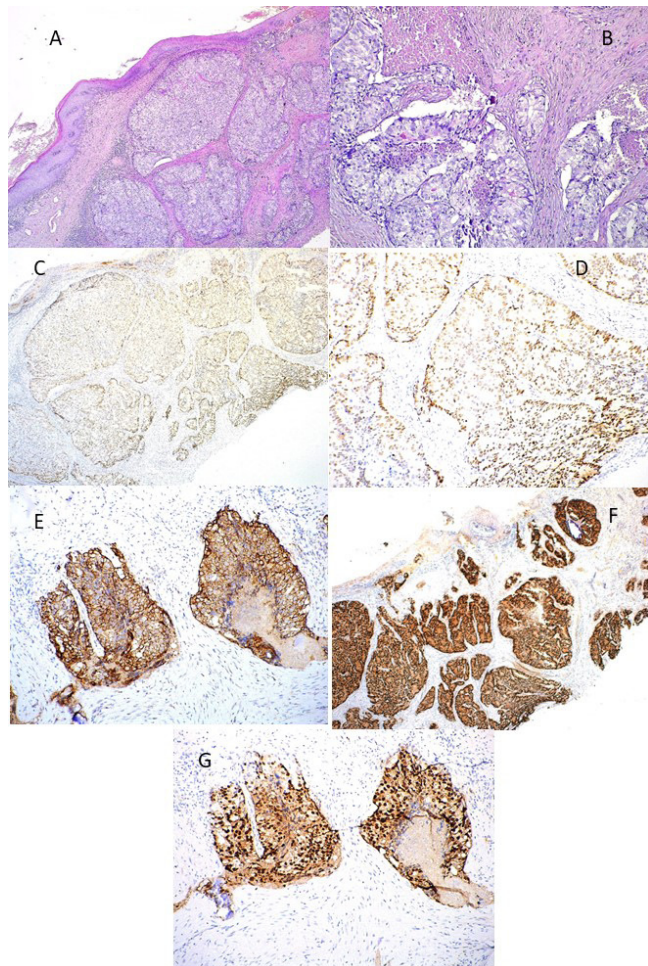
**Figure 1:** Malignant priapism, exophytic ulcerating lesion on glans penis.



**Figure 2:** Gross appearance of the resected specimen. 3A macroscopic view, 3B longitudinal section.

**Table 1:** The timeline of diagnostic and therapeutic interventions and outcome for the case.

Intervention	Date	Result
1. Urethroscopy	02.04.2022	No evidence of bladder tumor
2. Transrectal ultrasound-guided prostate biopsy (total PSA 8.269 ng/ml)	05.04.2022	Benign prostatic hyperplasia
3. Excisional surgical biopsy of an exophytic lesion of the glans penis	12.04.2022	Microscopic findings. Penile metastases from high-grade adenocarcinoma with a probable primary site from the colon (routine histopathological examination and immunohistochemistry)
4. Fibrocolonoscopy	04.05.2022	No evidence of tumor
5. Incisional biopsy on the dorsal surface of the penis. During the procedure the cavernotomy did not demonstrate any significant bleeding, raising concerns for a vascular etiology of the priapism	02.06.2022	Microscopic findings. If the possibility of primary colon adenocarcinoma is clinically ruled out, it should be assumed that the carcinoma is primary penile adenocarcinoma originating from the periurethral glands
6. Total penectomy with perineal urethral reconstruction was the treatment of choice	02.07.2022	Gross appearance of the surgically resected specimen (Figure 2). Microscopic findings. Corpora cavernosa and corpus spongiosum infiltrated by a metastatic prostatic adenocarcinoma; the edges of resection are free from carcinoma (Figure 3)
7. Repeated transrectal ultrasound-guided prostate biopsy (total PSA 13.54 ng/ml)	04.08.2022	Microscopic findings. Prostate acinar adenocarcinoma, Gleason 8 (4+4), grade group by WHO 4
8. Open retropubic prostatovesiculectomy and suprapubic cystostomy	13.08.2022	Microscopic findings – prostate acinar adenocarcinoma, Gleason 8 (4+4), grade group by WHO 4 at sites of perineural and perivascular invasion; left and right seminal vesicle involved by the described carcinoma (TNM: pT3bNxM1; R0; N+pV+)
9. Hormonal therapy was offered for metastatic prostate adenocarcinoma	Sept 2022	Antiandrogens
10. As a whole follow-up period lasted 19 months after his first visit to a doctor		The effects were good after several months of treatment and at control examination four months later the patient was well and showed no signs of disease progression (General health status – ECOG performance status 1). Follow-up of 14 months.



**Figure 3:** Histological and immunohistochemical characteristics of the malignant priapism. **3A.** H.E.  $\times 25$  Stratified squamous keratinizing epithelium, subepithelial nests, and brands of atypical cells with bright eosinophilic cytoplasm and vesicular nuclei; **3B.** H.E.  $\times 100$  Tumor cells with bright eosinophilic cytoplasm with vesicular nuclei and visible nucleoli and mitoses in some places. Presence of “dirty” necrosis in glandular lumens, microfocus of necrosis, single acini; **3C.** CDX2  $\times 25$  and **3D.** CDX2  $\times 100$  positive nuclear expression in tumor tissue; **3E.** AMACR  $\times 100$  positive cytoplasmic expression in tumor tissue; **3F.** NKX3.1  $\times 25$  and **3G.** NKX3.1  $\times 100$  strongly positive nuclear expression in tumor tissue.

### Discussion

Several solid tumors have been known to metastasize to the penis and cause malignant priapism. The prostate and bladder are the two most common primary organs [3,4]. Metastases to the penis are a manifestation occurring late in advanced stage of all types of tumors and is often associated with short survival. The primary tumors with metastases to the penis occur most frequently in the age group 60 to 80 [5]. Based on these observations we present a case of a patient with metastatic penile cancer from prostate adenocarcinoma since such patients were reported in very few cases. Several investigators have described possible metastatic mechanisms for primary tumor spread to the penis – local direct infiltration (from malignancies of the bladder, prostate and rectum), arterial embolism, retrograde venous and lymphatic spread (the most common way due to vast communication between the pelvic organs and dorsal venous system of the penis via retrograde lymphatic flow) or instrumental spread (cysto-urethroscopy, transurethral resections of prostate or bladder) [6]. In our specific case the ischemic priapism was due to the invasion of cancer cells into both corpora cavernosa blocking the venous draining, without

blocking the arterial flow causing a complete blockage and consequent priapism. As in our patient, prostate cancer is among the most common primary malignancies metastasizing to the penis, accounting for one-third of all cases [7].

Regardless of the site of primary tumor, the most common clinical symptoms according to their frequency were malignant priapism and metastatic penis enlargement (20-83%), urinary retention, penile nodules and ulcerations, perineal pain, diffuse or localized penile swelling, dysuria, and hematuria [1,6,7]. In our case malignant priapism was the initial clinical presentation of metastatic prostate carcinoma.

Diagnosis of metastases to the penis is usually made by biopsy of the corpora. For any malignant priapism, however, corporal biopsies are considered the most direct method of evaluating the primary site of neoplasm [6]. Thus, it is essential to obtain material as early as possible to differentiate between metastases to the penis and primary penile tumors.

Various treatment modalities, all of which may be accepted as palliative, consist of local excision, partial or total penectomy, suprapubic urinary diversion, radiotherapy, and chemotherapy. To date, no method has been shown to be superior to others, except for wide local excision (in case of single nodes) and total penectomy [8,9].

The rationale to treat this patient with this rare cancer was the ECOG performance status 0 and no comorbidities. In our case, radical surgery of penis (total penectomy) and prostate (radical prostatectomy) was applied successfully due to his good general health status. Suprapubic diversion of urine was taken after radical prostatectomy.

Immunohistochemical examination (IHC) with p63, CK-34bE12, PSA, TTF1, CK7, NapsinA, CK20 showed negative expression; CDX2-moderately expressed nuclear signal in tumor parenchyma suggested penile metastases from colorectal adenocarcinoma considering a negative previous needle prostate biopsy. Cytoplasmic positivity for AMACR did not support prostatic adenocarcinoma due to its non-specificity and positivity also in colorectal carcinomas. In the performed tests of serum PSA, the same was high (8.483-11.382 ng/ml). The result of the colonoscopy for tumor lesion was negative. This necessitated the expansion of the IHC panel with NKX3.1, traditionally used as a diagnostic biomarker for prostate cancer and other metastatic lesions originating in the prostate, was positive with strong nuclear staining in the tumor parenchyma.

Clinical data and immunophenotype led to assumption to be penile metastasis from prostatic acinar adenocarcinoma with high Gleason score. From the performed immunohistochemical interpretation, we concluded that PSA is an unreliable marker and should be carefully used in routine practice, as well as the positive CDX2 expression in prostate adenocarcinoma metastasis [7]. In unclear metastatic lesions, an expanded panel of IHC with specific markers, as well as good collaboration between clinicians and pathologists should be made.

As far as we know, this is the first case in our country describing not only the difficulties in diagnostic approaches, but also the making of an informed treatment decision.

Nonetheless of the site of origin or subsequent management, most similar cases have shown very poor prognosis [10]. According to the published follow-up data, after initial treatment patients have an average survival time of approximately 9

months (range 6-18 months) [10,11]. Interestingly, patients with priapism as the presenting symptom from metastases originating from a non-urological malignancy had a worse prognosis compared to those with metastases from urological malignancies and without priapism [10]. After treatment completion and 14 months follow-up, our patient is alive and well (ECOG performance status 1). This case demonstrates the potential clinical benefit of early detection and then appropriate management as crucial factors to improve survival.

### Conclusion

The penis may be a site of metastases from numerous primary sites especially to old patients. They usually indicate that the primary tumor is at advanced stage and the prognosis is very poor. Corporal biopsies are considered an effective method for diagnosis of the primary tumor. Radical surgical treatment of both malignancies is an effective option for patients in good general health status; they should be monitored closely during treatment to avoid morbidity and followed-up postoperatively to establish signs of disease progression.

### Declarations

**Conflict of interest:** The authors have no conflict of interest to declare. No funding source was involved in this study.

**Ethical approval:** Our institutions do not require ethical approval for publishing case reports.

**Author contributions:** YD, SZ, DK, and ID were involved in diagnosis and surgical treatment of the patient, SS made histological analyses of the tissue specimens and provided the histological images used in the article, ID wrote the manuscript that was approved by all the authors.

**Informed consent:** The patient provided an informed consent statement.

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