Case of Malignant Priapism and Review of the Literature

Haris Ahmed, Sammy Elsamra, MD, and Mark Sigman, MD

Introduction

Cancer metastasis to the penis is rare and manifestation as a priapism is even more uncommon. We present the case of a 73 year old man with urothelial carcinoma of the bladder status-post radical cystectomy who presented with malignant priapism as the first sign of cancer recurrence.

Case Presentation

The patient was a 73 year-old Caucasian man who presented to the emergency room with penile edema and pain of unclear etiology. Three months prior he underwent a radical cystectomy, bilateral pelvic lymph node dissection, and ileal-loop urinary diversion for multifocal high grade T4aN2Mx urothelial carcinoma (with involvement into prostate and seminal vesicles and extensive lymphovascular invasion but negative margins). He was initially diagnosed with high grade non-muscle invasive urothelial bladder carcinomatwo years earlier and was noted to have progression to muscle invasive disease 18 months later despite treatment with intravesical **Bacillis Calme-Guerin** (**BCG**). The patient's exam was remarkable for penile edema and tenderness throughout corpora cavernosa and glans penis. Three months prior to procedure, a **computerized tomography** (**CT**) of the abdomen and pelvis revealed no evidence of metastatic disease. He was diagnosed with a malignant priapism vs. metastatic infiltration to penis and subsequently underwent **magnetic resonance imaging** (**MRI**) of his pelvis (and penis) revealing a focus of sacral metastasis but no definite penile metastasis. Penile edema and pain were refractory to corporeal cavernosal aspiration or phenylephrine injection and penile nerve blocks, respectively. Attempts at palliating the patient with large doses of continuous and **patient controlled analgesia** (**PCA**) narcotics were ineffective and effecting his mentation. Further, he developed some erythema on the penile shaft and a black eschar on the glans that was concerning for a cellulitis prompting treatment with vancomycin IV. Because of concerns associated with a high narcotic requirement and wet gangrene of the penis, he underwent a palliative radical penectomy three weeks after admission. Pathology revealed widespread urothelial carcinoma with lymphovascular invasion and extensive necrosis with positive proximal margins. His pain improved significantly. A subsequent CT of the abdomen and pelvis revealed metastatic disease to the lung, liver, and sacrum. He elected to be made comfort measures only and died seven weeks after admission.

Discussion

Priapism is the prolonged occurrence of penile erection in the absence of sexual stimuli. It may present as either low-flow or ischemic, when there is obstruction of venous drainage from the corpora cavernosa, or high-flow, due to increased arterial flow to the corpora. Priapism may be caused by numerous factors, the most common being medications such as alpha agonists, vasoactive agents, or antipsychotics, or any process that may increase the viscosity of blood such as polycythemia, hematologic malignancy, or sickle cell anemia.[1]

Metastasis of tumor to the penis can result in both ischemic, by occlusion of cavernosal outflow, or high-flow priapism, via arterial fistulization. This is extremely rare with roughly 100 cases reported to date according to a large contemporary review. The most common primary sites include the prostate, bladder, and recto-sigmoid, but reports of metastasis from kidney, testes, lung, stomach, bone, ureter, hepato-biliary, and urethra have been reported.[2,3]

The penis has a rich vascular supply, which it shares with nearby organs. Metastasis can occur via retrograde venous and lymphatic routes, arterial spread, direct extension, and possibly implantation secondary to instrumentation.[4] Diagnosis can be facilitated by CT and MR imaging, and is definitively made by either biopsy or intracorporeal aspiration.[5]

As invasion of the penis is generally by metastatic spread, it usually represents disseminated disease, and carries with it a poor prognosis. Patients are generally in poor health with other sites of spread arising before penile metastasis.[2,3] However, cases of isolated penile metastasis have been reported, occurring several years after treatment of the primary disease. Malignant priapism has also, in one case besides the present case, been reported as the initial indication of cancer recurrence.[4]

Treatment should be catered to the patient, focusing on their general health, the extent of metastasis, the severity of their symptoms, and their overall prognosis, as well as the type of priapism when present. Often treatment is limited in

Abstract

Malignant priapism, or priapism secondary to locally invasive or metastatic cancer, is a rarely reported phenomenon with a very poor prognosis. We report on a case of malignant priapism secondary to metastasis of urothelial carcinoma of the bladder in a 73 year old gentleman who underwent a radical cystectomy with negative margins but positive nodal disease three months prior to presentation. The rapid progression of disease and resultant demise of this patient is unusual even when compared to the limited available literature. Rapid progression to muscle invasion despite intravesical immunotherapy, histologic involvement of the prostate and seminal vesicles with extensive lymphovascular invasion, and the rapid development of metastasis are all consistent with the aggressive nature of a cancer that has metastasized to the penis. Further, while conservative penile sparing therapy may be a goal in patients with limited life expectancy, ongoing pain and infectious concerns may force the physician to resort to a penectomy.

these patients to palliative and supportive care. Priapism can be initially managed with intracavernosal irrigation and instillation of phenylephrine, as well as shunts. In cases of high-flow priapism, which can be caused by arterial rupture secondary to tumor invasion, embolization of the internal pudendal artery may be an option[6]. While pain is not a prominent feature in penile metastasis, it can be severe in the case of malignant priapism. Dorsal nerve section or total penectomy may be employed in cases of intractable pain. Overall, local excision, partial or total penectomy, external beam radiation, and chemotherapy have not shown to improve survival, though brachytherapy has been used in cases of penile metastasis and has been shown to control local disease progression for up to one year.[3]

Survival in patients presenting with malignant priapism is usually less than one year. According to one study, the time interval between primary tumor and penile metastasis ranged from three to 60 months, with an average of 19 months. The time interval between penile metastasis and death was 0.25 months to 18 months, with an average of six months, and no patients surviving beyond 18 months.[2] With penile metastasis, patients with rectal primaries appear to have somewhat better survival with the longest on record being over nine years. The longest survival recorded for a GU primary is seven years in the case of a prostatic adencarcinoma. Still, the average survival in patients with genitourinary primary is only 47 weeks, even with treatment, giving those with metastatic disease to the penis a very poor overall outcome.[3]

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Haris Ahmed is a fourth year medical student at UMDNJ-New Jersey Medical School.

Sammy Elsamra is a fifth year urology resident at the Alpert Medical School of Brown University.

Mark Sigman is professor and chair of the division of Urology at the Alpert Medical School of Brown University.

Disclosure of financial interests

The authors and or spouses/significant others have no financial interest to disclose.

Correspondence

Sammy Elsamra, MD
Chief Resident in Urology
Rhode Island Hospital
2 Dudley Street, Suite #175
Providence, RI 02905
phone: (401) 444-5795
fax: (401) 444-6947