Case Report

Malignant priapism: Penile metastasis originating on a primary prostate adenocarcinoma

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Abstract

Malignant priapism is a definition invented in 1938 by Peacock, defined as a persistent erection, not related with sexual activity, caused by cavernous sinus and associated venous systems invasion with malignant cells. Penile secondary lesions are rare entities. Primary locations are usually the pelvic cavity organs, namely the prostate and the bladder as the most common ones. Priapism as a first manifestation of these kinds of lesions is even rarer. The aim was to present a 52-year-old patient harboring a penile metastasis that originated in the primary prostate adenocarcinoma, manifesting itself as a "common" priapism. The patient referred to the emergency room presenting with a priapism and nodules at the *coronal sulcus*, without previous similar episodes. His evolution until properly diagnosed was catastrophic with multiple lymph nodes, bone and organ involvement, and with his demise soon after from serious bleeding and congestive heart failure, almost 2 months after he first came to the emergency room. We review the literature concerning malignant priapism, diagnosis, and current treatment and survival perspectives.

Key Words: Malignant priapism, penile metastasis, priapism, prostate cancer

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Received: 23.04.2014, **Accepted:** 24.06.2014

INTRODUCTION

We describe a case report regarding a 52-year-old patient harboring a penile metastasis that originated in the primary prostate adenocarcinoma. The patient referred to the emergency room presenting with a priapism and nodules at the *coronal sulcus*, without previous similar episodes.

Priapism represents a persistent erection, partial or complete, which lasts for >4 h after sexual arousal or orgasm. It can appear even without any relation with sexual stimulation. Nonischemic, arterial or high flow priapism is a persistent erection caused by an anomaly at the cavernous arterial inflow. The corpus cavernosum is typically tumescent but not rigid and the penis is not painful.[1]

Access this article online	
Quick Response Code:	Website: www.urologyannals.com
	DOI: 10.4103/0974-7796.152030

Malignant priapism is a definition invented in 1938 by Peacock, that describes it as a persistent erection, not related with sexual activity, caused by cavernous sinus invasion as well as associated venous systems with malignant cells.[2]

Penile secondary lesions are rare entities. Primary locations are usually the pelvic cavity organs, namely the prostate and the bladder as the most common ones.[3] Priapism as a first manifestation of these kinds of lesions is even rarer.

CASE REPORT

We present a 52-year-old male, cook, that referred to the emergency room with a persistent erection, nontender, associated with mild penile discomfort and perineal pain, that began and progressed for about a month, aggravating during

this time, accompanied by dysuria and polaquiuria. He also described a 14 kg weight loss in the previous 2 months. He denied fever or other urinary complaints.

Clinical observation revealed a partial erection, with rigid (but nontender) cavernous bodies [Figure 1]. There was no evidence of perineal trauma or urethrorragia. He had small soft and round pink nodules, along the *coronal sulcus* [Figure 2]. Rectal exam was positive for an enlarged prostate, tender, with a central soft nodule, with about 15 mm of diameter.

Laboratory evaluation revealed normochromic and normocytic anemia (Hb: 10.7 g/dL), leukocytosis (13.08 × 10^9/L) without neutrofilia (75.5%) and a raised C-reactive protein (15.2 mg/dL). Urine analysis was positive for leucocituria (125 cel/uL) and erythrocytes (80 cel/uL). We assumed a diagnosis of acute prostatitis with a high flow priapism, of unknown etiology. Patient was admitted as an in-patient and started on antibiotics (ceftriaxone 2 g e.v. id) and NSAID (diclofenac 50 mg e.v. tid).

Symptoms persisted even after 5 days of antibiotic therapy. We reexamined the patient every day, and priapism persisted, with *coronal* inflammatory nodules and a *de novo* positive renal Murphy's sign. Rectal exam was again performed on the 5th day, and it was superposable, with an enlarged and painful prostate, with central nodularity. Hemoglobin was of 9.6 g/dL and C-reactive protein had gone up (19 mg/dL).

The toraco-abdominal computadorized tomography (CT)-scan showed bilateral pulmonary infarcts, without any abscesses, with multiple pulmonary nodules of unknown etiology and also multiple prostatic abscesses [Figure 3].

Penile and trans-retal ultrasound revealed a lateral and distal hypoechogenicity nodule on of the left cavernous body (most likely an arterio-sinusoidal fistulae), and also a hyperechogenic lesion/collection distal to the right cavernous corpus, with a stable arterial flow and a prostatic abscess. We aspirated pus and performed a bacterial exam, which was negative for aerobes and anaerobes bacteria. The histopathological exam didn't reveal any additional information. At this time, the patient was discharged against doctor's orders.

Patient was re-admitted to the Infectious Diseases Department after 2 weeks because of pelvic pain, asthenia and obstructive urinary symptoms, with a new perineal lump or mass. He had an important leukocytosis, of 22.000 × 10^9/L. C-reactive protein went up to 24 mg/dL. Urine analysis revealed leucocituria and positive nitrites. We ordered a new CT scan of the abdomen and pelvis that showed multiple mediastinic and supraclavicular lymph node enlargements. The lung parenchyma showed multiple nodular formations, noncalcified, which suggested

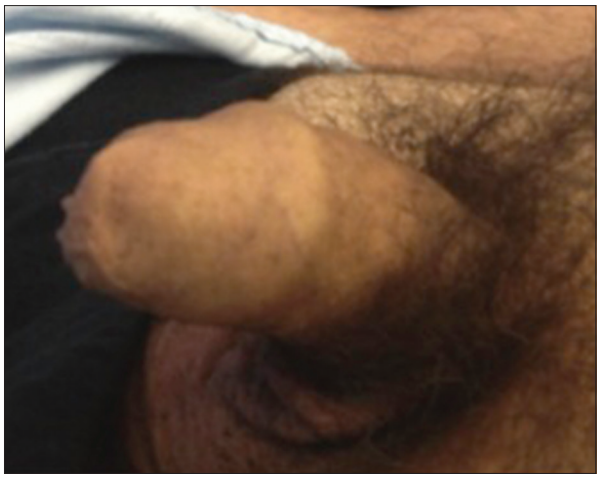

Figure 1: Priapism at the emergency room

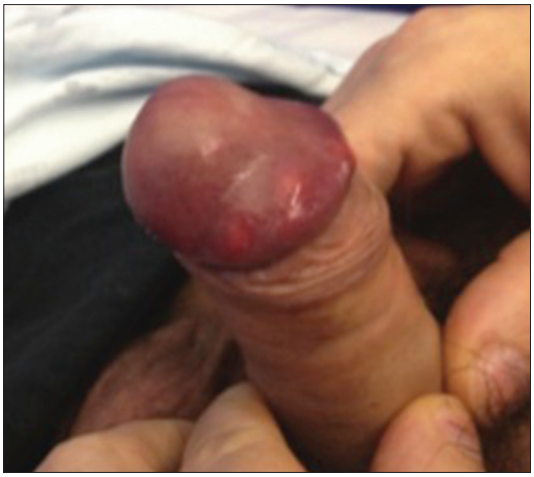

Figure 2: *Coronal sulcus* inflammatory nodules

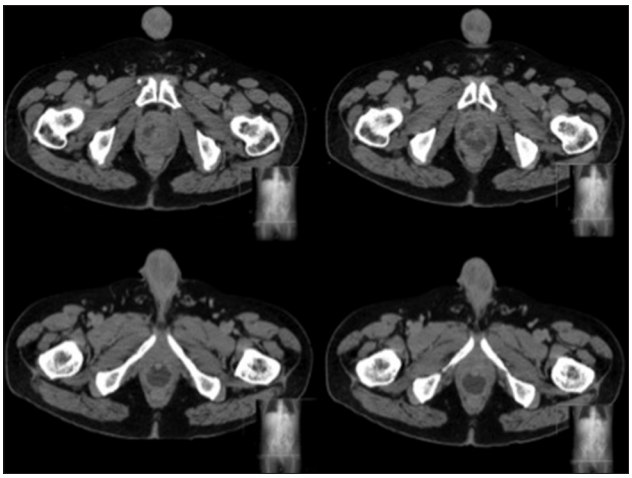

Figure 3: Toraco-abdominal computerized tomography-scan showing bilateral pulmonary infarcts, multiple pulmonary nodules and also multiple prostatic abscesses

secondary lesions. It also revealed a probable bone metastasis at the posterior arch of the 9th left rib. The liver showed a right lobe nodular formation, hypodense, with about 22 mm

of diameter, with similar multiple hypodense lesions in both suprarenal glands, related with metastasization.

The patient showed an important left ureterohydronephrosis caused by the invasion of the left ureter, with a pelvic mass involving the bladder, the prostate and the root of the penis, as well as bulky inguinal adenopathies and small ones on the peri-rectal, perineal, and right parieto-colic fat.

We decided on a transrectal biopsy of the prostate as well as a transperineal biopsy of the pelvic mass. Both revealed a Gleason 8 (5 + 3) adenocarcinoma of the prostate, extensively necrotic, and started bicalutamide 150 mg id. The prostate-specific antigen value, as of this time, was of 4.56 ng/mL.

His symptoms worsened. He started to have proctalgia associated with defecation, as well as rectal bleeding with hemodynamic repercussion (his hemoglobin levels went down to 5.4 g/dL), demanding urgently a transfusion.

A tumoral mass, coming out of the anal canal and the anus was identified, with an active bleeding. Biopsy of the mass revealed a tubulous-vilous adenoma, with high grade dysplasia. He was then scheduled to perform hemostatic palliative radiotherapy (20 Gy/5 fractions). On the 1st day of treatment, the patient develops congestive heart failure, with his demise soon following on the same day.

DISCUSSION

The first report of malignant priapism dates to 1870 by Eberth, that described a primary rectum adenocarcinoma that metastasized to the penis.[4] Abeshouse e Abeshouse described, up until 1961, another 140 cases.[5] More recently, Cherian *et al*. in 2006,[6] compiled 372 case reports, and in 2011, on the last published review, the number of cases totaled 394.[7] It is, indeed, a very rare first presentation of cancer, especially, when it comes to a primary prostate adenocarcinoma.

A Medline research included the key-words "malignant priapism," "penile metastasis," "malignant priapism" and "secondary" AND "malignancy" AND "penis." We found, another 6 more cases of penile metastases totaling 400 cases, beginning in 2011, this one being the 401st patient report found. The true incidence of penile metastasis can even be bigger because about 12% of the cases described are mostly asymptomatic and are only identified on autopsy.[3]

The penis has a rich and complex vascular circulation, in direct connection with the pelvic organs. In spite of it, penile metastases are rare findings, usually representing late findings of systemic disease and generally as multiple painless nodules, palpable, that can involve the cutaneous surface and ulcerate, resembling a syphilitic lesion. Differential diagnosis must include primary tumors of the penis, syphilis, tuberculosis, cancroid lesions and nonspecific inflammatory lesions.[8] Genitourinary and gastrointestinal lesions are the most common primary neoplastic diseases to metastize.[9] From this latter group, prostate cancer and urothelial tumors are the one that most frequently metastasize to the penis.[7,10] Other primary tumors described that metastasize to the penis are: renal tumors, lung, testicles, colon, and rectum.[6,11]

Prognosis is very bad, independently of the dissemination mechanism. The expected half-life of these kind of patients is generally very low, of approximately 9 months, with a global half-life of about 18 months.[7] In this case, in particular, the patient ended up dying after 2 months of its first manifestation of priapism in the emergency room.

There are a few different physiologic mechanisms proposed to explain malignant high and low debt priapism. Abeshouse e Abeshouse in 1961 and later Jacob Cherian in 2006 proposed five main mechanisms:

Venous retrograde route

Communicating venous plexus from the penis with pelvic venous plexus associated with a retrograde reflux. This mechanism might explain most of secondary tumours that originate in the prostate, the bladder, recto-sigmoid colon, as well as most lesions on cavernous bodies and glans.[5,6]

Lymphatic retrograde route

Penis, bladder, and posterior superficial prostate drains to the external iliac nodules. The lymphatics from the lower rectum also drain to the inguinal nodes and then to the iliac ones. By a mechanism of permeability or by embolization, neoplastic cells disseminate to the penis, especially to the cutaneous surface.[6,12]

Arterial dissemination route

Rare, may explain sarcomatoid metastasis.[6,12]

Direct extension route

Highly invasive prostatic or bladder tumors, in direct anatomic relation with the penis.[6,12]

Secondary to instrumentation

Improbable mechanism; isolated lesions of the corpus spongiosum without involvement of the corpus cavernosum or glans are practically inexistent.[6,12]

Malignant priapism is an invasion of the penile cavernous sinus with malignant cells originating on its venous drainage routes, without affecting arterial irrigation. Venous drainage blockade restricts sinus drainage provoking an erection.[13] Another explanation that may contribute to a malignant priapism

includes erectile nervous route sensitization by local tumoral infiltration. Both high and low flow mechanisms occur, but a high flow mechanism is thought to be the most common one, by a reverse arterial flow in dyastole.[7] Clinical manifestations of penile metastasis may include: a palpable penile mass or nodule, ulceration, lower urinary tract symptoms and malignant priapism in 20-50% of patients.[6,7,14]

In an article of Lin *et al*., in 2011, the most recent review,[7] malignant priapism's incidence as a first manifestation is of about 24% (7 in 29 cases, from 2006 to 2011). In the first table [Table 1], we identify the total number of published cases that occur with a penile metastasis.

Biopsy or an aspirate of the corpus cavernosum are the most common methods to obtain an histopathological diagnosis of a penile metastasis or a primary tumor of the penis, the latter having a more favorable diagnosis.[15,16] Cavernosonography is useful to delineate the involvement of the cavernous bodies, but invasive.[17] Imaging is the most reliable method to determine the existence and involvement of the corpus cavernosum and to plan a surgical intervention.[3] Ultrasound must be the first line exam if there is a suspicion of the penile metastasis, and then, during follow-up. It's useful for local staging (primary disease) as well as when there is a suspicion of secondary lesions.[6,18] Cavernous or spongiosum nodules normally present themselves with variable echogenicity and vascularized.[18] CT and magnetic resonance imaging (MRI) are both reliable and precise when it comes to confirm a diagnosis and establish the extent of the disease.[19] MRI is more accurate to establish the differential diagnosis, when compared to CT or genital ultrasound,[6] and it must be this the method chosen.[20] In the MRI, penile metastases have a low intensity signal in T1, and isointense in relation to the adjoining cavernous bodies. In T2, the metastatic focus is hypointense and contrast with the hyperintese signal of the corpus cavernous. It's common to exist gain of contrast by the metastatic focus, after its administration. MRI is also useful to determine if the primary cancer is confined to the genitourinary tract, the prostate or the recto-sygmoid colon, and even to determine lymphatic metastatic envolvement.[21]

Table 1: Total number of published reports that identify the origin of penile metastasis

Primary location	Absolute number	Percentage
Prostate	129	32.25
Bladder	121	30.25
Recto-sigmoid	54	13.5
Kidney	30	7.5
Remaining colon	16	4
Esophagus, stomach and small intestine	6	1.5
Testicles	12	3
Lung	9	2.25
Bone	4	1
Upper airway	4	1
Ureter	2	0.5
Hepatobiliar	2	0.5
Others	11	2.75
Total	400	

Recommended treatments for metastatic lesions of the penis are: local lesion excision, partial penectomy or total penectomy, in case of refractory pain, or obstructive urinary symptoms caused by infiltration of the cavernous corpus. Radiotherapy and chemotherapy are recommended as soon as the local tumor in no longer surgically feasible. The outcome of the patient is mostly dependent on his clinical status, primary tumor, extension of malignity, or the presence of metastatic disease that the modality of treatment chosen.[3,14] Corpus cavernous metastization represents evidence of disseminated disease in other locations in about 80-90% of patients, most of them dying in about 1 year.[2,17] Multiple metastasis are directly related to the death cause. Even so, there are reports published that reveal half-life's of about 7-9 years after the first evidence of penile metastasis.[22]

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How to cite this article: da Silva Gaspar SR, Nunes A, Dias JS, Lopes T. Malignant priapism: Penile metastasis originating on a primary prostate adenocarcinoma. Urol Ann 2015;7:391-5.

Source of Support: Nil, **Conflict of Interest:** None.