

Penile Gangrene: An Unusual Complication of Priapism in a Patient with Bladder Carcinoma

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INTRODUCTION

Penile gangrene is an infrequently encountered disease entity.^[1] Penile gangrene complicating priapism seems to be of even rarer occurrence.^[2-4] Several etiological factors have been associated with penile gangrene, ranging from self-inflicted penile strangulation to diabetes mellitus and chronic renal failure patients on dialysis.^[5-12] Limited references are found in the English literature on penile gangrene following priapism; an earlier paper reviewed the previously reported 13 cases in the literature.^[2,3] Majority of the cases of priapism are idiopathic in origin and are usually of the ischemic type;^[13,14] the presentations are classical and the treatment involves active resuscitation of the patient, followed by early urgent decompression of the intracavernosal pressure to achieve detumescence and prevent complications that are bound to follow ischemic injury to the endothelial lining of the corpora cavernosum. Malignant metastases to the penis are a rare cause of priapism.^[15] The presence of an initially unrecognized bladder carcinoma in a patient with ischemic type of priapism and the penile gangrene that followed its management made this case distinctive and thus warrants documentation.

CASE REPORT

A 40-year-old farmer was referred to our unit for

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ABSTRACT

A 40-year-old, apparently healthy farmer presented with a 4-day history of progressively painful penile erection with no known predisposing or precipitating factor. He had an emergency El-Ghorab shunt which resulted in almost complete detumescence. He was noticed to have developed ischemic changes of the distal part of the penile skin which progressed to gangrene of the distal part of the penis on the 4th day post intervention. Abdomino-pelvic ultrasound revealed an intravesical mass and urine and corpus cavernosa aspiration cytology were positive for malignant cell. The patient, however, declined further treatment and was discharged against medical advice.

Key words: Bladder cancer, penile gangrene, priapism

persistent, painful penile erection of 4 days duration. The penile erection was spontaneous in onset; there was no known predisposition or precipitating factor. There was no history of similar occurrence in the past, he was not a known sickle cell disease patient and there was no history of use of aphrodisiac. He was neither a known hypertensive nor a diabetic patient. There was some degree of lower urinary tract symptom; however, he presented with no hematuria, necroturia or suprapubic mass and had never been admitted in the past.

At presentation, he was in moderate painful distress, not pale, anicteric and well hydrated. The chest examination was normal. The pulse rate was 100 beats per minute and he had normal blood pressure. The abdominal examination revealed nothing significantly abnormal; however, the penis was erect, to about 110°, tender and slightly edematous. The testes were intrascrotal and palpable; the scrotum was slightly edematous too. Digital rectal examination, which was done much later, revealed a relatively fixed pelvic mass.

The hemoglobin was 11 g/dl, the white cell count was $8.7 \times 10^9/l$ and the differential white cell counts were within normal range of limits. The erythrocyte sedimentation rate (ESR) was 5mm/hour. The renal function evaluation was within normal with sodium 135 mmol/l, potassium 3.6 mmol/l, bicarbonate 22

mmol/l, urea 4.5 mmol/l and creatinine 124 μ mol/l. The random blood sugar was 7.0 mmol/l and the fasting blood sugar was 5.5 mmol/l. The abdomino-pelvic ultrasound, which was done after institution of management, revealed a left laterally located intravesical mass which measured 5 × 5 × 3 cm in dimensions; this prompted urine and corpus cavernosa aspiration cytology and these were positive for malignant cells.

The patient had emergency decompression of the intracavernosal pressure by modified El-Ghorab technique which resulted in almost 90% detumescence and this was sustained. About 48 hours following operation, the distal penile skin was noted to have changed color and this progressed to frank gangrene of that portion of the penis [Figures 1 and 2]. The patient was counselled for debridement of the penile gangrene with or without partial penectomy, but he declined and he later discharged himself against medical advice.

DISCUSSION

Penile gangrene has been described as a hallmark of severe systemic vascular disease.^[6] The present case report does not fit into most of the common causes of penile gangrene.^[1-12] It was preceded by the classical presentation of ischemic type of priapism that usually warranted no further evaluation, especially in a setting where cavernosa blood gas analysis is not routinely practiced because of lack of facility. The patient consented to and had a shunt procedure which resulted in an appreciable detumescence.

The presence of metastatic malignant disease as the predisposition to priapism has been documented. It commonly follows hematological malignancy and sometimes occurs in association with solid organ tumor as found in the present report.^[2,13-16] The hematological malignancies, understandably so, form a slug that occlude the venous drainage of the corpus cavernosa, leading to the engorgement of the sinusoidal space with low oxygen tension venous blood resulting in priapism. Priapism was considered to be the initial mode of clinical presentation in as much as 40-50% of malignant penile metastasis, as found in this index case report. Thus, clinicians need to be cautious of this presentation.^[14,15,17]

Although a full clinical and histological evaluation of the index patient could not be undertaken because the patient discharged against medical advice, the



Figure 1: Wet gangrene that follows the initial ischemic changes



Figure 2: Dry gangrene that supervenes later

presence of bladder mass on ultrasound evaluation and the positive urine cytology for malignant cell gave a high suspicion index for bladder cancer. Several mechanisms for tumor metastasis to the penis have been proposed and they include direct extension of the primary tumor to the penis, antegrade arterial dissemination from the primary tumor, seeding from instrumentation, retrograde lymphatic and venous spread and extension along the nerves. Aside from seeding from instrumentation, for which such history was not obtained from the patient, all other mechanisms were possible to explain the malignant priapism in this case.^[18]

For logistic reasons, only corpus cavernosa aspiration for cytology was available to affirm our suspicion of penile metastasis as the cause of the priapism; however,

the histological diagnosis of penile metastatic disease can further be confirmed using several other modalities that include cavernosography, computerized tomography scan, magnetic resonant imaging as well as core-needle biopsy of the corpus cavernosum.^[15,19] The core-needle biopsy seems to be the preferred option due to its more reliability in the assessment of the presence of malignant cell infiltration and the extent of invasion. It also allows for immunophenotypical characterization of the tumor cell infiltrate to confirm the precise nature of the malignancy.^[14]

Hypercoagulability is a well-documented paraneoplastic syndrome with apparent vascular thrombosis, occurring in as much as 11% of patients with malignant diseases, especially those with mucinous cancers.^[16] Inappropriate activation of the coagulation cascade due to interaction of the malignant cells with the vascular endothelium, platelets, coagulation and fibrinolytic systems are the pathological basis of the hypercoagulability that occurs in such patients.^[16] These could probably explain the occurrence of penile gangrene in the index patient.

Although paraneoplastic syndrome occurrence is not a usual finding with bladder carcinoma, the occurrence of adenocarcinoma of the bladder, which could not be confirmed in this case report, could probably explain the association between malignant priapism and the penile gangrene that follows its management, either as a cause or effect, through the mechanisms described earlier. Superficial thrombophlebitis, deep venous thrombosis, pulmonary embolism, non bacterial thrombotic endocarditis, visceral thrombosis, thrombotic microangiopathic hemolytic anemia, digital and cerebral microvascular arterial thrombosis and apparent disseminated intravascular coagulopathy are the clinical manifestations of cancer-associated hypercoagulability,^[20] and this could affect the several arterial supplies of the penis.

Variety of treatment options have been described in the literature for the management of penile metastasis based on the spectrum of presentation.^[15,21,22] Even at that, if the patient presents with priapism, a glans-to-cavernosa shunt is an exception rather than the rule. The treatment options for penile metastatic diseases are almost always palliative. The diagnosis of penile metastasis as the cause of priapism requires a high index of suspicion to avoid glans-to-cavernosa shunt which may be complicated as found in this patient.

In conclusion, penile gangrene following malignant priapism is a rare occurrence. Partial or total penectomy may be required, in addition to other modes of treatment, in the management of such a patient.

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