

Priapism with penile gangrene: An unusual presentation of multiple myeloma

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ABSTRACT

Penile gangrene is very rare sequel of ischemic priapism. Previous published reports have shown its occurrence in patients with sickle cell disease, urethral carcinoma, bladder carcinoma, thrombotic thrombocytopenic purpura, idiopathic, traumatic, etc. Ischemic priapism with penile gangrene as an initial presentation of multiple myeloma has not been reported. We present a 44-year-old patient of multiple myeloma presenting with ischemic priapism and penile gangrene requiring partial penectomy.

INTRODUCTION

Ischemic priapism is a surgical emergency necessitating urgent intervention.^[1] It can be either idiopathic or secondary to use or abuse of toxic or pharmacological substances, hematological disorders such as sickle cell disease, leukemias, penile metastases, and certain neurological disorders. Permanent erectile dysfunction and rarely penile gangrene are sequelae of priapism. Priapism with penile gangrene has been reported in patients with sickle cell disease, urethral cancer, thrombotic thrombocytopenic purpura,^[2] bladder carcinoma,^[3] trauma, etc. However, priapism with penile gangrene has not been reported in a patient of multiple myeloma so far. We present a unique case report of a patient of multiple myeloma who presented with ischemic priapism and penile gangrene.

CASE REPORT

A 44-year-old man presented with persistent painful erection and blackish discoloration of penile skin for 4 days. The erection was spontaneous in onset without

any precipitating factor such as drug intake and sexual stimulation. He did not have similar episodes in the past. On physical examination, penis was rigid, tender to touch with multiple fluid filled blebs and blackish discoloration involving penile shaft skin and glans [Figure 1a]. Investigations showed hemoglobin of 10 g/dl, total leukocyte count of 9700/mm,^[3] and serum creatinine of 1.0 mg/dl. Urgent cavernosal blood aspiration and saline irrigation followed by injection phenylephrine were given. The patient had partial detumescence, but priapism recurred within 6 h. Hence, a Winter's shunt was created, and the patient had detumescence with partial improvement in color of glans and penis. Next day, the patient had a sudden onset of blackening of the right index finger and foot associated with severe pain. In view of multiple sites of peripheral ischemia and penile gangrene, the patient underwent further investigations. Color Doppler of bilateral upper and lower limbs and 2D echo was normal. His coagulogram was normal and his serum creatinine rose to 1.8 mg/dl, serum albumin was 3.1 g/dl, and total protein was 9.82 g/dl suggesting reversal of albumin to globulin ratio. He was investigated further by serum protein electrophoresis and immunofixation which revealed a prominent "M" band and lambda light chain confirming the diagnosis of

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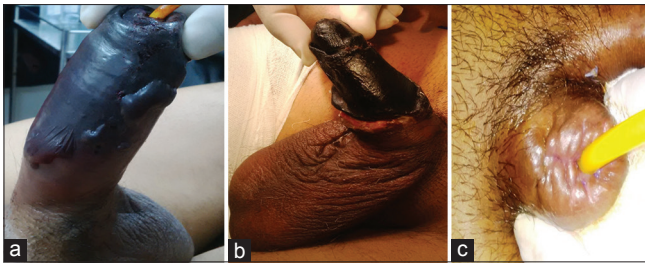


Figure 1: (a) Erect penis with gangrenous changes, (b) blackish discoloration of penis with clear line of demarcation before surgery, (c) penile stump after partial penectomy

multiple myeloma. Urgent plasmapheresis was performed. Ischemic changes involving the index finger reverted but those of the penis and foot worsened [Figure 1b]. The patient underwent partial penectomy [Figure 1c] and right below knee amputation. Histopathology was suggestive of penile gangrene with infiltration of myeloma cells in corpora cavernosa. Immunohistochemistry was carried out to confirm plasma cells using CD 38 and 138 and lambda light chain restriction [Figure 2]. The patient received seven cycles of bortezomib-based therapy. At present, the patient is doing well at 1 year follow-up without any recurrence of priapism.

DISCUSSION

Priapism is defined as involuntary, painful, and prolonged erection of penis lasting more than 4 h unrelated to sexual stimulation and unrelieved by ejaculation. Ischemic priapism accounts for >95% of cases of priapism. Underlying hematological causes such as sickle cell disease, thalassemia, leukemia, G6PD deficiency, fat emboli associated with hyperalimentation, and rarely multiple myeloma^[4,5] need to be evaluated in cases of ischemic priapism. Penile gangrene is a rare sequel of priapism. In our case, priapism with penile gangrene was the initial presentation. Khoriaty *et al.*^[6] in their paper reviewed around 13 cases of penile necrosis following priapism with sickle cell disease, as most common etiology. They suggested that tight compressive bandage around the penis and local infection was responsible for the development of penile necrosis in majority of cases. Penile gangrene with priapism has also been reported in a case of thrombotic thrombocytopenic purpura.

Our index case had neither tight compressive dressing nor local infection as a responsible factor for penile gangrene. However, the occurrence of peripheral limb gangrene led to further investigations leading to the diagnosis of multiple myeloma. He underwent partial penectomy, plasmapheresis and bortezomib chemotherapy, and limb amputation for

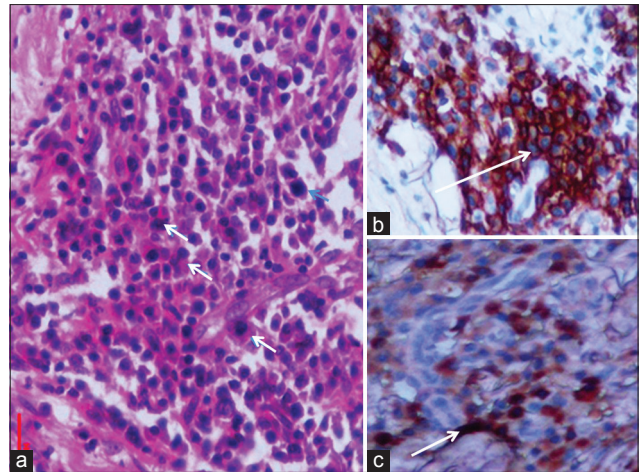


Figure 2: Panel of photomicrographs. (a) High power picture of plasma cells in sheets, a few of them having enlarged pleomorphic nuclei and binucleation (white arrows) (H and E, $\times 500$). (b and c) Highlighted the positively stained cells in immunohistochemistry. (b) The plasma cells showing membrane positivity for CD138 seen as brown color (white arrow) (Peroxidase anti-peroxidase, $\times 500$). (c) Cytoplasmic positivity for lambda light chain seen as brown color (white arrow) (Peroxidase anti-peroxidase, $\times 500$)

gangrenous leg. Hyperviscosity state in multiple myeloma might be responsible for the development of penile and peripheral limb gangrene in our patient.

The presence of penile gangrene in a setting of priapism without any evidence of local infection warrants detailed evaluation for hyperviscosity and hypercoagulability disorders. To the best of our knowledge, priapism with penile gangrene as an initial presentation of multiple myeloma has never been reported.

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