

## CASE REPORT

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# Recurrent priapism as the presenting sign of undiagnosed multiple myeloma: A case report

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## ABSTRACT

**Introduction:** Priapism is a urological emergency which poses a high risk of long-term sexual dysfunction for patients if left untreated. Although the majority of priapism cases are secondary to sexual enhancement medications, there are many underlying medical disorders that may lead to unexplained and recurrent cases of priapism.

**Case Report:** We present the case of a 53-year-old male with schizoaffective disorder and chronic lower back pain on no medications who presented multiple times to various emergency departments for recurrent priapism requiring aspiration. Ultimately, the patient presented to the emergency department after a fall, and labs revealed hypercalcemia, with serum protein electrophoresis and bone marrow biopsy confirming multiple myeloma.

**Conclusion:** Multiple myeloma continues to be a rare underlying etiology of recurrent priapism. This case represents the importance of performing a thorough investigation in patients with recurrent priapism for hematologic and oncologic disorders, despite there being no other insidious symptoms or signs.

**Keywords:** Emergency, Hematologic, Multiple myeloma, Priapism

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## INTRODUCTION

The emergency department continues to be a remarkable setting due to the broad range of medical presentations, often undifferentiated and unannounced. Occasionally, a case of priapism arrives, and often, it is relieved with simple needle aspiration through the corpus cavernosum and second line medications including phenylephrine and terbutaline [1]. To date, the most common cause of priapism continues to be sexual enhancement medications such as phosphodiesterase inhibitors, which make up two-thirds of the cases [2, 3]. Although the majority of providers may be well-versed with the remaining underlying etiologies of recurrent priapism, such as sickle cell disease, illicit drug use, medication side effects such as trazodone, and neurologic disorders, it is important to consider hematological oncological diseases such as multiple myeloma as the underlying cause. We present the case of a man who had presented to emergency department multiple times to receive needle aspiration of his priapism, until he was ultimately diagnosed with multiple myeloma.

## CASE REPORT

A 53-year-old male with schizoaffective disorder not on medications, and chronic lower back pain which he's

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had for over seven years after a motor vehicle accident started to present to various emergency departments multiple times over the course of 12 months for priapism which occurred upon awakening in the morning. The patient would attempt self-treatment at home by walking up and down the stairs, which he reported would often lead to detumescence. During his presentations to the emergency department, the patient declined the use of sexual enhancement drugs, the use of illicit drugs, personal or family history of illness, sickle cell trait or disease, hematological or oncologic illness. During these presentations, the patient did not have any other medical complaints or symptoms of concern. Furthermore, review of systems during these presentations were consistently negative for fatigue, fever, body aches, recurrent infection, weight loss, fractures, headache, chest pain, shortness of breath, abdominal pain, difficulty with urination, diarrhea, or constipation.

The patient had established primary care and had been followed by a urologic surgery clinic, pending the insertion of a caverno-dorsal shunt. This procedure had not yet been performed, and the patient had continued to present for recurrent needle aspirations with successful detumescence during each visit. It is important to note that while the patient was encouraged to undergo laboratory testing in the emergency department to evaluate for underlying etiologies, the patient had work-related commitments and requested to leave to follow-up with his primary care provider and urologic specialist.

Ultimately, the patient presented to the emergency department on his own, after experiencing a mechanical fall landing onto his back, worsening his chronic back pain. Computed tomography upon presentation revealed mild canal stenosis without any masses, lesions, or other acute findings. However, laboratory investigations performed upon presentation revealed the following relevant values: White blood cell count of 3.3 thousands per microliter (K/uL) (Reference [Ref]: 4.80–10.80 K/uL), Hemoglobin 7.3 grams per deciliter (g/dL) (Ref: 14.0–18.0 g/dL), red blood cell distribution width 16.3 % (Ref: 12.2–16.1%), platelet count of 121 K/uL (Ref: 150–400 K/uL), blood urea nitrogen 23.0 milligrams per deciliter (mg/dL) (Ref: 5.0–20.0 mg/dL), creatinine 1.39 mg/dL (Ref: 0.70–1.20 mg/dL), and calcium 10.6 mg/dL (Ref: 8.5–10.5 mg/dL).

The patient was admitted for the abnormal laboratory findings, and underwent extensive testing, including a protein electrophoresis revealing the following: Alpha One value of 3% (Ref: 2–8%), Alpha Two value of 9% (Ref: 6–14%), Beta One value of 47% (Ref: 6–12%), Beta Two value of 3% (Ref: 4–8%), Gamma percentage of 12% (Ref: 10–20%), Albumin percentage of 27% (Ref: 30–70%), M-Spike 3.83 grams per deciliter (g/dL) (Ref: <0.00 g/dL), with significant presence of IgA monoclonal antibodies. Bone marrow biopsy was performed, confirming the final diagnosis of plasma cell CD138+ multiple myeloma. Furthermore, immunoglobulin kappa

free light chain level was 12.42 mg/dL (Ref: 0.33–1.94 mg/dL).

The patient underwent plasmapheresis for hyperviscosity syndrome, decreasing his IgA levels from 7000 mg/dL (Ref: 70–400 mg/dL) to 2455 mg/dL after a single session. The patient was also started on first line multiple myeloma treatment consisting of Cyclophosphamide, Bortezomib, and Dexamethasone, and discharged with close follow-up. Chart review reveals that the patient has had decreased incidents and visits for priapism since initiation of treatment for multiple myeloma.

## DISCUSSION

Priapism is defined as a state of penile erection in the absence of sexual stimulation, or when abnormally prolonged following sexual activity [1]. Clinically, the current literature has described a state of prolonged erection harmful if longer than 4 hours [4]. Priapism can lead to penile ischemia and necrosis of the smooth muscle tissue and tissue fibrosis [5]. These injuries caused by priapism can lead to long-term and often irreversible sexual dysfunction [4]. It has been suggested that irreversible smooth muscle loss can occur as early as 6 hours [6, 7]. Furthermore, it has been documented that 90% of individuals who experience priapism longer than 24 hours will develop erectile dysfunction [8]. Therefore, untreated and unaddressed priapism is considered an emergency. There exists two general pathophysiological categories of priapism: ischemic priapism, also known as low-flow priapism, and high flow priapism. Ischemic priapism constitutes 95% of priapism cases, and it is caused by decreased blood return from the penis, leading to venous congestion and painful rigidity within the penis [1]. High flow priapism is caused by high arterial flow into the corpus cavernosum, causing a continuous erection that may last days to weeks. Generally, high flow priapism is not painful and is not considered a urologic emergency. Venous blood gas samples taken directly from the corpus cavernosum upon aspiration can be analyzed to determine the dichotomous nature of the priapism, although treatment of the two is grossly similar.

First line treatment for priapism entails aspiration of blood from the corpus cavernosum of the penis using sterile needle aspiration technique. If needle aspiration does not lead to successful detumescence, phenylephrine, a pure alpha agonist can be injected into the corpus cavernosum to cause vasoconstriction of the penile arteries to decrease blood flow into the penis. Terbutaline, a selective beta-2 agonist, can also be used as an intramuscular injection both as an adjunct or monotherapy to relax the smooth muscles within vessels to increase blood drainage from the penis. Recurrent priapism can be treated with tunneling and distal shunt insertion by urology [1].

It is prudent, however, to address the underlying cause of priapism. The most common cause of priapism has been sexual enhancement medications such as phosphodiesterase-5 inhibitors such as sildenafil and tadalafil [2, 3]. Other common causes include sympathomimetic drugs such as cocaine, and antidepressant medications such as trazodone [9, 10].

Although chronic illnesses such as sickle cell disease, polycythemia, and thalassemia has been understood as a common cause of priapism, undiagnosed hematologic and oncologic conditions may be overlooked as the reason behind priapism, likely due to the relative low odds of a patient with undiagnosed hematologic disorder presenting to the emergency department with priapism as their only symptom [11, 12].

Multiple myelomas, a plasma cell cancer with a variable 5-year overall survival rate between 40% and 80% depending on the stage, have an incidence of approximately 1.8% of all new cancers diagnosed in the United States each year [13, 14]. Modern literature has described priapism incidence in multiple myeloma patients upward of 3–5% [15]. Understanding that general hematologic malignancies may often manifest as recurrent priapism as the only sign of the disease may lead to early diagnosis, treatment, and quality of life enhancement for the patient. For this matter, American Urology Association recommends hemoglobin electrophoresis as an appropriate workup in scenarios of recurrent priapism with no clear underlying cause [1].

## CONCLUSION

It is prudent for emergency medicine providers to consider hematologic and oncologic disorders when presented with a case of unexplained priapism, even more so if the priapism is recurrent. In some patients, like the respective patient in this case, priapism may be the only sign of a sinister underlying etiology. A comprehensive workup may lead to early diagnosis and treatment of a disease with high mortality if left untreated, such as multiple myeloma.

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## Author Contributions

Shayan Azizi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Christopher Costa – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Grace Bomann – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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#### **Guarantor of Submission**

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#### **Conflict of Interest**

Authors declare no conflict of interest.

#### **Data Availability**

All relevant data are within the paper and its Supporting Information files.

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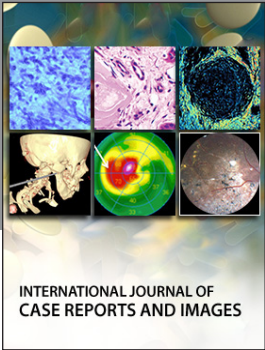
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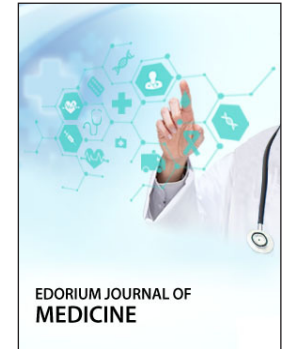


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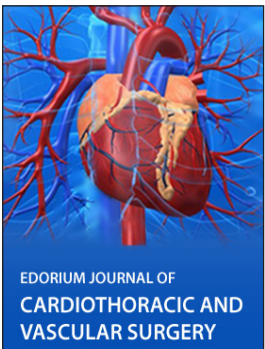


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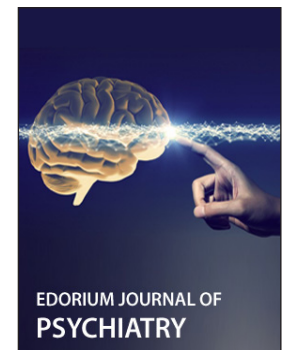
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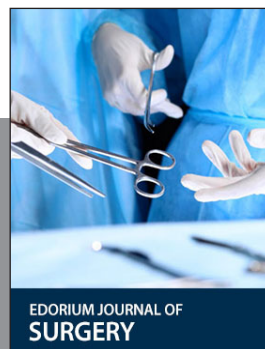
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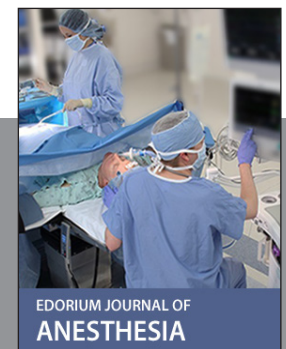
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