A Case Report of Priapism With Unusual Presentation and Clinical Course

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Abstract
This is a case report on a patient with an unusual presentation and clinical course of priapism. It further discusses treatment options with reflection on current literatures and guidelines. 48 year old patient presented with a history of more than 50 episodes of priapism, each lasting for five minutes. Patient had history of brain tumor that was resected and had since been in remission. On examination and further biochemistry assessment revealed conflicting clinical findings, making it difficult to ascertain the type of priapism in this case. The patient, however, recovered from the acute attacks of priapism after 24 hours of conservative management and no obvious cause had been identified on post-discharge follow-up. Priapism, despite being rare, is a medical emergency. This case report reflected upon the limitations of treatment guidelines and the lack of level one evidence to support treatment decisions. © 2017 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Despite being relatively rare, priapism is an important pathology that warrants the correct treatment in a timely manner. This case report presents a patient with an atypical clinical course of priapism, and will further discuss on the ambiguity of clinically findings with treatment choices and treatment responses.

Case history

A 48-year-old Caucasian man presented to the emergency department with a history of persistent erection following a whole body spasm for a few seconds. Patient reported experiencing 50 episodes throughout the day and each episode lasted for 5 minutes. There was no pain associated with each erection.

Patient had a background history of cerebral tumor that was resected initially in 1995, with recurrence in 2013, and subsequently had 2 years of monthly chemotherapy. Patient had been in remission since the last chemotherapy.

On examination, patient was mildly distressed, while being stable. A focused urology exam reviewed an erected penis without evidence of ischemia. Examinations of other systems including neurology exam, were unremarker.

Patient received urgent investigation and management. A full panel of blood tests including full blood count, liver function, renal function, C-reactive protein, coagulation markers were normal. Subsequently, patient was given oral pseudoephedrine and underwent cavernosal aspiration urgently. A total amount of 380 mL (ml) of bright red blood was aspirated, which indicated high-flow priapism as a diagnosis. However, the blood gas analysis showed an ambiguous result (Table 1).

Patient’s priapism persisted despite aspiration. Urgent urological intervention was considered and patient was transferred to the emergency theater. Whilst patient was in the anesthetic bay, a second aspiration was attempted following intra-cavernosal injection of phenylephrine. Approximately another volume of 100 mls of blood was aspirated and patient’s priapism subsequently resolved. No surgery was performed.

Patient was admitted to the ward for conservative monitoring. Patient was further evaluated with CT scans of the brain, abdomen and pelvis, which revealed no evidence of recurrence of brain tumors or distal metastasis.

However, patient further developed another episode of priapism that required further medical review in the ward. This episode occurred when patient was showering and resolved spontaneously after lasting for 5 minutes. No further episodes of priapism occurred. Patient was subsequently discharged on the following day.

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Priapism is defined as a persistent erection of the penis or clitoris that is not associated with sexual stimulation or desire.\(^1\) It is a relatively rare condition, with an incidence of 0.73 per 100,000 men per year.\(^2\)

Clinically, there are two main types of priapism, namely ischemic and non-ischemic. A subtype of ischemic priapism named recurrent priapism is characterized by repeated erections of short duration. Ischemic priapism commonly presents with a painful, stiff erection and warrants urgent management. Nonischemic priapism usually presents after trauma, without a painful or rigid erection, and resolves spontaneously.

History and physical examination are important in assessing the cause of priapism. A number of risk factors are listed in Table 2, which should be elicited in details as part of the initial assessments in the emergency department. In this case, patient had a history of brain tumor that was resected without evidence of metastasis or new recurrence. It was unclear whether patient was at higher risk of developing priapism after patient’s brain tumor was resected. Thus far, no literature had commented on the incidence of priapism following major brain surgery.

Further investigation and management should be performed concomitantly to avoid complications, such as erectile dysfunctions, which can significantly impair patient’s quality of life.\(^3\) Corpora blood gas analysis can be performed to distinguish between ischemic and non-ischemic priapism, as well as an attempt to reduce blood volume accumulated in the erected penis. According to the American Urological Association,\(^1\) treatment of priapism depends on the type of priapism, and should follow the algorithm in Fig. 1.

However in this case, the blood gas analysis rather revealed an ambiguous result (Table 1). Clinically this case represented towards a recurrent priapism, which was usually considered as a type of ischemic priapism. However, blood gas analysis yielded arterial blood, which contradicted clinical findings.

Response to treatment was also unusual in this case. Patient’s priapism still persisted despite of the initial performance of blood gas, which had aspirated 350 mls of blood out. Despite

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**Table 1**
The patient’s corporal blood gas analysis showed an ambiguous result, making it more difficult to define the type of priapism in this case.

|                  | pH     | pO\(_2\) (80–100 mm Hg) | pCO\(_2\) (35–45 mm Hg) |
|------------------|--------|------------------------|------------------------|
| **Patient results** | 7.44   | 42 mm Hg               | 28 mm Hg               |
| **Ischemic priapism** | Acidemia | Hypoxemia              | Hypercarbia            |
| **Non-ischemic priapism** | Normal pH | Normal value           | Normal value           |

**Table 2**
Important risk factors of priapism.

| History and Examination Findings | Risk factors                  |
|----------------------------------|-------------------------------|
| Prior episodes                   | Medications e.g. use of recreational drugs |
| Hematological disease, especially sickle cell anemia | Malignancy, e.g. brain tumors or metastasis |
| History or findings of penile or perineal trauma | |

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**Figure 1.** Treatment algorithm for priapism.
of contradicting clinical findings and blood gas analysis, patient’s management actually followed the ischemic priapism pathway (Fig. 1). However, patient suffered another acute episode during showering. This episode lasted 5 minutes and resolved spontaneously without any treatment. Data was limited in terms of the treatment efficacy, and therefore whether the additional episode priapism was attributable to treatment failure, insufficient dose or treatment complication, remained yet to define.

The presentation, clinical findings, blood gas analysis as well as treatment choice and response were atypical and unseen in previous literature. It is important to realize that, given such an important pathology that impacts greatly on patient’s quality of life, the safety and efficacy of various treatments are not well established. Management is empirical and derived from case reports or case series. There are no randomized controlled trials or evidence-based guidelines in the current literature.4

Conclusion

In summary, this case report discussed an unusual case of priapism. It further reflected on the current limitations of treatment guidelines and the lack of level-one evidence to support treatment decisions. Evidence-based guideline should be established and is important for future researchers, given current management is only empirically based.

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