

High-flow priapism after T-shunt and tunneling in a patient with ischemic priapism

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ABSTRACT

Priapism is defined as an erection lasting for more than 4 hours without sexual stimulation. It is grouped into 3 subtypes: ischemic (low-flow), nonischemic (high-flow), and stuttering priapism. Herein we describe a rare event of high-flow state as a result of conversion from a delayed ischemic priapism after a T-shunt with tunneling. To our knowledge, there is a paucity of reported cases, and the pathophysiology is still unclear. Clinicians should be aware of this uncommon but known scenario in case of penile tumescence after shunting procedure for ischemic priapism; penile Doppler ultrasound and selective pudendal angiography represent essential tools for diagnosis and treatment of this rare condition. In delayed ischemic priapism persisting for >36 hours, patients should be counseled about the irreversible damages of the cavernosal muscle and erectile dysfunction to consider an early penile prosthesis implantation with a satisfactory long-term functional outcome, decreasing the risks related to a distal shunt procedure.

Keywords: Arteriovenous malformations; high-flow priapism; ischemic priapism; priapism.

Introduction

Priapism is defined as a prolonged penile erection lasting for more than 4 hours in the absence of sexual stimulation and remains despite orgasm. It can be categorized into 3 different subtypes: ischemic (low-flow), nonischemic (high-flow), and stuttering.

In ischemic priapism (IP), inhibition of venous outflow leads to a form of compartment syndrome, and the lack of arterial inflow determines a hypoxic state. If conservative treatment with aspiration and instillation of sympathomimetic agent fails in inducing detumescence, more invasive shunting procedures are needed to prevent progressive smooth muscle changes with ischemic damage, corporal fibrosis, and irreversible erectile dysfunction.^[1]

In case of delayed IP persisting for >36 hours, early penile prosthesis implant (PPI) has been proposed as a treatment option for refractory erectile dysfunction management avoiding further corporal fibrosis that results from a de-

lay in surgery.^[2] Penile shortening with loss of length postoperatively is one of the most frequent reasons for dissatisfaction in men with a PPI.

The American Urological Association (AUA) guidelines on priapism, published in 2003 and reviewed in 2010, do not refer to PPI for refractory IP considering the procedure as an end-stage option for stuttering priapism.^[3] Conversely, the European Association of Urology (EAU) guidelines state that relative indications for immediate PPI are IP lasting >36 h or failed first-line treatments, including shunt procedures; although in delayed cases, PPI might be considered ahead of shunt surgery.^[4]

Nonischemic priapism (NIP) generally follows a penile or perineal trauma, owing to laceration of a penile artery creating an arterial-lacunar fistula, but it may also complicate the surgical manipulation of the penis.^[4]

We describe a particular case of an arterial-lacunar fistula after a T-shunt procedure with

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tunneling for delayed IP converting a veno-occlusive into a high-flow priapism.

Case presentation

This study was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The patient provided written informed consent to publish the case (including publication of images).

A 40-year-old man presented to the emergency room with a priapism lasting over than 48 hours; only 6 hours earlier, he underwent an unsuccessful drainage of the corpora cavernosa and intracavernous injection of alpha-mimetic drug at another institution.

He had a history of type 1 diabetes mellitus and a previous episode of IP 10 months ago, which was successfully managed with drainage and intracavernous injection. The patient denied drug use, known hematologic conditions, or genital trauma. Blood count, coagulation profile, and sickle-cell preparation showed normal levels.

At presentation, his physical examination revealed a painful complete erection of both corpora cavernosa with a quite tumescent glans; blood count, coagulation, and sickle-cell profiles had normal results, whereas corporal blood gas from the apex of the corpora cavernosa revealed values consistent with that of a low-flow state.

According to guidelines, a three-piece PPI was proposed to the patient, but he refused.^[4] Therefore, a T-shunt procedure with tunneling was performed using sequential Hegar dilators (up to 10 F diameter) to improve the corporal drainage. As described by Garcia et al.,^[5] the dilators are inserted through each glans incision, orientated slightly laterally to avoid the urethra, and advanced to the proximal corpora. Incision sites are closed, and

compressive dressings are avoided to favor drainage through the shunts. Detumescence was obtained intraoperatively at the level of the proximal two-third of the penis, and the distal one-third of the shaft maintained mild tumescence probably because of post-ischemic hyperemic state and local edema despite evacuation of trapped ischemic blood from the corpora.^[6] During the hospital stay, a quite complete detumescence was maintained through constant execution of the “milking maneuver.” Surprisingly, on postoperative day 4, the penis showed an attitude of resumption of a complete nonpainful erection. Corporal blood gas was taken, with the values consistent with that of a high-flow state. A penile Doppler ultrasound study confirmed high velocity in both the right and left cavernosal arteries (63 and 56 cm/s, respectively) (Figure 1); selective pudendal angiography was performed showing arteriovenous fistulous lesions apparently originating from the right bulbar and cavernous arteries.

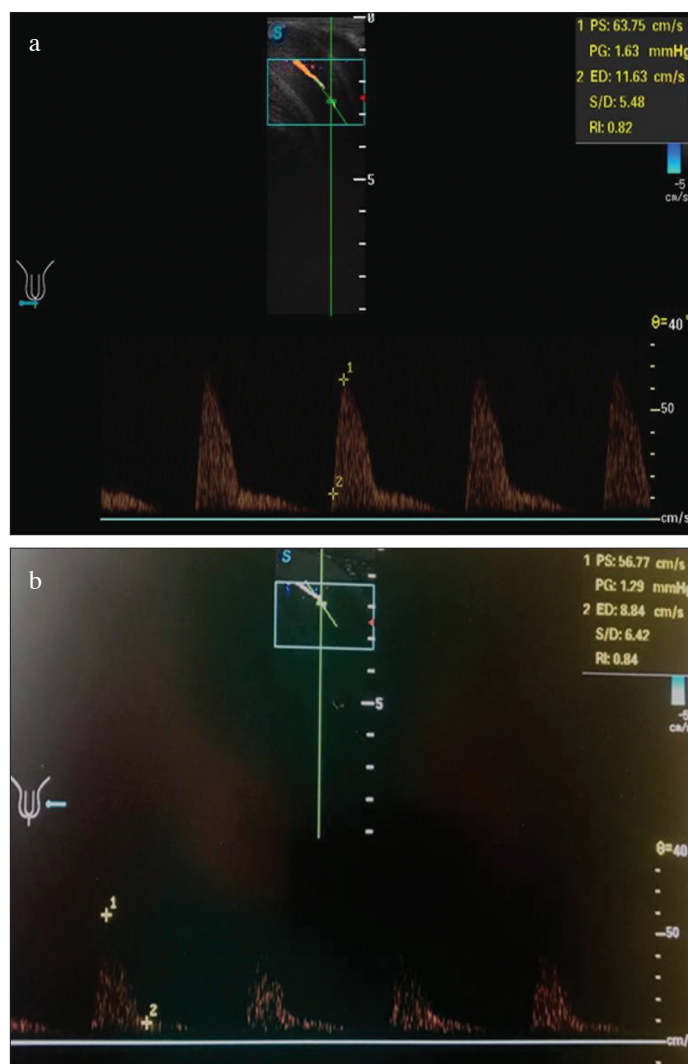


Figure 1. a, b. Postoperative day 4: penile Doppler ultrasound study shows high-velocity flow in the cavernosal arteries consistent with a high-flow state

Main Points:

- High-flow priapism represents a poorly understood complication for some patients with idiopathic or recurrent ischemic priapism.
- High-flow state that occurs after the treatment of a veno-occlusive priapism should always be considered in differential diagnosis of the patients in whom treatment fails.
- Increased utilization of T-Shunt with tunneling may increase the conversion of low-flow priapism to high-flow state.
- In delayed ischemic priapism, patients should always be counseled about the non-negligible effect on erectile function, and an early malleable or inflatable penile prosthesis implantation can represent a safe solution with a decreased rate of complications and improved long-term outcomes.

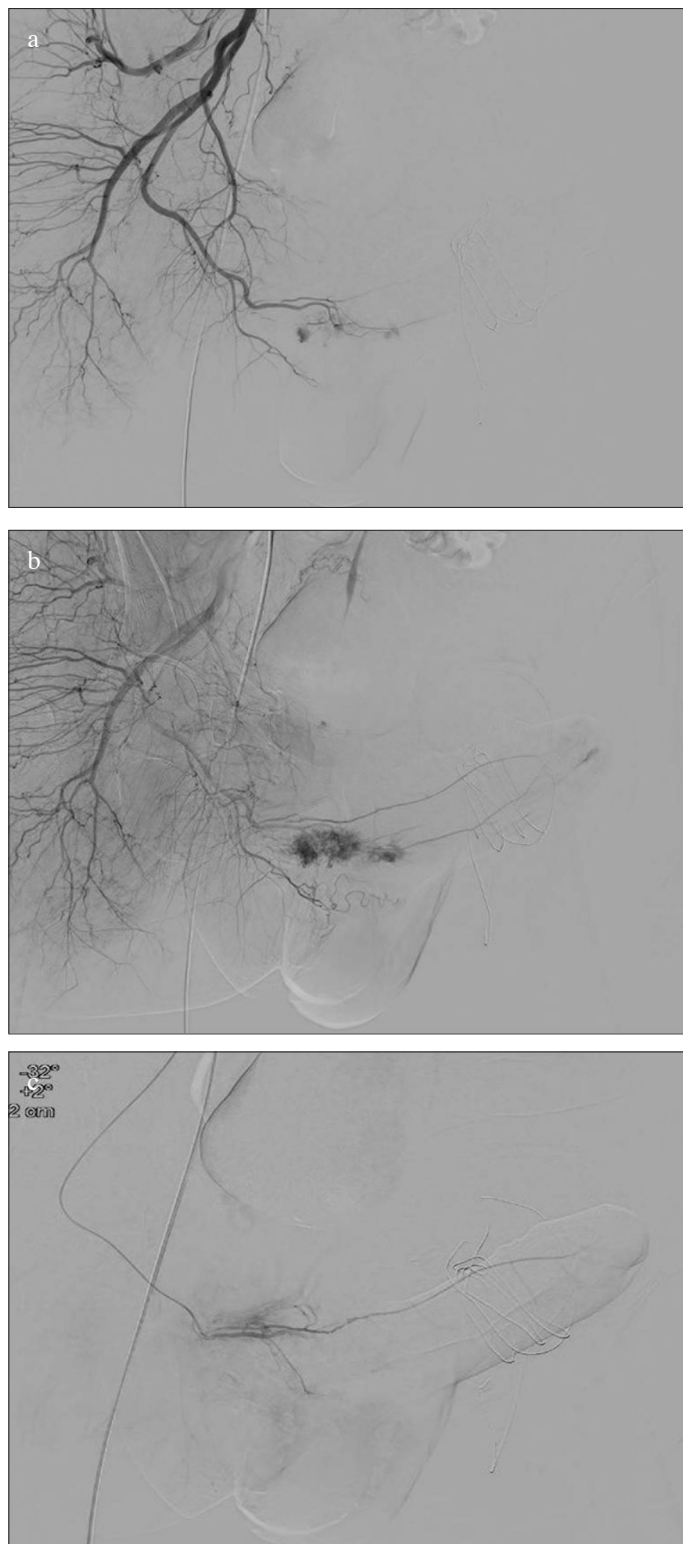


Figure 2. a-c. Selective pudendal angiogram of the left dorsal artery shows arteriovenous fistula originating from the right bulbar and cavernous arteries (a, b). Embolization with reabsorbable gelatin sponge material resulting in a complete obliteration (c)

After appropriate superselective catheterization, embolization was carried out with reabsorbable gelatin sponge material resulting in a complete obliteration (Figure 2).

After 72 hours without a renewing erection, the patient was discharged. At 3-month follow-up, the patient reported that he was able to achieve only partial erection without satisfaction during sexual intercourses; therefore, he underwent a three-piece PPI surgery with a definitive optimal functional outcome.

Discussion

IP is characterized by the inhibition of venous outflow that leads to impairment of arterial flow with a secondary risk of cavernosal fibrosis and erectile dysfunction. Shunt procedures are recommended when the aspiration and injection of pharmacological agents fail to achieve penile detumescence or in case of delayed IP. Both the AUA and EAU guidelines recommend surgical intervention using first distal shunts and then proximal shunts.^[3,4] However, in IP lasting >36 hours, shunt surgery might not reverse the ischemic damage; thus, long-term refractory erectile dysfunction would still be inevitable.^[7]

Delayed PPI might be associated with lower levels of satisfaction for the patients so that early implantation in IP has been proposed owing to the fact that it provides resolution of compartment syndrome along with the treatment of erectile dysfunction with a decreased rate of complications and improved long-term outcomes.^[8]

Herein we reported an interesting case of NIP presented after a T-shunt procedure. The pathophysiology is still unclear and poorly understood. Conversion of low-flow to high-flow state as a result of recent surgery has been described in relation with the tunneling portion of the distal shunting^[9] because of an iatrogenic arteriolar-sinusoidal fistula with shearing of the cavernosal artery^[10] or dorsal artery.^[11] Concurrent mechanisms may be represented by the depletion of vasorelaxing factors of the cavernosal system during the ischemic state^[12] and the local inflammatory vasodilatation with hyperemia owing to the release of nitric oxide and cytokines.^[13]

Is this risk correlated with an increased use of T-shunt with tunneling? This is difficult to establish because current literature provides case studies from which it is difficult to draw definitive statements, but clinicians should be aware in case of penile tumescence after shunting procedure for IP that may suggest a conversion to high-flow state.

In this setting, penile Doppler ultrasound is an essential diagnostic tool in absence of established etiology, and a selective pudendal angiogram can confirm the diagnosis allowing the treatment of NIP through embolization of the vascular abnormality.

However, it is fundamental to note that patients should always be counseled about the non-negligible effect on the erectile function because after 36–48 hours of IP, the damages of the cavernosal muscle are irreversible. In such cases, an early malleable or inflatable penile prosthesis implantation represents a recommended and technically safe solution to achieve a satisfactory functional outcome,^[2] minimizing all the possible sequelae related to the distal shunt procedure.

Informed Consent: Written informed consent was obtained from patient who participated in this case.

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