Successful Management of Recurrent High-Flow Priapism Treated with Selective Arterial Embolization: A Case Report

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ABSTRACT

Introduction: High-flow priapism is rare, uncontrolled arterial inflow, preceded by penile or perineal trauma and arterial-lacunar fistula. There are several ways to treat high-flow priapism, i.e., conservative management, the use of ice packs, mechanical decompression, surgery, and super-selective arterial embolization. Embolization is currently widely accepted in patients who fail from conservative management. This study aimed to report the use of Gelfoam and microcoil embolization in recurrent high-flow priapism compared to PVA embolization.

Case Study: A 36-year-old man complained of prolonged erection. The erection occurred three days before admission while waking up in the morning, not accompanied by either sexual stimulation or pain. There was a history of fall four days ago in the afternoon, with the patient's groin hitting a rocky ground. Physical examination revealed an erect penis, which felt warm, with an EHS of 4. Blood gas analysis of the corpus cavernosum showed bright red blood with a pH of 7.47, pCO_2 of 23.6, pO_2 of 145, HCO_3 of 17.3, BE of -6, and SaO_2 of 99%. Doppler ultrasound examination of the penis showed high-flow priapism. Embolization with PVA was performed, and there were decreased complaints. A few hours later, the erection occurred. Reevaluation was then performed and continued with embolization using Gelfoam and microcoil. There were immediate successful results (EHS of 3) accompanied by a decrease in symptoms. Long-term follow-up has shown a return to normal erectile function six months following the injury.

Conclusion: Priapism may happen due to various etiologies. Differentiating high-flow and low-flow is paramount during the acute phase because of different treatment strategies. Conservative management may be applied to high-flow priapism. If conservative management fails, embolization may be attempted. The choice of embolization agent must be taken into account.

CASE REPORT

CASE REPORT

A 36-year-old man came with a chief complaint of a prolonged erection. Erection has occurred continuously for the past three days in the morning after waking up without sexual stimulation. The erection was not accompanied by pain. There was a history of falling while working four days prior, with the position of the groin hitting rocky ground; the groin felt painful after the fall. There were no complaints of bloody meatal discharge and hematuria. The last time he had sex was four days before. The patient had no history of neurological diseases, leukemia, or other hematological abnormalities. A history of drug use that stimulates an erection was denied. There was no history of hypertension and diabetes mellitus.

On physical examination, we found an erect penis, palpably warm, with an erectile hardness score (EHS) of 4 (Figure 1). On the perineum, there appears to be a hematoma with tenderness. Based on examination of the corpus cavernosum, blood gas analysis showed bright red blood with a pH of 7.47, pCO_2 of 23.6, pO_2 of 145, HCO_3 of 17.3, BE of -6, and SaO_2 of 99%. Doppler ultrasound examination of the penis revealed high-flow priapism (Figure 2).

An angiography examination was conducted, revealing the presence of an extravasation in the right bulbar artery and the left helical artery (Figure 3). Afterward, the embolization procedure was conducted using bilateral polyvinyl alcohol (PVA), revealing the absence of both fistula and leakage. Partial detumescence occurred with the erection hardness score reaching a value of 3. Subsequently, compression therapy was done. Nevertheless, after a few hours passed, there was a further increase observed in the score measuring the hardness of the erection. The Doppler ultrasonography examination revealed the presence of proximal high-flow arterial lesions in the identical anatomical site as previously observed.

Subsequently, we conducted an embolization treatment using Gelfoam and microcoils, wherein no apparent signs of fistulas and extravasation were observed (Figure 5).

MONITORING AND EVALUATION

One week post-embolization, we did an evaluation, and there was partial detumescence with an EHS of 3; three months post-embolization, it was seen that the EHS was 1 and the patient had a normal erectile function.

DISCUSSION

Etiology and demographics

Priapism is a medical condition characterized by a prolonged and persistent penile erection lasting for a duration exceeding four hours in the absence of any exciting sexual activity. Priapism is divided into two categories: low-flow (ischemic) and high-flow (non-ischemic) [1]. With an estimated prevalence of 1.5 per 100,000 males, high-flow priapism is a rather uncommon medical problem [2-4].

The most prevalent kind of priapism, low-flow priapism, makes up 95% of all instances and is characterized by a painful, prolonged erection with unusual corpora cavernosa rigidity brought on by a problem with venous blood outflow from this tissue mass [5]. There are several other hemoglobinopathies, such as sickle cell disease and thalassemia, as well as any hypercoagulable condition that can produce low-flow priapism. Vasoactive drugs, such as those used to treat erectile dysfunction, are estimated to be the cause of at least 25% of cases of this condition [6]. Less often occurring high-flow priapism is typically the consequence of physical trauma or injury. In addition, congenital artery abnormalities, malignancy, and iatrogenic damage during surgical procedures can cause paresthesia. For some, the disease has no known underlying cause or explanation [5-7].

Clinical and imaging findings

Our case report describes trauma-induced high-flow priapism in a 36-year-old male. These symptoms manifest subtly due to the formation of an arterio-cavernosal fistula four days after experiencing trauma in the perineal or genital area [9]. This delayed presentation may be attributed to the resolution of arterial spasms caused by the injury, or it could be explained by the recanalization of a previously blocked penile artery. Interestingly, about 66% of patients exhibit prompt penile erection subsequent to trauma, but in the remaining cases, priapism manifests within an interval ranging from 1 to 72 hours. This observation indicates that the development of substantial hemodynamic fistulas may result from initially minor vascular anomalies [2-11]. In fact, approximately 99% of all fistulas occur in the internal pudendal artery and its branches, with bulbourethral arteries accounting for around one-third of cases [10]. When arteriocorporal fistula appears in these blood vessels, arterial overflow that remains unregulated may lead to recurrent high-flow priapism. [12]. However, there are case studies and literature reviews on high-flow priapism. There are certain cases of high-flow priapism, the causative factor of which remains unknown. This condition is called idiopathic high-flow priapism. These studies describe that idiopathic high-flow priapism often presents in pediatric population [7-11].

Treatment and prognosis

High-flow priapism treatment is not an emergency due to the minimal risk of persistent complications; however, patients with untreated long-term conditions have reported decreased efficacy [10]. Initially, conservative treatment including observation, and penile compression must be performed as the condition might self-resolve. However, failure of these methods requires further treatments. [10]. In the first 24–48 hours of onset, inducing cavernosal vasospasm through the application of ice and perineal compression is considered the initial step in conservative management of these cases; this facilitates thrombus formation and fistula obliteration. Angiography and selective artery embolization should be considered if the condition ceases to improve [8].

There are two types of embolic agents: permanent (i.e., polyvinyl particles, glue, and microcoils,) and non-permanent (i.e., Gelfoam, sponge, or autologous blood clots). It is important to note that the effectiveness of all of these occlusive agents is nearly comparable; nevertheless, permanent materials have a higher incidence of complications but a lower likelihood of recurrence compared to temporary materials [9].

To date, there has been considerable debate regarding the most effective materials for embolization. Various materials, including gelatin sponges, micro-coils, autologous blood coagulation, and polyvinyl alcohol (PVA) can be applied for superselective arterial embolization [13]. Selective temporary embolization is generally done using an autologous Gelfoam or autologous clot. Permanent embolization, on the other hand, is achieved via coiling or acrylic adhesive.

The application of an absorbable material leads to temporary blockage, hence decreasing the likelihood of erectile dysfunction despite the presence of a higher rate of recurrence. In contrast, the non-absorbable microcoil is preferable in preventing recurrence

but carries a higher risk of future impotence, in particular when bilateral embolization is performed [13]. Permanent materials have the potential to produce a more long-lasting occlusion than absorbable ones, resulting in a decreased risk of priapism recurrence. However, there is a chance of post-interventional persistent erectile dysfunction (ED), particularly in situations when there is a bilateral fistula. For a few circumstances that do not respond to treatment with the main temporary materials, permanent materials should be opted for instead. A more advanced and contemporary technique of angiography has been shown to effectively mitigate the risk of developing erectile dysfunction (ED) due to permanent embolism, an effect that has been compared to that of temporary embolism. [12].

In this case report, there is a recurrence of priapism after embolization with PVA material. PVA in the initial treatment was chosen as a material because it is a superselective embolic agent that is able to penetrate the microvascular tissue. [16,17], When recurrence occurs, we decided to perform selective embolization with the permanent agent Gelfoam and coils in the right cavernosal artery. In conclusion, our patient presented recurrent high-flow priapism; treating this problem with double-agent selective embolization has been proven to be effective and safe.

Differential Diagnosis

Differentiating high-flow priapism from low-flow priapism is crucial for determining the appropriate treatment course. Based on clinical findings, high-flow priapism is typically painless, while low-flow priapism is often accompanied by severe pain and tenderness in the penis. Erection characteristics in high-flow priapism include the condition where the erection is typically a pulsatile sensation; in low-flow priapism, the erection may lack pulsation. The gland appearance of high-flow priapism is typically normal in color; in low-flow priapism, the glands may be cyanotic due to venous congestion.

Doppler ultrasound also allows the differentiation of high-flow from low-flow priapism. A list of imaging findings is shown in the differentiating table.

A cavernous blood gas analysis, which verifies the storage of venous blood within the corpora cavernosa, can also be performed. It is advised to perform a cavernous blood gas study to differentiate between ischemic and non-ischemic priapism. Lower partial oxygen pressure (pO_2 < 30 mmHg), a higher partial carbon dioxide pressure (pCO_2 > 60 mmHg), and a decline in pH (< 7.25) are found in ischemic priapism. The corporeal blood gas study in non-ischemic priapism shows normal arterial blood gas values of pH 7.40, pCO_2 < 40 mmHg, and pO_2 > 90 mmHg [5,21].

TEACHING POINT

Priapism may happen due to various etiologies. Differentiating high-flow from low-flow is paramount during the acute phase because of different treatment strategies. Patients with high-flow priapism may be treated initially with conservative management. If conservative management fails, embolization may be attempted. The choice of embolization agent must be taken into account. Especially in cases of recurrent priapism, the utilization of double-agent Gelfoam and microcoil has been proven to be effective and safe.

AUTHORS' CONTRIBUTIONS

Alfryan Janardhana: Followed up the patient, wrote the manuscript.

Besut Daryanto: Processed the patient, followed up the patient, wrote the manuscript, reviewed the manuscript.

Andri Kustono: Processed the patient, followed up patient, wrote the manuscript, reviewed the manuscript.

Bayhaqi Nasir Aslam: Processed the patient, followed up the patient, wrote the manuscript, reviewed the manuscript.

CONSENT

Written Informed Consent was obtained from the patient for publication

HUMAN AND ANIMAL RIGHTS

Ethical approvals were signed by Health Research Ethics Commission of Dr. Saiful Anwar General Hospital and reported in letter number 400/010/CR/102.7/2023

QUESTIONS

Question 1: Which of the following is the primary goal of embolization in high-flow priapism?

a. To decrease arterial inflow to the corpora cavernosa
b. To increase venous outflow from the corpora cavernosa
c. To induce fibrosis of the corpora cavernosa
d. To prevent erectile dysfunction

Explanation Question 1: The correct answer is A.

The primary goal of embolization in high-flow priapism is to decrease arterial inflow to the corpora cavernosa

[The primary goal of managing high-flow priapism is to occlude the fistula without damaging the surrounding normal arteries or the patient's ability to maintain an erection]

Question 2: Which of the following is the most common type of embolic agent used in high-flow priapism?

a. Gelfoam
b. Polyvinyl alcohol particles
c. Steel coils
d. Sclerosing agents

Explanation Question 2: The correct answer is B.

PVA Particles are relatively non-reactive and have a high success rate of occluding the feeding arteries to the corpora cavernosa. In addition, PVA particles are biocompatible and do not degrade over time.

[Various material, including gelatin sponge, micro-coils, autologous blood coagulation, and polyvinyl alcohol (PVA), can be applied for superselective arterial embolization]

Question 3 : Which of the following is a potential complication of embolization for high-flow priapism?

a. Impotence
b. Infection
c. Tissue necrosis
d. All of above

Explanation Question 3: The correct answer is D.

Impotence estimated that up to 20% of patients will experience some degree of erectile dysfunction; infection on the corpora cavernosa can lead to tissue death, and necrosis will occur and become a life threat if it occurs. [Excessive arterial inflow with high oxygen levels and prolonged erection, caused by guanylate cyclase enzyme activation, may be detrimental to the cavernosal smooth muscle and connective tissue matrix, leading to irreversible corporal fibrosis and consequently erectile dysfunction]

Question 4: Which of the following is the most common indication for embolization in high-flow priapism?

a. Failure of conservative therapy
b. Recurrent priapism
c. Severe pain
d. All of the above

Explanation Question 4: The correct answer is D

The indications for embolization in high-flow priapism are persistent erection, failure of conservative management, severe pain, and recurrent priapism after embolization.

[Within the first 24–48 hours of onset, conservative management is considered the first step in the management of these cases, during which perineal compression and application of ice are used to induce cavernosal vasospasm in order to facilitate clot formation and fistula obliteration. If the condition doesn't improve, angiography and selective artery embolization should be taken into consideration]

Question 5: Which of the following is the most accurate statement about the success rate of embolization for high-flow priapism?

a. Embolization is always successful in resolving high-flow priapism
b. Embolization is successful in resolving high-flow priapism in approximately 70–90% of cases
c. Embolization is successful in resolving high-flow priapism in approximately 50% of cases
d. Embolization is successful in resolving high flow priapism in approximately 20–30% of cases

Explanation Question 5: The correct answer is B

The success rate of embolization in high flow priapism is around 70–80%

[The success rate is 80%, but there is still a significant failure or recurrence rate, ranging 30–40%]

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FIGURES

A B

Figure 1: A 36-year-old male came to the emergency room with a chief complaint of the penis appearing erect with an EHS of 4 and not accompanied by pain

(A) Anterior side

(B) Lateral side

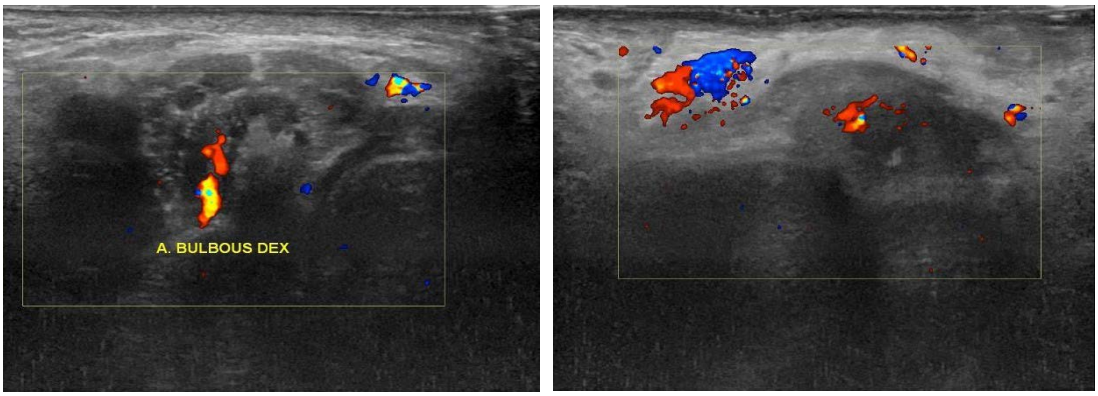

Figure 2: Doppler ultrasound: Flow can be seen in the right and left corpus spongiosa with a yin-yang sign (proximal). The appearance of a high-flow vascular lesion in the corpus spongiosum (proximal)

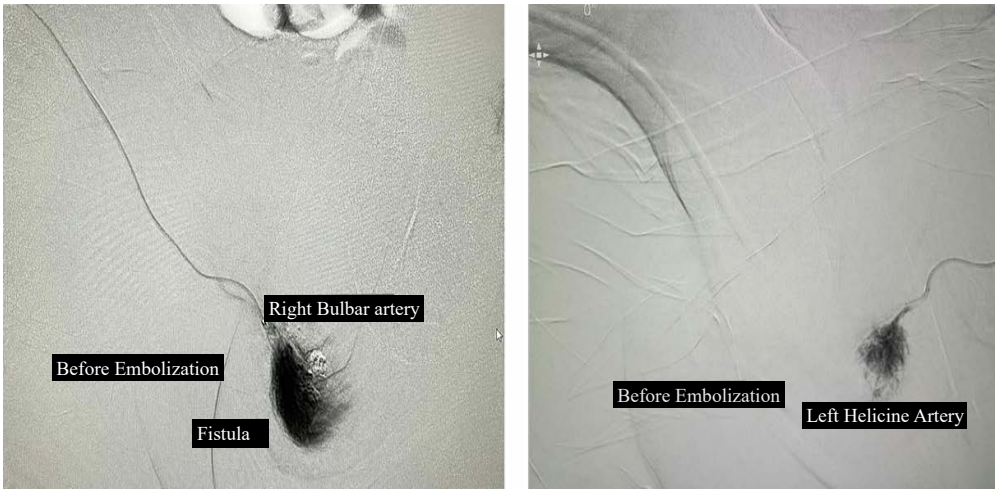

Figure 3: Angiography Examination: Visible extravasation of the right bulbar artery and the left helical artery on angiography.
[Technique: Puncture of the right and left common femoral artery was performed under ultrasound guidance. The 0.035" wire is entered under fluoroscopy guidance Cobra catheter size 5Fr with 0.035" hydrophilic wire used for navigation to the right and left internal pudendal artery, and A 2.4 Fr Merit microcatheter with a 0.014" floppy microwire is used for super selective catheterization]

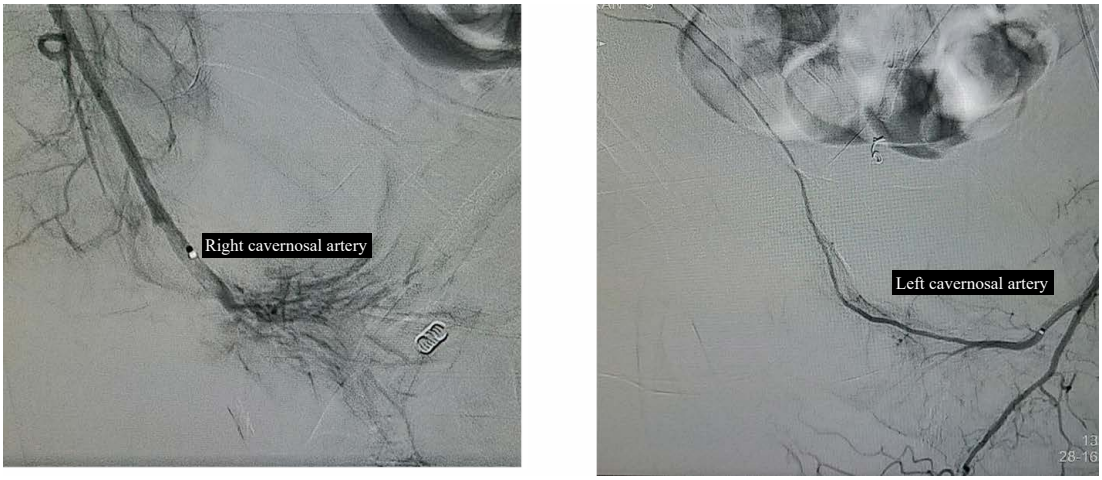

Figure 4: Angiography examination: After the first attempt of embolization, there is an extravacation in the right cavernous artery, and the left cavernous artery has improved with no extravasation.

[Technique: Puncture of the right and left common femoral artery was performed under ultrasound guidance. The 0.035" wire is entered under fluoroscopy guidance Cobra catheter size 5Fr with 0.035" hydrophilic wire used for navigation to the right and left internal pudendal artery, and A 2.4 Fr Merit microcatheter with a 0.014" floppy microwire is used for super selective catheterization]

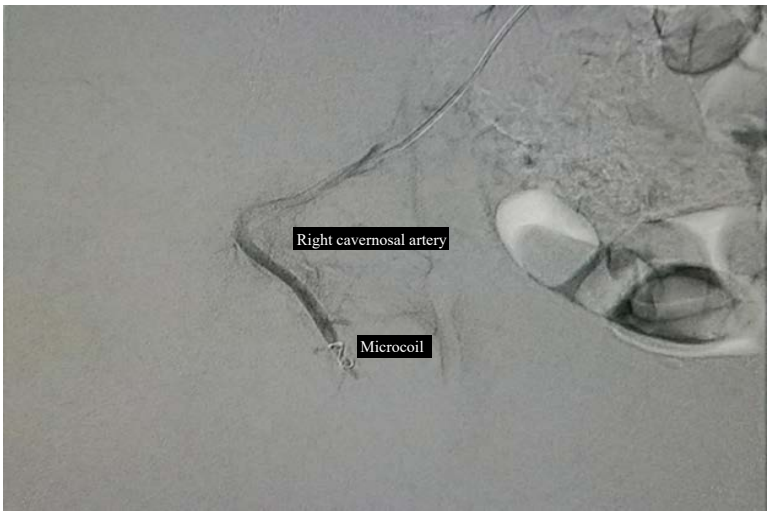

Figure 5: Angiography Examination: After the second attempt, selective embolization with microcoils and Gelfoam at right cavernosal artery was performed; no fistulas and extravasation were found

[Technique: Puncture of the right common femoral artery was performed under ultrasound guidance. The 0.035" wire is entered under fluoroscopy guidance Cobra catheter size 5Fr with 0.035" hydrophilic wire used for navigation to the right internal pudendal artery, and A 2.4 Fr Merit microcatheter with a 0.014" floppy microwire is used for super selective catheterization]

Table 1: Summary table of high flow priapism

Summary Table	High-Flow Priapism
Etiology	Arteriocorporeal fistula caused by trauma
Incidence	Approximately 0.3 to 1.5 per 100,000 men per year
Gender ratio	Male
Age Predilection	Middle age
Risk Factor	Penile or perineal trauma, spinal cord injury, Peyronie's disease
Treatment	Conservative using mechanical compression or ice packs, if the patient failed to show results from the conservative management, embolization with superselective embolization may be selected. Surgery may be necessary to repair the fistula and is only considered a last resort treatment.
Prognosis	Depends on the promptness of treatment. Permanent erectile dysfunction is more likely to occur if the treatment is delayed. The permanent agent selective embolization is less invasive and showed improvement of the patient's symptoms with no side effects in follow-up evaluation
Finding on imaging	Ultrasonography: - Increased arterial flow, presence of an arteriocorporeal fistula. Angiography: - Extravasation of the right bulbar artery and the left helical artery form a fistulation to helicine artery. - Extravasation of the right cavernous artery. - Coil embolization of the right cavernosal artery.

Table 2: Differential Table for High-Flow Priapism

	Doppler ultrasound	**MRI**
High Flow Priapism	- Arteriocorporal fistula may be present. - Elevated penile artery Doppler velocity.	- Flow voids in cavernosa may be visualized.
Low Flow Priapism	- Thrombosis of the corpus spongiosum or corpus cavernosa. - The cavernosal artery/arteries reveal a decreased or absent color flow or spectral Doppler.	- Not indicated due to time-consuming. - Abnormal increased signal in the corpora indicating a thrombus.

ABBREVIATIONS

PVA = Polyvinyl Alcohol Particle; EHS = Erectile Hardness Score; ED = Erectile Dysfunction

KEYWORDS

Arteriocorporeal fistula; Embolization; High Flow Priapism; Interventional Radiology; Penile Trauma; Recurrent Priapism