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Case Report

Endovascular intervention with high-flow priapism: A case report ☆,☆☆

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ABSTRACT

High-flow priapism is a rare condition in the form of priapism unrelated to sexual stimulation. The disease causes a decline in quality of life and has the risk of causing long-term erectile dysfunction if not treated promptly. We report a case of a 48-year-old male patient with prolonged high-flow erection after trauma who received endovascular intervention with n-butyl-2 cyanoacrylate biological glue. Results after 40 days of intervention and use of sildenafil 25 mg/day, the patient has an erection and has normal sexual activity. Cavernous arteriovenous fistula causing priapism with high flow is a rare condition. Computed tomography helps supplement information about feeding vessels as well as accompanying injuries for comprehensive assessment before treatment. Currently, there are many treatment methods, but transcatheter arterial embolization is modality of choice for achieve clinical efficacy and can safely and flexibly reduce the risk of secondary erectile dysfunction.

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Introduction

Priapism is defined as a persistent and painful erection lasting more than 4 hours without sexual stimulation with an

incidence rate of 0.3 to 1.0/100,000 men each year, with the most common age being 40-50 years old [1]. According to pathophysiology, priapism is divided into 2 types: ischemic (low flow), nonischemic (high flow). Nonischemic priapism includes 3 main types:

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- Trauma: most commonly due to trauma to the penis or pelvis causing arteriovenous fistula.
- Neurological: autonomic imbalance such as due to spinal cord injury or cauda equina syndrome.
- Post-fistula: results from reactive hyperemia in response to hypoxia and ischemic acidosis lasting more than 24 hours [2].

High-flow persistent erection is often diagnosed by clinical symptoms, color Doppler ultrasound, computed tomography, or contrast-enhanced magnetic resonance imaging, and digital subtraction angiography are the gold standard in diagnosis and treatment [3].

We report a case of a patient with high-flow priapism after perineal trauma with complications of cavernous arteriovenous fistula treated with endovascular intervention.

Case report

A 48-year-old male patient was admitted to the hospital because of priapism after a high-fall accident causing perineal injury for 3 days. The patient has no medical history, is married and has 5 children. Clinical examination: bruising of the perineum, mild, and continuous erection of the penis, no pain, normal urination, no swelling, or bruising on both sides of the scrotum.

On 256-row computed tomography image with contrast enhanced: the right cavernous artery separating from the internal pudendal artery is slightly dilated compared to the left. The right cavernous venous sinuses enhanced early in the arterial phase, with no leakage of contrast from the cavernous body (Fig. 1), suggesting right cavernous arteriovenous fistula.

After a computed tomography scan 1 day, the patient had a digital subtraction angiogram and found a small pseudoaneurysm in the cavernous artery with unilateral cavernous arteriovenous fistula. The patient received intervention to embolize the fistula with a mixture of n-butyl-2 cyanoacrylate (Histoacryl): lipiodol in a ratio of 1:4. The last control showed complete blockage of the cavernous arteriovenous fistula and small pseudoaneurysm (Fig. 2).

After 2 days of intervention, the patient no longer had erections and was given sildenafil 25 mg/day to support erections. After 40 days intervention and daily use of sildenafil, the patient has an erection and has normal sexual activity.

Discussion

Priapism erection is an erection unrelated to sexual stimulation lasting more than 6 hours, the erection is limited to the corpus cavernosum without affecting the corpus spongiosum. There are 2 different types of priapism: low-flow and high-flow [4].

Low-flow priapism clinically manifests as a condition in which the penis is completely erect, causing continuous pain due to blockage of venous outflow, causing cavernous distension by low-oxygenated venous blood. This condition leads to hypoxia, acidosis, ischemia and penile fibrosis, leading to erectile dysfunction and impotence if not treated urgently [4]. Meanwhile, high-flow prolonged priapism, first described by Burt et al (1960) after coital trauma, is a less common form of priapism, leading to damage to the cavernous artery or one of the branches of the penile artery, leading to a cavernous arteriovenous fistula with a small pseudoaneurysm of the cavernous artery as in our reported case, and pseudoaneurysm [5].

Clinical diagnosis of high-flow priapism is based on history of perineal trauma, clinical examination shows mild priapism, no ischemia [6]. Usually, a period of several days between the injury and the onset of priapism can be explained by the formation of a physiological blood clot by the body after the injury that seals the damaged artery and can slough off. then in the edema and inflammation phase [7]. In fact, this was the natural progression of high-flow priapism in our case: 2 days after the injury, the patient was reported to have no symptoms except pain and ecchymosis of the genitals, on the third day the patient had continuous penile erection. Treating high-flow priapisms is not as urgent as low-flow priapisms because the outflow from the veins is not obstructed; thus, stasis and ischemia are avoided. These patients are at low risk of

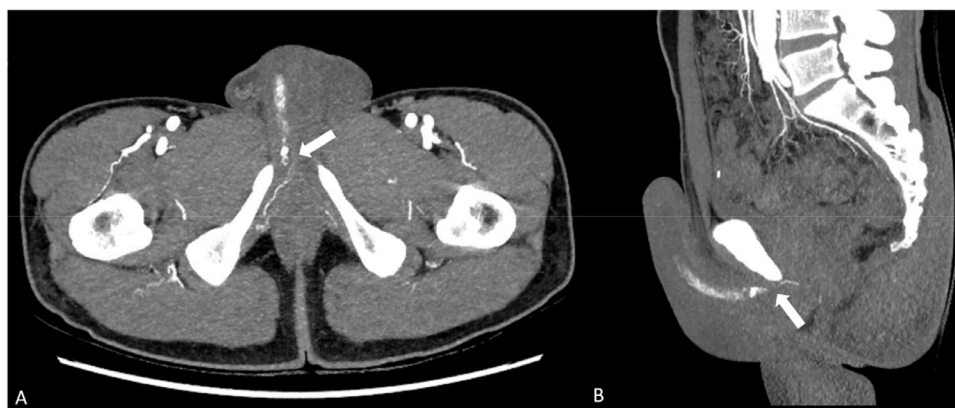


Fig. 1 – Contrast enhanced 256-row computed tomography scan of the abdomen and pelvis (arterial phase). (A, B) Axial and Sagittal: The right cavernous artery is slightly dilated, the right cavernous venous sinus enhanced early in the arterial phase suggest cavernous arteriovenous fistula (arrow).



Fig. 2 – Endovascular intervention of the lesion. (A) Internal iliac angiogram shows cavernous arteriovenous fistula (black arrow). (B) Selective angiography of the internal pudendal artery shows arteriovenous (white arrow). (C) Selective cavernous angiography shows a right arteriovenous fistula supplied by unilateral cavernous arteries and a small pseudoaneurysm of the right cavernous artery (white arrow). (D) After embolization with a mixture of Histoacryl and lipiodol, complete obliteration of the arteriovenous fistula and small pseudoaneurysm was achieved.

permanent complications, although reduced post-treatment erections have been reported in untreated patients with long-term disease [8].

Doppler ultrasound is the first-line diagnostic imaging method used to diagnose high-flow priapism due to its sensitivity and specificity of 100% and 73%, respectively [9]. Abnormal color Doppler signs are markedly increased flow in the cavernous body and arterialization of the cavernous veins with a typical “low impedance, high velocity” arterial waveform. However, ultrasound has many limitations: first, it is difficult to determine the exact location of the cavernous artery tear; second, the location, and number of collateral vessels and fistula vessels from the opposite side as well as difficulties in assessing accompanying injuries, so it is necessary to conduct a computed tomography scan of the abdomen and pelvis with injection before treatment.

Regarding therapeutic management, some authors recommend medical treatment in high-flow priapism because there

is no tissue ischemia and it can resolve spontaneously without danger. However, the disadvantage of medical treatment is that structural changes that can occur due to excessive arteriovenous blood flow can lead to long-term impotence as well as social difficulties. Treatment options include ice packs, intracavernous injection of alpha agonists, surgical ligation of the cavernous artery, and transcatheter arterial embolization. Among them, transcatheter arterial embolization is the preferred therapy for HFP, with a similar success rate to surgical ligation and a higher rate of preserving sexual function and is widely accepted. For cases that do not respond to conservative treatment methods, selective embolization of the cavernous arteriovenous fistula should be considered. Shigehara Kazuyoshi et al. reported priapism that was successfully treated by occlusion of the internal pudendal artery with both temporary (autologous blood clot, gelfoam) and permanent embolic materials (metal coil, n-butyl-2 cyanoacrylate and polyvinyl alcohol particles) [1]. Temporary materials are

often used for initial embolization because the rate of subsequent erectile dysfunction complications is approximately 32% of cases within 1 month of treatment, in 40.0% within 1~3 months, at 16.0% within 3~6 months and at 12.0% after more than 6 months; permanent erectile dysfunction occurs in <10% of cases. Meanwhile, the failure rate when using temporary materials is 60%-70% [1]. Permanent materials may contribute to a more durable obstruction than absorbable materials, which may achieve higher clinical success rates than temporary materials. However, there is a risk of permanent erectile dysfunction after intervention, especially in cases of bilateral fistula. In the reported case, the patient had a right cavernous arteriovenous fistula and an accessory feeding artery from the left cavernous artery, it was embolized with Histoacryl biological glue; after 40 days intervention and use sildenafil 25 mg/day, the patient has an erection and has normal sexual activity.

Conclusion

A rare disorder that causes priapism with high flow is cavernous arteriovenous fistula. For a thorough assessment prior to intervention, computed tomography can help complement information concerning feeding vessels and concomitant damage. Many therapeutic options are available today, but transcatheter arterial embolization is the most common technique for achieving clinical efficacy and can safely and flexibly lower the risk of secondary erectile dysfunction.

Author's contributions

Le TD and Nguyen MD: Case file retrieval and case summary preparation. Le TD, Tran QL, and Nguyen MD: preparation of manuscript and editing. All authors read and approved the final manuscript.

Availability of data and materials

Data and materials used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

Our institution does not require ethical approval for reporting individual cases or case series. Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

Patient consent

Informed consent for patient information to be published in this article was obtained.

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