CASE REPORT

Endovascular treatment of high-flow priapism using a combination of embolic materials: A case report and a review of the literature

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ABSTRACT

Background and aim: Priapism is a pathological condition defined as a prolonged and often painful erection, unrelated to sexual stimulation, that persists more than 4 hours. It is generally classified into different types based on its etiology and pathophysiology. High-flow (non-ischemic) priapism is a rare condition and typically results from perineal or penile trauma, which can lead to the formation of an arteriovenous fistula. This report details a case of high-flow priapism treated successfully with superselective arterial embolization, highlighting the diagnostic process, therapeutic approach, and follow-up outcomes.

Case presentation: We present the case of a 29 years-old male patient admitted to the hospital after bicycle trauma that occurred 24 hours earlier. Doppler ultrasound was performed and revealed a left-sided arteriovenous fistula. Selective catheterism of the left internal iliac artery showed an arteriovenous fistula fed by left pudendal artery, so a superselective microcatheterism was performed to reach the site of fistula. Transcatheter arterial embolization was carried out using gelatin sponge followed by the deployment of two detachable microcoils. Doppler ultrasound was repeated 24 hours after the embolization procedure, demonstrating a significant reduction of arteriovenous fistula with minimal residual flow, which wasn't deemed clinically significant. In the days following the procedure, the symptoms resolved completely, and the patient was discharged with neither recurrences nor complications.

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Conclusions: Endovascular embolization is safe and effective in the treatment of the high-flow non-ischemic priapism, offering a low rate of complications, short recovery time, and successful resolution of symptoms, preserving erectile function. (www.actabiomedica.it)

Key words: priapism, high-flow priapism, embolization, arteriovenous fistula, trauma

Background and aim

Priapism is a pathological condition defined as a prolonged and often painful erection, unrelated to sexual stimulation, that persists more than 4 hours. It is generally classified into different types based on its etiology and pathophysiology; understanding these distinctions is crucial for accurate diagnosis and appropriate treatment (1). The diagnosis of priapism relies on a through medical, sexual, surgical history, physical examination that includes the genitalia and perineum, corporal blood gas measurement at the initial presentation of priapism and penile Doppler ultrasound. Low-flow (veno-occlusive, ischemic) priapism accounts for more the 95% of the cases and results from impaired blood outflow from the penis, leading to hypoxia and acidosis in the erectile tissue (PO_2 typically less than 30 mmHg, pH < 7.2 at corporal blood gas). Thus, it is a true urological emergency; if left untreated, ischemic priapism can result in days to weeks of painful erections and prolonged ischemia, leading to cavernosal fibrosis and permanent loss of erectile function (2). Low-flow priapism can be considered a compartment syndrome, with various potential causes, though it is often idiopathic or associated with hematological disorders such as sickle cell disease (SCD) (3). Paraneoplastic diseases, spinal cord injuries, recreational substances like marijuana and cocaine, and second-generation antipsychotics are fewer common causes (4). First line treatment involves penile blood aspiration and intracavernosal injection of vasoactive agents; if these are unsuccessful, surgical intervention with distal corporoglanular shunt, with or without tunneling, should be considered (5).

Placement of a penile prosthesis may be necessary for patients with acute ischemic priapism that has been untreated for more than 36 hours or is refractory to surgery. Intermittent priapism is a recurrent form of ischemic priapism characterized by short-lived episodes of abnormal, painful tumescence that are self-limiting. It is seen almost exclusively in men with homozygous SCD. High-flow (non-ischemic) priapism is much rarer and typically results from perineal or penile trauma, which can lead to the formation of an arteriovenous fistula (AVF). In non-ischemic priapism, blood continues to flow into the penis, the blood is oxygenated (pO_2 > 90 mmHg, pCO_2 < 40 mmHg, pH 7.40), but it is not effectively trapped, resulting in a partially rigid, often painless erection (1). Corporal aspiration in non-ischemic priapism is generally diagnostic, showing high oxygen tension in intracavernosal blood, but imaging is required to confirm the presence and location of AVFs (6). The management of high-flow priapism is significantly different from low-flow priapism, as it is not a medical emergency due to the lack of ischemia. However, treatment is still necessary to prevent long-term complications such as erectile dysfunction. This report details a case of high-flow priapism treated successfully with superselective arterial embolization, highlighting the diagnostic process, therapeutic approach, and follow-up outcomes.

Case presentation

We present the case of a 29 years-old male patient admitted to the hospital after bicycle trauma that occurred 24 hours earlier. The patient presented with a partial rigid erection lasting more than 4 hours.

Blood gas analysis of the corpus cavernosum was performed, revealing a mean pH of 7.40 and a pO_2 of 92 mmHg, which supported the diagnosis of high-flow priapism. Doppler ultrasound was performed and revealed a left-sided AVF (Peak systolic velocity (PSV) 63,8 cm/s) (Figures 1, 2).

Written informed consent was obtained, and the patient underwent arteriography with the aim of embolizing the AVF. After administering local anesthesia, through a 5-Fr right femoral artery access, both internal iliac arteries were investigated using a selective diagnostic 5-Fr catheter (Cobra 5 Fr).

Selective catheterism of the left internal iliac artery showed AVF fed by left pudendal artery. After choosing the optimal projection, a superselective microcatheterism of the internal left pudendal artery was done using a 2.7-Fr microcatheter (Progreat®, Terumo), reaching the site of fistula (Figure 3).

Transcatheter arterial embolization (TAE) was performed using gelatin sponge. Then, due to the inadequate

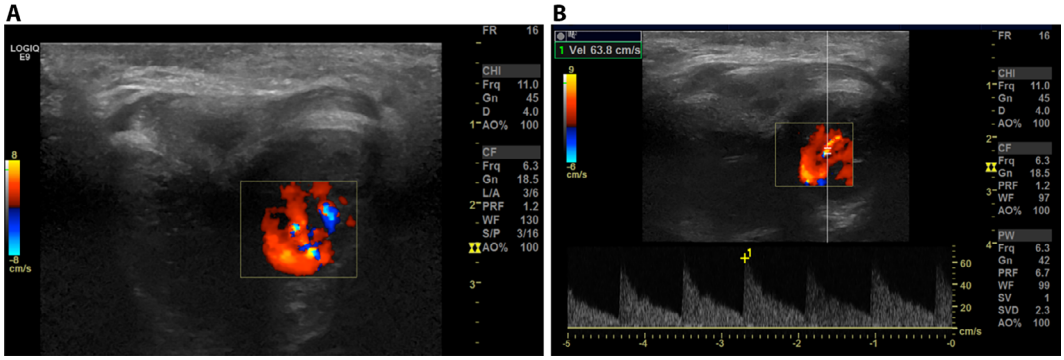

Figure 1. Doppler ultrasound shows a left-sided AVF, which looks like a dilated vascular lake with turbulent flow (A) and high flow velocity with high peak systolic velocity, PSV 63,8 cm/s (B).

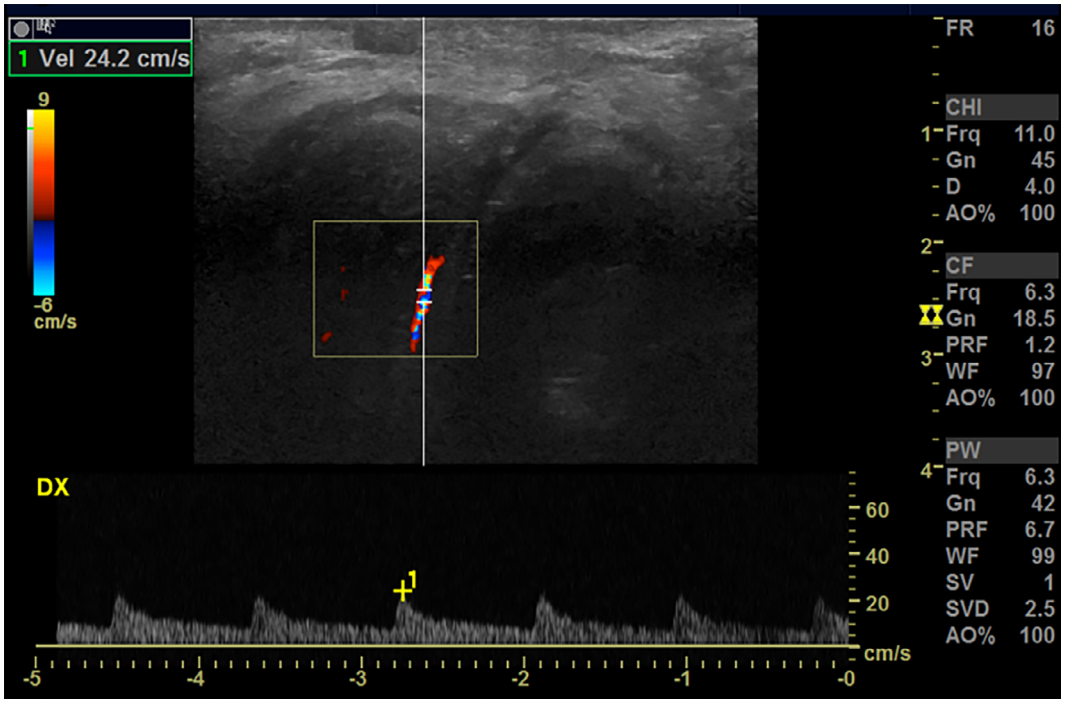

Figure 2. Normal flow on the right side (PSV 24,2 cm/s).

stop flow, complete embolization was achieved by the deployment of two detachable microcoils (Ruby Coil Soft, 3mmx5cm, Penumbra) (Figure 4).

The final angiograms documented deafferentation of the AVF. Right internal iliac arteriography showed normal penile and pudendal vasculature (Figure 5).

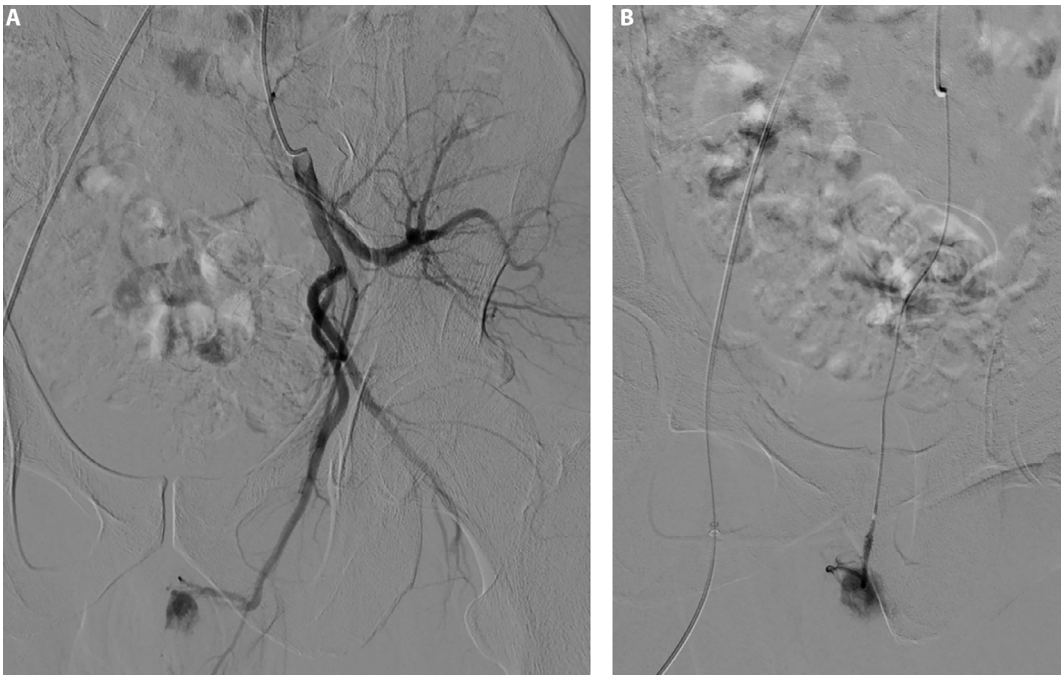

Figure 3. Selective angiogram of the left internal iliac artery with Cobra 5-Fr catheter shows left penile AVF fed by left internal pudendal artery (A). Superselective 2.7-Fr microcatheterism of left internal pudendal artery (B).

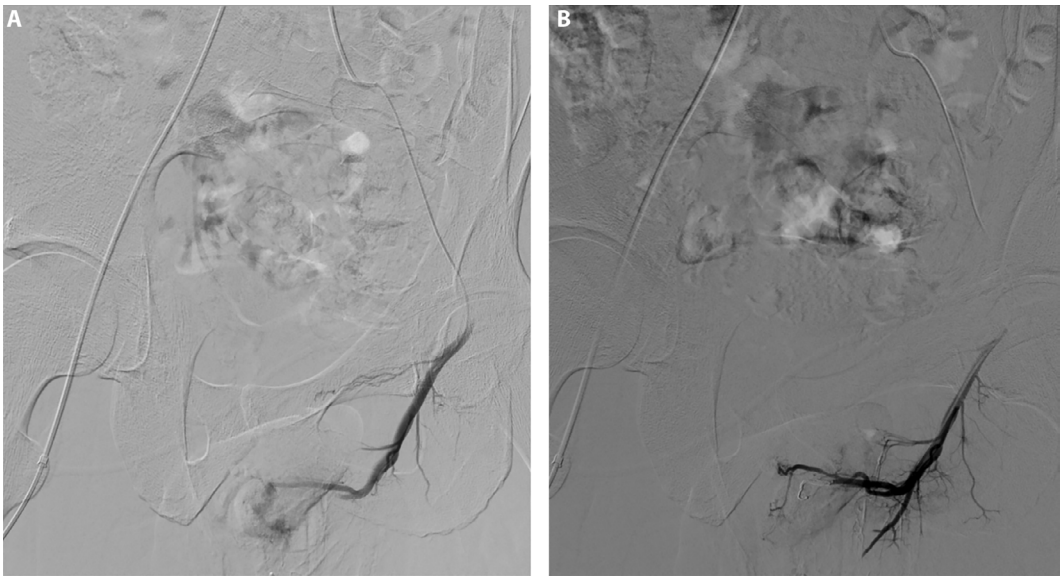

Figure 4. The fistula was embolized using gelatin sponge (A) followed by the deployment of two detachable microcoils (B).

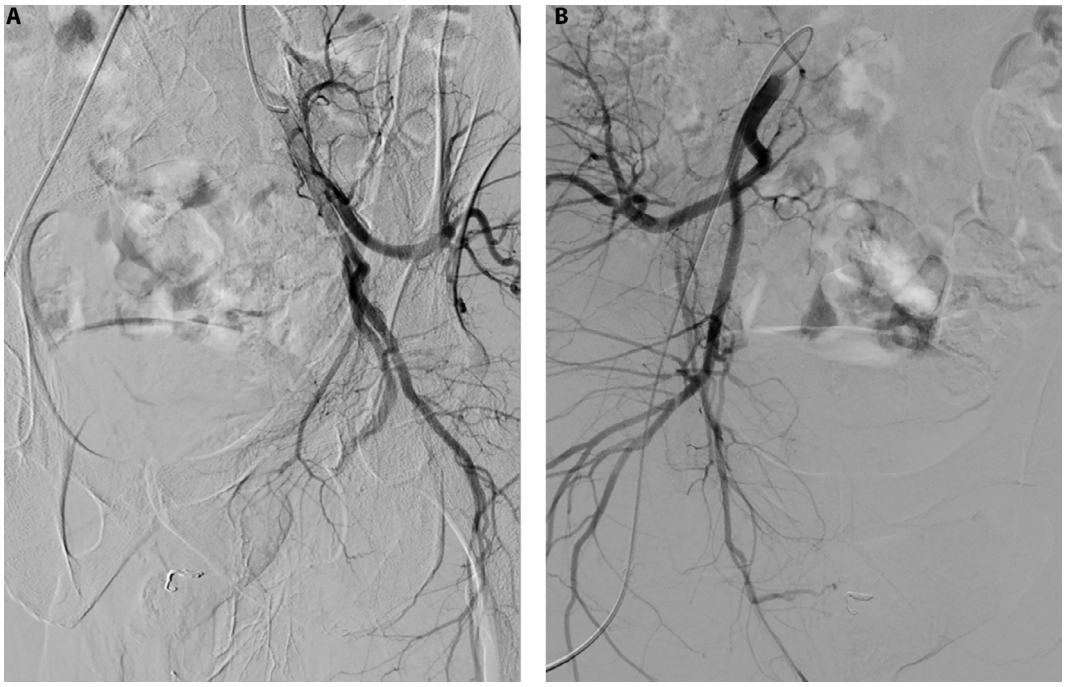

Figure 5. The final angiograms documented deafferentation of AVF (A). The selective angiogram of the right internal iliac artery showed normal opacification of the penile and perineal vasculature (B).

Doppler ultrasound was repeated 24 hours after the embolization procedure, demonstrating a significant reduction of AVF with minimal residual flow, which wasn't deemed clinically significant (PSV 41 cm/s) (Figure 6). B-mode ultrasound showed a hyperechoic area in the site of embolization with gelatin sponge, probably related to the presence of air microbubbles trapped during the gelatin sponge preparation.

In the days following the procedure, the symptoms resolved completely, and the patient was discharged with neither recurrences nor complications.

Discussion

High-flow priapism, also known as non-ischemic priapism, is a rare condition, accounting for approximately 5% of all cases of priapism (7). There are no precise epidemiological estimates quantifying the prevalence of high-flow priapism in the general population due to its rarity, and most of the available information comes from case reports or small case series. It is characterized by a prolonged, often painless erection, caused by an excessive influx of arterial blood into the corpus cavernosum. This condition is frequently caused by AVF or other vascular anomalies of the penis or urogenital system, most commonly following direct penile or perineal trauma, as observed in the case presented (8, 9). In high-flow priapism, an abnormal arteriovenous shunt bypasses the normal regulatory mechanism of erection, resulting in an unregulated influx of arterial blood into the penis (10). Doppler ultrasound is recommended to distinguish between ischemic and non-ischemic priapism, and to locate the site of fistula (5). A high-frequency transducer is used to visualize the cavernosal arteries from their origin in the perineum along the ventral aspect of the penile shaft. The procedure is performed with the patient standing with the legs in the frog position with the scrotum elevated, ensuring proper visualization (11). Doppler ultrasound can identify the site of cavernosal injury in more than 70% of cases (6). The characteristic waveform pattern of non-ischemic priapism is low resistance with high arterial flow. The affected cavernosal artery measurement shows a PSV exceeding 50 cm/s and an end diastolic velocity

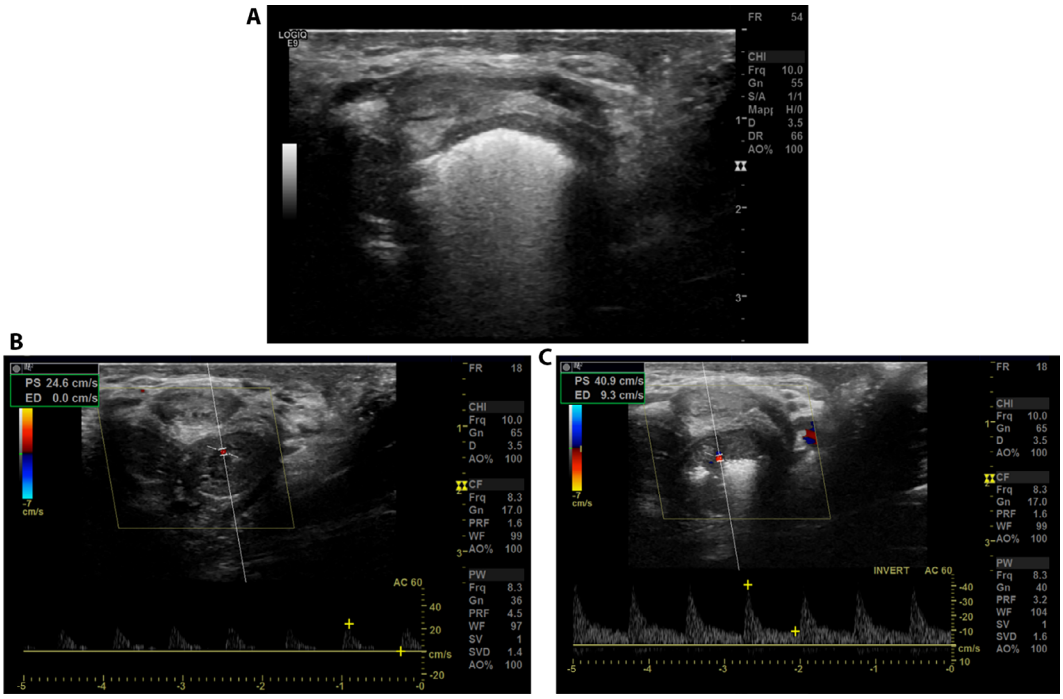

Figure 6. B-mode ultrasound showed a hyperechoic area in the site of embolization with gelatin sponge (A). Doppler ultrasound demonstrated a reduction of AVF with minimal residual flow (PSV 40,9 cm/s) (B, C).

(EDV) of over 10 cm/s with an associated resistive index (RI) of less than 0.7 (12). In contrast, in low-flow priapism, cavernosal arterial flow typically appears as a high-resistance, low-velocity waveform (11). Furthermore, doppler ultrasound can also be useful in assessing the effectiveness of embolization (13). It is common to observe a gradual resolution of the doppler ultrasound waveforms for up to six months following the resolution of the priapism. This is believed to be caused by reactive hyperemia during the healing process and potentially delayed healing of small arteriovenous shunts that are not distinguishable as AVF. Arterial embolization, regardless of the material used, has a success rate of approximately 89% and offers several advantages, including a brief treatment period, a short recovery time and an effective therapeutic outcome (14, 15). The primary goal of embolization is to close the AVF, while preserving the adjacent normal tissue and maintaining erectile function (16). In our case, we present the successful endovascular treatment of post-traumatic high-flow priapism with evidence of cavernosal AVF. Despite a small residual flow observed on Doppler ultrasound the day following the embolization, the patient's symptoms completely resolved. Numerous case reports and review articles report various embolic materials, which are commonly categorized into two groups: temporary occlusive agents such as gelatin sponge, and permanent occlusive agents including coils, plugs and glue (17). Other reviews analyze different types of embolic materials, with no significant difference regarding overall improved erectile function (1, 18). The use of permanent materials may cause a longer-lasting obstruction compared to absorbable materials. Liu et al. suggest that microcoils, as a permanent material, are more effective and provide accurate positioning within the targeted vessel. They suggested that microcoils were a more reliable agent, even in cases of recurrence and long-standing priapism. While theoretically, microcoils may increase the risk of permanent vessel occlusion, potentially leading to erectile dysfunction, no major ischemic events have been reported in the literature. Superselective embolization with microcoils has demonstrated results in achieving detumescence while preserving erectile function, especially in younger patients with a unilateral AVF (16). According to

Arrichiello et al., all materials guarantee a good clinical success (70%), with the best results achieved using gelatin sponge (89%) and the worst using polyvinyl alcohol (PVA) (70%). Gelatin sponge has a duration of action of five/six weeks after injection, allowing detumescence without causing permanent vascular occlusion (19). PVA (300–500 μm) are preferred when the preliminary angiographic studies show multiple small caliber fistulas, due to their ability to penetrate the microvascular circulation; furthermore, PVA injection must be performed with gradual pressure, to avoid proximal reflux (20). Additionally, patients treated with gelatin sponge experience a low recurrence rate (13%), while those treated with PVA have a higher recurrence rate (28%). N-butyl cyanoacrylate (NBCA), as an adhesive liquid embolic agent, is rarely used, with only a few case series reported in the literature (20). Although Ethylene-vinyl alcohol copolymer (EVOH), a non-adhesive liquid embolic agent, has been demonstrated to be safe and effective in arterial embolization while it is not employed in high-flow priapism (21). In the reported case we opted for embolization with gelatin sponge, followed by placement of microcoils. The decision to combine two different embolic materials was based on the high flow rate of the fistula, which could have led to the injection of high volume of gelatin sponge, with potential difficult detumescence of cavernous bodies or, more severely, to cavernous ischemia, inflammation, and fibrosis. We also adopted this technique because the risk of complication using coils is reported less common in young patients with unilateral left AVF, such as our case (16). Embolization can lead to potential complications such as ischemia/necrosis, inflammation, abscess formation, and erectile dysfunctions (22, 23). However, the reported rate of complications in the literature is very low, with an erectile dysfunction occurring in approximately 15% of cases (1). It has been reported that penile erectile dysfunction is mostly due to delayed embolization, which can lead to secondary development of penile fibrosis (24). In our patient, treated after 24 hours to the trauma, no complications related to the endovascular procedure were observed. Surgical treatment is rarely used but may be considered when endovascular embolization fails. Although successful surgical ligation of the arterial-lacunar fistula has been described in the literature, this intervention carries a significant risk of complications such as erectile dysfunction (reported to occur in up to 50%), penile gangrene, gluteal ischemia, purulent cavernositis, and perineal abscess (10, 14, 25, 26).

Conclusions

We presented a case of high-flow priapism successfully treated with embolization using microcoils and gelatin sponge, achieving technical and clinical success without any periprocedural complications. Endovascular embolization is safe and effective in the treatment of the high-flow non-ischemic priapism, offering a low rate of complications, short recovery time, and successful resolution of symptoms, preserving erectile function. The choice of the embolic agent should be tailored to each patient, and the procedures should be carried out by well-trained and experienced operators. In trauma-related high-flow priapism, prompt intervention is critical to minimize the risk of secondary vascular complications, and delaying treatment should be avoided to ensure optimal outcomes.

Ethic approval: The present study was conducted at our institution, in full accordance with ethical principles, including the World Medical Association Declaration of Helsinki and the additional requirements of Italian law. Furthermore, the University of Bari, Italy, classified the study as being exempt from ethical review as it carries only negligible risk and involves the use of existing data that contain only non-identifiable information about human beings. The patient signed a written informed consent form.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g., consultancies, stock ownership, equity interests, patent/licensing, arrangement etc-) that might pose a conflict of interest in connection with the submitted article.

Authors contribution: Conceptualization, N.L.; methodology, N.L., I.V., G.C.; validation, N.M., A.M., G.L.; resources, N.L., I.V.; data curation, N.L., I.V.; writing—original draft preparation, I.V., G.L., A.M.; writing—review and editing, N.L., N.M.; supervision, N.M., N.L., A.A.S.I.; project administration, N.L., N.M., I.V., A.A.S.I. All authors have read and agreed to the published version of the manuscript.

Declaration on the use of AI: None.

Consent for publication: Informed consent was obtained from the subject involved in the study.

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