American Journal of Medical Case Reports, 2020, Vol. 8, No. 8, 237-239
Available online at http://pubs.sciepub.com/ajmcr/8/8/6
Published by Science and Education Publishing
DOI:10.12691/ajmcr-8-8-6

A Rare Case of Recurrent Idiopathic Low-flow Priapism that Developed into a High-flow Priapism

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Received April 02, 2020; Revised May 04, 2020; Accepted May 11, 2020

Abstract Priapism, according to the American Urological Association is defined as a persistent penile erection that continues hours beyond sexual stimulation; typically, greater than 4 hours. Although priapism is a rare condition and has an unpredictable course in most presentation, it affects 5.36 per 100,000 male subjects per year [1]. Priapism is a urological emergency and delay in treatment or refractory cases can result in cavernous smooth muscle necrosis, fibrosis and penile shortening [2]. There are 2 categories of priapism-namely low-flow (ischemia, veno-occlusive) and high-flow (non-ischemic, arterial) [3,4]. There is a subset of ischemic priapism known as stuttering priapism which presents with recurrent incidences of ischemic priapism varying in length and is usually self-limiting [4]. Low-flow priapism occurs when an occlusive process inhibits the relaxation of the corpus cavernosum, thus the outflow of blood is impaired. The conditions associated with low-flow/ischemic priapism are as follows: sickle cell disease, vasoactive drugs, neoplastic diseases of the penis, urethra, prostate, bladder, kidney, gastrointestinal tract, leukemia, polycythemia, traumatic injury, hyperlipidemic parenteral nutrition, hemodialysis, heparin treatment, Fabry disease and neurologic conditions [3]. On the other hand, high-flow priapism occurs when there is increased arterial blood flow or pooling of blood. Conditions associated with high-flow priapism include traumatic arterio-cavernous fistula, vasoactive drugs, penile revascularization surgery, and neurologic conditions [3]. The mechanism of penile erection is a multifocal phenomenon that involves the nervous system, molecules (nitric oxide, cGMP, calcium), enzymes, and blood vessels. We present an interesting case of a patient with a history of recurrent priapism who converted from a low-flow priapism to a high-flow priapism, thought to be secondary to an arterio-cavernous fistula. Upon further review of PubMed and NIH database, there has been only few of such cases reported. We discuss the diagnostic process and management of high-flow priapism in this report.

Keywords: *priapism, low-flow, high-flow, ischemia, non-ischemic*

Cite This Article: Obed Agyei, Sakshi Joshi, Jeffrey Nalesnik MD, Arman Froush DO, and Everardo Cobos MD, "A Rare Case of Recurrent Idiopathic Low-flow Priapism that Developed into a High-flow Priapism." *American Journal of Medical Case Reports*, vol. 8, no. 8 (2020): 237-239. doi: 10.12691/ajmcr-8-8-6.

1. Case

The patient is a 42-year-old male with prior history of low-flow priapism that had been successfully managed with corporal shunt placement who presented to the Emergency Department complaining of a 36-hour history of priapism with associated pain.

On presentation, a penile block, intra-cavernous phenylephrine and terbutaline were performed with minimal response or detumescence. Next step in management of our patient was blood gas analysis of corporal vein. Bright red blood was aspirated from the corporal vein and sent for venous blood gas. Results were consistent with high-flow priapism *pH 4.3 (normal 7.37 - 7.44); PCO2 39 mmHg (normal 40-50) ; PO2 146 mmHg (normal 25-47)*. Subsequent penile blood gas also supported the diagnosis of high-flow priapism: *pH 7.43 (normal 7.37 -7.44); PCO2 30 mmHg (normal 35-48); PO2 121 (normal 83 -108*).

Given the patient's minimal response to penile block, intra-cavernous phenylephrine and terbutaline, a decision was made to perform a diagnostic angiography which revealed asymmetrically dominant left internal pudendal artery. A prophylactic unilateral Gelfoam embolization of the Left internal pudendal artery was performed. Post fluoroscopic images demonstrated improved detumescence.

It is important to note that our patient had a negative sickle cell screening and thus we ruled out priapism secondary to a vaso-occlusive phenomenon. The patient was not on any medications that could cause priapism (ie. trazodone). His CBC was within normal limits, thus ruling out polycythemia, thrombocytosis or any blood dyscrasia.

2. Images

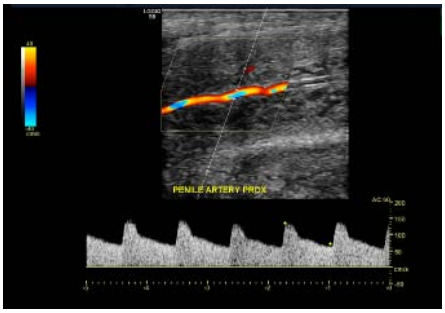

Figure 1. This figure reveals an ultrasound Duplex Flow of Penile artery that was initially performed. The velocity is 150cm/sec which indicates penile rigidity

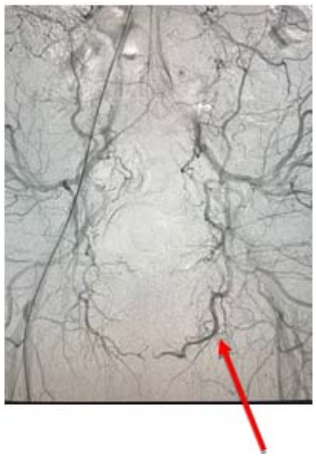

Figure 2.

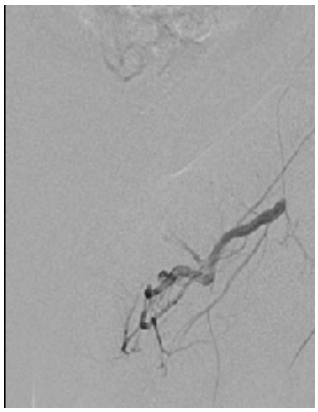

Figure 3.

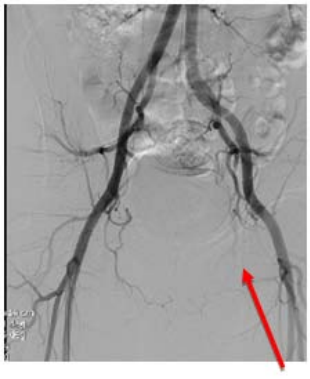

Figure 4.

Figure 2, Figure 3, Figure 4 demonstrates Transcatheter Gelfoam partial embolization of L. internal pudendal artery. In Figure 2, the arrow points to a larger and asymmetric L. internal pudendal artery prior to embolization. Figure 3 and Figure 4 shows post embolization arteriography which demonstrates decrease in flow of the L. internal pudendal artery.

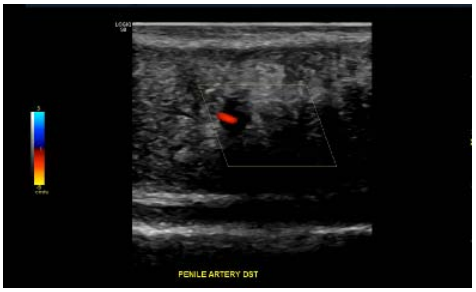

Figure 5. Ultrasound Duplex Flow of Penile artery that was performed post embolization of L. internal pudendal artery. Velocity of 40cm/sec which indicates partial resolution of tumescence

3. Discussion

There are three types of priapism: low-flow, high-flow, and stuttering. Low-flow priapism is usually ischemic and includes the subset of stuttering priapism. Low-flow priapism is a medical emergency, unlike its high-flow counterpart. If not treated in a timely manner, low-flow priapism can cause ischemic damage and can lead to permanent erectile dysfunction and infertility. On the other hand, high-flow priapism is usually associated with either penile/perineal trauma, vasoactive drugs, penile revascularization surgery, and neurologic conditions. Essentially there is a communication between the cavernous artery and the lacunar spaces of the cavernous tissue, which allows blood to bypass the protective helicine arteriolar bed that is responsible for the high physiologic vascular resistance within the penis [11].

On presentation, the proper evaluation would include differentiating between the different types of priapism. It is important to know the duration of the episode, severity of pain, previous history of priapism, history of pelvic or genital trauma, hematological disorders and medications.

One can also further evaluate via corporal vein blood gas. In low-flow priapism, one will observe dark blood, and blood gas will show low pO2 levels indicating deoxygenated venous blood. However, in high-flow priapism, the aspirate will be bright red blood with an elevated pO2 indicating oxygenated arterial blood flowing through the penis.

Phenylephrine, a pure alpha-adrenergic agonist, is considered the drug of choice for low-flow, ischemic priapism. Sympathomimetics such as phenylephrine induce contraction of the cavernous smooth muscle, and thus allow for venous outflow, which then allows for therapeutic drainage of the ischemic blood [12]. It is worth noting that a surgical shunt procedure in patients with recurrent priapism can induce an iatrogenic fistula which establishes an outflow channel from the corpora cavernosa, bypassing the pathological veno-occlusion [13]. In rare

instances such as our patient, arterio-cavernous fistula can eventually convert the low-flow into a high-flow priapism.

For high-flow priapism, if detumescence does not occur spontaneously or with an initial trial of phenylephrine, selective arterial embolization is the recommended treatment [14]. This approach has been shown to be highly effective and a well-tolerated procedure.

4. Conclusion

This case highlights the rare complication of high-flow priapism after corporal shunt placement in a with recurrent priapism. It is vital for clinicians to obtain a detailed history and perform the necessary physical exams as this may change the management of a patient's condition. More importantly, clinicians should obtain the appropriate laboratory tests such as penile arterial or venous blood gasses to different between low-flow and high-flow priapism. This case also reveals the importance of clinician collaboration with urology and interventional radiology to attain the maximum clinical outcome.

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