Superselective Arterial Embolization of Arteriovenous Malformation of Internal Pudendal Artery, a Rare Cause of Hematuria: A Case Report

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Abstract

Arteriovenous malformation (AVM) is a common form of vascular malformation, an abnormal direct communication between an artery and a vein without passing through the capillaries. AVM may just present as hematuria without plain or other symptoms. The article presents a case of a 52-year-old male with gross hematuria diagnosed as AVM of internal pudendal artery, which was successfully managed with superselective arterial embolization using temporary embolization materials.

Keywords

hematuria, arteriovenous malformation, TAE, interventional therapy

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Background

Hematuria is defined as the presence of red blood cells in the urine and is a problem commonly encountered by primary care physicians. Hematuria is the most common symptom of urinary system diseases (Bolenz et al., 2018; Davis et al., 2012), categorized as gross hematuria and microhematuria. Gross hematuria is less common than microscopic hematuria, but is more often associated with dangerous causes. Thus, the clinician should turn his or her attention to the patient with gross hematuria. Many diseases may cause hematuria, such as malignancy, benign tumor, infection, menstruation, urolithiasis, trauma, urinary anatomic disease, renal disease, chemical, and hematologic/vascular disease (Joshua et al., 2017; Peterson & Reed, 2019). The commonest cause is infection of the lower urinary tract, particularly infection of the bladder (Bin_ Dahman & Aljabry, 2019). According to a report, about 98% of hematuria is caused by diseases of the urinary system itself and only 2% caused by diseases of the whole body or adjacent organs of the urinary system (Linder et al., 2018). Malignancy is the commonest reason for gross hematuria, such as renal cancer, bladder cancer, and prostate cancer. Arteriovenous malformation (AVM) is a rare cause of gross hematuria.

AVMs are abnormal formations of blood vessels that shunt arterial blood directly into veins without passing through the capillaries. These vessels usually are crooked, dilated, and with thick vessel walls (Uller et al., 2014). The exact etiopathology of AVMs is unclear. The lack of blood flow and oxygen in the capillaries can lead to tissue damage in the affected areas. AVMs usually lead to the formation of a dilated sac-like connection, arteriovenous aneurysm, and so forth. Particularly, arteriovenous aneurysm is a fatal lesion. AVMs most commonly affect the head and neck (47.4%), followed by the extremities (28.5%). Extracranial AVMs are focal and diffuse. However an AVM that is associated with the internal pudendal artery is uncommon.
Surgical treatment is complex due to location of lesions and endovascular approach is usually the first choice among the treatment options, such as superselective vascular embolization (Jesinger et al., 2013; Papadakos et al., 2008). The article presents a case report of AVM of the internal pudendal artery, which was successfully treated by transarterial embolization with gelatin sponge and pieces of silk line.

Case Presentation

A 52-year-old male visited a doctor because of a history of repeated episodes of unexplained painless gross hematuria in the past two decades. During the past 20 years, he had visited a local hospital four times for massive hematuria, which had alleviated with conservative medical treatment. This episode was so violent that there was no time for cystoscopy. He denied any other accompanying symptoms including fever, headache, periorbital edema, joint pain, abdominal or flank pain, weight loss, skin rash, bleeding tendency, dysuria, or intermittent micturition. Hematuria was not preceded by upper respiratory tract infection or related to urinary trauma or exercises. No history of blood transfusion or other special diseases existed. Urine analysis (UA), coagulation function test, and kidney function tests (KFTs) were repeatedly normal.

Further examinations were preformed to investigate the cause of hematuria. The measurements, echogenicity, and density of kidney were normal when investigated by computed tomography (CT) urography and urinary ultrasonography (US), which excluded the hydrenephrosis, masses, or stones of the urinary tract. To exclude hematuria caused by vascular lesions, the patient underwent selective transfemoral pudendal arteriography. The arteriography was performed under local anesthesia via the right femoral artery with a 5 French sheath. Both internal iliac arteries were selectively catheterized, and there were no abnormal findings after contrast agent injection in both internal iliac arteries. However, the left pudendal artery was catheterized with a 1.7 French microcatheter and abnormal staining and extravasation of contrast agent of distal vascular branches was found, which confirmed the existence of AVM (Figure 1). Superselective embolization of the left
internal pudendal artery was performed with infusion of gelatin sponge until complete embolization of the lesion. After embolization, control angiograms showed no evidence of AVM or contrast extravasation while the perineal blood flow remained sufficient (Figure 2). Hematuria disappeared and the color of urine became clear within 6 hours.

The hematuria recurred without any accompanying symptoms 2 years later. A digital subtraction angiography was performed again, and abnormal staining and contrast agent extravasation of the both internal pudendal artery distal vascular branches were found (Figure 3). The patient chose transcatheter embolization (TAE) as he did 2 years ago. Superselective embolization of the bilateral pudendal arteries was executed with infusion of gelatin sponge plus natural silk segments. No existence of extravasation of contrast medium outside the vascular spaces was found (Figure 4). The difference from the first time was the use of natural silk segments and the slow speed of embolization. After long-term follow-up of 10 years, no recurrence and no postoperative complications such as erectile dysfunction and ischemic necrosis were found.

**Discussion**

Hematuria is often a common symptom of urinary system disease. A good medical history, physical examination, and focused laboratory investigations for persistent or

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**Figure 3.** Imaging of digital subtraction angiography (DSA) demonstrates the existence of arteriovenous malformation (AVM). AVM lesion is the white arrow shown in both A and B. A: the right; B: the left.

**Figure 4.** Imaging of superselective embolization of both internal pudendal arteries. Arteriovenous malformation (AVM) lesion disappeared without extravasation of contrast medium outside the vascular spaces. A: the right; B: the left.
The lesions of AVM are often diffuse, involving multiple tissue planes and important structures, and the possibility of cure is rare (Richter et al., 2007). The goal of treatment usually is to control the malformation. Intervention focuses on alleviating symptoms (e.g., bleeding, pain, ulceration), preserving vital functions (e.g., vision, mastication), and improving a deformity. The management includes embolization, resection, or a combination of the two. Resection may provide the best chance for long-term control, but the re-expansion rate is high and extirpation may cause a worse deformity. Embolization is not a curative method with the lesion’s final re-expansion after embolization. Embolization is the most commonly method used preoperatively to reduce blood loss during resection or for a palliation of unresectable lesions (Greene & Orbach, 2011; Liu et al., 2010; Zaki Ghali et al., 2019).

In the present case, the treatment for AVM is embolization. Superselective arterial embolization of the internal pudendal artery is an accurate diagnostic tool as well as an elective and safe treatment for the control of bleeding. Embolic materials available for embolization include coils, ethanol, balloons, gelfoam particles and powder, and autologous clots (Po et al., 2012). Absorbable gel-fоam particles, compared with permanent materials such as polyvinyl alcohol particles and trisacryl gelatin microspheres, are preferred because they are temporary embolic materials and without many complications. The complications of embolization for pelvic arteries include infection, necrosis of muscle or penis, nerve injury, bladder or ureteral infarction, bleeding or hemorrhage, bowel infarction, thigh or buttock claudication, and impotence (Vaidya et al., 2018). In this case, gelatin sponge and natural silk segments were used in order to preserve the normal erectile function of the patient. A very important reason for recurrence without necrosis of penis was the use of gelatin sponge the first time, which is the most common temporary embolization material. Thus, the second time the doctors chose gelatin sponge plus natural silk segments as the embolic material and infused slowly. The aim was
to give sufficient time for the formation of collateral circulation. It was the most important reason that no necrosis of penis occurred in 10 years after the treatment. In conclusion, an AVM of the internal pudendal artery is a rare cause of recurrent painless gross hematuria. In clinical work, if common causes of unexplained hematuria occur, such as infection, trauma, or stone, they deserve high attention and a high index of suspicion is necessary for abnormal vascular issues, especially AVM. Imaging detections such as Doppler US, CTA, and angiography should be performed in such patients to reach an accurate diagnosis. It is important to protect functions of penis, while choosing the treatment of AVM of the internal pudendal artery. The selection of embolization materials and the process of operation also should be carefully handled.

**Declaration of Conflicting Interests**

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**Ethical statement**

Ethics approval was not required for this study.

**Patient consent**

The patient of this case study consented to the publication of his information.

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