

## Eruption of blood: Arteriovenous malformation of the penile urethra

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### Abstract

While arteriovenous malformations (AVMs) are a common congenital or post-traumatic abnormality, male genital AVMs are rare and have been described in the scrotum or penis in pediatric patients.<sup>1,2</sup> We describe a 34-year-old male presenting with recurrent spontaneous penile urethral bleeding found to have an AVM of the penile urethra. While angiography has traditionally been helpful, magnetic resonance imaging (MRI) can aid in the diagnosis and characterization of these lesions.<sup>3</sup> Each case of male genital AVM provides a unique challenge to manage depending on the presenting complaint, as there are no guidelines to direct treatment.<sup>4</sup>

### Case report

A 34-year-old male presented with a history of blood erupting from the urethral meatus during erections. These episodes were quite painful, disturbing, and he noted a significant curvature with an erection (looking similar to Peyronie's disease). There was a history of a previous penile injury from a motorcycle accident. Physical exam showed a circumcised penis with a meatus that looks grossly normal. There was a palpable mass distally along the right ventral penile shaft similar to a Peyronie's plaque. This plaque was non-tender, located proximal to the glans corona, and more associated with the penile urethra than the corporal bodies. Flexible cystoscopy identified venous lakes running visibly within the penile urethra. These lakes would intermittently and spontaneously rupture during sleep, leaving the patient in a pool of blood that he treated by penile wrapping and compression. This caused him significant psychosocial issues and he had avoided sexual activity for years. Initially, transurethral attempts with cauterization were tried at a community hospital, but proved unsuccessful. The patient was then referred to our centre for further evaluation.

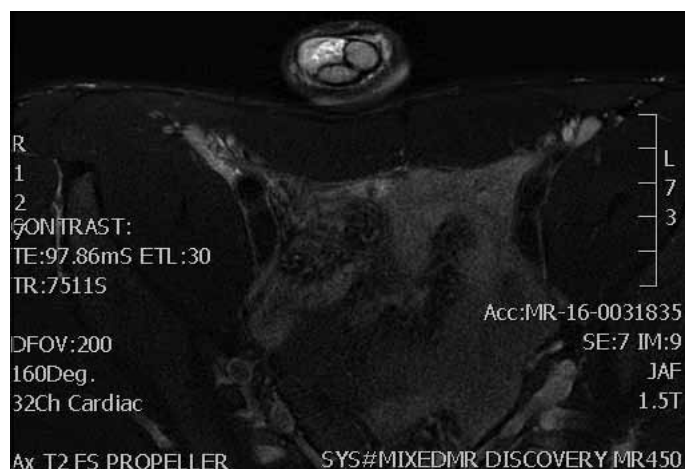
Although angiography with embolization was discussed, we were concerned about risks of erectile dysfunction and urethral structuring. After discussion with radiology, a mag-

netic resonance imaging (MRI) was ordered to characterize the abnormality and T2-weighted MRI findings characterized the area as a "serpiginous tangle of vessels in the corpus spongiosum" within the distal penile urethra (Fig. 1). The lesion was separate from the corpus cavernosum. MRI identified multiple small flow voids (patent vasculature), though it did not demonstrate direct communication between the lesion and the urethra (Fig. 2). Surgical treatment was offered in the form of exploration with consent including resection of the affected penile urethra, buccal mucosal grafting, and first-stage penile urethral reconstruction.

A degloving circumcision was performed, followed by a ventral urethrotomy on the penile urethra after attempts to free the penile urethra from the corporal bodies failed due to scarring and tethering of the penile urethral abnormality to the underlying corporal bodies. Upon urethrotomy, the arteriovenous malformation (AVM) ran for 3 cm and replaced a large portion of the penile urethra, leaving a small strip of normal urethra mucosa on the left lateral side (Fig. 3). The decision was made to resect the diseased penile urethra containing the AVM with proximal and distal closure of the spongiosum to ensure the vascular abnormality was halted. Given the large defect, buccal grafting in a staged fashion incorporating the remaining urethral plate was required. Proximally, a temporary, mid-shaft ventral neo-meatus was created. Distally, the meatus and urethra remained intact before opening into the staged area. Dressings were applied and a catheter was inserted for five days to allow maturation of the neo-meatus. At the two-month followup, the grafted area took well and no further penile bleeding was reported. Additionally, the prior curvature during erection was gone and the patient awaits a second stage urethral closure.

### Discussion

AVMs are the result of fast-flow vascular abnormality where in the normal capillary network is missing; the feeding arteries drain directly into the draining veins.<sup>5</sup> AVMs are most commonly found in the head and neck, though they may occur in any organ system.<sup>6</sup> AVMs of the male genital tract are extremely rare and the literature is largely limited to case



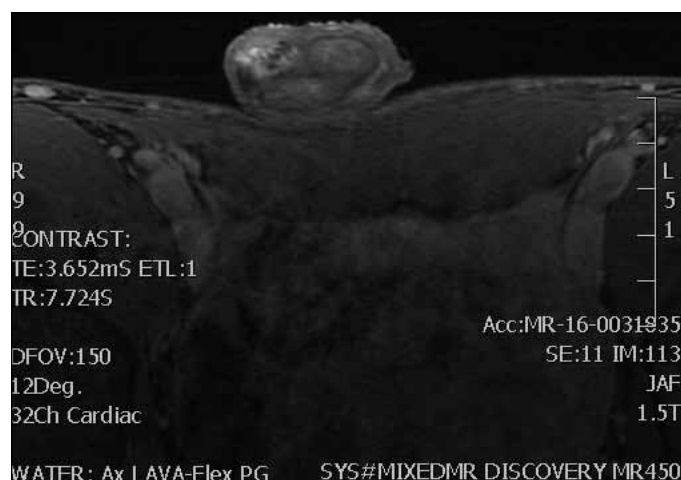
**Fig. 1.** T2-weighted images demonstrating a serpiginous tangle of vessels present in the posterior aspect of the corpus spongiosum. Left and right corpus cavernosa are unremarkable.

reports. Penile and scrotal case reports have been identified, though a literature search showed no cases of penile urethral AVM or corpus spongiosum AVM,<sup>1,7,8</sup> making this the first reported case.

AVMs may be congenital or traumatic in etiology and are often progressive in nature. Clinical manifestation is most often in the second or third decade of life.<sup>5</sup> A characteristic finding is profuse, pulsatile bleeding.<sup>5</sup> Imaging is essential in diagnosing AVMs. Doppler ultrasonography is effective in identifying flow patterns within the shunt,<sup>5</sup> but MRI provides incredible soft-tissue contrast and is extremely helpful in identifying the vascular anatomy of the penis.<sup>9</sup> Computed tomography (CT) is not the best choice if the AVM is thought to be within soft tissue.<sup>5</sup> Catheter arteriography is the gold standard, however, MRI angiography provides comparable information.<sup>10</sup>

Treatment is reserved for symptomatic lesions, and there is a wide variety of treatment options.<sup>11</sup> Embolization is a minimally invasive technique and is the first-line option in the treatment of AVMs.<sup>5</sup> Angioembolization may also be offered before resection to help shrink the size of the AVM. Other treatment options include: surgical excision, cryotherapy, YAG laser treatment, and sclerotherapy.<sup>4</sup> The lesion of our patient was located adjacent to the penile urethra and within the corpus spongiosum. Given the location, the risks of angioembolization, including erectile dysfunction and urethral strictures, were too great to offer this as a treatment option. Surgical excision is a reasonable consideration for voluminous formations<sup>8</sup> so after discussing the risks and potential benefits of surgery, excision was selected by the patient.

The first report of penile AVM being successfully treated by surgical excision dates back to 2000, when Go et al described a case of congenital penile AVM in an infant wherein venous dilatation of the prepuce was documented



**Fig. 2.** Multiple small flow voids enhancing post-contrast, most in keeping with an arteriovenous malformation.

at one year of age.<sup>2</sup> Surgical excision yielded positive results with no recurrence at six-month followup.<sup>2</sup> In our patient, the AVM was traumatic in origin, small at 3 cm, and sharply localized with a dramatic presentation. As such, potentially curative surgical excision was offered.

The rarity of urogenital AVMs necessitates a case-by-case approach to treatment. In our patient, the unique combination of the location of the AVM within the corpus spongiosum, spontaneous bleeding with erections, and curvature of the penis at the site of the malformation have not been described in the literature. Given the close proximity of the AVM within the corpus spongiosum to the urethra, excision of the penile urethra was necessary, resulting in a significant portion of the urethra removed. This mandated buccal grafting to replace the penile urethra excised with a staged surgical approach.



**Fig. 3.** Urethrotomy exposing 3 cm arteriovenous malformation adjacent to penile urethra

**Competing interests:** The authors report no competing personal or financial interests.

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