Urethral venous malformation: an unusual cause of recurrent post-coital gross haematuria in association with haemospermia

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ABSTRACT

Three cases of recurrent post-coital haematuria are described. Extensive protracted investigations pinpointed urethral varicosities as the likely cause. All patients were successfully treated with diathermy fulguration.

KEYWORDS

Urethral – Varicose – Venous – Malformation – Haemangioma – Haemospermia – Haematuria

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Bleeding per urethram is a common urological symptom which prompts urgent investigation, aimed primarily at excluding malignancy. The differential diagnosis in these patients is considerable. We present three cases of urethral bleeding associated with post-coital ejaculation that was found to be caused by urethral venous malformations, a condition that is rare but probably under-reported.

Patients and Methods

The case notes of three patients who were identified as suffering from post-coital urethral bleeding were reviewed. Due to the rarity of the disease and the lack of standardisation in the diagnosis and treatment, personal communication with each surgeon was necessary for a more accurate description of the intra-operative findings and details of the surgical treatment. A search and review of the literature published in English was performed using the PubMed database.

Results

The three patients in this series were treated by three different urological surgeons, all practising in the same unit. Cases are reported both separately and in comparison (Table 1).

Case report 1

A 58-year-old man represented to the department of urology in 2007 with a history of haemospermia and haematuria spanning 20 years. The patient reported that episodes

of haemospermia and haematuria were provoked by ejaculation. He was first investigated for frank haematuria 18 years previously when it was reported that there was no abnormality of the urinary tract. He was re-investigated for haematuria 10 years later when large 'varicose veins' were noted in the bulbar and membranous urethra, coursing over the external sphincter muscle. Three years previously, he was again referred complaining of haematuria. On this occasion, a flexible cystoscopy was reported as normal. Over the next 2 years, the patient was admitted to hospital on three occasions with further episodes of heavy haematuria and the passage of clots. Investigations including an intravenous urogram, renal/pelvic ultrasound scan and magnetic resonance imaging (MRI) of the pelvis were reported as normal. His most recent cystourethroscopy, however, demonstrated large, abnormal branching veins in the bulbar urethra (Fig. 1) on either side of the verumontanum and around the bladder neck. In addition, within the bladder there were abnormal sub-mucosal veins high on the posterior bladder wall. Judicious 'spot-welding' of these veins at the bladder neck, alongside the verumontanum and in the bulbar urethra was performed using diathermy coagulation. At 6-month review, the patient was free of post-coital haemospermia and haematuria for the first time in 20 years.

Case report 2

A 41-year-old man was referred with a 10-year history of intermittent haematuria with clots. This had always been associated with sexual activity and ejaculation. There was

no significant past medical history and no abnormalities on clinical examination. The patient denied any perineal discomfort or lower urinary tract symptoms suggestive of chronic inflammatory prostatitis. The patient underwent a cystourethroscopy demonstrating congested veins crossing the external sphincter. The abnormal vessels were diathermied and the patient, to date, has remained continent and symptom-free.

Case report 3

A 43-year-old man presented with a 5-year history of post-coital urethral bleeding. He had previously been investigated by cystourethroscopy, pelvic and renal ultrasound scans and a pelvic MRI scan, that failed to reveal the cause of bleeding. The patient had a repeat cystourethroscopy to exclude urethral venous malformations, as these were suspected to be the cause of bleeding. For this reason, the patient was instructed to ejaculate in the theatre prior to the investigation and moderate reverse Trendelenburg position

Figure 1 Operative sketch of venous malformation in the bulbar urethra at the level of the external sphincter.

was employed; the rationale being that both these manoeuvres would exaggerate any vascular abnormality and render it more visible and amenable to treatment. At endoscopy, enlarged veins were noted just distal to the verumontanum which were carefully diathermied. At 6-month review, the patient had no further post-coital urethral bleeding.

Discussion

Post-ejaculatory haematuria in association with haemospermia is rare.^{1,2} Urethral varices as a cause of this condition were reported in 4.6 % of 174 men investigated for haemospermia by Leary and Aguilo.³ More recently, Papp *et al.*⁴ described urethral varices as being the underlying cause in 4–7% of 122 patients being investigated for haemospermia. In earlier case reports,^{1,2} the venous abnormalities were fulgurated successfully with no adverse effect on continence. Arnold *et al.*⁵ treated urethral varices in 20 women and 5 men with a combination of coagulation and urethral dilatation and reported a success rate of 86%. Others have published case reports of massive urethral bleeding from urethral varicosities associated with congenital diseases or major venous malformations necessitating major surgical procedures.^{6,7}

Other rare causes of urethral bleeding include urethral polyps,⁸ ectopic prostatic tissue⁹ and urethral haemangiomas.^{10,11} Haemangiomas appear to be a separate entity and are described as sessile vascular lesions usually near the prostatic apex.^{10,11}

The condition we describe is uncommonly reported in the literature and, in addition, is not mentioned in the classical textbooks as a distinct urological condition. One such case could represent a medical curiosity and two, an extreme coincidence. However, the fact that three different surgeons treated patients with the same condition in the same unit within a short period of only a few months indicates that it is probably an under-reported condition of unknown incidence.

Table 1 Summary of patients' symptoms and treatment outcome

Patient 1

History	Hemospermia, severe hematuria post-ejaculation
Investigations	Multiple cystourethroscopies, intravenous urogram, renal/pelvic ultrasonography, pelvic MRI
Treatment	Electrocoagulation of abnormal veins
Outcome	Symptom-free at 6-month review, continent

Time to diagnosis and treatment 20 years

Patient 2

History	Clot hematuria post-ejaculation
Investigations	Cystourethroscopy
Treatment	Electrocoagulation of abnormal veins
Outcome	Symptom-free at 6-month review, continent

Time to diagnosis and treatment 10 years

Patient 3

History	Post-coital urethral bleeding
Investigations	Multiple cystourethroscopies, renal/pelvic ultrasonography, pelvic MRI
Treatment	Electrocoagulation of abnormal veins
Outcome	Symptom-free at 6-month review, continent

Time to diagnosis and treatment 5 years

Increased awareness of this cause of urethral bleeding is essential since the identification of urethral venous malformations may be hindered by the fact that they are not always easily visualised.^{1,2,5} The vascular pattern in the urethra and the prostate is subject to some variation and venous malformations may be overlooked if the operator has a low index of suspicion. In our first case, a number of cystourethroscopies were reported as showing no obvious abnormality and the patient endured the symptoms for almost two decades until definite treatment was instituted. In the second patient, the diagnosis was made 10 years following the onset of symptoms. Similarly, the third patient in our series had an initial endoscopic assessment that failed to identify the vascular lesion and there was a lag time of 5 years between the onset of symptoms and diagnosis and treatment. The present report emphasises that varices as a cause of urethral bleeding must be considered when common pathology has been excluded.

All three cases were treated by electrocoagulation; because the abnormal vessels were very close to the external urethral sphincter, actually crossing it in the first two patients, there were concerns about continence post-operatively. Extreme care was taken when fulgurating the lesions and, as a result, all patients remained fully continent. For such patients, laser therapy may be a better option for treating these lesions¹² as the highly defined area in which energy is applied and the precision of tissue destruction of laser therapy may have advantages. Many different types of lasers are established in urological practise and more are being introduced.¹³ With a small depth of penetration and a wavelength very near the absorption peak of water, Holmium, Erbium and Thulium lasers are probably the most suitable for the treatment of urethral varices, offering increased precision and safety, though at the cost of haemostasis.

Conclusions

Urethral varicosities are an unusual cause of recurrent post-coital urethral bleeding. In the three cases we present, the diagnosis was established 5–20 years after the onset of symptoms, a fact that indicates the elusive nature of the disease and the need for increased awareness by the urologic community. Careful coagulation of the engorged veins was a safe and effective treatment in all cases.

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