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Inflammation and infection

Female urethral stricture from chronic graft-versus-host disease

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ABSTRACT

Female urethral strictures are rare with limited literature to guide management. We present a case of a 27 year old female diagnosed with a urethral stricture secondary to chronic graft-versus-host disease. This diagnosis should be considered in females presenting with obstructive voiding symptoms and have risk factors for stricture development.

Introduction

Female urethral strictures are uncommon but can have significant functional impact and cause lower urinary tract symptoms (LUTS). It is estimated 10% of women with obstructive voiding symptoms will have a true anatomical urethral stricture.[1] Patients tend to present with impaired flow, urgency, frequency and dysuria, as well as recurrent urinary tract infections (UTIs) or urinary retention.[2]

Common aetiologies include trauma, iatrogenic injury and radiation.[2] Along with history and examination, cystoscopy and voiding urethrography can aid diagnosis (Fig. 1).[3] Standard management has been urethral dilatation, with success rates in excess of 50%, however previous dilatation is predictor of failure.[2] Reconstructive methods with genital skin flaps or oral mucosal grafts can have favourable long term outcomes.[1]

Chronic graft-versus-host disease (cGVHD) is a systemic immune disorder where grafted donor cells react against histocompatibility host antigens.[4] It is primarily T-cell-mediated and the most common late complication of allogeneic hematopoietic stem cell transplantation.[4] It can involve all organ systems, causing a variety of clinical manifestations. Skin, oral mucosa, liver and gut are well-documented sites of involvement, however genital cGVHD is rare and often undiagnosed. It can present with dryness, dysuria and dyspareunia.[5] Genital cGVHD is known to cause vaginal stenosis[5] however there is no mention in literature of it leading to female urethral strictures.

Case presentation

A 27-year-old female was referred to urology outpatient clinic with LUTS and recurrent UTIs. Her main complaints were poor flow and a sensation of incomplete emptying. Renal tract ultrasound showed a post-void residual volume of 164ml but otherwise unremarkable bladder and upper tracts (Fig. 2). She had documented *Escherichia coli* UTIs that had been appropriately treated with antibiotics.

The patient's medical history was significant for Hodgkin lymphoma,

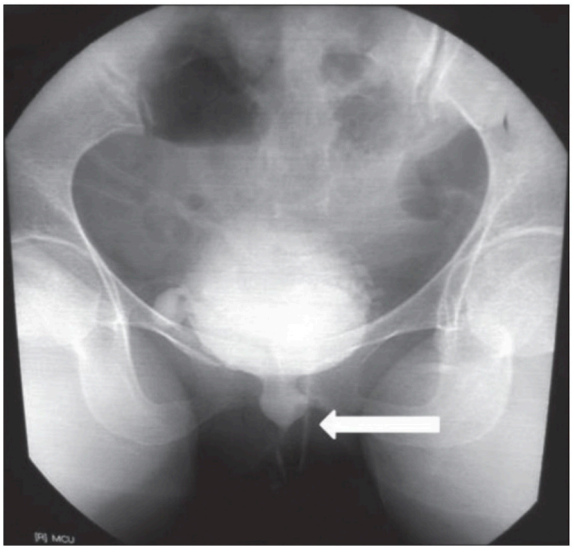

Fig. 1. Micturating cystourethrogram showing a distal urethral stricture with proximal urethra dilatation (white arrow).[3]

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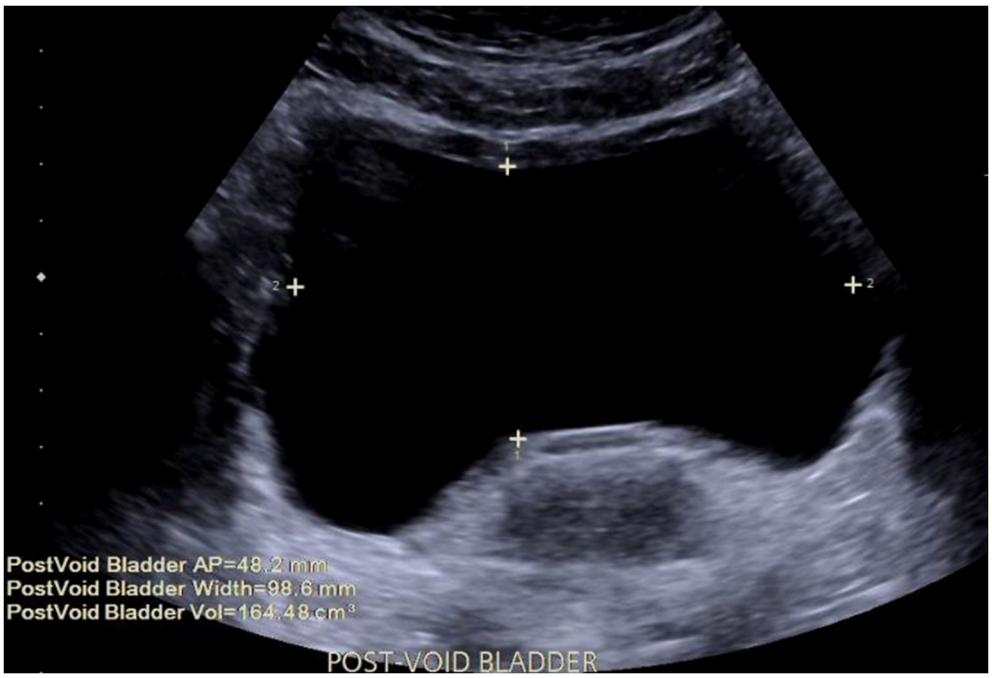

Fig. 2. USS showing post-void bladder volume of 164ml.

treated with chemotherapy followed by relapse requiring unrelated peripheral blood stem cell transplantation at age 16. The patient subsequently developed cGVHD primarily affecting her oral mucosa, skin and vagina, which was managed with tacrolimus as per haematology as well as vaginal self-dilatation as per gynaecology.

A recent examination in gynaecology clinic showed a normal vulva, vagina and cervix, but on review in urology clinic, a pin-hole urethral meatus consistent with distal urethral stricture was noted. The patient's urethra was serially dilated using rigid sounds, followed by flexible cystoscopy, showing a distal urethral stricture but normal proximal urethra and bladder.

During a phone review 6 weeks later, the patient reported her symptoms had resolved with much improved flow and a resolution of her sensation of incomplete emptying. She has had no further urinary tract infections and will have serial follow-up to ensure a sustained result.

Discussion

Female urethral stricture disease is an uncommon entity and its incidence is likely underestimated. It should be considered when females present with symptoms of BOO and have risk factors for development of strictures. It is important that the condition is detected and managed appropriately to not only improve a patient's symptoms and quality of life, but to prevent recurrent UTIs and the development of urinary retention and obstructive uropathy.

Female genital cGVHD is a recognised clinical entity, however its association with urethral pathology is not documented in literature. Male urethral strictures are documented in cGVHD[5] and we believe this is the cause of this patient's stricture given the chronic inflammatory nature of cGVHD and the absence of other risk factors for stricture formation. Clinicians should be mindful of the possibility of BOO when these patients present with LUTS or radiological signs suggestive of retention.

Options for treatment are both medical and surgical. Immunosuppression is the mainstay of reducing the risk of further anatomical deterioration. Surgical management of female urethral strictures has traditionally been urethral dilatation, however more complex reconstructive methods have been developed and may have more favourable long-term outcomes. The role of these methods in a chronic condition such as cGVHD is yet to be ascertained and warrants further investigation as to their efficacy.

Conclusion

Urethral stricture disease is an uncommon cause of LUTS in females but can have detrimental impacts on quality of life. It is an important diagnostic consideration for haematologists, gynaecologists and urologists in patients with genital cGVHD. A thorough history, and pelvic and urethroscopic examination are essential.

Consent

Verbal and written consent was obtained from the patient involved in this case for specific details of her case to be documented and published.

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Declaration of competing interest

There are no conflicts of interest to declare.

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