CASE REPORT

Acquired male urethral diverticulum: a rare entity treated in a one-stage procedure

José Carlos Santos,¹ Nidia Rolim,² Renato Lains Mota,² Hélder Monteiro³

¹Department of Urology, Centro Hospitalar Lisboa Ocidental, Lisboa, Portugal

²Department of Urology, Centro Hospitalar de Lisboa Ocidental, EPE, Lisbon, Portugal

³Centro Hospitalar de Lisboa Ocidental, Lisboa, Portugal

Correspondence to

Dr José Carlos Santos, jose.c.pereirasantos@gmail.com

Accepted 12 November 2016

SUMMARY

Acquired male urethral diverticulum is a rare entity with most of the literature revolving around case reports or small case series. Up to two-thirds of cases are acquired, mostly as a result of trauma, stricture or infection. Infrequently, some cases develop as a complication of urological procedures, or even penile clamping. We present the case of an adult male with lower urinary tract symptoms, recurrent urinary infections and a history of multiple surgeries to treat a complicated perineal fistulae disease. With the help of imaging techniques, a bulbar urethral diverticulum was discovered. Owing to the symptomatic nature of the diverticulum, an open procedure was performed with excision and primary urethral anastomosis. No urinary symptoms were reported and follow-up imaging and flowmetry demonstrated very good functional outcome.

During these procedures, there were no reports on lesions of the urethra, bladder or intestine. The decision to perform an intestinal diversion was obvious after the third attempt without success. The last MRI performed showed no new fistulae tracts but there were some small fibrosis in relation with the previous documented tracts ([figure 1](#)).

He is HIV positive under medication (triple highly active antiretroviral therapy regimen) with good serological and clinical control. No other clinically significant data were reported.

On physical examination, a normally implanted urethral meatus was observed and no penoscrotal mass was visible. A fully functioning colostomy stoma was located on the left lower abdominal quadrant. No other pathological changes were registered.

BACKGROUND

The clinical scenario and the rare presentation with perineal fistulae disease make this an original and interesting case. At the same time, it is an ideal opportunity to review possible diagnosis when evaluating lower urinary tract symptoms (LUTS) in male patients.

Owing to the low incidence of urethral diverticula in men every opportunity to document and share the surgical findings should be taken.

Overall, we find that this case is a wonderful learning opportunity and that it will raise a very interesting debate.

CASE PRESENTATION

We present the case of a 54-year-old man who was referred to our urology department due to penile mass tumescence during voiding and multiple urinary tract infections (UTIs). He mentioned the need to manually compress the mass in order to complete voiding. The symptoms had gradually increased in severity over the past 12 months. At the same time, multiple UTIs were diagnosed with isolates of *Escherichia coli* and *Klebsiella pneumoniae* in different urine cultures.

The patient also had a history of a complicated complex posterior perineal fistula which required multiple surgeries in the general surgery department, and was only solved after intestinal diversion with colostomy. From the surgical reports, a complex perineal fistulae with intersphincteric and trans-sphincteric components was initially treated with fistulotomy through a transanal approach. Two more surgical interventions were performed through the same approach without success.

INVESTIGATIONS

Both urine and blood samples were collected for culture with neither showing signs of active infection.

A free flowmetry showed a peak flow of 18 mL/s. The curve represented a prolonged intermittent flow time that could be explained by the need to manually compress the diverticulum in order to drain the residual urine. A cystourethroscopy revealed a small millimetric ostium in the bulbar urethra without any signs of urethral stricture.

Retrograde urethrography uncovered a bulbar diverticulum with 4 cm maximum length ([figure 2](#)) without any signs of strictures or urointestinal fistulae. A pelvic MRI showed a urethral saccular dilation suggestive of a diverticulum without new fistulae tracts or abscess.

DIFFERENTIAL DIAGNOSIS

The symptoms reported by the patient are highly suspicious of urethral pathology. Urethral stricture, fistulae and diverticulum were all possible diagnosis. Multiple UTIs in the adult males can raise the suspicion of bladder outlet obstruction.

A history of perineal fistulae should raise the awareness of a possible connection between the urinary and intestinal tracts. No symptoms of fecaluria or pneumaturia were recorded, making this hypothesis less likely. The history of intestinal surgeries for posterior perineal fistulae, plus the need to perform a temporary colostomy optimised the infectious events related to the urinary tract. MRI was performed in order to evaluate the perineal anatomy after the intestinal diversion and the clinical status of the perineal disease.

CrossMark

To cite: Santos JC, Rolim N, Mota RL, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-216670

Figure 1 Pelvic T2 MR showing one of the perineal fistulae tracts at initial presentation (small arrow).

Figure 2 Initial retrograde urethrogram showing a bulbar diverticulum without any signs of stricture.

The lack of other LUTS typically related to a urethral stenosis, including weak and thin urinary flow and, longer flow time also made this hypothesis less likely. Both the ureteroscopy and urethrography failed to show any narrowing of the urethral lumen.

A contrast-enhanced examination was recommended in order to analyse the lower urinary tract anatomy. In this case, a urethral bulbar diverticulum was clearly seen, without any leaks or fistulae.

TREATMENT

Owing to multiple UTIs and the symptomatic nature of the diverticulum, a surgical approach was recommended. A urine culture was performed prior to the surgical procedure which showed no signs of active infection.

A perineal approach was chosen through an inverted U incision in the posterior perineum in extreme lithotomy position (figure 3). Dissection and isolation of the bulbar urethra was carefully performed due to fibrosis involving the diverticulum (figure 4). In order to facilitate the procedure, methylene blue was instilled through the urethra in order to colour the urethral

Figure 3 Perineal incision with the inverted U incision in extreme lithotomy position.

Figure 4 Perineal dissection showing the bulbar urethra already dissected (thick arrow).

Figure 5 Diverticulum excision showing the diverticulum mucosa (thick arrow). The distal urethral top can clearly be seen (narrow arrow).

mucosa. Primary excision of the full diverticulum and an end-to-end urethral reconstruction were preformed (figure 5).

No postoperative events were recorded. A urinary catheter was left in place for 3 weeks with a full range antibiotics administered during 10 days.

OUTCOME AND FOLLOW-UP

No additional urinary symptoms or UTIs were reported, and 2 months after surgery a retrograde urethrography showed anatomical adequate reconstruction of the urethra (figure 6). A free urinary flow showed a normal trace with a free Qmax of 19 mL/s.

DISCUSSION

Although male urethral diverticulum is a rare entity, it should not be overlooked when assessing male urinary symptoms and cases of multiple UTIs. Even though the congenital cases are more common, one should remember that any urethral injury, infection surgery or even penile clamping could lead to a saccular dilation of the urethra.^{1 2} A possible mechanism for posterior urethral diverticulum development is after anorectal malformations treatment, with an estimated incidence of 12–18%.³ No case reports of male anterior urethral diverticulum related to perineal/anorectal pathology were found in the literature.

Although penile mass tumescence was the main symptom in this case report, the literature shows a different pattern of presentation. Cinman *et al*⁴ have the biggest case series with 22 cases of successfully treated male urethral diverticula in the past 10 years. Only 23% reported of penoscrotal mass and 32% of recurrent UTIs. Most of the cases were localised in the bulbar urethra. Although no case had a history of perineal fistulae, half

Figure 6 Postoperative retrograde urethrogram showing a clearly defined urethra without any strictures or diverticulum.

the cases had previously demonstrated stricture or trauma. Alphs *et al*⁵ showed only 2 out of 13 patients with penoscrotal swelling in their case series. Multiple UTIs were the most common symptom.

In women, MRI is the best imaging modality for the diagnosis and surgical planning for urethral diverticulum.⁶ In male patients, contrast-enhanced fluoroscopy could be sufficient to confirm the diagnosis,¹ although other imaging options should be considered when evaluating certain cases when disease-related anatomic changes are suspected.

The best surgical treatment should be individually based. In this case, due to the urinary symptoms and the multiple UTIs, a surgical solution was decided. Primary excision and full urethral permeability maintenance were the core objectives.

Cinman *et al* reported six cases of success with primary excision and end-to-end urethral anastomosis, with very good outcomes. The decision regarding the type of reconstruction was based on the size and location of the urethral gap after the diverticulum excision.

Alphs *et al* reported a series of 13 cases that were successfully treated with surgery. Six cases were treated with end-to-end anastomosis after surgical excision of the diverticulum, due to a small urethral defect (<4 cm).

Learning points

- ▶ Although a rare entity, one should not overlook male urethral diverticulum as a possible aetiology for multiple urinary tract infections or penile mass swelling.
- ▶ A simple contrast examination confirms the diagnosis and a one stage surgical procedure is possible with good results.

Contributors JCS wrote the main text, the other authors reviewed and helped in the final selection of the pictures available. The overall and scientific qualities were guaranteed by RLM and HM (guarantors).

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Peralta J, Reis M, Rabaça C, *et al.* Acquired male urethral diverticulum: a complication following artificial urethral sphincter implantation. *BMJ Case Rep* 2013;2013:pii: bcr2013201542.
- 2 Kalra S, Srinivas P, Manikandan R, *et al.* Urethral diverticulum: a potential hazard of penile clam application for male urinary incontinence. *BMJ Case Rep* 2015;2015:pii: bcr2015209957.
- 3 Alam S, Lawal TA, Peña A, *et al.* Acquired posterior urethral diverticulum following surgery for anorectal malformation. *J Pediatric Surg* 2011;46:1231–5.
- 4 Cinman N, Mcaninch JW, Glass AS, *et al.* Acquired male urethral diverticula: presentation, diagnosis and management. *J Urol* 2012;188:1204–8.
- 5 Alphs HH, Meeks JJ, Casey JT, *et al.* Surgical reconstruction of the male urethral diverticulum. *Urology* 2010;76:471–5.
- 6 Ockrim JL, Allen DJ, Shah PJ, *et al.* A tertiary experience of urethral diverticulectomy: diagnosis, imaging and surgical outcomes. *BJU Int* 2009;103:1550–4.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow