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INVITED ORIGINAL ARTICLE

Male Health

Syringoceles of Cowper's ducts and glands in adult men

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Cowper's syringoceles are uncommon, usually described in children and most commonly limited to the ducts. We describe more complex variants in an adult population affecting with varying degrees of severity, the glands themselves, and the complications they may lead to. One hundred consecutive urethrograms of patients with unreconstructed strictures were reviewed. Twenty-six patients (mean age: 41.1 years) with Cowper's syringoceles who were managed between 2009 and 2016 were subsequently evaluated. Presentation, radiological appearance, treatment (when indicated), and outcomes were assessed. Of 100 urethrograms in patients with strictures, 33.0% demonstrated filling of Cowper's ducts or glands, occurring predominantly in patients with bulbar strictures. Only 1 of 26 patients with non-bulbar strictures had a visible duct/gland. Of 26 symptomatic patients, 15 presented with poor flow. In four patients, a grossly dilated Cowper's duct obstructed the urethra. In the remaining 11 patients, a bulbar stricture caused the symptoms and the syringocele was identified incidentally. Eight patients presented with perineal pain. In six of them, fluoroscopy and magnetic resonance imaging (MRI) revealed complex multicystic lesions within the bulbourethral glands. Four patients developed perineoscrotal abscesses. In the 11 patients with strictures, the syringocele was no longer visible after urethroplasty. In three of four patients with urethral obstruction secondary to a dilated Cowper's duct, this resolved after transperineal excision ($n = 2$) and endoscopic deroofing ($n = 1$). Five of six patients with complex syringoceles involving Cowper's glands were excised surgically with symptomatic relief in all. In conclusion, Cowper's syringocele in adults is more common than previously thought and may cause lower urinary tract symptoms or be associated with serious complications which usually require surgical treatment.

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Keywords: complications; Cowper's glands; syringocele; treatment; urethral stricture

INTRODUCTION

Cowper's glands, also known as bulbourethral glands, are paired exocrine structures situated within the urogenital diaphragm. They are named after the 18th century London surgeon/anatomist William Cowper; however, their discovery is credited to the French surgeon Jean Mery in 1699.¹ Their ducts drain into the ventral bulbar urethra distal to the membranous urethra. They are subject to a number of congenital and acquired pathological processes.

Cowper's syringoceles are uncommon and usually described in the pediatric population. They are defined as cystic dilatation of Cowper's ducts, and radiological and clinical classifications are based on the degree of duct dilatation and whether they communicate or not with the urethra.^{2–4}

In our practice, we have become aware of a number of adult patients diagnosed with Cowper's syringoceles which are not only limited to the ducts but also involve, with varying degrees of severity, the glands themselves. The purpose of this article is to highlight this uncommon, but possibly unappreciated pathology, with specific emphasis on these more complex variants having the potential for serious complications. A modification of the current classifications is proposed in order to take these into account.

PATIENTS AND METHODS

The urethrograms of 100 consecutive patients with urethral strictures in a single tertiary Reconstructive Urology Unit at University College

London Hospitals (UCLH), London, UK, were reviewed by a single consultant urologist. These were patients who may or may not have had endoscopic intervention for their stricture but not urethroplasty. This study did not require research ethics committee approval because it was a retrospective review which did not alter the management of the patients in any way. It was carried out in accordance with the principles laid down in the Declaration of Helsinki. All study participants gave verbal informed consent. Subsequently, our prospectively compiled outcome database was searched retrospectively to identify patients who were treated for Cowper's syringocele between January 2009 and December 2016 or in whom this was identified as an incidental finding during the diagnostic process for various lower urinary tract or pelvic/perineal symptoms. Demographic data, clinical presentation, radiological assessment, treatment (when indicated), and outcomes were documented and evaluated. These are summarized in **Table 1**.

Patients were followed up after having been treated surgically at 3, 6, and 12 months and yearly thereafter. Follow-up was by clinical evaluation supplemented by a patient reported outcome measure⁵ questionnaire in the latter cases in the series. Flow rate was also evaluated. Ascending and descending urethrogram was performed at 3 months, 1 year, 2 years, and 5 years postoperation. Magnetic resonance imaging (MRI) was used to follow-up those patients with complex syringoceles.

RESULTS

Of the 100 urethrograms in patients with strictures, 33.0% demonstrated filling of Cowper's ducts or glands to some extent (**Figure 1**). This was seen twice as commonly on the ascending study and occurred predominantly in patients with bulbar strictures. Only 1 of 26 patients with non-bulbar strictures had a visible duct. Previous urethrotomy or dilatation made no difference to the incidence of the positivity for duct/gland nor did stricture length and tightness.

Over the 6-year period between January 2009 and December 2014, 26 patients with Cowper's syringoceles were managed. Their mean age was 41.1 (range: 19–63) years. Fifteen (57.7%) patients presented with symptoms suggestive of lower urinary tract obstruction, primarily poor urinary flow. All patients had an ascending/descending urethrogram as part of their diagnostic evaluation. In four of these patients, a grossly dilated Cowper's duct was shown to fill up and obstruct the urethra on a descending study (**Figure 2**). In the remaining 11 patients, a bulbar urethral stricture was demonstrated which was the cause of their symptoms, and a Cowper's syringocele was identified as an incidental finding. In four of these patients, the bulbar stricture was located distal to the urethral ostium of Cowper's duct and the duct presumed to be dilated (markedly in three) secondary to obstructive voiding. The remaining seven patients with a stricture proximal to Cowper's duct opening were associated with only minimal filling of the duct on retrograde urethrogram. None of the syringoceles associated with obstructive symptoms and urethral strictures extended up to the level of the perineal membrane. Physical examination was unremarkable in all patients.

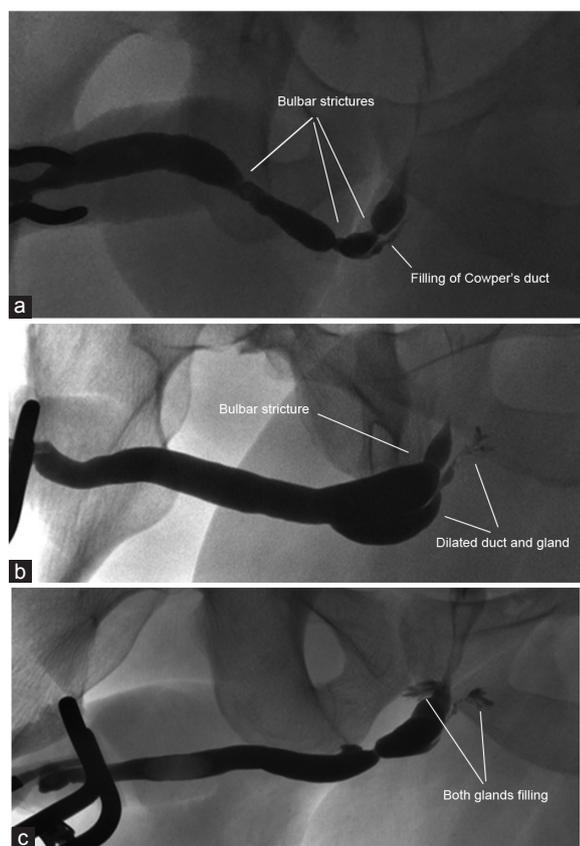


Figure 1: Appearance of syringocele on urethrography. (a) Filling of Cowper's duct only. (b) Filling of the duct and gland on one side. (c) Bilateral syringocele of duct and gland.

Eight patients presented with a history of troublesome perineal pain, associated with ejaculation in one. In six of these patients, fluoroscopic evaluation revealed grossly distended Cowper's ducts extending toward the level of the urogenital diaphragm and involving the glands themselves (**Figure 3**). All patients were evaluated by MRI which showed high signal intensity complex multicystic lesions within the bulbourethral glands on T2-weighted images (**Figure 4**). In all but one patient, these were in continuity with the dilated ducts. Again, no abnormality was detected on physical examination.

Four patients presented acutely with large perineo-scrotal abscesses resulting from infection of a large syringocele of Cowper's gland. This discharged spontaneously onto the perineum in one patient while the other three patients required surgical incision and drainage. The fifth patient developed a chronically discharging sinus in the perineum having previously been asymptomatic. Urethrogram demonstrated filling of the bulbourethral duct and gland with a tract coming off the gland and extending downward onto the perineum (**Figure 5**).

In those in whom the fluoroscopic appearance of a dilated duct was associated with a urethral stricture ($n = 11$), this resolved after treatment of the stricture by urethroplasty (dorsal augmentation using buccal mucosal graft [$n = 9$] and excision and primary anastomosis [$n = 2$]). Radiologic evidence of stricture recurrence was present in one patient at 2 years, but there was no radiological evidence of syringocele.

Three of the four patients with obstructive lower urinary tract symptoms secondary to compression of the urethra by a grossly dilated Cowper's duct had resolution of their symptoms after transperineal excision of the syringocele ($n = 2$) and endoscopic deroofting ($n = 1$). There was also no evidence of urethral obstruction on postoperative descending urethrogram in these patients. In the fourth patient, symptoms were not severe and he has been managed conservatively. To date, he has not experienced any infective complications, and regular ultrasound scans showed that he is voiding to completion.

Five of the six patients with large complex syringoceles involving Cowper's glands were managed by surgical excision via an abdominoperineal approach, with cure of their symptoms in all patients (sixth patient pending surgery). Follow-up MRI shows mild residual cystic dilatation of Cowper's gland in one patient who remains asymptomatic and is therefore being managed expectantly. One patient developed sphincter weakness incontinence after the surgery. This has been successfully treated by implantation of a bulbar artificial urinary sphincter. The five patients with infective complications of Cowper's gland syringoceles were also successfully managed by surgical excision after resolution of the acute episode.

DISCUSSION

The paired Cowper's glands are located within the urogenital diaphragm. An accessory pair of glands lies in the corpus spongiosum. These may drain separately into the urethra or join the main ducts.⁶

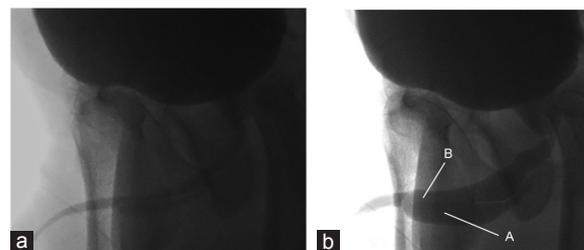


Figure 2: (a) Ascending study showing retrograde filling of Cowper's duct. (b) On descending study, dilated duct (A) is shown to obstruct distal bulbar urethra (B).



Figure 3: Urethrogram showing grossly distended Cowper's glands.

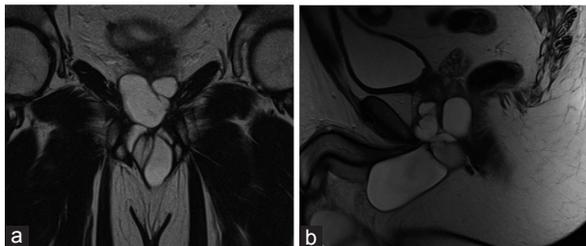


Figure 4: Multiloculated gross syringocele of bulbourethral glands as seen on MRI; (a) coronal and (b) sagittal view. MRI: magnetic resonance imaging.

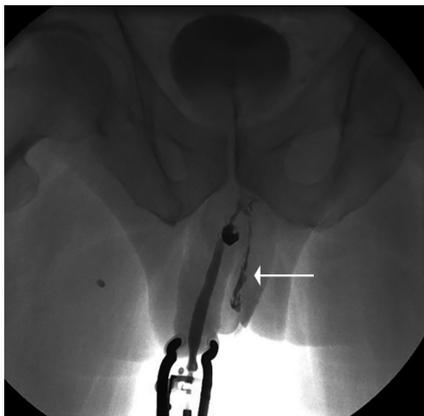


Figure 5: Urethrogram showing a fistulous tract (arrow) coming off the bulbourethral gland and extending downward onto the perineum.

Under the control of dihydrotestosterone, endodermal epithelium in the urogenital sinus is induced to form buds at the site of the future bulbous urethra. By week 11, Cowper's ducts arise from the buds, and by week 16, there is differentiation into compound tubuloalveolar secretory tissue.⁷ The bulbourethral glands are analogous to Bartholin's glands in females, and their secretions contribute to urethral lubrication during sexual intercourse besides having an immunological function.

The pathophysiology of congenital Cowper's syringoceles remains unclear; however, studies in mice have suggested that absent

or reduced levels of transforming growth factor-beta 2 (TGF- β 2) may be responsible.⁸ Acquired duct dilatation may result from post-inflammatory obstruction after infection or prolonged urethral catheterization.⁹

The incidence of Cowper's syringocele is difficult to determine accurately, particularly because many are asymptomatic. Moreover, the literature is sparse and limited to individual case reports or small case series. They appear to be more prevalent in children,^{2,4,10} but are nevertheless being reported more frequently in the adult population.^{6,11,12} This present report, to our knowledge, is the largest published series of Cowper's syringocele in adults to date.

Diagnosis is based on a thorough urological history. The primary differential diagnosis in adults is a urethral diverticulum. However, these usually present with post-void dribbling which was absent in all patients described in this series. The presence of a perineal mass has been described with syringoceles;¹³ however, in all our patients, even those with gross cystic dilatation of the glands (**Figure 3** and **4**), this was not evident on physical examination. Urethrography, in particular a voiding study, is diagnostic when there is a communication between the ducts and the urethra.¹⁴ If not, this may be shown indirectly as a mass compressing the ventral aspect of the bulbar urethra. Otherwise, cystic dilatation of the duct can be identified on transperineal ultrasound of the urethra. In our series, all complex syringoceles involving the bulbourethral glands were investigated by MRI both as a diagnostic tool but also to clearly delineate their anatomy and relationship to adjacent structures when planning surgical excision.

The striking finding from this study is that 1 in 3 patients with a radiological urethral stricture shows evidence of a Cowper's duct. More specifically, a Cowper's duct is more or less associated with a short sharp stricture of the junction between the proximal and middle segments of the bulbar urethra. It has been postulated that a stricture at this site is of "congenital" origin and known as a Cobb's collar¹⁵ or Moorman's ring.¹⁶ Third, the mere presence of a Cowper's duct is not usually a problem to the patient in its own right; indeed, these smaller syringoceles involving only the duct would probably have gone undiagnosed had the patients not presented with stricture-related symptoms. It is usually only when the full length of the duct up to the gland is involved and visible radiologically or when the duct and gland are distended that the patient has a problem directly from the syringocele. Distended ducts were more likely to cause obstructive symptoms in the absence of stricture. When the gland itself is principally involved, pain is more commonly a presenting feature and the biggest risk is of recurrent infection, abscess formation, and fistulation out through the perineal skin.

Cowper's syringoceles have been classified by Maizels *et al.*² into four types depending on their fluoroscopic appearance: (1) simple syringocele with a minimally dilated duct; (2) perforated syringocele with patulous communication with the urethra and looks like a diverticulum; (3) imperforate syringocele with a dilated bulbous duct; and (4) ruptured syringocele with a residual mucosal flap which acts as a "ball valve" to cause obstruction. However, this classification is solely radiological and has no bearing on clinical symptoms. Therefore, other authors have proposed modifications to this classification to reflect clinical presentation. Bevers *et al.*³ recommend grouping simple, perforated, and ruptured syringoceles together as "open" syringoceles which maintain a patent communication with the urethra, in contrast to the imperforate or "closed" variant which do not. Campobasso *et al.*⁴ classify syringoceles into "obstructive" (Maizels' Groups 2 and 3²) and "nonobstructive" (Maizels' Groups 1 and 4²) depending on whether they restrict urine flow or not. **Supplementary Table 1** summarizes these classifications and their correlation with reported syringocele-related symptoms.

Table 1: Symptoms, radiological appearance, and treatment of patients with Cowper's syringocele

Patient number	Age (year)	Clinical presentation	Diagnosis	Radiological appearance	Management
1	19	Obstructive LUTS	Urethrogram	Grossly dilated Cowper's duct causing urethral obstruction	Transperineal excision
2*	42	Obstructive LUTS; recurrent UTIs	Urethrogram	Moderately dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
3	63	Obstructive LUTS	Urethrogram	Grossly dilated Cowper's duct causing urethral obstruction	Conservative management
4*	43	Obstructive LUTS	Urethrogram	Moderately dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
5	58	Perineal pain worse on ejaculation	Urethrogram and MRI	Grossly distended cystic appearance of Cowper's gland	Awaiting surgical excision
6	56	Recurrent UTI; penoscrotal abscess	Urethrogram and MRI	Distended cystic appearance of Cowper's gland	
7*	53	Obstructive LUTS	Urethrogram	Mild filling of Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
8	56	Obstructive LUTS; hematuria	Urethrogram	Grossly dilated Cowper's duct causing urethral obstruction	Endoscopic deroofting
9	43	Perineal pain	Urethrogram and MRI	Grossly distended cystic appearance of Cowper's gland; no communication with urethra	Surgical excision
10	55	Perineoscrotal abscess	Urethrogram and MRI	Grossly dilated Cowper's ducts and glands	Surgical excision
11*	43	Obstructive LUTS; recurrent UTIs	Urethrogram	Grossly dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
12*	40	Obstructive LUTS	Urethrogram	Mild filling of Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
13*	35	Obstructive LUTS	Urethrogram	Mild filling of Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
14	53	Chronically discharging perineal sinus	Urethrogram and MRI	Fistulous track coming off Cowper's duct; cystic dilatation of glands	Surgical excision
15	41	Perineal pain	Urethrogram and MRI	Grossly distended Cowper's duct and glands	Surgical excision
16*	42	Obstructive LUTS	Urethrogram	Moderately dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
17	31	Perineal pain	Urethrogram and MRI	Grossly dilated Cowper's duct and glands	Surgical excision
18	36	Obstructive LUTS	Urethrogram	Grossly dilated Cowper's duct causing urethral obstruction	Transperineal excision
19*	24	Obstructive LUTS; recurrent UTIs	Urethrogram	Grossly dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
20	38	Perineal pain	MRI	Grossly distended Cowper's duct and glands	Surgical excision
21*	40	Obstructive LUTS	Urethrogram	Mildly dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
22*	21	Obstructive LUTS	Urethrogram	Moderately dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty
23	53	Perineoscrotal abscess	Urethrogram and MRI	Grossly dilated Cowper's ducts and glands	Surgical excision
24	39	Abscess discharging into perineum	Urethrogram and MRI	Grossly distended Cowper's duct and glands	Surgical excision
25	27	Perineal pain	Urethrogram and MRI	Grossly distended Cowper's duct and glands	Surgical excision
26*	20	Obstructive LUTS	Urethrogram	Moderately dilated Cowper's duct associated with obstructive bulbar stricture	Urethroplasty

*Incidental finding. LUTS: lower symptoms; UTIs: urinary tract infections; MRI: magnetic resonance imaging

However, all three classifications fail to include the more advanced variants of syringocele involving the bulbourethral glands which are presented in this series. We hypothesize that based on the pressures within the duct, there is a gradual progression from mild to increasingly prominent duct dilatation with eventual involvement of the gland itself. Based on this, the proposed classification takes into consideration whether the ducts or the glands are effected and how severely dilated they are, which has a direct implication on clinical presentation and indication for treatment. Small, mildly dilated ducts are often asymptomatic incidental findings and do not usually require treatment. More obviously, dilated ducts are the ones which will give rise to obstructive symptoms and which require treatment by endoscopic deroofting,^{17,18} transperineal ligation,¹⁹ or formal surgical excision.²⁰

At the extreme end of the spectrum, syringoceles involving Cowper's glands themselves lead to important complications such as perineal pain, abscess formation, and perineal fistulae and therefore also require surgical excision.

Notwithstanding the fact that this is the largest series of Cowper's syringoceles reported in the literature, the patient cohort size remains limited. Follow-up for most patients in the series is intermediate term with few patients having been followed up beyond 5 years. Having said that, at least in the case of strictures, if these are going to recur they will do so most commonly within the first 2 years after surgery. Further follow-up of these patients is nonetheless warranted.

All urethral strictures in this series were treated using autologous grafts (when a graft was necessary) which remain the gold standard urethroplasty technique (with or without the presence of associated

syringoceles). Various tissue-engineering techniques for urethral reconstruction are currently being investigated;²¹ however, none has as yet made it to routine clinical practice. The current treatment of large, complex syringoceles of Cowper's glands also remains open surgical excision and reconstruction as was the case in all patients in this series. Nevertheless, advances in minimally invasive surgery, particularly robotic urologic reconstructive surgical techniques over the last few years,²² could indicate a shift toward these procedures being performed using robotic assistance in the not too distant future.

CONCLUSION

Cowper's syringocele in adult men may be more common than previously thought and even though many are incidental findings, others may be a cause of lower urinary tract symptoms or be misdiagnosed as urethral diverticula or urethral duplication. Others still may be associated with serious complications such as infection, abscess formation, and fistulation. The current definition and classifications fail to appreciate cystic involvement of the bulbourethral glands as the extreme end of the Cowper's syringocele spectrum. These are commonly the ones that are associated with the more significant symptomatology and complications and which usually require surgical treatment.

AUTHOR CONTRIBUTIONS

SB contributed to manuscript preparation and data analysis. AF performed data collection and analysis. SI and MD made contributions to data collection; DEA reviewed the manuscript. ARM is the senior operating surgeon for all cases described in the series. All authors read and approved the final manuscript.

COMPETING INTERESTS

All authors declared no competing interests.

Supplementary Information is linked to the online version of the paper on the *Asian Journal of Andrology* website.

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Supplementary Table 1: Existing classifications of Cowper's syringoceles and relation to symptomatology

	<i>Open</i>	<i>Closed</i>	<i>Obstructive</i>	<i>Nonobstructive</i>
Maizels' equivalent	Simple Perforated Ruptured	Imperforate	Perforate Imperforate	Simple Ruptured
Symptoms	Postmicturition dribble UTI Urethral discharge Hematuria Perineal pain	Obstructive LUTS Dysuria Perineal pain	Obstructive LUTS Perineal pain	Postmicturition dribble UTI Urethral discharge Hematuria

LUTS: lower urinary tract symptoms; UTI: urinary tract infection