Urology Case Reports 46 (2023) 102306

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Cowper's gland syringocele in an adult: The great imitator

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A B S T R A C T

Cowper's gland syringocele (CGS) is the cystic dilation of its duct. It is an uncommon urological condition and is thought to be more commonly encountered in pediatric urology. However, it is in adults that CGS poses a diagnostic challenge because of its "chameleon-like" clinical presentation that may masquerade multiple urological etiologies. In this population, where urological conditions are more prevalent, CGS may present as bladder outlet obstruction, recurrent urinary tract infections, gross hematuria, urinary retention, perineal pain, or abscess.

1. Introduction

Cowper's glands are situated on each side of the posterolateral membranous urethra within the urogenital diaphragm in the deep perineal pouch. The gland secretion is part of the seminal fluid and lubricates the male urethra during the sexual arousal.[1] To the urologist, the Cowper's gland becomes clinically significant in its pathologic state. Cowper's gland syringocele (CGS) is the cystic dilation of the gland's duct. CGS may be congenital or acquired, and is most frequently diagnosed in children and young adolescents. Some reports indicate that patients with congenital CGS may have additional associated urinary tract pathologies. This may suggest that the detrimental effects of this lesion are already initiated in utero.[2] In adults, CGS poses a diagnostic challenge because of the higher prevalence of urological conditions and propensity for imitating multiple urological symptoms. Classification systems have been proposed based on radiologic and endoscopic appearance. Maizels M. et al., in 1983 described a system that classified CGS into four types: simple, imperforated, perforated and ruptured. However, a simpler more useful classification differentiates obstructive from non-obstructive CGS.[3] Currently, there is no consensus as to which is the best diagnostic modality, and which is the best treatment approach. Although classification systems for types of CGS have been proposed, based on radiologic and endoscopic appearance or clinical symptoms, these are not helpful in deciding on the ideal treatment approach.

2. Case report

A 49-year-old man presented to the urology clinic with a one-year history of recurrent urinary tract infections (UTI) and sepsis. He presented to our clinic 2 weeks after the last UTI episode. The patient stated that during UTI episodes he had a significant decrease in his force of stream. During the last UTI episode CRP 180, PSA 1.0 ng/ml, and urine was positive for *Pseudomonas Aeruginosa and Proteus Mirabilis*. Past medical history was significant for left nephroureterectomy at the age of 18-years for hypertensive disease secondary to a left, hydronephrotic, multicystic non-functioning kidney. In the clinic, genitourinary physical examination was unremarkable but the postvoid residual volume was 190 ml. Ultrasound, done a week earlier, reported a normal right kidney, and an enlarged prostate estimated at 46 cc. Initial cystourethroscopy showed a normal urethra. CT urogram was normal except for a periurethral cyst 3.5 cm just distal to the external urethral sphincter (EUS). MRI suggested Cowper's syringocele (Fig. 1. A, B). Cystoscopy was carried out again and this time with contraction of the pelvic floor and a right sided urethral mass was seen bulging into the urethral lumen just distal to the EUS (Fig. 2, see cystoscopy video). Subsequent Retrograde urethrogram indicated an imperforate CGS. Patient opted for conservative treatment. He is 21 months since last infection with normal voiding pattern, emptying well and random perineal pain episodes.

3. Discussion

Cowper's gland syringocele is rare but well documented. The diagnosis of CGS in the pediatric population seems straightforward since the

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https://doi.org/10.1016/j.eucr.2022.102306
Received 19 October 2022; Received in revised form 6 December 2022; Accepted 15 December 2022
Available online 16 December 2022
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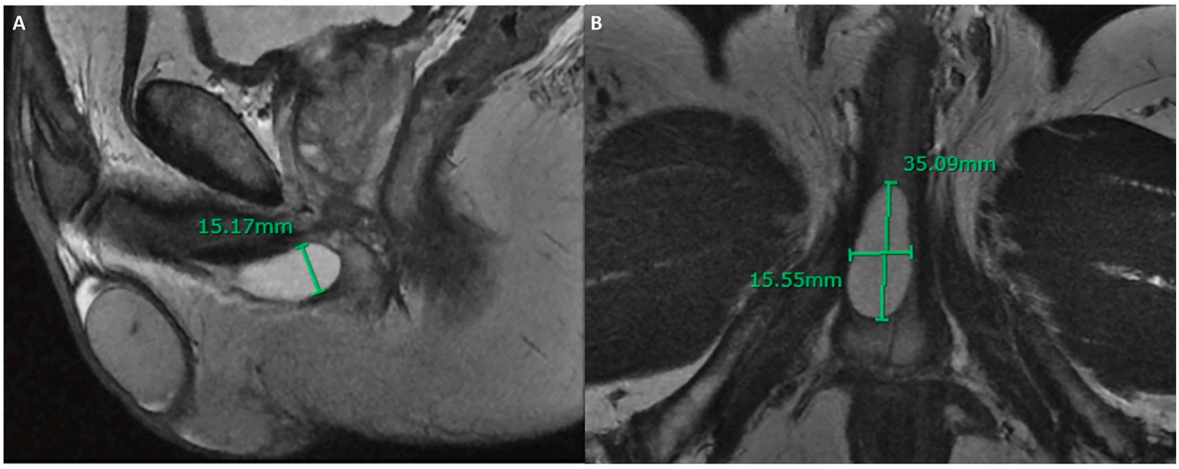

Fig. 1. MRI Cowper's Gland Syringocele **A:** Sagittal view, **B**: Transversal view.

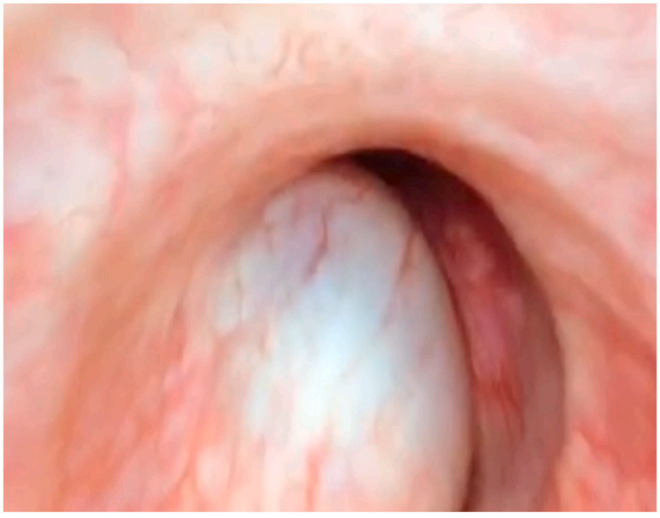

Fig. 2. Cystoscopy: Obstructive Cowper's gland syringocele.

vast majority (95%) of patients are diagnosed secondary to symptoms such as obstruction or UTIs.[4] CGS may "fly under the radar" and present in an adult. In one study only 5% of patients with CGS were in their 5th decade.[5] CGS poses a diagnostic challenge in the adult population. The reason may be that CGS has a "chameleon-like quality", imitating multiple urologic conditions in a population where these are more prevalent. Second, since most cases are diagnosed in the pediatric population most urologists may be unfamiliar with this etiology. Finally, as in our case initially, cystoscopy may easily miss an imperforated type syringocele. The technique of contracting the pelvis/bulbo-spongiosus muscle to cause the syringocele to bulge into the urethral lumen is helpful in making the diagnosis. If CGS is suspected an MRI should be the exam of choice to confirm the diagnosis. However, this etiology should be suspected in the adult with a history of recurrent UTIs, obstructive symptoms, history of upper tract abnormalities and PSA not elevated during the acute phase infection.

Our patient opted for conservative treatment with clinic follow-ups every 3 months. He has been asymptomatic for nearly 2 years. Although surgical treatment (endoscopy versus open repair) is an acceptable option in treating these patients post-void dribbling and urethral stricture formation are possible complications. There are currently no clear guidelines regarding the optimal approach to patients with this etiology.

4. Conclusion

Cowper's gland syringocele is a rare condition that may give rise to multiple urological symptoms. General urologists should be familiar with and aware of this etiology. Cystoscopy may easily miss the diagnosis even with 3.5 cm lesions. MRI should be done to confirm the diagnosis.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.eucr.2022.102306.

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