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Case Report

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Prostatic Utricle Cyst – A Rare Case of Obstructive Azoospermia

Md Faisal Islam[1], Hamudur Rahman[2], Ashraf Rahman[3], Md Sazid Imtiaz[4], Mohammad Mahfuzur Rahman[5], Md Firoz Mian[6]

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Abstract

Prostatic utricle cyst resulting from Mullerian duct remnant is a rare congenital condition in males and is often associated with structural anomalies in external genitalia. While most cases remain asymptomatic, clinical presentation ranges from lower urinary tract voiding symptoms, urinary tract infection, epididymitis, rectal mass, hematospermia, and obstructive azoospermia. Transrectal ultrasound is the reliable modality of investigation to distinguish prostatic utricle cysts causing obstructive azoospermia, especially when complemented with characteristic semen analysis parameters. Magnetic resonance imaging is also beneficial when ultrasound is inconclusive. While vasography provides excellent information regarding anomalies in the reproductive tract, it should be performed when surgery is planned. Urethrocystoscopy remains one of the definitive approaches to diagnosing prostatic utricle cysts, and transurethral resection showed excellent outcomes while being minimally invasive. In this article, a rare case of obstructive azoospermia by prostatic utricle cyst is presented, and clinical symptoms, diagnosis, and management are discussed.

Introduction

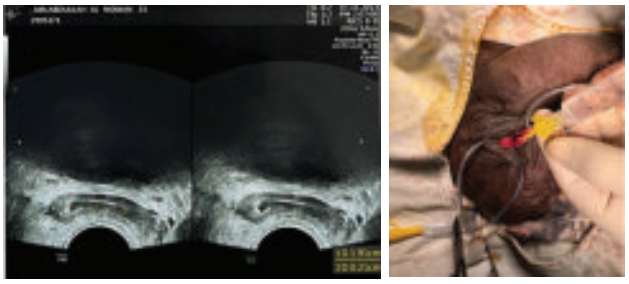
Cystic enlargement of the prostatic utricle, a vestigial remnant of the Mullerian duct, is a rare condition in males. It is present in up to 4% and 1% in newborns and adults respectively[1]. The incidence of prostatic utricle cysts is 11% to 14% in association with hypospadias or intersex anomalies and up to 50% in the presence of perineal hypospadias[2]. Majority of prostatic utricle cysts remain asymptomatic but may present clinically as lower urinary tract voiding symptoms, urinary retention, epididymitis, rectal mass, or obstructive azoospermia. Symptoms are determined by relative cyst size, the degree of obstruction of the bladder neck or seminal vesicles and ejaculatory ducts, and whether there is an associated infection[3]. Although obstructive azoospermia may be acquired congenitally, such as in Young's syndrome, which presents with bilateral vas deferens abnormalities, it commonly develops as a sequela of prostatic and ejaculatory duct cysts, childhood inguinal hernias, epididymitis, and vasectomy[4]. Prostatic utricle cyst may cause partial or complete ejaculatory duct obstruction. Complete ejaculatory duct obstruction is associated with azoospermia and low semen volume; however, partial obstruction has a variable clinical presentation, including oligozoospermia with low or normal semen volume[5]. We report a rare case of obstructive azoospermia due to a prostatic utricle cyst.

1. Professor of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh
2. Assistant Professor of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh
3. Assistant Register of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh
4. MS Phase B Resident of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh
5. Assistant Register of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh
6. MS Phase B Resident of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh

Correspondence: Prof. Md. Faisal Islam, Professor of Urology, National Institute of Kidney Diseases & Urology (NIKDU), Dhaka, Bangladesh, Email: faisaluro@gmail.com

Case Report

A 32-year-old unmarried gentleman presented with watery, low ejaculate volume during masturbation for the past 14 years. He had normal libido as well as erection and gave no history of testicular trauma, sexually transmitted diseases, hematospermia, childhood mumps, frequent respiratory infection, anosmia, and visual disturbance. His past medical history is significant in taking anti-tubercular therapy empirically for an epididymal nodule about 12 years back. He had no history of surgical intervention as well as taking any anabolic steroids. He is a nonsmoker and non-alcoholic. The scrotal examination was unremarkable, with bilateral normal-sized testes, normal epididymis, vas deferens, and no varicocele was found on either side. Digital rectal examination revealed a painless elongated cystic mass over the left lobe of prostate. Laboratory data, including blood count and urinalysis, were unremarkable. Urine GeneXpert for Mycobacterium tuberculosis was negative. Semen analysis revealed lowejaculate volume (0.5 ml) as well as azoospermia and absence of fructose. Hormonal ass, including follicular stimulating hormone and testosterone were normal. Normal spermatogenes were found on testicular biopsy. Abdominal ultrasound was normal; however, scrotal ultrasound showed bilateral epididymitis, and transrectal ultrasound revealed linear cystic lesion in seminal vesicle (Fig 1). Magnetic resonance imaging demonstrated a cystic lesion in the left seminal vesicle (Fig 2). Therefore, an intraoperative vasogram was performed to visualise the obstruction site and aid in surgical planning. Water soluble contrast media was injected via a 24-gauge cannula (Fig 3) towards the epididymis as well as the ejaculatory duct. Right-sided



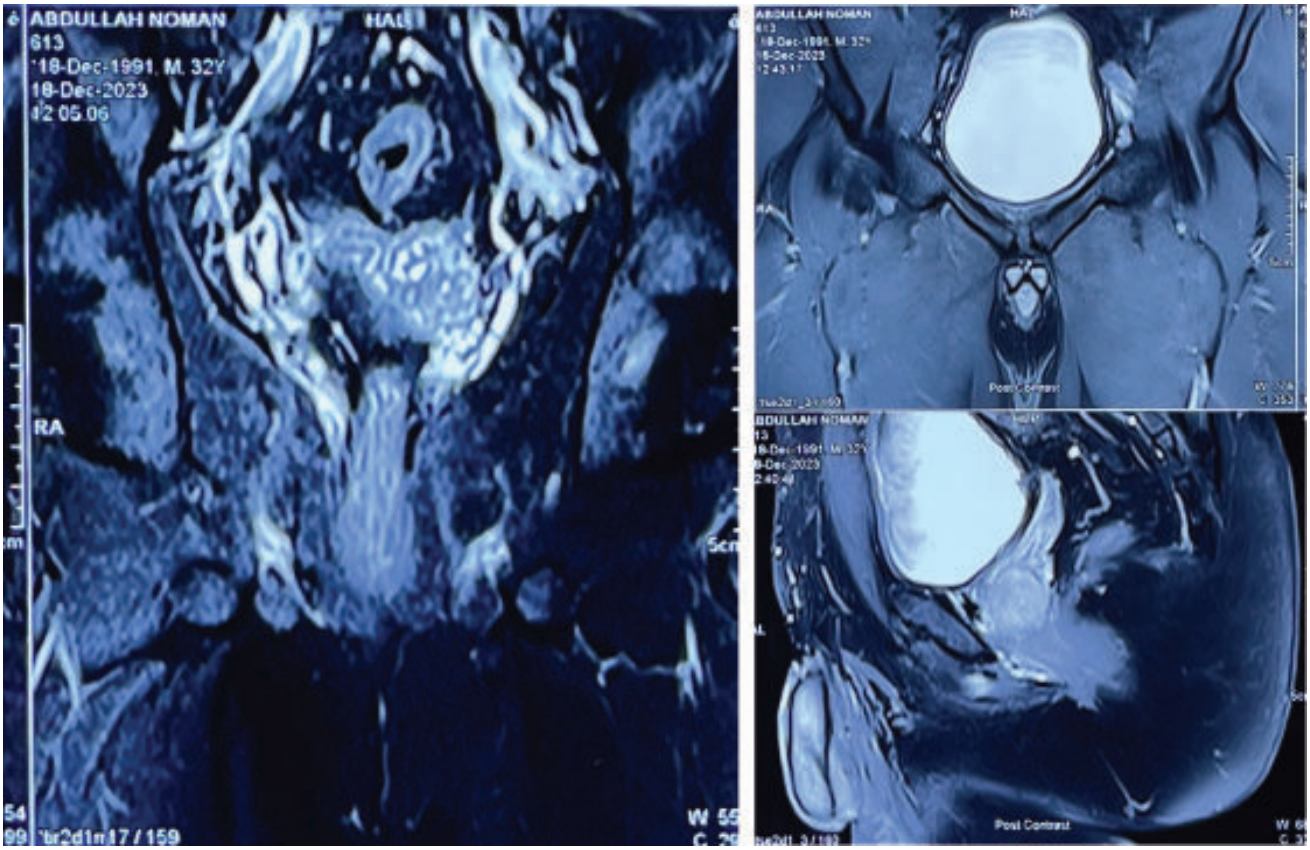


Fig 2: *MRI showing left seminal vesicle cyst indicated by fluid hyperintensity in both T2 weighted and post contrast film.*

vasogram opacified vas deferens and seminal vesicle as well as a pear-shaped dilated area at the level of urinary bladder (Fig 4). Left-sided vasogram revealed contrast material was travelling freely through the left vas deferens and seminal vesicle.

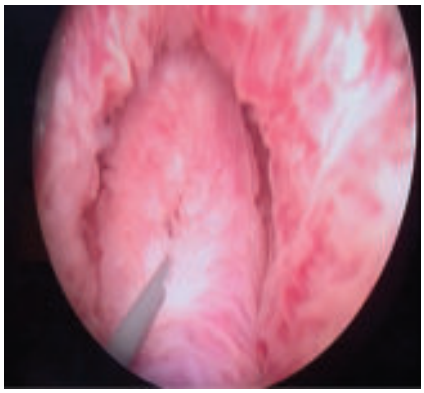


Fig 3: *Vasogram*

The patient was placed in the lithotomy position, and a urethrocystoscopy was performed. Urinary bladder appeared normal, although a bulging was seen at the verumontanum. Ejaculatory duct orifices were not seen. Using counterpressure from the anterior rectal wall, swelling with a slit-like opening was noted in themidline at the apex ofthe verumontanum. A guidewire (0.18 French) was inserted, and 6 French ureteroscope was introduced through the opening (Fig 5-a). A cystic lesion in prostatic utricle was found, as well as opening of both ejaculatory ducts into the cyst

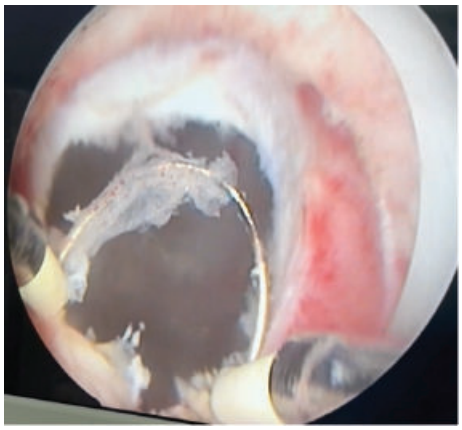


Fig 4: *Bilateral vasogram showing peer shaped dilated area at the level of neck of urinary bladder*

cavity (Fig 5-b). The cyst was communicating with the posterior urethra. Transurethral endoscopic wide deroofing of prostatic utricle cyst was performed by wire loop (Fig 5-c). The unroofed prostatic utricle cyst contained clear fluid. An 18-French Foley catheter was placed without irrigation for five days postoperatively. There were no postoperative complications or infections. Normal urinary continence was documented after surgery. Histopathology of resected tissue showed fibrosis. 21 days following surgery, his semen analysis parameters were semen volume 01 ml, sperm concentration 06 million/ml, total sperm count 11 million/ejaculate with 28% motility.

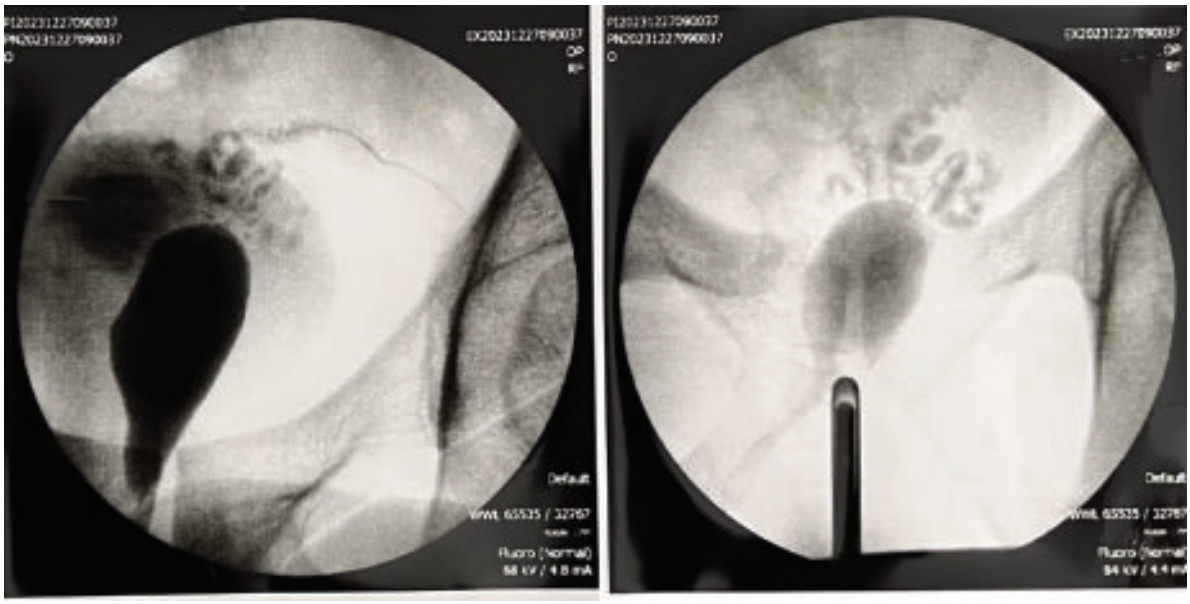
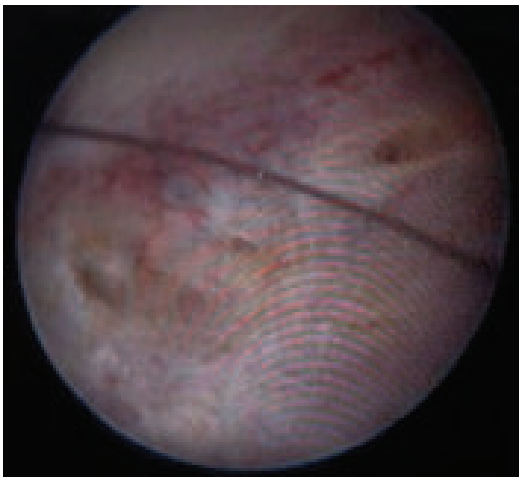


Fig 5: *(a) Prostatic utricle cyst (b) Inside cystic cavity including opening of ejaculatory duct (arrow marked) (c) Deroofing of prostatic utricle cyst*



Discussion

The prostatic utricle is a vestigial cavity, opening at the top of the verumontanum, located between the termination of the two ejaculatory ducts at the posterior border of the prostatic urethra. It becomes pathological when it is dilated. This dilatation is most often congenital and is referred to as a prostatic utricle cyst. It derives from an anomaly in the involution of the caudal end of the Müllerian apparatus due to a deficiency in the anti-Müllerian hormone affecting predominantly males younger than 20 years[2]. Utricle cysts are pear-shaped and can be revealed by signs of obstructive azoospermia, such in our case, urinary tract infection, pelvic pain, hematospermia, epididymitis, urethral discharge, or asymptomatic discovery, particularly if it is small. Although our patient had never experienced any subjective symptoms suggestive of a sexually transmitted disease, epididymitis, or prostatitis, prostatic utricle cysts are known to cause epididymitis, which may be due to retrograde infection of the urine remaining in the cyst or compression or obstruction of the ejaculatory duct by the cyst[6]. The ejaculatory ducts enter the urethra on the sides of the verumontanum but are not part of the utricle. Since the paramesonephric derivatives (utricle and Mullerian remnants) and the mesonephric derivatives (vas, seminal vesicle and ejaculatory duct) come from separate embryologic systems, it would not be expected that a prostatic utricle cyst would incorporate the ejaculatory ducts. Some recent case reports revealed the ejaculatory duct or distal vasa entered the cyst, contributing sperm to its contents[7]. In the case of ectopic opening of the ejaculatory duct into prostatic utricle cyst, sperm are usually detained in the cysts, where it is not suitable for them to live, which results in oligospermia and asthenospermia[8]. Sperm quality is even worse if infection and stones of the cyst are complications. Diagnosis of obstructive azoospermia is suggested by the finding of an ejaculate volume of < 1 ml in a patient with azoospermia. The ejaculate consists of thin fluid which fails to coagulate. The p^H of the fluid is between 6 and 7, and fructose is absent. The ejaculate usually lacks the characteristic odor of semen[4]. The differential diagnosis must always be considered, particularly a seminal vesicle cyst, urethral cyst, bladder diverticulum and most importantly, Müllerian duct cysts. Müllerian duct cysts are usually not communicating with the prostatic urethra. They are round and are frequently encountered in adults (20-40 years) with normal external genitalia. While prostatic utricle cysts are tubular or vesicular in shape, they are most commonly observed in pediatric and adolescent patients (<20 years of age) with hypospadias, cryptorchidism and sexual dysplasia and usually in communication with the prostatic urethra[9]. After suspicion of the diagnosis based on clinical elements, transrectal ultrasonography is proved to be a reliable diagnostic tool in men with obstructive infertility, especially when combined with seminal analysis. It allows to delineate pelvic mass of cystic appearance, intraprostatic and median in relation to the neighbouring organs such as the prostate and the urethra[10]. Although ultrasonography is the first-line examination, MRI allows multiplanar acquisition for better precision of the dimensions of the cyst, to localise it in the adjacent structures, in particular the rectum, bladder, and seminal vesicles, as well as its communication or not with the prostatic urethra[11]. Serum testosterone and follicular stimulating hormone are useful in ruling out occult hypogonadism. Testicular biopsy is not essential but may be done as confirmatory tests in a setup where sperm retrieval can be performed as well. In case of diagnostic unconformity, operative vasography can help identify obstruction level in the seminal pathway. Although it is a useful radiological modality in managing obstructive azoospermia, it should be performed if definitive reconstructive surgery is planned[10]. Certainly, the most useful investigation for a prostatic utricle cyst is urethrocystoscopy to identify the utricular orifice in the posterior urethra. Surgical treatment can also be performed with the aid of a urethrocystoscope. Appropriate surgical procedures have been described to treat symptomatic prostatic utricle cyst. Endoscopic dilatation of the orifice of the utricle, followed by catheterization and aspiration of its contents with or without sclerotherapy, and

transurethral resection of the anterior wall of the cyst are two less invasive techniques; they make it possible to avoid the complications of surgery, especially infertility and impotence. Transurethral resection of the prostatic utricle cyst can be performed using the Holmium laser with reliable and effective results[12]. Surgical resection is recommended for utricles with recurrent symptoms. Traditional open-surgical approaches require high operational skills and may damage adjacent tissues. Laparoscopic excision of prostatic utricle is suitable for surgeons skilled in advanced laparoscopic techniques. This technique reduces intrusion to the retrovesical space, provides a clear vision, and reduces the time required for recovery[13].

Conclusion

The cyst of the prostatic utricle that is not associated with an abnormality of the external genitalia, cryptorchidism or renal agenesis constitutes an entity rarely reported in the literature. Most of the patients remain asymptomatic. This poses a challenge in diagnosis and management. Obstructive azoospermia due to a prostatic utricle cyst is one of the male factor infertility that can be corrected surgically. Modern imaging techniques and endoscopic procedures allow us to overcome diagnostic challenges and achieve excellent surgical outcomes.

References

1. Desautel MG, Stock J, Hanna MK. Müllerian duct remnants: surgical management and fertility issues. J Urol [Internet] 1999 [cited 2024 Jan 24];162(3 Pt 2):1008–13. Available from: https://pubmed.ncbi.nlm.nih.gov/10458422/
2. Shebel HM, Farg HM, Kolokythas O, El-Diasty T. Cysts of the Lower Male Genitourinary Tract: Embryologic and Anatomic Considerations and Differential Diagnosis. https://doi.org/101148/rg334125129 [Internet] 2013 [cited 2024 Jan 24];33(4):1125–43. Available from: https://pubs.rsna.org/doi/10.1148/rg.334125129
3. Desautel MG, Stock J, H A MK. Mullerian Duct Remnants: Surgical Management And Fertility Issues. 1999.
4. Sharlip ID. Obstructive azoospermia or oligozoospermia due to Mullerian duct cyst. 1984.
5. Lotti F, Corona G, Cocci A, Cipriani S, Baldi E, Degl'Innocenti S, et al. The prevalence of midline prostatic cysts and the relationship between cyst size and semen parameters among infertile and fertile men. Human Reproduction [Internet] 2018 [cited 2024 Jan 25];33(11):2023–34. Available from: https://dx.doi.org/10.1093/humrep/dey298
6. Saito T, Komeya M, Usui K, Kuroda S, Takeshima T, Takashima K, et al. A case of obstructive azoospermia secondary to genitourinary tract infection caused by a prostatic utricle cyst. Asian J Androl [Internet] 2022 [cited 2024 Jan 26];24(5):558. Available from: /pmc/articles/PMC9491043/
7. Gomes AL, Freitas Filho LG, Leão JQS, Heinisch AC, Carnevale J. Ectopic opening of the vas deferens into a Müllerian duct cyst. J Pediatr Urol 2007;3(2):151–5.
8. Albakr A, Arafa M, Majzoub A, El Ansari W, Qasem M, Al Said S, et al. A rare abnormality of ejaculatory duct opening in the bladder trigone in a 33-year-old male associated with primary infertility: Case report and literature review. Andrologia [Internet] 2022 [cited 2024 Jan 26];54(11):e14627. Available from: https://onlinelibrary.wiley.com/doi/full/10.1111/and.14627
9. Hester AG, Kogan SJ. The prostatic utricle: An under-recognized condition resulting in significant morbidity in boys with both hypospadias and normal external genitalia. J Pediatr Urol [Internet] 2017 [cited 2024 Jan 26];13(5):492.e1-492.e5. Available from: https://pubmed.ncbi.nlm.nih.gov/28319024/
10. Donkol RH. Imaging in male-factor obstructive infertility. World J Radiol [Internet] 2010 [cited 2024 Jan 26];2(5):172. Available from: /pmc/articles/PMC2999021/
11. Gevenois PA, Van Sinoy ML, Sintzoff SA, Stallenberg B, Salmon I, Van Regemorter G, et al. Cysts of the prostate and seminal vesicles: MR imaging findings in 11 cases. AJR Am J Roentgenol [Internet] 1990 [cited 2024 Jan 26];155(5):1021–4. Available from: https://pubmed.ncbi.nlm.nih.gov/2120929/
12. Manjiri S, Luthra K, Padmalatha SK, Shetty J, Patil N. Symptomatic prostatic utricle: various approaches for treatment. Annals of Pediatric Surgery [Internet] 2020 [cited 2024 Jan 26];16(1):1
7. Available from: https://aops.springeropen.com/articles/10.1186/s43159-020-00062-8
13. Liu B, He D, Zhang D, Liu X, Lin T, Wei G. Prostatic utricles without external genital anomalies in children: Our experience, literature review, and pooling analysis. BMC Urol [Internet] 2019 [cited 2024 Jan 26];19(1):1–7. Available from: https://bmcurol.biomedcentral.com/articles/10.1186/s12894-019-0450-z