

Recurrent Urinary Tract Symptoms In A 40-year-old Man with Unilateral Right Intravesical Ureterocele: A Case Report

Sule MB^{1*}, Sa'idu SA¹, Ma'aji SM¹, Danfulani M¹, Yunusa GH¹, Umar AU², Gele IH³

¹Radiology Department, Usmanu Danfodiyo University, Sokoto

²Radiology Department, Gombe State University, Gombe

³Radiology Department, Usmanu Danfodiyo University Teaching Hospital, Sokoto

*Corresponding Author

Dr. SULE Muhammad Baba

Article History: | Received: 10.11.2020 | Accepted: 26.12.2020 | Published: 03.01.2021 |

Abstract: Ureterocele is defined as a congenital abnormality characterized by cystic dilatation of the distal intravesical portion of the ureter. It has an incidence of about 1 in 500 to 1 in 4000 individuals and shows a female preponderance and a ratio of about 4-6:1 (Female: Male) with prevalence among the Caucasians. This is a forty-year-old male who was referred to the radiology department for intravenous urography (IVU) following history of recurrent right loin pain since childhood and also right hydronephrosis following several abdominopelvic ultrasounds scans. The IVU showed bulbous dilatation of the distal intravesical portion of the right ureter with surrounding circumferential filling defect within the contrast opacified bladder giving the so-called "Cobra-head" appearance of intravesical/orthotopic ureterocele. Dilatation of the right ureter and collecting systems were also demonstrated. The complimentary ultrasonography showed a cyst within a cyst appearance on the right conforming to intravesical ureterocele, right hydronephrosis was also demonstrated. The patient had courses of antibiotics with subsequent Transurethral resection in the referring health care facility. We report a case of intravesical ureterocele in a 40-year-old man with recurrent loin pain and urinary tract symptoms since childhood.

Keywords: Duplex collecting system, Intravesical ureterocele, Urinary tract infection, Cobra-head appearance.

Copyright © 2021 The Author(s): This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

INTRODUCTION

Ureterocele is defined as a cystic dilatation of the distal intravesical portion of the ureter, the term ureterocele was first used by Leshnew in 1912 [1]. The pelvicalyceal system drained by the ureterocele is often obstructed [1, 2].

The etiology of ureterocele is basically speculative, it's been suggested that ureteroceles have an obstructive etiology due to delayed and incomplete reabsorption of the membrane that separates the ureteral bud from the mesophrenic duct in the embryo [1, 3].

Ureteroceles are seen to affect the right and left side ureter equally, and most often associated with a duplicated collecting system with affectation of simple/single ureter in about 20% of cases [4-7]. Ureteroceles in a duplicated collecting system are

reported as a known and recognized documented occurrence [8].

Ureteroceles are occasionally asymptomatic, but often with a sequela of hydronephrosis and reflux which may lead to permanent renal damage [8].

Ureteroceles are also classified as either intravesical or extravesical with respect to their location [8, 9] the intravesical ureterocele is completely located within the bladder and associated with good renal function. However, intravesical ureteroceles may cause obstruction and therefore referred as stenotic ureteroceles [8].

Extravesical ureteroceles has a portion of the ureterocele in the bladder neck or urethra and associated with poor renal function. Extravesical ureteroceles if causes obstruction, it is referred as

sphincteric stenotic ureterocele. Extension of a ureterocele beyond the bladder neck into the urethra, this is referred to as cecoureterocele or a prolapsed ureterocele [8, 10].

Ureteroceles may affect both ureters equally and seen in about 10% of cases, slight affectation of the left ureter more than the right is reported by some authors [8, 11-13]. A ureterocele in the ureter inserting at the normal site is called simple or orthotopic, and if it occurs at the ureter inserting caudal and medial, it is considered ectopic [1, 8, 9, 12].

About 60-80% of ureteroceles are ectopic in comparison to the intravesical forms, and 80% of ureteroceles are associated with the upper moiety of a complete duplication system and often seen in the pediatric age group, but when seen in adult, most are often intravesical and of the single system [12, 14, 15].

Stephens further classified ureteroceles as either intravesical ureterocele; these may either be stenotic in 40%, or non-obstructive in 5%, and ectopic ureterocele: these may be sphincteric in 40%, sphincterostenotic in 5%, cecoureterocele in 5% or blind in another 5% [2, 12].

Ureteroceles often present with varying symptoms ranging from hematuria, purulent urine, abdominal pain, renal failure, life threatening sepsis, frequent urinary tract infections, pyelonephritis, and to no any symptoms at all and usually detected incidentally or by antenatal ultrasonography [8, 12, 15].

Ureteroceles are reported to coexist with certain urinary tract anomalies, some of these are; duplicate system, multicystic kidney disease, polycystic kidney disease, horseshoe kidney, crossed fused renal ectopia, posterior urethral valve, hypospadias and myelomeningoceles, most of these associations are rather very rare [3, 4, 16-19].

Ureteroceles are diagnosed by imaging, these are ultrasonography, intravenous urography, and voiding cystourethrography to mention but a few, these detect the location, the size, affectation of the upper tracts, anatomy and function of the renal system and the clinical condition with other clinical features may lead to the choice of the imaging modality [2, 12, 15].

Ureteroceles are basically treated by medical and surgical approach, the medical treatment comprises of antimicrobial agents to treat any coexisting urinary tract infections. The surgical treatment is by transurethral resection and

unroofing of the ureteroceles and endoscopic meatotomy [4, 20, 21].

CASE REPORT

A forty-year-old male presented to the radiology department for intravenous urography following history of persistent right loin pain and hydronephrosis on several abdominopelvic scans.

He looks physically stable with no discomfort. He is well hydrated and not pale on physical examination. The patient had normal pulse rate of 72 beats/minute, blood pressure of 120/70mmHg, and normal respiratory cycle of 14 cycles per minute.

The patient had history of occasional and mild hematuria, recurrent right loin pain radiating to the groin and right testicle, recurrent supra-public pain with occasional urethral discharge.

The patient had urine microscopy that yielded the growth of *Escherichia Coli*, the urinalysis showed pus cells, white cells and red blood cells with traces of protein. The blood electrolyte, urea and creatinine were however within normal limits.

The IVU showed good excretion bilaterally with right hydroureteronephrosis (figure 2). The intravesical ureter on the right showed filling of bulbous terminal end with surrounding halo of filling defect likened to a 'Cobra head' appearance of intravesical or orthotopic ureterocele (figure 1 & 2).

No calculus (area of double density or filling defect) was demonstrated in the distal right ureter or bladder. No bladder mass was seen, bladder wall was normal and no significant post-voidal residue volume was demonstrated.

Complimentary ultrasonogram of the abdomen and pelvic region showed a cystic dilatation of the distal intravesical portion of the right ureter with a urine distended bladder lumen giving the so-called cyst-within-cyst appearance of intravesical or orthotopic ureterocele. There is associated dilatation of the right collecting system (single system noted) and right ureter; hydroureteronephrosis. Normal contralateral urinary tract, normal abdominal organs and situs were also demonstrated.

A diagnosis of a right intravesical or orthotopic ureterocele in a forty-year-old man with recurrent right urinary tract symptoms and complaint of recurrent right loin pain was established.

The patient was treated at the referring health care centre, had a course of antimicrobial agents for the treatment of the urinary tract infection. He also had surgical excision of the

ureterocele following a transurethral resection, and reported to have done well and got discharged home afterwards by a relation.



Fig-1: A conventional delayed pyelogram phase of an IVU showing dilated distal end (intramural) of the intravesical portion of the right ureter with Surrounding or circumferential filling defects giving the so called giving the so-called "Cobra head" appearance of intravesical ureterocele, distal right ureter (pelvic part) and residual contrast medium after voiding. This is a non-significant residual volume excluding urine retention



Fig-2: A pyelogram phase of an IVU; right dilated calyces and pelvis, prominent right ureter with a dilated and contrast filled distal outpouch/sacculation of the distal right intravesical ureter and surrounding circumferential filling defect; these together form the so-called "cobra-head" appearance of an intravesical normally situated orthotopic ureterocele



Fig-3: Ultrasonograms of the right urinary tract, showing a mildly distended urinary bladder with the ureterocele on the left image appearing as a cyst-within-a cyst, and dilated calyceal moieties of the right kidney on the right image

DISCUSSION

Ureterocele is a congenital disorder that causes a submucosal cystic dilatation of the terminal ureter, it has a fibromuscular wall lined with urothelium that swells and causes an abnormality in the flow of urine. These appearance leads to a fluid-filled structure within the bladder lumen and often referred to as a cyst within a cyst [8]. The index case also was not an exception to these, had the appearance of a cyst within a cyst following ultrasonography in figure 3. Thereby conforming to this literature.

Four types have been described by Brueziere[22], these are; ureterocele with single ureter (10%), ureterocele with total duplication and intravesical development (10%), ureterocele with total duplication and extravesical development (62%) and ureterocele with ectopic ureter (39%). The index patient has a ureterocele with a single system seen in about 10% of cases, thereby conforming to what was reported by Brueziere.

Ureterocele may either be intravesical or ectopic, with the intravesical form been often associated with a single ureter while the ectopic ureteroceles involves the upper of duplicated ureters[23], the index case is an intravesical ureterocele and associated with a single ureter/collecting system conforming to this literature.

Ureteroceles are more common in females (mostly children) and almost exclusively seen in caucassions [24], the patient under review happens to be an adult male and of African ancestry invariance to this literature.

Ureteroceles have been shown to rarely associate with conditions like Klinefelter's syndrome (49, XXXXY) and agenesis of the ipsilateral kidney (more with intravesical form) and other forms of

urinary tract and renal anomalies [3, 4, 16-19, 25, 26].

Ureteroceles may affect both ureters equally and seen in about 10% of cases, slight affectation of the left ureter more than the right is reported by some authors [8, 11-13], and the index case has a right sided ureterocele, and invariance to these literatures.

Pseudo-ureterocele may mimic ureterocele; this is otherwise an obstruction of normal intramural ureter. This is often associated with tumors (bladder tumor, commonly in adults) and edema (mostly from impacted ureteral calculus or instrumentation) [27]. The index case was that of a true intravesical ureterocele, he had no associated bladder tumor or impacted ureteral calculus or history of past instrumentation, and invariance to this literature.

Ureteroceles often present with varying symptoms ranging from hematuria, purulent urine, abdominal pain, renal failure, life threatening sepsis, frequent urinary tract infections, pyelonephritis, and to no any symptoms at all and usually detected incidentally or by antenatal ultrasonography [8, 12, 15], the patient under review had most of these features thereby conforming to these literatures.

Ureteroceles are diagnosed by imaging, these are ultrasonography, intravenous urography, and voiding cystourethrography to mention but a few, these detect the location, the size, affectation of the upper tracts, anatomy and function of the renal system and the clinical condition with other clinical features may lead to the choice of the imaging modality [2, 12, 15]. The index case was diagnosed following an IVU and abdominopelvic ultrasonography, these showed the location and form with associated affectation of the upper renal tract thereby conforming to these literatures.

Ureterocele are basically treated by medical and surgical approach, the index case was not an exception, and he had courses of antimicrobial agents for the treatment of the urinary tract infection and subsequently had surgical excision of the ureterocele, thereby conforming to these literatures.

CONCLUSION

The cases of recurrent urinary tract symptoms should be imaged by abdominopelvic ultrasonography and IVU to rule out possibility of ureterocele for immediate institution of management and to prevent consequent obstructive uropathy and nephropathy in this group of patients.

REFERENCES

- Chhetri, P. K., & Malla, S. K. (2010). Ureterocele—A case report. *Journal of college of Medical Sciences-Nepal*, 6(2), 38-41.
- Merlini, E., & Chiesa, P. L. (2004). Obstructive ureterocele—An ongoing challenge. *World journal of urology*, 22(2), 107-114.
- Chawla, A., Reddy, S., Natarajan, K., Thomas, J., & Sasidharan, K. (2008). Posterior urethral valve associated with orthotopic ureterocele. *Indian Journal of Urology*, 24(4).
- Xie, D., Klopukh, B., Nehrenz, G. M., & Gheiler, E. (2017). Ureterocele: Review of Presentations, Types, and Coexisting Diseases. *International Archives of Urology and Complications*, 3(1).
- Ribeiro, J. C. (2009). Unusual presentation of bilateral intravesical ureterocele. *Actas urologicas espanolas*, 33(3), 335-335.
- Penkoff, P., & Bariol, S. (2015). Urethral calculus originating from ureterocele and causing urinary retention. *ANZ journal of surgery*, 85(11), 892.
- Cichoń, W., Malik, L., Maciukiewicz, P., & Kotuła, J. (2014). Bilateral ureterocele in adult—a case report and review of literature. *Przegląd lekarski*, 71(8), 460-461.
- Vasey, G. M., & Michael, K. (2005). Ureterocele with duplicated collecting system: an antenatal and postnatal sonographic comparison. *Journal of Diagnostic Medical Sonography*, 21(1), 49-55.
- Emedicine: Ureterocele. (2020). Accessed December 19th 2020. www.emedicine.com/health.
- Hansen, W. F., Cooper, C. S., & Yankowitz, J. (2002). Ureterocele causing anhydramnios successfully treated with percutaneous decompression. *Obstetrics & Gynecology*, 99(5), 953-956.
- Sozubir, S., Lorenzo, A. J., Twickler, D. M., Baker, L. A., & Ewalt, D. H. (2003). Prenatal diagnosis of a prolapsed ureterocele with magnetic resonance imaging. *Urology*, 62(1), 144.
- Atta, O. N., Alhawari, H. H., Murshidi, M. M., Tarawneh, E., & Murshidi, M. M. (2018). An adult ureterocele complicated by a large stone: A case report. *International journal of surgery case reports*, 44, 166-171.
- Scultz, K., Toda, L.Y. (2016). Genetic basis of ureterocele. *Curr Genomics*, 17:62-69.
- Coplen, D., Duckett, J.W. (1995). The modern approach to ureterocele. *J Urol*, 153:169.
- Shokeir, A.A., Nijman, R.J.M. (2002). Ureterocele: an ongoing challenge in infancy and childhood. *BJU Int*, 90:777-783.
- Zerin, J. M., Baker, D. R., & Casale, J. A. (2000). Single-system ureterocele in infants and children: imaging features. *Pediatric radiology*, 30(3), 139-146.
- Mikuma, N., Adachi, H., Takatsuka, K., Yokoo, A., Wada, H., & Takahashi, A. (1996). Ectopic ureterocele with a horseshoe kidney in an adult. *International journal of urology*, 3(3), 243-244.
- Novak, M. E., Baum, N. H., & Gonzales Jr, E. T. (1977). Horseshoe kidney with multicystic dysplasia associated with ureterocele. *Urology*, 10(5), 456-458.
- Tsujimoto, Y., Oka, T., Noguchi, T., Fujii, T., Miyagawa, Y., Takano, Y., Kanno, N. (1999). A case of crossed fused kidney with simple ureterocele. *Nihon Hinyokika Gakkai zasshi. The Japanese journal of urology*, 90(12), 920-923.
- Sales, F. R. D., Dourado, G. A. C., Ribeiro, A. C. M., de Holanda Madeira Barros, H., Cristino, D. S., Castro, A. A. G., & Mesquita, F. J. C. (2020). Case Report: Prolapsed Ureterocele—A Differential Diagnosis of Urethral Cysts. *Case Reports in Urology*, 2020.
- Oueslati, A., Saadi, A., Chakroun, M., Zaghib, S., Bouzouita, A., Derouiche, A., & Chebil, M. (2020). Endoscopic meatotomy in the treatment of ureterocele: results in adult patients. *The Pan African Medical Journal*, 36.
- Brueziere, J. (1992). Ureterocele. *Ann Urol*, 26: 202 - 211.
- Idweini, Y. (2018). Infant's giant obstructive ureterocele causing bladder neck obstruction. *Urol Nephrol Open Access J*, 6(3), 104-106.
- Waihenya, C. G. (2008). Prolapsed single system ureterocele in a female adult: case report. *East African medical journal*, 85(4), 197-200.
- Kojima, Y., Hayashi, Y., Murayama, T., Sasaki, S., Mogami, T., Kohri, K. (1991). *Urol Int*; 63: 212-214.
- Ketabachi, A.A. (2003). Intravesical and unilateral ureterocele with ipsilateral kidney agenesis. *Journal of Kerman University of Medical Sciences*, 10: 1-9.

27. Dahnert, W. (2001). Radiology review Manual.
7th ed. Philadelphia. Wollers kluwer: *Lippincolt*

Williams and willians; 1002.