An adult ureterocele complicated by a large stone: A case report

Omar N. Atta, Hussein H. Alhawari *, Muayyad M. Murshidi, Emad Tarawneh, Mujalli M. Murshidi

Jordan University Hospital, Queen Rania Street, Amman 11942, Jordan

ARTICLE INFO

Article history:
Received 8 January 2018
Received in revised form 22 January 2018
Accepted 22 February 2018
Available online 28 February 2018

Keywords:
Case report
Cystolitholapaxy
Ureterocele
Ureteral calculus

ABSTRACT

INTRODUCTION: Ureterocele is a cystic dilatation of the lower part of the ureter. It is a congenital anomaly that is associated with other anomalies such as a duplicated system, and other diseases. It poses a great challenge owing to its numerous types and clinical presentations. Its incidence is 1 in every 4000 individuals. One of its presentations in the adult population is the presence of a stone, usually a solitary stone, inside the ureterocele.

CASE PRESENTATION: We are reporting a case of an adult ureterocele complicated by a large calculus; managed endoscopically with transurethral deroofing of the ureterocele followed by cystolitholapaxy. A literature review was also conducted.

DISCUSSION: The pathogenesis of ureteroceles is not well understood, however many proposed mechanisms exist with the incomplete dissolution of chawalla membrane being the most accepted one. The type of ureterocele and age at presentation will help guide the appropriate investigation and management, nevertheless certain goals of treatment should apply to all cases. Adult ureterocele is usually clinically silent but it may co-exist with other conditions such as a ureteral calculus and in these conditions it can be managed endoscopically.

CONCLUSION: Ureteroceles complicated by stones can be effectively managed with endoscopic resection or incision of the ureterocele coupled with stone removal, however long term follow up is required to monitor for hydronephrosis and iatrogenic vesicoureteric reflux.

© 2018 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Ureterocele is a congenital abnormality with a cystic dilatation of the lower part of the ureter, often associated with other anomalies like a stenotic ureteric orifice or a duplicated system along with other clinical sequelae. They could lead to various effects with regard to obstruction, reflux, continence, and renal function [1]. Ureteroceles may be intravesical (orthotopic) or extravesical (ectopic) [2]. It occurs in 1 out of 4000 individuals and it is 4 times more common in females than in males. It remains a challenge in terms of diagnosis and treatment due to its variable types and clinical presentations [1]. We are reporting a case of a left intravesical ureterocele and a calculus within the ureterocele in an adult patient who was treated with transurethral deroofing followed by cystolitholapaxy and stone removal. This work has been reported in line with the SCARE criteria [3].

2. Case report

A 58-year-old male patient with hypertension and non-insulin-dependent diabetes mellitus, who presented to our academic institute complaining of mild intermittent left flank pain for 1 year, recently associated with few episodes of gross hematuria and dysuria but no other complaints. He has no previous surgical history. Laboratory investigation showed Hg 12 gm/dl, creatinine 1.1 mg/dl, normal coagulation profile, uric acid 8.1 mg/dl, urinalysis on admission showed few white blood cells but no red blood cells or bacteria. Abdominal x-ray showed semi radiopaque stone in the area of the bladder and another semi radiopaque stone in right kidney (Fig. 1). Urinary tract computerized tomography (CT) scan without contrast showed a large stone at the left vesicoureteric junction measured 2.5 × 2 cm in cross-section with marked left hydronephroerephorosis. It also showed a right staghorn stone filling the right renal pelvis and lower calyces measuring 2.7 × 3.2 cm causing moderate right hydronephrosis (Figs. 2 and 3). Urinary tract ultrasonad showed the ureterocele and the stone within (Fig. 4). On the next day the patient underwent endoscopic operation under general anesthesia. The patient was put in lithotomy position, a 22F cystoscope was introduced into the bladder, the right ureteric orifice was identified and a large left intravesical ureterocele was
Serum creatinine has been within normal range. We are planning to repeat renal US at 6 and 12 months. We are also planning to do micturating cystourethrogram at about 12 months postoperatively to check for any evidence of vesicoureteral reflux.

3. Discussion

Ureteroceles are a cystic dilation of the distal aspect of the ureter that is located either within the bladder or spanning the bladder neck and urethra. It is a developmental anomaly and while its pathogenesis is unknown, several theories have been proposed, however the most accepted mechanism is failure in regression of the Chwalla membrane which is a membrane between the urogenital sinus and the developing ureteral bud.

The incidence of ureteroceles is 1:4000 individuals, occurring 4 times more in females with a slight predominance on the left side and 10% of the cases being bilateral.

Ureteroceles have diverse presentations ranging from life-threatening sepsis, renal failure, recurrent urinary tract infections (UTIs), to no symptoms at all being detected incidentally or by antenatal ultrasonography. These variable presentations are a reflection of the numerous types of ureteroceles, hence there are multiple classification systems such as the Stephens classification.

seen with no apparent left ureteric orifice noticed which goes with the fact that most intravesical ureteroceles have stenotic orifices (Figs. 5 and 6), then a 24F resectoscope was inserted, deroofing of the ureteroceles with cutting current was performed and the stone was visualized and nudged with the loop into the bladder (Figs. 7–10). Cystolitholapaxy was performed at the same session and stone fragments were removed (Figs. 11 and 12). The operation was concluded with insertion of a 3-way 20F Foley catheter and irrigation was started. The Foley catheter was removed the next day and postoperative abdominal x-ray and urinary tract CT scan without contrast showed no residual stone fragments in the bladder (Figs. 13 and 14).

Repeat renal and bladder US done at about three months postoperatively showed stable mild left hydroureterohydronephrosis (Figs. 15 and 16).
which depends on the size and location of the ureteric orifice or the functional based classification by Churchill, however due to their complexity these systems have gained less popularity and more simplified system was established by the American Academy of Pediatrics is more frequently used which classifies ureteroceles into intravesical (orthotopic) ureterocele or ectopic (if part of the ureterocele extends to the bladder neck or urethra permanently) [7].

According to Stephens classification, intravesical ureteroceles may be stenotic (40%) or non-obstructive (5%), while ectopic ones may be sphincteric (40%), sphincterostenotic (5%), cecoureterocele (5%) or blind (5%) [8]. In most series 60–80% of ureteroceles are ectopic as opposed to intravesical and 80% of ureteroceles are associated with the upper moiety of a complete duplication [9], this is more evident in the pediatric age group, but when found in adults they are usually intravesical of single system [6].

In keeping with previous arguments, the clinical presentations will vary with age; in pediatric age group the presenting condition is usually recurrent UTIs or urosepsis, incontinence, failure to thrive, urinary tract calculus, abdominal mass, bladder outlet obstruction and vaginal or urethral prolapse, while in the adult population the diagnosis is usually made incidentally, sometimes it presents with intermittent flank pain, recurrent urinary tract infection or calculus [10,11].

The diagnostic imaging starts with ultrasound due to its availability and non-invasive nature, it is also an excellent modality in this condition as it can show the cystic dilatation in the pos-
Fig. 9. The stone is now well inside the bladder cavity.

Fig. 10. The left ureteric orifice after the deroofing.

Fig. 11. Removal of stone fragments with punch forceps.

Fig 12. The removed stone fragments.

terior wall of the bladder and sometimes it can provide valuable information regarding the duplicity of the system [9].

Intravenous urography (IVU), although less commonly performed nowadays may show poor function of the affected side with delayed excretion or no excretion at all, however if the renal parenchyma retains some function a characteristic cobra head sign can be seen due to the opacified urine inside the intravesical ureterocele surrounded by halo sign produced by the wall of the ureter, it is worth mentioning that IVU can still be of importance in cases of confusing anatomy [8].

Voiding cystourethrogram is an essential part of the evaluation as it will detect the presence of vesicoureteral reflux, moreover renal nuclear imaging shows the function of renal tissue [8].

The numerous clinical presentations, the type of the ureterocele and the age of the patient are all many clinical variables that will guide the appropriate choice of management as there is no single method suffices for all cases and thus the management should be individualized, nevertheless the goals of management should be applied to all cases and these include maximal preservation of renal function, prevention and treatment of vesicoureteral reflux (VUR), unobstructed drainage of all functioning parenchyma, prevention of bladder outflow obstruction, maintaining continence and the removal of any potential source of infection [6].

In general, ureteroceles may be treated with endoscopic incision, upper pole partial nephrectomy, and complete reconstruction at the bladder level or non-operative (conservative) treatment [9].

With regards to our case, the ureterocele was complicated with
a ureteral calculus which is not uncommon according to the literature as the incidence of this particular condition lies between 4% and 39% and most stones are solitary and are formed due to stasis and/or infection [10,12]. According to Chtourou et al. who performed a study about stones in ureterocele in 20 adult patients who were all treated by endoscopic horizontal meatotomy with stone fragmentation and extraction, concluded that endoscopic meatotomy is easy to perform and gives good results and the associated stones constitute an additional argument in favor of endoscopic treatment [13]. There are also multiple case reports in which many of these conditions were managed with endoscopic resection. Endoscopic treatment includes transurethral puncture and transurethral incision; these are applicable mainly to the intravesical types and may be curative in up to 90% of cases [14,15], however these patients required long-term follow-up to monitor renal function, symptoms and occurrence of vesicoureteric reflux [14].

4. Conclusion

Ureteroceles represent a clinical challenge in term of diagnosis and management due to their variable presentations and types, thus treatment has to be individualized to each case and its co-existing pathology. Ureteroceles which are complicated by stones can be effectively managed with endoscopic resection but require long term follow up.

Conflicts of interest

The authors declare that no conflicts of interest exist.

Sources of funding

The authors declare that no funding sources were used.

Ethical approval

Since this was just a case report and patient's confidentiality was assured, there was no need for ethical approval based on our institution regulations.
Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

1. Omar N. Atta: He was a part in the primary inpatient managing urology team. He also participated in data collection, data analysis and interpretation, and writing the paper.
2. Hussein H. Alhawari: Consulting nephrologist. He also participated in literature review, writing the paper and review of the final manuscript.
3. Muayyad M. Murshidi: Participated in data collection, data analysis and interpretation, and writing the paper.
4. Emad Tarawneh: Consulting radiologist. He also participated in data collection and literature review.
5. Mujalli M. Murshidi: Operating surgeon. He also participated in data collection and literature review.

Guarantor

All above authors accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

References

[1] F.D. Stephens, Etiology of ureteroceles and effects of ureteroceles on the urethra, Br. J. Urol. 40 (1968) 483–487.
[2] J.C. Sander, A.N. Bilgutay, I. Stanaszek, C.J. Oh, N. Janzen, E.T. Gonzales, et al., Outcomes of endoscopic incision for the treatment of ureteroceles in children at a single institution, J. Urol. 193 (2015) 662–666, http://dx.doi.org/10.1016/j.juro.2014.08.095 (PMC free article) [PubMed].
[3] R.A. Agha, A.J. Fowler, A. Saetta, I. Bara, S. Rajmohan, D.P. Orgill, The SCARE Group. The SCARE Statement: consensus-based surgical case report guidelines, Int. J. Surg. Part A 36 (December) (2016) 396.
[4] P.W. Jeffrey, Embryogenesis of ureteral anomalies: a unifying theory, Aust. N. Z. J. Surg. 58 (1988) 631–638.
[5] K. Schultra, L.Y. Toda, Genetic basis of ureteroceles, Curr. Genomics 17 (2016) 62–69, http://dx.doi.org/10.2174/138920291666151014222815 (PMC free article) [PubMed].
[6] A.A. Shokeir, R.J.M. Nijman, Ureteroceles: an ongoing challenge in infancy and childhood, BJU Int. 90 (2002) 777–783, http://dx.doi.org/10.1046/j.1464-410X.2002.02998.x (PubMed).
[7] K.I. Glasberg, V. Braren, J.W. Duckett, et al., Suggested terminology for duplex systems, ectopic ureters and ureteroceles. Reports of the Committee on Terminology, Nomenclature and Classification. American Academy of Pediatrics. J. Urol. 132 (1984) 1153–1154.
[8] E. Merlini, P.L. Chiesa, Obstructive ureterocele-an ongoing challenge, World J. Urol. 22 (June) (2004) 107–114.
[9] D. Coplen, J.W. Duckett, the modern approach to ureteroceles, J. Urol. 153 (1995) 169.
[10] M.S. Murshidi, Orthotopic and ectopic ureteroceles in children, Int. Urol. Nephrol. 22 (1) (1990) 45–56 (PMID:2380002).
[11] A. Muhammed, M.Y. Hussaini, B. Ahmad, M.N. Hyacinth, K.D. Garba, Ureteroceles in adults: management of patients in Zaria, Nigeria, Arch Int. Surg. 2 (2012) 24–28.
[12] A. Dominici, F. Travaglini, M. Maleci, V. di Cello, M. Rizzo, Giant stone in a complete duplex ureter with ureteroceles, Urol. Int. 71 (2003) 336–337.
[13] M. Chitourou, S. Sallami, H. Rekik, M.V. Binous, I. Khaiar, A. Horncham, Ureteroceles in adults complicated with calculi: diagnostic and therapeutic features. Report of 20 cases, Prog. Urol. 12 (December (6)) (2002) 1213–1220.
[14] G. Monfort, G. Morisson-Lacombe, M. Coquet, Endoscopic treatment of ureteroceles revisited, J. Urol. 133 (1985) 1031–1033.
[15] B. Blyth, G. Passerini-Glazel, C. Camufllo, H.M. Snyder, J.W. Duckett, Endoscopic incision of ureteroceles: intravesical versus ectopic, J. Urol. 149 (1993) 556–559.

Open Access
This article is published Open Access at sciedirect.com. It is distributed under the IJSSCR Supplemental terms and conditions, which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.