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Endourology

Single-system orthotopic ureterocele with calculus masquerading as a bladder tumour – A case report and review of literature

Florian Löcherbach*, Pirmin Wolfsgruber, Stephen Wyler, Maciej Kwiatkowski

Cantonal Hospital Aarau, Department of Urology, Tellstrasse 25, 5001, Aarau, Switzerland

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Introduction

As cystic dilations due to a congenital wall weakness of the distal ureter,[1] ureteroceles tend to be found in autopsy with an incidence of 1/500 to 1/4000. According to literature solitary stones within such a single-system ureterocele are quite common with a prevalence of about 4%–39%.[2] However the diagnosis of such a situation is largely based on typical diagnostic signs such as the cobra-head-sign or the changing size of the ureterocele during sonographic evaluation as well as the direct proof of the ureterocele in cystoscopy and MRI-/CT-imaging. Still there are rare situations in which the diagnostic findings aren't as obvious to interpret as one might think. Such a case which is of interest to each clinically working urologist we present at hand. As to our knowledge there has only one such case been reported in literature so far.[3]

Case presentation

A 59-year old Patient was transferred to us by an outward Radiology after diagnosing a calcified right-sided bladder tumour (approx. 2cm in diameter) with a consecutive ureteral obstruction and a therefore massively dilated right-sided distal ureter in a CT-scan. The patient himself initially consulted his primary physician due to right-sided lower abdominal pain so that the formerly mentioned CT-scan was performed (see Figs. 1 and 2). Further investigation by cystoscopy within our clinic confirmed that the tumour was highly suspicious of being a urothelial carcinoma. Therefore a transurethral resection of the tumour was planned. Unfortunately no urinary cytology was acquired.

Intraoperatively cystoscopy once again confirmed the highly suspicious findings to most likely be an urothelial carcinoma and therefore transurethral resection was begun. However, after the first slices were made, the tumour revealed itself to be an intravesically prolapsed ureterocele containing a calculus instead of being a urothelial carcinoma. The calculus was extracted under slight bipolar resection of the ureterocele-wall as well as lithotripsy and finally a Double-J-Stent was implanted.

In the postoperative follow-up period the Double-J-Stent was removed 4 weeks after surgery and sonographic as well as CT-based controls showed unsuspicious renal drainage on the right side with a residual dilatation of the distal right ureter and pelvis.

Histological analyses of the resected tissue revealed no malignancy and the analysis of the extracted calculus showed it to be 100% Calcium-Oxalate-Monohydrate (Whewellit).

Discussion

As the incidence of ureteroceles in adults is very low and diagnosis is often delayed, there is scarcely literature about an adequate diagnostic algorithm as well as the proper treatment available.[4]

Most recommendations derive from pediatric surgery and suggest an individualized diagnostic approach with e.g. MRI/CT-scans, cystoscopy, cystography, sonography and isotope renogram fitted separately for each patient as there is no “one fits them all”-approach.[4] This is of special importance as typical diagnostic signs such as the cobra-head-sign or the changing size of the ureterocele during sonographic evaluation might not be present in every patient. As our case at hand shows, even a combined diagnostic approach with CT-scan, sonography and cystoscopy can lead each urological practitioner astray.

Depending on the diagnostic findings further treatment should be equally fitted for each patient and focus on the prevention of further complications such as vesico-ureteral reflux, recurrent UTIs and loss of

* Corresponding author.
E-mail address: florian.loecherbach@gmx.de (F. Löcherbach).

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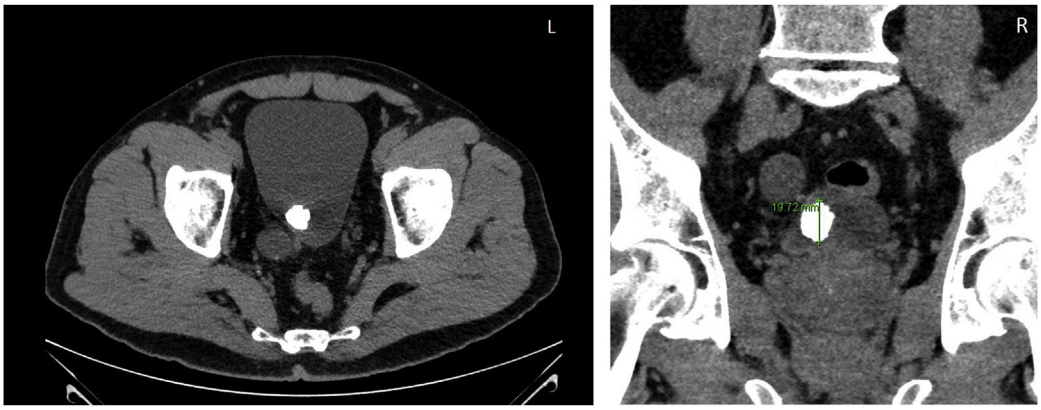

Fig. 1. Transversal (left) and frontal (right) slices of the initial CT-scan. The calcified tumour as well as the massively dilated right Ureter can be seen at the right-sided bottom of the bladder, both measuring about 2 cm in diameter.

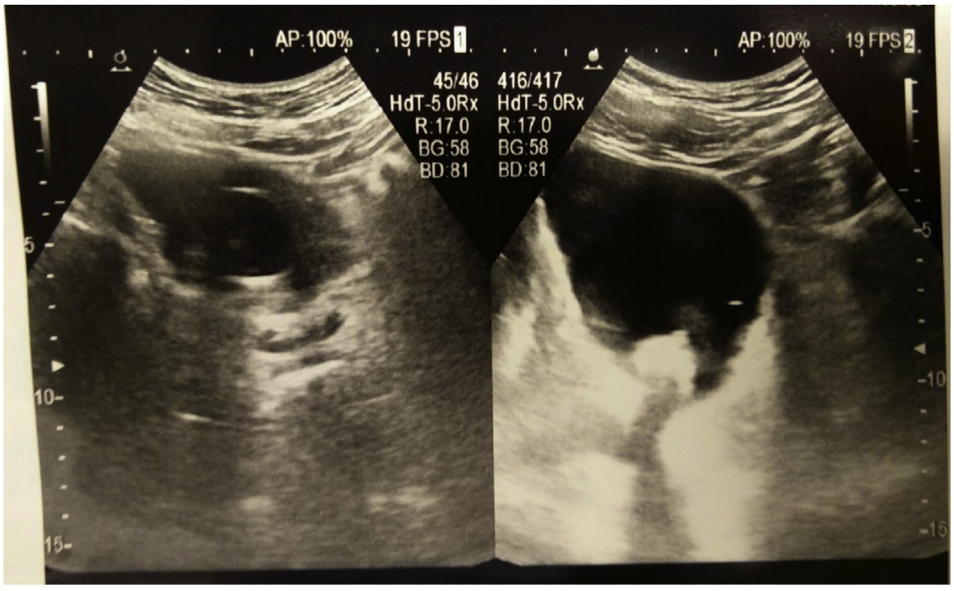

Fig. 2. Preoperative transversal (left) and sagittal (right) ultrasound imaging. A calcified tumour is to be found on the right-sided bottom of the bladder. There was no typical cobra-head-sign or a changing size of the tumour indicating a ureterocele present during the examination.

renal function. In more complex cases with duplex renal collecting systems including blindly ending ureters, hypoplastic renal segments which are drained by the ectopic ureter or infrasphincteric ureteral orifices ablative surgical approaches including nephrectomy, hemine-phrectomy and ureterectomy are mostly considered to be the therapy of choice.[4]

However, it is commonsense, that ureteroceles in adults tend to be less complicated and are more likely to be small and single-system with orthotopic ureteral orifices. The proper treatment, as it is in children with single-system orthotopic ureteroceles as well, is accordingly transurethral incision/resection of the ureterocele[1,4,5] and stenting the ureter as we did with our patient.

Still the main complication of this technique seems to be vesico-ureteral reflux. We therefore recommend regular follow-up by ure-throcystography, which we will perform on our patient during one of his next consultations. Furthermore follow-up after transurethral inci-sion of a ureterocele should include CT-urographic controls as well as sonographic controls.

As it comes to the long-term outcome after endoscopic de-roofing or puncture of orthotopic and even complicated, ectopic, ureteroceles re-cent publications suggest that the paradigm shifts slowly towards en-doscopic treatment as it is safe and long-term effective. Jawdat et al.[5] just published a summary of 78 cases which had a median follow-up of 12 years after endoscopic puncture of orthotopic as well as ectopic ureteroceles. It shows that in the group of patients with orthotopic ureteroceles who had undergone an endoscopic puncture of the ur-eterocele, there was scarcely additional surgery necessary. Within the group of patients suffering of an ectopic ureterocele more patients re-quired secondary surgery, but there still was no statistical significance between those two groups ($p = 0.716$) supporting Chowdhary et als[4] statement that even those patients can be managed endoscopically. Furthermore a significant number of patients who suffered of a new onset of vesico-ureteral reflux after the procedure showed spontaneous regression.

According to Jawdat and Chowdhary[4,5] non-functional, undrained, renal segments as a complication of ectopic ureteroceles should no longer be an indication for major surgery such as (partial-)ne-phrectomy.

Conclusion

The case presented at hand shows that due to its rareness ureter-oceles in adults pose a diagnostic and therapeutic challenge to each urologist and might be misinterpreted as a more common pathology such as a urothelial carcinoma of the bladder. The diagnostic algorithm should therefore be multimodal including sonography, CT-/MRI-scans,

cystoscopy and in some cases an isotope renogram and cystography. The therapy of choice for orthotopic single-system ureteroceles is transurethral incision, de-roofing and Double-J-Stenting of the ureter. As it comes to more complicated, ectopic, ureteroceles endoscopic incision should be evaluated as first-line treatment as well as the latest data suggest that major surgery even in these cases is rarely necessary and can therefore be avoided.

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Declarations

The authors declare that there is no conflict of interests regarding the publication of this paper.

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.eucr.2018.09.002.

References

1. Harris CR, Alwaal A, Yang G, Eisenberg ML, Breyer BN. *Adult Ureterocele Presenting with Ureteral Obstruction and Urosepsis during Pregnancy.* 2016; 2016:6.
2. Mandal S, Goel A, Kumar M, Goyal NK, Sankhwar SN. Spontaneous erosion of ureterocele wall by stone: rare images. *Urol Oktober.* 2012;80(4):e43–e44.
3. Thiel DD, Petrou SP, Broderick GA. Orthotopic ureterocele masquerading as a bladder tumor in a woman with pelvic pain. *Int Braz J Urol Dezember.* 2005;31(6):549–551.
4. Chowdhary SK, Kandpal DK, Sibal A, Srivastava RN. Management of complicated ureteroceles: different modalities of treatment and long-term outcome. *J Indian Assoc Pediatr Surg.* 2014;19(3):156–161.
5. Jawdat J, Rotem S, Kocherov S, Farkas A, Chertin B. Does endoscopic puncture of ureterocele provide not only an initial solution, but also a definitive treatment in all children? Over the 26 years of experience. *Pediatr Surg Int Mai.* 2018;34(5):561–565.