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PEDIATRIC UROLOGY

Original Article

Transurethral incision of ureteroceles in paediatric age group

Pediyatrik yaş grubunda üreterosellerin transüretral insizyonu

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ABSTRACT

Objective: Ureteroceles are a great clinical challenge because of variations in anatomy and clinical presentations. We present our experience with primary transurethral incision of ureteroceles in children.

Material and methods: Data of thirteen children managed for ureterocele from 2009 to 2016 was retrospectively analyzed with respect to age, sex, clinical presentation and symptomatology, type and localization of ureterocele, investigations, surgical management and follow-up.

Results: A total of 13 patients with ureteroceles were managed. There were 7 males and 6 females. Six were neonates with antenatal diagnosis of ureteroceles. Five patients presented with urinary tract infection and two were diagnosed during ultrasound for abdominal pain. The ureteroceles were on the right side in 7 patients and left in 6 patients. Six patients had a duplex system-five on right side and bilateral in one. Two patients had ureteroceles in solitary kidney. Four patients had associated hydronephrosis and hydroureter and two had only hydronephrosis alone. One patient had bilateral grade III reflux in the bilateral lower moieties of the patient with bilateral duplex system. Two patients had poorly functioning kidney on radionuclide scan. All patients underwent cystoscopic incision of the ureteroceles. Eleven had intravesical ureteroceles and two had large caeco-ureteroceles. Two patients required ureteric reimplantation during follow-up.

Conclusion: Though the approach of managing a patient with ureterocele should be individualized, transurethral incision remains valuable as a primary intervention with regular follow up. It may even prove to be the only intervention required in most of the patients.

Keywords: Duplex moiety; transurethral incision; ureterocele.

ÖZ

Amaç: Üreteroseller anatomi ve klinik belirtilerde değişkenlikler nedeniyle klinik açıdan zorlu bir patolojidir. Bu çalışmada çocuklardaki üreterosellerde primer transüretral insizyonla ilgili deneyimlerimizi sunuyoruz.

Gereç ve yöntemler: 2009 ile 2016 yılları arasında tedavi edilen 13 çocuğun verileri yaş, cinsiyet, klinik belirtiler, semptomatoloji, üreteroselin tipi ve yerleşimi, cerrahi tedavi ve takip açısından geriye dönük incelenmiştir.

Bulgular: Üreteroselli toplam 13 hasta (7 erkek ve 6 kız çocuk) tedavi edilmiştir. Altı hasta doğum öncesi üreterosel tanısı konmuş yenidoğandı. Beş hasta idrar yolu enfeksiyonu belirtileri göstermiş ve ikisine karın ağrısı nedeniyle yapılan ultrason sırasında tanı konmuştu. Üreteroseller 7 hastada sağ, 6 hastada sol taraftaydı. Altı hastanın beşinde sağ ve birinde her iki tarafta çift toplayıcı sistem mevcuttu. İki hastada üreteroselli tek böbrek vardı. Dört hastada üreteroseller hidronefroz ve hidroüreterle birlikte olup iki hastada yalnızca hidronefrozla üreterosel birlikteliği saptandı. Bir hastada her iki tarafta çift taraflı toplayıcı sistemle birlikte bilateral III. derece reflü mevcuttu. İki hastanın radyonüklit taramasında renal işlev bozukluğu saptandı. Hastaların hepsinde sistoskopla girilerek üreterosellere transüretral insizyon yapıldı. İki hastada intravezikal üreteroseller ve diğer iki hastada büyük çekoüreterosel mevcuttu. İki hastada takip sırasında üreter reimplantasyonuna gerek duyuldu.

Sonuç: Üreterosel saptanan hastalarda tedavinin bireyeselleştirilmesine rağmen; düzenli takipli birincil girişim olarak transüretral insizyon değerini korumaktadır. Hatta hastaların çoğunda gerekli tek girişim olduğu bile kanıtlanabilir.

Anahtar Kelimeler: Çift toplayıcı sistem; transüretral insizyon; üreterosel.

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Introduction

Ureterocele or cystic dilatation of the terminal ureter are a clinical challenge because of variations in anatomy and clinical presentations.[1,2] Ureteroceles occur in association with single or duplex collecting systems and may be intravesical (orthotopic) or extravesical (ectopic).[2] Vesicoureteric Reflux (VUR) may occur in upper moiety or combination of moieties. Presentation can be symptomatic with urinary tract infection (UTI) or asymptomatic (hydronephrosis).[2]

The optimal approach of management remains controversial. However, the management goals remain the same -maximal preservation of renal function, prevention and treatment of VUR, non-obstructed drainage of all functioning parenchyma, prevention of bladder outlet obstruction or any bladder wall defects, maintaining continence and the removal of any potential source of infection thereby minimizing surgical morbidity.[1,2] Transurethral incision of the ureterocele is an easy, relatively non-invasive and attractive option for primary management of patients with ureterocoles.[2]

We present data of thirteen children with ureteroceles who were primarily managed with transurethral incision.

Material and methods

This is a retrospective observational study involving thirteen children with ureteroceles managed at our institution from 2009 to 2016. Data records of these thirteen children were reviewed and analyzed with respect to age, sex, clinical presentation and symptomatology, type and localization of ureterocele, investigations done, surgical management and follow-up. Written and informed consent was taken from parents of all these patients.

All patients underwent abdominal ultrasonography (USG), voiding cystourethrography (VCUG) and renal radionuclide scans followed by cystoscopic incision of the ureterocele. The incision was made at the infero-medial aspect of the ureterocele and on the dependent portion of large ureteroceles. USG and cystoscopy were repeated after 6 weeks to confirm adequacy of incision. At follow up, six-monthly USG, annual VCUG (for secondary reflux) and radionuclide scans were done. Patients with persistent hydronephrosis and hydroureter and with gradual decrease in glomerular filtration rate (GFR) and renal function underwent reconstructive surgery.

Results

Demographic details

Thirteen children with ureterocele were treated over a 6 year period. There were 7 boys and 6 girls. Six patients were diagnosed antenatally. The others presented between 1 month to 9 years.

Symptomatology

Six patients were managed in the neonatal period in view of antenatal diagnosis of ureterocele. Five patients presented with urinary tract infections and two were diagnosed at abdominal USG for abdominal pain.

Associated anomalies

One patient had bilateral palpable undescended testes; one had a rectovestibular fistula and the patient with solitary kidney had

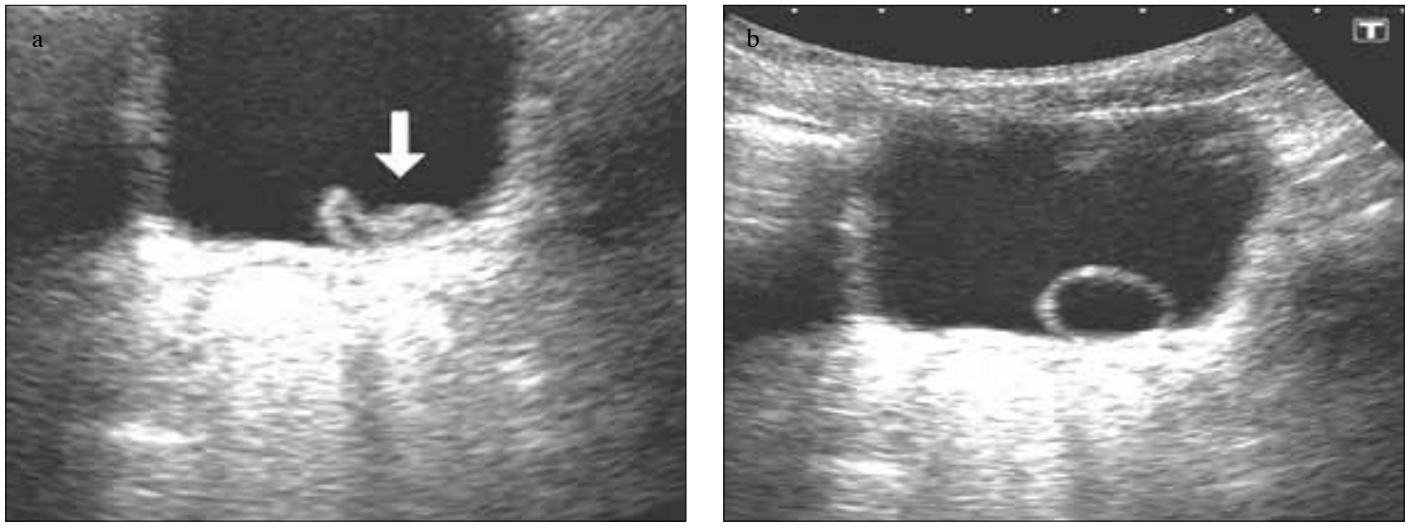

Figure 1. a, b. (a) Ultrasound image of a simple left ureterocele in a normal kidney during filling phase. (b) Ultrasound image of a simple left ureterocele in a normal kidney during emptying phase

Turk J Urol 2017; 43(4): 530-5
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an associated coarctation of aorta and contralateral palpable undescended testis.

Investigations

USG, VCUG and radionuclide scans were done for all patients. Ureterocele was diagnosed on ultrasonography in 11 patients (Figure 1a and b); six being post-natal confirmations of antenatal findings. Ureterocele was on the right side in 7 patients and left in 6 patients. 6 patients had a duplex system-5 on right side and bilateral in one. Four patients had associated hydronephrosis and hydroureter and two had only hydronephrosis alone. Two patients had ureteroceles in solitary kidney.

Five patients underwent Intravenous pyelography (IVP) for confirmation of diagnosis (Figure 2). VCUG revealed bilateral grade III reflux in the bilateral lower moieties of the patient with bilateral duplex. No VUR was detected in other patients. Two patients had poorly functioning kidney on radionuclide scan-one in a patient with solitary kidney and the in the upper moiety of the patient with duplex. In the other eleven patients, scan showed bilateral good functioning renal units.

Management

Eleven patients had intravesical ureteroceles. Two patients had large caeco-ureterocele. All patients underwent cystoscopic incision of the ureteroceles using a Bugbee electrode (Figure 3).

Follow-up

The patients were assessed after 6 weeks with a repeat USG and check cystoscopy (Figure 4). Patients were followed up with six-monthly USG and annual VCUG and radionuclide studies. Two patients required ureteric reimplantation in view of persistent hydronephrosis and hydroureter with gradual decrease of GFR and renal function. During follow-up, the patient with bilateral duplex having grade III VUR in the lower moieties had complete resolution of VUR on expectant management and USG showed no evidence of hydronephrosis or hydroureter. Renal scan also showed good renal function bilaterally.

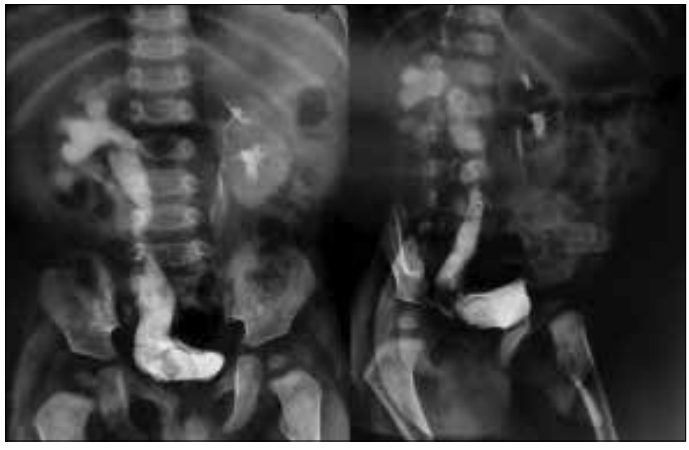

Figure 2. Intravenous pyelogram image of a patient with bilateral duplex systems showing ureterocele

Discussion

Ureterocele is a cystic dilatation of the distal intramural ureter which results in obstruction of urine flow, dilation of the ureter and renal pelvis and loss of renal function.[3] It was first classified in 1954 by Ericson[4] as either simple or ectopic depending on the

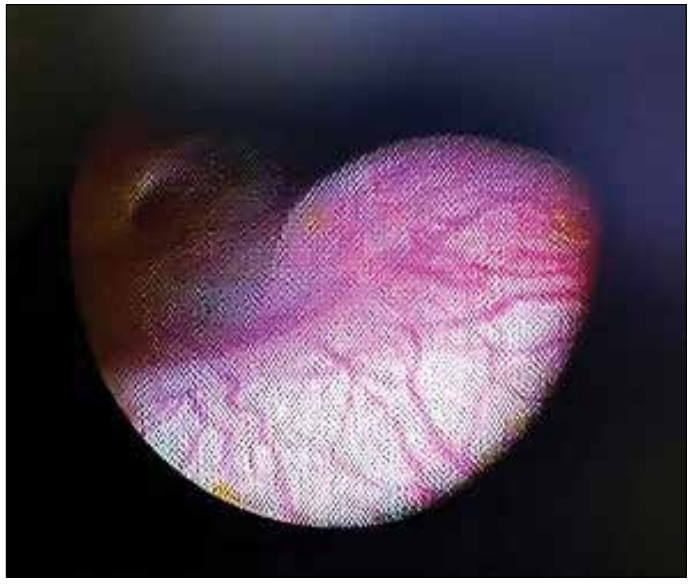

Figure 3. Cystoscopic image of a patient showing ureterocele in right duplex system

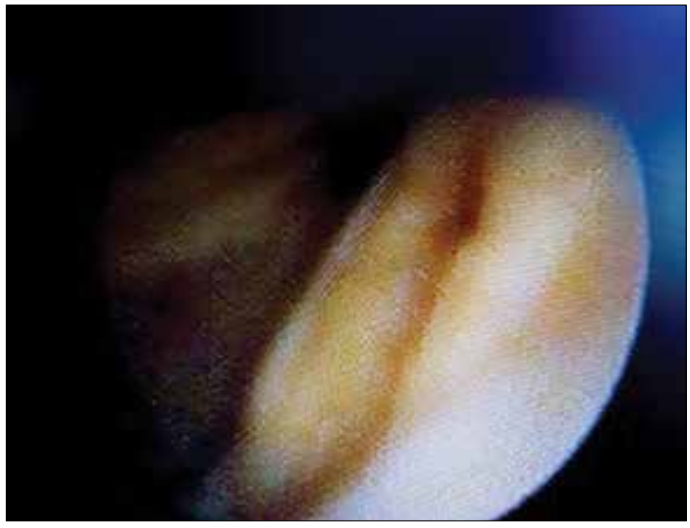

Figure 4. Cystoscopic image of the same patient after 6 weeks of transurethral incision showing the collapsed wall of the previously incised ureterocele

location of the ureteric orifice. The orifice in a simple non-duplex ureterocele is on the trigone of the bladder while the orifice in an ectopic ureterocele is either at the bladder neck or the posterior urethra.[1] In 1968, Stephens[5] classified ureteroceles based on the size and location of the ureteric orifice into four categories: Stenotic (narrow orifice within the bladder), sphincteric (wide orifice within the internal sphincter), sphinctero-stenotic (narrow orifice within the internal sphincter) and caeco-ureterocele (blind-ending ureterocele extending down the urethra). Churchill et al.[6] have classified ureteroceles based on the total amount of renal tissue or renal units at risk of damage from obstruction or high-grade reflux - Grade 1: ureterocele segment only affected; Grade 2, both segments of one kidney affected and Grade 3: both kidneys affected. The classification established by the American Academy of Pediatrics[7] as intravesical (entirely within the bladder) or ectopic (some portion is situated permanently at the bladder neck or in the urethra) is the most widely used currently.

Ureteroceles are known to occur in approximately 1 in 4000 children and are most common in Caucasians.[1,3] Ureteroceles are 4-6 times more common in females which holds true in adult population too.[3] There is a slight predominance on the left side, with approximately 10% bilateral disease.[3] An ectopic ureterocele has been reported to be four times more common than an intravesical ureterocele.[8] Eighty percent ureteroceles in infants are associated with the upper pole moiety of a duplex system and sixty percent of these have an ectopic location.[1,3,9] The upper pole in these cases tends to be dysplastic or poorly functioning.[3] Single-system ectopic ureteroceles are less common and are most often found in males.[1,3]

Studies have described series of family cases of ureterocele in both single and double systems, suggesting the possibility of genetic predisposition.[3,10] The etiology is unknown. Chwalla[11] had first suggested that ureterocele develops due to intrauterine obstruction of a membrane (the Chwalla's membrane). However, this didn't explain the development of the ureteroceles with a patulous ureteric orifice in the urethra. Tanagho[12,13] suggested that ureterocele formation may be related to the timing of absorption of the mesonephric duct into the urogenital sinus.

Studies in literature state that 90% of the patients with ureterocele are diagnosed before the age of 3 years.[14,15] There is an increasing number of antenatally diagnosed patients with ureteroceles. In young children, the most common clinical presentation is a UTI. Some may have an insidious clinical course and may present as failure to thrive or abdominal or pelvic pain. Sometimes, an obstructed renal unit may result in a palpable abdominal mass. An ectopic ureterocele may prolapse thereby causing urethral obstruction.[1] In girls, a prolapsed ureterocele may present as a vaginal mass.[16] A large intraurethral ectopic ureterocele which may render external urinary sphincter lax and inefficient can cause incontinence. Haematuria is an infrequent complaint. In older children and adults, the ureteroceles are simple with a normal or mildly dilated single collecting system. They are incidentally discovered with no symptoms; however, most children may present with symptoms of UTI. Stasis and infection predispose the patient to stone formation in the ureterocele and upper urinary tract.[1]

The ureterocele varies in size from a tiny cystic dilatation of the submucosal ureter to that of a large balloon that almost completely fills the bladder.[1] Histologically, the wall of the ureterocele is composed of attenuated smooth muscle bundles and fibrous tissue.[1] It is covered by vesical mucosa and lined with ureteric mucosa.[17]

The whole urinary system should be evaluated. USG is easy to perform, non- invasive and probably the best imaging modality for making the diagnosis.[18] It is the recommended screening method after the first urinary tract infection.[14] VCUG diagnoses ureterocele and detects VUR.[14] Reflux occurs into the ipsilateral lower pole in almost half of the patients but in 25% of patients, the contralateral system is also affected.[14] VCUG also helps in following up patients with preoperative VUR diagnosis or to detect newly forming refluxes after endoscopic intervention.[14] IVP, though not the currently preferred method, displays anatomical pathology characteristics and the non-functioning upper poles and helps to determine the management protocol of the surgical procedure.[14] Radionuclide studies help to assess the distribution of function in the duplex kidney and are also helpful for detecting and follow up of scarred tissue and non-functioning upper poles.[14,19]

The management of each patient with ureterocele must be individualized based on the clinical and pathophysiological characteristics of each patient.[1,14] Factors influencing the choice of management include the presentation of the patient, age of the patient, type of ureterocele, function of each renal segment if associated with a duplex system, the presence or absence of reflux in other segments and/or associated infection.[1,20]

The aim of management of ureteroceles includes maximal preservation of renal function, prevention and treatment of VUR, non-obstructed drainage of all functioning parenchyma, prevention of bladder outlet obstruction or any bladder wall defects, maintaining continence and the removal of any potential source of infection thereby minimizing surgical morbidity.[1,6,21]

The timing of surgical intervention is critical and controversial.[1] Primary transurethral ureterocele incision/puncture in neonate age-group helps to relieve the obstruction of both the involved segment and the lower pole ureter and bladder neck as well.[1,2] This allows recovery of function in the involved segment

Turk J Urol 2017; 43(4): 530-5
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and prevents infection.[1,2] Even if it does not result in a cure, it allows the delay of definitive treatment until the child is older, when this decompressed system could be reconstructed more easily.[1] After ureterocele incision, all patients need to be followed up for VUR, UTI and hypertension.[22] In a recent study, the authors have suggested early endoscopic incision as the first-line treatment of ectopic ureterocele.[23]

However, some authors are against this early endoscopic treatment in neonatal period. [1] They argue that this approach rarely constitutes definitive therapy and does not improve overall renal function significantly; it may also commit the patient to future unnecessary lower tract reconstruction.[1,22] Husmann et al.[24] recommends its routine endoscopic ureterocele incision in neonates with high-grade VUR but avoided this approach in patients with no pre-existing VUR.

Nevertheless, endoscopic ureterocele incision is simple, minimally invasive, requires only a short anaesthesia and can often be undertaken as an outpatient procedure. [1] Most studies in literature suggest that endoscopic treatment of intravesical ureteroceles is likely to be successful and definitive.[1,20,23,24] Blyth et al.[20] have reported that endoscopic ureterocele incision was definitive, enabling treatment in 93% of intravesical ureteroceles. Pfister et al.[23] have reported that endoscopic treatment alone proved effective in 14 of 16 intravesical ureteroceles in neonates. Thus, transurethral incision can be the only required intervention in most patients.[2,22] These patients should be kept on regular follow-up.

The main aim, however, remains the same-upper tract preservation.[1] The upper tract can be preserved by endoscopic incision with or without surgical reconstruction of the urinary tract. [1] The various surgical options are upper pole heminephrectomy without lower tract reconstruction, expectant management alone, lower tract reconstruction alone or with total nephroureterectomy (in non-functioning renal units).[1] It is difficult to compare these methods because they are appropriately applied to patients with different clinical presentations.[1]

In upper pole heminephrectomy without lower tract reconstruction: The upper pole segment is removed and the ureterocele is aspirated from above.[1] Patients with ureteroceles who have a non-functioning upper segment and low-grade or no VUR are amenable to this approach. [1]

Upper pole heminephrectomy with lower tract reconstruction: This combined approach or complete reconstruction is indicated in patients who clearly have no function of the upper pole system but high-grade reflux into the ipsilateral lower pole ureter or contralateral ureter.[1,19] This operation is technically challenging, particularly in neonates; reimplantation being difficult.[1] Therefore, some surgeons recommend a two-stage approach.[1,25] King et al.[26] have reported improved success with delayed ureteric reimplantation after initial ureterocele decompression with heminephrectomy.

Upper tract preservation: Patients with ureteroceles with some function of the upper pole segment are best managed with lower tract reconstruction alone - the ureterocele is dissected off the bladder till the point where it joins the lower pole ureter.[1] After that, both the ureters are dissected as a unit, tapered if required and reimplanted submucosally.[1,17] In patients with severe anatomical abnormalities of the bladder or ureter, ureteropyelostomy or uretero-ureterostomy may be required.[1,26,27] Ureteropyelostomy is preferred as it prevents the yo-yo reflux which can detrimentally affect urinary drainage and may lead to stasis, infection and ureteric dilatation.[1]

Expectant management: Non-operative management has been proposed in asymptomatic neonates with antenatally detected ureteroceles.[1,28,29] Rickwood et al.[28] and Jee et al.[29] have reported expectant management of antenatally detected ureteroceles for a period of 2.3 years and 24 months respectively. However, further data on the natural history of these ureteroceles is required before this can be suggested as a reasonable option.[1] In adults, ureteroceles may often be incidental findings that require no treatment.[1] Sometimes, they may contain a small calculus which can be extracted endoscopically by ureteric meatotomy.[1] They are less likely to have postoperative reflux in the incised ureterocele.[1]

Total nephroureterectomy: Excision of the ureterocele with complete nephroureterectomy is indicated in children with massive lower pole ureteric reflux and no function of both upper and lower renal segments.[1]

In conclusion, the approach of managing a patient with ureterocele should be individualized. Transurethral incision remains valuable as a primary intervention with regular follow up. Transurethral incision may prove to be the only intervention required in most of the patients.

Ethics Committee Approval: Authors declared that the research was conducted according to the principles of the World Medical Association Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects", (amended in October 2013).

Informed Consent: Written informed consent was obtained from the parents of the patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – H.S., C.T.; Design – H.S., C.T.; Supervision – H.S.; Resources – H.S., C.T.; Materials – H.S., C.T.,

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Hasta Onamı: Yazılı hasta onamı bu çalışmaya katılan hastaların ailelerinden alınmıştır.

Hakem Değerlendirmesi: Dış bağımsız.

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