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Outcomes of Endoscopic Incision for the Treatment of Ureterocele in Children at a Single Institution

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Abstract

Purpose—We assessed outcomes in children with ureterocele after transurethral incision at our institution between 2001 and 2014, focusing on end points of vesicoureteral reflux, improvement of hydronephrosis and need for second surgery.

Materials and Methods—We performed chart reviews of 83 patients, collecting information including age at transurethral incision, gender, renal anatomy, ureterocele location, indication for transurethral incision, and preincision and postincision vesicoureteral reflux and hydronephrosis status. Patients were divided into those with single system and duplex system ureteroceles, and intravesical and extravesical location for analysis. Statistical evaluations were performed with the Wilcoxon rank test and Fisher exact test.

Results—Transurethral incision was performed at a mean age of 34.2 months in patients with single system ureterocele and 8.9 months in those with duplex system ureterocele ($p < 0.0001$). Cure rates (improvement of hydronephrosis and absence of vesicoureteral reflux) were 55.6% in patients with single system ureterocele and 14.9% in those with duplex system ureterocele ($p = 0.0031$). Rates of de novo reflux into the ureterocele moiety were 27.8% for patients with single system ureterocele and 56.2% for those with duplex system ureterocele ($p = 0.0773$). Patients with single system ureterocele required significantly fewer second surgeries (3.8%) than those with duplex system ureterocele (73.7%, $p < 0.0001$).

Conclusions—Patients with single system ureterocele may benefit from endoscopic incision. Transurethral incision was definitive in all such patients except 1 in our study. Although most patients with duplex system ureterocele will need a second operation, transurethral incision remains of value in this population in instances of sepsis or bladder outlet obstruction, or to facilitate planned reconstruction when the child is older.

Keywords

endoscopy; kidney diseases; patient outcome assessment; ureterocele; urethra

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Ureteroceleles occur in association with a single or duplicated collecting system and are classified as intravesical (orthotopic) or extravesical (ectopic), in which a portion of the ureterocele is situated permanently at the bladder neck or in the urethra.¹ Reflux may occur in any moiety or combination of moieties. Presentation can be symptomatic (eg infection) or asymptomatic (hydronephrosis). Goals of ureterocele management include decompression, avoiding vesicoureteral reflux, preventing urinary tract infection, and minimizing the number and invasiveness of operative interventions. Variations in ureterocele anatomy and mode of presentation can pose a clinical dilemma regarding management, and the optimal approach remains controversial. Transurethral incision is an attractive treatment option, given its relative ease and noninvasiveness.

We reviewed the records of patients who underwent transurethral incision to assess postoperative outcomes. Primary end points were improvement in hydronephrosis, de novo reflux into ureterocele moiety, cure (defined as improved hydronephrosis and no reflux postoperatively) and need for secondary surgery. We hypothesized that children with single system or intravesical duplex system ureteroceleles would have less post-transurethral incision reflux and undergo fewer second procedures than children with extravesical duplex system ureteroceleles, as reported previously.^{2,3}

MATERIALS AND METHODS

We conducted an institutional review board approved retrospective chart review of all patients who underwent TUI at our institution between 2001 and 2014. Incision was made using either a Bugbee™ electrode or resecto-scope (16 and 67 patients, respectively). Demographic information, including age at TUI, gender and type of ureterocele (DSU or SSU and intravesical or extravesical), was collected for each patient. Ureterocele size, indication for TUI, preincision and postincision VUR status, and preincision and postincision hydronephrosis status of each moiety were recorded. TUI was considered curative if patients demonstrated improved hydronephrosis and no VUR postoperatively.

A total of 83 children (median age 3.5 months) underwent TUI of 98 ureteroceleles between 2001 and 2014. This cohort represents 36.4% of 228 children presenting to our clinic for initial evaluation of ureterocele during this period. Mean length of followup after TUI was 23.9 months. Objective size information was available for 70 ureteroceleles, and revealed a range of 3.9 to 43 mm in maximal dimension (mean 15, median 13). Indications for transurethral incision were UTI with sepsis and/or fever in 27 patients, UTI with no mention of sepsis or fever in 5, hydronephrosis/preservation of renal function in 48 and acute renal failure, bladder outlet obstruction and abdominal pain in 1 each. Objective functional data were not obtained in all patients preoperatively. Therefore, the validity of operating for renal function preservation was unclear in several patients.

Of 83 patients 22 underwent preoperative dimercaptosuccinic acid scan, of whom 20 had DSU. Only 2 patients had a nonfunctional upper pole. Patients with hydro-nephrosis, documented salvageable function in the ureterocele moiety and no associated VUR underwent TUI for a potentially definitive procedure, while other patients underwent TUI

with the understanding that open reconstruction might be required subsequently. In general salvageable function was considered to be greater than 10% of function on the affected side.

Patients were divided into 2 groups for further analysis, ie those with SSU and those with DSU. Statistical evaluations were performed with the Wilcoxon rank test and Fisher exact test (2-tailed) using SAS®, version 9.3. Findings were considered statistically significant at $p < 0.05$.

RESULTS

Patients with SSU underwent surgical intervention at a mean age of 34.2 months. Patients with DSU were younger on average, undergoing TUI at a mean age of 8.9 months (table 1). The SSU group contained approximately 3 times more males than females, while the inverse was true for the DSU group. Because the management of ureterocele is controversial and dependent on several factors, the consultants at our institution did not use a standard followup protocol after TUI. However, almost all patients (78 of 83, 94%) underwent postoperative ultrasound, and the majority of patients (67 of 83, 80.7%) underwent postoperative VCUG to check the reflux status of each moiety and to further characterize the ureterocele after incision. A total of 22 patients underwent postoperative dimercaptosuccinic acid scan.

Single System Ureteroceles

Bilateral ureteroceles were present in 42% of children with SSU and 7% of those with DSU. All single system ureteroceles were intravesical. Of 26 patients in the SSU group 24 had preoperative and postoperative ultrasound available for review, with 87.5% showing improvement or resolution of hydronephrosis and 12.5% displaying stable hydro-nephrosis (table 2). TUI was curative in 56% of patients with SSU for whom preoperative and postoperative ultrasound and postoperative VCUG data were available.

De novo reflux was seen in 28% of patients with SSU with preoperative and postoperative VCUGs available for review. One of these patients had high grade de novo ipsilateral VUR and persistent contralateral VUR, and underwent a second surgery consisting of excision of ureterocele and bilateral ureteral reimplant. This is the only patient in the SSU cohort who required secondary surgery. The remaining patients with SSU had low grade de novo ipsilateral VUR and have been followed clinically with (2 patients) or without (2) antibiotic prophylaxis. No patient has had further problems with UTI. The plan for these patients is to obtain another VCUG and to consider surgical intervention if VUR persists.

The single instance of persistent contralateral VUR seen in the SSU case necessitated secondary surgery, as discussed. No instances of de novo or resolved contralateral VUR were observed. We compared those treated with Bugbee puncture vs incision with a resectoscope and found no difference in outcomes. Therefore, the subgroups were combined.

Duplex System Ureterocele

Our 57 DSU cases were divided into those involving intravesical (54%) and extravesical (46%) ureterocele. Of patients with a preoperative VCUG negative for ipsilateral upper pole reflux and postoperative VCUG available the rate of de novo reflux into the ipsilateral upper pole moiety after TUI was 43.7% in the intravesical group and 68.7% in the extravesical group ($p = 0.2852$, supplementary table, <http://jurology.com/>), and 56.2% for all duplex system ureteroceles. De novo VUR into the contralateral ureter was observed in 23.5% of the intravesical group and 15.4% of the extravesical group ($p = 0.6725$). Of the 6 patients with intravesical DSU with preoperative VCUG negative for ipsilateral lower pole VUR and postoperative VCUG available de novo ipsilateral lower pole VUR occurred in only 2. Of the 15 patients with extravesical DSU with preoperative VCUG negative for ipsilateral lower pole VUR and postoperative VCUG de novo ipsilateral lower pole VUR similarly developed in only 2. The difference in rates of de novo lower pole VUR between these 2 groups was not statistically significant.

Of 31 patients with intravesical ureterocele 21 (67.7%) underwent secondary surgery, compared to 21 of 26 (80.8%) with extravesical ureterocele ($p = 0.3681$). Indication for secondary surgery was nonfunctioning moiety in 3 patients with intravesical DSU and VUR in 18, of whom 11 demonstrated persistent reflux in at least 1 moiety with or without de novo reflux in another moiety. Only 2 patients underwent secondary surgery for the indication of de novo ipsilateral upper pole VUR alone. In 1 patient the indication for secondary surgery was de novo contralateral VUR, which likely represented unmasking of inherent reflux on that side related to trigonal distortion. Four patients underwent secondary surgery for reflux that unfortunately could not be identified as new or persistent, given the lack of preoperative VCUG.

Indication for secondary surgery was VUR in 19 of 21 patients with extravesical DSU undergoing secondary surgery and nonfunctioning ipsilateral upper pole in 1. The remaining patient underwent ureteroureterostomy for preservation of renal function in the setting of improved but still moderate upper pole hydronephrosis and good upper pole function. Of the 19 patients who underwent secondary surgery for VUR 8 exhibited persistent reflux in at least 1 moiety with or without de novo reflux in another moiety, 10 had de novo reflux alone and 1 had VUR that could not be identified as new or persistent, given that preoperative VCUG was not done in this patient.

The majority of patients in the intravesical (65.5%) and extravesical (79.2%) subgroups had improved hydronephrosis after TUI ($p = 0.3630$). The cure rate was 11.1% in the intravesical group (3 of 27 patients) and 20.0% in the extravesical subgroup (4 of 20 patients, $p = 0.4380$).

DISCUSSION

Snyder and Johnston in 1978 reported a 100% postoperative reflux rate after TUI, leading them to propose initial reconstruction at the bladder level as the preferred approach.⁴ The topic was revisited by Monfort et al in 1985, who supported TUI for urgent relief of obstruction in an ill neonate as a first step in decompressing the urinary tract to facilitate

later reconstruction and as possible definitive treatment in cases without de novo VUR.⁵ Tank agreed that TUI may facilitate planned later reconstruction by allowing the “obstructed ureters to return toward normal.”⁶ We previously described alternative methods of primary reconstruction at the bladder level, which required tailoring in 4 of 6 ureters, vs TUI as an initial procedure to address the more immediate problems associated with the ureterocele (sepsis, obstruction, prolapse) and to facilitate future reconstruction.⁷

Reoperation rates have often been used as a primary outcome measure to evaluate the success of TUI. In multiple studies it has been noted that the reoperation rate is higher in patients with DSU vs SSU.^{3,8,9} This observation holds true in our series and is consistent with the results of a recent meta-analysis by Byun and Merguerian.¹⁰ Multiple studies have also revealed a significantly increased rate of secondary surgeries in ectopic vs orthotopic ureteroceles.^{2,3,8,9} However, a recent study by Castagnetti et al showed no difference in this or other outcomes, leading the authors to suggest that differentiating between orthotopic and ectopic ureteroceles is clinically useless.¹¹ It is noteworthy that the meta-analysis by Byun and Merguerian¹⁰ includes data from the trial by Castagnetti et al¹¹ and supports the hypothesis that reoperation rates are higher in patients with ectopic ureteroceles. The authors report that the excess risk has decreased with time, which they attribute to an increasing reoperation rate in the intravesical group. We are unable to evaluate reoperation rates of intravesical vs extravesical SSU due to the absence of patients with extravesical SSU in our cohort. Our data do not support a significant difference in the rates of reoperation between patients with intravesical and extravesical DSU.

The success rate in our SSU group was good, with improvement of hydronephrosis in 21 patients (87.5%), cure in 10 (56%) and the need for secondary surgery in just 1 (3.8%). These results are comparable to other reported success rates for single system intravesical ureteroceles.^{2,3} It is noteworthy that most patients in our SSU group were male, all ureteroceles were intravesical and patients underwent TUI at a significantly older age than patients in the duplex system group (average 34.2 vs 8.9 months).

The rate of new VUR into ureterocele when measured per child was not statistically different between patients with SSU and those with DSU. However, when this rate was calculated per ureterocele, the difference was significant ($p = 0.0137$). Additionally the success rate was not as good in patients with DSU in terms of cure rate and need for further surgery. Other studies have also demonstrated the rate of secondary surgery to be higher in patients with DSU vs SSU.^{9,10} We compared outcomes for duplex extravesical and intravesical ureteroceles and found no significant difference. The need for secondary surgery after TUI was 67.7% in the intravesical group and 80.8% in the extravesical group. These rates are comparable to rates reported in the literature.^{7,8,12,13} Our rates of de novo reflux into the upper ureter after incision were higher than in prior studies, at 68.7% for extravesical and 43.7% for intravesical ureteroceles. We incised the majority of ureteroceles with a resectoscope using a low transverse incision, which does not differ significantly from puncture with a Bugbee electrode.^{6,12} We also did not see a significant difference in outcomes with Bugbee electrode vs resectoscope in any of our groups, and, therefore, combined these subgroups, although our study might not have been powered to reveal a difference if one existed.

While it is useful to look at reoperation rates, it is important to realize that TUI is often undertaken knowing that additional surgery might be required. Therefore, one must consider not only the rate of secondary surgery, but also the surgical indications. Although our DSU intravesical group had a high reoperation rate (21 of 31 patients, 67.7%), only 2 patients underwent secondary surgery for the sole indication of de novo ipsilateral upper pole VUR. In contrast, 10 of 21 patients with extravesical DSU requiring reoperation had an indication of de novo ipsilateral upper pole VUR alone.

While many of our patients underwent secondary surgery for the indication of VUR, recent studies indicate that in a subset of patients post-TUI VUR may resolve on its own.^{14,15} It has been suggested that as long as decompression is achieved, secondary surgery may be unnecessary in the asymptomatic patient regardless of VUR status or upper pole function.¹⁶ For these reasons, and also because of evidence that nonfunctioning unobstructed upper pole moieties may be safely left in situ,¹⁷ reoperation rates in patients with DSU might begin to decrease in the future.

Limitations of our study include its retrospective nature and small sample size. In addition, all studies of interest were not available for all patients (eg not every patient underwent preoperative and postoperative ultrasound or postoperative VCUG to assess for cure), which can introduce bias. We only considered outcomes after TUI, and, therefore, we cannot comment on the efficacy of TUI compared to alternative primary treatment options, including nonsurgical management. These issues require further investigation.

CONCLUSIONS

When reconsidering an individualized approach to treatment of ureterocele, we wondered which patients would benefit from endoscopic incision. From our data we conclude that children with a single system intravesical ureterocele would benefit the most from endoscopic incision, as this approach was definitive in all but 1 such patient. The majority of children with duplex system ureteroceles have historically undergone secondary surgeries, many planned in the setting of preoperative VUR. This finding held true in our cohort. If additional surgery is planned, endoscopic incision remains valuable in the context of sepsis or to facilitate subsequent elective reconstruction. New evidence indicates that post-TUI reflux can be safely followed nonoperatively as long as it is asymptomatic, and it may resolve spontaneously. Therefore, TUI may prove to be the only required surgery in a growing number of patients with ureterocele.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Abbreviations and Acronyms

DSU	duplex system ureterocele
SSU	single system ureterocele

TUI	transurethral incision
UTI	urinary tract infection
VCUG	voiding cystourethrogram
VUR	vesicoureteral reflux

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Table 1

Patient demographics

	SSU	Intravesical DSU	Extravesical DSU	Overall
Av age at TUI (mos) *	34.2	3.6	15.3	8.9
Median age at TUI (mos)	9.7	2.7	2.2	2.5
No. gender: †				
Male	19	7	6	13
Female	7	24	20	44
No. bilat/total No. (%) ‡	11/26 (42.3)	2/31 (6.5)	2/26 (7.7)	4/57 (7.0)

* SSU vs DSU $p < 0.0001$ and intravesical vs extravesical DSU $p = 0.6711$ (both Wilcoxon rank test).

† SSU vs DSU $p < 0.0001$ and intravesical vs extravesical DSU $p = 1$ (both Fisher exact test).

‡ SSU vs DSU $p = 0.0003$ and intravesical vs extravesical DSU $p = 1$ (both Fisher exact test).

Table 2

Treatment outcomes

	SSU	DSU	p Value
No. improved hydronephrosis/total No. (%)	21/24 (87.5)	38/53 (71.7)	0.1564
No. de novo reflux into ureterocele/total No. (%):			
Measured per child	5/18 (27.8)	18/32 (56.2)	0.0773
Measured per ureterocele	5/22 (22.7)	19/33 (57.6)	0.0137
No. improved hydronephrosis + no VUR/total No. (%)	10/18 (55.6)	7/47 (14.9)	0.0031
No. second surgery/total No. (%)	1/26 (3.8)	42/57 (73.7)	<0.0001