ORIGINAL RESEARCH

Outcomes of Duplex System Ureteroceles in Pediatric Patients Managed Primarily with Transurethral Incision

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Transurethral incision of ureterocele (TUI-U) is a simple, quick, less invasive, and less expensive, and an effective procedure for the management of ureteroceles. Several studies have already shown its utility for primary management of ureteroceles but it has also been associated with the need for additional surgery. The authors reviewed charts of patients from their database to describe the outcomes of TUI-U done in ureteroceles associated with the upper pole moiety of a duplex system. They also looked into preoperative patient characteristics and post TUI-U outcomes that could influence the need for subsequent surgeries.

Materials and Methods: The authors identified patients from their duplex system database who presented with a ureterocele and underwent TUI-U. They reviewed the patient records of 25 patients who were included in the study to determine the outcomes of TUI-U in duplex system ureteroceles. Chi square and Mann Whitney U tests were used to determine whether preoperative patient features and post TUI-U outcomes were associated with secondary surgery.

Results: Out of 65 patients who had duplex system ureteroceles, 25 patients (38.4%) underwent TUI-U at a mean age of 1.51 years old. TUI-U alone was successful in improving the prevalent signs and symptoms of 15 patients (60%) in this group, while 10 patients (40%) had to undergo subsequent surgical procedures. Breakthrough urinary tract infection (UTI) post TUI-U was the only patient factor noted to be significantly associated with a secondary surgery for duplex system ureterocele ($p=0.027$).

Conclusions: TUI-U as primary treatment for duplex system ureteroceles is not yet widely accepted due to reported rates of morbidities and need for secondary surgery. Present data however show that TUI-U can be used as a primary procedure and even as a definitive procedure for this subset of patients with remarkable results in terms of symptoms resolution and improvement of upper tract profiles.

Key words: Ureteral surgery, endoscopy, Duplex system

Introduction

A ureterocele is a cystic dilatation of the distal end of the ureter as it enters the urinary bladder. It is often associated with a poorly functioning upper pole moiety of a duplex (double collecting) system and may be intravesical in location or below the bladder trigone, usually at the area of the bladder neck.[1] Ureteroceles may cause obstructive lower urinary tract symptoms and lead to recurrent urinary tract infections (UTIs). They can also affect ipsilateral and contralateral renal units by altering trigonal architecture resulting in varying degrees of reflux

and hydronephrosis, which may result in significant kidney injury over time.[2]

Variations in the clinical presentation and associated upper tract features make it challenging to propose an all-encompassing treatment protocol for ureteroceles.[3,4,5,6] Treatment goals would however be essentially the same- ureterocele decompression, control of UTIs, resolution of hydronephrosis and reflux, and prevention of renal damage.[6,7] The approach to patients should be individualized and dependent on the severity of presenting signs and symptoms at the time of consult.

Extensive surgery on the miniscule and immature anatomy of very young patients would not only prove to be a challenging task but is also associated with high risk of significant morbidities and complications. Postponing management however might lead to further renal impairment which could have otherwise been prevented.[1,7,8] Transurethral incision of ureterocele (TUI-U) is a simple, quick, less invasive, and less expensive, but effective procedure for the management of ureteroceles. Several studies have already shown its utility for the primary management of ureteroceles but it has also been associated with the need for additional surgery.[6,8,9,10]

The authors reviewed the records and assessed the outcomes of pediatric patients in the double collecting (duplex) system database who underwent TUI-U as primary treatment for duplex system ureteroceles. Their primary objective was to describe the outcomes of TUI-U in their subset of patients to lend support to its role as a possible definitive surgery for ureteroceles. They also looked into patient characteristics and post TUI-U outcomes that could influence the need for further surgical intervention.

Materials and Methods

Pediatric patients (<18 years old) with ureteroceles associated with an upper moiety of a duplex kidney, who consulted and eventually underwent TUI-U as primary treatment were included in the study. The clinical data of these patients were retrieved from the senior author's duplex system database (which involved 221 patients seen from January 1990 to May 2016) and reviewed. Twenty-five (25) of these patients (11.31%) met the inclusion criteria and thus comprised the study population. Chart review of each subject was done to determine preoperative characteristics and short- and long-term outcomes after TUI-U.

Ureteroceles were diagnosed preoperatively by renal and bladder (KUB) ultrasound and/or a voiding cystourethrogram (VCUG). Seven patients also had a renal scan done preoperatively. TUI-U was done by making a transverse incision approximately 3-5mm in length just above the junction of the ureterocele and bladder floor, using an endoscopic electrode. Patients were subsequently followed up for presence of UTI, persistence of the ureterocele, presence of reflux or hydronephrosis, or deterioration in kidney function. Individualized follow-up schedules and subsequent management were based on each patient's response to the primary TUI-U. Patients with recurrent or breakthrough UTI alone, or UTI with concomitant inadequate ureterocele decompression, or denovo vesicoureteral reflux underwent a secondary procedure as deemed appropriate by the attending urologist.

Utility of TUI-U was evaluated by comparing patients' clinical picture and upper tract status before and after the procedure. Rate of secondary surgery post TUI-U was also used to gauge the effectivity of TUI-U as definitive management for ureteroceles. Patients were followed up with an ultrasound, VCUG and/or renal scan post TUI-U as needed. Age at TUI-U, presence of UTI, and upper tract status preoperatively and on follow-up post TUI-U, were analyzed as independent variables to determine their correlation with success rates and the need for a subsequent surgical procedure after TUI-U using a Chi-square test. Mann-Whitney U analysis was used to determine if different grades of hydronephrosis and VUR present prior to TUI-U affected outcomes in terms of need for a second operation.

Results

A total of 221 patients were encoded in the duplex system database. From this cohort, 65

patients had ureteroceles associated with the upper moiety of a duplex system. Furthermore, 25 of their patients (38.46%) underwent TUI-U as primary treatment, for a total of 27 incised ureteroceles (23 unilateral, 2 bilateral) over a span of 26 years. The patients' average age at initial consult was 1.1 years (range: 0.01 to 5.5 years), with 20 (80%) female and 5 (20%) male patients (Table 1).

Table 1. Study population characteristics.

Duplex System Database	221 Patients
With Ureterocele	65 Patients
Study Population	25 Patients
Age at TUI-U (Mean, Range)	1.51 years, (0.016 to 5.6 years)
Gender	
Male	5
Female	20

Sixteen (16) patients presented with UTI on first consult (64%), while 11 (44%) were found to have a ureterocele during their prenatal checkup. Average age of patients at TUI-U was 1.51 years (range: 1 week to 5.6 years), and the operation was done within 6 months after the initial consult. Mean follow-up duration for the whole population was 3.08 years. The ureteroceles were intravesical in 24 patients (22 unilateral and 2 bilateral) and ectopic in 3 patients, being located at the area of the bladder neck. Preoperative hydronephrosis of the ipsilateral upper moiety and lower pole kidney were noted in 27 and 3 patients respectively. Grade 3 hydronephrosis was noted in the ipsilateral upper pole moiety in most cases (12 of 25, 48%), followed by Grade 4 (9 of 25, 36%) and lastly Grade 2 (4 of 25, 16%) hydronephrosis. There were 2 patients with hydronephrosis on the contralateral side (Grade 2 and Grade 4), while VUR was noted in the ipsilateral ureter in 6 patients (24%) during initial evaluation (Tables 1 & 2).

TUI-U was the only treatment necessary for 15 patients (60%) included in the study. Decompression of the ureterocele was noted in 14 of 15 patients (93.3%) and improvement of the hydronephrosis was noted in 13 of 18 affected upper pole moieties (72.2%) as of their last follow-up (mean 1.88 years, range 0.28 to 8.31 years). Two patients had ipsilateral hydronephrosis in the lower pole and one patient presented with a contralateral hydronephrosis, all of which showed improvement after TUI-U. Reflux was noted on the ipsilateral renal unit in 4 patients, with improvement in only 1 (25%) post ureterocele incision. There was no contralateral reflux on all patients preoperatively and no denovo reflux on the contralateral side after TUI-U. There were breakthrough UTIs in 3 of 9 patients during the course of follow-up (33.3%) but were eventually controlled and no secondary operation was needed (Table 3).

Table 2. Preoperative patient characteristics

	TUI-U only	with Second Procedure	Total
Patients (n)	15	10	25
Presentation on Consult			
UTI	9	7	16
Prenatal Checkup	7	4	11
Ureterocele			
Intravesical	14	10	24
Ectopic	2	1	3
Hydronephrosis			
Upper moiety	18	9	27
Lower moiety	2	1	3
Contralateral	1	1	2
VUR			
Ipsilateral	4	2	6
Contralateral	0	0	0

A subsequent operation was needed and carried out in 10 patients (40%) with an average follow up duration of 4.96 years (range: 1.02 to 19.9 years). The type of secondary procedure done was left to the discretion of the surgeon based on the clinical picture of the patient and information provided by imaging and work ups post TUI-U. The most common second procedure was total reconstruction (heminephrectomy with ureterocelectomy and ureteroneocystostomy) which was done in 4 patients (40%). Double barrel reimplantation was done in 2 patients (20%). Ureteroneocystostomy was also performed in 2 patients (20%), one of whom underwent bilateral ureterneocystostomy. One patient (10%) underwent heminephrectomy with ureterocelectomy, and another patient had 2 consecutive repeat TUI-Us (Table 4). These patients had recurrent or breakthrough UTIs with either denovo VUR (30%) or persistent ureterocele (20%). Two patients (20%) presented with UTI only and 3 patients (30%) underwent total correction because of a poorly functioning contralateral kidney noted on renal scan upon follow up post TUI-U (Table 5). Patients who underwent secondary procedures were also asymptomatic as of their last follow-up.

Discussion

Early experience with transurethral ureterocele management in the 1970s were met with unfavorable outcomes such as very high postoperative reflux rates, resulting in the reconstructive approach at the level of the urinary bladder (being proposed as the preferred initial treatment). TUI-U was later revisited in the mid 1980s for urgent relief of obstruction, with a definitive reconstruction should denovo VUR later occur.[6] Current literature would readily advocate TUI-U for single-system ureteroceles owing to higher reported success rates, as compared to duplex system ureteroceles.[7] However, up to 80% of ureteroceles are associated with the upper moiety of a duplex system and previous reports described these patients to have worse outcomes post TUI-U compared to those with single systems. Furthermore, these are the patients who would most likely present with symptoms that may warrant emergent management in the form of TUI-U, hence the authors wanted to focus on this subset of patients.[1,6,7,9,11]

Table 3. Summary of pre-operative patient characteristics and outcomes after TUI-U

Preoperative Characteristics/Outcomes		No. of Patients (%)	TUI-U only	Subsequent Operation
Study Population		25	15/25 (60%)	10/25 (40%)
UTI (on presentation)		16/25 (64%)	9/16 (56.25%)	7/16 (43.75%)
Improved		6/16 (37.5%)	6/9 (66.7%)	0
No Improvement/Recurred		10/16 (62.5%)	3/9 (33.3%)	7/7 (100%)
Ureterocele		N= 27	18	9
Improved		23/27 (85.2%)	17/18 (94.4%)	6/9 (66.6%)
No Improvement/Worsened		4/27 (14.8%)	1/18 (5.6%)	3/9 (33.4%)
Hydronephrosis (Ipsilateral)				
On Initial Consult		N= 27	18	9
Upper Moiety	Improved	16/27 (59.8%)	13/18 (72.2%)	4/9 (44.5%)
	No Improvement/Worsened/De Novo	11/27 (40.2%)	5/18 (27.8%)	5/9 (55.5%)
Lower Moiety On Initial Consult		N= 3	2	1
	Improved	2/3 (66.7)	2/2 (100%)	0
	No Improvement/Worsened/De Novo	1/3 (33.3%)	0	1/1 (100%)
Hydronephrosis (Contralateral)		N= 2	1	1
	Improved	1/2 (50%)	1 (100%)	0
	No Improvement/Worsened/De Novo	1/2 (50%)	0	1 (100%)
Vesicoureteral Reflux		N= 6	4	2
Ipsilateral	Improved	1/6 (16.6%)	1/4 (25%)	0
	No Improvement/Worsened/De Novo	5/6 (83.3%)	3/4 (75%)	2/2 (100%)
Contralateral	Improved			
	No Improvement/Worsened/De Novo			

Table 4. Subsequent secondary procedures after TUI-U

Procedure	No. of Patients
Total Reconstruction	4
Double Barrel Reimplantation	2
Bilateral Ureteroneocystostomy	2
Heminephrectomy with Ureterocelectomy	1
Repeat TUI-U	1

Table 5. Indications for secondary surgery

	Time of Onset post TUI-U (mean, range)	Number of Patients n (%)
Recurrent/Breakthrough UTI Only	1.69 years, 1.21-2.78 years	2 (20%)
UTI with denovo VUR to upper pole moiety	2.66 years, 2.09-3.01 years	3 (30%)
UTI plus persistent ureterocele/hydronephrosis/VUR	0.95 years, 0.88-1.02 years	2 (20%)
Non-functioning/Poorly functioning Contralateral Kidney	1.36 years, 0.52-2.68years	3 (30%)

Patients with ureterocoeles are commonly seen as referrals from general pediatricians after prenatal workup or neonatal screening in up to 30% of cases.[1] They may be asymptomatic in as high as 93% of cases but would occasionally present with febrile UTI or other non-specific symptoms (eg. hypertension, palpable mass).[7,8] The number of symptomatic patients in the present series was noted to be high at 60% (15 of 25 patients). These patients presented with febrile or recurrent UTI, and somehow reflect the predominant health-seeking behavior in the country, namely- unless patients become symptomatic, consult to a specialist would be delayed. This is in contrast to most reports from western countries wherein age at primary intervention for ureteroceles would be at less than 3 months of age.[6,7,8,10,11] For this study, mean age at TIU-U was 1.51 years (Table 1). Nevertheless, at 60% (15 of 25) cure or success rate post TUI-U (resolution of pertinent signs and/or symptoms necessitating surgical intervention), the outcome of the procedure was comparable to most reports.[8,10,12,13] Statistical analysis showed that age at primary procedure did not significantly influence the eventuality of a second surgery after TUI-U (p=0.794).

The present date showed that resolution of UTI post TUI-U was the only variable with a positive correlation with success rate (p=0.027). Outcomes pertaining to patients' upper tract status post-operatively did not necessarily influence the need for a second operation. TUI-U resulted in the resolution of UTI in 37.5% of the total population (6 of 16). UTIs were resolved in 66.7% (6 of 9) of patients who did not need a secondary operation. Episodes of UTI worsened, or recurrences were noted in all the cases (7 of 7) of patients who would eventually need further surgery (Table 3). Preoperative UTI was not significantly associated with the need for secondary surgery (p=0.99).

Incision of the ureterocele near the base, at its junction with the bladder floor, was effective in decompressing 85.2% (23/27) of ureteroceles in the present study. This technique creates a flap mechanism that theoretically prevents reflux after decompression, however, some reports show that denovo reflux can be observed in up to 56.2% of cases post TUI-U.[8,6] The present series showed only 11.1% (3 of 27) denovo reflux after endoscopic incision and this was one of the more common clinical indications for secondary surgery (Table 5). The rate of ureterocele

decompression was noted to be higher in the group of patients who did not undergo secondary surgery (94.4% vs 66.6%), however, the association was not statistically significant (p=0.316).

Preoperative ipsilateral upper pole hydronephrosis, regardless of grade or severity, did not correlate with the need for subsequent surgery. Merlini, et al. reported that the upper moiety associated with a ureterocele is non-functional in up to 90% of cases and its contribution to the global renal function would only range from 4%-8%.[1] Recent evidence also suggests that non-functioning upper pole moieties may be left in place as they neither place the patient at risk for malignancy or of developing hypertension and infections; thus obviating the need for further surgery in the upper tracts.[6,14] While appearance or persistence of VUR was an indication for secondary surgery (Table 5), VUR status both preoperatively and postoperatively (denovo) did not correlate with the need for secondary surgery in this study. Similar findings have been previously reported, and some further advocate less invasive management for VUR in this age group because of the possibility of spontaneous resolution.[1,7,15] Sander, et al. reported that as long as ureterocele decompression is achieved, secondary surgery may not be needed regardless of VUR status or upper pole function.[6,14]

Mean time of onset of indications for secondary surgery should also be considered during follow-up of patients (Table 5). While the mean follow-up period for the whole group was 3.08 years, average follow-up period for patients who did not undergo secondary surgery (1.88 years) may not be long enough to observe the emergence of such ominous features. Continuous close follow-up is therefore recommended for these patients.

Conclusion

The present study showed a high success rate (60%) for TUI-U as primary treatment for duplex system ureteroceles, despite reservations from other studies. While some reports show that additional surgeries are warranted for persistence or emergence VUR and hydronephrosis, and other similar morbidities; in this series, recurrence or breakthrough UTI post TUI-U was the only variable significantly correlated with secondary surgery. TUI-U is an accepted modality in addressing urgent complications brought about by obstructing ureteroceles and may be useful in facilitating subsequent reconstructive surgeries. With close monitoring and diligent follow-up, TUI-U may also prove to be a definitive surgical option, even in cases of duplex system ureteroceles. Individualized management per patient is still necessary for such a disease entity with variable presentations, however, the surgical procedures may not always be extensive. The authors report their data obtained via a review of their database, and as previously mentioned, their data reflect results from other published studies. Nonetheless, well-designed prospective, controlled studies with similar objectives may in the future further validate the results.

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