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Lower urinary tract reconstruction for ectopic ureterocele: What happens in the long-term follow-up?

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ABSTRACT

Background: We aimed to analyze the long-term clinical and lower urinary tract function outcomes in children with duplex system ectopic ureterocele who underwent ureteroneocystostomy and ureterocelectomy.

Methods: Fifty-one patients (28 females, 23 males) who underwent a series of surgical interventions including lower urinary tract reconstruction in childhood for duplex system ectopic ureterocele in our center between 1998 and 2019, were retrospectively reviewed. The demographic and clinical data, surgical history, and the indication for ureterocelectomy were noted. Lower urinary tract dysfunction (LUTD) status was assessed through dysfunctional voiding symptom scores (DVSS) and uroflowmetry in all patients at the last follow-up. The clinical outcomes, and LUTD were evaluated.

Results: At the last visit at a mean follow-up of 117.18 ± 57.87 months after ureterocelectomy, ipsilateral persistent lower pole VUR was detected in 5.6% (3/54 renal units, 2 females and 1 male) of the cases, who were treated using the subureteric injection. Abnormal DVSS (median 11, range 9–15) was detected in 27.4% (14/51 pts) of the patients. Out of these, 57.1% (8/14 pts) had storage symptoms, 35.7% (5/14 pts) had voiding symptoms, and 7.1% (1/14 pts) had both storage and voiding symptoms while 71.4%(10/14 pts) had abnormal uroflowmetry findings (plateau shaped flow curve in 2, staccato shaped curve with sustained EMG activity in 3, tower shaped curve in 2, interrupted shaped curve in 3 patients). Five patients had elevated residual volume. Anticholinergics were administered to six patients who had overactive bladder symptoms. In addition, two girls required open bladder neck reconstruction due to stress incontinence caused by bladder neck insufficiency.

Conclusions: Our findings showed that clinical success was achieved using the lower urinary tract reconstruction with no need for re-operation in 90.2% of patients with duplex system ectopic ureterocele. However, LUTD was present in 27.4% of our patients in the long-term follow-up. Therefore, LUTD should be carefully assessed in the long-term follow-up of these patients.

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1. Introduction

Ureterocele is the congenital cystic dilation of the intramural ureter. Although nomenclature for ureterocele subtypes vary, ectopic ureteroceles are defined as a type in which some portion of the ureterocele is situated permanently at the bladder neck or in the urethra [1]. Approximately 80% of all ureteroceles are known to be associated with the upper pole of a duplex system. On the other hand, 60% of these duplex systems are accompanied by ectopic ureteroceles [1]. There is an optional stepwise approach for the management of ectopic ureteroceles with duplex system [2]. The mainstay of management is to reduce the risk of urinary tract infection (UTI), vesicoureteral reflux (VUR) and renal deterioration as well as avoiding bladder dysfunction in these cases [2,3]. Various factors such as the presence of vesicoureteral reflux, upper pole renal functional status, and bladder dynamics play a role in determining the approach defined as upper urinary tract (upper pole partial nephrectomy, ureteropyelostomy or upper to lower pole ureteroureterostomy, with or without reimplantation of lower pole ureter), lower urinary tract (ureterocelectomy and common sheath reimplantation) or total reconstruction (upper pole partial nephrectomy, ureterocelectomy and common sheath reimplantation). However, surgical indications may sometimes depend on the

Abbreviations: DMSA, ^{99m}Tc dimercaptosuccinic acid scan; DVSS, Dysfunctional voiding symptom score; EMG, Electromyogram; IQR, Interquartile range; LUT, Lower urinary tract; LUTD, Lower urinary tract dysfunction; PVR, Post-void residual volume; UTI, Urinary tract infection; VCUG, Voiding cystourethrography; VUR, Vesicoureteral reflux.

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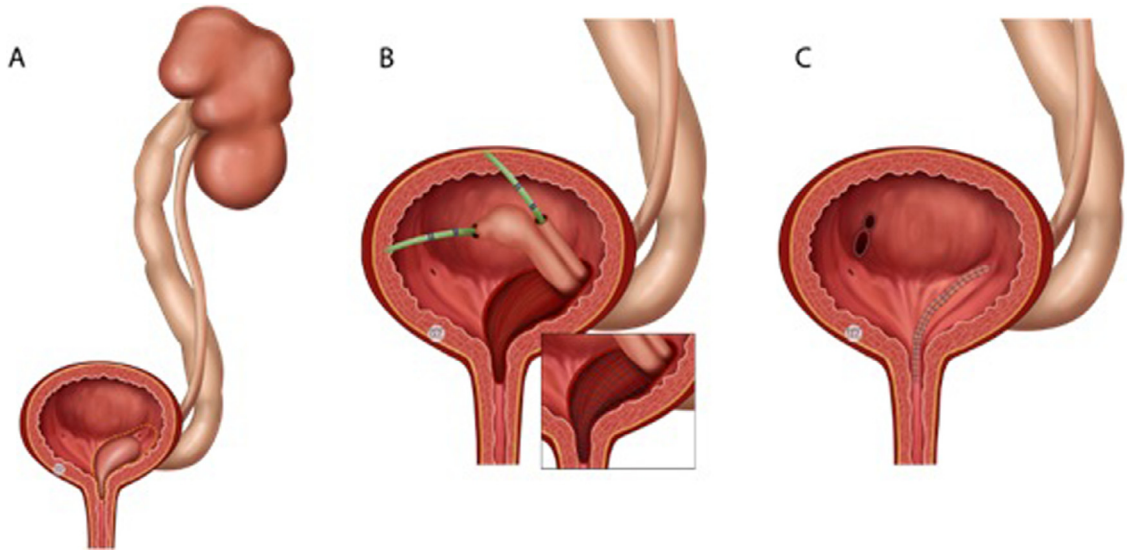


Fig. 1. Medical illustration of common sheath ureteral reimplantation following ureterocelectomy **A)** Incision line for common sheath ureteral reimplantation **B)** Ureters after dissection (Image on the right lower quadrant indicates the closure of the detrusor defect) **C)** After cross trigonal reimplantation.

surgeon's preferences [3]. Among all approaches, endoscopic ureterocele decompression is widely used as the initial intervention for ectopic ureteroceles as well as for the intravesical ureteroceles. However, higher re-operation rates (40–60%) have been reported in the long-term follow-up of ectopic ureteroceles [3,4].

Several previous studies showed that there was a risk for a high bladder capacity with incomplete emptying, infrequent voiding, urinary urgency and incontinence regardless of the type of surgical intervention as an integral part of the disorder in ectopic ureterocele. However, the ratio of the need for lower urinary tract reconstruction (ureterocelectomy and common sheath reimplantation) was extremely lower in these studies (12–18%) [5,6]. Although ureteroceles have been reported to be associated with bladder neck defect, which can lead to urinary incontinence or detrusor myogenic failure [2], the data is scarce on the long-term outcomes of lower urinary tract functions in children with ectopic duplex system who underwent lower urinary tract (LUT) reconstruction.

In the present study, we hypothesized that the re-operation rates are very low after bladder level reconstruction in duplex system ectopic ureteroceles. In addition, particular group of these patients require treatment for LUTD in the long-term follow-up.

2. Patients and methods

The patients with duplex system ureterocele who were followed up in our center between 1998 and 2019, were retrospectively reviewed. The study received Institutional Review Board approval. All procedures performed in our study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Among 91 patients whose data were available, 26 had intravesical ureterocele and 65 had ectopic ureterocele associated with a duplex system. Of 65 patients with ectopic ureterocele, twelve underwent endoscopic puncture alone, whereas 53 patients required subsequent surgeries (Nephroureterectomy in two patients and LUT reconstruction in 51 patients). Children with single system ureteroceles, those with intravesical ureteroceles, and those with ectopic duplex system ureteroceles that did not undergo LUT reconstruction were excluded. The demographic, and clinical data were noted. Surgical history, surgery indication, the preoperative and postoperative evaluations including ultrasonography, voiding cystourethrography (VCUG) and ^{99m}Tc dimercaptosuccinic acid scans (DMSA) were recorded. In addition, MAG-3 was done to confirm the persistent ureteral dilatation in patients who underwent subsequent ureterocelectomy due to persistent hydroureteronephrosis.

The indications for performing LUT reconstruction are the presence of the persistent or *de novo* ipsilateral or contralateral VUR, or persistent hydroureteronephrosis at either pole of the collecting system after the endoscopic puncture. Of note, surgery for LUT is usually delayed for 2–3 years in order to wait for maturation of the bladder dynamics, and resolving of the dilation. Lower urinary tract dysfunction (LUTD) status was assessed using the dysfunctional voiding symptom score (DVSS) [7]. According to this tool, a score of 8.5 or above shows voiding abnormalities.

Uroflowmetry was routinely performed at each follow-up visit of the patients aged over five years whose toilet training was completed. Uroflowmetry findings at the last clinical visit were also recorded. In the evaluation of the uroflowmetry parameters, the voided volume, flow curve, maximum urine flow rate, pelvic floor electromyogram (EMG) and post-void residual (PVR) urine volumes were recorded in all patients as described by the International Children's Continence Society [8]. PVR was considered elevated if it exceeds 10% of the expected bladder capacity for age [8]. Urodynamic studies were performed on patients with dysfunctional voiding when needed, as well as on patients undergoing bladder neck procedures due to stress type urinary incontinence.

The same pediatric urology team performed all operations. In each endoscopic decompression case, puncture at the point where ureterocele meets bladder on the most distal and medial part of the ureterocele was used as the surgical technique. Patients who presented with febrile UTI after puncture were assessed via VCUG and those who showed VUR on the either pole underwent LUT reconstruction. Also, the patients who had no resolution of the urinary tract dilation underwent bladder level reconstruction. LUT reconstruction was defined as the excision of the ureterocele with surgical repair of the bladder neck and urethral defect, and ureteroneocystostomy was carried out using common sheath ureteral reimplantation with the Cohen technique (Figs. 1 and 2). VCUG was requested only for patients with febrile UTI after reimplantation.

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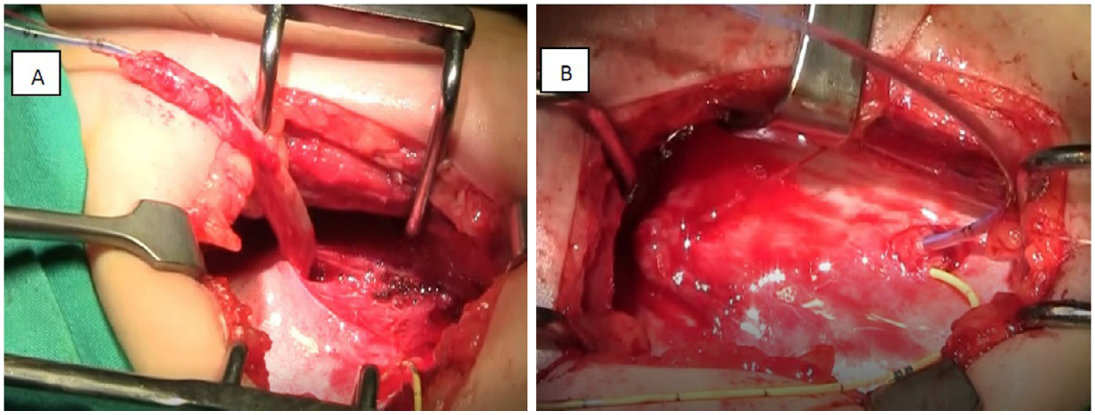


Fig. 2. Operative image of common sheath ureteral reimplantation following ureterocelectomy **A)** The detrusor defect after excision of the ureterocele (ureterocelectomy) **B)** Final appearance after repair of the detrusor defect and common sheath ureteral reimplantation.

2.1. Statistical analysis

Histogram, coefficient of variation, skewness and kurtosis, Q-Q plots, test of normality (Kolmogorov-Smirnov test, or Shapiro-Wilk test) were used for assessing the normality of the data [9]. Homogeneity of variance was evaluated using the Levene's test. Since all parameters showed non-normal distribution, the Mann-Whitney U test was used to compare two independent groups of non-normally distributed continuous variables [9]. The Chi-square test was used to compare the categorical data. The categorical data were shown as "number (percentage)". Among continuous variables, non-parametric data were shown as "median (IQR)". All statistical analyses were performed using the IBM Statistical Package for the Social Sciences (SPSS) Statistics 23 (IBM, Armonk, NY USA).

3. Results

Fifty-one patients had duplex system ectopic ureterocele (Three had bilateral ureteroceles resulting 54 renal units). The demographic, and clinical data of the preoperative period and surgical interventions are listed in Tables 1, 2 and 3. The study included 28 girls and 23 boys. The median age was 5 months (range 2 to 153 months) at the time of the initial surgery. Primary intervention was endoscopic puncture in 87.0% of the renal units (47/54). The main indications for LUT reconstruction were febrile urinary tract infections due to persistent ipsilateral lower pole VUR in 29.6% (16/54 renal units), *de novo* ipsilateral lower pole VUR in 20.3% (11/54 renal units) in addition to persistent ipsilateral dilation in 11.1% (6/54 renal units). The other indications are shown in Table 3. The incidence of *de novo* VUR of the upper pole after puncture was 14.8% (8/54 renal units). No postoperative surgical complications such as bladder outlet obstruction, progressive hydronephrosis, bleeding, leaks, or urosepsis were detected after LUT reconstruction. However, at the long-term follow-up, febrile upper urinary tract infection associated with ipsilateral persistent lower pole VUR was observed in 5.6% (3/54 renal units, 2 females and 1 male) after LUT reconstruction and were cured using the subureteric injection. The long-term renal functions were normal for age in all patients (Table 4).

At their last visit, at a median follow-up of 108 (IQR: 72–153, min: 24-max:264) months after LUT reconstruction, the median patient age was 13 years (range 6 to 27 years) and 27.4% (14/51 pts) of the patients had abnormal DVSS (median 11, range 9 to 15). Among them, 57.1% (8/14 pts) had storage symptoms, of whom 4 had urgency, 2 had urge incontinence, and 2 had stress incontinence. In addition, 35.7% (5/14 pts) had voiding symptoms, and 7.1% (1/14 pts) had both storage and voiding symptoms, whereas 10 of 14 patients with abnormal DVSS (71.4%) had abnormal uroflowmetry curve (plateau shaped flow curve in 2, staccato shaped curve with sustained EMG activity in 3, tower shaped in 2, interrupted shaped in 3 patients). Elevated PVR was detected in 5 out of these 10 patients (range 40–400 ml). These patients were further treated with urotherapy in the form of timed voiding, double voiding or biofeedback. Anticholinergics were used in 11.7% (6/51) of the patients who had overactive bladder symptoms.

No differences were detected regarding the rate of LUTD [10 (35.7%) vs. 4 (17.4%), $p = 0.152$], abnormal uroflowmetry findings [7 (25.0%) vs. 3 (13.0%), $p = 0.374$], and the rate of elevated PVR in accordance with the expected bladder volume for age [2 (7.1%) vs. 3 (13.0%), $p = 0.481$] between females and males, respectively (Table 4). In the long-term, 90.2% of the patients (85.7% of the females and 95.6% of the males) with duplex system ectopic ureterocele achieved clinical success with LUT reconstruction with no need for re-operation. Except for two females and one male who needed subureteric injection due to ipsilateral permanent lower pole VUR after LUT reconstruction, two patients required intervention due to stress incontinence. A girl aged 6 years underwent open bladder neck reconstruction three years after ureterocelectomy, whereas a girl aged 25 years underwent endoscopic bladder neck injection followed by an open bladder neck reconstruction (This patient underwent ureterocelectomy at the age of 2 years however, she was lost to follow-up until the intervention). The postoperative evaluations of the lower urinary tract functions of the patients in terms of gender in the long-term follow-up are shown in Table 4.

Our findings showed that out of a total of 14 (27.4%) patients having lower urinary tract symptoms (LUTS) and concurrent high DVSS scores, six (11.7%) patients used anticholinergics in addition to urotherapy due to their significantly severe symptoms. Two (3.9%) patients required additional open bladder neck reconstruction due to stress incontinence caused by bladder neck insufficiency. Only urotherapy in the form of timed voiding, double voiding, or biofeedback provided a cure in 3 (5.9%) patients presenting with mild LUTS. The remaining 37 asymptomatic (72.5%) patients had a median DVSS score of 2 (ranged 0 to 8) and they needed no treatment for LUTS. No significant differences were detected between female and male patients in terms of symptom severity (Table 4).

4. Discussion

The primary aim of the present study was to analyze the long-term clinical and lower urinary tract function outcomes in children with duplex system ectopic ureterocele who underwent LUT re-

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Table 1
Demographic and clinical data of 51 patients (54 renal units) in the preoperative period.

Parameters					
Patient ages at admission (years)			1 (1–1)		
min-max			1–13		
Gender (n,%)					
• Male			23 (45.1)		
• Female			28 (54.9)		
Type of Diagnosis (n,%)					
• Febrile UTI			12 (23.5)		
• Antenatal hydronephrosis			38 (74.5)		
• Incidental			1 (2.0)		
Side of ureterocele (n,%)					
• Left			26 (51.0)		
• Right			22 (43.1)		
• Bilateral			3 (5.9)		
Preoperative VUR side at initial admission (n,%)					
• Ipsilateral upper pole			4 (7.4)		
• Ipsilateral lower pole			25 (46.2)		
• Contralateral upper pole, in the presence of duplicated collecting system			0 (0.0)		
• Contralateral lower pole, in the presence of duplicated collecting system			2 (3.7)		
• Contralateral, in the presence of single system			7 (12.9)		
• None			16 (29.6)		
Preoperative VUR grade at initial admission (n,%)	**Grade I**	**Grade II**	**Grade III**	**Grade IV**	**Grade V**
• Ipsilateral upper pole	–	1 (1.9)	3 (5.6)	–	–
• Ipsilateral lower pole	1 (1.9)	3 (5.6)	10 (18.5)	6 (11.1)	5 (9.3)
• Contralateral upper pole, in the presence of duplicated collecting system	–	–	–	–	–
• Contralateral lower pole, in the presence of duplicated collecting system	–	1 (1.9)	–	1 (1.9)	–
• Contralateral, in the presence of single system	–	3 (5.6)	3 (5.6)	1 (1.9)	–

min: minimum, max: maximum, SFU: Society for Fetal Urology, UTI: urinary tract infection, VUR: vesicoureteral reflux.
Non-parametric data are shown as “median, (25th - 75th percentile)”. Additionally, minimum and maximum levels are also shown in the tables.
Categorical data are shown as “number (percentage)”.

Table 2
Clinical data of patients regarding surgical intervention.

Patient ages at the initial operation (months)		5 (2–12)	
min-max		2–153	
Type of operations (n,%)	**Initial operation**	**The second operation**	**Subsequent operations**
• endoscopic puncture	47 (87.0)	2 (3.7)	–
• heminephrectomy	2 (3.7)	1 (1.8)	–
• heminephroureterectomy	2 (3.7)	2 (3.7)	–
• ipsilateral ureteroneocystostomy + ureterocelectomy	–	27 (50.0)	7 (12.9)
• endoscopic ureteral injection	–	4 (7.4)	3 (5.6)
• contralateral endoscopic puncture	–	1 (1.8)	–
• ipsilateral ureteroneocystostomy + ureterocelectomy + contralateral ureteroneocystostomy	–	6 (11.1)	2 (3.7)
• ureteral stump excision+ ipsilateral ureterocelectomy	–	3 (5.6)	–
• bilateral ureterocelectomy + bilateral ureteroneocystostomy	–	4 (7.4)	–
• heminephrectomy + ipsilateral ureterocelectomy + ipsilateral ureteroneocystostomy	3 (5.6)	1 (1.8)	–
• heminephroureterectomy + ipsilateral ureterocelectomy + ipsilateral ureteroneocystostomy			1 (1.8)
• open bladder neck reconstruction	–	–	1 (1.8)
• endoscopic bladder neck injection	–	–	1 (1.8)
The rate of subsequent operations (n,%)		51 (94.4)	
Total number of operations		2 (2–3)	
min-max		1–4	

min: minimum, max: maximum.
Non-parametric data are shown as “median, (25th - 75th percentile)”. Additionally, minimum and maximum levels are also shown in the tables.
Categorical data are shown as “number (percentage)”.

construction. Our results showed that LUTD was present almost in one-quarter of patients in the long-term follow-up, although the surgery provided clinical success with no need for re-operation in almost 90% of patients with duplex system ectopic ureterocele.

Various treatment strategies exist in the management of ectopic ureterocele with duplex system. Endoscopic decompression (incision/puncture) is commonly the initial procedure in obstructed duplex system cases. In addition, some studies reported that an adequate single puncture or multiple punctures if required can provide good drainage from the obstructed ureterocele and may be effective in preventing vesicourethral reflux. Thereby, it decreases the incidence of subsequent surgeries due to *de novo* VUR [3,10]. However, the decompression of the upper urinary tract using various techniques may not be achieved with this method in 29–59% of the ectopic ureteroceles with duplex system [3,4,11,12]. Subsequent surgical procedures may be required even the rate of upper pole drainage reaches to 93% with a second endoscopic intervention [2,4,11,13]. In regards to these surgeries, common-sheath ureteral reimplantation with ureterocelectomy is the most common surgical procedure, especially in those with ipsilateral high-grade VUR. Of note, researchers have reported that 17.9% of the cases may require additional operations due to postoperative complications as-

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Table 3
Clinical data of patients before ureterocelectomy.

Patient ages at the time of ureterocelectomy (years) min-max			3 (2–4) 2–14		
Indications for subsequent ureterocelectomy procedures (n,%)					
• Persistent ipsilateral upper pole VUR			3 (5.5)		
• *de novo* ipsilateral upper pole VUR			8 (14.8)		
• Persistent ipsilateral lower pole VUR			16 (29.6)		
• *de novo* ipsilateral lower pole VUR			11 (20.3)		
• Persistent contralateral upper pole VUR in the duplex kidney			3 (5.5)		
• *de novo* contralateral upper pole VUR in the duplex kidney			0 (0.0)		
• Persistent contralateral lower pole VUR in the duplex kidney			2 (3.7)		
• *de novo* contralateral lower pole VUR in the duplex kidney			1 (1.8)		
• Persistent contralateral VUR in single system			4 (7.4)		
• *de novo* contralateral VUR in single system			0 (0.0)		
• Persistent hydroureteronephrosis			6 (11.1)		
Pre-ureterocelectomy VUR side (after ureterocele puncture) (n,%)					
• Ipsilateral upper pole			11 (20.3)		
• Ipsilateral lower pole			27 (50.0)		
• Contralateral upper pole, in the presence of duplicated collecting system			2 (3.7)		
• Contralateral lower pole, in the presence of duplicated collecting system			1 (1.8)		
• Contralateral, in the presence of single system			1 (1.8)		
• None			12 (22.4)		
Pre-ureterocelectomy VUR grade (after ureterocele puncture) (n,%)	**Grade I**	**Grade II**	**Grade III**	**Grade IV**	**Grade V**
• Ipsilateral upper pole	–	3 (5.6)	2 (3.7)	5 (9.3)	1 (1.9)
• Ipsilateral lower pole	–	4 (7.4)	9 (16.7)	8 (14.8)	6 (11.1)
• Contralateral upper pole, in the presence of duplicated collecting system	1 (1.9)	1 (1.9)	–	–	–
• Contralateral lower pole, in the presence of duplicated collecting system	–	1 (1.9)	–	–	–
• Contralateral, in the presence of single system	–	1 (1.9)	–	–	–
Presence of renal scar in DMSA before ureterocelectomy (n,%)					
• Ipsilateral upper pole			11 (20.3)		
• Ipsilateral lower pole			11 (20.3)		
• Contralateral upper pole			1 (1.8)		
• Contralateral lower pole			1 (1.8)		
• No scar			32 (59.4)		
Split renal functions (%) in DMSA before ureterocelectomy					
• Ipsilateral total function			41 (29.50–47.25)		
◦ Ipsilateral upper pole function			14.5 (10–20)		
◦ Ipsilateral lower pole function			24 (15–28)		
• Contralateral total function			59 (52.75–70.50)		

DMSA: ^{99m}Tc dimercaptosuccinic acid, min: minimum, max: maximum, VUR: vesicoureteral reflux.
Non-parametric data are shown as "median, (25th - 75th percentile)". Additionally, minimum and maximum levels are also shown in the tables.
Categorical data are shown as "number (percentage)".

sociated with the aggravation of hydronephrosis and obstruction, recurrent febrile UTI, VUR or voiding symptoms (e.g., elevated PVR, weak urine stream) in the follow-up of this surgery [14]. These results imply the importance of the individualized treatment for each patient.

In previous different studies, LUTD was reported as between 7.9–13% after endoscopic incision, whereas was reported as 13.4–18.4% after subsequent common-sheath reimplantation with ureterocelectomy for ectopic ureterocele [3,14,15]. In the two decades earlier series, the reported LUTD after total reconstruction were the high-capacity bladder with incomplete emptying (up to 50% of the patients) and urinary incontinence (about 10% of the patients) [16,17]. In a more recent study providing an overall view of ureteroceles, Paye-Jaouen et al. [2] reported that severe LUTD was observed at a very small rate of 6% in children with ureterocele and ureterocele decompression was a safe procedure that caused no significant LUTD. On the other hand, abnormal uroflowmetry results reached to 66% in those who underwent ureterocelectomy and ureteroneocystostomy as a subsequent surgery, whereas severe LUTD and abnormal DVSS, leading to patient admission were reported in 25% of the patients [2]. The older reports investigating the total reconstruction involving the upper and lower urinary tract, which was more commonly performed in the earlier decades in patients with ectopic ureterocele, revealed similar LUTD rate (20.3%) during a median follow-up of 9.6 years [18]. Our results are in accordance with the published series along the time (27.4%). Moreover, storage symptoms were detected in 9 (17.6%) of the patients, and a total of 7.8% had urinary incontinence in our study. Six (11.7%) patients responded well to a combination of urotherapy and anticholinergics, whereas urotherapy alone provided adequate improvement in 3 (5.9%) patients. Contrary to the literature, indicating the incidence of significant voiding phase problems including increased PVR around 50%, voiding symptoms was observed in 5 (9.8%) of our patients. In order to achieve urinary continence, additional surgery was required in only two (3.9%) patients due to stress incontinence. De Jong et al. [19] and Beganović et al. [18] stated that 8.1% and 6% of their patients, respectively, needed clean intermittent catheterization (CIC) following the open surgeries including ureterocele excision, however, our patients required no CIC procedure at all.

The possible etiologies in the development of postoperative LUTD have been suggested as the congenital defect on the bladder floor, changes in bladder neck configuration, damage to the bladder vascularization and innervation, iatrogenic injury to the bladder neck or external urethral sphincter due to surgery [14,15]. It has also been stated that myogenic detrusor insufficiency and the accompanying decrease in bladder compliance and elasticity of the bladder outlet may have caused LUTD [20]. Consequently, some researchers postulated that even if they are not operated, LUTD is a part of ectopic ureterocele [18]. Conflicting results have also been reported regarding the trigonal surgery as a cause of LUTD [15,18]. The rate of LUTD in patients with ectopic ureterocele undergoing various surgical treatments was found between 10 and 50% in different series, whereas this rate was 13–18% in the patient popu-

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Table 4
Postoperative lower urinary tract function evaluations of the patients in long term follow-up.

	Total population (n: 51, 100%)	Female (n: 28, 54.9%)	Male (n: 23, 45.1%)	p value
Patient age at final assessment (years)	13 (10–17)	12 (10–16.75)	13 (10–17)	†0.531
min-max	6–27	6–26	6–27	
Follow-up period after ureterocelectomy (months)	117.18 ± 57.87	111.86 ± 54.94	123.65 ± 61.87	ˢ0.480
min-max	24–264	24–264	36–252	
DVSS scores at the last clinical visit	2 (1–9)	3.5 (1–11)	2 (1–3)	†0.085
min-max	0–15	0–15	0–13	
Presence of LUTD at the last clinical visit (n,%)	14 (27.3)	10 (35.7)	4 (17.4)	‡0.145
LUTD subgroups (n,%)				
• Storage	8 (15.6)	7 (25.0)	1 (4.3)	‡0.152
• Voiding	5 (9.8)	2 (7.1)	3 (13.0)	
• Both	1 (1.9)	1 (3.6)	0 (0.0)	
• None	37 (72.7)	18 (64.3)	19 (82.6)	
Uroflowmetry curve at the last clinical visit (n,%)				
• normal (bell shaped)	41 (80.4)	21 (75.0)	20 (87.0)	‡0.374
• tower shaped	2 (3.9)	2 (7.1)	0 (0.0)	
• plateau shaped	2 (3.9)	2 (7.1)	0 (0.0)	
• staccato	3 (5.9)	2 (7.1)	1 (4.3)	
• interrupted	3 (5.9)	1 (3.6)	2 (8.7)	
Uroflowmetry Qmax at the last clinical visit (ml/min)	18 (14–22)	17.5 (14–22.75)	18 (15–21)	†0.955
min-max	3–57	3–57	6–31	
Uroflowmetry voided volume at the last clinical visit (ml)	265 (174–338)	265 (183.25–343.75)	266 (165–320)	† 0.520
min-max	114–595	118–595	114–413	
Uroflowmetry post-micturition residual volume at the last clinical visit (ml)	0 (0–28.25)	0 (0–28)	0 (0–30)	† 0.881
min-max	0–400	0–45	0–400	
The presence of elevated post-micturition residual volume at the last clinical visit (n,%) (greater than 10% higher than estimated bladder capacity)	5 (9.8)	2 (7.1)	3 (13.0)	‡ 0.481
Serum creatinine level at the last clinical visit (mg/dl)	0.53 (0.44–0.64)	0.53 (0.45–0.62)	0.50 (0.37–0.67)	† 0.595
min-max	0.33–0.73	0.34–0.73	0.33–0.69	
eGFR (CKD-EPI) level at the last clinical visit	146 (140–151)	147 (134.75–157.50)	146 (142–147)	† 0.489
min-max	122–198	122–166	140–198	

DVSS: dysfunctional voiding symptom scores, LUTD: lower urinary tract dysfunction, min: minimum, max: maximum, Qmax: Peak urine flow rate.
Uroflowmetry tests were confirmed to have voided volumes between 50% and%130 of estimated bladder capacity for age [(age+1) x 30 ml].
† Mann-Whitney U test; Data are shown as "median, (25th - 75th percentile)". Additionally, minimum and maximum levels are also shown in the tables.
ˢ Independent Samples t-test; Data are shown as "mean ± standard deviation".
‡ Chi-square test; Data are shown as "number (percentage)"*p <0.05 Asterisk (*) indicates statistical significance.

lation without ureterocele [17,18]. On the other hand, bladder dysfunction including a higher bladder capacity with incomplete emptying, infrequent voiding, urinary urgency and incontinence has been observed in several previous series, regardless of the type of surgical intervention as an integral part of the disorder in ectopic ureterocele [5,6]. Thus, it is reasonable to suggest that LUTD results due to a combination of both congenital, acquired and iatrogenic factors [15,17,18,20].

In our previous reported series of 18 patients with a mean follow-up of 5.4 years, all toilet-trained children were observed to have continence after LUT reconstruction [21]. However, other LUTD parameters were not evaluated except for postoperative continence status. On the other hand, our current results in the present study with longer follow-up period and higher number of patient participation indicate that LUTD may occur in approximately one-quarter of patients. Therefore, we conclude that LUTD is not a rare entity in the long-term follow-up after bladder level surgery. We suggest that the evaluation of LUT symptoms should be a part of clinical visits, since a substantial proportion of patients may face with LUTD in the long-term follow-up.

One of the most important limitations in the aforementioned studies is the inability to evaluate the presence of LUTD in very younger age groups such as infants and children below 5 years of age. Therefore, longer follow-up is needed for the assessment of LUTD. Most of these studies evaluating LUTD in the follow-up after LUT reconstruction have been published at least a decade ago. Therefore, the lack of a standard in the terminology and definitions of LUTD used in these studies may have caused misinterpretations. In addition, objective parameters such as uroflowmetry, urodynamics, and DVSS were not used as a method in evaluating the presence of LUTD in the majority of these studies. Generally, the only mentioned parameter was the postoperative continence. Since the patient evaluation was done in a modern and a standardized approach, we suggest that our study, with a relatively longer follow-up period, will contribute to the current data available on duplex system ectopic ureteroceles.

Our study also has some limitations. Only patients who had the symptoms of febrile urinary tract infections after LUT reconstruction underwent VCUG, therefore postoperative VUR rate might have been underestimated. On the other hand, retrospective study design and small sample size that only allow descriptive analysis in a single-arm study with no comparison cohort are the main limitations of this study. Although we had both patients with intravesical ureterocele and patients with ectopic ureterocele who underwent endoscopic puncture alone, the numbers of these patients were too small to provide sufficient statistical power for comparison analyses, especially for the LUT evaluation.

However, we compared the long-term clinical outcomes and LUTS for both genders in a subgroup analysis. Although we recommend routine visits per a protocol follow-up plan for all patients treated for congenital urinary pathologies, only the data of 51 patients who were followed up for duplex system ectopic ureterocele and underwent LUT reconstruction was accessible in this retrospective study. In the literature, researchers predicted that a higher rate of LUTD may postoperatively develop in females due to the higher incidence of duplex system ectopic ureterocele, the short urethra, and the different urethral closure mechanisms in females [5,6,11]. However, we attribute to the lack of significant gender differences in long-term follow-up in our study to the limited number of patients with a relatively high male ratio.

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5. Conclusions

Our results show that the persistent or *de novo* ipsilateral lower pole VUR and persistent ipsilateral dilation are the main reasons for the need for LUT reconstruction in the follow-up. Bladder level surgery provided sufficient clinical success with no need for re-operation in 90.2% of patients (85.7% of the females and 95.6% of the males) with duplex system ectopic ureterocele. However, it should be kept in mind that almost one-quarter of patients experience the symptoms of the LUTD in the long-term follow-up. Although the rate of LUTD in females seems to be higher, no significant difference is observed between males and females.

Compliance with ethical standards

Financial support

The authors declare that they have no relevant financial interests.

Ethical approval

All procedures performed in our study were in accordance with the ethical standards of the Institutional Research Committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was waived due to the retrospective nature of the study.

Declaration of Competing Interest

The authors declare that they have no conflict of interest.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jpedsurg.2022.09.002.

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