



# Endoscopic treatment of ureterocele in children: Results of a single referral tertiary center over a 10 year-period

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## Abbreviations

UC, Ureterocele; Ectopic UC, Ectopic Ureterocele

## Keywords

Ureterocele; Endoscopic treatment; Duplex collecting system; Ectopic ureterocele

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## Summary

### Introduction

The management of UCs remain controversial, especially for UCs with duplex collecting systems that still represent a great challenge in paediatric urology. Several approaches have been used and a shared management is not yet validated.

### Study aim

Aim of our study is to evaluate the results of the endoscopic treatment of UC comparing orthotopic single-system UC and ectopic duplex-system UC over a 10-year period in a single referral tertiary center. Success was defined as resolution of dilation, lack of urinary infections and preservation of renal function.

### Study design

We retrospectively reviewed medical records of children with a diagnosis of UC who underwent endoscopic puncture at our division from January 2009 to January 2019. Patients were divided in two groups: Group A composed of patients with ectopic UC associated with renal duplex system and Group B with orthotopic UC in single collecting system.

### Results

We identified 48 paediatric patients treated with transurethral primary endoscopic incision. Groups

result homogeneous for clinical and pathological characteristics. The only statistical significant difference between the two samples was the age at diagnosis (p value with Yates correction = 0.01).

### Discussion

We considered as a therapeutic success infections control and the elimination of obstruction with preservation of global kidney function. Based on that, our success rate after single (77%) or double (92%) endoscopic treatment is higher than data reported in literature. Differently from previous studies, vesico-ureteral reflux without UTIs was not considered as a failure of the procedure. The present study has some limitations: it is a retrospective and monocentric serie and it lacked a longer follow-up; on the other hand, it has been conducted on a quite large sample size and it is one of the few studies that compares the endoscopic treatment between orthotopic and ectopic UC.

### Conclusion

Our data report primary endoscopic puncture of ureterocele as a simple, effective, and safe procedure also in long-term follow up. This technique avoids the need for additional surgery in the majority of the patients, also in the case of an ectopic UC associated to a duplex system.

**Summary Table** Post treatment data were summarized in [Table 3](#).

	Persistent UTIs	Persistent VUR	"De Novo" VUR	UC sac bulging requiring second look
Group A	11 (42.3%)	1 (3.8%)	5 (19.2%)	6* (23.1%)
Group B	4 (18.2%)	1 (4.5%)	0 (0%)	2 (9.1%)
P Value (Yates correction)	0.14	0.54	0.09	0.36

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## Introduction

Ureterocele (UC) has been reported in 1 every 500 live births and 1:4000 in autopsy series [1]. UCs occur four to seven times more frequently in females than in males, and more commonly in Caucasians than in other races. Unilateral ureteroceles occur with similar frequency on the right and left side, and in 10 percent of cases there is bilateral involvement. This condition represents a clinical challenge because of the different anatomic and clinical presentations [2,3].

UC can occur in association with single or duplex collecting systems and the localization can be intra-vesical (orthotopic UC) or extravesimal (ectopic UC). Regarding clinical presentation, UC can be symptomatic, presenting mainly with urinary tract infections (UTIs), or asymptomatic. In this case, the initial diagnosis is consequent to the diagnostic investigations for a prenatal or postnatal hydronephrosis [3].

The management of UCs remains controversial, especially for UCs with duplex collecting systems that still represent a debated topic in paediatric urology. The goals of treatment are to eliminate the obstruction in order to preserve renal function, to protect the normal renal units, to control and avoid the UTIs, to maintain urinary continence, and to manage the vesicourethral reflux (VUR).

Several approaches, including endoscopic decompression, upper-pole partial nephrectomy and staged reconstruction, ureterocele excision and complete reconstruction have been used to achieve these goals [2–7]. Moreover, endoscopic puncture (EP) of UC as primary treatment has been widely reported and showed a high success rate in several pediatric urology series. Nevertheless, studies focusing on ectopic duplex system have been little reported and a shared management is not yet validated [4,5].

Aim of our study is to evaluate the results of the endoscopic treatment of UC comparing orthotopic single-system UC and ectopic duplex-system UC over a 10-year period in a cohort of 48 paediatric patients treated in a single referral tertiary center.

## Materials and methods

We retrospectively reviewed medical records of children with a diagnosis of UC who underwent endoscopic puncture at our division from January 2009 to January 2019.

Patients were divided in two groups: Group A composed of patients with ectopic UC associated with renal duplex system and Group B with orthotopic UC in single collecting system.

Preoperative evaluation included renal and bladder ultrasonography (US), voiding cystourethrography (VCUG) and MAG3 renal scan, as recommended by ESPU guidelines. Preoperative, peri and post-operative data were assessed. We analysed demographic data, antenatal or postnatal diagnosis, presence of hydronephrosis, clinical presentation with UTI, presence of VUR in the duplex system, age at surgery and complications. We also evaluated the persistence or the onset of UTI or VUR after the procedure and the need for a secondary surgery.

Both groups underwent the same follow up, that consisted in nephro-urologic evaluation with ultrasound scan 1, 3, 6 months and 1 year post procedure. VCUG and MAG 3 renography was performed only in patients with recurrent UTIs or if US scan show increasing ureteral dilatation with thinning of renal cortex.

Treatment failure is defined as no complete decompression (persistent upper tract dilatation) and persistence of ureterocele.

All data were analysed with the SPSS programme and the differences were statistically significant with p-values < 0.05.

## Surgical management

All patients underwent a single endoscopic incision of UC. The procedure was performed under general anaesthesia using an 8.5–9.5 Ch cystoscope with 5 Ch operative channel. Under direct vision the surgeon made a transverse incision extended from the distal aspect of the ureterocele to the bladder neck. The source of energy was an electrified monopolar hook or an Holmium laser 272 micron core fiber. At the end of the procedure a ureteral catheter (3 Fr) was used to verify the incision and removed before the end of the procedure. No bladder catheter was left in place.

## Results

Fifty-one patients were identified (24 females and 27 males), 3 patients were lost at follow-up and so 48 patients were considered for the purpose of this study. Age at diagnosis was between 0,1–198 months (IQR 21,425).

Twenty-six patients (16 females, 10 males) presented ectopic UC associated with renal duplex systems (group A) while twenty-two patients (7 females and 15 males) presented orthotopic UC (group B).

Descriptive features are listed in Table 1.

Associated anomalies were detected in 6 patients (3 for each group) and were represented by: polycystic kidney (1 case), Noonan Syndrome (1 case), spina bifida occulta (1 case), solitary kidney (2 cases), nephrolithiasis in congenital urethral stenosis (1 case).

Diagnosis was performed prenatally in 20 patients in group A (76.9%) and 11 patients in group B (50%).

Ureterocele was identified during abdominal US screening at birth in 4 children in group A (15.4%) and in 5 children in group B (22.7%); furthermore in six cases (2 group A and 4 group B) diagnosis was achieved later as a consequence of recurrent UTIs.

**Table 1** Descriptive features of both groups, percentage have been calculated on each group.

	Group A	Group B
Male	10 (38.5%)	15 (68.2%)
Female	16 (61.5%)	7 (31.8%)
Right sided	9 (34.6%)	14 (63.6%)
Left sided	16 (61.5%)	7 (31.8%)
Bilateral	1 (3.8%)	1 (4.5%)

One foreign child (4.5%) with orthotopic UC received a late diagnosis, because he came at our attention at the age of sixteen.

Twelve patients (5 group A and 7 group B) presented UTIs before treatment (in six cases despite prenatal diagnosis and antibiotic prophylaxis).

At US pre-treatment evaluation, all patients in group A and 21 children in group B presented hydronephrosis.

Vesico-ureteral reflux (Grade 2–4) was found at VCUg in 8 cases in group A (30,7%) and 3 cases in group B (13,6%), where it interested the ipsilateral lower moiety.

All these data are summarized in [Table 2](#).

All patients were treated with transurethral primary endoscopic incision. Punctures were performed using electrified hook in 24 patients in group A (92.3%) and in 18 in group B (85.7%). In Five patients (2 group A and 3 group B), the device used was Holmium YAG Laser. Complete immediate decompression was always obtained.

Mean age at endoscopy was 5.63 months (0.1–24 months, IQR 6.675) in group A and 41.34 months in group B (0.3–198 months, IQR 62.875).

No early complications occurred and all patients were discharged in 1 post-operative day.

The mean length of follow-up was 3.8 years (range 1–9 years, IQR 3.04).

Eleven cases in group A (42.3%) and 4 cases in group B (18.2%) presented recurrent postoperative UTIs. They were investigated with VCUg. Recurrent UC sac bulging and persistent upper tract dilatation was found in 6 cases in group A (23.1%) and 2 cases in group B. All patients in group B achieved a complete resolution after a second look endoscopic incision, while a third procedure was necessary in 2 cases in group A. The technique used was the same as the first treatment.

VUR was instead detected in 6 cases in group A (23.1%): in 1 case (3.8%) it was persistent, meanwhile in 5 cases (19.2%) it presented “*de novo*”. Two patients were successfully treated with antibiotic prophylaxis and clinical follow up, while 4 cases needed endoscopic VUR treatment with sub-mucosal injection of silicone (*Macroplastique*).

Only one patient had persisted VUR in group B (4.5%) and it was completely resolved with the same technique.

None of our patients reported renal function impairment after the procedures.

Groups result homogeneous for clinical and pathological characteristics. The only statistically significant difference between the two samples was the age at diagnosis (p value with Yates correction = 0.01).

Post treatment data were summarized in [Table 3](#).

## Discussion

Diagnosis of UC is achieved by antenatal ultrasonography in 75% of cases, while, after birth, the most common presentation is recurrent UTIs. However, the debate on the best clinical management of these children is still open [8–10].

As the EAU-ESPU guidelines 2021 underline about the UC management [1], the treatment should be based on symptoms, function and reflux as well on surgical and parenteral choices; the options vary from observation, endoscopic decompression, ureteral re-implantation, partial nephroureterotomy, to complete primary reconstruction. But the level of evidence in this case is 3 and the strength rating is weak [1].

The rationale for treatment is to preserve the most of renal function. The endoscopic correction of UC removes the obstruction on the urinary tract allowing to control infections, and to prevent or treat the VUR. In the case of ectopic UC with a subsphincteric outlet, urinary continence can only be restored by a ureteral reimplantation or eminephrectomy.

In the last two decades, the traditional aggressive treatment in the management of ureteroceles has changed to a more conservative approach by endoscopic puncture. Many reports indicated that these patients often require reintervention, such as ureteric reimplantation and upper pole partial nephrectomy owing to VUR either into the lower moiety of the ipsilateral kidney or into contralateral

**Table 2** Pre-treatment features.

	Group A	Group B	P Value (yates correction)
Prenatal diagnosis	20 (76.9%)	11 (52.4%)	0.10
UTIs	5 (19.2%)	7 (33.3%)	0.50
Hydronephrosis	26 (100%)	21 (95.4%)	0.88
VUR	8 (30.8%)	3 (13.6%)	0.29
Median age at diagnosis (months)	5.63	41.34	0.01

**Table 3** Post-treatment features (\*=two cases needed a third endoscopic treatment).

	Persistent UTIs	Persistent VUR	“De Novo” VUR	UC sac bulging requiring second look
Group A	11 (42.3%)	1 (3.8%)	5 (19.2%)	6* (23.1%)
Group B	4 (18.2%)	1 (4.5%)	0 (0%)	2 (9.1%)
P Value (Yates correction)	0.14	0.54	0.09	0.36

kidney or because of iatrogenic VUR into ureterocele moiety [11,12].

Nowadays endoscopic puncture or incision of the intravesical UC is considered the best approach, while in case of ectopic UC the management is controversial and debated and different studies are available in literature comparing the efficacy of endoscopic treatment with surgical management. It still remains a challenge for pediatric urologists.

Pearce et al. [13], retrospectively analyzed 17 patients with duplex UC with non-functioning upper kidney moiety that underwent heminephrectomy leaving the UC intact, showing that 53% of them needed a second surgery (endoscopic or surgical).

On the other side, Castagnetti et al. [11] published a review of 41 patients with UC in a duplex system who underwent endoscopic or surgical decompression of UC before one year of age. These data evidenced the efficacy of surgical decompressive treatment in 9/9 patients, while patients submitted to endoscopic approach required a second treatment in 28% of cases (9/32 pts).

Boucher et al. assessed the need for a second look after the primary endoscopic approach and found out a re-intervention rate of 61% (before 2002) and 42% (after 2002) for ectopic UC in duplex system and 42% (before 2002) and 10% (after 2002) for intravesical UC. Comparing their data with those of patients who underwent heminephrectomy, Authors concluded that endoscopic decompression is as effective as heminephrectomy in the treatment of UC.

Instead, Wang et al. [14] affirmed that a single procedure is rarely sufficient, and most of the patients require multiple procedures before they are 'treatment free'.

Moreover, the majority of studies that compare surgical and endoscopic approach, present some bias, because of the selection of the patients: in fact, the endoscopic decompression is often used for patients with VUR or if heminephrectomy is contraindicated. According to the literature, the surgical approach with heminephrectomy and/or excision of ureterocele is reserved only in few selected cases [14].

For these reasons, we decided to evaluate the efficacy of only the endoscopic approach, comparing the post-operative outcomes after the treatment of intravesical and extravesical duplex UC.

In our series the percentage of therapeutic success after a single endoscopic treatment was 77% (20 out of 26 patients) in group A and about 91% (20 out of 22 patients) in group B.

After a second endoscopic treatment we observed a 92% of healing in group A (24 out of 26 patients) and 100% in group B.

We considered as a therapeutic success both infection control, looking for preserving global kidney function, and the elimination of obstruction as well as the management of VUR. Based on that, our success rate after single or double endoscopic treatment is higher than data reported in literature.

After endoscopic treatment, none of our patients with ectopic UC in duplex system required subsequent surgery such as nephrectomy or heminephrectomy, nor ureteral derivations, and none of them showed infectious signs or

symptoms related to pyelonephritis, even those with a non or poorly functioning upper kidney moiety.

In our experience endoscopic treatment seems to be safe and effective. One of the main advantages of endoscopic puncture is the early decompression of UC and, therefore, a lower risk of developing pyelonephritis.

The lower approach to the duplex system UC can reduce the indications for partial nephrectomy even in non functioning kidneys if well drained. For this reason, we avoided an upper tract approach in all of our patients with no refluxing upper pole.

Regard on intravesical UC, several studies highlighted the effectiveness of decompression in more than 90% of cases after endoscopic puncture, with an incidence of iatrogenic VUR ranging between 0 and 10%.

In case of ectopic UC instead, success rate varies between 67% and 96% with a iatrogenic VUR rate between 27% and 56% [2].

According to recent literature, the high percentage of endoscopic "failures" are represented by the presence of a non-functioning hemi-kidney and the association with high grade VUR which represent the two main indications to surgical treatment after effective endoscopic decompression [15–23].

Although no long-term studies are available, Chertin et al. reported no complications associated with a non-functioning renal moiety left in situ after a 9-year follow-up [24], while Levy et al. reported only one case of high blood pressure over 115 patients treated with UC in duplex system effectively related to the scars of the lower pole of the contralateral kidney rather than the dysplastic upper pole [25].

In our sample, the incidence of post-operative VUR is lower than in literature: 23% in group A, of which only 19% new onset VUR, 5% in group B, with no case of new onset VUR without statistically significant differences between the two groups. About 33% of VURs spontaneously resolved, while the others were effectively treated with endoscopic meatoplasty, with total resolution of the VUR in medium-to-long-term follow-up.

Regard on VUR, Adorisio et al. reported a spontaneous resolution in 13/19 patients (68%), in a group of 46 patients underwent to endoscopic incision alone and also several recent studies show an high rate of asymptomatic VUR even after the suspension of prophylaxis [19].

Castagnetti et al. performed post-operative VCUG only in patients with persistent hydroureteronephrosis or UTI; in case of persistent but asymptomatic high-grade VUR, the patient was treated with prophylactic antibiotic therapy unless changes in the clinical picture or parental decision. Authors reported only 2 out of 41 patients (5%) requiring surgical treatment for symptomatic VUR (with recurring UTI). Castagnetti concludes that surgical indication after effective endoscopic decompression treatment should be given only in case of symptoms despite the presence of a VUR or a non-functioning upper pole and that the main goal of UC treatment should be an adequate decompression [11].

A bias in the evaluation of older cases is the different endoscopic technique used for ectopic UC, which, according to some Authors, could have increased the incidence of postoperative complications, such as iatrogenic VUR [26].

This hypothesis is supported by a recent meta-analysis that highlights that ectopia is not a risk factor for endoscopic decompression failure in patients with duplex system [27]. Over the years, endoscopic treatment improved thanks to the upgrade of technical devices and physiological knowledge; therefore we assisted to a reduction in the incidence of the major complication: the new onset VUR into the ureterocele moiety after incision.

The present study has some limitations: it is a retrospective and monocentric serie and it lacked a longer follow-up; on the other hand, it has been conducted on a quite large sample size and it is one of the few studies that compares the endoscopic treatment between orthotopic and ectopic UC.

## Conclusions

Our data report primary endoscopic puncture of ureterocele as a simple, effective, and safe procedure also in long-term follow up. This technique avoids the need for additional surgery in the majority of the patients, also in the case of an ectopic UC associated to a duplex system. In our experience, endoscopic UC puncture has success in 77% of cases of the ectopic UC with a single procedure and in 92% after second look. Moreover, endoscopic incision is associated with a low incidence of postoperative reflux into the affected moiety, that usually resolves spontaneously.

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