# Endourological treatment of ectopic ureterocele: our experience in the last 15 years

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#### ABSTRACT

**Objective.** To assess the efficacy of the endourological treatment of ectopic ureterocele in children in a large series and with a long-term follow-up.

**Materials and methods.** A retrospective, descriptive study of patients with ectopic ureterocele who had undergone surgery in our institution in the last 15 years was carried out. All patients were treated using an endourological approach, both for ureterocele and postoperative vesicoureteral reflux (VUR).

Results. 40 patients were treated -55% with left involvement and 5% with bilateral involvement. Mean age at diagnosis was 4.97 months, with diagnosis being established prenatally in 54.1% of cases. In all patients but one, endourological puncture of the ureterocele was conducted. Mean age at surgery was 6.96 months (0-1.11). Surgery was performed on an outpatient basis in 94.9% of patients. No perioperative complications were recorded. In the last 30 patients, preoperative voiding cystourethrography was not carried out. 72.5% of patients had postoperative VUR (44.8% into the upper pyelon, 10.3% into the lower pyelon, 17.2% into both, 6.9% into the contralateral system, and 20.7% into the bilateral system), but it was resolved with a single endoscopic procedure in 48.1% of cases (65% of patients were healed with two procedures). VUR was not endoscopically resolved in 3 patients who required ureteral re-implantation. 6 patients required heminephrectomy (n=3) or nephrectomy (n=3) as a result of functional impairment and infections.

**Conclusion:** The endourological treatment of ectopic ureterocele is a little aggressive and little invasive technique that allows the obstruction to be resolved on an outpatient basis, which means bladder surgery –if required– can be performed outside the neonatal period.

**KEY WORDS:** Ectopic ureterocele; Endourology; Minimally invasive surgery.

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### TRATAMIENTO ENDOUROLÓGICO DEL URETEROCELE ectópico. Experiencia en los últimos 15 años

#### RESUMEN

**Objetivo.** Evaluar la eficacia del tratamiento endourológico del ureterocele ectópico en niños en una serie amplia y con seguimiento a largo plazo.

**Material y métodos.** Estudio retrospectivo descriptivo de los pacientes con ureterocele ectópico intervenidos en nuestro centro en los últimos 15 años. Todos los pacientes se tratan por vía endourológica, tanto el ureterocele como el reflujo vesicoureteral (RVU) postoperatorio.

Resultados. Se trataron 40 pacientes, 55% eran izquierdos y 5% bilaterales. La edad media al diagnóstico fue de 4.97 meses siendo de diagnóstico prenatal el 54,1%. En todos los pacientes menos uno se realizó una punción endourológica del ureterocele. La edad media en el momento de la cirugía era de 6,96 meses (0-1,11). La cirugía fue ambulante en un 94,9% de los pacientes. No se registraron complicaciones perioperatorias. En los últimos 30 pacientes no se realizó cistouretrografía miccional preoperatoria. Un 72,5% de los pacientes presentaron RVU postoperatorio (44,8% a pielón superior, 10,3% a pielón inferior, 17,2% a ambos, 6,9% al sistema contralateral y 20,7% bilateral), pero este se resolvió con un único procedimiento endoscópico en un 48,1% de los casos (curación del 65% de los pacientes con dos procedimientos). El RVU no se resolvió de forma endoscópica en 3 pacientes que requirieron un reimplante ureteral. Seis pacientes precisaron heminefrectomía (n= 3) o nefrectomía (n= 3) por anulación funcional e infecciones.

**Conclusión.** El tratamiento endourológico del ureterocele ectópico es una técnica poco agresiva invasiva que consigue la resolución de la obstrucción de forma ambulante permitiendo diferir la cirugía vesical (si fuera necesaria) fuera del periodo neonatal.

PALABRAS CLAVE: Ureterocele ectópico; Endourología; Cirugía mínimamente invasiva.

# **INTRODUCTION**

Ureterocele is an infrequent cause of ureterovesical junction obstruction. In spite of the many articles published on this pathology, there is little consensus in terms of management. Regardless of the approach used, baseline treatment should alleviate ureteral obstruction, preserve renal function and continence, and prevent urinary tract infections (UTIs) and vesicoureteral reflux (VUR).

In this respect, there are three main approaches available –high reconstruction (heminephrectomy or ureteropyelostomy), full reconstruction (with ureterocele removal and ureteral re-implantation), and the endoscopic approach (endourological puncture). Even though ureterocele has been traditionally managed through open surgery, the development of endourology in the last years has turned it into one of the techniques of choice in this pathology<sup>(1)</sup>.

The objective of this study was to assess the efficacy of the endourological treatment of ectopic ureterocele in children in a large series and with a long-term follow-up.

# MATERIALS AND METHODS

A retrospective study of patients with ectopic ureterocele who had undergone surgery in our institution in the last 15 years was carried out. All patients with ureterocele associated with the upper pyelon of a duplex system were included, with the position of the ureteral meatus being non-orthotopic –either intravesical or not (nomenclature according to Glassberg et al.<sup>(2)</sup>).

Demographic variables (sex, and age at diagnosis and surgery), laterality, associated malformations, diagnosis, type of surgery, postoperative stay, postoperative UTIs, postoperative VUR, postoperative renal function, need for subsequent urinary tract surgeries, and follow-up time were collected. The information was gathered from patient documents and electronic medical records.

For over 15 years, the diagnosis and treatment protocol in our institution has included solely ultrasound diagnosis, without preoperative scintigraphy or voiding cystourethrography (SVCU). The approach is endourological in all cases.

The endourological treatment of ectopic ureterocele is carried out using a 9.5Fr cystoscope. The bladder and the urethra are explored. If the ectopic ureteral meatus is identified, the ureterocele is unroofed from the meatus to the bladder neck. If the meatus is not identified or cannot be guided, the ureterocele is widely punctured at the intravesical level. Both procedures are performed using a high power (30W) 3Fr monopolar electrode or a 270  $\mu$ m Holmium laser fiber, at the chief surgeon's discretion. No bladder or ureteral catheters are left in place postoperatively. The procedure is carried out on an outpatient basis.

In the postoperative period, serial control ultrasonographies as well as SVCU or ultrasound cystography are conducted. If the patient has VUR, it is endourologically treated up to twice. If VUR has not been resolved, a scintigraphy is performed. If the upper pyelon or both pyelons show functional impairment, heminephrectomy or nephrectomy are scheduled, respectively. However, if their function is preserved, ureteral re-implantation or uretero-ureterostomy are carried out.

The statistical analysis was conducted using the IBM SPSS Statistics software, version 22. Qualitative variables were expressed as percentage and absolute number, whereas quantitative variables were expressed as mean and confidence interval.

# RESULTS

Over the study period, a total of 40 patients underwent surgery. 75% (30/40) were female, 55% (22/40) of cases were left, and 5% (2/40) were bilateral. 3 patients had non-urological associated malformations –tetralogy of Fallot, aortic arch hypoplasia, and interventricular communication. In 37.5% (15/40) of patients, other urological malformations associated with the presence of ureterocele were found –8 contralateral renoureteral duplications (20%), 6 contralateral urinary tract dilatations (15%), 1 contralateral primary obstructive megaureter, 1 ectopic ureter on a non-duplicated system, and 1 ectopic ureter on a duplicated system without ureterocele.

Mean age at diagnosis was 4.97 months (0-11.2), with 89.5% of patients being in the first 6 months of life. 54.1% had been prenatally diagnosed. The remaining patients were diagnosed in the context of UTIs. 2 patients who had been prenatally diagnosed had an episode of preoperative UTI.

Ultrasound diagnosis was carried out in all cases, with 65% of patients having dilatation of the upper pyelon, 32.5% of both pyelons, and 2.5% of the lower pyelon.

All patients but one underwent ureterocele surgery. This patient was a 10-year-old girl with repeated UTIs, ureterocele associated with VUR, and functional impairment of the upper pyelon. Therefore, decision was made to endoscopically treat VUR only, which allowed clinical signs to disappear. Mean age at surgery was 6.96 months (0-13.3), with 79.5% of patients being under 6 months old. In 80.5% of the patients, ureterocele puncture was carried out at the bladder level, and in 19.5%, opening was conducted from the ectopic meatus.

94.9% of the patients underwent surgery on an outpatient basis. No perioperative complications were recorded. Only 1 patient required a second puncture as a result of persistent urinary tract dilatation at ultrasonography (de-obstruction rate: 97.5%).

In the follow-up period, 42.5% of the patients had at least one episode of UTI with fever, with a mean of 0.94 episodes (0.49-1.38) per patient, 69.2% of which required admission. In the SVCU or ultrasound cystography results, VUR was found in 72.5% (29/40) of the patients –44.8% into the upper pyelon, 10.3% into the lower pyelon, 17.2% into both pyelons, 6.9% into the contralateral system, and 20.7% bilaterally. At scintigraphy, 34.6% of the kidneys



had an adequate radiotracer uptake, with hypofunction of the upper pyelon in 46.2% of cases, and of the whole kidney in 19.2%. None of the patients has developed arterial hypertension in the follow-up period or has urinary incontinence.

All VUR patients but one underwent VUR surgery. This patient had a voiding dysfunction that was repaired with biofeedback, which allowed VUR to disappear. All patients who required VUR surgery underwent STING. In 3 patients only (11% of those who underwent VUR surgery), endoscopic healing was not achieved, so they required ureteral re-implantation. Owing to the functional impairment of the upper or both pyelons associated with repeated UTIs, 6 patients (15% of the series) required heminephrectomy (n=3) or nephrectomy (n=3).

Patients required a mean of 2.40 (1,9-2,8) surgeries –including the baseline procedure. With two endouro-logical procedures (de-obstruction and VUR treatment), 62.,% of the cases were resolved, and with two endoscopic treatments of VUR, 68.9% of VUR cases were healed. The patient flow diagram is featured in Figure 1.

Mean patient follow-up was 6.68 years (5.1-8.2).

# DISCUSSION

The classic management of ectopic ureterocele implies complex surgeries on neonatal patients. In addition, these surgeries may not offer definitive results and are not free from complications. Re-intervention rates of up to 10-62% have been reported in high reconstruction, 21.3% in the solely bladder approach, and 15-17% in the combined approach<sup>(3,4)</sup>. According to the literature, the rate of complications can be as high as 10%, and postoperative VUR can be present in 4-44% of the patients<sup>(3,4)</sup>.

Endourological surgery emerged in an attempt to conduct surgery in a less aggressive fashion. It was initially conceived as an intermediate step that would allow obstruction to be alleviated in order to conduct definitive surgery at an older age. Puncture and intravesicalization have success rates of 60-100%<sup>(3,5-17)</sup> of de-obstruction according to the technique used. However, as a result of the frequent occurrence of postoperative VUR, a large number of patients require a second open surgery procedure. In the first years of experience, 40-80% of the patients used to require other surgeries<sup>(3,5,10-12,15)</sup>, but thanks to the advances made in minimally invasive techniques and material, these rates have declined to 18-63%(1,6,8,9,15-18) (31.1% in our series, including re-implantations, nephrectomies, and heminephrectomies). Regarding the endoscopic technique, no differences have been demonstrated between unroofing and puncture, or between the monopolar scalpel, the laser, and the cold scalpel<sup>(13)</sup>. In a meta-analysis carried out in 2006<sup>(19)</sup>, the risk of re-intervention was only impacted by preoperative anatomy (ectopic vs. intravesical ureterocele, duplicated vs. simple system, and presence or not of preoperative VUR).

Postoperative VUR remains the main issue of concern once obstruction has disappeared. In our series, the rate of postoperative VUR was similar to that found in the literature, ranging from 30 to 90%<sup>(1,3,5,9,11-15,17,20)</sup>. The rate of postoperative VUR is probably influenced by the fact SVCU or ultrasound voiding cystography are systematically performed in all patients, regardless of the lack of clinical signs, contrarily to other authors who only conduct them in the presence of UTI<sup>(1,9,12)</sup>. On the other hand, in our institution's protocol, SVCUs are not carried out preoperatively but postoperatively, since the presence of VUR -which has been previously reported to range from 35% to  $65\%^{(1,3,6-10,12-17)}$  does not change the initial therapeutic approach. Consequently, it is uneasy to determine which percentage of such reflux is *de novo* and which one was present from the beginning or emerged as a result of the anatomical changes occurred following ureterocele de-obstruction. Regardless of its origin, we believe VUR management should be endourological, since it has demonstrated to be successful in 68.5% of patients. Even though many authors posit VUR has a natural tendency to resolve spontaneously<sup>(6-8,12,14-17)</sup>. This is also the case in our department, where surgical management of VUR is preferred in order to prevent UTIs with fever that may compromise renal functionality, especially considering it is a little invasive therapy with high effectiveness rates in the treatment of VUR. This is shown in the low need for uretero-vesical surgery found in our study (11% of patients undergoing VUR surgery, 7.5% of the total) vs. 9-70% in the literature. It should be mentioned that baseline endoscopic procedures allow bladder surgery to be carried out outside the first months of life, which facilitates ureterocele removal and re-implantation, both due to the size of the patient and to the fact ureteral remodeling is no longer required<sup>(3,5,16,20)</sup>. Indeed, no incontinence issues were observed in any of our patients.

A similar principle can be applied to the need for resection surgery of the renal parenchyma. Contrarily to what has been classically believed in terms of need for heminephrectomy in cases of little or non-functioning pyelons as a result of the theoretical risk of malignization and hypertension, the preservation of asymptomatic non-functioning pyelons is increasingly frequent<sup>(3,10,14,16)</sup>. With a heminephrectomy or nephrectomy rate of 15%, none of the patients in our series has developed arterial hypertension after a follow-up period of 6.8 years, even though 65.4% of pyelons had low or no uptake at renal scintigraphy. In a study by Adorisio et al.<sup>(16)</sup>, the upper pyelon was found to provide 4-8% of renal function. Following heminephrectomy, a mean of 1.25% was lost, whereas after endoscopic incision, it increased by 2.25%. Consequently, even little functioning pyelons should be preserved in asymptomatic patients, since heminephrectomy inevitably implies the resection of healthy renal parenchyma -in other studies, up to 6% of lower pyelon function<sup>(18)</sup>.

# CONCLUSIONS

The endourological management of ureterocele is a little invasive technique that can be applied neonatally in expert hands. It is a very little aggressive approach which allows for highly effective ureteral de-obstruction, with a success rate close to 100%. Although VUR is a frequent complication, it is often resolved using minimally invasive techniques, with bladder surgery being necessary in very few cases –and if it is required, it can be conducted outside the neonatal period, with a less dilated ureter, which facilitates re-implantation.

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