



Could surgical experience of adult endourologist overcome the learning curve of retrograde intrarenal surgery in children?

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Abstract

With the increasing of the prevalence of pediatric urolithiasis (1–5%), retrograde intrarenal surgery (RIRS) is emerging as preferred option for the management of stones in pediatric patients. Although the principles of RIRS developed in adults can be applied in children, also expert adult endourologists feel uncomfortable to approach young patients due to long learning curve that usually is expected to be required in this particular setting. The aim of the study was to compare peri- and postoperative outcomes of RIRS in pediatric and adult patients performed by a single surgeon expert in adult endourology (> 500 RIRS) with no experience in pediatric urology. Data on patient characteristics of 30 consecutive patients (15 adults and 15 children) undergoing RIRS at our institution were collected retrospectively from January 2016 to October 2018. Mean age for the pediatric group was 11.8 years (IQR 8–16) and for the adult group was 56 years (IQR 49–58). No significant differences between the two groups in terms of peri- and postoperative outcomes were found. The most common complication was hematuria in 2/30 patients (1 children vs 1 adults) and fever 2/30 (1 pediatric patient vs 1 adult) ($p = 1.00$) that required antibiotic treatment (Clavien Dindo 2). Median length of stay was 1 day (IQR 1–1 days) in both groups ($p = 1.00$). Stone-free rate was 86.7% in children and 80% in adults ($p = 0.624$). Our preliminary experience suggests that expert adult endourologist can manage successfully also pediatric cases with results comparable to adults and low complication rate.

Keywords Pediatrics · Urolithiasis · Renal stone · Ureterscopy · RIRS

Introduction

Stone disease is an important increasing clinical problem in pediatric urology practice. According to the most recent guidelines, extracorporeal shockwave lithotripsy (SWL) is the treatment of choice for renal stones up to 2 cm and has a corresponding recommendation grade of percutaneous nephrolithotomy (PCNL) for those between 1 and 2 cm [1, 2]. However, PCNL provides excellent results for the renal stone more than 1 cm thanks to its advantages such as single-step dilation and sheath placement, small skin incision, good working access for pediatric instruments, variable length,

and lower cost [3]. Indeed, PCNL thanks to the development of holmium laser, which has replaced open approach with a stone-free rate (SFR) over the 85% reported in the current literature in pediatric age population [3, 4, 5]. Otherwise, in adult population, European Association of Urology (EAU) guidelines suggest retrograde intrarenal surgery (RIRS) as a valid option for stones up to 2 cm with the routinary practice of this procedure in this setting of patient [2].

On the other hand, as reported by Suliman et al., there is an increasing use for RIRS also in pediatric stones disease treatment [6]. Different articles reported shorter hospital stay, lower radiation exposure and complication rate related to RIRS when compared to PCNL, especially for stones between 10 and 20 mm [7, 8, 9]. Evidences suggest that, although urinary stones are less prevalent in children than in adults, they are associated with significant morbidity and incidence is increasing [10]. In this background, a growing demand of surgeons able to face with RIRS in children is gaining attention considering that, in common practice, adult's endourologists appear reluctant in approaching this disease in pediatric age population due to the belief it may

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be challenging even for expert operators and require a specific learning curve. This often leads to forced uses of SWL and PCNL, which are not lack of complications [10, 11, 12].

The aim of this study is to evaluate the safety and feasibility of first series of RIRS performed in children by an expert adult's endourologist compared to procedures performed in adults in the same period: is a learning curve necessary in this specific setting?

Materials and methods

After institutional review board approval and informed consent from all the individual participants included in the study were obtained, we retrospectively reviewed preoperative and peri-operative data of 15 consecutive pediatric patients (group A) compared to a group of 15 consecutive adult patients (group B) affected by renal stone disease who underwent RIRS between January 2016 and October 2018.

Since July 2015 an interhospital center was created between the pediatric hospital and the adult hospital for the technology improvement of pediatric urology. In this setting, every complex pediatric case of urolithiasis was discussed in a multidisciplinary team involving a pediatric urologist, a pediatric nephrology, a radiologist and an endourologist or a mini-invasive surgeon of the adult field. All procedures were performed by the same surgeon who had a long expertise in adult renal stone disease (over > 500 RIRS completed).

Preoperative radiologic investigation was performed in adult patients with computer tomography (CT) urography. Pediatric patients had ultrasound or at least radiogram to establish localization and dimension of stones. The stone burden was calculated, as before reported, in mm² according to the multiplication of the two dimensions of the stone as seen in the preoperative plain abdominal radiograph [13].

Our study evaluated the localization of the stone, stone-free rate (SFR), pre-operative double-J ureteral stent implantation, operative time (OT), length of stay, complications (defined according to Clavien–Dindo classification) [14]. All the data were collected by medical doctors.

Categorical, continuous parametric and not-parametric variables were reported as frequencies and proportions or median and interquartile range (IQR). Pearson's Chi squared test was used for dichotomous variables. Median test was used for continuous variables.

Statistical significance in this study was set as $p \leq 0.05$. All reported p values are two-sided. Analyses were performed with SPSS version 20.0 (SPSS Inc, Chicago, IL, USA).

In the pediatric patient, a double-J stent placement is performed to improve the compliance and the dilatation of the ureter and to reduce the possible problem related to the access of the ureteroscopy. For what concern the adult patient we

performed a preoperative stenting only in case of urinary sepsis before RIRS.

The RIRS procedure is performed under general anesthesia with the patient in the dorsal lithotomy position. The bladder is entered either with a cystoscope or a semi-rigid ureteroscope. We used semi-rigid ureteroscope with an outer diameter of 7 Fr and a length 43 cm in the adult patient, instead in the pediatric patient have the same diameter but with a length of 34 cm.

Guidewire is sent to the ureter, under fluoroscopic guidance. Then we advanced the semi-rigid ureteroscope through the ureter under direct vision until the proximal ureter or the pelvic junction is achieved. After that, the semi-rigid ureteroscope is removed and the ureteral access sheath (UAS) is placed under the fluoroscope guidance with the auxilium of the guidewire. In the adult patient the outer diameter of the UAS was 11 Fr with a length of 35 cm and in the pediatric cases the outer diameter was 9.5 Fr and the length was 20 or 28 cm according to the age and height of the children. The flexible uretero-rensocope, the same in all the patients, is advanced directly under fluoroscopic control. All collecting system is observed under direct vision until the stone is found. Sometimes fluoroscopic vision or addition of a contrast agent can facilitate access to the stone.

A 30-W Holmium:YAG (Ho:YAG) laser generator was used for stone treatment both in adult and children. In both the cases, specific settings for dusting ($0.5 \text{ J} \times 15 \text{ Hz}$) or fragmentation ($1.0 \text{ J} \times 8 \text{ Hz}$) technique were set. When the stones are located in the lower calyx dislodgement in pelvis is usually performed. A fragmentation technique is preferred for harder stones; for the softer stones, a dusting setting is chosen. When the dusting setting was chosen, the stone is fragmented with the laser until clinically insignificant residual fragments (< 2 mm) are left. If stone analysis is desired or if the stones are fragmented, the little stone fragments are retrieved with a 1.7-Fr or 2.2-Fr basket catheter. At the end of the procedure, a double-ureteral stent is always placed.

If a residual stone up to 50 mm² is found after 70 min in the pediatric patient or 90 min in the adult patient, a new procedure is directly scheduled.

Normally, the SFR was evaluated with ultrasound and radiogram performed 2 weeks after surgery, if residual stones (up to 50 mm²) were found, a new RIRS was scheduled and we considered SFR not achieved.

Double-J stent was removed under general anesthesia in pediatric patients, while the adult patients underwent to the procedure in the outpatient department 3 week after surgery.

Results

There was no statistically significant difference between RIRS in group A and group B regarding gender, laterality, side of the stone, length of stay ($p = 1.000$), and OT

(group A: mean = 70, IQR 60–80; group B: mean = 80, IQR 63–105; $p = 0.466$). The data are collected and summarized in Table 1. We performed 1 (6.7%) and 5 (33.3%) SWL in the group A and B, respectively ($p = 0.068$), however, none of the procedures recorded a sufficient fragmentation to avoid a surgical procedure. 1 (6.7%) young patient experienced a previous RIRS; 2 (13.3%) patients of the group B had a PCNL in their medical history and 1 (6.7%) performed a RIRS, however, no statistical difference is recorded ($p = 0.283$).

While the mean size for stone surface area in group A was 90 (IQR 80–144) mm², for group B, it was 100 (IQR 90–165) mm². There was no statistically significant difference between the two groups regarding stone surface area ($p = 0.137$).

When the two groups were compared regarding SFR, no statistically significant difference was found ($p = 0.624$). In fact, 13 out of 15 patients in group A (86.7%) and 12 patients in group B (80%) were stone free after the procedure. All the patients, 2 (13.3%) in the pediatric and 3 (20%) in the adult group, respectively, who not achieve the SFR after the first procedure, were submitted to a second RIRS who gained the stone free without need of ulterior treatment.

There was no statistical difference between the two groups regarding complications ($p = 1.000$). Hematuria was noticed in one patient for both groups and fever occurred

in one patient for each group requiring antibiotic treatment (Clavien Dindo 2) and a longer length of stay (3 days).

We noticed statistically significant differences between the two groups only for preoperative double-J stent placement: it was placed in 8 out of 15 children patients (53.3%) in group A and 2 out of 15 adult patients (13.3%) in group B ($p = 0.02$).

Discussion

Evolution of treatment of urinary tract stones has followed innovations in surgical technology and expertise. Since the first paper on the use of ureteroscopy (URS) for stone treatment in children have been published by Ritchey et al. in 1988 [15], ureteroscopic management has become increasingly common in pediatric stone patients [16]. Recently, RIRS has gained attention as an effective method in the proximal ureter, collecting duct system, and, especially, lower calyx calculi with less invasiveness compared to PCNL [17]. The introduction of the European Working Time Directive in 2003 saw an increasing demand for focus on surgical competence through means such as learning curve identification [18]. An evidence-based definition of ‘learning curve’ has not yet been established, however, with regards to surgery, the learning curve is thought of as a representation of the improvement of a surgeon’s performance over time [19]. In

Table 1 Patients’ characteristics

	Group A (children; $n = 15$)	Group B (adult; $n = 15$)	p value
Gender, n %			0.256
Male	8 (53.3%)	11 (73.3%)	
Female	7 (46.7%)	4 (26.7%)	
Side, n %			1.000
Right	7 (46.7%)	7 (46.7%)	
Left	8 (53.3%)	8 (53.3%)	
Site of the stone, n %			0.296
Pelvis	7 (46.7%)	3 (20%)	
Upper calyx	4 (26.6%)	3 (20%)	
Medial calyx	2 (13.3%)	4 (26.6%)	
Lower calyx	2 (13.3%)	5 (33.3%)	
Previous surgery for stone treatment, n %	1 (6.7%)	3 (20%)	0.283
Age, median; IQR	11; 8–16	56; 49–58	0.605
Preoperative swl, n %	1 (6.7%)	5 (33.3%)	0.068
Preoperative stent, n %	8 (53.3%)	2 (13.3%)	0.020
Stone size mm ² , median; IQR	90; 80–144	100; (90–165)	0.137
Operative time, median; IQR	70; 60–80	80; 63–105	0.466
Length of stay, median; IQR	1; 1–1	1; 1–1	1.000
Complications, n %	2 (13.3%)	2 (13.3%)	1.000
Stone-free rate after first surgery, n %	13 (86.7%)	12 (80%)	0.624

IQR interquartile rate, Swl extracorporeal shockwave lithotripsy, mm millimeter

adult's endourology, efforts have been made to evaluate feasibility and efficacy of PCNL and URS focusing, moreover, on their learning curve. Quirke et al., indeed, reviewed 14 papers concerning PCNL and four papers focused on URS to identify learning curves in adult urolithiasis surgery. They outlined the learning curves for URS as 60 cases for operative time, 56 cases for fragmentation efficacy and 100 for complication rates [20]. Surprisingly, no learning curve has been evaluated in the literature for pediatric population regarding RIRS. In fact, the majority of studies reporting the learning curve of a procedure are related to oncological surgery in adult population [19], and in pediatric age population, the few existing concerns minimally invasive laparoscopic and robotic surgery [21]. In pediatric endourology, the number of cases is undoubtedly much lower than those in adult's field and there is a common sense of being inexperienced when facing with young patients. As such, learning curve in urolithiasis procedures for pediatric age population is poorly assessed. It is commonly thought that RIRS is relatively more difficult to perform in children due to the narrower space that makes it more challenging to maneuver the instrument. Two other key issues in this setting are the stone dust which can obscure vision after 30–45 min of fragmentation and the higher complications risk of pediatric kidney subjected to increased intrarenal pressure [22]. Moreover, while in adults, detailed preoperative imaging with CT scanning is the norm, in children ultrasounds is the preferred imaging modality and this could result in less anatomical information for surgeons and lack of stone characteristics at the moment of surgery [23].

Despite these limitations, the increasing demand for pediatric stone management in our daily clinical practice as tertiary referral center leads adult's surgeons to perform RIRS in this subset of patients without a high expertise in children. Our study wants to compare OT, length of hospital stay, stone size, complications and SFR after first surgery between the first 15 series of RIRS performed in children and 15 comparable RIRS performed in adults in the same period time by a single expert surgeon. The operator has previously reached the plateau in his learning curve performing more than 500 procedures in adults [20]. A similar study design has been conducted by Bayrak et al. that evaluated 70 cases of children undergoing PCNL by a surgeon who had performed 120 PCNLs on adults [13]. They divided the population in 2 groups of 35 consecutive patients: in group 1, mean age was 11 (range 1–16), while in group 2 was 8 (range 1–16). No significant difference was found in SFR between the two groups ($p=0.11$) [13]. The study demonstrated that an expert surgeon may effectively perform PCNL in young patients with comparable results even in a younger subset of population after the first approach.

In our study SFR was not significantly different between group A (86.7%) and group B (80%) ($p=0.62$). Moreover,

SFR in pediatric group was found in accordance with what was reported in the literature for series performed by other pediatric surgeons [8, 24, 25, 26]. According to Berardinelli et al., surgical experience influences the outcomes of RIRS especially in terms of safety. In this study they compared OT, complication rate, SFR in patients submitted to RIRS by surgeons with different expertise (< 100 procedures and > 400) and showed that, while SFR was comparable in the two groups, OT and complications were significantly lower in patients treated by more expert surgeons [27].

Our study confirmed these findings and reported no major complication (Clavien > 2) neither in pediatric nor in adult population. Median operative time was found shorter in children series perhaps due to the fact that preoperative stenting was performed more frequently in children as previously reported [28, 29].

Preoperatively, the presence of ureteral stent has been found statistically different when compared in both the groups. In our clinical practice, we preferred, when possible, double-J stent placement in children to improve the compliance and the dilatation of the ureter and to reduce the possible problem related to the access of the ureteroscopy. Furthermore, passive dilatation using a double-J stent could reduce long-term complications as it has been theoretically proposed that active dilatation of the ureteral orifice may cause reflux in children [30].

Moreover, a key issue of our strategy was the multidisciplinary approach of the complex pediatric cases: since the 2015, an interhospital center, involved the adult hospital and the pediatric hospital, allows to discuss the difficult cases and bring the know-how and the instrument of the adult world, as advanced endoscopic technology and the expertise of one adult endourologist to the pediatric field.

In conclusion all these findings showed that, although pediatric RIRS remains challenging, surgeons who have achieved high expertise in adult's field could confidently approach pediatric age population with efficacy and safety comparable with adults, even in his first series, as seen for other surgical procedures [31, 32].

However, our study has some limitations that we should report. This is only a preliminary experience and, although the number of cases performed by the surgeon is equal, it remains a small series. Anyhow the aim was to compare the outcomes of the initial procedures performed in children by an expert surgeon with those performed in the same period time in adults to encourage more endourologists to approach pediatric patients with more confidence in an era when stone diseases is becoming a real social problem also in children.

Conclusions

Our findings show that, after a good management in a multidisciplinary team, the RIRS is a safe and efficacy treatment in children when performed by an expert adult's endourologist with no previous experience in pediatric field. More studies are needed to confirm these findings but with a certain grade of confidence, a proper learning curve to approach stone disease in children is not strictly needed as long as the surgeon has reached his learning curve in adult age population.

Author contributions SS, AT, AC and LM conceived the original idea; AAG recorded the data; SS performed the statistical analysis; SS, LM and CC reviewed the final paper. All the authors contributed to the final manuscript.

Compliance with ethical standards

Conflict of interest The authors declare no conflict of interest.

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