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Development of ureteroneocystostomy in pediatric population: ten years of experience from Cipto Mangunkusumo National Referral Hospital, Jakarta, Indonesia

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ABSTRACT

Background: Ureteroneocystostomy is one of the most common surgery in pediatric urology. The procedure can be performed either laparoscopically or in open surgery fashion. Currently, there is limited data available regarding the outcome of pediatric ureteroneocystostomy in Indonesia. The aim of this study is to evaluate the efficacy and safety of pediatric ureteroneocystostomy in both laparoscopic and open surgery techniques in one of the largest referral hospitals in Indonesia.

Methods: This is a retrospective study in which all patients with age of 18 or below underwent ureteroneocystostomy in Cipto Mangunkusumo general hospital was included in ten years period. The outcome analyzed was the patients' demographic profile, procedure duration, blood loss, and length of stay of the patients.

Results: A total of 46 patients underwent ureteroneocystostomy from year 2010 to 2020 with 38 patients underwent open surgery and 8 patients underwent laparoscopic surgery. The median age of the patients was 5 years old, which predominantly female (60.9%). The most common urology condition was vesicoureteral reflux and ectopic ureter at 34.8%. The median procedure duration was significantly shorter in the open surgery than laparoscopic group (180 and 325 minutes, respectively; p=0.002). There was no significant difference in blood loss and length of stay between two groups.

Conclusion: The preferred ureteroneocystostomy technique used was open surgery, with the most common etiology of the patients underwent the procedures were vesicoureteral reflux and ectopic ureter. In addition, the open surgery approach also leads to a shorter procedure duration than laparoscopy.

Keywords: ectopic ureter; ureteroneocystostomy; vesicoureteral reflux.
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INTRODUCTION

Reconstructive surgery in the form ureteroneocystostomy has been widely accepted in the field of pediatric urology.[1,2,3] It is currently being advocated for different clinical indications, including embryologic malformation of the ureter (e.g., duplex ureter, ectopic ureter, ureterocele, primary congenital anomaly related with primary reflux, and etc) trauma (i.e., iatrogenic or non-iatrogenic), and stenosis or obstruction, and other factors involving the lower ureter and ureterovesical junction (UVJ).[2,3] The surgical procedure has been associated with good results in a robust number of published reports, and is available with diverse approaches and techniques, making it a highly developing subject that had undergone multiple improvements and modifications.[2,4,5]

Currently, available approaches for ureteroneocystostomy include open, laparoscopic, and robotic approaches, which are reported to be equally effective.[5] Open procedures showed 95.1% success rate per patient and 95.9% per ureter, while laparoscopic and robotic procedures resulted in a range of 95-99% success rates.[5,6] Although high, the surgery's success is highly operator-dependent and influenced by the varying selection of techniques available.[2,5,6] Current known techniques are divided as extravesical (e.g., Lich-Gregoir, Barry, Hodgson-Firlit-Zaons, and Paquin), intravesical (e.g., Politano-Leadbetter, Glenn-Anderson, Cohen, and Gil Vernet), and combination of both.[2,5,6] Different techniques may be more suitable for different conditions. Therefore operator expertise and clinical judgment serve as crucial factors associated with associated outcomes.[6,7] This suggests that the treatment patterns and outcomes should be different in different centers that provide ureteroneocystostomy.

In recent days, there are limited published data in the experience of ureteroneocystostomy, especially in pediatric age-group in Indonesia. Because this information should be relevant for

evaluation and improvements for future clinical practice in pediatric urology, this study would like to elaborate the past experience of ureteroneocystostomy in pediatric patients in Cipto Mangunkusumo National Referral Hospital, as well as showcasing the perioperative and post-operative outcomes resulting from the selections of procedures done by the surgeons.

METHODS

Design and samples

The study was a retrospective cohort study, investigating on all pediatric patients aged below 18 years-old who had ureteroneocystostomy performed in Cipto Mangunkusumo National Referral Hospital (RSCM) from year 2010 until 2020. The sampling method was total sampling, covering all pediatric cases documented in January 2010 until January 2020.

Inclusion and exclusion criteria

All patients undergoing ureteroneocystostomy done by the Department of Urology RSCM during 2010-2020 documented in RSCM local database who consented to be part of the study were included. All patients with no or inadequate follow-up records will be contacted for a call interview, and failure to achieve adequate information for data collection would be excluded.

Data collection

The study collected data from local database from Cipto Mangunkusumo National Referral Hospital and medical records. The data collected from patients' status included age, gender, diagnosis, duplex ureter status, prior treatment for the ureter, surgical side, approach of ureteroneocystostomy, and technique of ureteroneocystostomy; perioperative variable including anesthesia, operation duration, blood loss, and post-surgical length of stay; post-operative complications including persistent reflux, obstruction, diverticulum, leakage, infection, and urinary retention.

Data analysis

All data analysis was done using SPSS v.23 (IBM Statistics, New York). Comparison of open and laparoscopic approaches in their outcomes reported as non-parametric numerical variables (e.g., operation duration, blood loss, post-operative length of stay) was analyzed using Mann-Whitney test.

RESULTS

A total of 46 pediatric patients with median age of 5(1-18) years underwent ureteroneocystostomy were recorded during year 2010 until 2020 (Table 1). Indications for ureteroneocystostomy are classified as 3 groups. The highest number of cases found to be the indication for ureteroneocystostomy were vesicoureteral reflux (VUR) and ectopic ureter, followed by stenosis of the ureter. About a quarter of the cases had prior treatment such as nephrostomy and/or endoscopic treatment.

The comparison between the open and laparoscopic procedures can be seen in Table 2. One case of laparoscopic case in 2016 was converted into open, intraoperatively, which we did not include in the laparoscopic group. The reason of the conversion was the inability to perform ureteral tailoring at the time. More patients had been operated using the extravesical approach in comparison with intravesical. All intravesical approach was done in patient underwent bilateral ureteroneocystostomy.

Some patients need additional procedures due to the complexity of the case. Most of additional procedure performed was ureteral tailoring. It was performed in order to make a good tunneling or antireflux implantation, especially in tortuous ureter. Psoas hitch performed in a condition where the ureteral length was not adequate to do the reimplantation. One patient underwent bladder augmentation and Mitrofanoff procedure due to its complex diagnosis (small bladder capacity). Urethroplasty was performed in one patient with female hypospadias. The sigmoid mucosal laceration was identified in one patient in open procedure group and counted as intra-operative complication.

Post-operative complications between open and laparoscopic techniques can be seen in Table 3. There are 4 patients in open ureteroneocystostomy group that had late complications. Two patients had persistent VUR and were identified due to recurrent fever and UTI years

Table 1. Characteristics of the pediatric patients

Characteristics		Values
Age (years)		5 (1-18)
Gender Ratio		
	Male	18 (39.1%)
	Female	28 (60.9%)
Etiology		
	VUR	16 (34.8%)
	Stenosis of the ureter	14 (30.4%)
	Ectopic ureter	16 (34.8%)
	- Single collecting system	3 (18.75%)
	- Double collecting system	12 (75%)
	- Triple collecting system	1 (6.25%)
Side of Ureter		
	Left	22 (47.8%)
	Right	13 (28.3%)
	Bilateral	10 (21.7%)
	Allograft	1 (2.2%)
Prior Treatment		16 (34.8%)
Type		
	Open	38 (82.6%)
	Laparoscopic	8 (17.4%)

Table 2. Comparison between open and laparoscopic techniques in their perioperative variables

Procedures	Total	Open (n = 38)	Laparoscopic (n = 8)	p-value*
Extravesical				
Lich-Gregoir	42 (91.3%)	34 (89.5%)	8 (100%)	
Intravesical				
Politano-Leadbetter	4 (8.7%)	4 (10.5%)	-	
Additional Procedures **				
Psoas Hitch	1 (2.2%)	1 (2.6%)		
Ureteral Tailoring	12 (26.1%)	10 (26.3%)	2 (25%)	
Circumcision	5 (10.9%)	5 (13.1%)	-	
Bladder Augmentation + Mitrofanoff	1 (2.2%)	1 (2.63%)	-	
Urethroplasty	1 (2.2%)	1 (2.63%)	-	
Intra-operative complication				
No Complication	45 (97.8%)	37 (97.4%)	8 (100)	
Complication	1 (2.2%)	1 (2.6%)	-	
Operation Duration (minutes)	210 (120-450)	180 (120-450)	325 (240-390)	**0.002**
Blood Loss (ml)	50 (5-300)	50 (5-300)	30 (15-50)	0.05
Post-operative Length of Stay (days)	4 (2-20)	4 (2-20)	5 (2-8)	0.436

*Statistical analysis for operation duration, blood loss, and post-operative length of stay were using Mann-Whitney test for nonparametric variables.
** More than one additional procedure can be performed in one operation.

Table 3. Comparison between open and laparoscopic techniques in their complications

Procedures	Total n (%)	Open n (%)	Laparoscopic n (%)
Early			
Leakage	0	0	0
Infection	0	0	0
Urinary Retention	0	0	0
Late*			
Persistent reflux post-surgery	3 (6.5)	3 (7.9)	0
Ureterovesical obstruction	2 (4.3)	2 (5.3)	0
Diverticulum	0	0	0
Total	**5 (10.9)**	**5 (13.1)**	**0**

*More than one complication can occur in one patient

after surgery. One patient had UVJ partial stenosis and was identified due to progressive hydronephrosis during follow-up. The last patient with single kidney had both persistent VUR and partial UVJ stenosis and was detected due to decreased renal function parameters. All partial UVJ stenosis patients were treated by DJ stent insertion.

Among the complications, the highest number found were persistent reflux followed by ureterovesical obstruction. Further elaboration showed that the persistent reflux post-surgery was due to coexisting neurogenic bladder dysfunction.

DISCUSSION

Forty-six pediatric patients which had the procedure of ureteroneocystostomy were reported by our study showing a median age of 5 (1-18) years with 14:9 female to male ratio. Upon the ureter that has undergone reimplantation, mostly were documented on the left ureter, a quarter of the cases had duplicity of the collecting system, and a quarter had priorly been given treatment to overcome the problem. Amongst the indications that were present, the highest number of cases were due to VUR and ectopic ureter, mostly in the form of double collecting, followed by ureteral stenosis.

The low number of pediatric patients undergone ureteroneocystostomy from ten years of period in Cipto Mangukusumo National Referral Hospital is in accordance of the low occurrences of the underlying problems requiring ureteral reimplantation, such as VUR, in children.[2] It is also caused by the limitation of diagnostic tools, that not all hospitals can perform VCUG or renogram. Although the clinical indications for ureteroneocystostomy also included stenosis of the UVJ or distal ureter, trauma (often iatrogenic), VUR, fistulas, and malignancy, most of the cases requiring ureteroneocystostomy in children are related to VUR.[1,2,8] Our study has showed that the diagnosis that is related with VUR, namely double collecting system, ectopic ureter, and congenital anomaly associated with primary reflux covered large portion of the indication for ureteroneocystostomy in Cipto Mangunkusumo Hospital. The reported age that was found in our study showed that more than a half of the recruited

patients were aged below 4, indicating that more patients came to seek help in earlier years of their childhood. This is according to the theory whereas most problems such as VUR are asymptomatic, in addition less severe cases of primary VUR are associated with high rate of spontaneous resolutions over time.[1] The number of older children undergoing ureteroneocystostomy were present with indications such as double collecting system and stenosis of the UVJ, problems that may not resolve on their own that progressed overtime gradually.[1] There was an interesting finding in gender, where more patients in this study were female. Although previous studies suggested that reflux in younger patients is to be more common in males, our study included other indications for ureteroneocystostomy aside from VUR therefore such result occurred.[9]

Our study showed higher number of open procedures in comparison with laparoscopic procedures (38 vs 8 cases). The practice of laparoscopic ureteroneocystostomy was firstly done in RSCM in 2015. This may underlie the lower number of laparoscopic procedures in comparison with open. At first pediatric instruments for laparoscopy were not available, therefore it was rather difficult to operate especially in smaller children. It was not until year 2019 that the pediatric laparoscopic instruments are available in RSCM, which is in accordance with the high increase of laparoscopic procedures done in 2019.

RSCM does not perform non-surgical initial intervention for the indicated underlying disease using bulking agent. The sole reason for such condition, is the non-availability of bulking agents in Indonesian market, leaving surgery as the only options for the patients.

More procedures are done with extravesical approach in comparison with the intravesical approach (42 vs 4). All of the extravesical approaches that were done in Cipto Mangunkusumo hospital was Lich-Gregoir technique, while all of the intravesical approaches that were done was Politano-Leadbetter technique. There are 2 known approaches for the procedure for laparoscopic ureteroneocystostomy: transperitoneal (extravesical) and pneumovesical (intravesical). Both approaches are generally accepted. However, we adopted the transperitoneal approach, using Lich-gregoir technique which was done in all 8 of the laparoscopic patients. There is no significant difference in safety and efficacy in known literatures, although the result is very operator-dependent and should get better with more experience in performing the procedure.[8,10,11]

As surgery is the gold standard for most of the clinical indications mentioned above, the general principles of ureteroneocystostomy are the adequate ureter mobilization to preserve the vascularity, tunneling in the bladder wall, wide enough to accommodate an optimal length of the lower ureter and to provide a reinforcement to the implanted ureter with generous detrusor muscle support.[10] As also reported in our study, conventional method of ureteroneocystostomy is done with open approach.[1-3] The success rate of available procedure is 95.1% rate per patient and 95.9% per ureter, therefore the approach is generally chosen by the surgeons.[5,6] However, with the increase of laparoscopy expertise, the laparoscopic approach of ureteral reimplantation has been introduced and is associated with high success rate, similar to open procedure.[5-6,10-11] Our center has also done about 8 cases using the laparoscopic approach. Although the preference lies within the surgeon's expertise and experience, extravesical approaches are reportedly associated with lower morbidity and shorter hospital stay than intravesical techniques.[12] Similarly, our center has implemented more extravesical procedures over the period of 10 years, which is in accordance to the suggestion given by prior studies.[13,14] Based on previous reports, Lich-Gregoir, compared to the other extravesical approaches, is often preferred in children due to its high rate of success, less complications, and recovery of hydronephrosis.[10,15-17] Our center has also practiced the Lich-Gregoir method for all cases treated with extravesical approach.

The complications that occurred in our study were low and only occurring in open ureteroneocystostomy. The highest problem occurring in our study was persisting VUR post-operatively. This is however, was found to be caused by coexisting neurogenic bladder dysfunction that prevented total resolution. This is in accordance to the literature, whereas primary and secondary cause of VUR may occur together in one patient.[1,2]

Our study also did a brief comparison between open and laparoscopy approaches in ureteroneocystostomy. As also reported in other studies, our study did not show any notable differences between the two approaches in terms of the perioperative variables and complications, although no cases of complication were reported in the laparoscopic group.[4-6] The laparoscopic approach was found to be longer in duration, which may be due to the high difficulty of the procedure. The laparoscopic approach was also found to show lower blood loss in comparison with open procedure. Further studies are needed to compare the two approaches, as the number of patients in our study is too small to undergo statistical analysis comparing the two treatment approaches.

CONCLUSION

This study reported 46 cases of pediatric ureteroneocystostomy operated by urologist from Department of Urology, Cipto Mangunkusumo National Referral Hospital, from year 2010-2020. Among those cases, most were indicated due to VUR and ectopic ureter (mostly double collecting system). Open and laparoscopic ureteral reimplantation was done in our center. More extravesical approach was done in our center, in the form of Lich-Gregoir technique with low complication occurrences.

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COMPETING INTEREST

The authors declare there is no competing interest to disclose.

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ETHICAL STATEMENT

This study has been approved by Ethical Committee Faculty of Medicine, Universitas Indonesia/Cipto Mangunkusumo National Hospital, Jakarta, Indonesia.

AUTHOR CONTRIBUTION

All author had contributed for writing the original draft of the manuscript and agree for the final version of the manuscript for publication.

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