

## Original Research Article

# Revisiting ureteral substitution and its outcome in children: single centre 13 year experience

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## ABSTRACT

**Background:** Objective of this study is to explore various ureteric substitutes in pediatric age group and their outcomes.

**Methods:** Retrospective analysis was done from 2003-2016, of all patients operated in this hospital that had undergone ureteral replacement. Thirteen such patients (5 from initially published paper (1) and 8 new patients) were followed up to find conduit patency, renal function and related complications with their outcomes assessed.

**Results:** Ureteric substitution was done in 8 patients. Age of the patient ranged from 4 months to 8 years. Out of these for 2 patients monti tube was created; one with colon and other jejunum, rest of the 6 patient appendix was used. On follow up one patient had early appendico-ureteral leak requiring re-anastomosis and one patient had partial obstruction at 6 months corrected by dividing mesentery and untwisting appendix. With a mean follow up of 3.8 years all patients have preserved renal functions and drainage present. Also 5 patients of ureteral substitution performed at this institution and published prior were followed up with mean follow up of 10.4 years having no complaints with preserved function and unobstructed drainage.

**Conclusions:** This small series supports that not only appendix and ileum but even colon as well as jejunum should be considered as Monti's tube for ureteric replacement, when confronting with short ureter in paediatric age group.

**Keywords:** Monti tube, Ureteral substitution, Ureteric substitutes

## INTRODUCTION

Ureteral defects can be too long; requiring non-urinary tissue to overcome it. Such cases present a surgical challenge to provide low pressure drainage while avoiding urinary stasis and reflux. Since 1912, when appendix was introduced for substitution and gained wide popularity, authors have progressed to use small and even large intestines for it.<sup>1</sup> Creating an illeal ureter was first described by Schoemaker in 1906.<sup>2</sup> Years later it was modified and Young Monti's principle was used to do away with its drawbacks. Majority of this experience, as is evident from literature is mainly based on studies and case reports in adults.

Ureteric replacement in children has been reported infrequently. Authors present a single centre experience of various ureteric substitutes in 13 of this paediatric patients requiring same. In 5 cases the cause was iatrogenic: 3 after pyeloplasty for PUJO (pelviureteric junction obstruction), 1 ureteric injury during modified Duhamel procedure and 1 after pyelostomy for sepsis in case of PU valves).

Other 3 cases; 2 were due to VUJ (vesico ureteric junction obstruction) and one missed lower stricturous ureter in an operated case of pyeloplasty for PUJO. Other than these 5 patients from this previously published paper were followed up.

## METHODS

From 2003-2016, 13 patients had undergone ureteric replacement in this hospital. 8 patients were males and 5 were females, with their age ranging from 1 month to 9 years. For ureteric replacement colonic Monti's tube was used in 1 patient; Jejunal Monti's tube was used in 1 patient, ileal Monti's tube was used in 1 patient and remaining 10 patients appendix was used. All patients underwent thorough preoperative workup for diagnosis and renal function. These were then followed up for patency and drainage along with renal function and clinical outcome.

Amongst these in one patient antenatal ultrasound (USG) was suggestive of (s/o) mild left hydronephrosis and hydroureter presented at 1.5 year age at a peripheral hospital with high grade fever and pain in left side of abdomen. On investigating, USG was s/o left renal pelvis antero-posterior diameter as 2cm with moving echoes and an EC renogram showed left renal function as 10 %, he underwent a left pyelostomy for same. He was then referred at this centre at age of 2.5 years. His renal function tests and hemogram were normal, and renal scan s/o left renal function improved to 35%. Authors planned a definitive surgery. Intraoperatively through the pyelostomy, upper ureter could be cannulated only up to 5cm and on cystoscopy lower ureteric opening could not be cannulated. As the appendicular length was small and it being left sided, colon was used as Monti's tube and sutured above to the pelvis and lower end implanted into bladder extravasically to bridge the 8cm gap in between. A nephrostomy tube along with a trans- anastomotic tube was kept in situ. 10 days later trans- anastomotic tube was removed and a dye study done; patency and free flow were confirmed and then the nephrostomy tube was removed. On immediate follow up and now at 4 years after surgery DTPA scan shows preserved function with non-obstructive drainage. Renal profile is normal.

Similarly in a patient diagnosed with Posterior urethral valves; at 3 months age underwent fulguration of valves along with right pyelostomy for urosepsis at a peripheral centre other details of the same were not available. He then presented at 8 years with severe pain in left flank region. USG was s/o left renal multiple large stones for which he underwent laparoscopic pyelolithotomy and a nephrostomy tube kept in situ. He had no voiding complaints and micturating cystourethrogram showed no evidence of reflux or bladder abnormality. Dye study through nephrostomy was s/o pelviureteric junction obstruction (PUJO). Authors then posted him for a definitive surgery to attempt pyeloplasty, intraoperatively there was PUJO with upper third of ureter long stricture. Adhesiolysis was done for peri-pelvic dense adhesions; pelvis freed and repaired and a Jejunal Monti's tube interposed between the pelvis and lower ureter. 10 days later trans-anastomotic tube was removed and a dye study done; patency and free flow confirmed and then the nephrostomy tube removed along with drain removed.

One month later Right pyelostomy closure was done. On immediate follow up and now at 4 years after surgery DTPA scan shows preserved function with non-obstructive drainage. Renal profile is normal. No voiding complaints.

In all other 6 patients Appendix was used as ureteral substitute. Wherein a 7 months girl after ureteral appendicular substitution a week later on dye study showed non passage of dye but nephrostomy showed decreasing trend of output so 3 days later catheter was clamped intermittently; then nephrostomy output further decreased and on day 14 when a repeat dye study showed good drainage then the nephrostomy tube was removed. On immediate follow up and now at 3.5 years after surgery DTPA scan shows preserved function with non-obstructive drainage. In another patient, 1 month old girl with appendicular ureteral substitution done and had an uneventful postoperative course with free drainage across. On f/u at 6 months on scan there was obstructed pattern on drainage. She required exploration, which showed partial obstruction due to twisting of appendix by drag on mesentery subsequently appendicular artery was divided not compromising its vascularity through the neovascularisation developed. Six weeks later the DJ stent was removed and repeat scan showed preserved function with non-obstructive drainage. In another 3 years old boy having strictured left ureter after Duhamel surgery for Hirschsprung, underwent appendicular ureteral substitution. On day 5 of surgery there was excessive output in drain and dye study through nephrostomy showed there was leak at uretero-appendiceal junction. He was re-explored and uretero-appendiceal anastomosis refashioned obliquely. Repeat dye study on 7<sup>th</sup> day showed no leak and smooth passage of dye. PCN was then removed. On immediate and 5 years follow up renal functions are maintained and drainage is present.

These 8 cases and their outcomes are summarised in Table 1 and 2. In this study other five patients who were published prior, on further 9 years follow up have no complaints with repeat scans showing preserved renal function and drainage is present.<sup>1</sup> This protocol for evaluation in follow up was to perform ultrasound after removal of stent at 3 months and then 6 monthly. Along with scan at 6 months and yearly thereafter till 5 years postoperatively and at puberty. Repeat dye studies and early ultrasound or scan is to be done if any complaints or complication.

## RESULTS

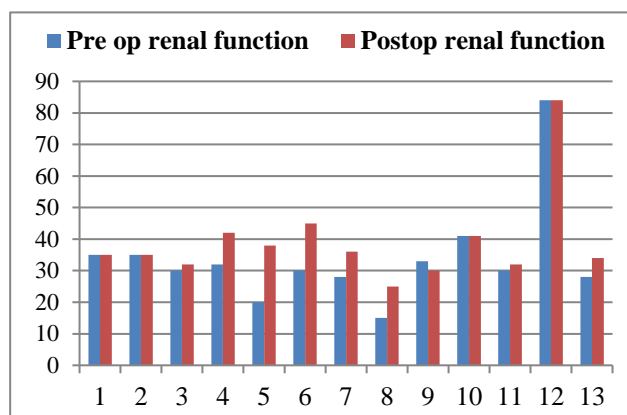
Out of 13 patients undergoing ureteral substitution at this institute, 8 patients were newly included and 5 patients were followed up from previous study. In these 8 patients age of the patient ranged from 4 months to 8 years. Out of these for 2 patients Monti tube was created; one with colon and other jejunum, in rest of the 6 patients appendix was used.

**Table 1: Demographics with case presentation and their evaluation.**

Case no.	Age	Sex	Pre presentation History	Diagnostic investigation	Gap
1	2.5 yrs	M	Failed left (LT) pyeloplasty at 1.5 yrs age with Lt pyelostomy done at peripheral center	Intraoperatively could not cannulate proximal ureter beyond 5cm + cystoscopy: ureteric opening could not be cannulated	8cm
2	8 yrs	M	Operated case of PU Valves, with Right(Rt) pyelostomy for urosepsis with Lt renal calculus	On dye study no passage of dye beyond PUJ Intraoperative : upper 1/3 <sup>rd</sup> ureter stricture	6cm
3	7 months	F	Failed pyeloplasty done at 5 months with PCN in situ	PCN gram non passage of dye beyond PUJ Intraoperative proximal ureter stricture, distal patent	4cm
4	3 yrs	F	Horshoe kidney	Intraoperative dye study : non passage of dye beyond proximal 2/3 <sup>rd</sup> ureter + cystoscopy ureteric opening could not be cannulated	5cm
5	1 yr	M	Case of Rt Vesicoureteric junction obstruction with end ureterostomy done for sepsis	Intraoperatively ureteric length inadequate	4cm
6	4 months	F	Failed Rt pyeloplasty at 1 month age with end ureterostomy done for sepsis for lower stricturous ureter	Previous surgical findings: deficient lower ureter	5.5cm
7	5 yrs	M	Rt failed pyeloplasty at 6 months age	Intraoperative finding of upper 1/3 <sup>rd</sup> ureter absent	5cm
8	3 yrs	M	At 2.5 yrs Modified Duhamel for Hirschsprung's disease; b/l pyonephrosis PCN done. Lt staghorn calculus	PCN gram: Rt side obstructed beyond midureter	6cm

**Table 2: Ureteric substitutions, their complications along with drainage and function on follow up.**

Case No.	Defect	Substitution	Post-operative complications	Preoperative renal function	Renal function and drainage on Follow up
1	8cm entire Lt ureter	Colonic Monti tube	None	Before pyeloplasty 10%, After pyeloplasty 35%	35% at 4 yrs f/u drainage present
2	6cm Rt upper 1/3 <sup>rd</sup> ureter	Jejunal Monti tube	None	After pyeloplasty 35%	35% at 4 yrs f/u drainage present
3	4cm RT upper 1/3 <sup>rd</sup> ureter	Appendix	At 1 week postop nephrostomogram showed non passage of dye, but clamped after 3 days and output decreased and hence tube removed.	Pre PCN 12% Post PCN 30%	32% 3.5 yrs f/u drainage present
4	5cm Lt lower 2/3 <sup>rd</sup> ureter	Appendix	None	32%	42% at 2 yrs f/u drainage present
5	4cm Rt lower ureter	Appendix	None	20%	38% at 4.5 yrs f/u drainage present
6	5.5cm Rt lower ureter	Appendix	Delayed at 6 months partial obstruction due to twisting of appendicular mesentery: divided as neovascularisation present	30%	45% at 3 yrs f/u drainage present
7	5cm Rt upper 1/3 <sup>rd</sup>	Appendix	None	28%	36% at 4.5 yrs f/u drainage present
8	6cm Rt lower 1/2 of ureter	Appendix	Early postoperative Uretero-appendiceal anastomotic leak re-explored and re-anastomosed	15%	25% at 5 yrs f/u drainage present
Follow up of previously Presented patients					
9	Lt entire ureter	Appendix	none on F/U	33%	30% at 8.8 yrs f/u drainage present
10	Rt distal 1/3 <sup>rd</sup> ureter	Appendix	none on F/U	41%	41% at 10 yrs f/u drainage present
11	Rt ureter	Illeal Monti Tube	none on F/U	Unobstructed drainage	32% at 10.5 yrs f/u drainage present
12	Rt lower ureter	Appendix	none on F/U	84%	Asymptomatic 12 yrs f/u drainage present
13	Rt upper ureter	Appendix	none on F/U	Unobstructed drainage	Asymptomatic 11 yrs f/u drainage present



**Figure 1: Comparison of preoperative and postoperative renal function.**

On follow up one patient had early appendico-ureteral leak requiring re-anastomosis and one patient had partial obstruction at 6 months corrected by dividing mesentery and untwisting appendix. With a mean follow up of 3.8 years all patients have preserved renal functions and drainage is present across. Also 5 patients of ureteral substitution performed at this institution and published prior were followed up with mean follow up of 10.4 years having no complaints with preserved function and

unobstructed drainage. The renal function in all patients stayed constant or improved as documented in scan except in 1 patient showing a 2% initial drop in function, however has unobstructed drainage on follow up dye studies (Figure 1). These preoperative renal functions in all 13 patients were compared with their renal function on longest follow up. These were then subjected to paired T test and the difference was found to be significant (Table 3, 4, 5).

**Table 3: Table 3 Paired samples statistics.**

Renal Function	Mean	N	Standard Deviation	Std. Error Mean
Preop Renal function	33.92	13	16.414	4.552
Postop Renal function	39.15	13	14.479	4.016

**Table 4: Paired sample correlations.**

Renal Function	N	Correlation	Sig
Preop Renal Function	13	0.918	0.000
Postop Renal function			

**Table 5: Paired samples test.**

	Paired Differences				T	dt	Sig (2 tailed)	
	Mean	Standard Deviation	Standard error of mean	95% Confidence Interval of the Difference				
				Lower				Upper
Preop Renal function - Postop Renal function	-5.231	6.521	1.809	-9.171	-1.290	-2.892	12	0.014

**DISCUSSION**

Long defect of ureter in adults and its substitution by appendix or small and large bowel is frequently reported. However in children there is very sparse literature, where mainly only appendix has been used that too in the form of isolated case reports and very few studies. This prompted us to report these 13 patients operated at this centre where various substitutes were used and outcomes assessed with long and short term follow up in paediatric patients.

In adults ureteric defect may result from chronic inflammatory diseases such as tuberculosis, retroperitoneal fibrosis, iatrogenic injuries during open and endourological surgery, neoplasms and radiation damage, and very rarely trauma.<sup>3,4</sup> The etiology in children is mainly due to congenital ureteric stenosis or strictures and iatrogenic, after pyeloplasty or reimplantations and other procedures. Rarely is it seen

due to tumor or trauma. Factors which influence the choice of the surgical procedures depend upon: renal back pressure changes, functional integrity, data of the defect as regards (site of the defect, length of the defect, the extent of the peri-ureteric fibrosis, whether the defect: is primary or recurrent as well as the presence of complications such as stones or urinary fistula), and surgeon's preference. The goals of intervention in ureteral defect correction are to preserve renal function and eliminate ureteric obstruction.<sup>3</sup>

Regarding surgical correction, short ureteral defects can easily be managed by end-to-end anastomosis or reimplantation. For longer distal defects, a pedicled flap of bladder wall (Boari's flap) with or without psoas hitch can be considered. Ileal loop replacement of the ureter for very long stricture or extensive ureteral loss is suitable. Appendicular and illeal tube have been used historically but even Jejunal and Colonic Monti's tube can be used with similar technique and avoiding the drawbacks of

previous option as is evident from this study.<sup>3,5</sup> Appendix although appears a gold standard and ideal for right sided replacements owing to its adequate narrow lumen for substitution in children and its good mesentery and pedicled blood supply there are limitations to it.<sup>6</sup> Sometimes inadequate length of appendix and its pedicle or its twisting or drag on trying to use it on left side can limit its usage. Also incrustations and stone formation have been reported with it.<sup>4,7</sup> This prompts us to use its alternative, a bowel loop segment. Initially a whole segment was used as a substitute but due to its drawbacks of lumen discrepancy, chances of reflux, excessive mucus formation, infections, loss of larger length of bowel and metabolic disturbances; now creating a tube from smaller bowel segment using a young Monti's principle is preferred.<sup>8-10</sup>

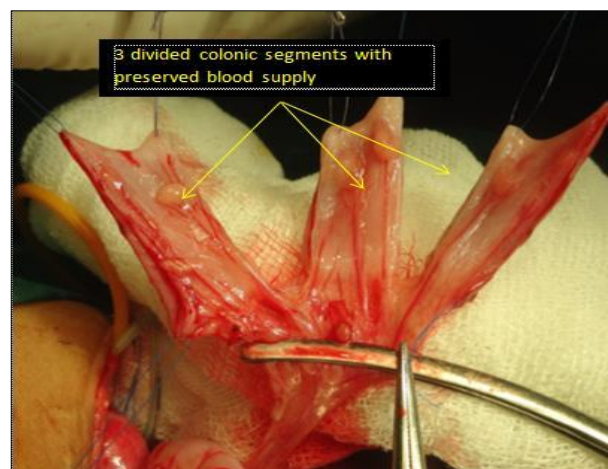
### **Technique and advantages and drawbacks**

The appendix has an irregular lumen that is approximately 8 French in diameter and the blood supply arises from the appendicular artery, which is a branch of the ileocolic artery, located in the mesoappendix. Appendiceal interposition is technically easy and the appendix can be readily mobilized with its blood supply.<sup>3</sup> The length-to-diameter ratio of the appendix, its contractility, and its peristalsis prevent urinary stasis.<sup>1</sup> Another advantage, is its small surface area, which results in negligible urine absorption, and this explains the lack of serum electrolyte abnormalities. The appendiceal lumen corresponds in caliber to that of the ureteral lumen and this allows secure anastomosis with the proximal or distal ureter. The location of the appendix facilitates mobilization and replacement of the upper, mid, or lower ureter.<sup>3</sup> The use of the appendix allows complete retro peritonealization of the anastomoses to both the ureter and the bladder. Authors have used appendix as a substitution in 10 of this patients where 8 patients had no complications and 1 had early complication of grade III Clavien-Dindo classification who required re-exploration and refashioning of anastomosis at uretero-appendicular junction. Second patient developed delayed partial obstruction due to twisting of appendix by drag on mesentery which required exploration and dividing appendicular artery not compromising its vascularity through the neovascularisation developed.

But the appendix is unavailable or unsuitable for reconstructive purposes when it is surgically removed beforehand or is too short to bridge the existing anatomic gap or sometimes when a defect is left-sided and mesentery of appendix is not long enough.

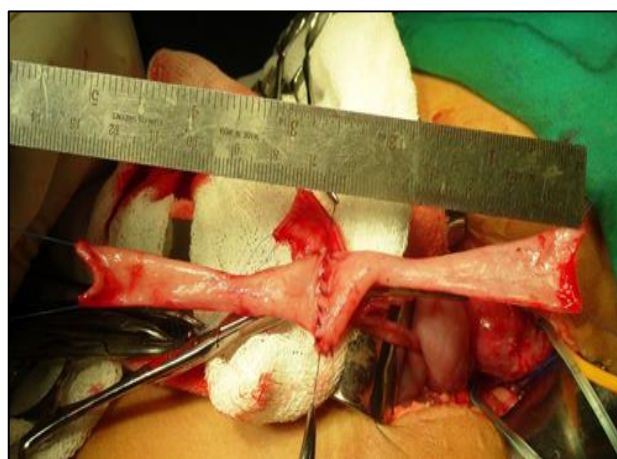
In such situations, although technically demanding bowel substitution is required and authors should be aware of its principles and technique. Around 5-7.5 cm of the terminal ileum or colon or jejunum was isolated on their vascular bed, according to the length required to compensate the ureteral defect. Then, according to the Yang-Monti principle, the isolated segment was further

subdivided into 2-3 equal parts (each 2.5cm - 3cm in length) with preservation of the individual blood supply (Figure 2).



**Figure 2: Colonic segments with preserved blood supply.**

The continuity of the bowel was re-established. Each segment was incised at its longitudinal axis close to the mesenteric border. Unfolding of the incised segments with suturing their adjacent ends were performed and resulted in the formation of an intestinal plate 2.5cm wide and 12-18cm long (according to the number of isolated segments) (Figure 3).



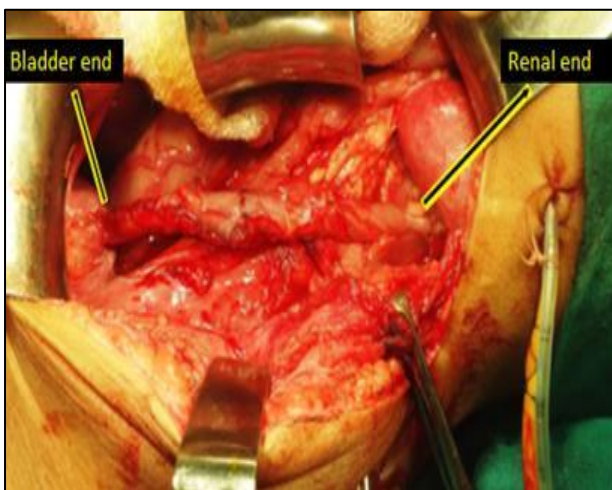
**Figure 3: Formation of intestinal plate.**

This plate was tubularized around 10 french infant feeding tube using 4/0 absorbable sutures (Figure 4). The proximal end of the tube was anastomosed to the ureter or renal pelvis while the distal end was anastomosed either to the bladder using a non refluxing Lich-Gregoir technique (Figure 5) or to the spatulated cephalic end of the lower ureter in cases with the presence of an intact healthy lower ureter. At the end of the procedure, a transanastomotic tube is brought out along with nephrostomy especially when pelvis was anastomosed at upper end or DJ stent is kept in situ and a urethral

catheter were left in. If there was excessive dissection or pelvis sutured, a drain was left in situ and output monitored. After 10 days trans-anastomotic tube was removed and a dye study was done through nephrostomy to confirm free passage and then drain and finally nephrostomy is removed. If DJ stent was kept it was removed after 6 weeks, then a scan is repeated or intravenous pyelogram done to confirm free passage and assess function. Authors have used a bowel Monti's tube in 3 of this patients; 1 with ileal Monti's, 1 as Colonic Monti's and 1 patient Jejunal Monti's tube. All 3 patients had no complications postoperatively and on f/u patency were maintained (Figure 6) and renal functions preserved.



**Figure 4: Tubularisation of intestinal plate.**



**Figure 5: Anastomosis of tube at pelvic and bladder end.**

Although the study is of limited number of patients authors found that same Yang Monti's principle can be used to create a colonic and Jejunal tube and used in place of ileum, where it was not available and/ or to avoid excessive mucus formation and metabolic disturbances as also for surgical ease as well as reducing chances of adhesions.

Authors found that use of Yang Monti's principle and using bowel Monti's tube has not been reported in children although it has been adequately used in adults.

Colonic Monti's tube for ureteral substitution is reported in only 22 adult patients none in children and in only three studies published so far. Several studies in adult patients using intestinal Monti's tube for ureteral substitution have reported encouraging results regarding surgical outcomes and renal function. These can be successfully duplicated in children as is evident from this study.



**Figure 6: Postoperative dye study demonstrating free flow across.**

Ureteral substitution itself is a rare requirement in paediatric patients. This explains few studies published on it, and usage of colon or jejunum for this is even rare.

Hence although 13 patients appears a small group, surgical technique and outcome; along with a significant difference in the postoperative renal functions as demonstrated by paired T test on follow up is important to be known.

## CONCLUSION

Authors are hereby reporting various techniques employed in the management of ureteral defects encountered in this series. No single procedure can address every situation and therefore the surgeon must be aware of the various techniques that have been and can be developed to treat such defects. This is the first occasion where a Colonic and Jejunal Monti's tube was used for ureteral substitution in children and is showing a favourable alternative.

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