

ORIGINAL RESEARCH

Clinical Study of Duplex Moiety in Children: A Retrospective and Prospective Study

K.V.Sathyanarayana¹, M. Santhi², Mandakini K.T³, Swapna Palakurthy³

¹Associate Professor, Department of Pediatric Surgery, Osmania Medical College, Koti, Hyderabad, Telangana, India.

²Associate Professor, Department of General Surgery, Osmania Medical College, Koti, Hyderabad, Telangana, India.

³Assistant Professor, Department of Pediatric Surgery, Osmania Medical College, Koti, Hyderabad, Telangana, India.

ABSTRACT

Background: To evaluate the incidence, clinical manifestation and management of duplex moiety in children and also to study the outcome of duplex system in children.

Materials and Methods: Retrospective and prospective study of children who underwent treatment for duplex moiety in a total of 24 cases for a period of 2 years.

Results: Of the 24 patients included in the study, n= 7 (29.1%) were in the age group of <1year, n= 10(41.6%) were in the age group of 1-3 year. Right side was involved more commonly. Upper moiety was more commonly involved in this study. UTI was most common presentation in this study. Out of 24 cases in this study, 22 cases were managed surgically and 2 cases managed conservatively. Cystoscopic puncturing of ureterocele was done in 2 cases of ureterocele in this study (n=2, 8.3%). 2 cases of PUJO were managed surgically by Anderson hynes dismembered pyeloplasty (n= 2, 8.3%). Out of 22 cases managed surgically from 24 cases, open surgeries were done in 17 cases(n= 17,77.27%) and 3 cases were managed by laparoscopically (n=3, 13.6) and cystoscopy was done in 2 cases (n= 2, 9.09%). Out of 22 cases managed surgically, in 20 cases there are no post op complications (n=20), 2 cases had complications(n= 2).

Conclusion: The anatomy and function of the duplex moiety are critical for making management decisions. In comparison to late identification, early detection of the duplex moiety and early management had a positive prognosis.

Keywords: Duplex moiety, Cystoscopic puncturing, ureterocele, Urinary tract infections, complications.

Corresponding Author:Dr. Swapna Palakurthy, Assistant Professor, Department of Pediatric Surgery, Osmania Medical College, Koti, Hyderabad, Telangana, India.

INTRODUCTION

Renal duplications are one of the common anomalies affecting genitourinary tract. Duplex kidney is defined as a renal unit comprised of two pelvicalyceal system and ureters.^[1] Their incidence is about 1% of all live births. Females are affected more commonly than males and this anomaly is bilateral in 17–33% of cases.^[2] The previous and current classifications of duplex kidney are based on its ureter status, which differentiates into two types, complete and incomplete duplex kidney. Affected children may present with, 1. antenatally detected hydronephrosis, 2. continuous urinary incontinence, 3. flank pain from an obstructed moiety, 4. recurrent urinary tract infections (UTI) and vesicoureteral reflux predisposing to renal

scarring and compromised renal function. Most common clinical presentation is UTI (pain abdomen, fever, pyuria). Upper moieties(UM) are associated with ectopic ureters, ureteroceles and non-functioning of UM. Ectopic ureters occur in females two to three times more commonly than in males.^[3] Lower moieties are associated with VUR and pelvi-ureteric junction obstruction. Investigation done to rule out duplex moiety is – now a days antenatal USG. Duplex moiety is being detected early on post-natal period.^[4] Initial investigation is USG followed by VCUG, DMSA/DTPA renogram and MR Urogram. These can be evaluated systemically and can be treated accordingly by medical or surgical intervention.^[5-7]

MATERIALS & METHODS

Retrospective and prospective study of children who underwent treatment for duplex moiety in a total of 24 cases. The study period was for 5 years (December 2015 to December 2020). Retrospective study period was 3 years and prospective study period was 2 years. The study was done in the Department of Pediatric surgery, Niloufer hospital for women and children, Red hills. Niloufer hospital is a tertiary care center for pediatric and maternal cases.

Sampling Technique:

All consecutive children satisfying the inclusion criteria who underwent treatment for duplex moiety will be included in the study.

Preoperative work up was done with routine investigations, Complete blood picture, Complete urine examination, Urine culture and sensitivity, Renal parameters (blood urea & serum creatinine) and specific investigations like Ultrasonography Abdomen & pelvis (KUB), 2. MCUG, 3. DMSA scan/DTPA renogram, 4. IVP, 5. MR UROGRAM.

Post operatively children were followed up by:

USG Abdomen (KUB) - 6 weeks after surgery Followed by 3 months. Followed by 6 months Followed by 1, 2 and 3 years Renal functional status assessed with Blood Urea & serum creatinine one month after surgery or whenever required, DTPA: 6 months and yearly once after surgery till 5 years. MCUG is done at 6 months and yearly after surgery or whenever required to assess grading of reflux, DMSA scan At 6 months and yearly after surgery to assess new scars. Routine urine examination: 1 month after DJ stent removal or as and when required. Simple statistics were used to analyse data. Case sheet proforma and data collected by history and investigations

Duplex moiety was diagnosed in all cases while evaluating for antenatally detected hydronephrosis, children who were presented with documented UTI or with symptoms of urinary incontinence, pain abdomen, fever, pyuria, straining during micturition. Systematic Preoperative work up was done in all cases and treated accordingly either by conservative or by surgical intervention.

The indications for surgery were: (i) Failure of chemoprophylaxis, i.e., recurrent symptomatic UTIs despite continuous medication. (ii) Presence of high-grade (IV or V) VUR, or grade III VUR with progressive scars. (iii) Persistent or progressive hydronephrosis. (iv) Poor or nonfunctioning of upper or lower renal moiety.

v) Decrease in function of moiety by 10% or presence of persistent hydronephrosis or increase in APD. vi) Ureterocele with obstructing features. vii) Ectopic ureter causing incontinence or.

All children underwent cystoscopy on operation table to rule out ureterocele and to know the location of ureteric orifices and also to rule out causes other than duplex moiety causing UTI.

RESULTS

Of the 24 patients included in the study, n= 7 (29.1%) were in the age group of <1 year, n= 10(41.6%) were in the age group of 1-3 year, n =3(12.5%) were in 3-6 years age group, n =2 (8.3%) were in 6-9 years age group and n= 2(8.3%) were in 9-12 year age.

Table 1: Demographic details in study.

Age (Years)	Number of casesN=24	Percentages
<1 yr	7	29.1%
1-3 yrs	10	41.6%
3-6 yrs	3	12.5 %
6-9 yrs	2	8.3%
9-12 yrs	2	8.3%
Gender		
Males	14	58%
Females	10	42%
Laterality		
Left	9	37.5%
Right	12	50%

Out of 24 cases in the study, no of cases involved on left side n=9(37.5%), no of cases involved on right side n= 12(50%), bilateral involvement n=3(12.5%). Right side was involved more commonly. Out of 24 cases, number of cases where upper moiety involved were n= 17 (71 %) and lower moiety involved n= 7 (29%) cases. Upper moiety was more commonly involved in this study.

Table 2: UTI was most common presentation in this study.

Clinical Presentation	Number of casesN=24	Percentages
Antenatally detectedHydronephrosis	8	33.3%
UTI	12	50%
Urinary Incontinence	3	12.5%
Epididymo Orchitis	1	4.1%

Out of 24 cases, Antenatally detected hydronephrosis were n=8(33.3%), UTI were n= 12 (50%), urinary incontinence was seen in n= 3 (12.5%) and n=1(4.1%) case was presented as left side epididymo-orchitis which is a rare presentation.

Out of 24 cases,6 patients presented with ureterocele (n= 6, 25%). Ectopic ureter was seen in 4 cases n=4, (16.6%), Non-functioning of upper moiety was seen in 7 cases n= 7, (29.1%), VUR was seen in 5 cases n= 5, (20.8%), 2 cases presented with pelvi-ureteric junction obstruction n=2, (8.3%). Most common presentation relatively was non-functioning upper moiety in this study.

Out of 24 cases in this study, 22 cases were managed surgically and 2 cases managed conservatively. Out of 22 cases, upper moiety nephroureterectomy was done in 9 cases (n= 9, 37.5%). Open ureteric reimplantation was done in 9 cases (n=9, 37.5%). Cystoscopic puncturing of ureterocele was done in 2 cases of ureterocele in this study (n=2, 8.3%). 2 cases of PUJO were managed surgically by Anderson hynes dismembered pyeloplasty (n= 2, 8.3%). Lastly 2 cases (n= 2, 8.3%) of low grade VUR were managed conservatively by antibiotics after urine culture and sensitivity along with prophylactic antibiotics. Out of 22 cases managed surgically from 24 cases, open surgeries were done in 17 cases (n= 17,77.27%) and 3 cases were managed by laparoscopically (n=3, 13.6) and cystoscopy was done in 2 cases (n= 2, 9.09%).

Out of 22 cases managed surgically, in 20 cases there are no post op complications (n=20), 2 cases had complications (n= 2). One case had superior pole collection after laparoscopic

upper moiety nephroureterectomy and one had post op urinary leak after ureteric reimplantation.

DISCUSSION

This study was done in the department of paediatric surgery at Niloufer hospital with a study period of 5 years 2015 - 2020 (3 years retrospective and 2 years prospective). A total number of 24 cases were included. The incidence of duplex moiety was 0.24%. The most common age group falls under 1-3 years of age. Youngest age at presentation was 2 months, and oldest age child was 11 years. Males are more commonly involved. Out of 24 cases 14 cases (58%) were of male gender and 10 cases (42%) were female. According to the literature most common gender involved in duplex moiety is female. In one study done by Sri Ram Krishnamoorthy et al in 2016, nearly 60% of the patients were females and bilateral in 10% of the cases.^[5]

In this study right side was involved most commonly about 12 cases (50%), left side involved was around 9 cases (37%). Bilateral involved were 3 cases (13%). In the study done by Krishnamoorthy et al the side of involvement was right side and bilateral involvement was in 10% cases.^[5]

In this study upper moiety involvement is associated with complications like ureteroceles, ectopic ureteric insertion, dysplastic changes or non-functioning kidney in renal parenchyma. Lower moiety was associated with VUR and PUJO. Upper moiety was involved in 17 cases (71%) and lower moiety involved was in 7 cases (29%).

In this study the most common clinical presentation was with UTI, out of 24 cases UTI was presented in 13 cases (54.1%), according to the literature the most common presentation is also the UTI. Eight cases (33.3%) presented as antenatally detected hydronephrosis which were followed up post-natally to have duplex moiety and 3 cases (12.5%) presented with urinary incontinence. In Whitten et al study 75% of cases were diagnosed antenatally by USG and followed up post-natally to have duplex moiety. Out of 24 cases 7 cases (29%) presented as non-functioning of upper moiety, which is the most common mode of presentation in this study.

Out of 24 cases 6 cases (25%) presented with ureteroceles in this study. Shanker et al study shows ureteroceles 27% of them are asymptomatic, they were followed up for 8 yrs. – none of them developed symptoms or required any intervention. In our study isolated ureteroceles were seen in 2 cases which presented bilaterally and underwent cystoscopic puncturing, 4 cases were associated with VUR. Two cases of recurrent UTI, after evaluation found to have ureteroceles. In the 4 cases presented as – ureteroceles with VUR, excision of ureteroceles and common sheath reimplantation done.

According to literature, 80% of ureteroceles are associated with the upper pole duplex system. The remainder are single system ureteroceles. Bilateral involvement occurs in 15% of cases (69,70). In our study all ureteroceles were associated with upper moiety (100%). Bilateral involvement was 33%. According to literature Management options for ureteroceles include endoscopic puncture and decompression, a simplified upper-tract approach, namely, heminephrectomy, or complete repair including upper-pole surgery, ureterocoele excision, and lower-tract reconstruction in a single setting. The same has been followed in the management in this study.

In our study out of 24 cases 5 cases presented with VUR which is around 20.8%. Two cases (grade II and grade III) were managed conservatively based on urine C/S report, 3 cases were managed surgically common sheath reimplantation. Lee et al. followed 1/3 of their patients with VUR and DS nonoperatively, and concluded that resolution rates of low-grade (I-II/III) reflux were comparable to those seen in SS.^[6] Spontaneous resolution occurred in over half of patients with grades I–III/V VUR and support consideration for initial conservative management with prophylactic antibiotics.

In our study out of 24 cases 4 presented with ectopic ureter that is around 16.6%. According to literature Ectopic ureter associated with ureteroceles can cause upper-pole hydronephrosis and obstruction, which leads to ipsilateral lower-pole reflux in 50% of cases.^[7] Contralateral reflux is seen in 25%. After systematic evaluation by said investigations out of 4 cases of ectopic ureter, 3 females had ectopic ureteric insertion in to the posterior wall of vagina and in 1 male the ectopic insertion was in to the posterior urethra. Out of 24 cases 2 cases (8.3%) presented with PUJO of lower moiety both cases presented on left side.

In our study the commonest mode of presentation was non-functioning of upper moiety which is around 29% followed by VUR, as it was also associated with ectopic ureter and ureterocoele. Isolated VUR was 20.8% only but when combined with associated other anomalies like ectopic ureter and ureterocoele, the incidence is coming up to 45.8% in total cases. Hence in our study the most common symptom was UTI, most common presentation was VUR and the least common presentation was PUJO. Upper moiety was most commonly affected and lower moiety least commonly affected.

Out of 24 cases all presented with symptoms, 2 cases were managed conservatively and 22 cases managed surgically as and when indicated. Two cases which managed conservatively were presented with low grade UTI 2 and 3. Followed up for 3 years and they showed low grading of VUR and no further increase in grading or scarring on further investigations. Husmann et al studied management of vur in duplex system, stated that low grade vur can be managed conservatively.^[8]

In our study, 9 cases (37.5%) underwent ureteric reimplantations by Cohen's technique. In all common sheath ureteric reimplantation were performed. Out of 9 cases 4 (44.4%) were ureteroceles associated with VUR, 3 (33.3%) cases of isolated high grade VUR and 2 (22.2%) cases presented with ectopic ureteric insertion. Sander et al observed a higher need for second surgery in children undergoing endoscopic incision of ureteroceles associated with VUR in the duplex system.^[9] In Krishnamoorthy et al study, 4 children had mild reflux following incision, but were asymptomatic and followed up with conservative measures.^[5] In our study the procedure followed were, for ureterocoele with VUR - excision of ureterocoele and common sheath reimplantation was done. For isolated VUR – common sheath ureteric reimplantation was done. For ectopic ureter- both combined extravesical and intravesical approach was done. Types of reimplantation done were 1) Cohens cross triangular 2) Politano-Leadbetter.

Out of 22 cases, 9 cases (40.9%) were managed by upper moiety nephroureterectomy. In this overall open surgery were performed in 8 cases and 1 case underwent laparoscopic upper moiety nephroureterectomy. Choi and Oh, in their series of 63 children with complete duplex system concluded that uretero-ureterostomy had been the most successful nephron-sparing additional surgery, especially if the upper moiety is salvageable.^[10] The management of non-functioning moieties has been a relatively easy task. Laparoscopic heminephrectomy has been the standard treatment of poorly functioning moiety. It is a relatively safe mode of treatment, reproducible with relatively low morbidity and better treatment outcomes.^[11]

Gundet et al., in his study on assessment of functional outcome 101 patients following heminephrectomy, observed that there was a mild decrease in the function of the remaining moiety in more than 50% of the patients.^[12] Dalsan et al. reiterated the need for following up all remnant moieties with nuclear scintigraphy even though the extent of functional damage is subtle.^[13] Hisamatsu observed that recurrent infections after heminephrectomy were all treatment outcomes based on the initial surgical approach to ectopic ureterocoele rather than isolated problems associated with a distal ureteral stump.^[14]

They concluded that total ureterectomy is unnecessary at the time of heminephrectomy for a poorly functioning moiety because the distal ureteral stump rarely causes a problem. Recently, a less invasive method for upper pole ablation has been described by Romao et al.

in an initial series of 9 patients. Simple laparoscopic ureteral ligation via placement of either two nonabsorbable polymer locking or titanium clips and transection was successful and asymptomatic in all patients at a mean follow-up of 27 months. Two cases of PUJO were managed laparoscopically out of 22 cases. Two cases out of 22 cases were managed by Cystoscopic puncturing of ureteroceles in our study. Shankar et al. observed in his study that in those with duplex system ureteroceles, 27% of them were asymptomatic and were followed up for 8 years and none of them required surgery or developed urinary tract infections. In krishnamoorthy et al series also, ureteroceles that were asymptomatic were followed up conservatively. In our study 2 cases of ureteroceles after puncturing were followed up, there was recurrence of any symptoms on follow up period.^[5]

CONCLUSION

In Upper Moiety commonly associated with ureteroceles, ectopic ureter, non-functioning or dysplastic changes. Lower Moiety commonly with VUR, PUJO accordingly as per literature. Systematic investigations are needed to support further management. Anatomy and functioning of the duplex moiety are important for making decision regarding the management. Early detection of duplex moiety and early intervention has good outcome on comparison to late detection.

REFERENCES

1. Horst M, Smith GHH. Pelvi-ureteric junction obstruction in duplex kidneys. *Br J Urol*. 2008;101:1580-4.
2. S. M. Whitten and D. T. Wilcox, "Duplex systems," *Prenatal Diagnosis*, 2001;21, no. 11, pp. 952–957.
3. Siomou Ekaterini, Papadopoulou Frederica, Konstantinos D Kollios, Photopoulos Andreas, Evagelidou Eleni, Androulakakis Philippos, et al. Duplex collecting system diagnosed During the first 6 Years of life After a first urinary tract infection: a Study of 63 children. *J Urol* 2, February 2006;175(2):678e82.
4. Ma R, Wu RD, Liu W, Wang G, Wang T, Xu ZD, et al. A new classification of duplex kidney based on kidney morphology and management. *Chin Med J (Engl)* 2013;126:615-9.
5. Krishnamoorthy S, Kumar SB, Babu R. Duplication Renal Anomalies in Children: A Single Centre Experience. *Int J Sci Stud* 2016;3(10):12-17.
6. Lee, D. A. Diamond, P. G. Duffy, and P. G. Ransley, "Duplex reflux: a study of 105 children," *The Journal of Urology*, vol. 146, no. 2, part 2, pp. 657–659, 1991.
7. Ma R, Wu RD, Liu W, Wang G, Wang T, Xu ZD, et al. A new classification of duplex kidney based on kidney morphology and management. *Chin Med J (Engl)* 2013;126:615-9.
8. D. Husmann, B. Strand, D. Ewalt, M. Clement, S. Kramer, and T. Allen, "Management of ectopic ureterocele associated with renal duplication: a comparison of partial nephrectomy and endoscopic decompression," *The Journal of Urology*, 1999: vol. 162, no. 4, pp. 1406–1409.
9. Sander JC, Bilgutay AN, Stanasel I, Koh CJ, Janzen N, Gonzales ET, et al. Outcomes of endoscopic incision for the treatment of ureterocele in children at a single institution. *J Urol* 2015;193:662-6.
10. Lee JH, Choi HS, Kim JK, et al. Nonrefluxing neonatal hydronephrosis and the risk of urinary tract infection. *J Urol* 2008;179:1524–8.
11. Wallis MC, Houry AE, Lorenzo AJ, Pippi-Salle JL, Bägli DJ, Farhat WA. Outcome analysis of retroperitoneal laparoscopic heminephrectomy in children. *J Urol* 2006;175:2277-80.

12. Gundeti MS, Ransley PG, Duffy PG, Cuckow PM, Wilcox DT. Renal outcome following heminephrectomy for duplex kidney. *J Urol* 2005;173:1743-4.
13. You D, Bang JK, Shim M, Ryu DS, Kim KS. Analysis of the late outcome of laparoscopic heminephrectomy in children with duplex kidneys. *BJU Int* 2010;106:250-4.
14. Hisamatsu E, Takagi S, Nakagawa Y, Sugita Y. Nephrectomy and upper pole heminephrectomy for poorly functioning kidney: Is total ureterectomy necessary?. *Indian J Urol*. 2012;28(3):271-274. doi:10.4103/0970-1591.102699