

<sup>a</sup>Division of Pediatric Urology, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA

<sup>b</sup>Division of Pediatric General and Thoracic Surgery, Cincinnati Children's Hospital Medical Center, Cincinnati, OH,

Correspondence to: A.C. Strine, Division of Pediatric Urology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, MLC 5037, Cincinnati, OH, 45229-3039, USA, Tel.: +1 513 636 4975; fax: +1 513 636 6753

Andrew.Strine@cchmc.org (A.C. Strine)

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## Clinical and urodynamic outcomes in children with anorectal malformation subtype of recto-bladder neck fistula



A.C. Strine <sup>a</sup>, B.A. VanderBrink <sup>a</sup>, Z. Alam <sup>a</sup>, M. Schulte <sup>a</sup>, P.H. Noh <sup>a</sup>, W.R. DeFoor Jr <sup>a</sup>, E. Minevich <sup>a</sup>, C.A. Sheldon <sup>a</sup>, J.S. Frischer <sup>b</sup>, P.P. Reddy <sup>a</sup>

#### Summary

#### Introduction

Patients with anorectal malformations (ARMs) have a high incidence of genitourinary anomalies. Those with a recto-bladder neck fistula may represent a high-risk group, but their long-term urologic outcomes are poorly described.

#### Objective

To evaluate the clinical and urodynamic outcomes in a large cohort of patients with an ARM subtype of recto-bladder neck fistula.

#### Materials and methods

A retrospective cohort study was performed of patients who had been treated for a recto-bladder neck fistula at the present institution since 2007. The primary outcomes were the ability to achieve urinary continence after 4 years of age, and development of a mildly decreased glomerular filtration rate (GFR) or worse (<89 ml/min/1.73 m $^2$ ). Continence was defined as the ability to store urine for 3–4 h during the day and 8 h overnight without leakage.

#### Results

Demographic and clinical data are provided in the Summary Table. The most recent urodynamic findings included the presence of detrusor overactivity in 30 (75%) patients, median leak point pressure of 56.0 cmH $_2$ O (range, 14–140), median functional cystometric capacity at 40 cmH $_2$ O of 125.5% age-expected capacity (range, 36–473%), and median maximum cystometric capacity of 131.0% age-

expected capacity (range, 44–473%). A mildly decreased GFR or worse developed in 13 (24%) patients. Of the 52 (78%) patients who were followed by pediatric urology at the present institution with a median follow-up of 30.9 months (range, 0.0–86.8), 35 (67%) were at least 4 years of age and could be assessed for continence. Continence was achieved in five (14%) patients voiding spontaneously and 15 (43%) performing CIC. Recurrent urinary tract infections (UTI) (OR 0.70, P=0.006) were an independent predictor of incontinence, while urethral anomalies (OR 1.40, P=0.03) were an independent predictor of chronic kidney disease (CKD) on multiple logistic regression analysis.

#### Discussion

The findings favorably compared with other studies, but were more robust due to the size of the cohort and breadth of urologic evaluation. Limitations included the retrospective design at a single institution. Incomplete clinical data and misclassification of continence may have lead to bias.

#### Conclusions

This large cohort of patients with an ARM subtype of recto-bladder neck fistula had a high incidence of genitourinary anomalies. They were rarely able to achieve continence with spontaneous voiding alone and were at risk of developing CKD, both of which were likely multifocal in origin. Long-term urologic follow-up is warranted for patients with a recto-bladder neck fistula.

	Urologic follow-up at the present institution $(n = 52)$	No urologic follow-up at the present institution $(n = 15)$	P-value
Median age at posterior sagittal anorectoplasty in months (range)	6.9 (0.0-100.8)	7.1 (2.7–78.9)	0.98
Posterior sagittal anorectoplasty at the present institution, <i>n</i> (%)	37 (71)	11 (73)	1.00
Re-operative posterior sagittal anorectoplasty, n (%)	26 (50)	6 (40)	0.57
Laparoscopic assistance, n (%)	12 (23)	3 (20)	1.00
Tethered cord, <i>n</i> (%)	22 (42)	4 (27)	0.37
Tethered cord release, n (%)	15 (29)	4 (27)	1.00
Median sacral ratio (range)			
Anteroposterior	0.60 (0.00-1.20)	0.57 (0.00-1.06)	0.47
Lateral	0.61 (0.00-1.20)	0.62 (0.00-1.47)	0.98
Recurrent UTI, n (%)	27 (52)	5 (33)	0.25
Epididymo-orchitis, n (%)	5 (10)	0 (0)	0.58
Pre-operative hydronephrosis, n (%)	26 (55)	5 (56)	1.00
Pre-operative VUR, n (%)	27 (57)	6 (55)	1.00
Solitary kidney, n (%)	21 (40)	4 (27)	0.38
Upper-tract anomalies (e.g. ureteropelvic junction obstruction, ureterovesical junction obstruction, ectopic ureter, ureterocele, renal fusion anomaly), <i>n</i> (%)	14 (27)	2 (13)	0.49
Hypospadias, n (%)	19 (37)	4 (27)	0.55
Urethral anomalies (e.g. PUV, urethral atresia, congenital urethral stricture, megalourethra), n (%)	14 (27)	1 (7)	0.16
Undescended testis, n (%)	18 (35)	5 (33)	1.00
Most recent median GFR in ml/min/1.73 m <sup>2</sup> (range)	108.0 (0—187)	106.5 (62–147)	0.96

### Introduction

The association between anorectal malformations (ARMs) and genitourinary anomalies is well established. Most studies have reported a 30–50% incidence of genitourinary anomalies in patients with ARMs. Their identification and appropriate management are critical, as they represent a significant source of morbidity and mortality in these patients [1]. However, it is challenging to stratify the risk for long-term sequelae of certain genitourinary anomalies when identified early [2].

A growing body of evidence has suggested that the severity of ARMs and presence of a tethered cord and abnormal sacrum predict a worse prognosis for urinary continence and preservation of renal function [1]. As one of the most severe ARMs in male patients, the recto-bladder neck fistula is characterized by a termination of the rectum at or near the bladder neck [3]; it occurs in approximately 10% of male patients with ARMs [4]. Several studies have recently observed a high incidence of genitourinary anomalies, urinary incontinence, and chronic kidney disease (CKD) in patients with a recto-bladder neck fistula; however, the majority of these studies included a small number of patients with a recto-bladder neck fistula,

and often did not provide a strict definition for continence or describe a comprehensive urologic evaluation [5-17].

The present study sought to evaluate the clinical and urodynamic outcomes in a large cohort of patients with an ARM subtype of recto-bladder neck fistula. It was hypothesized that they would have a higher incidence of genitourinary anomalies, incontinence, and CKD when compared to other published series of patients with less severe ARMs.

#### Materials and methods

After approval from the Institutional Review Board, a retrospective cohort study was performed of patients who had been treated for a recto-bladder neck fistula at the present institution since 2007. All patients were identified by the subtype of ARM and through a query of the institutional database. Demographic and clinical data were abstracted from their medical records.

The primary outcomes were the ability to achieve urinary continence after 4 years of age and development of a mildly decreased glomerular filtration rate (GFR) or worse (<89 ml/min/1.73 m<sup>2</sup>). Continence was defined as the ability to store urine for 3–4 h during the day and 8 h overnight without

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