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ORIGINAL ARTICLE

Management of urinary incontinence in girls with congenital pouch colon

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Abstract

Purpose This study describes the management of urinary incontinence (UI) in eight girls with congenital pouch colon (CPC) associated with anorectal malformation (ARM).

Methods From 2013 to 2015, six girls with CPC and UI underwent bladder neck reconstruction (BNR). Four girls had complete UI (CUI) and two girls partial UI (PUI). From 2019 to 2023, four girls, including two with failed BNR, underwent bladder neck closure (BNC) and augmentation cystoplasty (AC) with a continent stoma. Subtypes of CPC were Complete CPC ($n=7$) and Incomplete CPC ($n=1$). All girls had a double vagina; short, wide urethra; and reduced bladder capacity with an open, incompetent bladder neck (BNI). During BNR, a neourethra was constructed from a 1.5–2 cm-wide and 1.5–3-cm-long trigonal strip. During BNC, AC was performed using a 20 cm ileal segment ($n=3$) and by a colonic pouch segment, preserved during earlier colorraphy ($n=1$). Continent stoma included a Monti's channel ($n=3$) and appendicovesicostomy ($n=1$).

Results BNR produced moderate improvement of UI ($n=2$), while UI was still very severe ($n=4$). During BNC, intraoperative complications included iatrogenic vaginal tears ($n=4$). Early complications included partial dehiscence of the ileocystoplasty ($n=1$), partial adhesive small bowel obstruction ($n=1$), and difficulty in stomal catheterization with prolonged drainage from the pelvic drain ($n=1$). Late complications included unilateral grade II vesicoureteric reflux (n = 2) and vesicovaginal fistula (VVF) ($n=2$) needing trans-vaginal closure in one girl. Urinary stones ($n=2$) with stomal leakage of urine in one girl needed open cystolithotomy twice ($n=1$), and endoscopic lithotripsy ($n=1$). At follow-up, all patients have high overall satisfaction with the procedure and their continence status.

Conclusions BNC with AC and a catheterizable stoma satisfactorily achieves continence in girls with CPC and UI, vastly improving quality of life. If lower urinary tract (LUT) anatomy is favorable, BNR with/without AC can be the initial surgical procedure. BNC should be the primary procedure in girls with unfavorable LUT anatomy and for failed BNR.

Level of evidence IV

Keywords Congenital pouch colon · Urinary incontinence · Bladder neck reconstruction · Bladder neck closure · Continent diversion · Appendicovesicostomy

Introduction

Congenital pouch colon (CPC) is an unusual abnormality in which a pouch-like dilatation of a shortened colon is associated with an anorectal malformation (ARM). CPC is classified into 'Incomplete CPC' where the length of normal colon is adequate for performing pull-through surgery without need for preserving the colonic pouch by tubularization (coloplasty), and 'Complete CPC' where there is either absent or insufficient normal colon for a pull-through procedure and coloplasty is necessary [1].

The anomalous clinical anatomy of CPC in girls, including that of the associated genitourinary tract (GUT) anomalies, has been detailed earlier [2, 3]. All girls with CPC had a hemiuterus and vagina on each side in the pelvis with the terminal fistula of the colonic pouch usually opening in the vestibule, just posterior to the urethral meatus in an intervaginal position [2, 3]. Less commonly, the fistula opened in the urethra [3]. There was a high incidence of urinary incontinence (UI) ranging from 75% [2] to 81% [3]. UI was either

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partial (PUI), the girls passing urine at regular intervals but also having diurnal and nocturnal dribbling, or else complete (CUI) [2]. UI was due to a varying severity of a structural abnormality of the lower urinary tract (LUT), the main anatomical features responsible being a short, wide urethra leading to a partially or widely open bladder neck, often deficient posteriorly in an inverted V-shaped fashion, and reduced urinary bladder (UB) capacity [2, 3]. Due to the frequent 'high' or more cranial confluence of the urethral and vaginal openings and the tilt of the vaginas, vaginal pooling of urine after micturition is not infrequent and may worsen UI [4].

Due to the unique anatomical features in girls with CPC, UI is difficult to manage. In an earlier review article, we had reported 6 girls with UI in whom a Young–Dees–Leadbetter bladder neck reconstruction (BNR) had been performed with moderate improvement in 2 girls and little or no improvement in 4 girls [4]. Subsequently, 4 girls, including 2 of the 4 girls with poor results after BNR, have undergone surgical closure of the bladder neck (BNC), bowel augmentation- cystoplasty (AC), and construction of a catheterizable channel for clean intermittent catheterization (CIC). The present study describes the clinical profile, evaluation, and management of 8 girls in whom surgical treatment of UI has been performed.

Materials and methods

From 2013 to 2015, 6 girls with CPC and UI, aged 8 years to 21 years (mean, 14.4 years), underwent Young–Dees–Leadbetter BNR. Four girls had CUI, while 2 girls had PUI with ability to hold urine for a few minutes to half an hour in the daytime. From 2019 to 2023, 4 girls, including 2 from the previous group with unsatisfactory results after BNR, underwent BNC and AC with construction of a Mitrofanoff/Monti catheterizable channel. The two girls undergoing primary BNC had CUI with a very widely open or incompetent bladder neck (BNI) on CUS. The records of the 8 patients treated surgically for UI were studied and relevant findings including type of CPC, age at BNR and/or BNC, findings on outpatient examination, examination under anesthesia (EUA) and at CUS, results of radiologic studies and the operative findings were noted (Table 1). Seven girls had Complete CPC while 1 girl had Incomplete CPC. The confluence of the urethral and vaginal openings (3 openings) and, if visible, the opening of the terminal fistula of the colonic pouch (4 openings), was classified as being 'low' or near-normal (≤ 1.5 cm from the base of the clitoris) or 'high' (> 1.5 cm from the base of the clitoris) [2]. The terminal fistula of the colonic pouch ended as an intervaginal vestibular fistula (VF) in four girls, a distal urethral fistula ($n=1$), proximal urethral fistula ($n=1$) and was not identified ($n=2$). On EUA and CUS, in one girl with PUI (Case 3) the bladder neck was partially open while in 7 girls, the bladder neck was widely open. All patients had normal renal biochemistry.

X Rays of the sacrum were normal in all 8 patients. Ultrasound (US) examination showed normal kidneys without ureteric dilatation ($n=7$) and mild unilateral pelvicalyceal dilatation ($n=1$; Case 6). The UB was empty in 2 girls with CUI (Cases 2 and 6). A micturating cystourethrogram (MCU) ($n=4$; Cases 1–3, 5) showed a small capacity UB in all cases with evidence of an open, incompetent bladder neck (BNI). In one patient (Case 2), the vaginas were opacified with right-sided grade I vesicoureteric reflux (VUR) (Fig. 1) while in another girl with PUI (Case 3), there was some evidence of bladder neck patency (partial BNI). An intravenous urogram (IVU) ($n=3$; Cases 2, 5, 7), showed normal kidneys, suboptimally distended UB and BNI with, in one girl, vaginal filling with contrast.

Bladder neck reconstruction (BNR) procedure ($n=6$)

Access was obtained by a lower abdominal midline incision till the symphysis pubis, extended supra-umbilically if necessary. After extra-peritoneal dissection of the UB and retropubic dissection of the bladder neck and urethra, the UB was opened vertically in the midline. BNR was performed constructing a neourethra from a 1.5–2 cm-wide and 1.5–3 cm-long trigonal strip over an 8–12 Fr Foley's catheter (Fig. 2).

The ureters were usually low and laterally placed and in one patient (Case 4), achievement of a sufficient length of trigonal strip for tubularization necessitated cross-trigonal bilateral ureteric reimplantation at a more cranial level. Three patients had an inverted V-shaped defect at the bladder neck posteriorly (Fig. 2), and in 2 girls (Cases 2, 6), the defect was long, needing midline closure before constructing the neourethra. The neourethra was reinforced by double breasting of lateral flaps of de-epithelialized trigone and the bladder drained by a suprapubic (SPC) and a per-urethral catheter. The per-urethral catheter was removed after 10–14 days and the SPC 3 weeks after surgery.

Bladder neck closure (BNC), augmentation cystoplasty (AC), and Monti/Mitrofanoff procedure

Abdominal access via a vertical midline abdominal incision enabled dissection of the UB and retropubic exposure of the bladder neck and almost the entire urethral length. The short urethra and the frequent cranial opening of the urethral meatus aided this exposure. The UB was opened anteriorly in the midline extending the incision to the distal most accessible urethra (Fig. 2). By means of slightly oblique cuts, close to the midline, the bladder neck and urethra were laid open. Dissection of the widely open posterior margin of the bladder neck and urethra from the closely apposed anterior

Table 1 Details of patients undergoing surgery for UI ($n=8$)

Case no	CPC subtype; age (y) at BNR and BNC	UI (CUI/PUI)	Outpatient and EUA findings	CUS findings	Operative findings and procedure	Outcome of BNR and/or BNC
1	Incomplete CPC BNR- 21 y	CUI	'Low' confluence of 4 perineal openings; wide urethral meatus; intervaginal VF	Wide urethral opening; urethra < 1 cm long; BNI; small capacity UB; lateral, low-placed ureteric orifices	Small capacity UB, ureteric orifices normal caliber, very laterally placed BNR with 1.5 cm long urethral tube (over 10-Fr catheter). Vagino-fistula septum divided each side	**Poor** result
2	Complete CPC BNR- 19 y	CUI	'High' confluence of 3 perineal openings; excoriations + ve	Virtual absence UB neck; moderate capacity UB	Severe BNI; moderate capacity UB; long, posterior V-shaped defect UB neck; low, lateral ureteric orifices BNR with 2 cm urethral tube (8 Fr) after closure V-shaped defect. Vaginal septum divided	**Poo**r result
3	Complete CPC, appendix absent BNR- 12 y	PUI	'Low' confluence of 3 openings: wide urethral meatus	Wide urethra, 2 cm long; partial BNI; UB capacity moderate; lateral, low-placed ureteric orifices	Moderate capacity UB; ureters low-down, laterally placed BNR with 2.5 cm urethral tube (10 Fr)	**Fair result**: Diurnal dry period, 2- 3 h. Nocturnal UI + ve
4	Complete CPC BNR- 11 y	PUI	'Low' confluence of 4 openings; wide, patulous urethral meatus; Intervaginal VF; excoriations + ve	Virtually no urethra; BNI; UB capacity moderate; ureteric orifices not clearly made out; trigone small	Moderate capacity UB; ureteric openings very low and lateral, almost on anterolateral wall UB BNR with 2.5 cm urethral tube (8 Fr) after bilateral trans-trigonal ureteric reimplantation more cranially	**Fair result:** Diurnal dry period, 2 h. Nocturnal continence 3- 4 h
5	Complete CPC; short, stubby appendix BNR- 8 years BNC- 14 years	Earlier no UI detected; Later CUI	'High' confluence of 4 openings; urethral opening wide; Intervaginal VF	Urethral meatus wide; BNI; UB capacity small; trigone poorly developed; ureteric orifices lateral	Urethra short, wide; UB neck patulous, V-shaped defect posteriorly; UB capacity small; trigone poorly developed; ureteric orifices laterally placed BNR with 2.8 cm urethral tube (10 Fr) BNC at 14 years: Ileocystoplasty, BNC, Monti procedure	BNR: **Poor** result BNC: Final result Satisfactory

Table 1 (continued)

Case no	CPC subtype; age (y) at BNR and BNC	UI (CUI/PUI)	Outpatient and EUA findings	CUS findings	Operative findings and procedure	Outcome of BNR and/or BNC
6	Complete CPC BNR- 13 years BNC- 22 years	CUI	'High' confluence of 4 openings; Intervaginal VF; excoriations + ve	UB neck completely open, very small UB; ureters opening low on anterolateral UB wall. Long posterior V- shaped cleft extending to 1.5 cm proximal to ureteric openings	BNR with 3 cm urethral tube (10 Fr). Posterior V-shaped cleft closed before BNR BNC: Ileocystoplasty, BNC, Mitrofanoff procedure. RA flap over suture-line	BNR: P**oor** result BNC: Final result Satisfactory
7	Complete CPC. Primary BNC- 13 years	Earlier no UI detected; later CUI	'Low' confluence of 4 openings. Distal urethral fistula. opening within wide urethral meatus; excoriations + ve	Widely open UB neck, especially posteriorly; ureters opening low-down, laterally	[Primary surgery: TC and pull-through, segment of pouch preserved with leash of vessels and stoma; vaginal septum divided.] BNC: Posterior V- shaped defect UB neck; moderate-sized UB; Pouch segment-cystoplasty, BNC; Monti's procedure. RA flap over suture-line	BNC: Final result satisfactory
8	Complete CPC, short, stubby appendix Primary BNC- 11 years	Earlier no UI detected; later CUI	'High' confluence of 3 openings	Urethral opening 'Very High'; urethra very short. UB neck widely open; proximal urethral fistula just distal to UB neck	BNC: Posterior V- shaped defect UB neck**;** moderate-sized UB; ureters opening low-down, laterally; Ileocystoplasty, BNC, Monti procedure. RA flap over suture-line	BNC: Final result satisfactory

CPC Congenital pouch colon, *UI* urinary incontinence, *CUI* complete UI, *PUI* partial UI, *EUA* examination under anesthesia, *CUS* cystourethroscopy, *BNR* bladder neck reconstruction, *BNC* bladder neck closure, *VF* vestibular fistula, *BNI* bladder neck incompetence, *UB* urinary bladder, *TC* tubularizing colorraphy, *RA* rectus abdominis

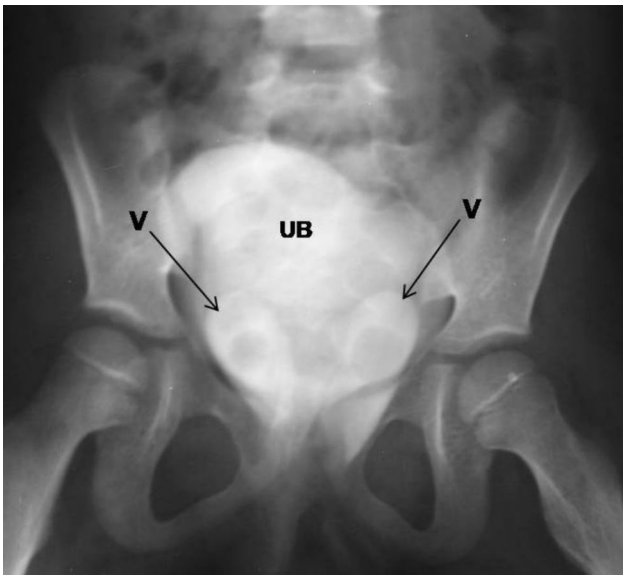

Fig. 1 Pre-BNR MCU film (Case 2) showing moderate capacity UB, open bladder neck, short, wide urethra, and filling of vaginas (arrows). UB, urinary bladder; V, vagina

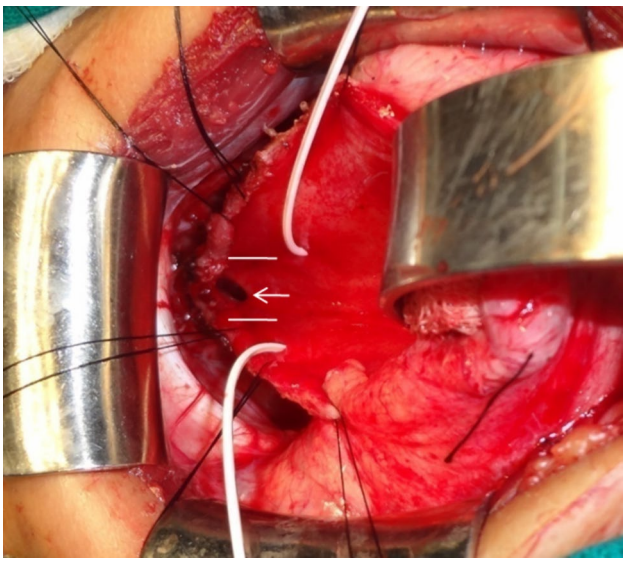

Fig. 2 Young–Dees BNR in 8-year-old girl (Case 5) showing open bladder neck with V-shaped posterior defect (arrow). Ureters have stents and lines mark incisions for constructing neo-urethral tube

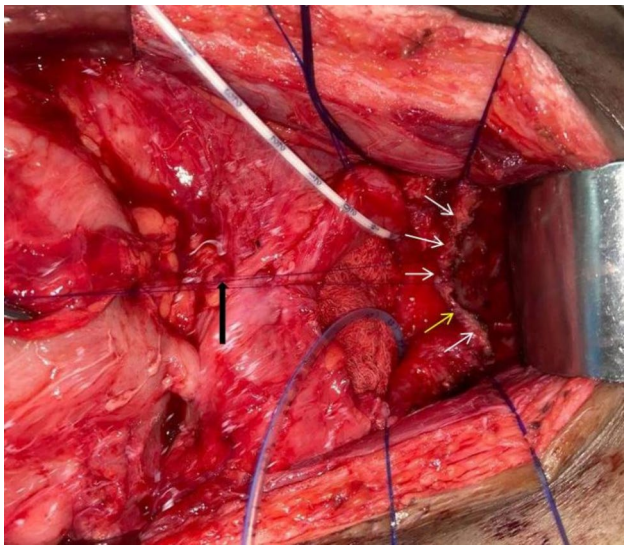

Fig. 3 Bladder neck and urethra laid open in 11-year-old girl (Case 8) at BNC before mobilization from vaginas. Arrow shows stay suture holding apex of bladder neck

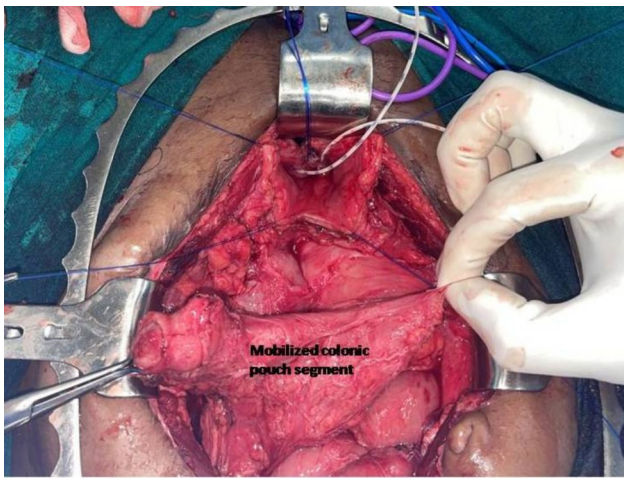

Fig. 4 [Case 7] Retention of a colonic pouch segment for AC showing partially closed anterior UB wall after BNC, split posterior UB wall, and retained pouch segment mobilized for colocystoplasty

vaginal walls was performed to create a 3–5 mm-wide margin (Fig. 3). Any iatrogenic rent in the anterior vaginal walls was closed in 2 layers with 4–0 Vicryl (Polyglactin, Ethicon Inc. USA).

The mobilized margins were rolled anteriorly and closed meticulously in the midline in 2 layers with 4–0 or 5–0 Vicryl. Dissection of bowel was often a lengthy, tedious procedure due to dense adhesions from previous abdominal surgeries. In 3 patients, a 25 cm segment of ileum 20 cm proximal to the ileocecal junction was isolated, the proximal 3 cm being used for a Monti's channel. The distal 20 cm was opened along the antemesenteric border. The incision in the anterior wall of the UB was carried down posteriorly in the midline to 2 cm proximal to the trigone (Fig. 4) and the posterior anastomosis of the ileocystoplasty performed. A Monti's tube was constructed over a 12–14-Fr catheter and tunneled into the right posterior wall of the native UB with the stoma in the right lower abdomen. In one patient (Case 6), the anatomy of the colon and appendix enabled an

appendicovesicostomy with an umbilical stoma. In another child (Case 7), a segment of the colonic pouch with its leash of vessels had been preserved during previous colorraphy and pull-through and brought out as a stoma. During BNC, this pouch segment was mobilized, laid open, and used for AC along with a Monti's procedure (Fig. 4).

In 3 patients (Cases 6–8), the bladder neck closure line was reinforced by an infra-umbilical, inferiorly based musculofascial flap of rectus abdominis (RA). A 20-Fr SPC was placed in the UB via an oblique tract along with a 10-Fr Foley's catheter through the catheterizable stoma. A pelvic drain was inserted. Externally draining 4-Fr ureteric stents were placed for 5 days to 1 week and the urinary reservoir irrigated with normal saline at 1 to 2 ml/ hour during this period. At 3 weeks postoperatively, the SPC was removed. A week later, the stomal catheter was removed and the child started on 3-hourly CIC through the continent stoma.

In all patients, the postoperative course and the occurrence and management of any postoperative complications was recorded. At follow-up, conducted monthly for 3 months and 3 monthly thereafter, urinary continence was assessed, any complaints noted, and further radiological assessment and management performed if necessary.

Results

There were no significant perioperative complications in the 6 girls undergoing BNR. At assessment on postoperative visits and at later follow-up, moderate improvement of UI in 2 girls (Cases 3, 4) was noted with a diurnal dry interval of 2–3 h and occasional stress incontinence. One girl (Case 4) had a nocturnal dry interval of 3–4 h, while the other girl had near-constant leakage of urine at night and needed diapers at night. There was no significant improvement after institution of CIC. These patients were offered the option of BA with/without a catheterizable abdominal stoma, but further surgical intervention was refused. In the remaining 4 girls, postoperatively UI was still severe and necessitated constant use of diapers. After counseling, only 2 of these 4 girls agreed to a further attempt to achieve continence by BNC, AC, and continent urinary diversion.

During BNC, an intraoperative complication in all 4 patients was iatrogenic tears in the vagina(s) during the difficult dissection of the rim of the bladder neck, distal fistula, and urethra from the vaginas. Table 2 shows the significant postoperative complications and their management in the 4 girls who underwent BNC. Within 2 years of BNC with AC, one patient (Case 5) required open cystolithotomy twice for a 1.5–2 cm stone in the urinary reservoir. In one instance, there was accompanying stomal leakage of urine which resolved after cystolithotomy. A recent dye study through the stoma shows the reservoir with an hour-glass appearance, grade II right-sided vesicoureteric reflux (VUR), and a closed bladder neck (Fig. 5).

At follow-up ranging from 1 to 5 years (mean, 2.5 years), apart from the girl with a minor VVF, 3 girls with BNC, CA, and a continent stoma are fully dry on diurnal 3-hourly and nocturnal 4–6 hourly CIC or OCD. The 4th patient (Case 8) with a minor VVF is satisfied with her overall continence status (Table 2) and is unwilling for an additional surgery. Overall satisfaction with the procedure is high in all patients. The patients have been advised irrigation of the reservoir with normal saline three-to-four times daily and whenever feasible, overnight catheter drainage (OCD).

Discussion

Apart from the studies described earlier [2, 3], there are only a few reports of UI in girls with CPC. Gharpure reported a girl with Complete CPC, double uterus and vagina, a

Table 2 Postoperative complications and their management in girls undergoing BNC procedure ($n=4$)

Complication; number	Management and outcome
Early complications	
Partial anastomotic dehiscence of ileocystoplasty, POD 7 [$n=1$; Case 6]	Reoperation; reinforcement of suture-line; healed
Transient adhesive bowel obstruction [$n=1$; Case 8]	Conservative management; resolved
Prolonged drainage from pelvic drain; difficulty in CIC [$n=1$; Case 7]	Additional 2 weeks indwelling stomal catheter drainage; resolved
Late complications	
Vesico-vaginal fistula (VVF) [$n=2$; Cases 5, 8]	6 mm VVF closed trans-vaginally in multiple layers; healed (Case 5) Diurnal dryness on 3–4 hourly CIC; minimal nocturnal wetting on OCD (Case 8; minor VVF)
Calculus/ calculi in reservoir [$n=2$; Case 5 (twice, once with stomal leak), Case 6]	Open cystolithotomy (2 times) [Case 5] Endoscopic (via Mitrofanoff channel) lithotripsy [Case 6]
Unilateral grade II VUR [n=2; Cases 5, 7]	Follow-up, monthly culture studies

POD postoperative day, *CIC* clean intermittent catheterization, *OCD* overnight catheter drainage, *VUR* vesicoureteric reflux

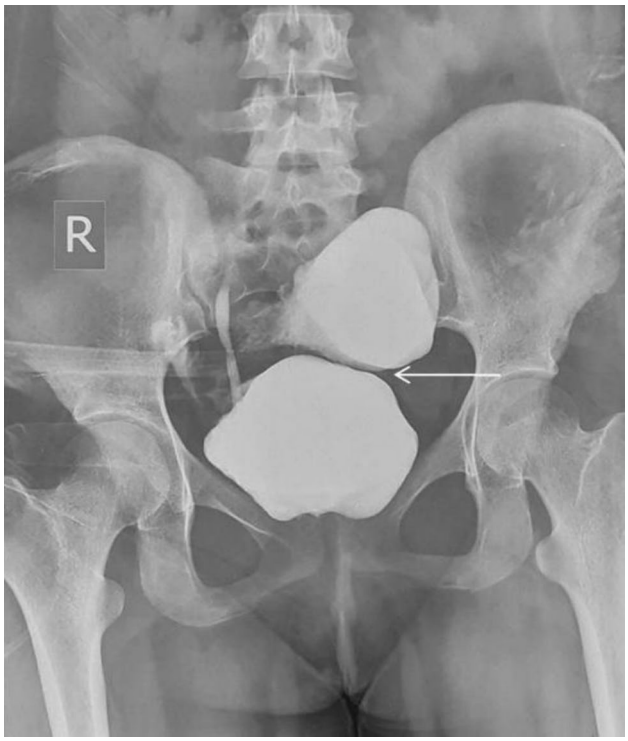

Fig. 5 Radio-contrast dye study via Monti's channel after BNC, AC, and Monti's procedure (Case 5), showing closed bladder neck, 'waisting' at site of ileocystoplasty (arrow), and grade I right-sided VUR

colovestibular fistula, and bladder hypoplasia with UI [5]. UI was managed by ureteric reimplantation into an ileal conduit [5]. Another report described 2 girls with Complete CPC; a 2.5 year-old girl with a recessed urinary meatus and UI, and a 14-year-old girl with CUI and a small, hypo-compliant UB [6]. Similar to our findings, in both cases, the sacrum was normal [6].

An important consideration in these patients is that there is a fairly wide spectrum of anomalous anatomy of the LUT, i.e., the urethra, bladder neck, UB capacity as well as of urinary continence [2, 3]. Accurate volumetric assessment of UB capacity is difficult because of the open bladder neck. Radiologic studies and CUS are important to study LUT anatomy and subjectively assess UB capacity. Detailed history-taking is essential as UI may be underestimated in infants and young girls. The parents of some infants stated that the girl passed urine normally in a stream, but further questioning revealed that on crying and/or while standing, the child leaked urine [3]. An earlier study reported that only 1/10 girls with CPC < 1-year age were assessed to have UI, while 9/ 12 (75%) older girls had UI [2].

Both PUI and CUI are major disabilities and need active intervention as the girls are almost always wet, need diapers, and have significant stress and psychological trauma. There are several anatomic reasons why BNR procedures may not be uniformly successful in treating UI in girls with CPC. The open bladder neck often has a posterior V- shaped defect that may need midline closure before constructing the neourethra. The UB is also thin-walled and the neo-urethral tube may not provide adequate muscular resistance. The trigone is small with low-placed ureteric openings [3] and constructing an adequately long urethral tube can be difficult. It may be necessary to reimplant the ureters at a more cranial level which is hazardous as the ureters are usually of normal caliber. In addition, UB capacity is subnormal for age. In 2 patients with a 'fair' result after BNR, AC was considered to increase UB capacity and the dry interval but was refused by the patients [4]. Additionally, a high and inaccessible urethral opening, as in several girls, may make CIC by this route difficult. However, as there is a spectrum of severity of UI, if BNI is not severe and UB capacity is not markedly reduced, BNR may be beneficial [4], especially if combined with AC and a catheterizable stoma.

It has been suggested earlier that if BNR procedures were unsatisfactory in managing UI in these girls, an effective option could be BNC and AC with a continent stoma [3]. The segment of bowel available for AC in the more common Complete CPC is limited by the normal proximal colon being either absent or very short, virtually ruling out colocystoplasty. Thus, the stomach or small bowel may be required for augmentation [3], although, especially in Complete CPC, using a substantial length of ileum for AC aggravates the risk of short-bowel syndrome. One useful feature in Complete CPC is that, as described earlier in 4 patients, during tubularization of the colonic pouch, a segment of the pouch can be preserved with its leash of vessels [3] for possible use for AC as in one of the patients reported here. Duci et al. also described five cases treated for CPC in two Italian centers and emphasized on the double-vascular arcade which allows pouch splitting and lengthening [7]. This enabled increasing UB capacity in one case and vaginal reconstruction in 2 patients [7]. Wester et al. reported two girls with CPC in whom vaginal reconstruction was performed by longitudinal splitting of the colonic pouch, using the redundant patch of pouch colon for the purpose [8]. A case of CPC where the colonic pouch was used to augment a high-pressure neurogenic bladder has also been reported [9]. It is important, however, that the colonic pouch has abnormal histology [10], and with the high incidence of development of calculi after AC, there is a long-term risk of malignant transformation of the pouch tissue. This should be kept in mind during the decision-making process and in cases where a pouch segment has been used for AC, endoscopic examination of the reservoir at regular intervals with biopsy of any suspicious area or lesion should be mandatory.

There are several reports describing BNC with/without AC as a successful means of achieving urinary continence in patients with a variety of conditions in whom other bladder

outlet surgery have failed [11–17]. In a report of 28 consecutive patients who underwent BNC with enterocystoplasty and Mitrofanoff diversion for structural and neurogenic conditions, 19 (68%) had undergone 20 unsuccessful bladder neck procedures before BNC [11]. BNC was initially successful in 27 (96.4%) patients [11]. One patient required subsequent closure of a postoperative VVF. In 11 patients, 16 additional procedures included stomal injection of bulking agents ($n=2$), stomal revision for stenosis ($n=2$) or prolapse ($n=1$), percutaneous lithotripsy ($n=1$), open cystolithotomy ($n=2$), lithotripsy for upper tract stones ($n=4$), repair of augment rupture ($n=3$), and retrograde ureteral stenting for stone ($n=1$) [11]. There were no observed cases of progressive or de novo hydronephrosis. Another report described 12 children in whom BNC with construction of a catheterizable stoma was performed with/without AC for bladder exstrophy, spinal dysraphism, and other conditions [12]. Although 6 patients needed reoperation, ultimate success rate was 100%. Nguyen and Baskin reported 12 children in whom BNC was performed for a variety of indications and found it to be effective for achieving urinary continence but with a high incidence of early and late complications as well as need for reoperation [13]. Complications included a urethral fistula, stomal leakage, stomal stenosis, and bladder stones [13]. Similar results have been reported in the other studies of children who have undergone BNC for refractory UI due to neurogenic bladder and/or other structural conditions [14, 15] or due to the vesical exstrophy complex [16]. Early and late complications after BNC appear to be related in part to compliance with CIC [13] and preoperative counseling to ensure a regular postoperative CIC and reservoir irrigation regimen is necessary [18]. In the long term, the most frequent complications are those related with catheterizable stoma and stones [16].

Our study also shows that BNC with AC and continent diversion is a satisfactory option to treat refractory UI in girls with CPC. However, one technical problem in performing BNC in these patients is the difficulty in closing the widely open UB neck with its often deficient posterior rim. In three of the four cases reported here, the incision in the anterior bladder wall was carried down just lateral to the midline on each side, laying open the bladder neck and short urethra completely. This extremely helpful maneuver is possible without pubic symphysiotomy as the more cranial urethral opening, the short urethra, and the widely open bladder neck greatly aid exposure and dissection. Dissection of the UB neck and urethra from the closely apposed distal remnant of the colonic pouch and the vaginas is also difficult. Iatrogenic tears in the vagina(s) are common and need repair. As an interposition layer after BNC, omentum and an RA muscle flap have been described [18, 19]. Omentum may not be easily accessible or available in girls with CPC because of the shortened colon and the previous surgeries. An inferiorly based RA musculofascial flap [19] is easily mobilized and is effective as interposition between the UB and vaginal suture lines. Another limitation in Complete CPC is that the appendix may not be available as a catheterizable channel as it is often absent/short and stubby or else duplicated [4]. A Monti procedure is then necessary, further increasing the technical difficulties.

An important technical point is that the native bladder should be opened widely posteriorly to prevent a narrow-mouthed anastomosis which can result in the augmentation segment behaving as a diverticulum predisposing to stone formation [18], a complication that occurred in one of our patients. Our follow-up period is relatively short, but 3 of the 4 patients required reoperation with one patient requiring 3 additional surgeries. Difficulty in stomal catheterization resolved over time and Hoebeke et al. also reported that only 2/8 stomal complications needed revision under anesthesia [14]. However, long-term urologic follow-up is essential as late occurrence of stomal complications and stone formation is not infrequent [11, 13]. The additional risk of malignancy in patients in whom a colonic pouch segment has been used for AC also needs monitoring.

In conclusion, although BNC and AC with a continent stoma is technically demanding with several early and late complications, it is overall a satisfactory option to achieve continence in girls with CPC and UI, vastly improving their quality of life. In girls with favorable LUT anatomy, BNR with/without AC may be considered as the initial procedure, especially if the urethral opening provides easy access for CIC. BNC should be the primary procedure for managing UI when in girls with unfavorable LUT anatomy and the procedure of choice after failed BNR.

Author contribution Study conception and design: PSY, KK, RC, VK, NP, and SRC (all authors). Data acquisition: KK, VK, NP, and RC. Analysis and data interpretation: PSY, NP, RC, and SRC. Drafting of the manuscript: KK (First draft), VK, RC, PSY, and SRC. Critical revision: RC, PSY, and SRC. All authors approved the final version of the manuscript.

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Data availability All data are available in the text and other material submitted for consideration for publication.

Declarations

Conflict of interests Nil. The authors have no competing interests to declare that are relevant to the content of this article.

Ethical approval This research study was conducted retrospectively from data obtained for clinical purposes and from management of the cases. We consulted extensively with the Institutional Ethics Committee of the institution who determined that our study did not need ethical approval provided that patient confidentiality was maintained.No other acknowledgements.

Consent to participate and publish Informed consent was obtained from all individual participants and/ or legal guardians included in the study.

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