A Case Series of Congenital Pouch Colon- A Rare Regional Variant of Covered Bladder Exstrophy

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Abstract

Introduction: The clinical, radiologic and cystourethroscopic (CUS) findings regarding the colonic pouch, its terminal colovesical fistula (CVF) and lower urinary tract (LUT) in 27 boys with congenital pouch colon (CPC) and associated anorectal malformation (ARM) are described and discussed.

Materials and Methods: Records of 27 boys with CPC in whom radiologic studies and/ or CUS yielded information about the colonic pouch, CVF, and the LUT were studied. Relevant clinical findings were recorded.

Results: CPC subtypes were Type I (n=1); Type II (n=13); Type III (n=1), and Type IV (n=12). Five patients (18.5%) had hypospadias and 3 (11.1%) unilateral undescended testis. X-Rays spine (n=25) showed normal sacrum (n=21) and partial sacral agenesis (n=4). Ultrasound (n=27) showed normal kidneys (n=21), unilateral malrotated kidney (n=1), unilateral hydronephrosis (n=2), and hydroureteronephrosis (n=3). Micturating cystourethrogram (n=12) showed a large, lobulated, smooth-walled urinary bladder (n=7) and vesicoureteral reflux (n=6). Contrast study of the colonic pouch (n=16) showed the pouch ending in a long wide CVF opening at the bladder neck with, in 11 patients (68.75%), filling of a large, lobulated urinary bladder. CUS (n=22) showed a competent bladder neck and an abnormal verumontanum pulled-up into the bladder neck. In 16 patients, the CVF opened in the trigone, just cranial to the right (n=13) or left (n=3) of the verumontanum.

Conclusion: CPC likely results from an early, 'localized' error in cloacal septation by the uro-rectal septum with faulty development of adjacent developing structures. The long-term clinical consequences on bladder dynamics and male fertility need attention.

Keywords

- Congenital pouch colon
- Anorectal malformation
- Colovesical fistula
- Verumontanum
- Embryology

Introduction

Congenital pouch colon (CPC) is an unusual abnormality in which a pouch-like dilatation of a varying length of shortened colon is associated with an anorectal malformation (ARM).[1] This condition has most frequently been reported from centers in northern India. CPC is classified into 4 subtypes (Types I- IV) based on the length of normal colon proximal to the colonic pouch.[2] In the more severe subtypes, either the normal colon is absent with the ileum opening directly into the colonic pouch (Type I) or the ileum opens into a short segment of cecum which then opens into the pouch (Type II). The less severe subtypes have a longer length of normal proximal colon (Types III and IV CPC).[2]

In males with CPC, the colonic pouch is generally believed to end distally in a relatively wide colovesical fistula (CVF) opening close to the bladder neck, although some studies have reported that the CVF enters the urinary bladder (UB) midway between its fundus and base;[3-4] its posterior wall;[2-5] or at any of three different sites on its posterior wall.[6] However, these assertions are not based on conclusive radiologic or endoscopic evidence[7], and only a few reports have documented the exact level of opening of the CVF by a contrast study or by cystourethroscopy (CUS) .[7-10] In a case of Type II CPC, a lateral film of a colostogram showed a long, relatively narrow fistula ending close to the bladder neck.[8] Similar findings on a distal loopogram were reported in a boy with Type II CPC.[9] In a case of Type I CPC, cystography and CUS showed a fistula between the bladder neck and the colonic pouch.[10]

In a review article on CPC from our center,[7] the clinical findings and results of investigations in 19 boys with CPC were briefly described. The present report presents a more detailed study of the radiologic and CUS findings of the colonic pouch, CVF and lower urinary tract (LUT) along with relevant clinical findings in 27 boys with CPC. The case material includes the 19 patients who formed part of the earlier review.[7] The discussion centers on the implications of these findings both in speculating on the possible embryo-pathogenesis of CPC and ARM as well as in optimizing management of these patients.

Materials and Methods

For this observational research study, after obtaining informed signed consent from the patients and/ or guardians, the medical records of males with CPC, managed from January 2007 to October 2022, were studied retrospectively. After consultation, the Institutional Ethics Committee of the institution determined that the study did not need formal ethical approval provided patient confidentiality was maintained. Patients in whom radiologic contrast studies and/ or CUS examination yielded information about the terminal CVF of the colonic pouch and the LUT were included in the study. Those with incomplete medical records were excluded. The 27 boys eligible for inclusion in the study were categorized into the subtypes of CPC (Types I – IV).[2] Relevant clinical findings, including those related to the lower genitourinary tract (GUT), were recorded.

X-rays of the lumbosacral spine (n= 25) were evaluated for gross appearance of the sacrum and the antero-posterior sacral ratio (APSR) was estimated.[11] In patients with sacral abnormalities or a low APSR, magnetic resonance imaging (MRI) of the spine was performed. Abdominal ultrasound (US) was available for all patients (n=27).

Due to logistics, institutional lacunae, and failure of all patients to come for regular follow-up, the radiologic contrast studies performed varied in the study group patients. A micturating cystourethrogram (MCU) (n= 12) was performed in patients with abnormal urologic findings on abdominal US or the finding of dilated ureter(s) at the time of primary/ definitive surgery. In 16 patients, a contrast study of the colonic pouch had been performed by injecting contrast material via a window colostomy or through the distal limb of an ileostomy or colostomy constructed proximal to the colonic pouch.

CUS examination of the LUT was performed in 22 patients. In 12 patients, CUS was performed just before disconnection (division-ligation) of the CVF during the definitive pull-through procedure (abdomino-PSARP), while in 10 patients, it was performed later to evaluate for VUR detected on MCU, or during the closure of the proximal diverting stoma. The findings regarding the urethra and bladder neck, position of the verumontanum, the opening of the CVF (if visualized), the trigonal area, ureteric orifices, and the UB were recorded.

Result

The significant abnormalities detected in the study group (n=27) are summarized in Table 1. The subtypes of CPC were Type I CPC (n=1; 3.7%); Type II CPC (n=13; 48.1%); Type III CPC (n=1; 3.7%), and Type IV CPC (n=12; 44.4%). Clinical findings related to the lower GUT included distal hypospadias without chordee (n=5; 18.5%), and unilateral undescended testis (n=3; 11.1%). Hypospadias was associated with penoscrotal webbing (n=1), partial penoscrotal transposition (n=2), and a bifid scrotum (n=2). All patients were passing urine normally. No patient had a significant abnormality of any other major organ system.

The 15 patients with Types I- III CPC (n= 15) had undergone a variety of preliminary and definitive surgical procedures while in all 12 patients with the less severe Type IV CPC, preliminary surgery consisted of a divided sigmoid colostomy proximal to the colonic pouch.

Radiologic investigations

X-Rays of the spine (n=25) showed a grossly normal sacrum (n=21; 84%) and partial sacral agenesis (PSA) (n=4; 16%). In patients in the first group, the APSR [11] ranged from 0.565- 1.1 (mean 0.775). The 4 patients with apparent PSA had APSR varying from 0.425- 0.50 (mean 0.475) and the MRI spine did not show any intraspinal abnormality. Abdominal US (n=27) showed normal kidneys (n=21; 77.8%), unilateral malrotated kidney (n=1), unilateral hydronephrosis suggestive of pelviureteric junction obstruction (n=2), and hydroureteronephrosis (n=3; bilateral- 2, unilateral- 1).

A MCU (n=12) had been performed in 10/15 patients with Types I- III CPC and 2/12 patients with Type IV CPC (n=12). In 7 patients (58.33%), the UB, although smooth-walled, appeared large and lobulated, often with a bilobed appearance **(Figures 1 & 2).**

Table 1: Summary of radiologic and cystourethroscopic (CUS) findings of the study (n= 27).

Study & Finding	N (%)
Radiologic findings	
MCU (n=12) 1. Large, smooth-walled, lobulated bladder	7 (58.33)
2. VUR (unilateral/ bilateral)	6 (50)
3. Posterior angulation of bladder neck at the junction with the proximal urethra	7 (58.33)
4. Retrograde filling of the colonic pouch	2 (16.67)
Contrast study of the colonic pouch (n=16) 1. Filling of the large, smooth-walled lobulated urinary bladder through long CVF at bladder neck	11 (68.75)
2. Bilobed appearance of urinary bladder on each side of long, wide CVF (Type IV CPC; n=10)	10 (100)
Cystourethroscopy findings (n=22)	
1. Verumontanum pulled up into bladder neck	22 (100)
2. Small, triangular verumontanum	6 (27.3)
3. CVF just proximal to verumontanum	16 72.7)
4. CVF not identified (in 6/10 patients in whom division- ligation of CVF done earlier)	6 (27.3)

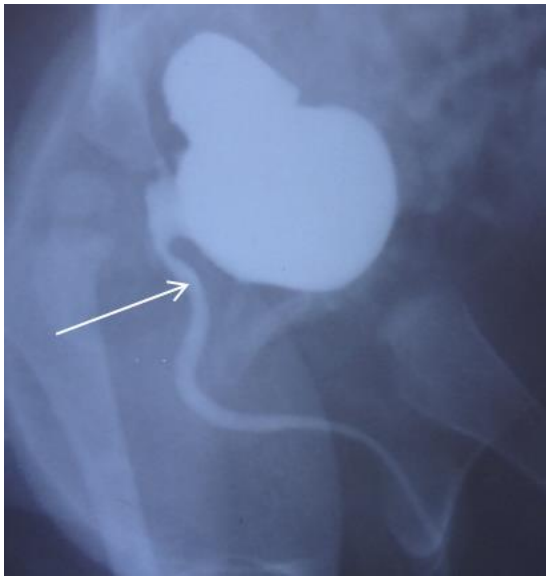

Figure 1: MCU of boy with Type III CPC showing a lobulated urinary bladder with posterior angulation of the bladder neck and the proximal urethra angled forward from the bladder neck (arrow).

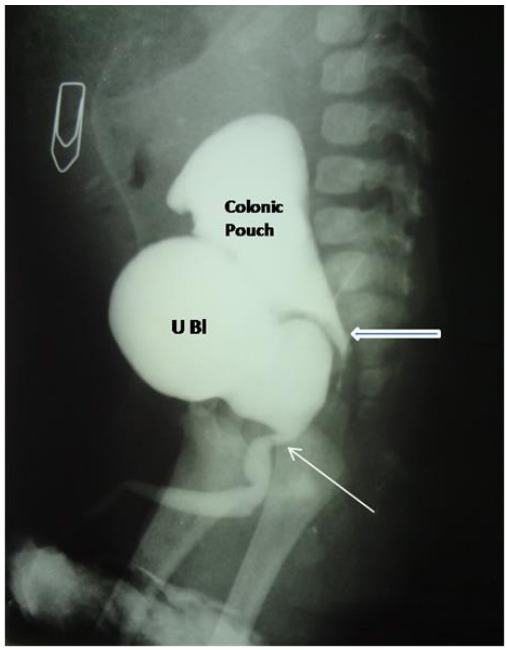

Figure 2: MCU of patient with Type II CPC showing large, lobulated urinary bladder (U Bl), and retrograde filling of the colonic pouch via the long CVF (arrowhead). Arrow points to anterior angulation of posterior urethra.

Another finding in 7 patients (58.33%) on lateral films was that the bladder neck at its junction with the proximal urethra was tented or angulated posteriorly and the proximal urethra angled forward from this point **(Figures 1 & 2).** Vesicoureteral reflux (VUR) (n= 6) was bilateral grade II, III, and IV in 1 patient each, unilateral grade II/ III (n=2), and unilateral grade IV VUR (n=1). In 2 patients (16.7%), during the micturating phase of the MCU, there was a retrograde filling of the colonic pouch through the patent CVF **(Figure 2).**

A contrast study of the colonic pouch (n=16) had been performed via a window colostomy (n=3; all Type II CPC) or through the distal limb of an ileostomy (n=1; Type I CPC) or colostomy (n=12; Type II [n= 2]; Type IV [n= 10]) constructed proximal to the colonic pouch. The transition from the dilated colonic pouch to the CVF was abrupt in all cases **(Figure 3).**

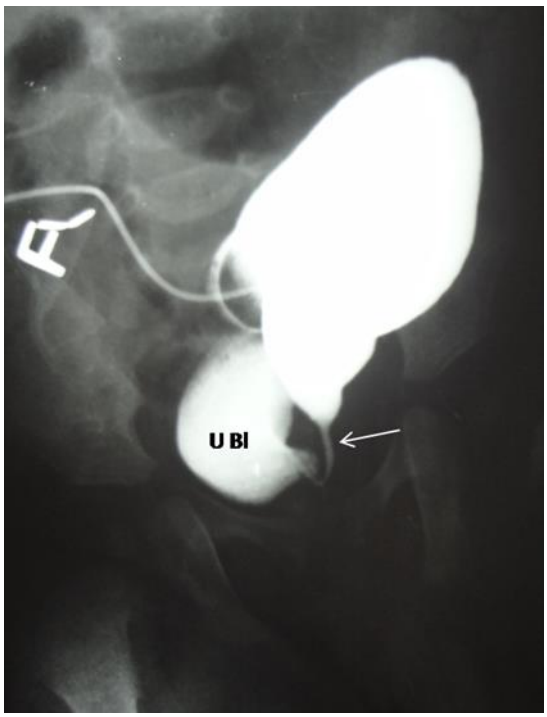

Figure 3: Distal colostogram of a child with Type III CPC showing the dilated colonic pouch with abrupt transition to a long, narrow CVF (arrow) opening at the bladder neck.

In addition to opacifying the colonic pouch, in 11 patients (68.75%), there was filling up of a large, smooth-walled, lobulated UB through the long CVF, opening at the bladder neck **(Figures 3-6).**

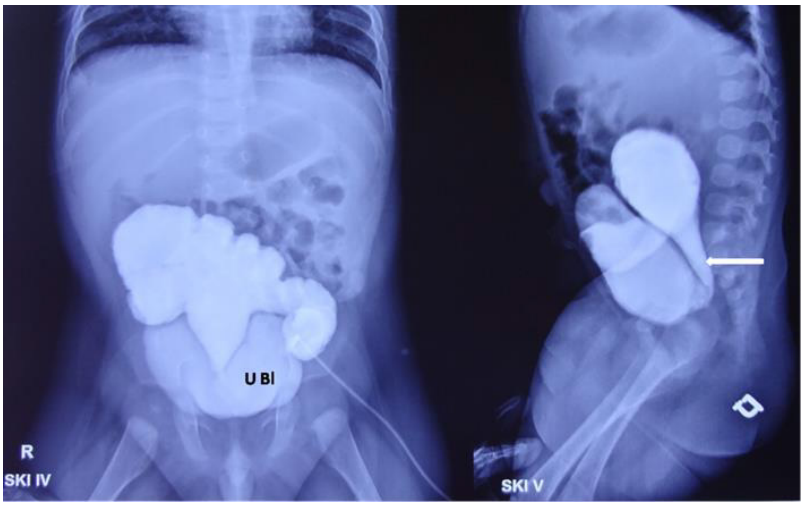

Figure 4: Distal ileostogram in a boy with Type I CPC showing the bilobed urinary bladder (U Bl) on either side of the long, relatively wide CVF (white arrow).

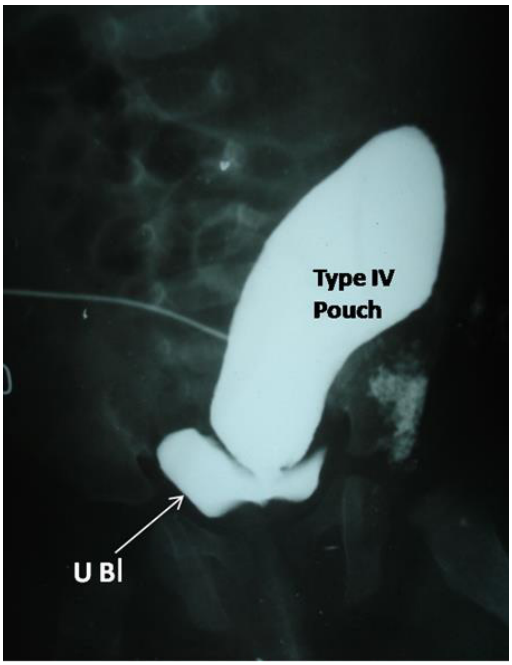

Figure 5: Distal colostogram in a boy with Type IV CPC showing the bilobed bladder (U Bl) on either side of the CVF.

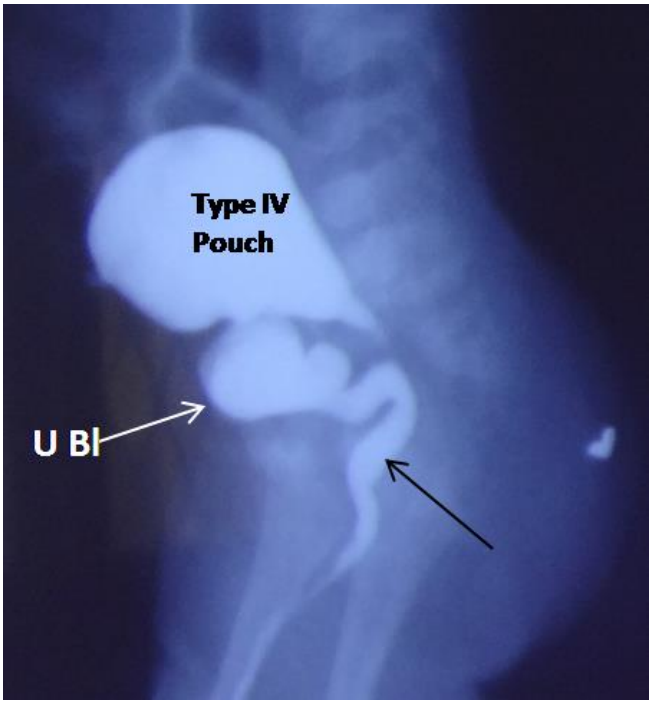

Figure 6: Distal colostogram in a boy with Type IV CPC showing filling of the lobulated urinary bladder (U Bl) through the long CVF. Note the similarity with the features seen in the MCU film in **Figure 2.**

In 10 patients with Type IV CPC, the UB had a bilobed appearance with each lobe on either side of the long CVF **(Figures 5 & 6).** In 2 patients with Type IV CPC, the distal colostogram opacified the UB and also the urethra **(Figure 6).**

Cystourethroscopic (CUS) findings (n=22)

CUS examination of the LUT was performed in all 15 patients with Types I-III CPC and 7/12 patients (58.33%) with Type IV CPC. The age at CUS examination ranged from 9 months to 5 years (mean 3.73 years). In all 22 patients, the bladder neck was competent. Difficulty in negotiating a cystourethroscope through the proximal urethra was noted in 3 boys with Type IV CPC, perhaps because of angulation of the urethra at its junction with the bladder neck.

In all 22 patients, the verumontanum was not at its normal position in the posterior urethra but was pulled- up into the bladder neck in the trigonal area **(Figures 7 - 9).**

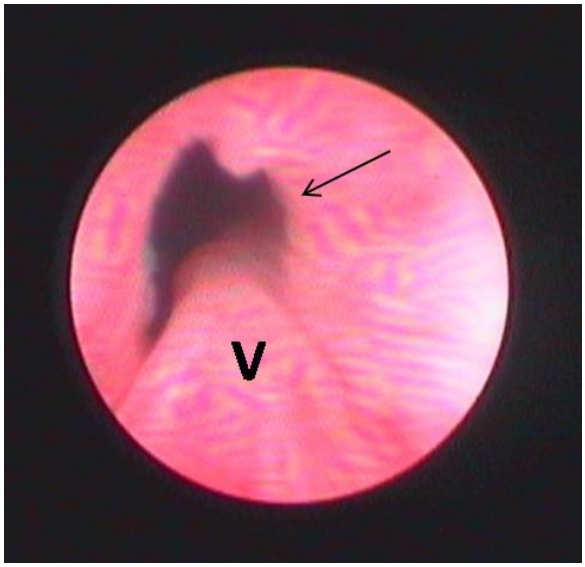

Figure 7: CUS in a boy with Type II CPC showing the verumontanum (v) entering the bladder neck (arrow)

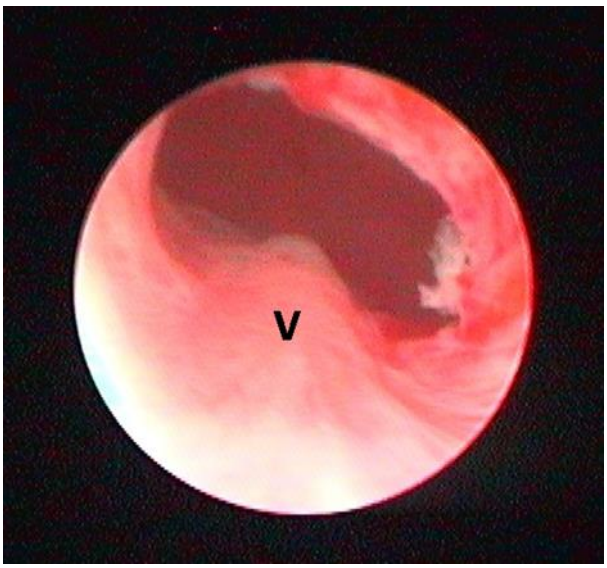

Figure 8: CUS in a boy with Type II CPC showing a relatively small verumontanum (V), CVF not seen (?As ligated earlier).

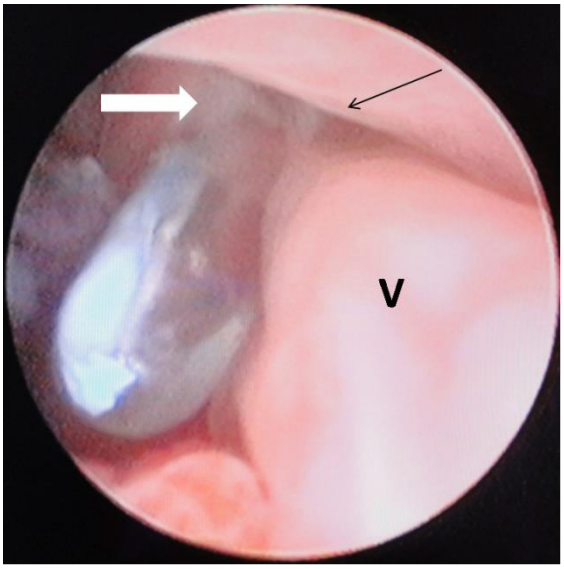

Figure 9: CUS of Type II CPC showing verumontanum (V) entering the bladder neck (black arrow). The white arrow shows the catheter emerging from the CVF above and to the right of the verumontanum.

The verumontanum was more triangular in shape than its usual hour-glass appearance, having the appearance of an inverted–V, and in 6 patients (27.3%), it was very small and triangular **(Figure 8).**

In the 12 (54.55%) patients in whom CUS was performed during definitive surgery before division-ligation of the CVF, the visualized fistula was usually wide enough to allow passage of a 5 or 6 Fr infant feeding tube or fine metal dilator. The fistula opened low down in the trigone, just proximal to the right of the cranial limit of the verumontanum in 10 patients **(Figure 9)**, or just to the left of the cranial limit of the verumontanum (n=2). The opening of the CVF could not be identified on CUS examination in 6/10 patients in whom division-ligation of the CVF had been performed earlier, while in 4 patients the opening was seen just above the verumontanum in the trigonal area. In 2 of these patients, an endoscope introduced through the fistula, led to a blind diverticulum with colonic mucosa. The ureteric openings were not clearly visualized in 12 patients. In the remaining 10 patients, the openings were seen to be somewhat more cranial and lateral in position than normal. In 2 patients,

unilateral dilatation of the ureteric orifice was present with a golf-hole appearance.

Discussion

In the usual male ARM, the highest termination of the rectum is also with a rectovesical fistula opening just above the bladder neck.[12] The exact relationship of the fistulous opening with the verumontanum can be determined only by CUS, as in our study. Such reports are rare; although a study of ARM with rectobladder neck fistula reported that of 40 patients who underwent cystoscopy, 16 (40%) had a higher than normal location of the verumontanum.[12] Koga et al[13] performed urethroscopy at the time of laparoscopic-assisted anorectal pull-through, and showed that a fine catheter, passed through the fistula, entered the urethra just cranial to the verumontanum in one of 3 cases considered to have a recto-prostatic urethral fistula on a distal loopogram. Significantly, with regard to our findings, Smith and Stephens[14] studied specimens of embryos and fetuses with ARM and reported that whenever the fistula issued into the urethra cranial to the normal site of the verumontanum, the vasa and prostatic tissue ascended with it. In rectovesical fistulae, these structures were lying within the bladder along with the verumontanum.[14] This ascent of the verumontanum and associated structures may explain the abnormal shape of the verumontanum noted in our study.

The traditional theories of cloacal septation by fusion of lateral folds or by the descent of a coronally oriented wedge of mesenchyme representing the urorectal septum (URS) were first challenged by van der Putte's studies on pig embryos[15] and Kluth's studies of the morphology of the caudal region in rats and 'short tail' mice embryos.[16] The authors stated that in ARM, the basic error is defective development of the dorsal part of the cloacal membrane (CM) and it is always short in ARM.[15-16] If the dorsal cloacal defect is large, the hindgut enters the UG part in a high position resulting in a high ARM while a minimal defect results in a relatively low ARM.[16] Nievelstein et al[17] in a study on human embryos, concluded that during cloacal differentiation, the apparent decrease in distance between the tip of the URS and the CM is most likely due to the unfolding process of the embryo changing the spatial relationship between the involved structures. Paidas et al[18] studied the differentiation of the human cloaca and

concluded that as the embryo becomes a fetus it lengthens, grows, expands, and rotates counterclockwise through *transformation* resulting in loss of caudal curvature, the rapid growth of the ventral cloaca, and a decrease of the distance between the URS and the CM but these structures do not fuse and there is no actual descent of the URS. The work of several other authors, however, suggests that actual descent of the URS and fusion of its tip with the CM do play a role in cloacal differentiation.[19-21]

It appears likely, therefore, that both *transformational changes* and descent of the URS play a vital role in cloacal partitioning and, as suggested earlier,[20-21] these processes are complementary. Apart from studies that show the descent of the tip of the URS keeping the opening of the Wölffian duct (WD) as a marker,[19-20] the very fact that in males, the fistula may open cranial to the verumontanum as in CPC, at the level of the verumontanum (corresponding to the openings of the vasa efferentia [WD]) or at varying levels caudally (bulbar urethra or the perineum) suggests that actual descent of the URS does occur.

In recent years, the molecular and genetic factors behind the causes of various congenital malformations have been studied in detail, especially in animal models subjected to the effects of teratogens. As regards CPC, considering its unique geographical pattern of occurrence, it is likely that an, as yet unknown, environmental teratogen[1-4] may affect the genes and factors such as *HOXD-13* and *BMP4* responsible for mesodermal differentiation and proliferation in the URS and the adjacent hindgut and urinary system.[22-24] This must be a very 'early' error as the CVF opens in the trigone, cranial to the verumontanum, suggesting its occurrence even before cloacal septation starts. As suggested by Sasaki et al[22] and others,[23-24] if the genetic pathways responsible for mesodermal differentiation in the URS are affected, the development of the mesenchyme of the adjacent hindgut can also be affected. Probably, the unique, specific teratogen affecting mesodermal differentiation in the URS in CPC also causes 'dysmorphogenesis' of the adjacent shortened hindgut with later development of the colonic pouch.[4-7] The large and flaccid UB seen in several boys in our study as well as reported earlier,[25-28] may result from additional involvement of the mesenchyme of the adjacent allantois which forms the UB.[22]

In our study, apart from the length of colon affected, the radiological and CUS findings regarding the CVF in boys with Type IV CPC were identical to those in the more severe Types I- III CPC. A variation in the cranial extent of the error in mesodermal differentiation in the URS and adjacent hindgut can account for the varying subtypes (Types I–IV) of CPC.[7] A unique finding in a distal dye study of the colonic pouch, especially in Type IV CPC, was the frequent bilobed appearance of the UB. This is likely to be due to the long CVF closely opposed to the trigone in the midline, tethering this region.

It is of interest that in the usual ARM with a recto-bladder neck fistula, there is a high incidence of associated major renal anomalies, severe sacral defects, and often, other major organ anomalies.[12] Our findings as well as several other previous reports and studies suggest that, at least in the Indian subcontinent, considering the very early embryologic defect in CPC, major other organ malformations are not very common[2-5,7,25] As in 21/ 25 (84%) patients in our series, the sacrum is also usually normal,[4,7,25] although there is a high incidence of hypospadias and undescended testes, both considered relatively minor abnormalities.[7,29] These findings suggests that CPC in the Indian subcontinent usually results from an early but very 'localized' error in cloacal septation by the URS along with faulty development of the structures adjacent to the URS, i.e. the post-arterial limb of the midgut and hindgut and, in some cases, the developing UB.

A review of the literature, however, shows that from outside the Indian subcontinent, major associated malformations including severe sacral anomalies, renal anomalies, major abnormalities of the external and internal genitalia, pseudo-exstrophy, cardiac defects, craniofacial defects, and facial dysmorphism have frequently been reported with CPC.[30-35] A few case- reports from India have also described a similar association of major anomalies.[36-38] In several such reports, the findings are very similar to those in the cases of *urorectal septum malformation syndrome* (URSMS) or the *caudal dysgenesis syndrome*.[39] Currarino[27] studied ARM with rectovesical fistula and described a simple benign form with a normal bladder and urethra, commonly unilateral/ bilateral VUR, usually a normal sacrum and kidneys, and an enlarged rectosigmoid in some cases. In one case illustrated the UB was large. Currarino[27] suggested that these cases may represent milder forms of CPC. Another

variant of ARM with rectovesical fistula was described, as represented by the autopsy findings of 11 cases detailed by Magnus.[40] All cases had multiple severe malformations of the urethra, major renal anomalies, phallic enlargement in girls, and major sacral abnormalities. The UB was often thick-walled and dilated with a greatly enlarged distal bowel (sigmoid-rectum) having an abrupt transition to normal, very similar to that in CPC.[27-40] It is therefore likely that cases of CPC with major, severe associated anomalies, usually reported from outside the Indian subcontinent, result from an 'early' (around 3rd-week gestation) but severe and generalized defect in mesodermal organization in the entire caudal development area. Most cases also have other major-organ defects, suggesting an early, more generalized defect of mesodermal organization similar to that responsible for URSMS.[39]

The clinical implications of our findings include the necessity of cautious and meticulous dissection of the long CVF, which is adherent to the trigone and opens at the bladder neck, very close to the openings of the vasa efferentia. In Type IV CPC, in view of the long CVF, it is important to construct a sufficiently proximal diverting colostomy, to facilitate pull-through of the colonic pouch during definitive surgery. Prior to definitive surgery, radiologic studies are essential for assessment of the anatomy of the colonic pouch and its associated CVF, the LUT anatomy, and also to rule out VUR. The UB morphology along with the verumontanum being pulled- up into the trigone proximal to the bladder neck can have clinical consequences on bladder dynamics and male fertility. Thus, long-term follow-up is essential.

Conclusion

Males with all subtypes of CPC had a long terminal CVF opening in the trigone just proximal to an abnormal verumontanum, which itself was pulled up into the bladder neck. The UB was often large and lobulated. Apart from a high incidence of VUR, other major abnormalities were uncommon. Identification of the specific teratogen responsible and the critical gene and signaling pathways that are affected should be the subject of further studies.

Ethical Consideration

Approval was obtained from the Ethics Committee of Lady Hardinge Medical College & Kalawati Saran Children's Hospital, New Delhi- 110001.

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Conflict of interests

All authors declare that they have no conflict of interest

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