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Congenital pouch colon: follow-up and functional results after definitive surgery

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Abstract

Purpose: In this study, functional results with regard to fecal continence levels and other parameters were studied in 22 patients with congenital pouch colon associated with anorectal agenesis (CPC) more than 3 years old who had undergone definitive pull-through surgery 1 to 13 years earlier. An attempt was made to formulate treatment protocols for management of fecal incontinence and other problems associated with CPC.

Methods: The study sample consisted of 14 males and 8 females. Three of the 8 female patients had had a cloacal malformation. The medical records of the patients were scrutinized and they were classified into 4 subtypes based on the length of normal colon proximal to the colonic pouch. The patients were further categorized into 3 groups based on the terminal bowel that had been pulled-through, namely, the ileum or colon proximal to the colonic pouch or a tubularized segment of the colonic pouch. The somatic growth of the patients was studied. Clinical assessment of fecal continence was performed by the Kelly and the Kiesewetter and Chang scoring systems. A computed tomographic scan of the pelvis with a barium enema was performed to assess the terminal bowel and its placement as well as the bony and muscular anatomy of the pelvis. The urinary system was assessed by a clinical history as well as by abdominal ultrasound and a micturating cystourethrogram. Various treatment modalities including dietary modifications, drugs, and enemas were instituted in patients with poor continence levels, and the response to treatment studied.

Results: Thirteen patients (59.2%), all with an ileal pull-through, had height and weight less than 50% of that expected for their ages. Overall fecal continence was "poor" in 17 patients and "fair" in only 5 patients. Patients with pull-through of either ileum or normal colon often had very frequent passage of liquid or semisolid stools, whereas the 4 patients with pull-through of tubularized colon had infrequent passage of semisolid stools with abdominal distension and bloating. One of these 4 patients had massive colonic redilatation necessitating surgical correction. Mucosal prolapse and perineal excoriations were frequent findings. Ultrasonography and micturating cystourethrogram showed hydroureteronephrosis and vesicoureteric reflux in 5 patients. Radiologic assessment revealed that there were no significant sacral abnormalities and the striated sphincteric musculature was well developed, although the levator ani was thinner than normal in 15 patients (68%). The bowel was very well placed in the sphincteric complex in 19 patients (86%). In 7 of the 13 patients who had pull-through of normal ileum or colon,

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some improvement in continence levels was seen 3 to 6 months after institution of dietary measures, loperamide, and saline-water enemas. Two of 3 patients with pull-through of tubularized colon improved to some extent with colonic washouts alone. Overall, quality of life was poor in the 22 patients.
Conclusions: Despite the fact that the sacrum is usually normal, the sphincteric musculature well developed, and the terminal bowel well placed without any anal strictures, long-term prognosis with regard to fecal continence, growth and development, and quality of life appears to be dismal for all subtypes of CPC, irrespective of the type of definitive surgery performed. Corrective measures also appear to be of limited value. Various newer management modalities for management of fecal incontinence may be considered, but in several patients a permanent abdominal stoma may be a more practical solution.

Congenital pouch colon associated with anorectal agenesis (CPC) is an unusual malformation in which a pouch-like dilatation of a shortened colon is associated with an anorectal malformation. The pouch usually terminates in a fistula communicating with the genitourinary tract. The severity of the condition varies from complete absence of normal colon, the distal ileum opening into the colonic pouch, to the presence of a nearly complete length of normal proximal colon, with only the rectum or rectosigmoid colon being affected [1]. A unique feature of CPC is that most large series describing this anomaly have been reported from centers in northern India [2-7].

There are very few studies detailing the results and long-term follow-up after definitive surgery in patients with CPC. In addition, many different treatment protocols have been used for the several anatomical variants of CPC so that comparison of results is difficult. In this study, we present a detailed evaluation of fecal continence levels and other parameters in 22 patients with CPC who had undergone definitive pull-through surgery 1 to 13 years earlier. The response to various corrective measures for treating fecal incontinence in these patients was also assessed. The options available for the management of fecal incontinence and other long-term problems seen in patients with CPC are discussed.

1. Patients and methods

The study sample consisted of 22 patients with CPC who had undergone definitive pull-through surgery at Kalawati Saran Children's Hospital, New Delhi, India, and were registered for follow-up from October 2002 to July 2004. The criteria for inclusion in the study were age more than 3 years, at least 1 year to have elapsed after the pull-through surgery (if performed without a protective ileostomy/colostomy) or after closure of the protective enterostomy, and informed parental consent.

There were 14 male and 8 female patients (male/female ratio, 1.7:1). The age at the time of evaluation ranged from 3 to 16 years (mean, 5.95 years), the last surgery having been performed 1 to 13 years (mean, 53 months) earlier. The medical records of the patients were scrutinized in detail. The patients were categorized into 4 group types based on the length of normal colon proximal to the colonic pouch, as described by Narasimharao et al [3] (Fig. 1): type I, normal colon is absent and the ileum opens directly into the colonic pouch; type II, the ileum opens into a short segment of cecum which then opens into the colonic pouch; type III, presence of a significant amount of normal colon between the ileum and the colonic pouch; and type IV, presence of near normal colon with only the terminal portion of the colon (rectum and a varying portion of the sigmoid) converted into a pouch.

Table 1 shows the categorization of patients on the basis of sex and the subgroup type of CPC. Of the 8 female patients, 3 had type I CPC with a cloacal deformity, 4 had type I/II CPC with a colovesical or colovestibular fistula, and 1 had a type IV CPC with a colovestibular fistula.

In 7 patients (31.9%), all with type I/II CPC, the initial surgical procedure had consisted of subtotal excision of the pouch after division/ligation of its fistula with the genitourinary tract, tubular colorraphy (TC), and an end-colostomy [5-8]. A window colostomy of the pouch had been performed in another center on a child with type II CPC. Excision of the colonic pouch with a proximal end ileostomy/colostomy, which is our current policy for all subtypes of CPC, had been performed in 14 patients (63.5%). These included 9 patients with type I/II CPC and all 5 patients with type III/IV CPC.

The age at the time of definitive pull-through surgery ranged from 11 months to 7 1/2 years. In all cases, the posterior sagittal route [9] had been used, with the additional abdominal component necessitated by the nature of this anomaly (abdomino–posterior sagittal anorectoplasty). Based on the procedure performed during definitive surgery, the 22 patients were categorized into 3 groups (groups I-III) as shown in Table 2. Group II included 3 patients in whom TC had been performed in the neonatal period. However, during definitive surgery, as the colonic tube had become a small, redilated pouch, it was excised and pull-through of normal proximal ileum performed.

The 3 patients with type I CPC and persistent cloaca had undergone reconstruction for the cloacal anomaly at the time of the pull-through surgery. One girl had a very short common cloacal channel, and a 2-cm posterior cut-back

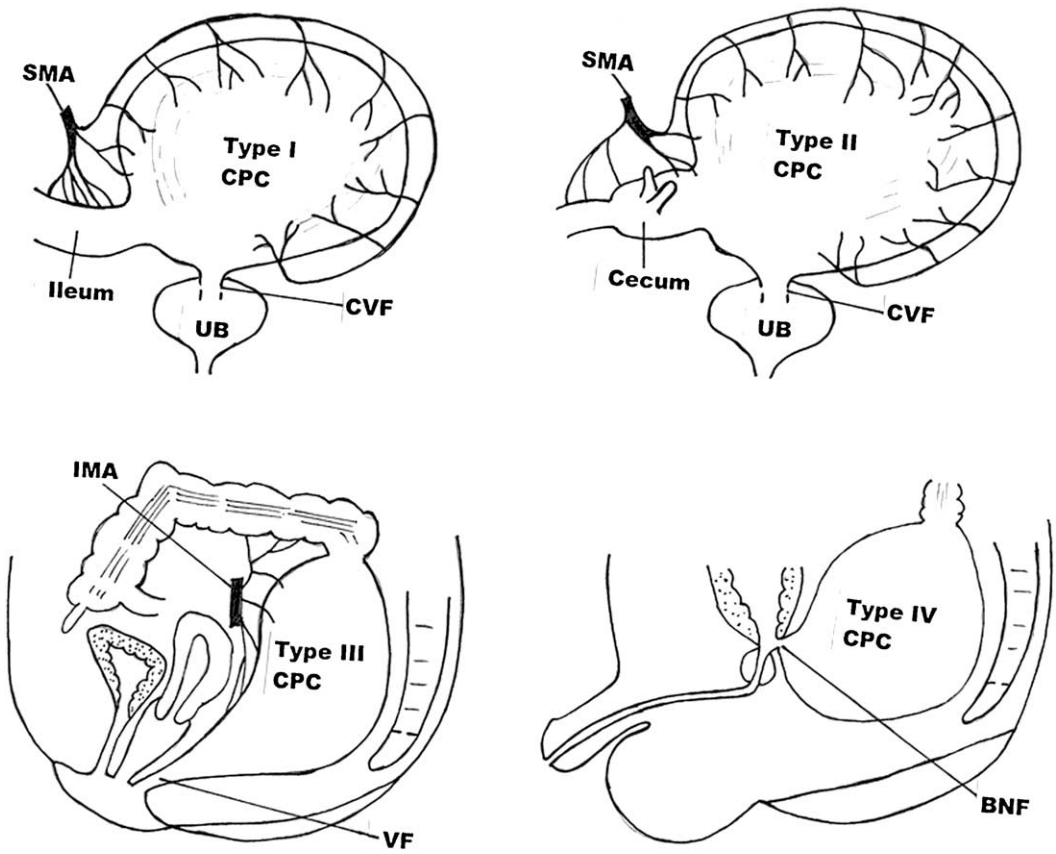

Fig. 1 Anatomical subtypes of congenital pouch colon associated with anorectal agenesis. SMA indicates superior mesenteric artery; IMA, inferior mesenteric artery; UB, urinary bladder; CVF, colovesical fistula; VF, vestibular fistula; BNF, fistula at bladder neck.

vaginoplasty had left her with a mild hypospadias. In 2 patients, the common channel was 3 and 7 cm long, respectively, the latter patient requiring a 7-cm graft from the mid-ileum for vaginal reconstruction.

The somatic growth of the patients was determined by comparing their height and weight against matched growth charts for Indian children [10]. Clinical evidence of malabsorption or any vitamin or mineral deficiencies was noted. Anal strictures were ruled out by digital examination or by passage of an adequately sized anal dilator.

Clinical assessment of continence was performed both by the Kelly [11] and the Kiesewetter and Chang [12] scoring systems. In the Kelly system [11], assessment of continence is based on leakage phenomena (staining/smearing and occurrence of accidental defecation or soiling) as well as strength of puborectalis muscle on digital examination. A total score of 5 to 6 points is considered "good"; 3 to 4 points, "fair"; and 2 points or less, "poor." The Kiesewetter and Chang [12] criteria assess continence qualitatively at 3 levels—good, fair, or poor—based on the degree of social acceptability achieved. Episodes of enteritis or enterocolitis, as evidenced by passage of loose, watery stools, often foul-smelling, accompanied by abdominal distension with or without fever, were also evaluated.

Urological evaluation included clinical assessment of urinary control and the pattern of micturition, biochemical renal function tests, an abdominal ultrasound, and micturating cystourethrogram. If any abnormality was detected in these tests, radionuclide diethylenetriamine pentaacetic acid and ^{99m}Tc-dimercaptosuccinic acid renal scans were obtained.

A barium enema/defecogram examination was performed to assess the placement and caliber of the terminal bowel, any dilatation of the proximal bowel, and any bony pelvic or sacral abnormality [11,13]. A computed tomographic scan of the pelvis with rectal contrast (barium solution) was performed, and the caliber of the levator ani and the external sphincter complex was graded as good, fair, or poor [14].

In patients with unsatisfactory fecal incontinence levels, corrective measures consisted of combinations of dietary manipulation, drug therapy, and enemas or rectal washouts.

Table 1 Categorization of patients based on sex and subgroup types of CPC (n = 22)

Type of CPC	Male	Female	Total
Type I	3	4	7
Type II	7	3	10
Type III	2	0	2
Type IV	2	1	3

Table 2 Clinical continence scores (n = 22)

Group	No. of patients (n)	Kelly's score	Kiesewetter score
Group I: pull-through of normal proximal colon (5-18 cm long) in patients with type II-IV CPC	5	0-2 = 3, 2-4 = 2	Poor = 3, fair = 2
Group II: pull-through of ileum in patients with type I/II CPC	13	0-2 = 12, 2-4 = 1	Poor = 12, fair = 1
Group III: pull-through of tubularized colonic pouch in patients with type I/II CPC	4	0-2 = 2, 2-4 = 2	Poor = 2, fair = 2

Dietary manipulation for patients with frequent episodes of diarrhea included advice to increase dietary fiber, ensure adequate food hygiene, and avoid foodstuffs that evoke increased frequency of stools. In patients of group I/II with very frequent passage of liquid stools, management consisted of dietary measures along with the motility-lowering drug loperamide (1-2 mg orally, 3 times a day) to reduce stool frequency and improve stool consistency. If these measures were successful to some degree, saline-water enemas or washouts (12.5 mL/kg body weight, twice daily) were used to increase the dry interval. In patients of group III, only twice-daily saline-water enemas were used. Protective skin creams and antifungal ointments were also used, wherever required.

2. Results

In 13 patients (59.2%), all from group II, the height and weight were less than the 50th percentile of the expected values for their age; in 5 patients (23%), these parameters were in the 50th to 80th percentile; and only 4 patients (18%) had normal somatic growth patterns. No patient had any obvious clinical evidence of vitamin, essential mineral, or fatty acid deficiency.

As shown in Table 2, in 17 patients (77.2%) continence was assessed as poor by both scoring systems. Continence was fair in 5 patients (22.7%) and no patient had good continence. Continence was worst in group II patients, with 12 (92.3%) of 13 having poor continence scores. In 18 patients (82%), all from groups I and II, the basic pattern of defecation consisted of frequent (5-25 times per day) passage of liquid or semisolid stools both during the day and night. Only 2 patients, both more than 10 years of age and from group I, reported voluntary bowel movements (2-4 times a week). In the 4 patients from group III, the pattern of defecation consisted of infrequent passage of small amounts of semisolid stools, the act of defecation usually being preceded by abdominal distension, bloating, and cramps. One of these patients had recurrent massive abdominal distension, relieved only partially after the passage of stools. He had had several episodes of severe enterocolitis in the past.

Fifteen patients (68%) had 1 to 2 episodes of enteritis/enterocolitis every month. Of these, 13 patients were from group II and 2 patients from group III. In the patients from group II, these episodes appeared to be acute infective exacerbations of their chronic diarrheal condition. Anal stenosis or strictures were not present in any patient. However, 18 patients (81.8%) had significant perineal excoriations and granulomas, usually with superadded fungal and bacterial infections (Fig. 2). Only 4 patients (all with fair continence) had a dry perineum. Four patients had a moderate degree of mucosal prolapse at the anal verge (Fig. 2).

Thirteen patients (59.2%) had normal urinary continence; 3 patients, all more than 5 years of age, had nocturnal enuresis; whereas 5 patients had both diurnal and nocturnal incontinence. This last group included 2 patients with a cloacal anomaly in whom urinary incontinence was present from birth, and 3 patients in whom it had manifested only after the definitive surgery. Persistent continual dribbling was noted in a girl operated on for a type I CPC with persistent cloaca. Renal function test (RFT) results were normal in all 22 patients. Ultrasound revealed unilateral hydroureteronephrosis (HUN) in 5 patients (22.7%), 3 of whom had a type II CPC, and 2 a type III CPC. Micturating cystourethrogram showed a low-lying bladder neck in 3 patients and unilateral vesicoureteric reflux (VUR) in the 5 patients who had HUN on ultrasound (grade III in 4 boys aged 3-5 years; grade I in a 15-year-old adolescent boy). Diethylenetriamine pentaacetic acid/^{99m}Tc-dimercaptosuccinic acid renal scans were normal in the 8 patients in whom they were performed.

On radiological assessment, the levator ani was well developed (good grading) in 7 patients (32%) and thinner than normal (fair grading) in 15 patients (68%) (Fig. 3), whereas the sphincteric musculature was usually well

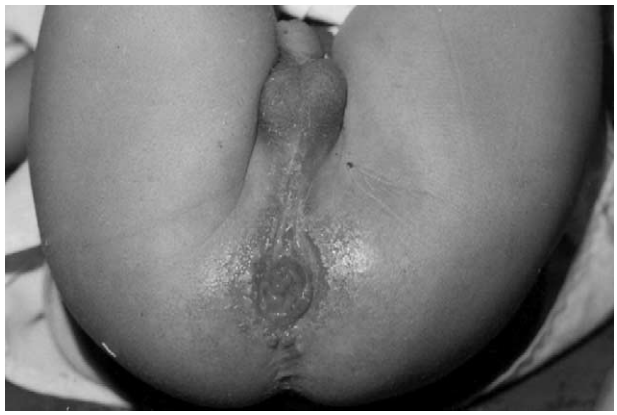

Fig. 2 Extensive perineal excoriations and mucosal prolapse in a child with an ileal pull-through.

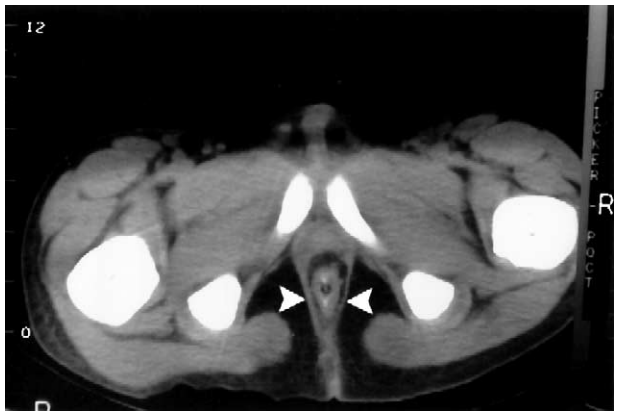

Fig. 3 Computed tomographic scan of the pelvis showing a relatively thin levator ani (arrowheads) and centrally placed bowel in a boy who had pull-through of normal proximal colon for type II CPC.

developed (Fig. 4). All 22 patients had an essentially normal sacrum with 4 to 5 sacral segments, with 2 patients having a minor degree of spina bifida at S3 to S4. The bowel was accurately placed anterior to the levator ani and within the center of the sphincteric muscle complex in 19 patients (86%) (Figs. 3 and 4), whereas in 3 patients, some eccentric placement of the bowel was present. Mesentery with presence of mesenteric fat was visible on the computed tomographic scan in 5 patients (22.7%). In 2 patients with type I/II CPC from group III, there was moderate redilatation of the tubularized colon, whereas in one 4-year-old boy, the tubularized colon had undergone massive redilatation (Fig. 5), necessitating resection of the redilated pouch and anastomosis of the normal proximal ileum to the distal remnant of the pouch just above the peritoneal reflexion.

The response to treatment of fecal incontinence could be evaluated at 3 and 6 months only in 16 patients (4 from group I, 9 from group II, and 3 from group III). Overall, little or no improvement was noted in 7 patients. In 2 patients

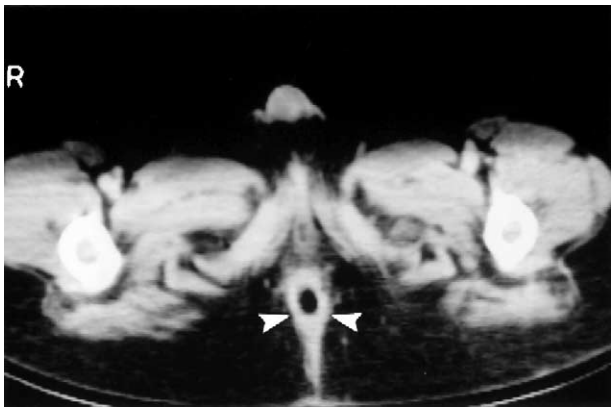

Fig. 4 Computed tomographic scan of the pelvis showing the ileum placed in the center of a well-developed striated muscle complex (arrowheads) in a boy who had type I CPC with an ileal pull-through.

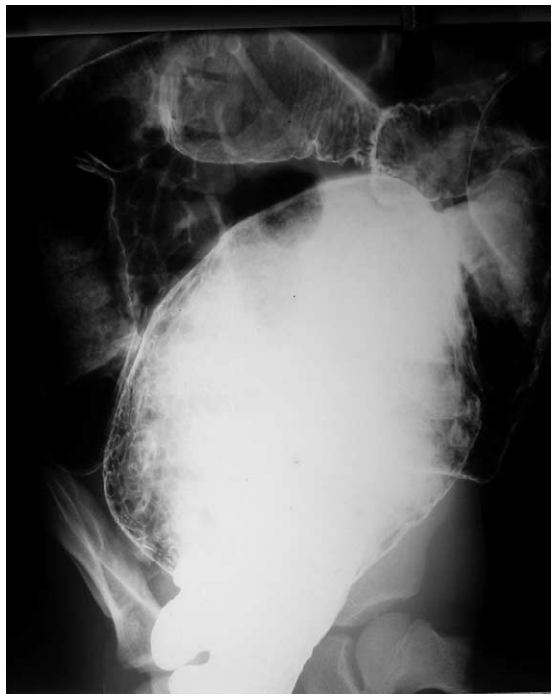

Fig. 5 Barium enema showing massive colonic redilatation in a 4-year-old boy who had undergone pull-through of a tubularized segment of the colonic pouch.

from group I and 5 patients from group II, continence improved from a score of 1 to 2 or 3 in 6 patients (with a dry interval of 4-6 hours after the enema), and from 0 to 1 in 1 patient. Healing of perineal excoriations, aided by the use of protective skin creams and antifungal ointments, was also noted in these patients. Five patients reported side-effects of loperamide therapy, necessitating reduction in its dose. In 2 patients from group III, the continence score improved from 2 to 3 after institution of colonic washouts, although symptoms of abdominal distension and bloating persisted, albeit to a lesser degree. Excision of the mucosal prolapse was beneficial in 3 patients.

Significantly, older children had better continence scores. However, the overall quality of life for the 22 patients was very poor. Of the 12 patients older than 5 years, 9 (75%) were yet to start schooling, and 2 of the 3 patients who had joined school had dropped out because of peer pressures, fecal incontinence, or chronic ill health.

3. Discussion

In a study conducted 3 to 8 years after pull-through surgery following coloplasty (TC) to the size of a 12- to 16-F tube, Wakhlu et al [8] found that in 16 of 18 patients, the tubularized colon had the caliber of a normal colon, whereas 2 patients had significant colonic redilatation. Subjective evaluation of fecal continence showed that 10 patients had good continence, 5 had acceptable continence with occasional soiling, whereas 3 patients were inconti-

nent. Other authors have also reported satisfactory results in such patients, although the degree of tapering of the pouch was not specified and only short-term results were reported [3,4,15-17].

Interestingly, a study performed on patients in whom excision of the colonic pouch with pull-through of normal proximal ileum or colon had been performed 3 to 8 years previously showed that weight gain and growth were in the 97th percentile, whereas 9 (81%) of 11 had socially acceptable continence [8]. Others have also reported similar good results in patients managed in this fashion [3,4,18].

Our findings and results are, however, in contrast to the favorable outcome as reported by these authors, perhaps because our assessment was more detailed and objective.

As also described in earlier reports [4,6,7], despite the very high nature of the associated anorectal malformation, the sacrum is usually normal in all subtypes of CPC and, surprisingly, even in those with a cloacal deformity. The sphincteric musculature and striated muscle complex are well developed, although the levator ani may be thinner than normal, perhaps as a consequence of the high pelvic location of the pouch and its terminal fistula [1,7]. During abdomino–posterior sagittal anorectoplasty, it is easy to place the terminal bowel in the correct anatomical position, with a minimal risk of damage to the pelvic nerves. None of the patients in this series had anal stenosis or anal strictures. Thus, several factors that are important for normal fecal continence are usually present in patients with CPC. Despite this, long-term outcome with regard to fecal continence appears to be dismal. Perhaps the most important reason for this is the complete absence of normal terminal bowel. The rectum is a natural reservoir whose absence leads to diarrhea and frequent, often unmanageable, loose stools [19,20]. The rectum is also important for sensation and proprioception [21], and an anorectal resistance zone in the distal rectum of 3.5 to 5.0 cm is very important for both gross and fine fecal control [22,23]. The value of the internal sphincter in preserving fecal continence has also been stressed [20,24,25]. A complete or near-complete absence of normal colon in type I/II CPC, made worse by the absence of the ileocecal valve (type I CPC), also predisposes to diarrhea and liquid stools with resultant fecal incontinence [1,7]. The presence of mesenteric fat around the terminal bowel in patients with an ileal or colonic pull-through would also impair proprioceptive ability. In addition, the diet in India is usually nonconstipating, and as most of our patients are from the poorer sections of society, episodes of infective diarrhea are very common.

It is interesting that continence levels were best in patients belonging to Group I or Group III. In Group I patients, this can be explained by the presence of a length of normal proximal colon as well as the ileocecal valve. In patients of group III, continence was fair in 2 of 4 patients. However, this appears to be largely caused by infrequent evacuation of semisolid stools collected in the poorly peristaltic tubularized colonic pouch.

Fecal continence was worst in patients with type I/II CPC belonging to Group II. These patients also showed evidence of growth retardation. This suggests that preservation of a segment of the colonic pouch by TC in patients with type I/II CPC does benefit the patients. However, this benefit has to be titrated against the complications resulting from retention of the inherently abnormal tissue of the colonic pouch whose histologic features include disorganized thinned-out muscle layers, splaying of the muscle fibers, and abnormalities of innervation [26]. These features probably account for the weak peristalsis characteristic of the colonic pouch, which leads to ineffective evacuation, stasis, and possible enterocolitis [2,16,26,27], as well as its marked tendency to undergo redilatation, even after adequate tapering [8,28]. Although the degree and timing of presentation with redilatation are unpredictable, it may be apparent even at the time of definitive surgery [1,7,29]. Massive life-threatening colonic redilatation necessitating surgical intervention has been reported in patients who had earlier undergone TC and pull-through surgery [8,28]. These considerations explain our current policy of excising the colonic pouch at initial presentation in all patients with CPC.

Assessment of the urinary tract is limited by the absence of urodynamic studies. Significantly, although the incidence of VUR in patients with CPC has been reported to be very high [1,3,5,7], only 5 patients in this series had VUR. Only one of these patients was more than 5 years of age, suggesting that VUR in patients with CPC improves with age.

It is very difficult to suggest management protocols for these unfortunate children. In patients of Group III, colonic washouts improved continence although chronic ill-health, bloating, and enterocolitis remained significant problems. Colonic motility–enhancing drugs such as cisapride have also been advocated for these patients [8], and daily low-dose metronidazole may reduce the risk of enterocolitis. These patients need close and regular follow-up for several years so that colonic redilatation with the possibility of disastrous consequences can be detected early [28].

In patients with pull-through of the ileum or normal colon, the absence of a reservoir or an adequate length of normal colon means that drug therapy and washouts cannot achieve satisfactorily long “dry” intervals. This is especially true for patients with an ileal pull-through. Long-term use of loperamide or other constipating drugs can also have deleterious effects. Constructing a J-shaped or S-shaped reservoir or pouch of the terminal ileum for washouts [1] or a “pouch colon graft” on the ileum that is pulled down [30] can be attempted to improve fecal continence. However, absence of any length of the normal anorectal resistance zone in patients with CPC indicates that these procedures may not provide satisfactory long-term benefits.

In recent years, several innovative techniques have been described for treating chronic fecal incontinence in children. An anal plug [31] is unlikely to be effective in patients with CPC as the degree of incontinence is usually quite severe. Gracilis muscle reimplantation and electrostimulation [32]

may not be necessary as the sphincteric musculature is usually well developed. More promising techniques appear to be the use of an artificial anal sphincter or sacral nerve electrostimulation [32].

It is somewhat encouraging that continence levels are better in older children and this may be caused by maturation of bowel function with time [33,34], enhancement of social motivation [34], or simply because, over time, older children learn to voluntarily contract the perineal musculature to minimize fecal leaks. In this group of patients, although limited by the absence of anal sensation and adequate anal resting pressures, an anal reeducation program using electrostimulation and "biofeedback" therapy may provide the best chances of sustained improvement in fecal continence levels [32,35,36].

In conclusion, this study confirms our earlier findings [1,7] that long-term functional results after pull-through surgery in patients with CPC are largely unsatisfactory. With the surgical techniques described and used so far in the literature, it appears that only patients with a fairly long segment of normal proximal colon have the potential to achieve a fair level of fecal continence. Especially in type I/II CPC, despite the use of the management options available to us at present, diarrhea and severe perineal excoriations may remain a major persistent handicap. A permanent abdominal end-enterostomy, although not acceptable to the majority of our patients or their parents, may well be a more practical long-term treatment option for several of these unfortunate children.

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