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Two-staged management for all types of congenital pouch colon

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ABSTRACT

Background: The aim of this study was to review our experience with two-staged management for all types of congenital pouch colon (CPC). **Patients and Methods:** This retrospective study included CPC cases that were managed with two-staged procedures in the Department of Paediatric Surgery, over a period of 12 years from 1 January 2000 to 31 December 2011. **Results:** CPC comprised of 13.71% (97 of 707) of all anorectal malformations (ARM) and 28.19% (97 of 344) of high ARM. Eleven CPC cases (all males) were managed with two-staged procedures. Distribution of cases (Narsimha Rao *et al.*'s classification) into types I, II, III, and IV were 1, 2, 6, and 2, respectively. Initial operative procedures performed were window colostomy (n = 6), colostomy proximal to pouch (n = 4), and ligation of colovesical fistula and end colostomy (n = 1). As definitive procedures, pouch excision with abdomino-perineal pull through (APPT) of colon in eight, and pouch excision with APPT of ileum in three were performed. The mean age at the time of definitive procedures was 15.6 months (ranges from 3 to 53 months) and the mean weight was 7.5 kg (ranges from 4 to 11 kg). Good fecal continence was observed in six and fair in two cases in follow-up periods, while three of our cases lost to follow up. There was no mortality following definitive procedures amongst above 11 cases. **Conclusions:** Two-staged procedures for all types of CPC can also be performed safely with good results. The most important fact that the definitive procedure is being done without protective stoma and therefore, it avoids stoma closure, stoma-related complications, related cost of stoma closure and hospital stay.

Key words: Anorectal malformation, congenital pouch colon, congenital short colon, staged procedures

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INTRODUCTION

Congenital pouch colon (CPC) is a congenital anomaly in which whole or part of the colon is replaced by a pouch-like dilatation that communicates distally with the urogenital tract by means of fistula and is uniformly associated with anorectal malformations (ARM). This anomaly is also described as congenital short colon and pouch colon syndrome.[1-6] CPC is seen much more frequently in northern, north western, and central part of India.[1-7] Few sporadic cases have also been reported from western India and other parts of the world.[8-13] The standard procedure for the management of CPC is a three-staged procedure.[1-3,5] We are herein reporting our experience with two-staged management for all types of CPC with a brief review of literature.

PATIENTS AND METHODS

This is a retrospective, single institution study of CPC managed in the Department of Paediatric Surgery over a period of 12 years from 1 January 2000 to 31 December 2011. CPC (n = 11) managed with two-staged procedures were reviewed in detail. The invertogram was the choice of the investigation during the initial admission to investigate the case of ARM. CPC was suspected/possible to diagnose (pre-operatively) with characteristic radiological findings [Figure 1a and b]. Distal colostogram was ordered prior to the definitive procedure [Figure 2]. The pre-operative preparation was done with normal saline washes twice daily for 2 days prior to operation, and the post-operative management was as the standard protocol for any exploratory laparotomy; i.e., intra-venous fluids, antibiotics, nasogastric aspiration, etc. Oral feedings allowed on fourth post-operative day in most of the cases. The outcome of the various surgical procedures was observed in the form of major surgical complications (needing surgical interventions) and mortality.

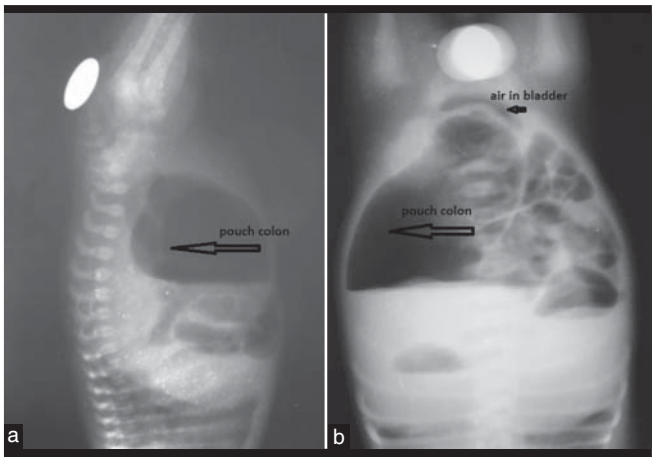


Figure 1: (a) Invertogram showing dilated colon (CPC), (b) Invertogram showing dilated colon (CPC) with gas in the urinary bladder

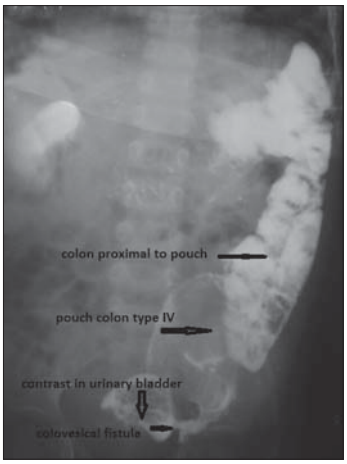


Figure 2: Distal cologram showing type IV CPC, colovesical fistula, and contrast in the urinary bladder

RESULTS

During a period of 12 years from January 2000 to December 2011, 707 patients with ARM were admitted and managed at the author's department of paediatric surgery. We classified our cases of ARM according to the Wingspread classification as low, intermediate and high. We classified our CPC cases as described by Narsimha Rao *et al*. in 1984. They classified CPC cases into four types and are as follows:

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 caecum which then opens into the pouch.

Type III: Presence of a significant length of normal colon between the ileum and the colonic pouch.

Type IV: Presence of near normal colon with only the terminal portion of colon (sigmoid and rectum) converted into a pouch.

CPC comprised of 13.71% (97 of 707) of all ARM and 28.19% (97 of 344) of high ARM. All the patients of CPC had a high variety of ARM. Demographics of CPC managed in the department of paediatric surgery over a period of 12 years are provided in Table 1. Fifty-three definitive procedures have been performed; 38 (71.69%) managed with three-staged, 11 (20.75%) with two-staged, and 4 (7.54%) with primary single-stage procedures during the study period. Details of the CPC cases (n = 11) managed with two-staged procedures are provided in Table 2.

The stages of surgical procedures performed during two-staged procedures in this study are as follows:

A. First stage of the two-staged procedure: The surgical procedures performed during the initial procedures are provided in Table 2. In six of our cases, window colostomies were created; in three patients window colostomies were created by general surgeons (not specialist in paediatric surgery) and referred to us for definitive procedures and in three cases window colostomies were created by us due to poor general conditions of the patients. Complications are frequently observed following initial operations for fecal diversion and includes mild skin excoriation in six, colostomy prolapse in four [Figure 3], and stoma stenosis in two.

B. Second stage of the two-staged procedure: During definitive operation, abdominal exploration was done through left pelvic incision. Colovesical fistula (CVF) was found in all the cases, and was identified, transfixed, ligated and cut. Pouch colon was excised including the stoma. Abdomino-perineal pull through (APPT) of proximal colon in (n = 8) [Figure 4]

Table 1: Demographics of congenital pouch colon (January 2000 to December 2011)

S. No.	Type of cases	Total (%)
Anorectal malformations (Wingspread classification)	Total number of low ARM	255 (36.06)
	Total number of intermediate ARM	108 (15.27)
	Total number of high ARM	344 (48.65)
	Total number of ARM (low + intermediate + high)	707
Congenital pouch colon	Total number of CPC	97
	Total number of male CPC	78 (80.41)
	Total number of female CPC	19 (19.58)
	Male: female ratio of CPC	78:19 (4.1:1)
Congenital pouch colon (Narsimha Rao *et al*. classification)	Type I	31 (31.95)
	Type II	10 (10.30)
	Type III	27 (27.83)
	Type IV	29 (29.89)

ARM: Anorectal malformations; CPC: Congenital pouch colon

Table 2: Details of congenital pouch colon (*n* = 11) managed with two-staged procedures

S. No.	Cases	1	2	3	4	5	6	7	8	9	10	11
Demographics	Age (days)	3	1	1	2	1	2	2	1	1	2	10
	Sex	M	M	M	M	M	M	M	M	M	M	M
	Weight (kg)	2.7	3	2.5	2.4	2.6	2.6	2.5	2.3	3	2.7	2.7
	Pre-operative diagnosis	Yes	No	No	No	No	No	Yes	No	Yes	Yes	Yes
	Types of CPC	III	III	III	II	III	II	IV	IV	I	III	III
First stage (initial procedures)	Window colostomy	Yes	Yes	–	Yes	Yes	–	–	–	Yes	Yes	–
	Proximal colostomy	–	–	Yes	–	–	Yes	Yes	Yes	–	–	–
	End colostomy	–	–	–	–	–	–	–	–	–	–	Yes
	Stoma location	LLQ	LLQ	LLQ	LLQ	LLQ	LLQ	LLQ	LLQ	RUQ	LLQ	LLQ
Complications of stoma	Skin excoriation	Mild	Mild	Mild	Mild	–	–	Mild	–	–	Mild	–
	Stoma stenosis	–	–	–	–	–	–	–	Yes	Yes	–	–
	Stoma prolapse	–	–	–	Yes	Yes	Yes	–	–	–	Yes	–
	Treatment	Medical	Medical	Medical	Revision	Medical	Revision	Medical	Revision	Revision	Medical	–
Two-staged (definitive procedures)	Age (months)	46	3	9	17	6	6	11	6	10	5	53
	Weight (kg)	10	4	7	7.5	6.5	7	9	5.5	8	8	11
	Incision	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic	Lt. Pelvic
	CVF ligation	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-
	Pouch excision	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
	Coloplasty	No	No	No	No	No	No	No	No	No	No	No
	APPT colon	Yes	Yes	Yes	No	Yes	No	Yes	Yes	No	Yes	Yes
	APPT ileum	No	No	No	Yes	No	Yes	No	No	Yes	No	No
Complications	Anal stenosis	–	–	–	–	–	Yes	Yes	Yes	–	Yes	–
	Mucosal prolapse	–	–	–	–	–	–	Yes	–	Yes	–	–
	Others	IntObst	Gangrene	Leak	–	–	–	–	Yes	–	–	Leak
	Treatment (surgical)	No	End stoma	Once	–	–	Once	Twice	Twice	Once	Once	Once
Follow-up	Duration (months)	1 5	0 (Lost FU)	0 (Lost FU)	3	6	6	51	42	15	1	0 (Lost FU)
	Cosmesis	Good	–	Good	Good	Good	Good	Good	Good	Good	Good	–
	Fecal continence	Good	–	–	Good	Good	Fair	Good	Good	Fair	Good	–

APPT: Abdomino-perineal pull through; CPC: Congenital pouch colon; Int Obst, Intestinal obstruction; Lt: Left; LLQ: Left lower quadrant; medical, Conservative/non-surgical; proximal colostomy, Colostomy proximal to pouch; RUQ: Right upper quadrant; stenosis, stoma/anal stenosis; FU: Follow up



Figure 3: Window colostomy for type III CPC, showing colostomy prolapse (case 10)

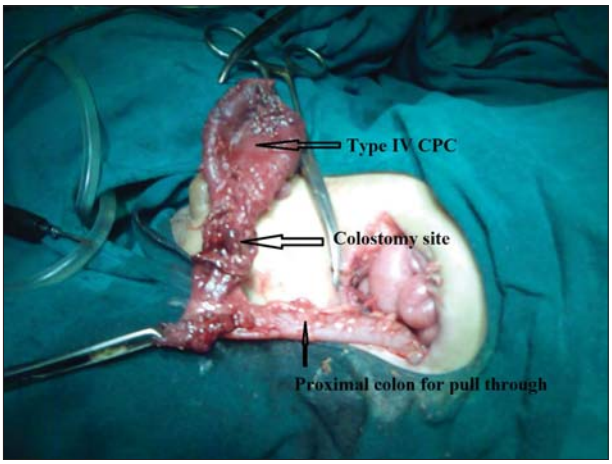


Figure 4: APPT of colon for type IV pouch colon (operative photograph)

and APPT of ileum in (n = 3) [Figure 5] were performed, depending upon the type of CPC and anorectoplasty accomplished. Intra-peritoneal drain was inserted in all, and the laparotomy was closed without protective stoma (colostomy or ileostomy). Three CPC cases with types 1 and 2 (cases 4, 6 and 9) were also treated with pouch excision and ileal pullthrough rather than coloplasty. In cases 4 and 6, colostomies were performed by general surgeons (not specialist in paediatric surgery) and referred to us for definitive procedures. In case 9, because of poor general condition on admission, a window colostomy was made through right supra-umbilical incision and was fixed at right upper quadrant of the abdomen. All the three above cases had stoma complications (prolapse in two and stoma stenosis in one) and needed revisions for correction of the complications. During exploratory laparotomies for definitive procedures in the above three cases, we found the coloplasty was not feasible, so pouch excision and ileal pullthrough were performed.

The mean age at the time of definitive procedures was 15.6 months, ranges from 3 to 53 months. Three of our cases returned for definitive procedures at 17, 46 and 53 months of age, respectively. The reason for delayed definitive procedures in the above three cases was poor follow-up after creation of stoma and delayed reporting for definitive procedures. In rest of the eight cases, the definitive procedures were performed within a year following colostomies. The mean weight at the time of the definitive procedure was 7.5 kg, ranges from 4 to 11 kg. Complications observed following initial operations for fecal diversion and following definitive APPT procedures, and these complications have been detailed in Table 2. Case 2 had gangrene of the pulled through colon and needed re-exploration and an end colostomy was created. He failed to return to us for further operative procedures. Case 3 had a minor colo-cutaneous fistula following the definitive procedure and needed minimal exploration and closure of the fistula in the follow-up period. In case 11, pouch excision and APPT of colon was performed. He developed major disruption of the laparotomy wound. He also had a minor fecal leak from the laparotomy wound, although he was also passing stool per-rectally. He needed repair of the disrupted wound and fecal leak there after he lost to the follow-up to us. The median follow-up after definitive procedures is 6 months with ranges of 1–51 months. Two of our patients lost to follow-up after definitive procedures, and another one (Case 2) who needed re-exploration with end colostomy for complication also failed to return to us for further operative procedures. The duration of the follow-up periods mentioned regarding the patients in Table 2 is the last visit of the patients with us after the definitive procedures. In cases 4, 5, and 6, the follow-up period were 3, 6 and 6 months, respectively. Case 10 also failed to return for follow-up after the definitive procedure. The reasons for not reporting to follow up may be either that they are doing well without any complications or reporting to other surgeons as most of the patients belong to the other cities. Fecal continence was assessed in the eight patients who were available for follow up. Continence was assessed by Kelly's method and observed as "Good" in six patients and "Fair" in two others on follow-up. There was no mortality following definitive procedures amongst above 11 cases included in the study.

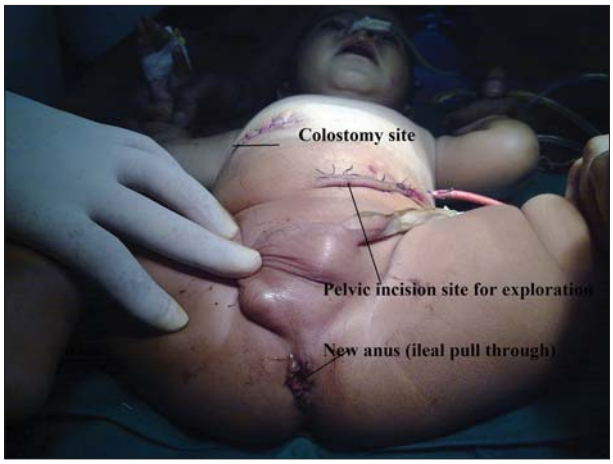


Figure 5: APPT of ileum for type I pouch colon (third postoperative day) showing the old stoma site, pelvic laparotomy site and new anus (case 9)

DISCUSSION

A CPC-like condition was first described in 1912 by Spriggs in a London hospital museum specimen with the absence of left half of the colon and rectum, but it was not named at that time.[1,2] Trusler *et al*. (1959) from Canada described a pouch-like dilation of shortened colon associated with high ARM, but that time also no name was given.[1,2,14] In 1972, Singh and Pathak from India reported six cases associated with imperforate anus and named this anomaly as "Short Colon".[15] Narsimha Rao *et al*. (1984) suggested the name "Pouch Colon Syndrome" and also proposed an anatomical classification of this condition, which has been widely accepted.[1,2,16]

The incidence of CPC among all the cases of ARM has been reported to occur from 2% to 18% and among the high ARM is 10–26% and almost all the series reported male preponderance.[1-6] High incidence (55.8%) of CPC with high ARM has been reported from Udaipur, Rajasthan,

India.[7] We managed 97 CPC cases during the study period of 12 years, and it comprised of 13.71% (97 of 707) of all ARM and 28.18% (97 of 344) of high ARM and we also observed male preponderance as 4.1:1. Almost all the series on CPC are reported from India; Lucknow, Delhi, Chandigarh, Varanasi, Bhopal, Kashmir, Udaipur, and Rohtak.[1-7,17,18] Recently, a series of seven cases of CPC have been reported from Saudi Arabia.[19] The embryogenesis and the exact cause of the unique geographical distribution of the CPC cases are yet to be ascertained.

CPC was first classified by Chiba *et al*. in 1976 and they classified it into five types.[1,2] A most widely accepted classification for CPC is based on the length of the normal colon present proximal to the dilated pouch; was given by Narsimha Rao *et al*. in 1984 and according to them, CPC is classified into four types.[1,2,16] Wakhlu *et al*. in 1996 simplified the classification as "partial short colon" (Type A) with the presence of >8 cm of normal colon proximal to the pouch and "complete short colon" (Type B) with the absence of normal colon or less than <8 cm of colon proximal to the pouch.[1] Gupta *et al*. in 2005 classified CPC into two types: "complete congenital pouch colon" and "incomplete congenital pouch colon" depending upon the presence or absence of the adequate normal colon for performing the pull-through operation.[2] Saxena *et al*. in 2008 classified CPC cases into five types based on the anatomic morphology.[7] CPC, which was not mentioned in the Wingspread classification of ARM in 1984, has now been accepted and included in the new International Classification (Krickenbeck) of ARM in 2005, as rare anomalies (being only regional).[20,21] We classified our cases of CPC according to Narsimha Rao *et al*. classification, and details are provided in Table 1.

A large loop of bowel with a single air fluid level occupying more than half of the total width of the abdomen on the plain abdominal X-ray, either erects or inverted, is almost diagnostic of CPC. The pouch is proximal to the pubococygeal line in the invertogram, and it is possible to diagnose pouch colon in about 75–90% of the cases.[1-7,17,18,22] In the present series of 11 cases included in this study, we were able to diagnose (pre-operatively) the case as CPC in five of eight, while three were referred from other cities for definitive procedures and the CPC was diagnosed during colostogram.

The standard procedure for the management of the CPC cases is three-staged procedures. The aim of the surgery is to utilize the available length of the colon for absorption and storage capacity, as well as for the capability for propelling the faecal matter onward with a continent anal opening. In cases with incomplete/partial pouch colon (types III and IV), an adequate length of the normal colon is present so that the pouch can be excised while the colonic function is still preserved. In cases with complete pouch colon (types I and II), as adequate length of the normal colon is absent, so the above objectives can be achieved only by tabularizing (coloplasty) the remaining colon.[1-3,5,6,17,18,23]

Three-staged management of CPC consists of:

A. Initial operative procedure: It includes either of one (a) proximal colostomy, window colostomy, or ileostomy with or without fistula division, (b) division of fistula and end colostomy, excision of pouch and end colostomy, and (c) division of fistula, coloplasty and end colostomy.
B. Definitive operative procedure: It includes either of one (a) pouch excision and APPT of colon, (b) pouch excision with abdomino-posterio sagittal anorectoplasty (PSARP), (c) division of fistula, coloplasty and APPT of coloplasty colon with proximal ileostomy, (d) division of fistula, coloplasty and sacro-APPT, (e) division of fistula, coloplasty/colorrhaphy and PSARP, and (f) pouch excision and APPT of ileum.
C. Colostomy/ileostomy closure: We performed two-staged procedures in 11 cases and the details are provided in "Results" section. The initial operative procedure is fecal diversion and the definitive procedure includes APPT without protective stoma. Two-staged procedures for CPC have certain advantages and are as follows: (a) It provides sufficient time for proper investigations and selection of the cases for two-staged and performing the definitive procedures without protective stoma. (b) As the stoma is excised during the definitive procedure in two-staged; therefore, stoma closure, its cost, hospital stay, and stoma-related complications are avoided. (c) Two-staged procedures can be performed for all types of CPC. (d) Definitive procedures are well tolerated at few months of age. (e) Complications observed following definitive procedures of two-staged are manageable with minor surgical procedures in the majority of the cases. (f) The only pre-requisite is the location of stoma in left lower quadrant of the abdomen, as the left pelvic incision is the preferred approach for definitive procedures for CPC. (g) Best way for planning the CPC patient for the future two-staged procedure is to create a stoma just proximal to pouch (2–3 cm proximal to pouch) and locate it at the left lower quadrant of the abdomen.

Disadvantages of two-staged procedures for CPC: (a) Definitive procedures are performed without protective stoma, so any major complications of pulled colon/ileum are difficult to manage. (b) Definitive procedures without protective stoma may not be a good option when performing a coloplasty/colorrhaphy for preserving the colon, as there is always a risk of suture line leak.

We strongly believe that doing definitive procedures for CPC at few months of the age, preferably after 4 months or weight of the child is 5 kg or more, is better than doing it on birth and also definitive procedures are well tolerated at few months of age than at birth. There is a need for more work on the definitive procedures without protective stoma for CPC to establish the procedures as an accepted technique.

Primary single-stage procedure for the management of all types of CPC in newborns has also been reported with better continence and cosmesis, with low morbidity and mortality.[4] At the present moment, the primary single-stage procedure for CPC is not advocated by most of the authors, as there is unacceptably high mortality associated with it (although it has certain advantages) and secondly definitive procedures at few months later have better outcome and are well tolerated.[1-3,5,6,23]

Complications are known to occur with CPC whether managed by staged procedures or the primary single-stage procedure.[1-5] These complications can be related to (A) complications of colostomy/ileostomy: peri-stomal skin excoriation, stomal bleeding, stoma stenosis, prolapse, recurrent urinary tract infections, fecaloma formation, diarrhoea, malnutrition, etc.,[1,2,4,5] (B) complications following colorrhaphy/coloplasty: suture line leaks, wound dehiscence, dilatation of colon, constipation, diarrhoea, etc.[1,2,5] and (C) complications following definitive pullthrough procedures: anal stenosis, stricture, mucosal prolapse, constipation, diarrhoea, perianal soiling, and incontinence.[1-4] Complications we observed in the cases managed with two-staged procedures are described in the "Results" section and also provided in Table 2.

The overall mortality of CPC continues to be very high and reported to occur in 10–45% of cases and was attributed to delayed presentation, poor general condition, and poor weight, presence of sepsis, types of CPC, associated lethal congenital anomalies, pouch perforation peritonitis, and complications following operative procedures.[1-4,23] Mortality following primary single-stage procedures for CPC is reported in 11% and attributed to other associated malformations.[4] Mortality following coloplasty has been reduced to about 5%, since it is being performed as a staged procedure.[5] There was no death following definitive procedures (two-staged) performed amongst eleven cases included in the study.

CONCLUSIONS

Two-staged procedures for all types of CPC can also be done safely with good results. The most important fact that the definitive procedure is done without protective stoma and therefore it avoids stoma closure, stoma-related complications, related cost of stoma closure and hospital stay. Best way to plan the case of CPC for the two-staged procedure is to create a stoma just proximal to pouch and create it at left lower quadrant of the abdomen.

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