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# Assessment of Nutritional Status of Patients of Congenital Pouch Colon Following Definitive Surgery

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## Abstract

### Purpose:

To assess the nutritional status in 31 patients of congenital pouch colon (CPC) who had undergone definitive surgery and closure of a protective stoma, if any, at least 1 year earlier and were below 14 years age.

### Materials and Methods:

The clinical history, demographic details, anthropometric measurements, and results of hematological and biochemical tests were recorded. In addition to collective data, analysis was also performed after grouping by age, subtype of CPC (Types I/II and Types III/IV CPC), and in Types I/II CPC patients, by whether the colonic pouch had been completely excised or else a segment preserved by tubular colorraphy (TC).

### Results:

Severe fecal incontinence (FI) was common (64.52%). Anthropometry showed a significant malnutrition in 53.85–95.45% patients, especially stunting which was most prevalent in the 0–5 years age-group. Serum Vitamin $B_{12}$, folate, and Vitamin D were lower than normal in 38.71%, 22.58%, and 74.19% patients, respectively, without statistically significant difference among the various groups studied. Patients with Types I/II CPC had a statistically significant higher incidence of anemia, low serum ferritin, and severe FI than patients with Types III/IV CPC. Patients with Types I/II CPC, managed by excision of the colonic pouch, had a higher incidence of severe FI, wasting, and thinness than those undergoing TC.

### Conclusions:

On follow-up of the patients of CPC, anthropometry shows a high incidence of malnutrition, especially stunting in the 0–5 years age-group. There is an adequate adaptation of fluid-electrolyte homeostasis. Although Types I/II CPC patients have a significantly higher incidence of anemia and severe FI than Types III/IV CPC patients, long-term anthropometric parameters are similar. In Types I/II CPC, preservation of the colonic pouch by TC offers long-term benefit.

**Key Words:** *Congenital pouch colon*, *malnutrition*, *mineral deficiency*, *tubular colorraphy*, *vitamin deficiency*

## Introduction

*C*ongenital pouch colon (CPC), an unusual abnormality in which a pouch-like dilatation of a shortened colon is associated with an anorectal malformation, has most frequently been reported from Northern India.[1,2,3,4,5,6]

CPC has been categorized into four subtypes based on the length of normal colon proximal to the colonic pouch.[1] In Type I CPC, normal colon is absent, and the ileum opens directly into the colonic pouch while in Type II CPC, a short segment of cecum is present between the ileum and the pouch. Type III CPC has a significant length of normal proximal colon (7–8 cm in the newborn) while with Type IV CPC, only the terminal portion of the colon (rectum and a varying length of sigmoid colon) is converted into the colonic pouch.[1]

It is evident that, following preliminary and definitive surgeries, patients with the more common and more severe forms of CPC (Types I/II CPC) are candidates for excessive fluid and electrolyte losses in the short term and in the long term for malnutrition and deficiencies of vitamins and other micronutrients and minerals. Although patients with Types III-IV CPC have longer normal colon, there is still a varying degree of the absence of normal colon.

There are few studies describing the long-term nutritional status and growth and development of patients of CPC.[2,4,7] There is no study at all in which a clinical and biochemical assessment for the presence of deficiencies of macronutrients and micronutrients has been performed. The present study attempts to provide a comprehensive nutritional assessment of patients of CPC in whom all surgical procedures have been completed.

## Materials and Methods

In this study, conducted from December 2012 to August 2014, the study population ($n$ = 31) consisted of patients of CPC who had undergone definitive surgery and closure of a protective stoma, if any, at least 1 year earlier and were below 14 years of age. Patients with significant malformations involving other major organ systems and those in whom the parents and/or patients refused consent for participation were excluded from the study.

The Institutional Ethical Committee clearance was obtained, and data were collected on a pretested proforma regarding the clinical and surgical history, demographic details including the socioeconomic (SE) status, dietary assessment including caloric value of the diet (adequate/inadequate for age) by the 24-h dietary recall method, anthropometric assessment, and the results of hematological and biochemical investigations. In addition to the collective data, the data were also analyzed after grouping the patients by age at assessment, subtype of CPC (Types I/II and Types III/IV CPC), and in patients of Type I and II CPC, by whether the colonic pouch had been completely excised or else a segment preserved by tubularization.

Clinical assessment Clinical examination was performed for physical signs of malnutrition such as pallor, hair changes like depigmentation, sparsity and alopecia, signs of Vitamin A deficiency, changes in the nails, skin changes such as dermatosis, desquamation or ulceration, bilateral pedal edema, and wasting. Assessment of fecal incontinence (FI) was performed using the Rintala and Lindahl score.[8] A continence score of 5 or less was graded as “severe” FI while a score between 6 and 9 was graded as “poor” fecal continence.[8]

Anthropometric assessments The standing height was measured using a stadiometer. The height reading was compared with the WHO reference standards 2006.[9,10] Body weight was taken and the reading compared with the WHO growth standards 2006.[9,10]

The body mass index (BMI) (weight in kg/height in meter$^2$) and the mid-upper arm circumference (MUAC) were also measured.

For the assessment of the nutritional status, percentiles and Z score were calculated for weight-for-height, weight-for-age, height-for-age, BMI for age, and MUAC for age using the WHO anthro- and anthro-plus software. On anthropometric assessment, the main indicators of protein-energy malnutrition (PEM) are “stunting” (low height-for-age) which signifies slowing in skeletal growth and is a marker of chronic undernutrition; “wasting” (low weight-for-height) which indicates a deficit in tissue and fat mass compared with that expected in a child of the same height and indicates acute undernutrition; and “underweight” (low weight-for-age) which is an indicator of both acute and chronic undernutritions.[11] MUAC also assesses the degree of “wasting” while the BMI assesses “thinness.”[11]

The *Z*-score was used to assess undernutrition. A Z-score cutoff point of $<-2$ standard deviation (SD) was used to classify low weight-for-age, low height-for-age, and low weight-for-height values as signifying moderate or severe undernutrition.[9,10] A BMI for age Z score of $<-2$ SD signified thinness.[9,10] As per the WHO growth parameters, all these parameters are applicable in children up to 5 years age. In the age-group of 5–10 years, all parameters are applicable apart from weight-for-height and MUAC. In children more than 10 years old, only height-for-age and BMI are applicable.[9,10]

Laboratory evaluations (hematology and biochemistry) Laboratory investigations included complete blood count and measurement of serum total proteins and albumin, lipid profile, blood urea, serum creatinine, serum electrolytes, serum calcium, ferritin, Vitamin $B_{12}$, folate, and blood glucose levels. The results were compared with the reference values.[12,13] Anemia was defined by the WHO's hemoglobin thresholds.[12,13]

The final results were tabulated and analyzed using standard statistical methods as per SPSS software (IBM Corporation, USA). The results were presented as mean ± SD for continuous variables and number (percentage) for categorical variables. Comparison between continuous variables was made using the Students' *t*-test. Categorical variables were compared using Chi-square test. The correlations between clinical, anthropometric, and laboratory evaluations were analyzed using appropriate tests. $P < 0.05$ was considered as statistically significant.

## Observations and Results

The age of the 31 patients ranged from 3 years to 14 years (mean 7.84 ± 4.10 years). There were 20 males (64.52%) and 11 females (35.48%), male:female ratio of 2:1.1. The subtypes of CPC were: type I CPC ($n$ = 2; 6.45%), Type II CPC ($n$ = 19; 61.29%), Type III CPC ($n$ = 3; 9.68%), and Type IV CPC ($n$ = 7; 22.58%). The definitive surgical procedures performed, according to the subtype of the CPC, are shown in Table 1.

According to Kuppuswamy's Socioeconomic Status Scale-2014,[14] a standard reference scale for the Indian population, 25 (80.65%) children belonged to lower or upper-lower SE status group while 6 (19.35%) children belonged to the lower-middle SE status group. All 31 children had a mixed diet including mainly green leafy vegetables, cereals, pulses, and milk. In 5 patients (16%), the diet included eggs or meat 1–3 times/week. On the assessment of the caloric and nutritive value of the diet,[15] it was categorized as inadequate in 28/31 patients (90.32%) and just adequate in only 3 patients (9.67%).

On clinical assessment, 23 children (74.19%) had pallor. No patient had evident physical signs of PEM or Vitamin A deficiency. Severe FI with continence score[8] of 5 or less, was present in 20/31 (64.52%) patients. These patients were passing small amounts of liquid or semisolid stools at a frequency of 8–20 times/day. The remaining 11 patients (25.81%) had a “poor” grade of fecal continence.[8] In 12 patients older than 5 years, a bowel management program consisting of oral loperamide once or twice daily and daily rectal washouts had been instituted. This resulted in reducing the frequency of soiling although significant improvement in continence scores did not occur.

The results of the anthropometric assessment of the study group are shown in Table 2, and the significant results of hematological and biochemical assessments are shown in Table 3.

**Assessment according to age grouping** The 31 patients were grouped into 3 age groups as applicable for anthropometric assessment by the WHO parameters.[9,10] In the age group 0–5 years ($n$ = 13; 41.94%), the mean age at assessment was 3.72 ± 0.60 years and the mean time duration from the last definitive surgical procedure was 1.54 ± 1.10 years while in the age group 5.1–10 years ($n$ = 9; 29.03%), these parameters were 8.50 ± 1.39 years and 6.1 ± 1.80 years, respectively. In the age group 10.1–14 years ($n$ = 9; 29.03%), these parameters were 13.11 ± 1.17 years and 9.2 ± 3.70 years, respectively.

On anthropometric assessment, all 13 children in the 0–5 years age group (100%) had stunting and the difference from the other age groups (44.4% in the 5.1–10 years age group and 66.7% in the 10.1–14 years age group) was statistically significant ($P$ = 0.011). Thinness (Low BMI for age) was more common in the older age groups (88.9% in the 5.1–10 years age group and 77.8% in the 10.1–14 years age group) as compared to the 0–5 years age group (5/9; 38.46%), the *P* value being 0.032.

#### Assessment according to subtypes of congenital pouch colon

The patients were grouped into two groups, Group A (Types I and II CPC; *n* = 21) and Group B (Types III and IV CPC; *n* = 10), based on whether or not there was a significant shortening of the length of the normal proximal colon. In both groups, the mean age at assessment and the mean time duration from the last surgery were comparable.

Clinical assessment showed that both severe FI (76.19% vs. 40%) and pallor (90.48% vs. 40%) were significantly more common in Group A patients than in Group B patients (*P* = 0.024 and 0.001, respectively). Significantly, on anthropometric assessment, the values of none of the 4 parameters compared showed any statistically significant difference. Anemia (66.7% vs. 40%), a low hematocrit (61.9% vs. 40%), and serum ferritin deficiency were more common in Group A patients than Group B patients, the differences in serum ferritin levels being statistically significant (*P* = 0.022). Vitamin $B_{12}$ deficiency was also more common in Group A patients (47.62% vs. 20.0%), a *P* = 0.070.

#### Assessment of Group A Patients (Types I/II Congenital pouch colon) according to surgical procedure performed

Broadly, patients from Group A underwent two types of definitive surgery. In Subgroup I (*n* = 9; 42.85%), the colonic pouch was excised and either a permanent end colostomy constructed (*n* = 1) or a pull through of normal ileum (*n* = 1) or of a short length of normal colon (*n* = 7) performed [Table 1]. Subgroup II consisted of 12 patients (57.15%) in whom tubular colorraphy (TC) with pull through was performed (*n* = 11) and 1 patient with a permanent end colostomy after TC.

In both subgroups, the mean age at assessment and the mean time duration from the last definitive surgery were comparable. Severe FI was present in all 8 patients from Subgroup I (100%) and in 6/11 patients from Subgroup II (54.55%), the difference being statistically significant (*P* = 0.030). Although thinness (88.9% vs. 41.7%; *P* = 0.037) and wasting were more common in Subgroup I patients, there was no statistically significant difference in the incidence of underweight children and stunting between the two subgroups. Serum Vitamin $B_{12}$ deficiency was more common in Subgroup I patients and Vitamin D deficiency in Subgroup II patients although, for both parameters, the number of positive cases in each subgroup was small and the difference in values was not statistically significant.

## Discussion

The colon absorbs water, sodium, and chloride, and secretes potassium, bicarbonate, and mucus.[16] It is also the site of digestion of certain complex carbohydrates and some proteins.[16,17] Thus, the absence of the colon or a significant reduction in its length, as in Types I/II CPC, can affect the nutritional status. It is also important that especially with Types I/II CPC, FI, often severe, is very common after definitive surgery.[5,7] The overall length of the ileum in Types I/II CPC may be short[5] and the ileocecal valve is absent in Type I CPC. The reduced intestinal "transit time" due to shortened bowel and an incompetent continence mechanism would have a deleterious effect on the absorption of fluids, electrolytes, and some nutrients. In some patients, as in 6 patients in our series, a preliminary or protective ileostomy, constructed during surgical management, results in defunctionalization of the terminal ileum, and colon for a varying period. This is significant as the terminal ileum is important for absorption of Vitamin $B_{12}$, bile salts, fat, and the fat-soluble vitamins. In addition, patients with CPC undergo multiple surgeries and as shown by a study of children subjected to ostomy (ileostomy/colostomy) procedures,[18] multiple surgeries and their complications, both early and late, can directly or indirectly influence nutritional status. All these problems are less severe with Types III-IV CPC.

In the present study, as expected in a government institution catering to the poorer sections of society, all 31 patients belonged to the lower SE strata[14] and had inadequate caloric and nutritive intake. Significantly, no patient showed evident clinical signs of major PEM, or of deficiencies of vitamins and micronutrients.

Although anthropometric assessment showed a high incidence of wasting, stunting, low MUAC, and of underweight children, it is relevant that the National Family Health Survey-3 (NFHS-3) (2005–2006) data[19] shows a very high incidence of significant malnutrition in Indian children with 48% of children <5 years age being stunted, 19.8% children having wasting, and 43% being underweight. These parameters are more frequently abnormal in children from families in the lowest wealth category and from the Northern Indian states.[19,20] However, our findings clearly indicate a higher than expected the incidence of both acute and chronic malnutrition, especially severe stunting, a specific marker for chronic malnutrition.

There are few detailed studies describing the long-term nutritional status and development of patients with CPC. The majority of reports[2,4,21] suggest that long-term growth and nutrition are not significantly affected. Wakhlu *et al*.[4] reported that 3–5 years after coloplasty (TC) and pull through, 8/10 patients had growth and weight gain within the $97^{th}$ percentile for their age. The stool frequency had diminished from 6 to 20/day 3 weeks after coloplasty to 3–5 per day 3 years later. Eleven patients with pouch resection and pull through of normal ileum or colon had growth and weight gain within the $97^{th}$ percentile.[4] Another study reported that 97/122 patients had good quality of life with normal growth and development after coloplasty and pull through.[21] Our findings of a high incidence of malnutrition on anthropometric assessment and persistence of severe FI, are at variance with these results[4,21] but are largely similar to those of an earlier, less detailed, study from our center which found that only patients with Types III/IV CPC or Types I/II CPC patients in whom pull through had been performed after TC had normal or near-normal growth patterns.[7] Another report also stated that patients with Types I/II CPC have severe recurrent diarrhea after pull through with poor weight gain and growth.[6]

The hematological assessment showed a high incidence of anemia (58.06%). However, the NFHS 3 data[19] showed the prevalence of anemia in 6–59-month-old children in India to be 69% and 69.9%, respectively in males and females, the incidence being more than 70% in the Northern states. The low Vitamin $B_{12}$ and folate levels in several patients (38.71% and 22.58% respectively) have also to be interpreted with caution as a high incidence of Vitamin $B_{12}$ and folate deficiency, especially Vitamin $B_{12}$ deficiency, has been reported in anemic children from Indian slums and in malnourished children.[22] Vitamin D levels were significantly low in 74.19% patients. However, a high incidence of Vitamin D deficiency has been reported in normal children.[23] Ritu and Gupta[24] found that Vitamin D deficiency has a prevalence of 70–100% in the general population all over the Indian subcontinent. It is also important that though serum Vitamin $B_{12}$, folate, and Vitamin D levels were low in a significant number of patients, there was no statistically significant difference among the various groups studied. Renal function tests and serum electrolyte levels were normal in all 31 patients, suggesting that in the long term, there is an adequate adaptation of the fluid-electrolyte homeostasis mechanism.

Stunting was the most common in the 0–5 years age-group (100%) and the difference from the other age groups was statistically significant. A possible explanation is that in underdeveloped countries including India, delayed but notable "catch-up" growth, especially skeletal growth, has been reported in chronically malnourished children as they get older.[25,26] In addition, most patients in the 2 older age groups were those in whom all operative procedures had been completed 5 years or more earlier, allowing a longer period of uninterrupted catch-up growth. Thinness was more common in the two older age groups, perhaps signifying continuing progression of relatively mild PEM in children in whom stunting had decreased with "catch-up" skeletal growth.[27]

Although patients with Types I/II CPC (Group A) had a statistically higher incidence of anemia, low serum ferritin and as expected, a higher incidence of severe FI than those with Types III/IV CPC (Group B), anthropometric assessment did not show significant differences between the two groups showing that in the medium-to-long-term, growth parameters are similar, irrespective of the length of normal colon present.

Patients with Types I/II CPC, managed by excision of the colonic pouch, had a higher incidence of severe FI ($P = 0.030$), wasting, and thinness than patients who had undergone TC. This suggests that preservation of a segment of the colonic pouch does provide long-term benefit to patients with the more severe Type I/II CPC, especially with regard to the degree of FI. At our center, before 2007, the colonic pouch was routinely excised during surgical management of Types I/II CPC. This was because the pouch lacks normal peristalsis, even after tubularization, and there is also a risk of marked redilatation of the tubularized colon.[5] However, the extremely poor functional results with this approach[7] led to the adoption of TC and pull through as the standard management for Types I/II CPC.

## Conclusion

It is evident that patients with CPC need monitoring and nutritional supplementation to manage PEM, vitamin, and micronutrient deficiency. Oral iron therapy and supplementation of Vitamin $B_{12}$ and folate may be beneficial, especially as the addition of cobalamin to iron and folic acid improves hemoglobin rise in nutritional anemia.[28] Early institution of high energy, good-quality protein intake is essential as catch-up growth is maximal before puberty and onset of epiphyseal closure.[29] In practice, sustained implementation of these measures may be difficult considering the poor SE status of our patients. Administration of constipating agents such as loperamide or diphenoxylate, especially to Type I/II CPC patients, may reduce fluid losses and improve fecal continence although marked improvement in continence scores may not occur. The increased intestinal transit time may also help in the absorption of nutrients.

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Conflicts of interest There are no conflicts of interest.

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# Figures and Tables

## Table 1

Definitive surgical procedure according to subtype of congenital pouch colon (*n*=31)

| Definitive surgery | Type I | Type II | Type III | Type IV | Total (*n*) | Percentage |
|---|---|---|---|---|---|---|
| Tubularization of colonic pouc[illegible] | [illegible] | [illegible] | [illegible] | [illegible] | [illegible] | [illegible] |

## Table 2

Anthropometric assessment of the study group (*n*=31)

| Anthropometric parameter | *n* | Percentage |
|---|---|---|
| Weight for height (wasting) (<5 years) | 7/13 | 53.85 |
| Weight for age (underweight) (<10 years) | 21/22 | 95.45 |
| [illegible] | [illegible] | [illegible] |

## Table 3

Significant results of hematological and biochemical assessment (*n*=31)

| Parameter | Number of patients with finding | Percentage of total patients |
|---|---|---|
| Hemoglobin (low) | 18 | 58.06 |
| Hematocrit (low) | 17 | 54.83 |
| Platelet count >350×10$^3$ cmm (high) | 8 | 25.80 |
| Total proteins <6.6 (g %) (low) | 9 | 29.03 |
| Albumin <3.5 (g %) (low) | 10 | 32.26 |
| Triglycerides<50 (mg %) (low) | 7 | 22.58 |
| Ferritin <23.9 (ng/ml) (low) | 25 | 80.65 |
| Vitamin $B_{12}$ <180 (pg/ml) (low) | 12 | 38.71 |
| Folate <5.9 (ng/ml) (low) | 7 | 22.58 |
| Vitamin D <75 (nmol/l) (low) | 23 | 74.19 |