

## PULL-THROUGH PROCEDURE FOR HIRSCHSPRUNG'S DISEASE: TRANSANAL OR TRANSABDOMINAL? A COMPARATIVE STUDY.

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**Aim:** To document our experience with the transanal pull-through procedure, to compare its results with the results of the one-stage open Soave procedure and to compare results one-stage procedures with the gold standard multi-stage.

**Methods:** The prospective part of the study included twenty-eight patients with biopsy-proven HD. The retrospective part of the study included 277 patients treated by Soave multistage procedure. Patients were randomized into; Group A: treated by the transanal pull-through procedure. Group B: treated by the trans-abdominal one-stage Soave pull-through procedure. Group C: treated by Soave multistage procedure.

**Results:** The operative bleeding, the operating time, the onset of oral feeding, the postoperative pain, the hospital stay, the length of follow-up and costs were the only statistically significant variables. Functional results were good in 92.86%, 85.71%, and 88.09% of patients of groups A, B and C respectively. Postoperative complications were seen mainly in groups C then B. For groups A and B, there had been no recurrence of obstructive symptoms. Hospital stay was significantly longer in the groups C and B. Cost was significantly higher in the group C than group B and group A.

**Conclusions:** In selected cases, one stage pull-through operation can be safely done. Transanal technique is superior to open Soave due to its simplicity, cost effectiveness, and less surgical morbidity. It can be tolerated very well in the newborns. The operative technique can be easily mastered. It could be carried out in older children with little morbidity. Long-term follow-up will be required to determine whether early total reconstruction produces better lifelong bowel function than traditional staged repairs.

**Key words:** Hirschsprung's Disease, Pull-through.

### INTRODUCTION

Hirschsprung's disease (HD) is a common cause of bowel obstruction. The time-honored approach to therapy was to perform a preliminary colostomy in normally innervated bowel and subsequent definitive pull-through procedure later. Performing defunctioning colostomy as a first step was because of the unsafely of doing the pull-through procedure in the neonatal period. Endorectal pull-through was described in 1964 by Soave.<sup>(1)</sup> In the same year, the technique was modified by Boley who performed the coloanal anastomosis during the pull-through.<sup>(2)</sup>

The last decade has seen an evolution in the surgical management of HD. The previous gold standard of two- or three-stage pull-through with a preliminary stoma has been replaced by a one-stage approach in many centers.<sup>(3-5)</sup>

Recently, minimally invasive approaches to the one-stage pull-through have become popular. These have consisted of pull-through utilizing laparoscopic abdominal and pelvic mobilization of the rectum<sup>(6-8)</sup> and the transanal procedure, which does not include any intra-abdominal dissection.<sup>(9,10)</sup> This transanal new endorectal technique involves rectal mucosectomy, aganglionic segment colectomy, and normoganglionic colon pull-through that are performed through the anus. No laparotomy or laparoscopy is required in this technique. Few reports are available concerning this technique in the literature. Meager reports are available that compares the transanal approach with the open Soave technique.<sup>(11)</sup> Moreover, no reports are available that discuss the comparison between transanal and transabdominal approaches to answer the following questions:<sup>(1)</sup> Are there any differences in the

short-term results?<sup>(2)</sup> Are there any differences in cost?<sup>(3)</sup> Is routine laparotomy necessary?

The design of the current study was to document our experience with the transanal pull-through procedure, compare its results with the results of the one-stage open Soave procedure and to compare results one-stage procedures with the gold standard multi-stage procedure by which results of any one-stage procedure for HD must be assessed.

## PATIENTS AND METHODS

To achieve inclusive appraisal of pull-through Soave approaches to HD, a combined prospective and retrospective study was designed.

The prospective part of the study included twenty-eight patients with biopsy-proven HD, selected out of fifty-seven cases, admitted in a period between November 2000 and December 2003 to Alexandria Main University Hospital. Patients excluded from the study had; a total colon disease, a previous colostomy, a long aganglionic segment exceeding the splenic flexure, a hugely dilated colon, a complicated HD, an ultrashort-segment HD or a non pathognomonic barium enema with poor discrepancy between narrow and dilated segments. Patients were randomized by the closed-envelop technique into two groups; Group A: included fourteen patients treated by the transanal pull-through procedure.<sup>(9)</sup> Group B: included fourteen patients treated by the trans-abdominal one-stage Soave pull-through procedure.<sup>(12)</sup> An informed consent was obtained from each patient. The study protocol was registered and approved by the committee of Postgraduate Studies and Medical Research, Faculty of Medicine, University of Alexandria. These patients were subjected to:

1. *Thorough history taking and clinical examination:* with special emphasis on delayed passage of meconium, lax abdominal distension, with or without bilious vomiting.
2. *Radiological examination:* plain abdominal X-ray, barium enema, and in some cases MRI to corroborate the diagnosis and define the site of the transition zone. (Fig.1,2)
3. *Biopsy and histopathological examination:* Noblett's suction or full thickness rectal biopsy was taken 2 cm above the dentate line.
4. *Preoperative preparation:* One day before surgery, 25 ml/kg of polyethylene glycol with electrolyte solution was administered every hour for 4 hours. However, if colonic cleanliness is achieved mechanically with our regimen of mandatory saline irrigations, no other mechanical preparation was needed. The newborn was kept on clear fluids for 48 hours before surgery. Intravenous third-generation cephalosporin and metronidazole were

administered. After surgery, antibiotics were continued for the first 72 hours.

5. *Operation:* transanal pull-through procedure<sup>(9)</sup> was carried out for patients of group A while the trans-abdominal one-stage Soave pull-through procedure<sup>(12)</sup> was carried out for patients of group B. (Fig. 3,4)

6. *Postoperative and follow up:* Approximately 10 days after surgery, the anorectum was dilated to a # 10 to 12 Hegar dilator. The mother's fifth finger was substituted either immediately or 1 week later. These daily dilations were discontinued when the anastomotic ring can no longer be felt. These dilations were carefully monitored and were usually required for 2 to 3 months after the surgery.

Operative details, duration (including anesthesia), bleeding (expressed as percent of the blood volume), blood transfusion, length of resected segment, postoperative recovery, immediate postoperative mishaps, duration of nasogastric tube insertion, onset of oral feeds, regaining of bowel motions, duration of hospital stay, postoperative complications, and functional results were analyzed in all patients. Postoperative functional results were considered good when the patient spontaneously evacuated soft stools at least once daily without enemas, laxatives, or changes on diet. A mean follow-up period of  $1.72 \pm 1.2$  years was applied to all cases, during which, full clinical assessment as regard general well being, weight gain, frequency of bowel motions, attacks of constipation and/or diarrhea, as well as the local wound condition (in group B). A follow up barium enema study was carried out for all cases of both groups.

The retrospective part of the study included 277 patients (Group C) with biopsy-proven HD who were treated by Soave multistage procedure<sup>(3,4)</sup>, selected out of 398 cases, admitted in a period between March 1978 and October 2000 to Alexandria Main University Hospital. The study did not include patients treated by one-stage procedure or other procedures except Soave multistage procedure. Patients with incomplete data or those lost for follow up were excluded.

### *Statistical Analysis:*

The Statistical Package for Social Sciences (SPSS) was utilized for statistical analysis and tabulation. Mean, standard deviation, and median were calculated. The three groups were compared by 1-way analysis of variance (ANOVA) using the Kruskal-Wallis test. The level of significance selected for this study was P less than or equal to 0.05..

## RESULTS

The comparison of the data of the three groups showed that the operative bleeding, the operating time, the onset of

oral feeding, the postoperative pain, the hospital stay, the length of follow-up and costs were the only statistically significant variables in this study Table 1. There were no statistically significant differences among groups with respect to gender, age, at the time of the pull-through, family history, length of the aganglionic segment, intra-operative complications or level of the transition zone.

Intra-operative complications were limited to a urethral mucosal injury from the Foley catheter in one child undergoing an open multistage Soave procedure. This was managed conservatively with an indwelling catheter for 48 hours postoperatively and was not associated with any sequelae.

The mean operative time did not include the time for frozen sections. The dissection required freeing the stoma and the abdominal adhesions in patients of group C, and the opening and closure of the laparotomy and the arduous mucosectomy in older and previously complicated patients in group C were the causes of the extended operative time and the increased operative bleeding in this group.

All patients of group A had ganglion cells confirmed by permanent section analysis of the proximal margin. Intra-operative blood loss was minimal in all. Acetaminophen was administered for postoperative analgesia; no child was given narcotics postoperatively. All patients started postoperative bowel function within the first 24 hours, and oral feeding was resumed the next 24 to 36 hours. During the first 2 weeks, bowel function was normal in all of them with one to three bowel movements per day. There were no intra-operative complications. Infection or anastomotic leak was developed in none of the patients. Mean follow-up had been secured for 1.7 years. Bowel movements range from one to three per day. No patient had soiled his diapers between bowel movements. Only one case developed constipation. No enterocolitis or intestinal obstruction had been observed. However, anal stricture had developed in two cases. One patient was submitted to rectal dilatations for 3 months; rectal dilatation was not performed on one patient because the parents did not grant permission.

Functional results were good in 92.86%, 85.71%, and 88.09% of patients of groups A, B and C respectively. Majority of patients began to pass stool usually 2-5 days postoperatively. Patients who remained constipated were

all controlled with high-fiber diet and an oral laxative. Eleven patients had minimal fecal soiling that was secondary to diarrhea in seven cases and was controlled with low-residue diet and loperamide. All patients of the study had normal sphincter tone. Not all of these patients, however, were necessarily continent. Patients of group C could only be assessed for continence, as they achieved normal bowel function between 3 to 4 years of age. Patients of the remaining groups could not have their continence assessed at time of the study as most of them were still in diapers. The continence rate for group C would be 94.95% (14 of 277).

Follow-up in some of patients of group C was as long as 21 years. This represented a very long-term follow up after multistage Soave pull-through procedure. All cases of groups A and B underwent postoperative barium enema after three months to demonstrate the area of descending colon and new rectum. In all cases, there was no constricting ring, stricture, or discrepancy.

Postoperative complications were seen mainly in groups C then B. 7 patients in group C presented with intestinal obstruction secondary to adhesions 6 months after the pull-through. We had not seen postoperative enterocolitis in groups A and B and attributed this in part to early and adequate dilatations. The seromuscular cuff was not drained in the entire study, and there were no sequelae. Thirteen patients of group C had enterocolitis (defined as fever and distension) that required antibiotics, rectal decompression and dilatations for 6 months. The four patients who had universal Hirschsprung's disease suffered from bouts of diarrhea. Three of them developed mucosal rectal prolapse while two of them suffered from anal stenosis. For groups A and B, there had been no recurrence of obstructive symptoms, and no patient had had to have a colostomy. Repeat surgery was necessary for eight patients of group C who had recurrent aganglionosis (missed segment) and one child of group C who had an anastomotic leak. None of the patients in the groups A or B required an additional operation. It should be mentioned that the perianal rash, seen almost universally after any Soave pull-through for Hirschsprung's disease, had been of much shorter duration and less severe in groups A and B patients who had not had a preliminary colostomy.

Hospital stay was significantly longer in the groups C and B ( $P < 0.05$ ).

**Table 1. Comparison of patients with HD operated by the three different approaches.**

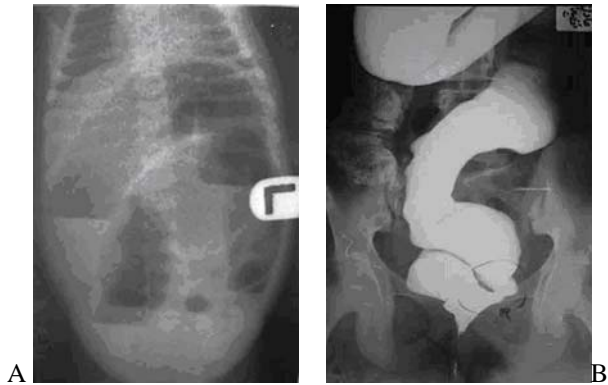
Parameter	Group A n = 14	Group B n = 14	Group C n = 277	ANOVA P value
Gender (M:F)	11:3	12:2	223:54	0.12
Age at pull-through (month)	2.5 ± 0.61	3.3 ± 1.4	15.1 ± 4.9	0.36
Family history	2 (14.3%)	1 (7.14%)	29 (10.47%)	0.71
Level of transition zone:				
Rectum	6 (42.86%)	5 (35.7%)	112 (40.43%)	
Rectosigmoid	4 (28.57%)	5 (35.7%)	89 (32.13%)	
Sigmoid	3 (21.43%)	4 (28.57%)	53 (19.13%)	
Descending colon	1 (7.14%)	0	13 (4.69%)	
Transverse colon	0	0	6 (2.17%)	
Total colon	0	0	4 (1.44%)	
Length of resected segment (cm)	24.2 ± 9.3	32.1 ± 5.7	43.2 ± 18.4	0.14
Proximal margin ganglion cells	14/14	14/14	269/277 (97.1%)	0.25
Operative blood loss (ml)	12.4 ± 7.1	20.3 ± 5.8	26.1 ± 13.9	0.02*†‡
Intra-operative complications	0	0	1 (0.36%)	
Operative time (min)	43.5 ± 2.2	91.3 ± 6.7	122.1 ± 9.4	0.03*†‡
Nasogastric tube duration (day)	1.2 ± 0.1	3.2 ± 1.7	3.6 ± 0.9	0.04*†‡
Onset of oral feeding (day)	1.5 ± 0.04	3.3 ± 1.4	4.5 ± 0.7	0.03*†‡
Onset of passing stools (day)	1.3 ± 0.5	3.2 ± 1.2	3.8 ± 1.1	0.04*†‡
Postoperative pain (day)	3.7 ± 0.2	6.4 ± 1.1	7.5 ± 2.3	0.03*†‡
Need for narcotics (%)	0	21.43	29.24	0.02*†‡
Hospital stay (day)	2.6 ± 1.3	8.1 ± 1.5	9.4 ± 6.3	0.02*†‡
Functional Results				
Good (once / day without help)	13 (92.86%)	12 (85.71%)	244 (88.1%)	
Constipation	1 (7.14%)	2 (14.3%)	19 (6.86%)	
Incontinence	0	0	14 (5.05%)	
Fecal soiling	0	0	11 (3.91%)	
Bouts of diarrhea	0	0	7 (2.53%)	
Postoperative complications				
Wound infection	0#	3 (21.43%)	28 (10.11%)	
Abdominal pain and distention	0#	2 (14.3%)	17 (6.14%)	
Rectal prolapse	0	0	4 (1.44%)	
Enterocolitis	0	0	13 (4.69%)	
Stricture or cuff narrowing	2 (14.3%)	4 (28.57%)	22 (7.94%)	
Adhesive obstruction	0	0	7 (2.53%)	
Perianal rash or excoriation	4 (28.57%)	7 (50%)	136 (49.1%)	
Anastomotic leak	0	0	1 (0.36%)	
Prolapse of colostomy	0	0	34 (12.27%)	
Recurrence (missed segment)	0	0	8 (2.89%)	
Follow up (year)	1.72 ± 1.2§	1.72 ± 1.2∞	13.4 ± 8.5	0.0001*†§

\* Significant difference between groups.

† Significant difference between A & B.

‡ Significant difference between A & C.

§ Significant difference between B & C.



**Fig 1. HD A:** a plain x-ray abdomen showing marked gaseous distension.  
**B, C & D:** Barium enema showing the site of transition zone.



**Fig 2. HD A:** Sagittal cut by MRI showing the level of transition zone.  
**B, C & D:** Coronal cuts by MRI showing the level of transition zone.



**Fig 3. Open Soave procedure;** showing the transition zone.



**Fig 4. The transanal endorectal pull-through procedure (TERPT) & the excised specimen.**

## DISCUSSION

The conventional multi-stage repair (colostomy followed by pull-through procedure) is the gold standard by which results of any procedure for HD must be assessed.<sup>(10)</sup> Currently, the one-stage Swenson, Duhamel, and Soave pull-through procedures have been accomplished by laparotomy<sup>(12)</sup> or laparoscopy.<sup>(13)</sup> The results are as good or better than those classically completed in 2 or 3 stages. The one-stage procedures, moreover, avoid the complications of colostomy and the costs of staged therapy.<sup>(12)</sup> The one-stage Soave pull-through is one of the more commonly used procedures. Trying to collect the material for this study, we were only able to carry it out in one third of cases because most patients with HD seen for the first time at our hospital had a colostomy, presented with complications, or were diagnosed too late to allow a one-stage pull-through. The variations of primary pediatric care in our country have given us the opportunity to compare these three forms of endorectal pull-through.

The TERPT approach had the advantages of a significantly shorter hospital stay, less need for narcotic analgesics, and lower cost when compared with the open approach. It eliminated the time taken to open and close the laparotomy and to perform the colectomy followed by the pull-through during open procedures. In this technique, neither abdominal nor intraperitoneal bowel opening was necessary; therefore, the risk of adhesion formation presumably decreased. In addition, it offered the best cosmetic results in the surgical correction of Hirschsprung's disease. Likewise, the endorectal dissection preserved the anorectal sphincters as well as the local blood supply and innervation and so fecal and urinary continence were not affected. It also helped avoid damaging pelvic structures such as the ductus deferens. In the current study, all children tolerated feedings and began passing stool the evening of surgery. This rapid return of bowel function might be attributed to two factors: First, the mucosal incision was above the dentate line obviating the administration of intra-operative or postoperative narcotics. Second, the procedure did not require manipulation of the intra-abdominal viscera.

Because the TERPT approach offered a smaller operative field, a potential hazard with this technique could be uncontrolled bleeding when the colon is being freed and mobilized. When colorectal vessels were being dissected and tied to free the colon, extreme care were exercised to obtain hemostasis. Likewise, the mobilization of the colon through the anus required retraction to maintain visibility. Because the sphincters were stretched, the functional results on them were not predictable at this time. In spite of this, all patients had normal bowel movements and no one presented soiling between bowel movements. However, these were preliminary data and our patients would be submitted to manometry for a complete evaluation (external

and internal sphincters) at the age of three years or as soon as their cooperation could be obtained.

The transitional zone level seen in the preoperative barium enema could be located higher during the operation. There had been good correlation of the observed transitional zone level between the barium enema and the surgical findings. There were two risks to this approach. The first was that the adequacy of the resection was solely dependent on accurate identification of the transition zone pathologically using frozen section analysis. The second was that total colonic involvement might be present. In addition, we think that TERPT would become laborious with more bleeding in older patients, those diverted, patients with previous enterocolitis, or patients with deep rectal biopsies had been taken.

None of the patients undergoing the TERPT approach had adhesive bowel obstruction. In contrast, the incidence of adhesive small bowel obstruction after open pull-through for Hirschsprung's disease had ranged from 0% to 2.53%, with most studies reporting between 5% and 10%.<sup>(11,12)</sup> Avoidance of this significant and often late complication represents another potential advantage of the TERPT approach. Lack of stricture in this study was because of early rectal dilatation until the anastomotic ring could not be detected. Postoperative enterocolitis was not encountered due to early diagnosis, preoperative colonic irrigations, antibiotics, early surgical correction of the Hirschsprung's disease, and early, adequate, and monitored postoperative anorectal dilatations. Our data suggested that: 1. There was no benefit to routine laparoscopic visualization, selective use of laparoscopy or minilaparotomy for those children at high risk for longer segment disease. 2. TERPT provided the same benefits without any increase in cost or operating time. 3. The avoidance of the intraperitoneal laparoscopic dissection reduces the potential risk to pelvic structures. 4. TERPT permits those surgeons who are inexperienced with advanced laparoscopic techniques to perform the operation. In addition, we had found that a pure transanal approach without laparoscopy was feasible because<sup>(1)</sup> a relatively long length of mucosa-submucosa could be dissected easily, obviating the need for laparoscopic or open dissection and<sup>(2)</sup> One cannot be able to distinguish easily the transition zone by laparoscopic visualization because of the preoperative colonic irrigation and decompression.

TERPT can be tolerated very well in the newborns. It could be carried out in older children with little morbidity. The operative technique can be easily mastered. Preliminary colostomy in the newborn with Hirschsprung's disease will be rarely indicated. Fecal continence is expected in the majority of these patients. Because approximately 75% of HD in children is limited to the rectosigmoid region, a majority may be treated using TERPT strategy. TERPT

procedure is suitable for classical uncomplicated rectosigmoid aganglionosis and those not exceeding the splenic flexure or having markedly dilated colon, because it is effective and promising. In patients with aganglionosis exceeding the splenic flexure, a complementary laparoscopy could be performed to release the colon. Laparoscopy with biopsy before TERPT may have a role in female gender with a strong family history that raise the suspicion of long segment disease. Without a radiological transition zone, a laparotomy with serial biopsies would be the ideal. Long-term follow-up is required to determine whether early total reconstruction produces better lifelong bowel function than traditional staged repairs.

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