



## The Surgical Correction by Endo Rectal Pull through of Hirschsprung's Disease: Evaluation of Late Outcome

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**Abstract: Background:** Harold Hirschsprung, a Danish pediatrician, is attributed to the first definitive description of the disease of his name. In the next 30 years, it has been reported that incomprehensible actions aimed at reducing sympathetic hyperactivity, at least temporarily, were successful in the treatment of HD. Complications can be classified either early (weeks to months) or late (months to years). Transanal Endorectal pull-through approach represents an important revolution in the treatment of Hirschsprung disease. **Objectives:** The aim of this study is to evaluate the post-operative late complications of Transanal Endorectal pull-through operation, clinically in-patient with Hirschsprung disease, we will focus on the fecal incontinence using Krickenbeck score. **Patients and Methods:** This retrospective cohort study was conducted on 20 children with Hirschsprung disease operated beyond during last 7 years in whom Transanal Endorectal pull through was indicated. For Patients with histopathologically documented HD underwent pull-through in the last seven years. Age > 4years (to be able to evaluate faecal continence). In addition, Patients were operated Trans anal pull through Soave and Swenson. **Results:** Our results showed that; as regard to enterocolitis was present in 16 patients (80%) and absent in 4 patients (20%). Patients with recurrent attacks of one to three times of enterocolitis was 12 patients (60%), patients with four to six times was 3 patients (15%) and patients with more than six times was 1 patient (5%). According to types of operation Transanal soave was done in patients (75%) and Trans anal Swenson was done in five patients (25 %). As regarding Voluntary bowel movements was, absent in 2 patients (10%) and present in 18 patients (90%). As regarding Soiling was, absent in 5 patients (25%) and present in 15 patients (75%) and divided in two grads, grade one in 12 patients (60%) and grade two in 3 patients (15%). As regarding Constipation was, absent in 4 patients (20%) and present in 16 patients (60%) and divided in two grads, grade one in 10 patients (50%) and grade two in 6 patients (30%).

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**Keywords:** Transanal Endorectal Pull-through Hirschsprung; Anal canal; abdominal distension

### 1. Introduction

Hirschsprung's disease (HD) is a common cause of neonatal intestinal obstruction that is of great interest to pediatric surgeons throughout the world. Prior reports concerning the historical origins ascribe the initial description of this condition to Fredericus Ruysch, a Dutch anatomist in Amsterdam in 1691. He described a 5-year-old girl with abdominal pain who did not respond to the usual treatment of the day to relieve pain, she eventually died (*Baltogiannis et al., 2003*).

HD is a relatively common neonatal developmental disorder of the enteric nervous system. It is characterized by the absence of ganglion cells in the myenteric and submucosal plexuses of the distal intestine. This results in absence of peristalsis in the affected bowel segment, and the development of a

functional intestinal obstruction. The pathogenesis and genetic basis of the disease is yet unclear (*Temple et al., 2012*).

The surgical management of HD has developed, from full-thickness rectosigmoid dissection (Swenson and Bill), endorectal dissection (Soave) and retrorectal pouch procedure (Duhamel), to a more recent primary repair that can be done transanally (*Sookpotarom, 2009*).

Langer et al. described the one-stage transanal pullthrough operation for HD. Since then, the procedure has become increasingly popular and compared with the traditional open laparotomy, the advantages of the operation are that it is minimally invasive; it eliminates the abdominal incision, so there is no abdominal scar, and it avoids complications of traditional laparotomy such as intestinal adhesion and

wound infection postoperatively. The operating time and hospital stay are also shortened (*Langer et al., 2007*).

The early complications include wound infection, bleeding and anastomatic complications as (leak, pelvic abscess, retraction of pull-through segment (*Teitelbaum et al., 2008*).

The late complications are usually present within the first few postoperative months. They include: enterocolitis, stricture and constipation (*Pastor et al., 2009*).

One potential problem in the transanal endorectal pull-through (TERPT) procedure is the greater amount of traction on the anal sphincters to perform the endorectal dissection and anastomosis. Such manipulation of the anal canal has been associated with significant problems in adult patients. Interestingly, in a previously published study from our institution in 2002, short-term manometric findings and stooling patterns showed no differences between the ABD and TERPT approaches. Specifically, no significant difference was noted when comparing postoperative anal resting pressure and loss or absence of the anorectal inhibitory reflex (*El-Sawaf et al., 2009*).

Distinctions between occasional soiling and significant incontinence are difficult. Although uncommon, the surgeon should consider the possibility of retained aganglionosis. Additional workup may include barium enema, manometer and rectal biopsy. MRI of the pelvis and lower back can be useful in selected patients. Most children will achieve satisfactory continence with time. Occasional soiling appears to improve over time (*Engum & Grosfeld, 2004*).

The anorectal function are greatly affected after the surgical treatment of the HD. It can be assessed clinically by interviews and questionnaires to the patients and the parents. It also can be assessed by investigations as endorectal ultrasound and anorectal manometry, Colonic transit time and defecography (*Dasgupta and Langer, 2008*).

#### **Aim of the Work**

To highlight the post-operative late complications of the patients who had been diagnosed and operated as Hirschsprung disease including enterocolitis and faecal incontinence. In addition, we will focus on the fecal incontinence using Krikenbeck score.

## **2. Patients and Methods**

Type of Study: Retrospective Thesis Study  
Setting: one group of children with HD operated at Benha children hospital with endorectal pull-through.

Study Period: last seven years .

Study Population.

#### **Inclusion Criteria:**

- Patients with histopathologically documented HD underwent pull-through in the last seven years.
- Age > 4years (to be able to evaluate fecal continence).
- Patients were operated trans anal pull through Soave and Swenson.

#### **Exclusion Criteria:**

- Previous colorectal surgery for HD other than rectal biopsy.
- Patients were excluded if they were younger than 3 years (before the age of toilet training).
- Patients had total colonic aganglionosis, had a colostomy at the time of evaluation, or required a reoperation.

Sampling Method Benha children hospital files records.

#### **Sample Size 20 patients**

**Ethical Considerations:** Approved by ethical committee of Ain-Shams university

We revised the data of the patients who had been operated HD at the last seven years from the medical records of Benha children hospital. Data such as demographic data, preoperative assessment and the operative details.

We collected the telephone numbers of the patients from patients files and contacted them to arrange an appointment and if the patient have an apology. The history was taken and evaluates the patient by telephone.

We used Krikenbeck score questionnaire for evaluation voluntary movement, soling and constipation single sheet for.

## **3. Results**

Analysis was performed using student T test for numerical data and chi squar and fischer exact test for categorical data (p value) was considered significant if it is less than 0.05 and non-significant if it is more than 0.05.

The table (1) showing the distribution of study sample according to gender and age variables, the total No. of Female was 5 which represent 25% of study sample and male was 15 which represent 75% of study sample. we classifying the age variable into two categories Age at time of evaluation in months with Mean of 60.55 and SD of 17.35. and Age at time of operation in months with median of 6 and range from 3 to 48. The Interval time between operation and evaluation (months) mean was 52.5 with SD of 13.26.

The Table (2) shown that enterocolitis was present in 16 patients (80%) and absent in 4 patients (20%). Patients with recurrent attacks of one to three times of enterocolitis was 12 patients (60%), patients

with four to six times was 3 patients (15%) and patients with more than six times was 1 patient (5%).

**Table No (1): Demographic Distribution of study sample**

		<b>Total no. = 20</b>
Gender	Female	5 (25.0%)
	Male	15 (75.0%)
Age at time of evaluation in months	Mean $\pm$ SD	60.55 $\pm$ 17.35
	Range	44 – 108
Age at time of operation in months	Median (IQR)	6 (5 – 6)
	Range	3 – 48
Interval time between operation and evaluation (months)	Mean $\pm$ SD	52.2 $\pm$ 13.26
	Range	34 – 90

**Table No (2): Recurrent attacks of enterocolitis**

<b>Recurrent attacks of enterocolitis</b>	<b>No</b>	<b>%</b>
None	4	20
1-3 attacks	12	60
4-6 attacks	3	15
More than 6 attacks	1	5

As regarding to types of operation Transanal soave was done in patients (75 %) and Trans anal Swenson was done in 5 patients (25 %).

**Table No (3): Types of Operation**

		<b>Total no. = 20</b>
<b>Etiology</b>	<b>HSD</b>	20 (100.0%)
Biopsy diagnosis	Aganglionic rectal biopsy	20 (100.0%)
Operative level	Recto-sigmoid	20 (100.0%)
Type of operation	Transanal Soave	15 (75.0%)
	Transanal swenson	5 (25.0%)
Number of enterocolitis attacks	Median (IQR)	2 (1 – 2.5)
	Range	0 – 7
Hospital admission	Median (IQR)	1 (0 – 2)
	Range	0 – 7
Home treatment	Median (IQR)	0 (0 – 1.5)
	Range	0 – 4

As regarding **Voluntary bowel movements** was, absent in 2 patients (10%) and present in 18 patients (90%).

**Table No (4): Contrast level**

		<b>Total no. = 20</b>
<b>Contrast level</b>	<b>Recto-sigmoid</b>	20 (100.0%)
Voluntary bowel movements	No	2 (10.0%)
	Yes	18 (90.0%)
Soiling	No	5 (25.0%)
	Grade 1	12 (60.0%)
	Grade 2	3 (15.0%)
Constipation	No	4 (20.0%)
	Grade 1	10 (50.0%)
	Grade 2	6 (30.0%)

As regarding to the age there is **no statistical significant** difference between soiling groups and the age variable as (p value) = 0.441.

As regarding to the Gender there is **no statistical significant** difference between soiling groups and the age variable as (p value) = 0.136.

As regarding to Interval time between operation and evaluation (months) there is **no statistical significant** difference between soiling groups and the age variable as (p value) = 0.653.

**Table No (5): P- test for statistical differences between age and gender variables and soiling groups**

		Soiling groups		Test value	P-value	Sig
		No	Yes			
		No. = 5	No. = 15			
Gender	Female	0 (0.0%)	5 (33.3%)	2.222*	0.136	NS
	Male	5 (100.0%)	10 (66.7%)			
Age at time of evaluation in months	Mean $\pm$ SD	55.20 $\pm$ 11.56	62.33 $\pm$ 18.88	-0.788•	0.441	NS
	Range	44 – 72	46 – 108			
Age at time of operation in months	Median (IQR)	5 (5 – 6)	6 (6 – 6)	-1.075 $\neq$	0.282	NS
	Range	4 – 7	3 – 48			
Interval time between operation and evaluation (months)	Mean $\pm$ SD	49.80 $\pm$ 11.01	53.00 $\pm$ 14.19	-0.457•	0.653	NS
	Range	40 – 66	34 – 90			

P-value > 0.05: Non significant; P-value < 0.05: Significant; P-value < 0.01: Highly significant

\*: Chi-square test; •: Independent t-test;  $\neq$ : Mann-Whitney test

As regarding to the type of operation there is **no statistical significant** difference between soiling groups and the Type of operation variable as (p value) = 0.799.

As regarding to the Number of enterocolitis attacks there is **no statistical significant** difference between soiling groups and the Number of enterocolitis attacks variable as (p value) = 0.964.

As regarding to the Numbers of Hospital admission there is **no statistical significant** difference between soiling groups and the Hospital admission No variable as (p value) = 0.964.

As regarding to Home treatment there is **no statistical significant** difference between soiling groups and the Home treatment variable as (p value) = 0.106.

**Table No (6): P- test for statistical differences between variables of (Type of operation, Number of enterocolitis attacks, Hospital admission No and Home treatment) with soiling groups**

		Soiling groups		Test value	P-value	Sig.
		No	Yes			
		No. = 5	No. = 15			
Type of operation	Transanal Soave	4 (80.0%)	11 (73.3%)	0.089*	0.799	NS
	Transanal swenson	1 (20.0%)	4 (26.7%)			
Number of enterocolitis attacks	Median (IQR)	2 (1 – 2)	2 (1 – 3)	-0.045 $\neq$	0.964	NS
	Range	0 – 7	0 – 6			
Hospital admission No	Median (IQR)	1 (0 – 2)	1 (0 – 2)	-0.046 $\neq$	0.964	NS
	Range	0 – 7	0 – 3			
Home treatment	Median (IQR)	0 (0 – 0)	0 (0 – 2)	-1.616 $\neq$	0.106	NS
	Range	0 – 0	0 – 4			

P-value > 0.05: Non significant; P-value < 0.05: Significant; P-value < 0.01: Highly significant

\*: Chi-square test; •: Independent t-test;  $\neq$ : Mann-Whitney test

As regarding to the gender there is **no statistical significant** difference between constipation groups and the age variable as (p value) = 1.00.

As regarding to the age there is **no statistical significant** difference between constipation groups and the age variable as (p value) = 0.517.

As regarding to Interval time between operation and evaluation (months) there is **no statistical significant** difference between constipation groups and the age variable as (p value) = 0.721.

**Table No (7): P- test for statistical differences between age and gender variables with Constipation**

		Constipation groups		Test value	P-value	Sig.
		No	Yes			
		No. = 5	No. = 15			
Gender	Female	1 (25.0%)	4 (25.0%)	0.000*	1.000	NS
	Male	3 (75.0%)	12 (75.0%)			
Age at time of evaluation in months	Mean $\pm$ SD	65.75 $\pm$ 28.78	59.25 $\pm$ 14.37	0.660•	0.517	NS
	Range	47 – 108	44 – 96			
Age at time of operation in months	Median (IQR)	5.5 (4.5 – 27)	6 (5.5 – 6)	-0.304 $\neq$	0.761	NS
	Range	4 – 48	3 – 12			
Interval time between operation and evaluation (months)	Mean $\pm$ SD	50.00 $\pm$ 8.91	52.75 $\pm$ 14.33	-0.362•	0.721	NS
	Range	42 – 60	34 – 90			

P-value > 0.05: Non significant; P-value < 0.05: Significant; P-value < 0.01: Highly significant

\*: Chi-square test; •: Independent t-test;  $\neq$ , Mann-Whitney test

As regarding to the type of operation there is **no statistical significant** difference between Constipation groups and the Type of operation variable as (p value) = 1.00.

As regarding to the Number of enterocolitis attacks there is **no statistical significant** difference between constipation groups and the Number of enterocolitis attacks variable as (p value) = 0.307.

As regarding to the Numbers of Hospital admission there is **no statistical significant** difference between Constipation groups and the Hospital admission variable as (p value) = 0.622.

As regarding to Home treatment there is **no statistical significant** difference between Constipation groups and the Home treatment variable as (p value) = 0.162.

**Table No (8): P- test for statistical differences between variables of (Type of operation, Number of enterocolitis attacks, Hospital admission No and Home treatment) with Constipation groups**

		Constipation groups		Test value	P-value	Sig.
		No	Yes			
		No. = 5	No. = 15			
<b>Type of operation</b>	Transanal Soave	3 (75.0%)	12 (75.0%)	0.000*	1.000	NS
	Transanal swenson	1 (25.0%)	4 (25.0%)			
<b>Number of enterocolitis attacks</b>	Median (IQR)	0.5 (0 – 4)	2 (1 – 2.5)	-1.022 $\neq$	0.307	NS
	Range	0 – 7	0 – 6			
<b>Hospital admission</b>	Median (IQR)	0.5 (0 – 4)	1 (0.5 – 2)	-0.493 $\neq$	0.622	NS
	Range	0 – 7	0 – 3			
<b>Home treatment</b>	Median (IQR)	0 (0 – 0)	0 – (0 – 2)	-1.400 $\neq$	0.162	NS
	Range	0 – 0	0 – 4			

P-value > 0.05: Non significant; P-value < 0.05: Significant; P-value < 0.01: Highly significant

\*: Chi-square test; •: Independent t-test;  $\neq$ , Mann-Whitney test

#### 4. Discussion

Transanal pull-through is the latest and the most popular way of operation for the treatment of HD. Firstly was introduced by De la Torre-Mondragon and Ortega Salgado in 1998. It is preferable to other treatment options because it prevents laparotomies, laparoscopies, and colostomies in many cases. TERPT is especially useful when aganglionosis is restricted to the rectosigmoid region although the short-term results of using total TERPT operations to treat HD

are satisfactory; the long-term results are still being evaluated (**Gunnarsdottir A et al 2010**)

This study aims to highlight the delayed postoperative functional outcome for patients who had been diagnosed and operated as HD (Transanal approach). We assessed three main outcomes that included: fecal continence, postoperative attacks of HAEC, and constipation.

Enterocolitis is a common complication of HD both before and after surgery. There is some evidence

implicating alterations in intestinal mucin production and the mucosal production of immunoglobulins, which presumably results in loss of intestinal barrier function and allows bacterial invasion (**Demehri FR et al 2013**).

In this study, 20% of cases did not develop attacks of enterocolitis following surgery, 60% had 1-3 attacks (mild), 15% had 4-6 attacks (moderate), and only 1 case (5%) had recurrent attacks (more than 6) with mean follow up time (34 – 90 months). Criteria of enterocolitis used this study was Fever, lethargy, decreased peripheral perfusion, distended abdomen, tachycardia, hypotension, explosive discharge of gas and stool on rectal examination, and perianal excoriation. We statistically tested whether factors like age at operation, type of operation (Soave versus Swenson) would influence postoperative HAEC but this is statistically non-significant. Every patient with attack of HAEC was admitted at least 3 to 5 days and clinical examination done such PR examination to enroll in or out any stricture or tight muscle cuff and X-ray abdominal erect to see intestinal edema. the patient then received intravenous antibiotics, rectal irrigation was done and nothing per oral to decompress the abdomen. For recurrent cases of HAEC, we did a new contrast enema or redo Histopathological examination for old slides to ensure there is no residual a ganglion segment. The result of both contrast enema and revision of the slides was normal. This higher rate of enterocolitis may be because of low threshold in diagnosing early cases of enterocolitis.

The rate of enterocolitis varies widely in published reports, from a higher rate of 54% by **Van Leeuwen et al. (2002)** to a lower rate of 4.55% in **Zhang et al. (2005)** study of which may attribute to criteria of enterocolitis used in different studies.

**Teitelbaum** found in his result that the rate of enterocolitis varies from 1.4% to 42%.

**Hadidi (2003)** reported a 4.4% enterocolitis rate utilizing the transanal approach in comparison to a 12% incidence in a preceding series of open pull-through procedures. This is a very low incidence of post-pull-through enterocolitis which differs from most of the studies reported in the literature.

**Akshay et al** are founded the frequency of post-operative enterocolitis in the current series is still less than reported following other surgical approaches the relative low incidence of enterocolitis after TERPT in the current series may be related in part to the short sero muscular cuff, the low coloanal anastomosis, and their policy of routine postoperative anal dilatation particularity in neonates and infants.

TERPT requires traction on the anal sphincters for end rectal dissection and anastomosis. Such manipulation of the anal canal may induce

overstretching of the anal sphincter muscle and has been suggested as a potential cause of fecal incontinence (**El-Sawaf et al 2007**).

The evaluation of fecal continence and bowel function in patients operated for HD is performed using parameters such as voluntary bowel movement, soiling, and constipation. Voluntary bowel movement is considered the most significant sign of fecal control. It is defined as the act of feeling the urge to use the toilet, and the ability to verbalize it and hold back defecation if necessary. Constipation is defined as the patient's inability to have a complete bowel movement and empty the rectum without any help, either by diet, laxatives or enemas. Finally, soiling is the involuntary leaking of small amounts of stool in the underwear that may provoke devastating effects on the patient's self-esteem and social behavior (**Levitt and Pena, 2005**).

In this study we used the Krikenbeck score is a newer score which is far simpler that assess the 3 basic problems encountered in HD patients namely: voluntary control, soiling, and constipation. Its main advantage is that it can divide patients into rather comparable groups with shared criteria in each, as patients with voluntary control versus involuntary group, soiling versus no soiling and constipation versus no constipation.

In this study, we found most of the cases (90%) had voluntary bowel movements. The last (10%) can be getting better by good bowel training and by changing bowel habits.

Regarding fecal soiling, 25% had no soiling, 60% had mild soiling (grade 1), and 15% had moderate soiling (grade 2) with no social problem; while we had no cases with severe post-operative soiling. The soiling group is examined for rectal examination and examination under general anesthesia to detect the level of coloanal anastomosis which is above the dentate line this group is correctable by laxative treatment and changing the diet style of this group. In contrivers the other group which the level of colo-anal anastomosis which is below the dentate line this group is uncorrectable and treated by bowel management for life.

**Ekema et al.** noticed in their study that Postoperative soiling accidents were observed occasionally in 4 patients (10%), often in 18 patients (45%). Soiling was not observed in 18 patients (45%). Soiling was observed at night in 16 patients (40%) and at day and night in 6 patients (15%).

As regarding Constipation in this study was, absent in 4 patients (20%) and present in 16 patients (60%) and divided in two grads, grade one in 10 patients (50%) and grade two in 6 patients (30%). Every patient with constipation full examination done and good history taken bout time and frequency of

defecation. We noted the first group with constipation grade one is correctable by changing lifestyle and modification in diet and not needed any medical treatment. In the second group with grade 2 constipation are getting better by enema and added laxative and we noted that after this treatment the bowel habit and frequency of defecation become good.

Multidisciplinary behavioral treatment is effective for incontinence and constipation after correction of HD. (**Schmiedeke et al 2008**).

In literature, we noticed that Zhang et al reported 5 cases of constipation and 3 of incontinence in a series of 58 patients after trans anal one stage pull through. Teitelbaum et al. found that constipation in 28% of 78 children. Elhalaby et al. reported that a complete continence in 35 out of 42 patients after one stage TERPT and 6 patients had constipation (14.3%). We found constipation in 16 patients.

This high incidence of constipation may be due to short follow up time of the study as this complaint tends to improve over time with the aid of laxatives and regular post-operative dilation.

**Ekema et al. (2003)** noticed in their study to 15 children reported that 7 patients (46.6%) had constipation.

**Zhang et al. (2003)** studied 76 children who underwent staged Swenson operation for HD; they showed that constipation occurred in 6.7% of the patient incontinence in 6.7% and soiling in 11% of patients.

**Zhang et al. (2005)** studied 58 patients who underwent transanal pull-through operation for HD, the results were soiling in 9 patients (15.5%) and constipation in 5 (8.6%) and no incontinence was obtained in any patient.

In other studies explained that the satisfaction of the parents with the results is a subjective object varies with the socioeconomic levels of the parents; their education and their residence (rural or urban) Moreover, one other point deserving recognition is that, currently, the purpose of the management of HD is not only to ensure survival, but also to achieve normal bowel function and, more importantly, to achieve and good quality of life in all possible ways. Surgery is not the end of the treatment. We feel it is necessary to emphasize the long-term regular follow up of patients after surgery for HD because it is the prerequisite for improvement of the quality of life. Strategies, including psychological counseling, medication therapy, and toilet training, are needed.

One limitation of the present study is the small sample size; we focused only on the management of HD patients with only transanal pull through, short time of follow up and some patients not responding to be evaluated.

Despite its limitations, this study demonstrates further clinical evidence for late complications of trans anal pull through for HD patients. It also provides data that will enable the power of broad-spectrum follow up for HD patients in the next studies.

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