Comparative Study between One Stage Total Transanal Endorectal Pullthrough and Assisted Transanal Soave for Treatment of Hirschsprung's Disease

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Purpose: *Was to compare one stage total transanal endorectal pullthrough and assisted transanal Soave (either by minilaparotomy or laparoscopy) for treatment of Hirschsprung's disease.*

Patients and methods: *This study was conducted on 40 pediatric patients with Hirschsprung's disease. The patients were divided into two groups; group (A); 20 cases with one stage total transanal endorectal pullthrough, and group (B); 20 cases with assisted transanal Soave (10 cases mini-laparotomy (B_1) and 10 cases laparoscopy (B_2)).*

Results: *Mean age of patients was 20.01 months for group A and 17.14 months for group B. The ratio of male to female was 3:1 and P value was highly significant <0.001. The mean operative time in group A was 102 min. While in group B it was 117 min. The onset of oral feeding in group A was with mean of 1.7 ±0.86 days while in group B with mean of 2.25 ±0.97days. The mean postoperative hospital stay in group A was 4.2 ±1.73 days and in group B was 5 ±1.87days. Complications were more common in group B than group A, and more common in subgroup B_1 than B_2.*

Conclusion: *Transanal endorectal pull-through is characterized by a shorter operating time, less bleeding, shorter hospital stay, less morbidity and earlier recovery than similar open pull-through procedures.*

Key words: *Hirschsprung's disease, transanal endorectal pull-through, assisted transanal.*

Introduction:

Hirschsprung's disease (HD) is a congenital aganglionosis of the submucosal and myenteric neural plexuses principally affecting the rectosigmoid or rectal segments of varying lengths. Most cases manifest during the neonatal period, but in rare instances, the disease is initially diagnosed in older children and adult patients.[1]

Since the seminal description of Hirschsprung's disease in 1889, there has been a gradual evolution in the surgical management of this condition. Recognition that Hirschsprung's disease arises from functional obstruction in the distal, aganglionic colon led Swenson to advocate resection of this segment. Later, Duhamel described a retrorectal anastomosis and Soave an extramucosal dissection, to minimize risk of neurovascular injury. Primary endorectal pull-through without enterostomy has gained in popularity since first described, being further modified to include minimally invasive approaches.[2]

The one-stage transanal endorectal pull-through operation (TEPT) was introduced in the late 1990s and had rapidly replaced traditional procedures in infants and young children in many surgical centers around the world.[3]

Single-stage pull-through, both with and without laparoscopic assistance, has enabled surgeons to perform definitive surgical correction at an earlier age than previously possible.[4]

Minimally invasive laparoscopic techniques gained popularity because of their superior cosmetic results and shorter hospital stay. A completely transanal approach without any intraabdominal dissection had generated considerable interest. This procedure has the potential advantages of lower cost, less risk of damage to pelvic structures, lower incidence of intraperitoneal bleeding and adhesion formation, and absence of any abdominal incision.[5]

Routine laparoscopic visualization or minilaparotomy is not necessary with the transanal approach, but it should be used in children who are at higher risk for long segment disease or if any difficulties encountered during the procedure.[6] The limitation of transanal pullthrough is aganglionic segment extending proximal to the sigmoid colon because of difficulty in achieving adequate mobilization of the colon.[5]

The aim of this work was to study and compare one stage total transanal endorectal pullthrough and assisted transanal soave (either by minilaparotomy or laparoscopy) for treatment of Hirschsprung's disease.

Patients and methods:

During the period from January 2013 to June 2015, this study was conducted on 40 pediatric patients with Hirschsprung's disease. The patients were divided into two groups; group (A); 20 cases with one stage total transanal endorectal pullthrough, and group (B); 20 cases with assisted transanal Soave (10 cases mini-laparotomy (B_1)and 10 cases laparoscopy (B_2)).

These patients were managed at pediatric surgery unit, Benha university hospitals and pediatric surgery department, Ain Shams university hospitals after obtaining approval from local ethical committee and after a fully informed consent taken from the parents discussing with them the operative procedure and the possible intraoperative and postoperative complications.

Inclusion criteria:

Hirschsprung's disease with transitional zone in the rectosigmoid (Classic type)

Exclusion criteria:

1. Previous colortectal surgery for Hirschsprung's disease other than rectal biopsy.
2. Associated congenital syndromes (e.g., Down syndrome).
3. Ultrashort or total colonic agangilionosis.
4. Generally unfit patients due to other causes as congenital heart diseases, …etc.

Preoperative Assessment:

1- Full clinical history taking including:

All the cases were presented with chronic constipation and abdominal distension. They had history of delayed passage of meconium (more than 48 hours).

2- Clinical examination including:

General condition, weight, abdominal examination: Abdominal distension, palpable colon, and rectal examination (presence of fecal matter, gush of stools, anal tightness and sphincteric state).

1- Laboratory tests:

Routine preoperative laboratories: CBC, coagulation profile (PT, PTT, and INR), liver and renal function tests (AST, ALT, albumin, and Na, K, Urea, creatinine).

2- Radiological examination:

Contrast (Barium or Gastrograffin) enema: For the transition zone (TZ), retained contrast on a postevacuation film, and abnormalities of the rectal mucosal folds.

3- Rectal biopsy:

All patients were submitted to full thickness rectal biopsy under general anesthesia. The definitive diagnosis of HD is based on histological evaluation of a rectal biopsy, looking for the presence or

Figure (1): An anal retractor is placed (Lone Star type).

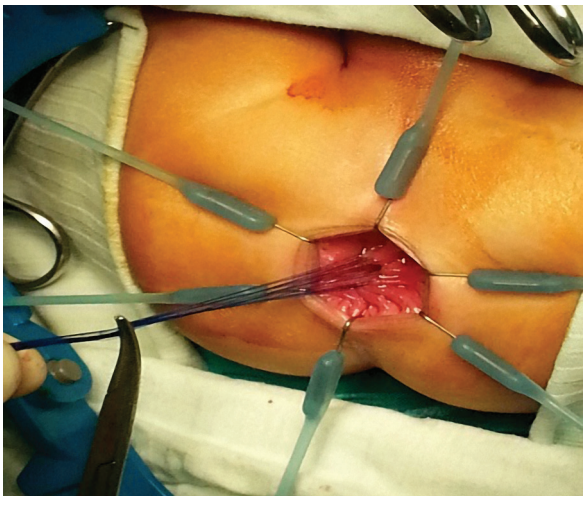

Figure (2): Mucosal cuff is tagged with multiple fine sutures

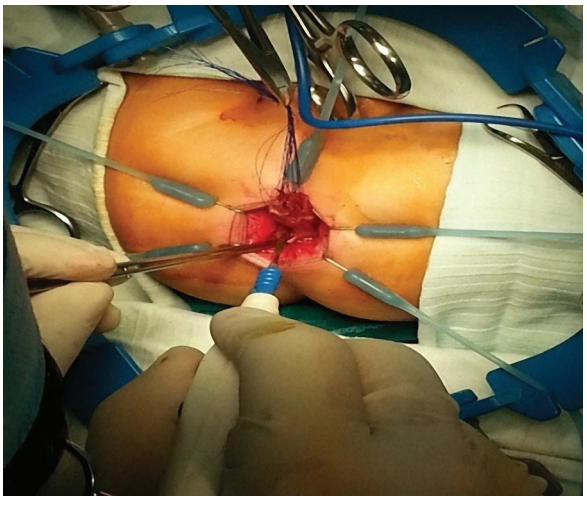

Figure (3): Rectal mucosa is circumferentially incised.

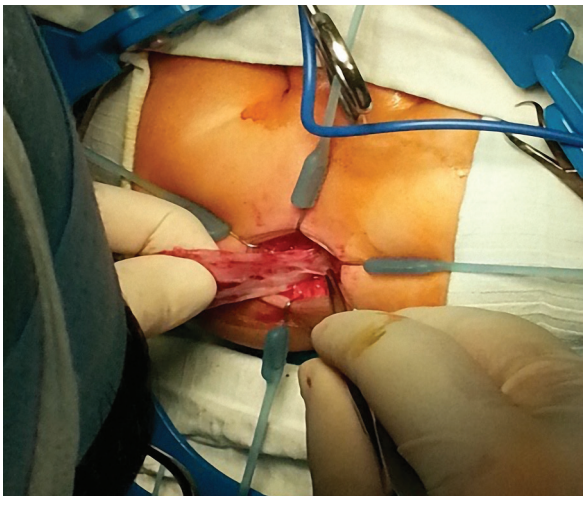

Figure (4): The submucosal plane is developed.

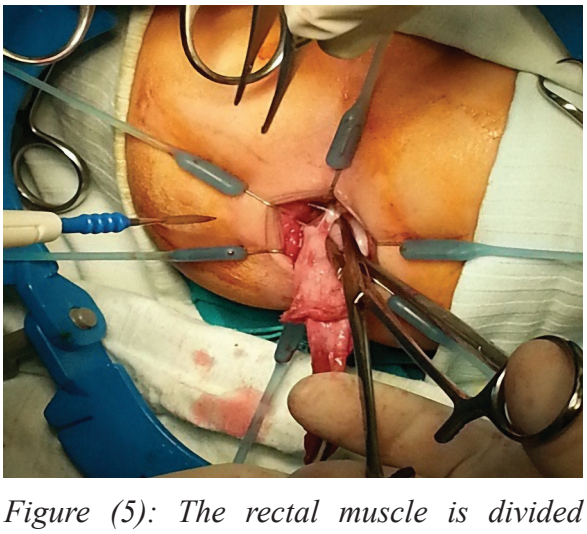

Figure (5): The rectal muscle is divided circumferentially.

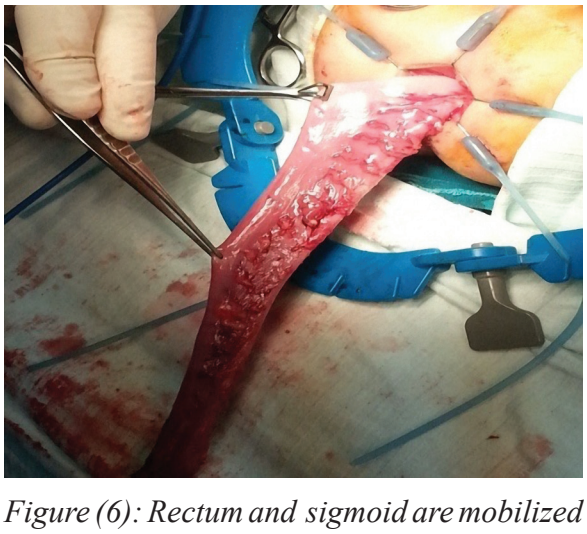

Figure (6): Rectum and sigmoid are mobilized out through the anus.

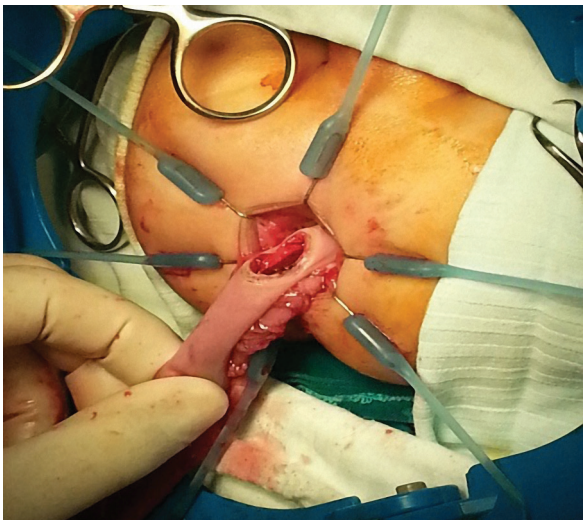
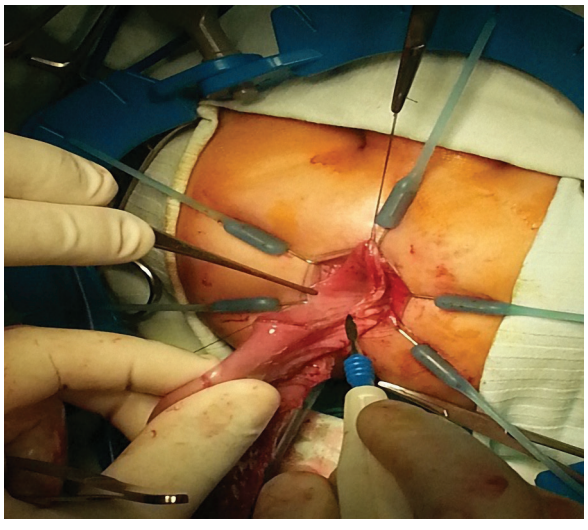

Figure (7,8): Resection of the aganglionic segment.

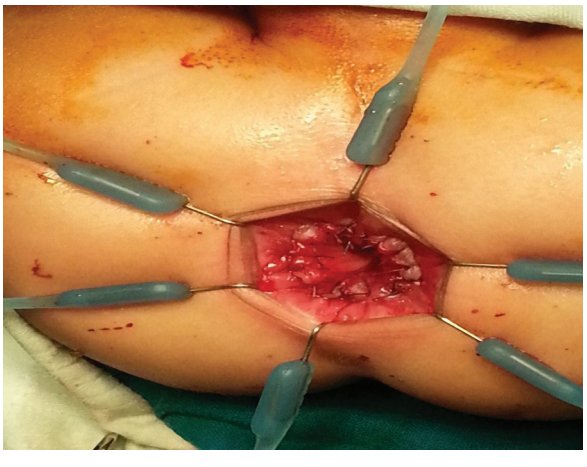


Figure (9,10): Coloanal anastomosis is performed using absorbable suture.

Figure (11): The photograph shows port placement for this operation.

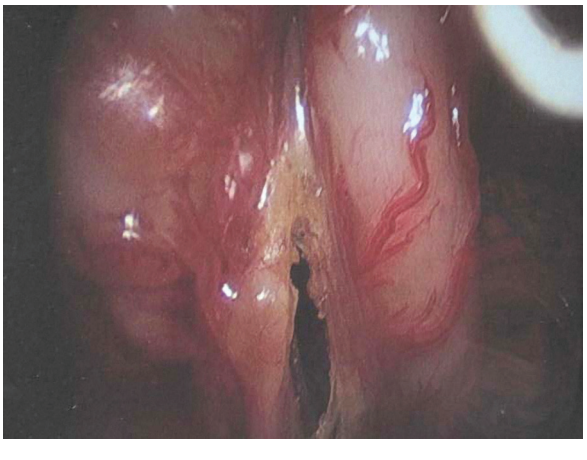

Figure (12): Window is developed through the rectosigmoid mesocolon.

absence of ganglion cells and the finding of hypertrophied nerve trunks

Operative procedure:

A) Preoperative preparation:

The status of nutrition, hydration, electrolyte, and acid-base balance must be optimal. All the patients were consulted by pediatrician to rule out other associated congenital anomalies.

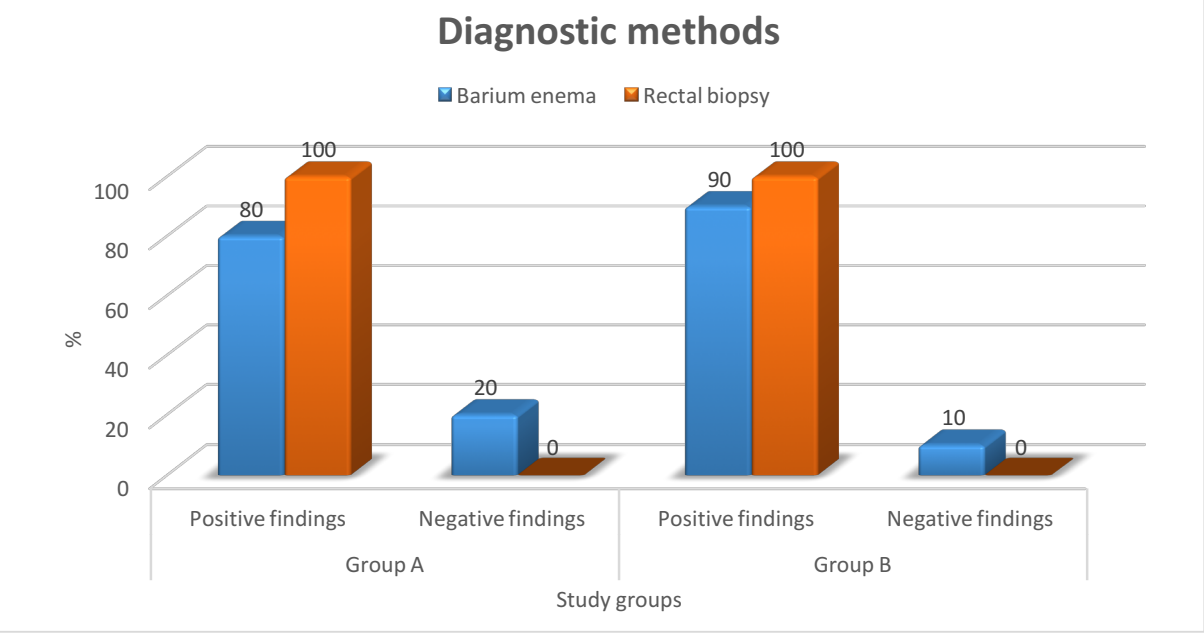

Figure (13): Diagnostic methods.

Table (1): Diagnostic methods.

Group	Diagnostic methods			
	Barium enema		Rectal biopsy	
	Positive findings	Negative findings	Positive findings	Negative findings
Group A (n=20)	16(80.0%)	4(20.0%)	20 (100.0%)	0(0.0%)
Group B (n=20)	18(90.0%)	2(10.0%)	20(100.0%)	0(0.0%)
B1(n=10)	8(80.0%))	2(20.0%)	10(100.0%)	0(0.0%)
B2 (n=10)	10(100.0%)	0(0.0%)	10(100.0%)	0(0.0%)

Bowel preparation:

All patients were on clear oral intake for 24 hours, after that they were kept fasting and intravenous fluids were started for another 24 hours. Warm saline colonic enemas (20cc/kg/enema) performed every 6 hours.

An intravenous intestinal antiseptic (Mitronidazole 15 mg/kg infused over one hour) and third generations cephalosporin (Cefotaxime 50mg/Kg/day) were used at induction of anesthesia.

B) Operative technique:

1. Technique of total transanal endorectal pull-through:

After general anesthesia. A urinary catheter of adequate size was used. The patient was placed at the end of the operating table with the feet fixed to a cross bar at the end of the table, the child was put into lithotomy position with knees and hips kept flexed to access to the patient's perineum.

An anal star retractor was placed, Also, retraction was achieved using eight perianal retraction 3-0 or 4-0 silk sutures to evert the anus and expose the rectum.

The rectal mucosa was circumferentially incised using the cautery, approximately 5-20 mm from the dentate line, and the submucosal plane was developed.

The submucosal plane was infiltrated with 1 in 200.000 adrenaline solution. The mucosal cuff was tagged with multiple fine sutures, which were used for traction.

The rectal mucosa was circumferentially

Table (2): Comparison between the two groups regarding operative data.

Group	Incision above the dentate line (mm)		Transitional zone from anal verge (cm)		Total length of resected colon (cm)		T1 & P1	T2 & P2	T3 & P3
	Mean ±SD	Range	Mean ±SD	Range	Mean ±SD	Range			
Group A (n=20)	9.7±3.29	5-15	13.05±5.39	5-23	25.1±7.52	10-34	1.52& 0.14	0.96 & 0.34	1.88 & 0.07
Group B (n=20)	11.65±4.70	5-20	11.55±4.38	4-18	20.9±6.54	10-30			
B1 (n=10)	13±5.46	5-20	10.9±4.36	4-18	19.7±6.11	10-28	1.31& 0.21	0.65& 0.52	0.81 & 0.43
B1 (n=10)	10.3±3.59	5-15	12.2±4.54	5-18	22.1±7.05	10-30			

t1&P1 for Incision above the dentate line; t2&P2 for Transitional zone from anal verge; t3&P3 for Total length of resected colon

Table (3): Comparison between the two groups in relation to total operative time.

Group	Total operative time (minutes)			T	P-value
	Mean	±SD	Range		
Group A (n=20)	102	20.8	60-140	1.79	0.08
Group B (n=20)	117	31.09	80-180		
B1 (n=10)	110	35.82	80-180	1.01	0.33
B2 (n=10)	124	25.47	80-160		

Table (4): Comparison between the two groups in relation to postoperative onset of oral feeding:

Group	Postoperative onset of oral feeding (days)			T	P-value
	Mean	±SD	Range		
Group A (n=20)	1.7	0.86	1-3	1.90	0.06
Group B (n=20)	2.25	0.97	1-4		
B1 (n=10)	2.4	1.17	1-4	0.68	0.50
B2 (n=10)	2.1	0.74	1-3		

Table (5): Comparison between the two groups in relation to postoperative hospital stay.

Group	Postoperative hospital stay (days)			T	P-value
	Mean	±SD	Range		
Group A (n=20)	4.2	1.73	2-7	1.93	0.06
Group B (n=20)	5.3	1.87	3-9		
B1(n=10)	5	1.88	3-9	0.71	0.49
B2(n=10)	5.6	1.9	3-9		

incised using the cautery, approximately 5-20 mm from the dentate line, and the submucosal plane was developed.

The endorectal dissection was then carried

Table(6): Comparison between the study groups regarding postoperative complications:

Group	Wound infection	Ileus	Stricture	Constipation	Fecal incontinence
Group A (n=20)	2(10.0%)	2(10.0%)	2(10.0%)	4(20.0%)	0(0.0%)
Group B (n=20)	4(20.0%)	3(15.0%)	3(15.0%)	6(30.0%)	0(0.0%)
B1 (n=10)	3(30.0%)	2(20.0%)	2(20.0%)	4(40.0%)	0(0.0%)
B2 (n=10)	1(10.0%)	1(10.0%)	1(10.0%)	2(20.0%)	0(0.0%)
Z1&P1*	0.88&0.37	0.48&0.63	0.48&0.63	0.73&0.46	-
Z2&P2*	1.12&0.26	0.63&0.53	0.63&0.53	0.97&0.33	-

*Obtained using the test of proportion (Z) for two samples; Z1 & P1 for Group A vs. Group B; Z2 & P2 for Group B1 vs. Group B2

proximally, staying in the submucosal plane. When the submucosal dissection had extended proximally to a point above the peritoneal reflection, the rectal muscle was divided circumferentially and the full thickness of rectum and sigmoid was mobilized out through the anus. This required division of rectal and sigmoid vessels, which could be done under direct vision using cautery or ligatures. When the transition zone was encountered, full-thickness biopsy sections were taken, and frozen section confirmation of ganglion cells was obtained.

The resection of the aganglionic segment was extended approximately 5–10 cm proximal to the identified transition zone. This modest extension of the dissection allowed for removal of dysfunctional bowel, which was often present proximal to the transition zone and not readily identified by rapid frozen section biopsy. The rectal cuff was grasped with Allis clamps on either side of the intended point of transection anteriorly or posteriorly. The intussuscepted cuff was trimmed and then returned to the pelvis.

2. Technique of Minilaparotomy assisted Transanal pull-through:

We used a small 5 cm incision over a skin crease, situated 2-4 cm above the left inguinal ligament. From this mininlaparotomy incision we Identified the transitional zone.

A) Mobilized and devascularized the colonic segment to be resected.

B) Before completing the perineal pull-through, the minilaparotomy incision was used to make sure that:

I. No intra-abdominal bleeding.

Ii. The colon was not over-stretched and anastmosis is not under tension.

Iii. No colonic torsion.

Iv. No mesenteric defect to allow internal hernia to occur.

3. Technique of Laparoscopic Assisted Transanal Pull-through:

The transition zone was located visually when possible. A seromuscular biopsy was obtained with laparoscopic Metzenbaum scissors for histologic leveling. Again, the resection of the aganglionic segment extended approximately 5–10 cm proximal to the identified transition zone.

Once inside the peritoneal cavity, the perineal and laparoscopic dissection planes were joined circumferentially. The muscular cuff should was divided. The rectum and colon were pulled down through the anus until the selected site of proximal colon resection was identified then coloanal anastmosis was done as mentioned in pure transanal pull-through.

Postoperative follow up:

A diet was introduced when there was evidence of bowel function, Prophylactic oral Metronidazole for 1–2 weeks was given postoperatively to avoid enterocolitis in the early postoperative period. Patients were followed up after 1 month, 6 months and 1 year from surgery.

Results:

Statistical analysis:

The collected data were summarized in terms of mean ±SD and range for quantitative data and number and percentages for qualitative data. Comparisons between the study groups were carried out using the Student's t-test to compare mean differences in two groups; the test of proportion Z-test to compare two proportions; the Chi-squared test (χ^2) and Fisher's Exact Test to compare more than two proportions as appropriate. Pearson's correlation coefficient (r) was used to assess the correlation of the studied data, when normally distributed. Statistical significance was accepted at P value <0.05 .

Table (1) The barium enema was diagnostic in 34 cases (85% of cases) 16 cases in group A (80%) and 18 cases in group B (90%); 8 cases in B1 (80%) and 10 cases in B2 (100%), The transition zone was present at rectosigmoid junction in 30 cases (88.2% of cases with barium positive findings -16 cases in group A and 14 cases in group B; 5 cases in B_1 and 9 cases in B_2), at the proximal part of rectum in 3 cases of (B_1) (8.8% of cases with barium positive findings), at the proximal part of sigmoid colon in 1 case in (B_2) (3% of cases with barium positive findings).

Table (2) shows there is no statistically significant difference between the two groups regarding operative data including; incision above the dentate line, transitional zone from anal verge, total length of resected colon.

Table (3) shows that the mean operative time in group A was 102 min. While in group B it was 117 min. The minimum operative time in Group A was 60 min. In 2 weeks old newborn and the maximum was 140 min. In 6 years old child, while The minimum operative time in Group B was 80 min. In 2 weeks old newborn and the maximum was 180 min. In 3 years old child. There is no any statistically significant difference between the two groups with P value 0.08.

Table (4) shows the onset of oral feeding ranged from one to three days in group A with mean of 1.7 ±0.86 days while in group B ranged from one to four days with mean of 2.25 ±0.97days (B_1 =2.4 and B_2 =2.1). P value of both groups; A and B was 0.06 while P value of subgroups; B1 and B2 was 0.50 which is statistically insignificant.

Table (5) shows the mean postoperative hospital stay in group A 4.2 ±1.73 days and in group B was 5 ±1.87days (B_1 =5 day and B_2 =5.6day, with p value 0.49). There was no significant difference between the two groups with p value 0.06. The minimum postoperative hospital stay in group A was two days and the maximum was seven days while in group B the minimum was three days and the maximum was nine days.

As regards the postoperative complications, **Table (6)** two patients (10%) in group A had mild ileus that responded to nasogastric tube and GIT rest within 2 days and three patients of group B (15%) had ileus (two patients in subgroup B_1 and only one in B_2), two patients in subgroup B_1 started oral feeding after 4 days and tolerated it well and did not need any further interference.

Two patients (10%) in group A and four patients (20%) of group B (three patients in subgroup B_1 and only one in B_2) had mild wound infection that responded to antibiotic therapy.

Anal stricture affected five patients in this study with a percentage of 12.5%, two patients in group A (10%) and three patients (15%) in group B (two patients in subgroup B_1 and one patients in B_2). Actually the anal stricture occurred in spite of the routine regular dilatation to all the patients in the study (routine anal dilatation were performed in all patients of both groups once or twice weekly for at least 3 weeks), and the five patients responded well to the anal dilatations that extended beyond the three weeks.

Ten patients (25%), four in group A (20%) and six (30%) in group B (four patients in subgroup B_1 and two patients in B_2), suffered from recurrent mild attacks of constipation after the pull-through procedure that responded to the transient use of laxatives.

There were no mortality in our study and also there were no instances of other complications such as postoperative bowel obstruction, anal incontinence, pelvic or

intra-abdominal abscesses, retraction or prolapse of the pullthrough segment or wound dehiscence.

Discussion:

This study was conducted on 40 pediatric patients with Hirschsprung's Disease, in whom transanal endorectal pullthrough was indicated. The Patients were divided into two groups; group (A); 20 cases with one stage total transanal endorectal pullthrough, and group (B); 20 cases with assisted transanal Soave (10 cases mini-laparotomy (B_1) and 10 cases laparoscopy (B_2)).

The patients of the study were 30 males and 10 females, group A included 14 males (70%) and 6 females (30%) while group B included 16 males (80%) , and 4 females (20%) B_1 included 7 male and 3 female while B_2 included 9 male and 1 female with a ratio 3:1, male to female and P value is highly significant; <0.001.

This finding is in agreement with The literature which describes a predominance of Hirschsprung's disease in males, from 3:1 to 4:1.[7]

The most common presentation in this study was constipation (100% in both groups) followed by inability to pass meconium (85% group A and 90% group B) , then abdominal distension (75% in both groups) and lastly bilious vomiting (25% group A and 20% group B). This findings were comparable with García-Arias and Ceciliano-Romero study on 130 patients at the National Children´s Hospital during the period 2000 to 2010, they identified bloating as the most frequent, followed by bilious vomiting, terminal meconium and constipation, for a 74%, 52%, 36% and 19%, respectively (García-Arias and Ceciliano-Romero, 2013.[8]

The barium enema was diagnostic in 34 cases (85% of cases) 16 cases in group A (80%) and 18 cases in group B (90%); 8 cases in B_1 (80%) and 10 cases in B_2 (100%), The transition zone was present at rectosigmoid junction in 30 cases (88.2% of cases with barium positive findings-16 cases in group A and 14 cases in group B; 5 cases in B_1 and 9 cases in B_2), at the proximal part of rectum in 3 cases of (B_1) (8.8% of cases with barium positive findings), at the proximal part of sigmoid colon in 1 case in (B_2) (3% of cases with barium positive findings)

The accuracy of barium enema as a diagnostic tool is reported to 63% by Hussain et al. 2002.[9] Ideally all the patients should have mucosal suction biopsy with acetyl cholinesterase staining of the nerve fibres (Zilur, et al. 2010).[10]

We were not fortuned in this regard and we did not have this facility. Therefore we rely mostly on full thickness rectal biopsy with H/E staining and rectal biopsy was diagnostic in all cases.

Operative time was favorable in group A compared to group B (the main 102 vs 117 min with P value 0.08). It was also favorable in group B_1 compared to group B_2 (110 vs 124 min with P value 0.33). In this study; although operative time was favorable in group A, but there is no any statistically significant difference between the two groups with P value 0.08. There was also a significant positive correlation between operative time and onset of oral feeding with p value <0.001, also there was a significant positive correlation between operative time and postoperative hospital stay with p value <0.001.

The mean operative time in group A (102 min) is significantly shorter when compared with the Egyptian multicenter study of El-Halaby et al. (Elhalaby, 2004)[11] 120.2 ±27.8 min, and with that of Teeraratkul (Teeraratkul, 2004)[12] 140 min. This difference may attributed to increasing learning curve and gaining experience.

The differences in the operative time between the different studies is due to that the operative time is influenced by many factors including age (the younger, the age, the less time is needed), preoperative enterocolitis, adherent musosa and intra-operative bleeding (the older the child the more the bleeding) (Hadidi, 2003).[13]

The onset of oral feeding ranged from one to three days in group A with mean of 1.7 ±0.86 days while in group B ranged from one to four days with mean of 2.25 ±0.97days (B_1

=2.4 and B_2 =2.1). P value of both groups; A and B was 0.06 while P value of subgroups; B_1 and B_2 was 0.50 which is statistically insignificant.

Most of the studies regarding both totally transanal and assisted transanal pull-through agree with our study in starting postoperative oral feeding. Shabbir, et al. (in their study of transanal pull-through) started the postoperative oral feeding 1-3 days postoperatively (Shabbir, et al. 2013).[14]

The mean postoperative hospital stay in group A 4.2 ±1.73 days and in group B was 5 ±1.87days (B1 =5day and B2=5.6day, with p value 0.49). There was no significant difference between the two groups with p value 0.06. The minimum postoperative hospital stay in group A was two days and the maximum was seven days while in group B the minimum was three days and the maximum was nine days. There was a significant positive correlation between age and postoperative hospital stay with p value <0.001.

Most of the studies regarding both totally transanal and laparoscopic assisted transanal pullthrough agree with our study in the postoperative hospital stay. Hadidi, (in his study of transanal pull-through) reported that postoperative hospital stay ranged around 3 days (Hadidi, 2003).[13] Shabbir, et al. (in their study of transanal pull-through), reported that postoperative hospital stay ranged around 5 day (Shabbir, et al. 2013).[14] As regards the postoperative complications, complications were more common in group B than group A, and more common in subgroup B_1 than B_2. Although these results, but there were no any statistically significant differences between the two groups . Two patients (10%) in group A had mild ileus that responded to nasogastric tube and GIT rest within 2 days and three patients of group B (15%) had ileus (two patients in subgroup B_1 and only one in B_2), two patients in subgroup B_1 started oral feeding after 4 days and tolerated it well and did not need any further interference.

Two patients (10%) in group A and four patients (20%) of group B (three patients in subgroup B_1 and only one in B_2) had mild wound infection that responded to antibiotic therapy.

It is believed that postoperative routine anorectal bouginage is an effective tool to prevent the occurrence of anal stricture and to decrease both the frequency as well as the severity of enterocolitis particularly in neonates and young infants (Hussam S. 2009).[15]

Anal stricture affected five patients in this study with a percentage of 12.5% , two patients in group A (10%) and three patients (15%) in group B (two patients in subgroup B_1 and one patients in B_2).

Rouzrokh et al. Reported anal stricture was 14% of patients in their study on TEPT procedure (Rouzrokh, et al. 2010).[16]

Ten patients (25%) , 4 in group A (20%) and 6 (30%) in group B (4 patients in subgroup B_1 and 2 patients in B_2), suffered from recurrent mild attacks of constipation after the pull-through procedure that responded to the transient use of laxatives.

Constipation may be expected to improve over time. Lifschitz and Bloss noted that 33% suffered from constipation after the initial operation, but only 9% reported persistent constipation after an average trof 5 years (Lifschitz and Bloss, 1985).[17]

There were no mortality in our study and also there were no instances of other complications such as postoperative bowel obstruction, anal incontinence, pelvic or intra-abdominal abscesses, retraction or prolapse of the pullthrough segment or wound dehiscence.

Continence is very important parameter that should be assessed as regards the treatment of Hirschsprung's disease, but unfortunately we could not assess it in this study due to the short period of follow up and also because all the patients of the study were 3 years or less with 75 % of them under one year, so to assess the continence this needs a large number of patients with varying ages and a long period of follow up. This notice was also reported by (Hadidi, 2003)[13] in his study in which the follow up ranged from 3 month to 3.5 years and he reported that this was too short to fully assess bowel function,

sexual function and continence.

Conclusion:

The advantages of totally transanal pull-through approach included its feasibility, improved cosmesis, that the procedure does not damage the pelvic structures, the reduced hospital costs, hospital stay, operating time, and overall improved quality of life, but overstretching of the internal anal sphincter remains a critical issue, which may impact the long-term continence outcome.

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