ORIGINAL ARTICLE

One stage Pull-through for Management of Hirschsprung Disease

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ABSTRACT

Keywords: Hirschsprung's disease; Pull-through; Soave procedures; Trans-anal; Trans-abdominal

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Background: Hirschsprung's disease is one of the commonly studied diseases with an incidence of about 1 per 5000 live births, males are more likely to be affected than females. Surgical management is rapidly changing with the most popular procedure is the single stage total trans-anal pull-through. **Objectives:** This study aimed to evaluate the management Hirschsprung's disease by one stage pull-through Soave procedure concerning perioperative, short-term outcomes and complications. **Methodology:** The study included 32 patients recruited from the Pediatrics Surgical Department, Assiut University Hospitals at the period between April 2015 and March 2019 and followed up at our clinic. **Results:** The advantages of this operation included a good cosmetic effect and a short hospital stay, and its safety has been proved by many studies, also surgical site infection was encountered only in 18.8% of cases in the trans-abdominal group, compared to no cases in the trans-anal group. In this study, post-operative incontinence was not significantly different between the two groups. Hospital stay was significantly longer in trans-abdominal than trans-anal group. **Conclusion:** According to this study trans-anal pull-through maneuver has the advantages of less incidence of ileus, constipation, pain, and incontinence, and shorter operative time and hospital stay above trans-abdominal one.

INTRODUCTION

Hirschsprung's disease (HD) was first presented in 1886 by Harald Hirschsprung, and now carries his name to the pediatric Congress in Berlin **(1).** The incidence of Hirschsprung`s disease ranges from 1 in 4400 - 1 in 7000 live births. The male-female ratio is 4:1 **(2)**. Among the families of children with HD, the incidence increases to approximately 6%, with a range of 2% to 18% **(3).** The diagnosis of HD is usually based on clinical history, radiological studies, anorectal manometry and histological examination of the rectal wall biopsy specimens **(4).**

Surgical management includes Leveling Colostomy: although a primary pull-through procedure is the preferred approach to children with HD, some children benefit

from an initial leveling colostomy **(5).** Swenson Procedure: although still used, the Swenson procedure has become less popular in recent years **(6)**. Duhamel Operation: has several advantages; ease of performance, reduction of anastomotic leaks and strictures, and retention of anal sensory receptors **(7).**

Soave's procedure: it is conventionally used in infants between 9 and 12 months of age, although several studies had reported primary Duhamel pull-throughs in younger patients **(8).** There are two techniques: Open Approach: typically, the child`s buttocks are brought to the end of the operating table, and the legs are padded and positioned on wooden skis or leg supports. Laparoscopic Approach: The basic operative principles are virtually the same with the laparoscopic approach **(9).** Trans-anal Approach: it has rapidly replaced the traditional procedure in many pediatric surgical centers around the world. The main advantage of TERPT is that it is minimally invasive. It does not require abdominal incision, and there should be no operation scar. The lack of intraabdominal dissection may decrease the chance of intraperitoneal adhesion formation and contamination. The risk of damage to pelvic structures is also reduced **(10).**

An assessment of the outcomes of HD different treatment modalities can generally be broken down into early and late findings, as well as the overall quality of life. **(11).** Complications after pull-through can be classified as early and late. Early postoperative complications include anastomotic leak and cuff abscess, bowel obstruction, perineal excoriation, stoma complications, and wound infection. Late complications include bowel obstruction, constipation, enterocolitis, incontinence, and stricture. There is some overlap between early and late complications. Factors that increase the risk of these complications include tension on the anastomosis or ischemia of the pull-through segment **(12).**

The current study aimed to evaluate the management of children with Hirschsprung's disease by one stage pull-through Soave procedures (trans-anal and trans-abdominal one stage pull-through) concerning perioperative, short term outcomes and complications.

PATIENTS AND METHODS

This RCT study was conducted between trans-anal and transabdominal one stage pull-through procedures for management of Hirschsprung's disease. The study included 32 patients recruited from the inpatients of the Pediatrics Surgical Department, Assiut University Hospitals at the period between April 2015 and March 2019 and followed up at our clinic. All patients aged 3 months to 16 years, from both sexes, have no operation before, associated with no other complex congenital malformations were included. Exclusion criteria involved patients with long segment Hirschsprung's disease, recurrence and those with associated complex other congenital malformations that necessitates other combined operation.

Preoperative assessment

All patients were subjected to: History taking, general examination, local examination of the abdomen for distension, scars, tenderness, and palpable masses and perianal region and PR examination for perianal fistula, position of the anus, anal wink, anal scars, and fissures.

Recruitment and randomization

All eligible patients scheduled for surgery were briefly informed about the study during the outpatient visit. After completion of the baseline assessment, participants were randomly allocated to one of the two intervention groups (**Group A:** 16 patients underwent trans-abdominal one stage pull-through procedure and **Group B:** 16 patients underwent trans-anal one stage pull-through procedure). Allocation was done by the biometrician based on a predetermined list generated with a blocked randomization SPSS procedure with a fixed block size. To prevent possible bias, study personnel involved in the recruitment and the baseline assessment did not have access to the randomization lists and were not aware of the block size.

Operative technique

I. Abdominal Trans-anal approach (Soave operation) (Fig. 1): supine position with Foley's catheter inserted in the bladder and nasogastric tube. The child`s buttocks were brought to the end of the operating table, and the legs were padded and positioned on wooden skis or leg supports. Left lower abdominal **(hockey stick)** or Pfannenstiel incision. Dissection involving mobilization of the aganglionic bowel and sufficient segment of the normally innervated bowel to reach the anus with an adequate blood supply without tension. Dissection was carried down to within 0.5 cm of the dentate line in the abdominal approach. An easy way to determine whether there is adequate length is to ensure that the ganglionic bowel be stretched over the pubis and to the level of the anal verge. Recognition that the internal sphincter may cause persistent obstructive symptoms has led us to pursue a more distal incision posteriorly into the internal sphincter during the definitive pull-through**.**

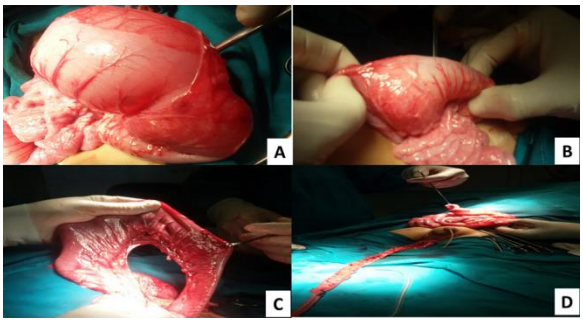

Fig. 1: Trans-abdominal approach (soave operation). (A) Colon prior to mobilization. (B) Identification of transitional zone. (C) Ligation of blood vessels. (D) Colon after complete mobilization and pull through

II- Pure Trans-anal pull through approach (Fig. 2). In Jack-knife position (we put buttocks at the end of the table) with Foley's catheter inserted in urinary bladder and nasogastric tube to the stomach). Four traction sutures were taken at 12, 3, 6, 9 O'clock from mucocutaneous junction to peri-anal skin if the retractors were not available. A circumferential incision was made 1cm above the dentate line to enter the submucosal plane. With meticulous hemostasis, the mucosa was dissected till the peritoneum was reached approximately 7-10 cm from anal verge. The seromuscular layer was incised circumferentially and the full thickness rectum was mobilized by electrocautery or ligature of its meso-rectum. Mobilization was continued from the narrow segment, transitional and the dilated segment till a near normal caliber colon is reached and a coloanal anastomosis was done as usual.

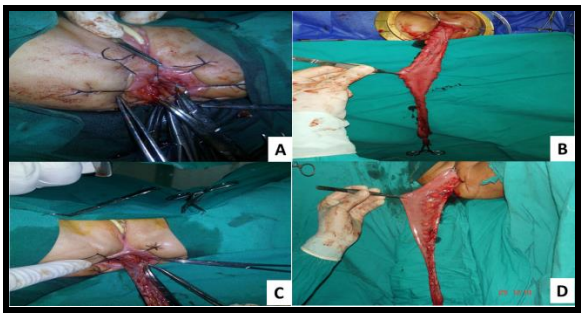

Fig. 2: Trans-anal approach (soave operation). (A) Traction sutures are taken at 12,3,6,9 O'clock from mucocutaneous junction to peri anal skin. (B) Skin retractors. (C) Dissection vessels. (D) Dissection muscles.

Statistical analysis

Data were verified, coded by the researcher, and analyzed using IBM-SPSS version 24. Descriptive statistics: Means, standard deviations, medians, ranges, and percentages were calculated. Test of significances: chi-square/Fisher's Exact test was used to compare the difference in distribution of frequencies among different groups. For continuous variables, independent t-test/Mann Whitney U test analysis was carried out to compare the means and medians for parametric/non-parametric data. A p-value < 0.05 was considered significant.

Ethical considerations

Approval for this study was obtained from Institutional review board (IRB) of Faculty of Medicine, Assiut University hospital prior to study execution. In addition, all participants/caregivers received a written consent form. The informed consent was clear and indicated the purpose of the study, and their freedom to participate or withdraw at any time without any obligation. Furthermore, participants' confidentiality and anonymity were assured by assigning each participant with a code number for the purpose of analysis only. The study was not based on any incentives or rewards for the participants and was abided to the guidelines of Helsinki Declaration and the CONSORT guidelines.

RESULTS

Of the 16 patients in the trans-abdominal group, 11 (69%) were males and 5 (31%) were females. The patients' age ranged between 1.5 and 15.5 years with a mean of 5.5 ± 3.3 years. Also, the mean patient's age at operation was 3.3 ± 3.3 years, with a median of 2.5 (0.5 – 15 years). Regarding trans-anal group, about four-fifth (n=13) of the sample was males and one-fifth (n=3) was females. **(Table 1).** Patients in group A were older than those in group B, and this was statistically insignificant (p > 0.002). As well, patients in in group A were older at time of operation than those in group B, and this was statistically insignificant (p > 0.043).

The average operative time was higher (ranged between 120 and 180 minutes with mean of 138 ± 24 minutes) in the transabdominal group than trans-anal group (ranged between 60 and 90 minutes with mean of 72 ± 12 minutes). Post-operative infection rates were illustrated in table 2. Rate of wound sepsis was higher in group A representing about 19% (n=3) compared with group B (0%) and this was statistically significant (p = 0.034) **(Table 2)**.

Table 1: Baseline Sociodemographic Characteristics of the studied Groups

	Trans-abdominal (n=16)	Trans-anal (n=16)	P-value
Age/years			
• **Mean ± SD**	5.50 ± 3.3	3.19 ± 2.2	**= 0.002***
• **Median (Range)**	5 (1.5 – 15.5)	2.5 (1 – 12.5)	
Sex			
• **Female**	5 (31.2%)	3 (18.8%)	= 0.343**
• **Male**	11 (68.8%)	13 (81.2%)	
Age at Operation/years			
• **Mean ± SD**	3.27 ± 3.1	2.21 ± 2.1	**= 0.043***
• **Median (Range)**	2.5 (0.5 – 15)	1.5 (0.3 – 11.8)	

***Mann-Whitney U test was used to compare the median differences.**
****Fisher's Exact test was used to compare the percentages between groups.**

Table 2: Operative and Data Comparison of the studied Groups

	Trans-abdominal (n=16)	Trans-anal (n=16)	P-value
Operative Time/hour			
• **Mean ± SD**	138 ± 24	72 ± 12	**= 0.002***
• **Median (Range)**	2.3 (2 – 3)	1.2 (1 – 1.5)	
Wound Sepsis			
• **No**	13 (81.2%)	16 (100%)	**= 0.034**
• **Yes**	3 (18.8%)	0 (0%)	
Enterocolitis			
• **No**	11 (68.8%)	13 (75%)	= 0.699
• **Yes**	5 (31.2%)	3 (25%)	
Retraction of the Rectum			
• **No**	15 (93.7%)	16 (100%)	= 0.500**
• **Yes**	1 (6.3%)	0 (0%)	

***Mann-Whitney U test was used to compare the median differences.**
****Fisher's Exact test was used to compare the percentages between groups.**

Regarding post-operative vomiting and distension, group A had statistically significant (p=0.003) higher rates (62.5%) compared with group B (12.5%). Likewise, rates of constipation were significantly (p = 0.039) higher (44%) in comparison with group B (19%). Also, partial disruption of the anastomosis was reported in only one case (6.3%) of group A. On the other hand, patients in group A had higher rates of post-operative paralytic ileus (37.5%) than group B (0%), and this was statistically significant (p = 0.018). Additionally, three patients (18.8%) in group A reported significant pain that was treated with strong analgesics, whereas pain was tolerable in group B (0%) and this was statistically significant (p = 0.034) **(Table 3)**. Regarding post-operative incontinence, group A had statistically insignificant (p = 0.723) lower rates (50%) compared with group B (56%). Likewise, rates of adhesive intestinal obstruction were insignificantly (p = 0.310) higher (6.2%) in comparison with group B (0%).

There was statistically significant (p = 0.001) longer duration of hospital stay among patients in group A (11.1 ± 6.4days) compared with patients in group B (5.0 ± 1.4 days). Additionally, proportion of patients underwent post-operative dilatation was higher in group A (94%) compared with those in group B (44%) and this was statistically significant (p = 0.002). Among those underwent dilatation by Hegar dilator 10-18, only one case (6.7%) failed in group A (trans-abdominal) and managed by strictureplasty **(Table 4).**

Table 3: Post-operative Complication Data Comparison of the studied Groups

	Trans-abdominal (n=16)	Trans-anal (n=16)	P-value*
Vomiting and Distention			
• **No**	6 (37.5%)	14 (87.5%)	= **0.003**
• **Yes**	10 (62.5%)	2 (12.5%)	
Constipation			
• **No**	9 (56.2%)	13 (81.2%)	= **0.039**
• **Yes**	7 (43.8%)	3 (18.8%)	
Bleeding			
• **No**	16 (100%)	15 (93.7%)	= 0.500
• **Yes**	0 (0%)	1 (6.3%)	
Partial disruption anastomosis			
• **No**	15 (93.7%)	16 (100%)	= 0.500
• **Yes**	1 (6.3%)	0 (0%)	
Paralytic Ileus			
• **No**	10 (62.5%)	16 (100%)	= **0.018**
• **Yes**	6 (37.5%)	0 (0%)	
Pain needs potent analgesia			
• **No**	13 (81.2%)	16 (100%)	= **0.034**
• **Yes**	3 (18.8%)	0 (0%)	
Incontinence			
• **No**	8 (50%)	7 (43.8%)	= 0.723
• **Yes**	8 (50%)	9 (56.2%)	

PO Adhesive Intestinal Obstruction			
• **No**	15 (93.8%)	16 (100%)	= 0.310
• **Yes**	1 (6.2%)	0 (0%)	
Stenosis			
• **No**	12 (75%)	14 (87.5%)	= 0.654
• **Yes**	4 (25%)	2 (12.5%)	

***Fisher's exact test was used to compare the percentages between groups.**

Table 4: Other Post-operative Data of the studied Groups

	Trans-abdominal (n=16)	**Trans-anal (n=16)**	**P-value**
Length of Hospital Stay/days			
• **Mean ± SD**	11.06 ± 6.4	5.00 ± 1.4	**= 0.001***
• **Median (Range)**	10.5 (1 – 20)	4.5 (3 – 7)	
Post-operative Dilatation			
• **No**	1 (6.2%)	9 (56.2%)	**= 0.002****
• **Yes**	15 (93.8%)	7 (43.8%)	
Response on Dilatation by Hegar Dilator 10-18			
• **Yes**	14 (93.3%)	7 (100%)	= 0.652**
• **Strictureplasty**	1 (6.7%)	0 (0%)	

***Independent t-test was used to compare the mean differences.**

****Fisher's Exact test was used to compare the percentages between groups.**

DISCUSSION

In 1998, De la Torre-Mondragon proposed a new treatment called single-stage trans-anal approach, which is more suitable for infants. This minimally invasive surgery with an anal approach has become an increasingly popular method for the treatment of HD, eliminating the risk of complications such as abdominal adhesions and pelvic nerve injury **(13)**. The advantages of TERPT include a good cosmetic effect and a short hospitalization time, and its safety has been proved by many studies. However, there are a variety of ways to choose surgery in clinical practice, and no consensus has been reached.

In our study the average age of patients in transabdominal group was higher than those in trans-anal group. Tannuri *et al*. reported similar results, that the patients were significantly older in the abdominal than trans-anal group. It had a mean value of 42 months compared to 11 months in the trans-anal group **(14)**. This difference could be

explained by differences in the medical setup of each country, and delayed referral from the surrounding rural areas.

In our study, the gender of the included cases was not significantly different between the two groups. Males represented 68.8 and 81.2% of cases in Groups A and B respectively. In agreement with or findings, Romero *et al*., reported that males represented 79.3 and 87.5% of cases in the transabdominal and trans-anal groups respectively, with no significant difference between the two groups **(15)**.

Regarding operative time in our study, it was significantly shorter in the trans-anal group. It had mean values of 138 and 70 minutes in Groups A and B, respectively. Romero *et al*. confirmed this where trans-anal maneuver had a mean operative time of 133 minutes compared to 204 minutes in the transabdominal approach **(15)**. This is because the trans-anal approach eliminates the time to open and close the laparotomy followed by the pull-through of the ganglionic colon in the transabdominal approach **(16).**

In our study, surgical site infection was encountered in 18.8% of cases in the transabdominal group, compared to no cases in the other group. It was evident that this complication was significantly associated with the transabdominal approach. On the contrary, Kim *et al*., reported nearly similar incidence of that complication in both groups. It occurred in 2 and 5% of cases in the transabdominal and trans-anal groups respectively **(17).**

In our study, enterocolitis occurred in 31.2 and 25% of cases in Groups A and B respectively, with no significant difference between the two groups. On the other hand, Hadidi reported slightly higher incidence of the same complication in the transabdominal group (12%) compared to the trans-anal one (4.4%)**.** The difference in the incidence of enterocolitis may be related to the length of aganglionic muscle cuff left behind and whether posterior midline myotomy was carried out **(18).**

In our study, post-operative vomiting and distension were encountered in 62.5 and 12.5% of cases in Groups A and B respectively, with a significant increase in its incidence in the transabdominal group. That would be reasonable with the increased incidence of post-operative paralytic ileus in the transabdominal group compared to the other group. Contrarily, Kim and his colleagues reported that the same complication was noted in 6 and 4% of cases in the transabdominal and trans-anal groups respectively **(17).**

The incidence of post-operative bleeding did not show any significant difference between the two study groups. It occurred in 0 and 6.3% of cases in the groups A and B respectively. However, Onishi *et al* reported that the trans-anal approach was associated with a significant decrease in blood loss compared to the transabdominal approach **(19).** Elrouby *et al* in a recent study confirmed the previous findings **(20).** In our study, partial anastomotic disruption was encountered in 6.3% of cases in the transabdominal group versus no cases in the trans-anal group. Visser *et al*. reported the higher incidence of anastomotic leakage for the transabdominal pull-through procedure (16%) compared to the lower leakage rate for the trans-anal procedure (4%) **(21).** On

the other hand, Stansrud *et al* negated the incidence of such complication in their study which included 28 and 24 cases in the trans-anal and transabdominal groups, respectively **(22).**

In our study, paralytic ileus was diagnosed in 37.5% of the transabdominal cases versus no cases in the trans-anal group. There was a significant increase in this complication with the transabdominal approach (p = 0.018). In the same context, Hadidi reported that passage of first bowel motion was noted on the 4th post-operative day in the transabdominal group compared to the passage of the 1st bowel motion on the 2nd days in the trans-anal group, indicating less incidence of ileus and early return of bowel movements with the trans-anal approach **(18).**

In our study, post-operative fecal incontinence was not significantly different between the two groups (p = 0.723). It was reported by 50 and 56.2% of cases in groups A and B, respectively. This could be explained by the direct sphincteric damage or nervous damage caused by the operative manipulation. Likewise, Romero *et al*., confirmed the previous findings as no significant difference was noted regarding the incidence of incontinence neither in children younger nor older than 5 years (p = 0.15 and 0.17 respectively **(15).**

Of note, although classical transabdominal pull-through involves little trans-anal manipulation, pelvic dissection may lead to lesion of regional nerves and impairment of urinary bladder function and fecal incontinence **(14).** During the follow up period scheduled in our study, adhesive intestinal obstruction was encountered in only one case in the transabdominal group (6.2%), with no difference between the two groups. In line with our findings, intestinal obstruction was detected in 9.8% of cases in that group versus no cases in the trans-anal group **(20).** In Sosnowska *et al*., adhesive intestinal obstruction was the most common complication after surgery for that disease and occurred in 10 (34%) children. Eight children did not respond to conservative treatment and underwent surgical treatment **(23).** The incidence of postoperative bowel obstruction can be decreased using laparoscopically assisted procedures, which is recommended by its proponents **(24).**

In our study, stenosis was insignificantly encountered in 25 and 12.5% of cases in Groups A and B respectively. All cases were well-managed by frequent dilatation, while only one case required stricturoplasty in the transabdominal group. Stensrud *et al*., reported no significant difference between the two approaches regarding the incidence of strictures that was encountered in 43 and 21% of cases in the trans-anal and transabdominal groups respectively **(22).**

In our study, the hospitalization was significantly prolonged in the transabdominal group compared to the trans-anal one. Of course, the increased complication rates in the transabdominal group will have its impact on the prolongation of the hospital stay. Likewise, **Romero *et al*.,** reported that the hospitalization showed significant increase with the transabdominal approach. Hospital stay had mean values of 9.8 and 17.7 days in the trans-anal and transabdominal groups respectively **(15).**

Our study has some limitations, it is a single center study. Additionally, the study lacks long-term follow up after the two procedures. The economic costs of both procedures should have been evaluated as well. These drawbacks should be well-covered in the upcoming studies.

CONCLUSION

In our study advantages of trans-anal maneuver include a good cosmetic effect and a short hospitalization time, and its safety has been proved by many studies, also surgical site infection was encountered in 18.8% of cases in the transabdominal group, compared to no cases in the other group. Increased incidence of post-operative paralytic ileus in the transabdominal group compared to other one. In our study, the hospitalization was significantly prolonged in the transabdominal group compared to the trans anal one. So, according to our study trans anal pull through has the advantage of shorter operative time, less incidence of ileus, constipation, pain, incontinence, and less hospital stay, above transabdominal pull through so we recommend doing trans-anal pull-through to patients with Hirschsprung disease.

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