Comparative study between two-stage trans-abdominal Duhamel procedure and two-stage Soave pull-through in treatment of Hirschsprung's disease: Clinical evaluation

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Abstract

Background: Hirschsprung's disease (HD) is a congenital anomaly affecting the bowel of neonates, due to a defect in the ganglion cells presence in the colon in a caudo-cranial fashion, various surgical techniques used to correct the aganglionic segment, *Duhamel* procedure, *Soave* pull-through were used to correct the defect.

Objectives: This study was conducted to compare the clinical outcome of two-stage *Duhamel* procedure and two-stage *Soave* pull-through in HD management.

Patients and method: 40 patients diagnosed with Hirschsprung's disease in the pediatric age group were enrolled in this study, 20 patients operated by *Duhamel* procedure, and 20 patients operated by *Soave* pull-through.

Results: Enterocolitis was detected in 40% of patients after *Soave* pull-through while it was 5% in patients subjected to Duhamel procedure (p=0.020). Stricture was detected in 15% of cases subjected to *Soave* pull-through, Patients subjected to *Duhamel* procedure didn't show stricture without significant difference. Regarding faecal continence, patients operated on by *Duhamel* procedure had better results compared to two-stage *Soave* pull-through (p<0.001). Regarding constipation, the differences were not significant between both groups.

Conclusion: Two-stage trans-abdominal *Duhamel* procedure has significantly better results as regards post-operative continence, constipation, and enterocolitis than *Soave* endorectal pull-through in the management of HD.

Keywords: Hirschsprung's disease; *Duhamel*; *Soave*

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Introduction

In 1691 Harald Hirschsprung explained the cause of constipation as megacolon, the derangement of peristaltic wave of the distal aganglionic portion of the intestine was reported by *Robertson* & *Kernohan* in the 1938. *Zuelzer* & *Wilson* demonstrated a functionally disturbed rectosigmoid segment with absent ganglion cells in 1948. Thereafter, *Whitehouse* & *Kernohan* proved such finding. **(Jensen and Frischer, 2022)**

Histological examination of HD revealed aganglionosis in the myenteric plexus which lies between the smooth muscle layers of the bowel wall as well as the submucosal plexus which is found within the submucosal layer of the bowel wall in a part or whole intestinal tract**. (Taguchi et al., 2019)**

Distended abdomen is the commonest symptom of HD in neonates, the prevalence of abdominal distension is about ninety three percent, associated with bilious vomiting, anorexia, and constipation. However, Hirschsprung associated enterocolitis (HAEC) is the most serious complication, as it is the leading cause of morbidity and mortality. **(Taguchi et al., 2019)**

The most applied protocol of treatment for HD patients include rectal irrigation and a primary one stage pull-through procedure avoiding the need for the staged procedure including an initial colostomy then definitive Pull-through. **(Hutchings et al., 2023)**

Soave procedure was created to decrease the possibility of pelvic structures injury by dissection in the submucosal endorectal plane and passing the pull-through ganglionic bowel through a "cuff" consisting of rectal musclosa and serosa then anastomosing the ganglionic segment with the anal canal one centimeter above dentate line. *Duhamel* approach consist of bringing down the normal ganglionic intestine through the bloodless plane retrorectal and joining the two walls to make a new lumen, by end to side anastomosis in which the anterior wall is aganglionic and the posterior wall is ganglionic.(**Langer**, **2012)**

In this study, we compare the outcome between the two-stage trans-abdominal *Duhamel* procedure and two stages *Soave* pull-through in the treatment of pediatric patients diagnosed with HD.

Our study aimed at comparing two-stage trans-abdominal *Duhamel* and two-stage *Soave* pull-through in managing pediatric patients having HD at the Pediatric Surgery Department, Faculty of Medicine, Alexandria University.

Patients and methods

This comparative prospective interventional clinical trial included all patients who were admitted to our Pediatric Surgery Department, Faculty of Medicine, Alexandria University starting from January 2022 to January 2023 being diagnosed as having HD with a leveling colostomy aged between 3 months old and 16 years old. Patients requiring redo procedure or who were treated with one stage procedure weren't included in our study.

Cases were categorized in a non-randomized fashion into 2 groups, the 1st one included cases who were treated by *Duhamel* procedure and Group II included patients who were treated by *Soave* pull-through. Those patients were followed up for one post-operative year.

Patients were assessed preoperatively as regard the demographic data (age, gender, and weight), laboratory tests (complete blood count, coagulation profile), and the type of stoma (Colostomy whether sigmoid or transverse, ileostomy, simple loop, divided). Also, preoperative contrast enema and biopsy (rectal or from colostomy) were recorded.

The operative records were revised, and the following data were collected: the type of the procedure whether *Soave* or *Duhamel*, the operative time, the need for a covering stoma at the end of the procedure, the need for blood transfusion, the length of the resected aganglionic

intestine as well as the result of its histopathological examination.

The postoperative follow up was done at the end of the 1st postoperative month, at the 3rd month, at the 6th month and at the end of the 1st postoperative year as regards the development of any complication as leakage, intestinal obstruction, enterocolitis, anastomotic stricture and/or constipation. Constipation and faecal continence were also scored according to the PICSS (Pediatric Incontinence Constipation Scoring System) proposed by Fichtner- Feigl et al. which is questionnaire scoring system used in the evaluation of fecal incontinence, constipation, and the ultimate scores of the PICSS were calculated from the summation of each individual value. The highest score was 32 for incontinence and indicates complete continence with a score less than 10 is considered as incontinent while score of 20-22 points considered normal for continence. The maximum score for constipation is 29 that there is no constipation, but 20-22 score is within normal. **(Fichtner-Feigl et al., 2003; Holschneider & Puri, 2008)**

Also, postoperative follow up contrast enema was done at the end of the 6th post-operative month.

Statistical analysis

Data were collected, recorded to be analysed via SPSS software version 20.0. (Armonk, NY: IBM Corp). Qualitative data were expressed using number and %. The Shapiro-Wilk test had been utilized to confirm the normality of distribution. Quantitative data were explained by range (min & max), mean ± SD, median and IQR. Statistical significance is considered if $p<0.05$.

Results

The current study included 40 patients, divided equally between the two studied groups. There were 29 male cases (72.5 %) and 11 female cases (27.5 %) with no significant differences between both groups, (**Table.1**).

The age of the studied cases ranged from 6 months to 15 years old with a significantly lower age in patients of Group II **(**10.74±7.18) than in patients of Group I (64.62±54.85), **(Table.1)**.

The weight of our patients ranged between 6 kg and 45 kg; a significantly lower weight was measured in patients of Group II (9.45±2.19) than in patients of Group I (23.30±12.39), **(Table.1).**

Table 1. Comparison between the 2 studied groups based on the demographic data

Demographic data	Group I (n = 20)		Group II (n = 20)		Test of sig.	p
	No.	%	No.	%		
Gender						
Male	14	70.0	15	75.0	χ^2=0.125	0.723
Female	6	30.0	5	25.0		
Age (months)						
Min. – Max.	6.0 –180.0		6.0 –36.0		t=4.356*	<0.001*
Mean ± SD.	64.62 ±54.85		10.74±7.18			
Median (IQR)	37.80 (24.0 –123.0)		8.0 (6.50 –12.0)			
Weight (kg)						
Min. – Max.	6.0 –45.0		6.0 –15.0		t=4.922*	<0.001*
Mean ± SD.	23.30 ± 12.39		9.45 ± 2.19			
Median (IQR)	20.0 (14.50 –34.50)		9.50 (8.0 –10.50)			

As regards the type of stoma done in the first stage; there were different types among the studied patients. (**Table.2**)

Table 2. Comparison between the 2 studied groups based on the level of stoma in the 1st stage

Type of stoma done	Group I (n = 20)		Group II (n = 20)		χ^2	p
	No.	%	No.	%		
Ascending simple loop colostomy	2	10.0	0	0.0	2.105	^{FE}p=0.487
Lower descending simple loop colostomy	1	5.0	6	30.0	4.329	^{FE}p=0.090
Right transverse simple loop colostomy	7	35.0	0	0.0	10.744*	^{FE}p=0.009*
Sigmoid simple loop colostomy	3	15.0	13	65.0	10.417*	^{FE}p=0.001*
Simple loop ileostomy	7	35.0	1	5.0	5.625*	^{FE}p=0.044*

The length of the resected aganglionic segment among our studied patients was 5-110 cm; this was significantly shorter in Group II (13.80±7.32) than in patients of Group I (65.25±33.50) as shown in (**Fig.1, Table.3).**

Table 3. Comparison between the 2 studied groups based on the length of the aganglionic segment

Length of a ganglionic segment	Group I (n = 20)	Group II (n = 20)	U	p
Min. – Max.	15.0 –110.0	5.0 –30.0	26.0*	<0.001*
Mean ± SD.	65.25 ± 33.50	13.80 ± 7.32		
Median (IQR)	75.0 (35.0 –90.0)	10.0 (10.0 –17.50)		

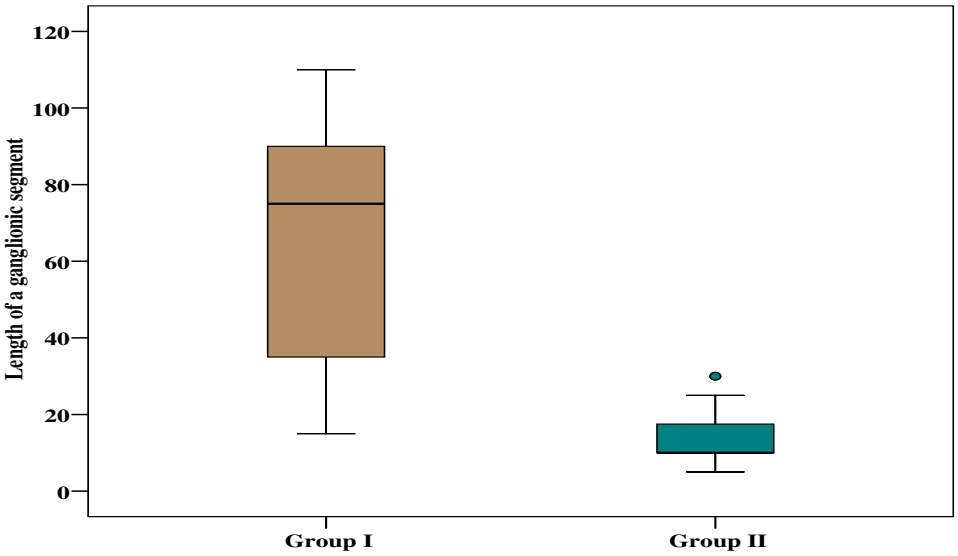

Fig.1. Comparison between the 2 studied groups based on the length of a ganglionic segment

The operative time ranged from 45-180 minutes, it was shorter in group II (80.25±19.77) than group I (105.5±27.24) with statistically significant difference between both groups, **(Table 4).**

Table 4. Comparison between the two studied groups according to operative time

Operative time (min.)	Group I (n = 20)	Group II (n = 20)	U	p
Min. – Max.	60.0 – 180.0	45.0 – 120.0	80.500*	0.001*
Mean ± SD.	105.5 ± 27.24	80.25 ± 19.77		
Median (IQR)	100.0(90.0 – 115.0)	80.0 (60.0 – 90.0)		

Blood loss ranged from 10-30 ml; the blood loss was significantly less in group II (13.50±4.62) than in group I (19.0±6.20) without the need for blood transfusion in either operation, **(Table.5).**

Table 5. Comparison between the 2 studied groups based on the amount of intra-operative blood loss

Blood loss amount (ml)	Group I (n = 20)	Group II (n = 20)	U	p
Min. – Max.	10.0 – 30.0	10.0 – 20.0	101.0*	0.005*
Mean ± SD.	19.0 ± 6.20	13.50 ± 4.62		
Median (IQR)	20.0 (15.0 – 22.50)	10.0 (10.0 – 20.0)		

A protective proximal stoma was constructed at the end of the procedure in eleven patients of Group I (55%). In contrast, no patient in Group II required proximal stoma; this was statistically significant. **(Table.6)**

Table 6. Comparison between the 2 studied groups based on temporary stoma

Temporary stoma	Group I (n = 20)		Group II (n = 20)		χ^2	p
	No.	%	No.	%		
No	9	45.0	20	100.0	15.172*	<0.001*
Yes (Simple loop ileostomy)	11	55.0	0	0.0		

The length of the resected segment in Group II (18.50±7.63) was significantly shorter than in patients of Group I (69.50±2.64), **(Table.7).**

Table 7. Comparison between the 2 studied groups based on the length of the resected segment.

Length of resected segment	Group I (n = 20)	Group II (n = 20)	U	p
Min. – Max.	20.0 –110.0	10.0 –35.0	26.0*	<0.001*
Mean ± SD.	69.50 ± 32.64	18.50 ± 7.63		
Median (IQR)	80.0 (40.0 –95.0)	15.0 (15.0 –22.50)		

As regard the postoperative complications the follow up revealed significantly higher rate of its development in Group II in comparison with Group I ($p \leq 0.05$). Anastomotic leakage was encountered in a single patient belonging to Group II. No single case showed intestinal obstruction. Also, anastomotic stricture was found by rectal examination during the postoperative follow up visits in 3 patients; all of them were in Group II and they were managed by regular dilatation. The difference between the incidence of different postoperative complications between the two groups was not significant except enterocolitis which was significantly elevated in Group II (8 patients; 40%) compared to Group I (one patient; 5%), (**Table.8**).

Table 8. Comparison between the 2 studied groups based on different parameters of post-operative complications

Variables	Group I (n = 20)		Group II (n = 20)		χ^2	^{FE}p
	No.	%	No.	%		
Leakage	0	0.0	1	5.0	1.026	1.000
Obstruction	0	0.0	0	0.0	–	–
Enterocolitis	1	5.0	8	40.0	7.025*	0.020*
Stricture	0	0.0	3	15.0	3.243	0.231
Need for dilatation	0	0.0	3	15.0	3.243	0.231

The questionnaire measuring the degree of postoperative constipation as titrated by the PICSS revealed almost the same intermediate score in both groups without significant difference. Considering that a score ranging between 20–22 points is normal; the mean of the calculated score in both groups indicated approximately normal post-operative bowel habits. **(Table.9)**.

Table 9. Comparison between the two studied groups based on constipation score

Constipation score	Group I (n = 20)	Group II (n = 20)	t	p
Min. – Max.	21.0 –25.0	21.0 –25.0	0.630	0.533
Mean ± SD.	22.0 ±1.21	22.30 ±1.75		
Median (IQR)	21.0 (21.0 –23.0)	21.0 (21.0 –24.0)		

Patients of Group I showed a significantly better level of continence (27.65±2.89) than in patients of Group II (21.95±1.85) as measured by the PICSS, **(Table .10).**

Table 10. Comparison between the 2 studied groups based on faecal continence score

Fecal continence score	Group I (n = 20)	Group II (n = 20)	t	p
Min. – Max.	20.0 –29.0	20.0 –29.0	7.432*	<0.001*
Mean ± SD.	27.65 ±2.89	21.95 ±1.85		
Median (IQR)	29.0 (29.0 –29.0)	22.0 (22.0 –22.0)		

Post-operative barium enema follow up was done only in 2 patients in each group due to incomplete patient compliance with no stricture nor dilatation, there were no statistical significance, (**Table.11**).

Table 11. Comparison between the two studied groups according to post-operative contrast study

Post-operative contrast study	Group I (n = 20)		Group II (n = 20)		χ^2	^{FE}p
	No.	%	No.	%		
Not done	18	90.0	18	90.0	0.000	1.000
Done	2	10.0	2	10.0		

Discussion

Hirschsprung's disease could be treated either in one stage or in a staged procedure with a preliminary temporary intestinal stoma. The decision whether to do a stoma or to proceed to a single-stage procedure is based upon the patient's clinical status. Also, complicated cases of HD are usually treated with a preliminary stoma with the definitive procedure taking

place after stabilization of the patient's status, also after partial normalisation of the megacolon, by resecting the aganglionic segment with stoma closure, either at the same time or later on. **(Sosnowska et al., 2016)**

Several procedures have been described as the definitive surgery in the treatment of HD; the commonly used procedure is Duhamel operation which entails a retro-rectal pull-through approach that doesn't need rectal resection. Rectal stump posterior wall is anastomosed with the anterior wall of the proximal ganglionic segment using a crushing clamp to make a wide pouch. Thus, a portion of the aganglionic rectum is kept connected with a segment of ganglionic colon to create a wide pouch.**(Mao et al., 2018)** Another procedure is *Soave* approach which includes removing the mucosa of the rectum with retaining a "cuff" of rectal muscle and the anastomosis of the proximal ganglionic segment to the anal canal one centimetre above the dentate line.**(Mao et al., 2018)**

Males are affected than females with a ratio of 4:1, however, this fact is less evident in long segment HD, as male: female ratio reaches 1:1–2:1 and in contrast it's reversed in case of total colonic aganglionosis, as this ratio becomes 0.8:1. **(Holschneider & Puri, 2008)** In our study male: female (n=29, n=11) respectively with a ratio 2.6:1.

Short segment HD refers to aganglionosis extended to the sigmoid colon that has been proved to be the commonest type of this pathology with an incidence rate varying from 57.7% to 87.1%. On the other hand, cases with long segment HD are less common, with a documented frequency of 26%. In total colonic HD the incidence fall to 2.8%. **(Taguchi et al., 2019)**

In our study the length of the aganglionic intestinal segment ranged from 15-110 cm in patient operated by *Duhamel* procedure while it ranged from 5-30 cm in patient operated by *Soave* pull-through.

The diagnosis of HD by contrast enema alone is not enough, so cases showing a normal contrast enema finding with clinical symptoms and signs of HD should have rectal biopsy to confirm or exclude the aganglionosis. **(Montalva et al., 2023)**

In our study the contrast enema was done in only 7 patients (17.5%), one in Group II and 6 in Group I as most of the studied patients were presented to our unit with a previous colostomy.

The operative duration was significantly shorter in patients treated with *Soave* procedure (80.25±19.77) than in those who were treated by *Duhamel* procedure (105.5±27.24). A longer operative duration was observed in **Tannuri et al. (2009)** study who reported 232±82.7 (range, 125-480) min in two stage *Duhamel* procedure. Also, a slightly longer operative duration was reported in two-stage *Soave* pull-through by **Shakya et al. (2010)** 133±14.18 min.

Intra-operative bleeding showed significant reduction in cases who were treated by *Soave* procedure in comparison with those who were managed by *Duhamel* procedure as the blind retro-rectal dissecting may result in in more blood loss during *Duhamel* procedure, On the contrary **Soper and Figueroa (1971)** observed a higher level of blood loss during *Soave* procedure.

The titrated blood loss was generally lower in our study in both groups than in similar studies; for example two-stage *Duhamel* procedure showed a range of blood loss of 50±0.20 (40-90 ml) in **Agarwal and Kundu (2019)** study, while in our study it was 19.0±6.20 (10.0 – 30.0 ml).

Another study carried out by **Shakya et al. (2010)** revealed 31±13 mL (20–100 mL) blood loss while it was 13.50±4.62 (10.0 – 20.0 ml) in our study.

Complications following *Soave* pull-through included leakage in a single patient, enterocolitis in 40% of operated cases and anastomotic stricture in 15% of

such patients who were treated by regular dilatations.

Askarpour et al. (2019) evaluated the results of trans-abdominal *Soave* pull-through in 160 cases of HD. The post-operative follow-up revealed the development of enterocolitis in 24 patients; 15%; being the most frequent post-operative complications in this study. Anastomotic stricture was noticed in eleven patients; 7% and anastomotic leakage in 6 patients; 4%.

Enterocolitis developed in a single patient (5%) operated by *Duhamel* procedure without any incidence of anastomotic leakage, stricture, or obstruction. A systematic review conducted for the follow-up of *Duhamel* procedure done in 161 patients revealed that enterocolitis occurred in two patients 7.45%. The rate of postoperative anastomotic stricture was none 0%, postoperative anastomotic leakage occurred in 3.03%.**(Wang et al., 2023)**

In our study the questionnaire measuring the degree of postoperative constipation as titrated by the PICSS revealed almost the same intermediate score in both groups without significant differences. The titrated score in the two groups revealed almost normal bowel habits with a mean score around 22.

However, in a study done by **Davidson et al. (2022)** cases with *Duhamel* procedure had documented constipation as a frequent post operative complain. In a study including 160 children diagnosed as HD who were subjected to *Soave* pull-through, the documented postoperative results were constipation (n=24; 15%). **(Askarpour et al., 2019)**

In our study, although patients operated by *Soave* procedure showed a calculated continence score within the range of normal levels by PICSS (21.95±1.85); it was significantly lower than the level calculated in patients who were treated by *Duhamel* procedure ((27.65±2.89).

In **Wang et al. (2023)** study the least encountered complication in two stages *Duhamel* was faecal incontinence which developed in two patients only (1%). **Askarpour et al. (2019)**documented fecal incontinence after transabdominal *Soave* procedure in 1% of patients in his study. **(Askarpour et al., 2019; Khazdouz et al., 2015)**

No rectal spur was detected in our study, however in a study carried out by *Minford JL et al.* observing the results of *Duhamel* procedure; it was seen in 6 out of 33 (18%) children, five of them had poor bowel functional outcomes. **(Minford et al., 2004)**

A study conducted by **Langer (2004)** observed that out of forty nine patients referred with marked obstructive symptoms following a pull-through for HD, 2 children presented with rectal spur which was resected later.

Conclusion

Our study showed better outcome following two stage *Duhamel* procedure regarding continence and constipation score compared to two stage *Soave* pull through, also, the rate of post-operative enterocolitis was higher in patients operated with *Soave* pull-through. We recommend *Duhamel* procedure for treatment of HD, however, a prospective randomized study including larger group of patients with longer follow up periods is recommended for confirmation.

List of abbreviations

HD: Hirschsprung's Disease
HAEC: Hirschsprung-associated enterocolitis
PICSS: Pediatric Incontinence Constipation Scoring System
IQR: Inter quartile range**,**
SD: Standard deviation**,**
χ^2: Chi square test,
t: Student t-test,
p: p value for comparing between the studied groups,
***:** Statistically significant
FE: Fisher Exact test
U: Mann Whitney test

Min: minimum
Max: maximum
N: number

Declarations

Ethics approval and consent to participate:

Parents of all the studied patients have signed an informed consent and the ethical committee of the Faculty of Medicine, Alexandria University has revised and approved our study. (Serial number: 0201678, 19-5-2022)

The corresponding author as well as the co-authors confirm that all methods were performed by the ethical standards as laid down in the Declaration of Helsinki and its later amendments or comparable ethical standards.

Consent for publication: Not applicable.

Availability of data and materials:

The datasets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: AK designed the work and performed the operation, SS revised the collected data, AM. Collected and analysed the data, AE. drafted and revised the manuscript. All authors have written and approved the manuscript and agreed to be personally accountable for their contributions and have ensured that questions related to the accuracy or integrity of any part of the work, even ones in which they were not personally involved, were appropriately investigated, resolved, and the resolution documented in the literature. AM; The corresponding author verified that all of the contributing authors have approved the manuscript before submission and that all the names and order of them were correct. He also complied with the transparency and reproducibility standards of both the field and journal by ensuring that all authors receive the submission and all substantive correspondence with editors, as well as the full reviews, verifying that all data, figures, materials (including reagents), and code, even those developed or provided by other authors.

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