

## Original Research Article

# A PROSPECTIVE AND COMPARATIVE STUDY OF FACTORS INFLUENCING THE OUTCOME OF DUHAMEL AND SOAVE PROCEDURES DONE FOR HIRSCHSPRUNG'S DISEASE

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### ABSTRACT

**Background:** To assess the factors influencing the functional outcomes of Surgery for Hirschsprung's Disease, and to compare Duhamel and Soave Pullthrough procedures based on Postoperative complications and Quality of life.

**Materials and Methods:** A total number of 30 cases were studied from 2018 to 2021 at Government General Hospital/Guntur Medical College, Guntur, and the follow-up period varied from 6 months to 18 months. This study was done to compare the results of both the Pull through procedures (Duhamel and Soave) with respect to age at definitive procedure, gender, aganglionic segment type, postoperative course, and outcomes of both procedures based on postoperative Complications and Quality of life. The analysed data were compared with other series in the literature and discussed. A master chart dealing with all aspects has been designed and presented. Statistical analysis is done with SPSS VERSION 16 and the Chi square test.

**Results:** Both the Groups was matched and the results was studied regarding Voluntary Bowel movements, Soiling, Constipation, Micturition disturbance, Quality of life, Postoperative Enterocolitis and with features of Bowel Retraction and Perineal Excoriation. Voluntary Bowel movements were present in 80% of study subjects operated by Duhamel procedure compared to 86.7% of study subjects who underwent Soave. Soiling was observed in 6.7% of study subjects who underwent the Duhamel procedure and is 20% with Soave. Constipation was observed in 6.7% of study subjects who underwent the Duhamel procedure and is 13.3% with Soave. Soiling and constipation was a major complaint following Soave surgery than with Duhamel procedure. Constipation was slightly more common in the Soave than Duhamel group (24 percent vs. 4 percent;  $p=0.04$ ), while the soiling rate was comparable in the Duhamel (21 percent) and Soave (8 percent) groups ( $p=0.26$ ). 13.3% with Duhamel procedure, and 6.7% with Soave procedure had a history of enterocolitis. Diarrhoea with explosive stool was found to be 13.3% each in study subjects who got operated by Duhamel and Soave procedures, respectively. Bloody stool was among 13.3% of study subjects who got operated by Duhamel procedure, when compared to soave it is only 6.7%. Perineal Excoriation was seen in 20% of subjects with Duhamel procedure compared to 6.7% with Soave.

**Conclusion:** The incidence of voluntary bowl movements, soiling and perennial excoriation is better in soave than Duhamel. Retraction and

constipation is more with soave. There was no significant difference between the outcomes of the two procedures, and in the light of the present findings, both the procedures appear similar in terms of efficiency and associated complications. Both the procedures have their own advantages. The Quality of life in patients who underwent Duhamel and Soave procedures appears to be almost similar in our study.

**Keywords:** Pullthrough, Constipation, Soiling, Excoriation, Micturition, Enterocolites.

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## INTRODUCTION

Hirschsprung's Disease is a developmental disorder of the intrinsic component of the Enteric nervous system that is characterized by the absence of the ganglion cells at Meissner's plexus of the submucosa and Auerbach's plexus of the muscularis in the distal bowel beginning at the internal sphincter and extending proximally for varying distance.<sup>[1]</sup> The Aganglionosis is limited to the rectosigmoid in over 80% of patients and remaining patients, the aganglionosis extends beyond and even to small bowel. Total intestinal aganglionosis with an absence of ganglion cells from the duodenum to the rectum is the rarest form of Hirschsprung's disease.

The incidence of Hirschsprung's disease is estimated to be 1 in 5000 live births and it appears to be more common in males, with a male to female 4:1, but it is less evident in long-segment where it is 1.5–2:1. Once considered a deadly disease, Surgical treatment has reduced mortality to 3% in the developed countries and is responsible for non-specific symptomatology, including chronic constipation and neonatal intestinal obstruction.<sup>[4]</sup> Patients with Hirschsprung's disease are primarily diagnosed in the neonatal period. The clinical presentation was a distended abdomen, with history of delayed passage of meconium. Older children usually presents with chronic constipation distended abdomen and failure to thrive. Approximately 10% of patients with Hirschsprung's disease present with Enterocolitis. The diagnosis relies mainly upon the histopathological examination of rectal biopsies.

Treatment of the Hirschsprung's disease aims to resect the aganglionic bowel and pull through the normal ganglionic bowel to the rectum.<sup>[5]</sup> Various pull-through procedures are described for Hirschsprung's disease with varying functional outcomes. The Surgery for Hirschsprung's disease has changed from a multistage approach to a single stage, in the recent years. It is unclear which one of the pull-through techniques yields substantially better outcomes. Most of the Paediatric surgeons stick to the surgical procedures of their choice.<sup>[7]</sup> The Duhamel procedure for Hirschsprung's disease was introduced in 1956 and the Soave procedure in 1960 and has been used since then, as a one or two stage procedure with a recent trend towards single stage procedure.

The Swenson procedure involves resection of the Aganglionic segment down to the rectum, and an oblique anastomosis is performed between the

normal colon and the distal rectum. The Duhamel procedure is designed to bring the normal colon down through a bloodless plane between the rectum and the sacrum and joining the two walls to create a new lumen, which was Aganglionic rectum anteriorly and is normally innervated colon posteriorly. Duhamel procedure has earned popularity due to technical ease, minimal anal stretching, and better visibility in the entire procedure. The Soave procedure was designed to avoid the risks of injury to pelvic structures by doing a submucosal endorectal dissection and placing the normal ganglionic bowel within Aganglionic "cuff".

## MATERIAL AND METHODS

The present study, "A comparative study of outcome of Duhamel and Soave Procedures done for Hirschsprung's Disease" is a Prospective study which has been carried out at Department of Paediatric Surgery, Government General Hospital, Guntur from 2018 to 2021. A total number of 30 cases were grouped as group D and group S, and the follow-up period varied from 6 months to 18 months. This study was done to compare the results of both the pull through procedures (Duhamel and Soave) with respect to Age at definitive procedure, Gender, Aganglionic segment type, Post-operative course, and Outcomes of both procedures based on postoperative complications and Quality of life. The analysed data were compared with other series in the literature and discussed. A master chart dealing with all aspects has been designed and presented. Statistical analysis is done with SPSS VERSION 16 and the Chi square test. Inclusion criteria was child age is more than one year, those who underwent stage I (colostomy) at our institute and as a two stage procedure. Age less than one year and those who underwent stage I procedure out of our institute was excluded. All the patient were underwent barium enema and colostomy site biopsy for ganglion cells at the time of stage I procedure.

## RESULTS

Total number of 30 cases were followed those who undergone procedures for Hirschsprung's disease from 2018 to 2021. The ages were between 12 to 24 months with a minimum follow-up period of 6 months. [Table 1]

In the total study group of 30 patients, 21 were males and 9 were female. [Table 2]

In the study group of 30 patients, Duhamel procedure was done in 15 patients where 20% were long segment cases and 80 % are short segment cases. Soave procedure was performed in 15 patients where long segment cases are 13.3 % and short segment cases are 86.7%. [Table 3]

Voluntary Bowel movements is seen in 80 % of patients who are treated by Duhamel procedure and in 86.7 % of patients treated by Soave procedure. [Table 4]

Soiling is seen in 6.7 % of patients who are treated by Duhamel procedure and in 20 % of patients treated by Soave procedure. [Table 5]

Constipation is seen in 6.7 % of patients who are treated by Duhamel procedure and in 13.3 % of patients treated by Soave procedure. [Table 6]

Micturition disturbance is seen in 6.7 % of patients who are treated by Duhamel procedure and no patients treated by Soave procedure had micturition disturbance. [Table 7]

The Quality of life between Duhamel and Soave pull-through patients is compared based on each dimension using HAQL(Parents). [Table 8]

Diarrhoea with explosive stool is seen equally (13.3% each) in both the study groups Diarrhea with foul smelling stool is seen more in patients treated with Duhamel procedure (20%) than in patients treated with Soave procedure (13.3%). Bloody stool is seen more in patients treated with Duhamel procedure (13.3%) than in patients treated with Soave procedure (6.7%). History of Enterocolitis is seen more in patients treated with Duhamel procedure (13.3%) than in patients treated with Soave procedure (6.7%). [Table 9]

Bowel Retraction is seen in none of the treated by Duhamel procedure and is seen in 1(6.7%) patient treated by Soave procedure. [Table 10]

Perineal Excoriation is seen in 20 % of patients who are treated by Duhamel procedure and in 6.7 % of patients treated by Soave procedure [Table 11]

**Table 1: Distribution based on age**

Age	Group D	Group S
12 – 18 months	5 (33.3%)	7 (46.7%)
18 – 24 months	10 (66.7%)	8 (53.3%)
Mean SD	18.93 ± 3.28	19.00 ± 3.85
Chi square test = 0.55, p=0.45 not significant		

**Table 2: Distribution Based On Gender**

Sex	Group D	Group S
Male	9 (60%)	12 (80%)
Female	6 (40%)	3 (20%)
Chi square test = 1.43, p=0.23 ( Not significant)		

**Table 3: Distribution Based On Aganglionosis Type**

Level	Group D	Group S
Long segment	3 (20%)	2 (13.3%)
Short segment	12 (80%)	13 (86.7%)
Chisquare test = 0.24 , p=0.62 ( Not significant)		

**Table 4: Voluntary Bowel Movements**

Bowel movement	Group D	Group S
Yes	12 (80%)	13 (86.7%)
No	3 (20%)	2 (13.3%)
Chisquare test =0.23 , p=0.63 (Not Statistically significant)		

**Table 5: Soiling**

SOILING	Group D	Group S
Yes	1 (6.7%)	3 (20%)
No	14 (93.3%)	12 (80%)
Chisquare test =1.15, p=0.28(Not Statistically Significant)		

**Table 6: Constipation**

Constipation	Group D	Group S
Yes	1 (6.7%)	2 (13.3%)
No	14 (93.3%)	13 (86.7%)
Chisquare test =0.37, p=0.54 ( Not Statistically significant)		

**Table 7: Micturition Disturbance**

Micturition	Group D	Group S
Yes	1 (6.7%)	0 (0%)
No	14 (93.3%)	15 (100%)
Chisquare test = 1.03 , p=0.30 ( Not significant)		

**Table 8: Quality of Life**

Quality of life	Soave	Duhamel	P value
Voluntary bowel movements	13 (86.66%)	12 (80%)	0.63
Soiling	3 (20%)	1 (6.7%)	0.28
Constipation	2 (1.3%)	1 (6.7%)	0.54
Micturition disturbances	1 (6.7%)	0 (0%)	0.30
Social functioning Mean ± SD	2.33 ± 0.48	2.53 ± 0.51	0.27
Emotional functioning Mean ± SD	2.40 ± 0.50	2.53 ± 0.51	0.48
Physical functioning Mean ± SD	2.40 ± 0.63	2.60 ± 0.50	0.53
Overall QOL	2.35 ± 0.38	2.56 ± 0.17	0.06

**Table 9: Postoperative Enterocolitis with Features**

Enterocolitis	Group D	Group S	P value
Diarrhoea with explosive stool	2 (13.3%)	2 (13.3%)	1
Diarrhoea with foul smelling stool	3 (20%)	2 (13.3%)	0.62
Bloody stool	2 (13.3%)	1 (6.7%)	0.54
Postoperative Enterocolitis	2 (13.3%)	1 (6.7%)	0.54

**Table 10: Bowel Retraction**

Bowel retraction	Group D	Group S
Yes	0 (0%)	1 (6.7%)
No	15 (100%)	14 (93.3%)

Chisquare test =2.14 , p=0.14 (Not Statistically significant)

**Table 11: Perineal Excoriation**

Perineal excoriation	Group D	Group S
Yes	3 (20%)	1 (6.7%)
No	12 (80%)	14 (93.3%)

Chisquare test =1.15 , p=0.28 (Not Statistically significant)

## DISCUSSION

The mean age of the study subjects who underwent the Duhamel procedure is 18.93±3.28 months compared to 19±3.85 months operated on by the Soave procedure. Among study subjects aged 12 – 18 months, 33.3 % were operated on by Duhamel procedure, whereas 66.7% in the age group of 18-24 months were operated on by Duhamel procedure. There appears to be statistically no significant relationship between the age and type of procedure in the current study. Whereas, in a study conducted by Bing X et al., the mean age of the study subjects was found to be 15.26±2.71 months for those who underwent soave procedure in their study, and in another study done by Parahita IG et al., 21 mean age for study subjects who underwent Soave procedure was 25.4 ± 41.0 months whereas for study subjects who underwent Duhamel procedure was 43.7 ± 48.1 months which is quite contrasting to the current study.

In the current study, among study subjects who underwent the Duhamel procedure, 60% were males, and 40% were females, whereas study subjects who underwent Soave procedure males were 80% compared to 20% females. In a study done by Parahita IG et al., 52 males and 19 females underwent Soave procedure compared to 23 males and six females operated by Duhamel procedure. Whereas, in another study conducted by Gunadi et al., study subjects underwent the Duhamel procedure. Males were 71.8%, and

females were 28.2%. Compared to this, male study subjects operated for soave procedure were 84.1%, which is similar to the current study, and females in this study accounted for 15.9%. Widyasari A et al.,<sup>[10]</sup> in their study, found that for study subjects who underwent the Duhamel procedure, 79% were males and 21% were females. For subjects who underwent the Soave procedure, the percentage of males was 92%, whereas females were 8%.

In the current study group, in patients who underwent Duhamel procedure 20% had long segment Aganglionosis whereas 80% had short segment Aganglionosis. In study subjects who underwent Soave procedure, 13.3% had long segment Aganglionosis and 86.7% had short segment Aganglionosis. Widyasari A et al.<sup>[10]</sup> in their study stated that for study subjects who underwent Duhamel procedure, 14% had long segment Aganglionosis whereas 86% had short segment aganglionosis and for study subjects who underwent Soave procedure, 8% had long segment aganglionosis compared to 92% who had short segment aganglionosis. In another study conducted by Gunadi et al.,<sup>[9]</sup> study subjects who were operated by Duhamel procedure had long segment aganglionosis of 15.4 % and short segment Aganglionosis of 84.6% and in cases operated by Soave procedure had long segment Aganglionosis of 25% and short segment Aganglionosis of 75%. The above two studies findings are quite relevant to the current study findings wherein the current study the percentage differences of study subjects having long



and short segment Aganglionosis may be accounted different geographic distributions or may be due to the genetic and race complexes.

In the current study, Voluntary Bowel movements were present in 80% of study subjects operated by Duhamel procedure compared to 86.7% of study subjects who underwent soave procedure. In a study conducted by Widyasari, A et al,<sup>[10]</sup> voluntary bowel movement was found in 93% study subjects who were operated by Duhamel procedure compared to 88% study subjects who underwent Soave procedure whereas in another study conducted by Mattioli G et al,<sup>[11]</sup> 68.5% had voluntary bowel movements who were operated for Duhamel procedure. Soiling was observed in 6.7% of study subjects who underwent the Duhamel procedure and in 20% of study subjects who underwent the Soave procedure. In a study conducted by Widyasari A et al,<sup>[10]</sup> soiling was found in 21% of study subjects who were operated by Duhamel procedure compared to 8% study subjects who underwent Soave procedure and in another study conducted by Mattioli G et al,<sup>[11]</sup> 5.3% study subjects who underwent Duhamel procedure had a complaint of soiling which is low when compared to the current study. Constipation was observed in 6.7% of study subjects who underwent the Duhamel procedure and in 13.3% of study subjects who underwent the Soave procedure. In a study conducted by Widyasari A et al,<sup>[10]</sup> the constipation rate was significantly higher in the Soave 24% than the Duhamel groups 4%. Constipation rate is higher in the Soave than Duhamel group. The risk of constipation following the Soave procedure is increased ~ 8.5-fold higher than the Duhamel procedure according to their study. He attributed this finding might be caused by an anastomotic stricture or “rolling down” of the rectal muscular cuff following the Soave procedure. In our study 30 HSCR Patients, Micturition disturbances is seen in 1 (6.7%) patient who underwent Duhamel procedure and no cases treated by Soave procedure had micturition disturbances. In a study conducted by Moore SW et al,<sup>[12]</sup> significantly lower incidence of micturition disturbances was seen in patients treated by Soave procedure than in patients treated by Duhamel procedure.

In our study of 30 HSCR Patients (Duhamel: 15 HAQL Parents vs. Soave: 15 HAQL Parents). for the quantitative study, the mean HAQL score was 2.56 and 2.35 for the Duhamel and Soave groups respectively. For the qualitative study, interviewed patient's parents expressed how their child's life had improved after surgery. However, soiling and constipation was a major complaint following Soave surgery than with patients treated by Duhamel procedure. Moore et al,<sup>[12]</sup> conducted a study on Clinical outcome and long-term Quality of life after Surgical correction of Hirschsprung's disease where One hundred seventy-eight of 330 patients were recalled after undergoing surgery for histologically proven Hirschsprung's disease (H. Results: The

Long-term functional results were comparable for Soave and Duhamel procedures. Assessment of the complications demonstrated is significantly ( $P < .01$ ) lower incidence of constipation and micturition disturbance following Soave procedure when compared to Duhamel procedure. Neurological impairment and length of an Aganglionic segment beyond the rectosigmoid area appeared to influence the functional outcome. Constipation was mainly associated with the Duhamel procedure. Functional assessment by different scoring methods showed that 86 (74.7%) of the 115 patients above the age of 4 had an excellent anorectal function and appeared well-adjusted. Twenty-two patients (19.2%) had relatively minor long-term problems, but seven (6.1%) had persistent fecal soiling resulting in psychosocial maladjustment.

In the current study Functional outcomes in Hirschsprung's disease patients after transabdominal Soave and Duhamel procedures was done in our hospital, Results are there were 30 patients (9 males and 6 females in Duhamel vs 12 males and 3 females in Soave  $p= 0.23$ ). Following Duhamel and Soave pull-through, 80% and 86.7 percent of patients, respectively, had Voluntary bowel movements ( $p=0.63$ ). Constipation was slightly more common in the Soave than the Duhamel groups (13.3 percent vs. 6.7 percent;  $p=0.04$ ), while the soiling rate was comparable in the Duhamel (6 percent) and Soave (20percent) groups ( $p=0.28$ ). Widyasari et al<sup>10</sup> conducted a study on Functional outcomes in Hirschsprung disease patients after transabdominal Soave and Duhamel procedures where A retrospective study at a Teaching Hospital. Results are, There were 53 patients (23 males and 2 females in Soave vs. 22 males and 6 females in Duhamel,  $p= 0.26$ ). Following Duhamel and Soave pull-through, 93% and 88 percent of patients, respectively, had Voluntary bowel movements ( $p=0.66$ ). Constipation was slightly more common in the Soave than Duhamel group (24 percent vs. 4 percent;  $p=0.04$ ), while the soiling rate was comparable in the Duhamel (21 percent) and Soave (8 percent) groups ( $p=0.26$ ). Furthermore, after the Soave treatment, the risk of constipation increased in female patients, which was almost statistically significant ( $p=0.05$ ).

13.3% of study subjects operated on the Duhamel procedure, and 6.7% of study subjects who underwent the Soave procedure had a history of enterocolitis. Saleh W et al,<sup>[13]</sup> in their study found that for study subjects who underwent Soave's procedure, enterocolitis was found in 8% compared to study subjects who underwent Duhamel's procedure enterocolitis 7.6%. In another study done by Vinit K Thakur, Sandip K Rahul et al,<sup>[15]</sup> the percentage of enterocolitis for Duhamel procedure study subjects was found to be 8.33%. In another study conducted by Askarpour S et al,<sup>[14]</sup> In which study subjects were subjected to soave procedure, the percentage of Enterocolitis was found to be 15%. Most of the studies were having fewer values

when compared to the current study findings. Diarrhoea with explosive stool was found to be 13.3% each in study subjects who got operated by Duhamel and Soave procedures, respectively. There is no significant association between the type of procedure performed and the presence of Diarrhoea with explosive stool in the current study. Diarrhoea with foul-smelling stool was found in 20% of study subjects who underwent the Duhamel procedure compared to 13.3% of study subjects who underwent the Soave procedure. In the present study, the occurrence of bloody stool was among 13.3% of study subjects who got operated on by Duhamel procedure. In contrast, only 6.7% of study subjects who underwent the Soave procedure had an occurrence of bloody stool in the present study. In the current study, the bowel retraction was seen among 6.7%(1 in15) of study subjects operated on by the Soave procedure. There was no retraction among all the study subjects who underwent the Duhamel procedure. Perineal Excoriation was seen in 20% of study subjects who got operated by Duhamel procedure compared to 6.7% study subjects who underwent Soave procedure. 80% of study subjects who underwent Duhamel procedure and 93.3% study subjects who got operated by soave procedure had no Perineal Excoriation. In comparison to the current study Vinit K Thakur, Sandip K Rahul et al.<sup>[15]</sup> In their study stated that perineal Excoriation was observed in 5.56% study subjects who underwent Duhamel procedure.

## CONCLUSION

The incidence of voluntary bowl movements, soiling and perennial excoriation is better in soave than Duhamel. Retraction and constipation is more with soave. There was no significant difference between the outcomes of the two procedures, and in the light of the present findings. both the procedures appear similar in terms of efficiency and associated complications. Both theprocedures have their own advantages. The Quality of life in patients who underwent Duhamel and Soave procedures appears to be almost similar in our study.

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