

## Short-Term Complications Between Duhamel and Soave Procedures in Hirschsprung Patients: A Systematic Review

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### Abstract

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Hirschsprung's disease, or congenital megacolon, is a birth defect where ganglions are absent in the gut wall, leading to constipation and intestinal obstruction. It affects 1 in 500 births worldwide and, if untreated or treated late, significantly impacts children's growth and quality of life. Understanding the short-term complications of the Soave and Duhamel methods in Hirschsprung disease patients. A systematic review was conducted based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. In our initial PubMed search and hand-searching, we identified 36 articles. After screening titles and abstracts, four were relevant for discussion. We included three articles in our systematic review after reading the full texts. Enterocolitis was more frequent with the Duhamel procedure (28%) compared to the Soave procedure (10%). The risk of Hirschsprung-associated enterocolitis (HAEC) increased in patients with a history of enterocolitis in both treatment groups. Two studies indicate that short-term complications such as constipation and bloating occur more frequently after the Soave procedure than the Duhamel procedure. Based on the three studies we discussed, HAEC was more frequent in the Duhamel procedure than in the Soave procedure, with significant differences. Constipation was more common during the soave procedure. Both procedures may cause complications such as voluntary bowel movement, soiling, and fecal incontinence, with no statistically significant differences. Minor complications in the Soave procedure include poor weight gain and strong-smelling flatulence. In the Duhamel procedure, minor complications include hardened stools and poor appetite.

**Keywords: Short-Term Complications, Hirschsprung Patients, Systematic Review.**

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## INTRODUCTION

Hirschsprung's disease, also known as congenital megacolon, is a congenital disorder characterized by the absence of ganglions in the gut wall, which causes constipation and obstruction in the intestine (Martínez et al., 2024). Hirschsprung disease is one of the most commonly found developmental disorders occurring in 1/500 births worldwide (Klein & Varga, 2020; Wang et al., 2022). Untreated or late treatment of Hirschsprung disease significantly affects children's growth and quality of life (Koo et al., 2023).

Hirschsprung disease is caused by the failure of ganglion cells to migrate to the colon during pregnancy. This congenital abnormality can obstruct the colon, most commonly affecting the rectosigmoid area (Mueller & Goldstein, 2022). However, it can be found throughout the entire large intestine and rarely affects the small intestine. Hirschsprung

disease presents significant challenges in surgery because it can lead to severe complications and even death if not promptly treated (Moore & Elhalaby, 2020; Mueller & Goldstein, 2022). The primary treatment for Hirschsprung disease is surgical therapy aimed at resecting the aganglionic segment of the intestine and restoring its function (Moore & Elhalaby, 2020).

Several surgical procedures, such as the Swenson, Soave, and Duhamel procedures, are commonly practiced to treat Hirschsprung disease (Langer, 2022). However, the laparoscopic Swenson procedure takes a longer time to perform and produces more intraoperative blood loss (Ietto et al., 2021). The most commonly used procedures are the Soave and Duhamel procedures, considered the most effective for managing this condition. In Duhamel's technique, there is minimal pelvic dissection performed, leaving the native rectum in place and creating a side-to-side colorectal anastomosis. Soave's approach involves removing the diseased mucosa and submucosa, pulling the aganglionic intestine through the muscular cuff, and performing end-to-end coloanal anastomosis (Khairi et al., 2024; Mattioli et al., 2021). Both procedures have been proven effective in reducing obstructive symptoms caused by aganglionic intestines. However, these procedures remain debated among surgeons, especially regarding short-term complications.

Current literature presents limited short-term complication evaluation comparisons between Duhamel and Soave techniques for Hirschsprung disease. Hence, in this systematic review, we want to evaluate the short-term complications that might occur in both methods. By understanding the possible complications, surgeons can minimize post-operative morbidity and optimize long-term outcomes, thereby improving the quality of life for pediatric patients with Hirschsprung disease. This systematic literature review aims to address this gap by synthesizing existing evidence and thoroughly evaluating short-term complications following the Duhamel and Soave procedures.

## **RESEARCH METHODS**

### **Literature Search**

The literature search was conducted manually using the PubMed database and hand-searching online. Synonyms, terms, and other expressions were evaluated using MeSH text terms to compile suitable keywords for the literature search. The keywords used for the literature search were: (Hirschsprung Disease) OR (Congenital Aganglionic Megacolon) AND (Duhamel Procedure) OR (Duhamel Operation) AND (Soave Procedure) OR (Soave Operation). We conducted our systematic review based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The literature search results underwent several processes, including eliminating duplicates, screening full titles, abstract screening, and full-text screening.

### **Inclusion and Exclusion Criteria(s)**

The inclusion criteria for research articles were: (1) original articles published in English; (2) studies conducted on human subjects; (3) original research papers, such as case-control studies, clinical trials, and cohort studies; (4) articles with Duhamel and Soave procedures in the title; (5) complications reported after performing Duhamel and Soave procedures; and (6) studies conducted during 2014 - 2024. The exclusion criteria for research articles were: (1) studies not conducted on human subjects (animal studies, in-vitro studies); (2) non-original

research papers, such as reviews, meta-analyses, preliminary studies, abstracts, letters to the editor; (3) duplicate studies; (4) studies written in languages other than English; (5) studies that are not accessible.

### Quality Assessment

We used the Newcastle-Ottawa Scale to assess study quality. This scale is designed to evaluate cohort studies and assess four aspects of the study: cohort selection, comparability of cohorts, and outcome assessment (Stang, 2010).

## RESULTS AND DISCUSSION

### Literature Identification

In the initial search in the PubMed database and hand-searching, we identified 36 articles. After screening the titles and abstracts, we found four articles relevant for discussion. After reading the full texts, we included three articles in our systematic review.

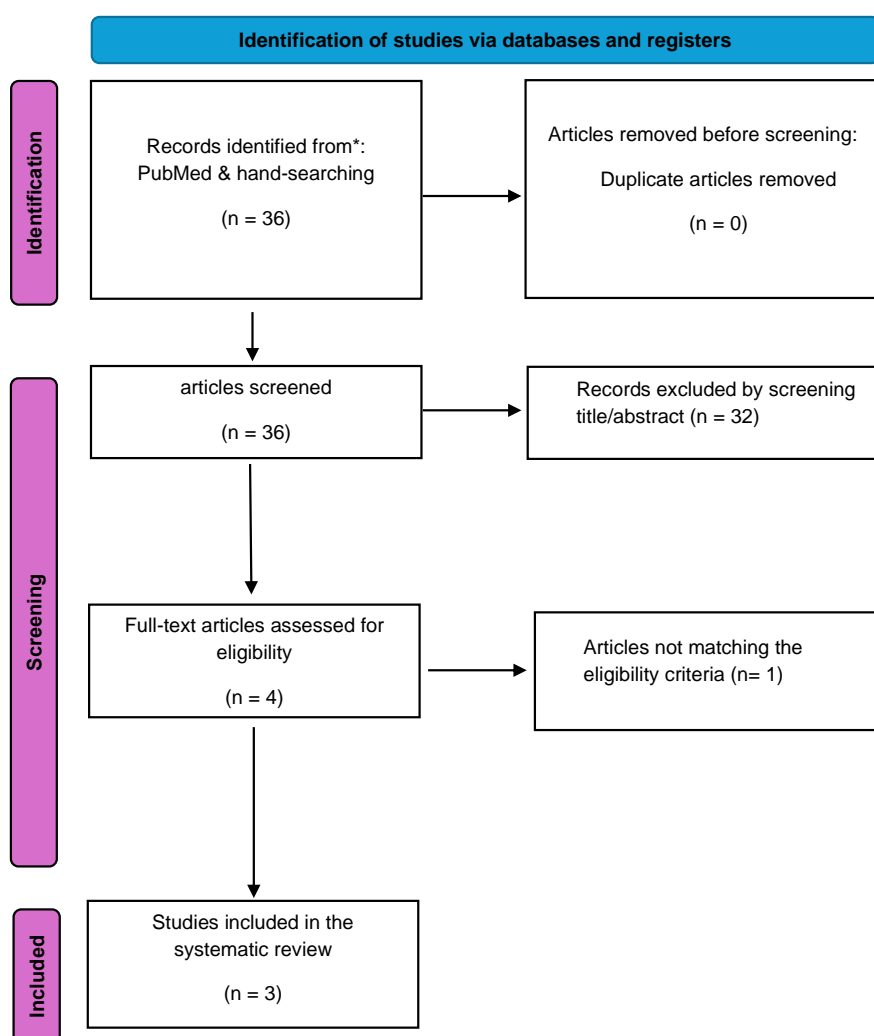


Figure 1. Preferred Reporting Items of Systematic Review and Meta-Analyses (PRISMA) Flow Diagram

### Quality Assessment

For methodological quality assessment, we utilized the Newcastle-Ottawa Quality Assessment Scale. All studies included in this systematic review scored 8 – 9 points, which

indicates that the literature included in this systematic review is of good quality.

### Study Characteristics

A total of three literatures were included in this review. The literature consists of cohort studies and was published between 2014 and 2024. We identified that the male gender (77%) was more frequent than the female gender (23%). All of the literature included in this systematic review was conducted in Indonesia. The included literature performed the Soave procedure on 101 subjects (62%) and the Duhamel procedure on 63 subjects (38%). The Duhamel procedure was performed on 54 male patients (79%) and 14 female patients (21%). Meanwhile, there was a total of 80 male patients (79%) and 21 female patients (21%) who received the Soave procedure. The population sample varied throughout the study, from paediatrics to adults.

**Table 1. Study Characteristics**

Characteristics	Soave	Duhamel
<b>Total Participants</b>	101 (62%)	63 (38%)
<b>Gender:</b>		
• <b>Male</b>	80 (79%)	54 (79%)
• <b>Female</b>	21 (21%)	14 (21%)

### Short-Term Complications Identified

One study explains that enterocolitis is a complication of Hirschsprung's disease treatment using the Soave and Duhamel techniques. Cases of enterocolitis were found more frequently with the Duhamel procedure (28%) than with the Soave procedure (10%) (Parahita & Makhmudi, 2018). In the study by Parahita et al. (2018) the risk of Hirschsprung-associated enterocolitis (HAEC) increased in patients with a history of suffering enterocolitis in both Soave and Duhamel treatment groups. Additionally, the risk of having enterocolitis increased in patients who received the Soave procedure with long-segment aganglionosis.

Two studies indicate that short-term complications such as constipation and bloating occur more frequently after the Soave procedure than the Duhamel procedure (Saysoo et al., 2020; Widyasari et al., 2018). In a study by Widyasari et al. (2018) the Soave procedure produced a higher constipation rate compared to the Duhamel procedure (24% Vs. 4%) with a statistically significant difference. Furthermore, constipation was more commonly found in female subjects who underwent the Soave technique. Hence, female subjects who received the Soave technique had a ~21.7-fold higher risk of experiencing constipation compared to male subjects (Widyasari et al., 2018). In Saysoo et al. (2020) qualitative study, the Soave and Duhamel procedures can cause constipation accompanied by hardened stools.

Various minor short-term complications can be found in both the Soave and Duhamel procedures. Patients receiving the Soave or Duhamel procedure can experience voluntary bowel movement (VBM). However, the incidence of VBM in both procedures was not statistically significant (Widyasari et al., 2018). Likewise, fecal incontinence can also happen in both procedures. Other minor complications found with the Soave procedure are bloating, poor weight gain, and foul-smelling flatulence. In the Duhamel procedure, another possible complication is a decreased appetite and hardened stool

(Saysoo et al., 2020).

**Table 2. Short-Term Complications of Soave and Duhamel Procedure**

Author	Location	Subject	Procedures	Result
(Parahita & Makhmudi, 2018)	Yogyakarta, Indonesia	Male: 75 Female: 25	Soave: 71 Duhamel: 29	<ul style="list-style-type: none"> <li>• HAEC with a significantly higher frequency after the Duhamel procedure (28%) compared to the Soave procedure (10%) (P = 0.03)</li> <li>• Increase HAEC risk in patients with long-segment aganglionosis who received Soave procedure (P = 0,015)</li> <li>• History of pre-operative enterocolitis (P &lt; 0.001) cause higher risk of developing HAEC after pull-through surgery.</li> </ul>
(Widyasari et al., 2018)	Yogyakarta, Indonesia	Male: 45, Female: 8	Soave: 25 Duhamel: 28	<ul style="list-style-type: none"> <li>• VBM: Soave (88%), Duhamel (93%)</li> <li>• Constipation: Soave (24%), Duhamel (4%)</li> <li>○ Risk of constipation increase in female patients</li> <li>• Soiling: Soave (8%), Duhamel (21%)</li> </ul>
(Saysoo et al., 2020)	Yogyakarta, Indonesia	Male: 9, Female: 2	Soave: 5 Duhamel: 6	<ul style="list-style-type: none"> <li>• Soave procedure: Frequent bloating, poor weight gain, flatulence with strong odor, and occasional fecal incontinence</li> <li>• Duhamel procedure: Hardened stools, poor appetite, and occasional fecal incontinence at night</li> </ul>

Hirschsprung disease is characterized by the lack of ganglions in Meissner's plexus and Auerbach's plexus in the rectum and extends to the intestine due to neural crest migration failure during intestinal development (Parahita & Makhmudi, 2018; Widyasari et al., 2018). Surgical intervention for Hirschsprung disease involves removing the aganglionic bowel and connecting it to the normally innervated bowel to the anus. The surgical intervention aims to remove the aganglionic colon and connect the normal innervated intestine to the rectum to restore rectum function (Furness et al., 2023). Several surgical techniques can be performed in Hirschsprung disease. Currently, the most commonly used surgical techniques are Duhamel and Soave. Duhamel's and Soave's procedures offer the advantage of preserving the innervation of the rectum and bladder (Montalva et al., 2023).

In the Soave procedure, the aganglionic rectal muscular cylinder is persevered as it will be necessary for protection during the dissection of the pelvis. Pelvic dissection is

performed to avoid contamination during muscectomy (Lefèvre & Parc, 2011). Rectal muscectomy is performed proximal to the dentate line, extending to the proximal level of the intraperitoneal rectum. Rectal muscle is separated circumferentially, and the full thickness of the rectum and sigmoid colon is removed through the anus. After identifying the transition zone, the presence of ganglion cells is confirmed through the frozen section. The rectal muscular is divided longitudinally, and the bowel was transected above the proximal normal biopsy site. The operation ended with coloanal anastomosis to the dentate line (Feldman, 2021).

The Duhamel technique's principle excludes the rectum rather than removes it. The normal innervated colon proximal side is pulled through a separation of the rectal space, and the colon will appear at the posterior side of the anal canal wall. A large enterotomy connects the excluded rectum with the pulled-through colon, creating a new rectum with an anterior aganglionic and a posterior ganglionic bowel. Histology specimens are taken utilizing frozen sections at the appropriate sites. The final decision on the extent of resecting the aganglionic colon is made according to the biopsy result. Following the resection, the surgeon performs colorectal anastomosis and removes the septum between the rectum and the ganglionic colon (Saysoo et al., 2020).

Following the surgery, patients with Hirschsprung disease can experience digestive problems. In this systematic review, we only included studies that compared short-term complications of the Soave and Duhamel procedure for Hirschsprung disease. From the three studies in this systematic review, the short-term complications that might occur include constipation, voluntary bowel movement issues, enterocolitis, foul-smelling flatulence, poor appetite, poor weight gain, fecal incontinence, and hardened stool.

One of the most severe complications is enterocolitis. The exact cause of enterocolitis is unknown and may be multifactorial. Several causes have been identified, including younger age, more extended aganglionic segment, abnormalities in chromosome anastomosis stricture, and malnutrition, which can lead to secondary infections in the intestine. It is also caused by disturbances in mucosal barrier function and changes in innate immune response (Eshel Fuhrer et al., 2024; Lewit et al., 2022). Enterocolitis is an inflammation of the digestive tract. In Hirschsprung's disease, enterocolitis is characterized by fever, abdominal distension, and diarrhea (Parahita & Makhmudi, 2018).

In one literature, the incidence of enterocolitis was higher in patients after the Duhamel procedure compared to the Soave procedure. In this systematic review, we found that the frequency of enterocolitis was higher in the group of patients who underwent the Duhamel technique (28%) compared to the Soave group (10%) (Parahita & Makhmudi, 2018). After the surgery, risk factors for developing HAEC include intestinal obstruction due to adhesive disease following the surgery and stricture or anastomotic leak (Lewit et al., 2022). Enterocolitis after the Duhamel procedure may be caused by secondary rectal achalasia due to the remaining aganglionic rectum, resulting in partial obstruction. The occurrence of HAEC in the Soave procedure is associated with an anastomotic stricture. In this study, the Soave procedure produces less HAEC than the Duhamel procedure. According to this study, the protective effect of the Soave procedure is due to fecal incontinence, a complication that commonly occurs following the Soave procedure (Parahita & Makhmudi, 2018).

Patients who experienced enterocolitis before surgery are more likely to experience

enterocolitis again after surgery. enterocolitis is due to disturbances in the gut microbiome before surgery, such as reduced *Lactobacillus* and *Bifidobacteria* species that cause reduction of immunoglobulin A secretion and protease production to inhibit endotoxin of *Clostridium Dificile* (Demehri et al., 2016; Xie et al., 2022). Additionally, alteration in short fatty acid also contributes to the occurrence of HAEC post-surgery. Other factor that contribute to the occurrence of HAEC includes long segment agangliosis. More extended segments of agangliosis cause more impairment to the bowel immune system and proximal obstruction. Hence, the intestine is more susceptible to stasis and increasing potential pathogenic bacterial multiplication (Ostertag-Hill et al., 2024; Parahita & Makhmudi, 2018).

Obstructive symptoms such as abdominal distention, vomiting, bloating, and constipation are commonly found post-operative for Hirschsprung disease (Sutthartarn et al., 2023). Constipation, mentioned by two of the included studies, is the most common short-term post-operative complication in Hirschsprung's disease. Constipation was more frequently found patients who underwent the Soave technique than in the Duhamel group. Post-operative constipation can occur due to mechanical obstruction, abnormal innervation after the surgery due to the pathology error or transitional zone pull-through, achalasia of the internal anal sphincter, and functional constipation from stool-holding behavior. According to Widyasari et al. (2018), following the Soave procedure, constipation occurs due to stricture at the rectal muscle anastomosis. In addition, residual aganglionic sleeve in the Soave procedure can remain contracted, causing a compressive action and influencing the peristaltic movement of the colon, thereby contributing to constipation post-surgery (Dickie et al., 2014). Additionally, constipation was more commonly found in female study subjects in the Soave procedure. However, more constipation was found in female study subjects due to hormonal factors (Rao, 2022). Other obstructive symptoms, such as bloating, may occur in subjects who received the Soave procedure (Saysoo et al., 2020).

There was no significant difference between VBM and soiling between the Soave and Duhamel procedures (Widyasari et al., 2018). In Saysoo et al. (2020) study, Duhamel and Soave procedure can cause fecal incontinence. In the Soave procedure, the internal anal sphincter is prone to be injured during the dissection of mucosa and submucosa, and the damage to the external sphincter and puborectalis due to vigorous anal dilation and excessive anal canal traction leads to fecal incontinence (Sun et al., 2012). However, fecal incontinence was also associated with poorer psychogenic functioning (Panicker et al., 2020). Accompanying short-term complications such as poor weight gain and flatulence with strong odor might occur after the Soave procedure. In contrast, hardened stools and poor appetite can be found after the Duhamel procedure (Saysoo et al., 2020).

This study's strength is that this is the most recent study that evaluated short-term complications of the Soave and Duhamel procedures for Hirschsprung disease. To our knowledge, this systematic review only consists of the most recent studies (2014-2024). This systematic review also includes both quantitative and qualitative studies. However, this study has various limitations. First, most included studies were designed retrospectively with small sample sizes and varying disease severity, which could introduce bias into this systematic review. Second, the included studies covered a narrow time range (2014–2024). During these years, improvements in surgical techniques, perioperative management, complication prevention, and treatment would certainly affect

the overall prognosis of patients. This could also introduce bias and affect the analysis results. Lastly, the definitions and evaluation criteria for complications were not standardized (especially for fecal continence, constipation, and enterocolitis), which could also introduce bias and affect the analysis results.

## CONCLUSION

Based on the three studies we discussed, HAEC was more frequently found in Duhamel than in the Soave procedure, with significant differences. Constipation was more commonly found in the Soave procedure than the Duhamel procedure. Both procedures may cause complications such as voluntary bowel movement, soiling, and fecal incontinence that are not statistically significantly different. The minor complications in the Soave procedure are poor weight gain and flatulence with a strong odor. In the Duhamel procedure, there are hardened stools and poor appetite.

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