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Development of postoperative obstructive symptoms in patients with Hirschsprung disease

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ABSTRACT

Hirschsprung disease (HSCR) is a rare congenital disorder of the intestine, typically diagnosed in neonates presenting with delayed passage of meconium and signs of bowel obstruction. The gold-standard and life-saving procedure is the surgical resection of the segment of the colon lacking ganglion cells. A notable number of patients, nevertheless, experience long-term postoperative complications that significantly affect quality of life. This review analyzes 315 studies published between 2013 and 2025, focusing on complications after surgical treatment for HSCR, especially fecal incontinence (reported in up to 83.3% of cases), constipation (up to 66.7%), and Hirschsprung-associated enterocolitis (HAEC) (up to 48.5%). Several factors influence treatment outcomes, including age at operation, type of surgical procedure, length of aganglionosis, one-stage or multi-stage surgery, and the presence of HAEC before the operation. Patients with associated syndromes, e.g., Down syndrome, manifest a higher risk of bowel dysfunction. Children with HSCR frequently experience a lower quality of life than their peers, specifically in continence and psychosocial well-being. In contrast, adults report slightly more favorable long-term quality of life, especially in psychological and social domains. Differences in research plans, participant numbers, diagnostic criteria, and follow-up methods contribute to discrepancies in complication rates. In conclusion, although surgical intervention is essential, long-term multidisciplinary care and standardized follow-up are key for optimizing patient outcomes and quality of life.

Keywords: Hirschsprung disease, fecal incontinence, constipation, postoperative outcomes, HAEC

1. INTRODUCTION

Hirschsprung disease (HSCR) is a congenital intestinal disorder caused by the absence of ganglion cells in the distal bowel. It occurs in approximately 1 in 5,000 live births, which makes it one of the most common causes of intestinal obstruction in newborns. The disease develops during the embryonic period, when neural crest cell migration in the intestines is disturbed. It results in an aganglionic colon without bowel movements (Faticato & Mattioli, 2019). In about 80% of cases, doctors diagnose HSCR in neonates. The main clinical symptoms include delayed

meconium passage along with bowel obstruction and Hirschsprung-associated enterocolitis (HAEC) (Morera & Nurko, 2024). Surgical intervention is the only treatment for HSCR. The three main procedures are Swenson, Soave, or Duhamel (Ralls et al., 2017). Despite the innovation of surgical treatment, up to 60% of patients develop long-term postoperative complications, including constipation, fecal incontinence, and HAEC (Munnangi et al., 2023).

2. REVIEW METHODS

We investigated available publications documenting long-term postoperative outcomes in patients with Hirschsprung disease, including clinical trials, systematic reviews, and meta-analyses. The literature review included specialized textbooks and publications in the PubMed database, using keywords such as Hirschsprung's disease, constipation, incontinence, postoperative outcomes, and HAEC. Included were English-language studies with full-text availability. The scope of this review included 315 articles published between 2013 and 2025. The PRISMA flow diagram (Figure 1) presents the selection process.

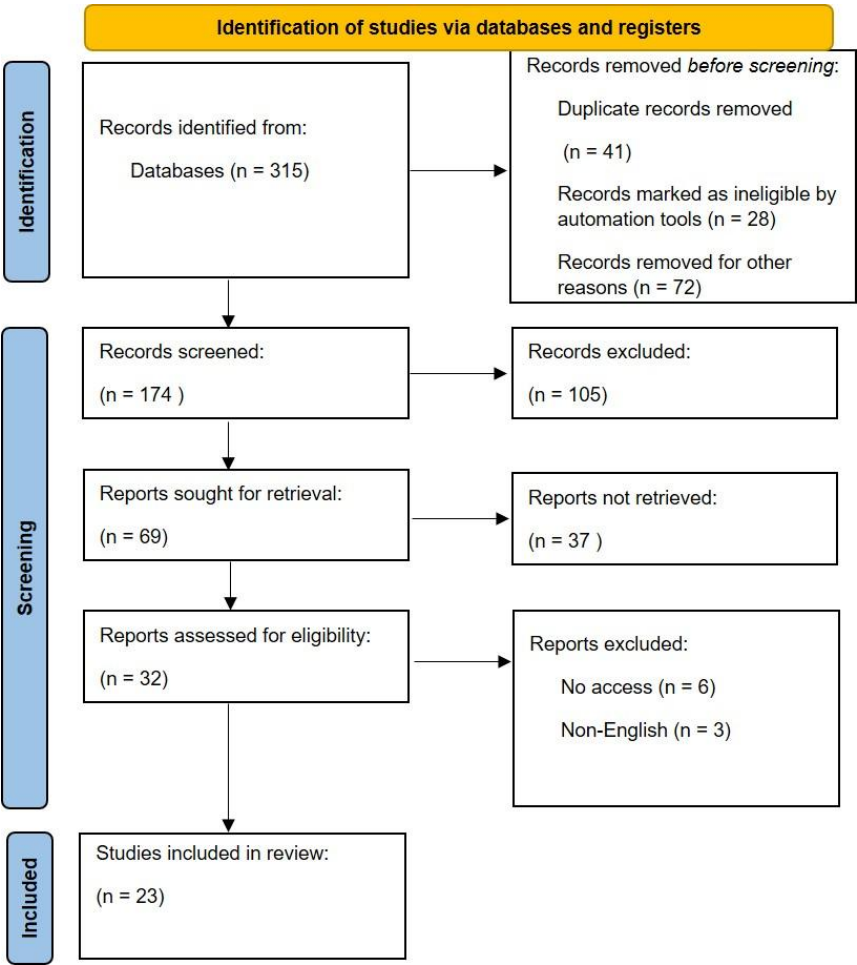


Figure 1. PRISMA flow diagram of the study selection process.

3. RESULTS AND DISCUSSION

Epidemiology

The prevalence of HSCR ranges from 1 to 2.6 per 10,000 live births. It is a sex-dependent disease, with a male-to-female ratio of 2.8-4 males per 1 female. In families with a history of HSCR, the risk of recurrence is 200 times higher than in the general population. The condition may occur as an isolated disorder in approximately 70% of cases or in association with chromosomal abnormalities such as Down syndrome and genetic syndromes, including Waardenburg and Haddad syndromes. Up to 30% of patients have additional anomalies involving the renal and urinary system (Morera & Nurko, 2024).

Pathophysiology

HSCR develops as a result of incomplete migration of the neural crest cells in the gastrointestinal tract during the 5th–12th gestational weeks. It leads to aganglionosis, which presents as a lack of intestinal motility (Faticato & Mattioli, 2019). HSCR can be classified based on the extent of aganglionosis. Short-segment HSCR, where aganglionosis does not extend beyond the sigmoid-descending colon junction, is the most common and occurs in around 72% of cases. Long-segment HSCR, develops when aganglionosis occurs beyond the sigmoid colon and descending colon junction. The most extensive forms are total colonic aganglionosis (TCA) and total intestinal aganglionosis (<20 cm of intestine has ganglion cells); the latter is the rarest type (<1% of cases) and has a high mortality rate (Montalva et al., 2023).

Diagnosis

The diagnostic process consists of three main aspects: clinical presentation, radiological findings, and histopathological confirmation. Infants with HSCR develop a complex of specific symptoms, including delayed passage of meconium beyond 24 h and bilious vomiting, along with abdominal distension and intestinal obstruction. Some newborns may show signs of enterocolitis (e.g., fever, diarrhea, and rectal bleeding). In extreme cases, intestinal obstruction and distension lead to bowel perforation (Montalva et al., 2023).

Contrast enema reveals a difference between ganglionic, dilated proximal bowel and aganglionic, narrow distal colon. Despite this, in up to 50% of newborns with TCA, the intestine may appear normal on imaging tests. To confirm the disease, a rectal biopsy is needed; the absence of ganglion cells confirms HSCR (Montalva et al., 2023). Symptoms such as abdominal distension, palpable large faecaloma, and HAEC are typical for older children with HSCR. Other signs are malnutrition and failure to thrive, anemia, and a history of chronic constipation. X-rays show fecal stasis, air-fluid levels, and a cut-off sign (Ouladsaiad, 2016).

Treatment

Surgical operation is the only definitive treatment for HSCR. The aim is to resect the aganglionic segment and restore bowel continuity by anastomosing ganglionated bowel to the anus. The most widely performed procedures include the Swenson, Soave, and Duhamel techniques (Ralls et al., 2017). Currently, transanal endorectal pull-through (TERPT) is the preferred surgical approach because it is less invasive and has lower complication rates than laparoscopic endorectal pull-through (LERPT) (Gunadi et al., 2021).

Postoperative Complications

Complications after surgical treatment for HSCR occur in nearly 60% of patients (Munnangi et al., 2023). Some of these complications, including anastomotic insufficiency, neorectal retraction, and rectal stenosis, alongside conditions such as prolonged ileus and adhesive intestinal obstruction, occur in the immediate postoperative period. Delayed complications are a more chronic issue, which manifests as constipation, fecal incontinence, Hirschsprung-associated enterocolitis (HAEC), and genitourinary dysfunction (Moore, 2016).

Hirschsprung-Associated Enterocolitis (HAEC)

HAEC affects 6–60% of patients before surgical treatment and 25–42% after. The etiology of HAEC is not fully known. The highest incidence rate occurs during the first two years of life. Clinical presentation includes abdominal distension and foul-smelling, explosive stools, along with less specific fever, lethargy, and dehydration. Untreated HAEC may lead to severe conditions, such as sepsis, intestinal perforation, and failure to thrive. The treatment includes antibiotics and rectal irrigation, as well as botulinum toxin injections into the sphincter. In extreme cases, stoma formation may be necessary (Morera & Nurko, 2024).

Fecal Incontinence

Fecal incontinence presents in around 40% of patients after surgery and occurs as the uncontrolled passage of stool. The etiology of soiling is multifactorial and includes sphincter dysfunction, abnormal rectal sensation, intestinal hypermotility, and obstruction-related mechanisms. Management depends on the underlying cause. The treatment of sphincter dysfunction and sensory abnormalities consists of a structured bowel routine and pelvic floor rehabilitation. Hypermotility is managed with dietary modification and anti-motility agents. In cases of constipation, treatment should focus on identifying and correcting the underlying obstructive pathology (Ahmad et al., 2021).

Constipation and other obstructive symptoms

Obstructive symptoms such as constipation, abdominal distension, bloating, and difficulty passing stools occur in around 8-30% of cases. Many factors cause obstruction, and treatment of this complication entails identifying them. Anatomical problems, e.g., an anastomotic stricture or a twist in the pull-through segment, are managed with repeat surgery. Similar treatment is required for obstruction created by aganglionosis of the pulled-through segment. Functional factors require alternative solutions; e.g., botulinum toxin injections can manage internal sphincter achalasia, and stoma creation may be necessary to treat a motility disorder (Ahmad et al., 2021).

Postoperative complications across studies

The Nordic study used the Bowel Function Score questionnaire to evaluate long-term bowel function in patients following transanal endorectal pull-through (TERPT) or laparoscopic endorectal pull-through (LERPT). Constipation was reported in 25% of patients, while none of the patients aged over 18 years reported this symptom. Soiling and fecal accidents were observed in 77% and 47% of participants, respectively. Both constipation and fecal incontinence were more common in younger patients. Furthermore, these complications occurred more frequently in syndromic patients, e.g., with Down syndrome ($p = 0.023$ and $p < 0.001$, respectively). One-third of patients had postoperative Hirschsprung-associated enterocolitis. HAEC occurred similarly in both sexes. Patients who had frequent fecal incontinence more often experienced HAEC ($p=0.006$). Additionally, patients reporting abdominal discomfort after bovine milk ingestion more frequently developed HAEC ($p < 0.001$) (Bjørnland et al., 2017).

A Swedish study analyzing changes in long-term postoperative complications in pediatric patients found a reduction in constipation from 41% to 14% and in laxative use from 48% to 30% over time. However, soiling rates did not change significantly. These results suggest that incontinence may be a more prevalent long-term complication compared to constipation (Granström et al., 2013).

Postoperative incontinence affected 31.3% of children, and constipation developed in 25% of patients in a Malaysian cohort. Both conditions occurred in 12.5% of patients. To obtain this information, clinicians used the Pediatric Incontinence/Constipation Scoring System. While comparing these clinical findings with anorectal manometry data, the researchers noted that the rectoanal inhibitory reflex was present in 37.5% of cases post-surgery. Despite those observations, no clinically important correlation was proven between bowel function and anorectal manometry findings (Tan et al., 2023).

In a Chinese cohort of 119 patients, clinicians identified several risk factors involved with the appearance of postoperative complications such as HAEC (27.7%), fecal incontinence (18.5%), and constipation (6.7%). The risk determinants included surgical method and one-stage operation. The Soave procedure was associated with constipation. In contrast, improved surgical techniques were associated with a lower complication rate. The extent of aganglionosis influenced the appearance of HAEC, which occurred more often in patients with long-segment HSCR (36.4%) and TCA (61.4%) (Gao et al., 2022).

Another Chinese study compared outcomes between two cohorts divided by age at surgery: less than 3 months or more than 3 months. Clinicians noted high rates of fecal incontinence in both groups (<3 months: 78.3%; 3–12 months: 83.3%). Their constipation rates did not differ significantly. In the younger group, HAEC occurred in 69.1% of participants before surgical treatment, and in 30.9% after ($p < 0.001$). Preoperative HAEC occurred in 27.1% of older children and postoperative HAEC in 28.1% of them ($p=0.867$) (Zhang et al., 2024).

A study from Thailand demonstrated bowel dysfunction in 64.95% of patients after pull-through surgery. HAEC was the most frequent complication (46.39%). According to the research, preoperative enterocolitis correlated with postoperative HAEC ($p=0.031$). The study determined that the second risk factor for HAEC development was gestational age <37 weeks at the time of surgery ($p=0.051$). Other complications, such as constipation, non-retentive and retentive incontinence, occurred in 20.62%, 22.68%, and 15.46% of participants, respectively. Patients treated using the Duhamel procedure more often had constipation and retentive fecal incontinence ($p=0.026$). Non-retentive incontinence occurred more often in children undergoing surgical treatment at an age of less than 6 months ($p=0.040$) (Surasen et al., 2024).

Evaluation using the Krickenbeck classification revealed that 18% of patients experienced grade 1 soiling after TERPT. It occurred more commonly in children undergoing surgery at ≥ 4 years of age than in those who had it earlier. Constipation occurred in 8% of patients overall. Children with complications after TERPT were significantly more prone to develop constipation ($p = 0.04$). 82% of patients achieved voluntary bowel movements (Gunadi et al., 2021).

A comparative study of surgery at age <31 days versus delayed surgery (children aged ≥31 days) found no statistically significant differences in the prevalence of HAEC, constipation, or fecal incontinence between groups, despite variation in reported rates (Kastenberget al., 2021).

Good bowel function after surgical treatment for HSCR was described by 81.1% of Iraqi patients; 14.2% of them had mild dysfunction, and 4.7% had severe dysfunction. Factors such as one-stage or multi-stage surgery influenced the development of postoperative complications (p < 0.01). Persistent constipation and HAEC occurred in 26.8% and 15% of patients, respectively, and both complications were more common after multi-stage surgery. Social anxiety developed in 19% of patients, which correlated with the occurrence of severe bowel dysfunction or persistent constipation (Aboud, 2024).

An Indonesian study found higher HAEC rates following the Duhamel procedure (28%) than following the Soave technique (10%). Patients after Soave surgery with long-segment HSCR had a higher risk of HAEC than those after the same procedure with short-segment HSCR (p=0.015). Preoperative HAEC was strongly associated with postoperative occurrence, particularly following Duhamel procedures (Parahita et al., 2018).

In an American study of patients diagnosed later in childhood with HSCR (median age during diagnosis: 3.4 years), constipation was reported as the most common persistent postoperative complication (42.9%). To manage this outcome, patients used mostly oral laxatives (39.3%), whereas only 7.1% of them were treated with suppositories. Even fewer children (3.6%) received enemas. In this cohort, HAEC and soiling were not as common (17.9% and 16.7%, respectively) (Ostertag-Hill et al., 2024).

Factors like sex, age at operation, surgical approach, and length of resection did not predispose to the development of constipation, incontinence (soiling), or HAEC, according to the Tunisian study. Constipation was reported in 16% of patients and fecal incontinence in 35% (Zitouni et al., 2025).

A large Dutch study found that fecal incontinence was considerably more frequent in patients with HSCR than in controls, especially in younger participants (37.6% vs 6.1%; p < 0.001). Constipation had a similar prevalence in both the study and control groups. Adults reported lower rates of incontinence (16.8%), suggesting that incontinence improves with age. Children more frequently used laxatives or bowel management as a treatment for constipation. Likewise, more individuals from the younger group required bowel management for incontinence. Younger age and redo surgery were identified as significant risk factors for fecal incontinence (Meinds et al., 2019). The findings of these studies are summarized in Table 1.

Table 1. Comparison of postoperative complications in patients with HSCR.

Study	Postoperative HAEC	Postoperative constipation	Postoperative fecal incontinence/soiling	Main findings
Bjørnland et al., 2017	~ 33%	25%	Soiling: 77% Fecal accidents: 47%	Constipation, soiling and fecal accidents were less common in older patients.
Granström et al., 2013	-	14%	Soiling (loose stool): 59% Soiling (solid stool): 56%	Constipation decreased over time (from 41% to 14%).
Tan et al., 2023	-	25%	31.3%	No clinically relevant correlation with anorectal manometry findings.
Gao et al., 2022	27.7%	6.7%	18.5%	HAEC was associated with long-segment HSCR and TCA; the Soave procedure was associated with constipation.
Zhang et al., 2024	Younger group: 30.9% Older group: 28.1%	-	Younger group: 78.3% Older group: 83.3%	No significant differences in postoperative outcomes between the age groups.

Surasen et al., 2024	46.39%	20.62%	Non-retentive: 22.68% Retentive: 15.46%	Preoperative HAEC correlated with postoperative HAEC. The Duhamel procedure was associated with a higher risk of constipation and retentive incontinence.
Gunadi et al., 2021	-	8%	Grade 1 soiling: 18%	Soiling occurred more commonly in older children.
Kastenberg et al., 2021	Neonatal cohort: 36.7% Delayed cohort: 48.5%	Neonatal cohort: 59.2% Delayed cohort: 66.7%	Neonatal cohort: 30.6% Delayed cohort: 15.2%	No significant differences in postoperative complication rates between the neonatal and delayed surgery groups.
Aboud, 2024	15%	26.8%	-	Complications were more common after multi-stage surgery.
Parahita et al., 2018	After the Duhamel procedure: 28% After the Soave procedure: 10%	-	-	Preoperative HAEC and the Duhamel procedure were associated with postoperative HAEC.
Ostertag-Hill et al., 2024	17.9%	42.9%	16.7%	Constipation was the most common persistent complication.
Zitouni et al., 2025	5.7%	16%	35%	No association with sex, age at surgery, approach, or resection length.
Meinds et al., 2019	-	Children: 22% Adults: 22%	Children: 37.6% Adults: 16.8%	Incontinence was less common in adults than in children.

The quality-of-life study on patients with HSCR compared with the general population shows differences between children and adults. Analysis of younger patients showed significantly lower scores in domains such as self-esteem, behavior, and general health. However, results were slightly different for children with constipation than for those with fecal incontinence. The group with HSCR and constipation had considerably lower results in all categories, and patients with HSCR and fecal incontinence had similar outcomes only in topics such as behavior and general health. In contrast, adults reported similar or even improved quality of life compared with the general population (Meinds et al., 2019).

Table 2. Comparison of postoperative bowel function in children with and without Down syndrome.

Study	Patients with HSCR and Down syndrome	Non-syndromic patients with HSCR	p-value
Kwendakwema et al., 2016	HAEC: 38%	HAEC: 41%	0.71
Pini Prato et al., 2019	Fecal incontinence: 53.3%	Fecal incontinence: 25%	0.0301
	HAEC: 42%	HAEC: 25.9%	0.1786
	Constipation: 30%	Constipation: 13.7%	0.0929

Postoperative bowel function in children with HSCR and Down syndrome was similar to that of non-syndromic children. HAEC appeared in 38% of syndromic patients and 41% of non-syndromic patients. Children after primary pull-through had a comparable incidence of HAEC to those after two-stage surgery (Kwendakwema et al., 2016). In another study comparing postoperative bowel function in children with Down syndrome to non-syndromic children, results were similar. In syndromic patients, the results were as follows: 42% reported HAEC, 30% constipation, and 53,3% fecal incontinence. In non-syndromic groups, 25,9% of children had HAEC,

13,7% constipation, and 25% fecal incontinence. Analysis showed that problems with continence were statistically more frequent in patients with Down syndrome ($p=0,0301$) (Pini Prato et al., 2019). The findings of studies comparing patients with and without Down syndrome are summarized in Table 2.

Summary

Long-term complications after surgery for HSCR occur very often. The most common is fecal incontinence (15.2%-83.3%). In the long term, incontinence is more persistent than constipation. The prevalence of constipation (6.7%-66.7%) depends strongly on surgical technique, age at surgery, and the occurrence of other postoperative complications. It is more common in younger patients. Children more often require treatment for constipation, such as enemas or laxatives.

Postoperative HAEC (10%-48.5%) is a very prevalent complication. There are many variables affecting this outcome: preoperative HAEC, redo surgeries, younger gestational age, TCA, and long-segment HSCR. Types of surgical procedures may impact long-term bowel function. Both the Soave and the Duhamel techniques are associated with a higher risk of constipation in some studies. In contrast, the Duhamel method is associated with retentive incontinence and HAEC as well. Additionally, more persistent constipation can be related to having multi-stage surgery instead of one-stage surgery. The timing of the operation, early versus delayed, did not show any obvious difference in constipation or fecal incontinence rates.

Children with Down Syndrome and HSCR have more frequent bowel dysfunction than those without. There is no higher occurrence of HAEC in syndromic patients. However, patients with Down Syndrome are more prone to fecal incontinence and constipation.

The quality of life for patients with HSCR differs between children and adults. Younger patients have lower scores on domains such as self-esteem, general health, and behavior than the general population. However, adults with HSCR had higher scores in domains such as overall quality of life, physical or psychological health, and social relationships than their controls.

4. CONCLUSION

While fecal incontinence, constipation, and Hirschsprung-associated enterocolitis are common long-term complications of surgical treatment for Hirschsprung disease, research shows a wide spread in their frequency across individual studies. This wide gap is indicative of differences in sample size, diagnostic criteria, study design, and follow-up methods. A variety of factors modify postoperative outcomes, including age at surgery, type of procedure, extent of aganglionosis, and the need for multi-stage operations. Preoperative HAEC and redo surgeries are strongly associated with an increased risk of postoperative complications. Constipation often improves over time and becomes easier to manage. All complications can considerably affect the quality of life. Therefore, standardized research approaches, long-term multidisciplinary follow-up, and advanced patient education are essential to optimize outcomes in patients with HSCR. Moreover, larger, more precise studies are needed to identify the exact causes of postoperative complications and determine the most effective way to prevent them.

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Informed consent

Not applicable.

Ethical approval

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Conflict of interest

The authors declare that they have no conflicts of interest, competing financial interests or personal relationships that could have influenced the work reported in this paper.

Data and materials availability

All data associated with this study will be available based on the reasonable request to corresponding author.

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