Original Article

Short-term and Long-term Predicted Outcomes after Swenson's Pull-through Procedure for Hirschsprung Disease

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Abstract

Background: Hirschsprung disease (HD) is a congenital deficiency of myenteric and parasympathetic ganglion cells in the last portion of the bowel, resulting in a loss of function and consequently causing intestinal blockage and clinical symptoms of a distended abdomen. **Objectives:** This study aimed to follow up and investigate the early and late complications of patients diagnosed with HD after definitive surgical treatment by Swenson's technique. **Materials and Methods:**A prospective cross-sectional study was conducted on 38 patients ranging from neonates to adolescents, diagnosed with HD and who underwent Swenson's technique. The initial suspicion of HD by clinical features, and the final diagnosis by rectal and colonic biopsies, and contrast enemas could help in the diagnosis of HD. Postoperative care included short-term and long-term follow-up after Swenson's pull procedure for HD. The extension of aganglionosis in HD was evaluated to determine the type of HD. **Results:** The results of this study revealed that 38 patients diagnosed with HD had a male-to-female ratio of 3.2: 1. The larger age group of patients (23, 60.5%) was infants. Constipation, delayed meconium, and abdominal distension were expressed in 23 (60.5%), 15 (39.5%), and 38 (100%) cases, respectively. Short-segment type had a higher frequency of 31 (81.6%). The wound infection formed a major earlier complication in 15 (39.5%) patients. Later complications were postoperative enterocolitis in 14 (36.8%) patients. Constipation represented in 8 (21.1%) patients and fecal incontinence in 7 (18.4%) patients. Perianal excoriation was seen in 19 (50%) patients, 27 (71.1%) patients suffered from abnormal foul-smelling stool odor, and 13 (34.2%) patients had abnormal bowel sounds and abdominal distension. **Conclusion:** Mostly, HD is diagnosed at an age <1 year, and males are the most predominant sex at diagnosis. Most short-term complications are related to wound infection and pus formation, whereas long-term complications are related to efficiency for good fecal continence, constipation, enterocolitis, and behavioral issues.

Keywords: Hirschsprung disease, postoperative outcomes, Swenson's pull-through procedure

Introduction

Hirschsprung disease (HD) is a congenital deficiency of myenteric and submucosal nerve supply (parasympathetic ganglion cells) in the last portion of the bowel resulting in loss of function and consequently intestinal blockage[1] and clinical symptoms of a distended abdomen.[2]

HD is classified into short-segment HD, which involves a deficiency of ganglion nerve cells in the rectum and sigmoid parts of the colon, long-segment HD, which involves a deficiency of ganglion nerve cells in parts of the colon before the sigmoid colon,[2] and total-segment HD, which involves a deficiency of ganglion nerve cells in all parts of the colon and part of the small intestine (<50 cm before the ileocecal valve).[3]

HD is embryologically related to the absence of migration of cells from the neural crest to the large bowel resulting in intestinal blockage, and this represents a predominant etiology of intestinal blockage in the newborn age group.[4,5] HD involves various levels of the

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distal part of the intestine, starting from the internal anal sphincter.[6]

The sex distribution is four males to one female in HD, and the type of treatment ranges from colostomy in newborn babies to final surgical repair, which is done as the children get older with increasing age due to gaining more body weight.[7]

The prevalence is 1/5000 newborn births, and a wide range of symptoms, particularly in the newborn age group, extending from enlargement of the abdomen, difficulty in passing meconium defecation, problems receiving the proper amount of feeding, and vomiting with or without bile.[8]

The most important diagnostic procedure for HD is a rectal biopsy, which should include thick specimens to ensure a full-layer thickness sample.[8] Definitive treatment is by surgery, including many different types of surgical procedures for treatment (Swenson, Duhamel, and Soave), modern surgical procedure for rectosigmoid HD involves the surgical removal of the diseased part of the intestine and its replacement by pulling the healthy part, commonly done through an endorectal pull-through (ERPT) that could be performed by total transanal or laparoscopic ERPT.[9]

There is a higher risk of postoperative complications in HD, which can reach 60%.[10] However, early complications may include fecal leakage, loose and watery stools, which may take numerous months to return to normal,[11] and perianal itching.[12] While later complications are the inability of the internal anal sphincter to relax, stenosis occurs in intestinal anastomoses, some parts of intestinal aganglionosis, incontinence of feces, and lastly, recurring enterocolitis,[13-17] which carries a higher risk of mortality in the HD pediatric age group.[18-20]

The term "functional outcome" is related to the effect of the pull-through post-operation, which means the efficiency for good fecal continence, community behavior, and this effect for a longer period on the life behavior of children.[12,21]

This study aimed to follow up and investigate the early and late complications for patients diagnosed with HD after definitive surgical treatment by Swenson's technique.

Materials and Methods

Study design and patients

A prospective cross-sectional study was conducted on patients ranging from neonates to adolescents who were diagnosed with HD and underwent Swenson's technique between September 2018 and June 2021 in the Pediatric Surgical Unit of the Babylon Maternity and Pediatric Teaching Hospital, Babylon, Iraq.

A total of 38 patients diagnosed with HD ranging in age between 1 month and 14 years, who visited the clinic of Babylon Maternity and Pediatric Teaching Hospital and had gastrointestinal features suggestive of having suspicious of this disease such as enlargement of the abdomen, difficulty in passing meconium, and problems receiving the proper amount of feeding, especially for newborns.

Rectal colonic biopsies were regarded as an important diagnostic procedure in HD and should be obtained to obtain full-layer thickness samples. In addition, contrast enemas could also help in the diagnosis of HD.

Preoperative assessment included previous surgical, drug, and medical information, whereas nutritional status and age at presentation concerning the presence of other illnesses represented the most significant risk factors to be evaluated before surgery.

Postoperative care included short-term and long-term follow-up after Swenson's pull-through procedure for HD. The extension of aganglionosis in HD was evaluated to determine the type of HD. Presence of early and late complications included abscess or infection at the surgical site, occurrence of perianal excoriation or fistula or leakage from the anastomotic operation site, gastrointestinal abnormal function like abnormal sounds, constipation, incontinence, enterocolitis, and obstruction.

Inclusion and exclusion criteria

Inclusion criteria included newborns who should continue to keep track of earlier and later complications that were established after a serial process of Swenson's pull-through technique as a definitive procedure for HD, and the patients are below 15 years of age. Exclusion criteria should mean follow-up is 3 years, so all patients who lost follow-up before 3 years are excluded from the study.

Statistical analysis

The statistical software for the Statistical Package for the Social Sciences version 27.0 (IBM Company, Chicago, IL, USA) was used for the description, analysis, and display of the data.

Ethical approval

All individuals involved in this study were informed, and consent was obtained verbally from parents before the collection of samples. The study was approved by the Committee on Publication Ethics at the Babylon Health Directorate under reference number 0899 on August 12, 2023.

Results

A total of 38 patients diagnosed with HD were divided into 23 (60.5%) infants below 1 year old and the remaining 15 (39.5%) children exceeding 1 year old; 76.30% were

Table 1: Preoperative clinical presentation of patients with Hirschsprung disease (HD)

Total patients with HD (n = 38)			
Parameters		***N***	**%**
Sex	Male	29	76.30
	Female	9	23.70
Age (years)	<1	23	60.50
	1–5	6	15.80
	6–10	7	18.40
	11–14	2	5.30
Congenital malformation		2	5.30
Delayed meconium		15	39.5
Abdominal distension		38	100
Constipation		23	60.5
Vomiting		15	39.50

Table 2: Type of Hirschsprung disease (HD)

Total patients with HD (n = 38)		
Parameters	***N***	**%**
Short-segment	31	81.60
Long segment	6	15.80
Total colonic aganglionosis	1	2.60

male and 23.7% were female, with a male-to-female ratio of 3.2:1.

The larger age group comprising 23 (60.5%) were infant babies, remaining 6 (15.8), 7 (18.4%), and 2 (5.3%) patients represented the 1–5, 6–10, and 11–14 years age groups, respectively. Constipation, delayed meconium, and abdominal distension were the more common clinical presentations at the time of diagnosis, expressed in 23 (60.5%), 15 (39.5%), and 38 (100%) patients, respectively, whereas vomiting and congenital anomalies were less common, found in 15 (39.5%) and 2 (5.3%) patients, respectively. One case had one case of congenital heart disease in Down syndrome, and one case had congenital vesicoureteric reflux with renal failure as illustrated in Table 1.

Rectal biopsy was performed at the time of colostomy, and short-segment aganglionosis showed more percentage of 31 (81.6%), whereas long-segment and total colonic aganglionosis were shown in 6 (15.8%) and 1 (2.6%) patients, respectively [Table 2].

The wound infection formed a major earlier complication 15 (39.5%), whereas only one case (2.6%) each experienced pus discharge from the rectum, perianal, fistula, and anastomotic leakage [Table 3].

However, later complications needed follow-up for 3 years period. Postoperative enterocolitis had a high frequency in 14 (36.8%) patients. Postoperative constipation was represented in 8 (21.1%) patients. Fecal incontinence was observed in 7 (18.4%) patients. The mortality occurred

Table 3: Early postoperative complications

Outcomes	***N***	**%**
Wound infection	15	39.50
Pus discharge from the rectum	1	2.60
Perianal fistula	1	2.60
Anastomotic leakage	1	2.60

Table 4: Later postoperative complications

Total patients with Hirschsprung disease (n = 38)		
Parameters	***N***	**%**
Mean follow-up time	3 years	
Postoperative enterocolitis	14	36.80
Perianal excoriation	19	50
Postoperative constipation	8	21.10
Fecal incontinence	7	18.4
Mortality	2	5.30
Abnormal bowel sound and abdominal distension	13	34.20
Redo pull-through	2	5.30
Adhesive intestinal obstruction	2	5.30

in 2 (5.3%) patients due to the presence of one due to congenital heart disease in Down syndrome and the other due to renal abnormalities with vesicoureteric reflux. Adhesive intestinal obstruction occurred in 2 (5.3%) patients.

Additionally, 2 (5.3%) patients did not improve after a long period and had continued clinical symptoms of megacolon, requiring should be managed by Redo pull-through management. Perianal excoriation showed in 19 (50%) patients, and 13 (34.2%) patients experienced abnormal bowel sounds and abdominal distension as shown in Table 4.

Discussion

In this study, most of the patients with HD were infants. About 22 (60.5%) out of 38 children had HD below the age of 1 year old, whereas the remaining 15 patients were more than 1 year old. They were undergoing Swenson's technique, which involved an earlier colostomy during the initial presentation of the diagnosis then colectomy for the abnormal aganglionic affected part, and then closure of the colostomy to establish a normal passage of stool from the anal region. Age presentation was >90% in the earlier age group and mostly found in the neonatal age group.[22] Raghunath *et al*.[23] exhibited an age group that was most newborns or early infant age group 84.4%, postsurgical constipation showed 38.5% and was treated by enema and laxative, and fecal soiling 30.8%.

In addition, in the current study, males showed 3.2 times more than females, in addition to more presenting clinical features such as 39.5% delayed meconium, 100%

abdominal distension, 60.5% constipation, in addition to 39.5% vomiting and two cases of congenital malformation including one case with Down syndrome and another case with congenital vesicoureteric reflux.

Joseph Mabula *et al*.[24] showed an age presentation between 7 days and 10 years, with 5.5% of the neonatal age group and 94.5% of the age group > 1 year old, with a male-to-female ratio of 3.6:1, and clinical presentation like constipation in 94.5%, abdominal distention in 92.7%, vomiting in 22.7%, and one case with Down syndrome.

Histopathological readings are an excellent diagnostic parameter to make the final decision about HD,[25] with many entities regarding rectal biopsy,[26] and provide higher and more accurate results than contrast enema for the diagnosis of HD.[27]

During HD colostomy operation in this study, multiple intestinal biopsies were taken from various parts of the large intestine and sent for histopathological diagnosis type of HD disease, where the short-segment had a higher percentage of 81.6%, whereas the long segment and total colonic aganglionosis were found 15.8% and 2.6%, respectively.

Joseph Mabula *et al*.[24] showed that the anatomical distribution of short-term, long-term, and total colonic aganglionosis segments presented 67.3%, 16.4%, and 0.9%, respectively.

This current study, after Swenson's pull-through technique, within 2 days, should follow the starting of intestinal movement and then begin food ingestion, as these are considered the most crucial points after surgery. The most common wound infection formed a major earlier complication in 39.5%, whereas only 2.6% had presented with pus discharge from the rectum, perianal fistula, or/and anastomosis leakage.

However, the results revealed that long-term complications continued for 3 years, and postoperative enterocolitis had a high frequency of 36.8% and could be improved by using intravenous fluid, electrolyte balance, and antibiotic therapy. Postsurgical constipation represented 21.1% and could be managed by using oral laxatives and dietary modifications.

Fecal incontinence occurred in 18.4% and was treated via washing 1 day by enema and improvement. After the operation, 5.3% of cases of death occurred after 1 and 6 months due to congenital heart disease in Down syndrome and other congenital vesicoureteric reflux with renal failure, respectively. Adhesive intestinal obstruction occurred in 2 (5.3%) patients and should be treated by open laparotomy. The results also revealed that 5.3% of the patients did not recover well after the procedure and had clinical symptoms of megacolon, which should be managed by a redo pull-through. Perianal excoriation was shown in 50% and 34.2% of the patients, who had abnormal bowel sounds and abdominal distension.

Fecal continence is an important aim in HD surgery to restore normal bowel evacuation. In the bowel,[28] good results with normal fecal continence were about 87.8% in Swenson's surgery and 42.9% in Duhamel's surgery. However, there were some cases of fecal incontinence ranging from 1% to 32.7%.[29-31]

Muhammad Saleemb *et al*.[32] revealed that postoperative enterocolitis occurred in 26.6%, Postsurgical constipation in 16.1%, fecal incontinence in 37.5%%, wound infection in 9.4%, anatomical leak and perianal fistula in 3.1%, pus from the rectum, and adhesive intestinal obstruction in 4.7%.

Chien-Chung Lee *et al*.[33] showed that short-segment had a higher percentage of 70.4%%, whereas long-segment and total colonic aganglionosis were found in 11.3% and 18.18% of cases. Delayed passage of meconium occurred in 29.54%, preoperative constipation in 52.27%, adhesive intestinal obstruction in 13.63%, anastomosis leakage and entero-cutaneous fistula in 6.8%, perianal excoriation in 70.45, postoperative constipation in 15.9%, and death in 13.63%.

In addition, enterocolitis commonly occurs through a period of 2 years from the beginning of the operation, with a distribution of about 18% in the perioperative period, but about 30% in association with suffering from this illness and death. Patients were presented with a swollen abdomen, diarrhea, lethargy, and high temperature.[34,35] Enterocolitis can lead to functional blockade of the large intestine.[36]

The mechanism of HD is not well understood yet and it might occur due to multiple factors such as bowel blockage, inheritance, widespread microbial infections,[34,35] and preoperative malnutrition.[37]

Enterocolitis can be prevented by prophylactic injection of botulinum and irrigation of the rectum,[34,38] and there is a supposition that anorectal dilatation procedure has a role in decreasing the frequency and intensity of enterocolitis with inhibition of anal stenosis.[39]

Many researchers have supposed that incontinence results from injury to the anal sphincter,[40,41] which might result from laparoscopic surgery[42] or a second operation that might lead to a weak anastomosis within the anal tract or due to a lack of an anal tract (partial or total loss of anus) after the pull-through procedure.[43]

Conclusion

Mostly, HD is diagnosed at an age <1 year old, and males are the predominant sex at diagnosis. The most common type is short-segment, whereas long-segment and total colonic aganglionosis are of lesser extent. Most short-term

complications are related to wound infection and pus formation, whereas long-term complications are related to efficiency for good fecal continence, constipation, enterocolitis, and behavioral issues.

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Conflicts of interest
There are no conflicts of interest.

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