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Evaluation and treatment of the patient with Hirschsprung disease who is not doing well after a pull-through procedure

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KEYWORDS
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Enterocolitis;
Obstruction;
Incontinence;
Soiling;
Bowel management

Ideally, after operative management of Hirschsprung disease, a child should thrive, avoid recurrent episodes of abdominal distention and enterocolitis, and be fecally continent. However, there is a small group of patients that do not do well after their pull-through procedure. The purpose of this article is to describe our algorithm for the work-up and management of the post pull-through patient with Hirschsprung disease who is not doing well. These children can be categorized into 2 distinct groups: (1) those who are soiling, and (2) those who suffer from distention and enterocolitis. Both of these patient types can be systematically treated with a combination of bowel management, dietary changes, and laxatives, and, potentially, a redo operation, with the goal of having a clean, and happy child.

The operative management of Hirschsprung disease has evolved since first described by Orvar Swenson in 1948,[1] including various surgical procedures involving a full-thickness rectosigmoid dissection (Swenson), an endorectal dissection (Soave), a retrorectal pouch procedure (Duhamel), and a transanal pull-through.[2-5] Procedures are now less invasive, often primary repairs are done compared with previous 2 or 3 stage procedures, and laparoscopy is utilized.[4,6-10] Each of the surgical procedures developed involve removing the aganglionic bowel, pulling through ganglionic bowel, while preserving the anal canal and sphincter mechanism, and thus preserving continence.

There is very little published on the long-term results after operative management of Hirschsprung disease. Outcomes are likely varied given differences in the surgery performed, the age of treatment, the involved aganglionic segment, the amount of colon resected, and the patient's and parent's perception of fecal continence.[4,11-13] The most commonly reported long-term functional problems after definitive surgical management of Hirschsprung disease are constipation, fecal incontinence, and enterocolitis. The incidence of each is unknown and their definitions vary widely. If these issues are not dealt with, they can have a considerable effect on the child's overall well-being, as well as their physical, emotional, and social development.[11]

We reviewed our series of patients with Hirschsprung disease who had a pull-through performed at other institutions, and presented to us thereafter with different postoperative problems. Based on this experience, we have developed an algorithm for the investigation and treatment, including surgical intervention, of a child with Hirschsprung disease who is not doing well after operative management.

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Initial evaluation

Our protocol when a patient is not doing well after surgery for Hirschsprung disease begins with a detailed history and physical examination. In particular, in the history, it is important to investigate the patient's bowel habits, such as any history of constipation, enterocolitis, abdominal distention, failure to thrive, incontinence, use of antimotility agents, such as loperamide, laxative use, or need for dilations or irrigations. In addition, it is vital to know the type of surgery that was previously performed.

We perform a water-soluble contrast enema on all patients. This allows us to visualize the anatomy that exists after the initial operation, and gives us insight into the motility of the colon and any structural anomalies that may account for the patient's symptoms.

All of these patients then have an examination under anesthesia. This inspection looks for (1) the integrity of the anal canal, (2) the presence of a stricture, (3) the status of the sphincters, (4) the presence of a large rectal pouch, or (5) the presence of a palpable Soave cuff. A full-thickness rectal biopsy is also performed for histologic evaluation for patients with obstructive symptoms.

Categorization of the patient with Hirschsprung disease who is not doing well after pull-through procedure

There are 2 distinct subsets of patients we have identified who are not doing well after their operation for Hirschsprung disease. These include (1) those who are soiling, or fecally incontinent, and (2) those patients who are distended, have repeat episodes of enterocolitis, and may have failure to thrive.

The Hirschsprung patient who is soiling

Ideally, after operative management of Hirschsprung disease, fecal incontinence should not occur as the patient is born with a normal continence mechanism; normal anal canal sensation and normal sphincters. We define continence as the ability to have voluntary bowel movements without soiling and without the need for enemas. For this to occur, there must be normal anal sensation, voluntary sphincter control, and appropriate colonic motility. An abnormality in any of these components can lead to partial or total incontinence. Pseudo-incontinence is an entity in which all the physiological mechanisms for continence are intact, but the child continues to soil. Such a patient has the potential for bowel control with the appropriate medical therapy.

It is extremely important to evaluate each of these patients systematically. After the history and physical examination, the anus must be examined closely, usually under anesthesia, to determine whether the anal canal is intact or disrupted, and whether the anus itself is open or closed (Figure 1). Disruption of the dentate line from a dissection started too low or damage to the anal sphincters as a result of excessive stretching will result in decreased anal sensation and loss of voluntary sphincter control. In addition, the contrast enema will disclose whether the colon is dilated or nondilated. From this image, we can infer whether the colon is hypomotile or hypermotile.

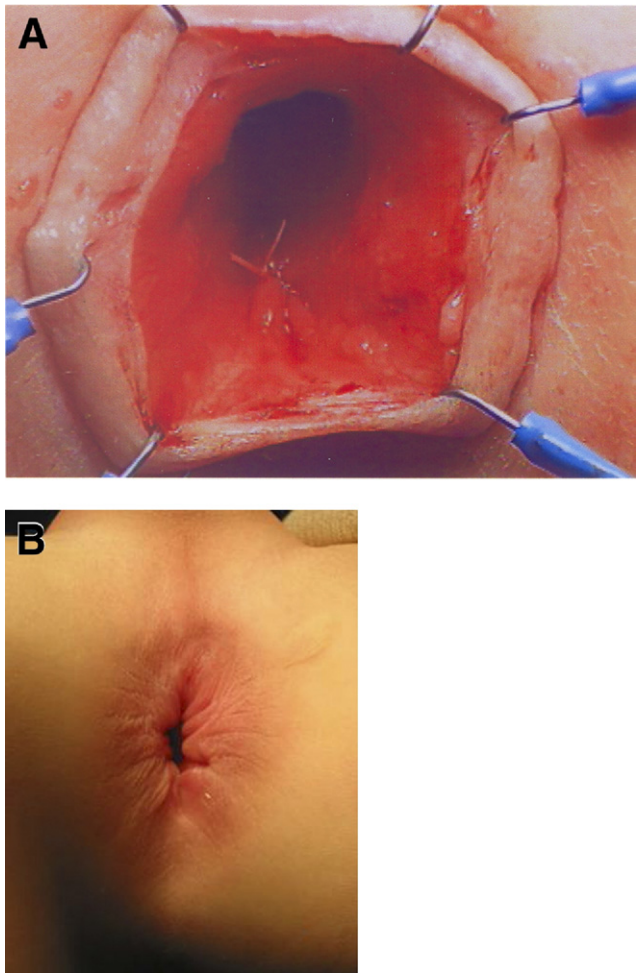


Figure 1 (A) Loss of the anal canal, there is no visible dentate line, meaning that the pull through procedure dissection was begun too low. (Reprinted with permission.[16]) (B) Destruction of sphincter mechanism leaving an "open anus," likely from excessive stretching during the dissection.

Children with a large dilated colon on contrast study and a history of constipation are considered hypomotile (Figure 2A). This group of patients is treated with a daily, senna-based, laxative. The amount of senna administered is titrated with a "laxative trial" in which daily abdominal radiographs are taken to determine whether the colon has emptied with the previous day's laxative. Through trial and error, we adjust the amount of laxatives so the child has 1 to 2 soft bowel movements a day, with no accidents, and a radiograph showing a colon empty of stool. Those patients achieving this goal, in retrospect, were pseudoincontinent, and only had difficulty because of overwhelming constipation. The

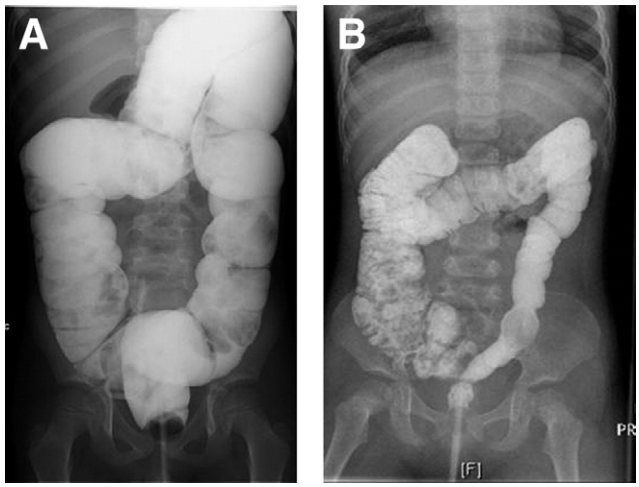


Figure 2 (A) Contrast enema of a dilated hypo motile colon. (B) Contrast enema of a nondilated hypermotile colon. (Reprinted from Levitt MA, Peña A. Fecal Incontinence and Constipation, in Pediatric Surgery (ed 5), with permission from Elsevier.)

patients who fail this laxative trial (continue to soil even though radiologically the colon is empty) are truly incontinent. These patients require daily bowel management with administration of large volume (500–750 mL), saline-based enemas.[15]

Children with a nondilated or normal caliber colon are considered hypermotile (Figure 2B). This group of patients is initially managed with loperamide, pectin, and dietary modifications that include limiting meals to 3 per day, no snacks, and encouraging of constipating foods. If a patient is able to achieve continence with this regimen, they are also considered to have been pseudoincontinent. For such children, changing the motility of the colon and subsequently the consistency of their stool, allows them to have voluntary bowel control. However, the patients in this group who are unable to have voluntary bowel movements require bowel management consisting of a combination of a daily small volume enema (200-500 mL), slowing the colon down with medications, such as loperamide, and firming up the consistency of the stool with pectin and a constipating diet.

If patients have incontinence despite medical treatment and have an intact anal canal and sphincters, enemas are given and then a trial of medical management without enemas is offered every 6-12 months, because their anatomical features give them the potential for bowel control. Once again, colonic motility and stool consistency are manipulated so the child will have 1 to 2 well-formed stools per day, which gives them the best possible chance of achieving voluntary bowel control. Because the child now knows what it feels like to be clean after daily enemas, there may be an increased awareness on his/her part to achieve continence. By contrast, those patients with a damaged or absent anal canal or damaged sphincters are maintained on bowel management with enemas, and are good candidates for a Malone appendicostomy.

In our previously published results, we reviewed 130 patients referred to our institution's bowel management program with soiling after operative management of their Hirschsprung disease.[16] Of 130 patients in this group, 68 had primary complaints of soiling and 33 had dilated colons on their contrast enema and had a history of constipation. After initial treatment with laxatives, 6 patients became continent on laxatives alone and were considered pseudoincontinent. and 27 did not respond to laxatives and went on to bowel management with large-volume enemas. Eighty-five percent (23/27) of them were clean with daily enemas.

Of the 68 patients who were soiling, 35 had a nondilated colon on their contrast enema and had a tendency toward diarrhea. These patients were managed with loperamide, pectin, and a constipating diet. Six of these patients went on to have voluntary bowel movements and no longer soiled, and therefore were recategorized as pseudoincontinent. Of the remaining patients, 79% (23/29) became clean with the addition of a small-volume enema added to the loperamide, pectin, and the constipating diet.

In the initial evaluation of these patients, 5 of the 68 patients (7%) had a damaged anal canal (Figure 1), all of whom had true incontinence requiring long-term bowel management with enemas. This stresses the importance of maintaining the integrity of the anal canal and sphincters during the initial operation to preserve both anal canal sensation and the sphincter mechanism.

The Hirschsprung patient with distention and enterocolitis post pull-through procedure

Abdominal distention after operative management of Hirschsprung disease can be due to either a pathological or an anatomical problem. A possible sequela of patients prone to abdominal distention is enterocolitis. Although the exact cause of enterocolitis is unknown, certain factors, such as fecal stasis, can precipitate and aggravate the problem. It is hypothesized that the stasis can result in proliferation and mucosal invasion by intraluminal flora resulting in a local and systemic inflammatory response. The clinical criteria for enterocolitis include fever, abdominal distention, and diarrhea; however, this broad definition makes it difficult to determine its actual incidence. Individual reports in the published data quote an incidence of anywhere between 5% and 42%.[17-21] The presence of chronic enterocolitis can lead to failure to thrive.

On initial assessment of patients, particularly those a year out from their pull-through with abdominal distention with or without enterocolitis, we again review the history and physical examination, evaluate the anal canal under anesthesia, and perform a rectal biopsy. We then implement a regimen of rectal irrigations with normal saline. The goal of irrigations is to "wash" the lining of the bowel to help eliminate any liquid stool or flatus, which is different from enemas. Aliquots of 10–20 mL of saline at a time are

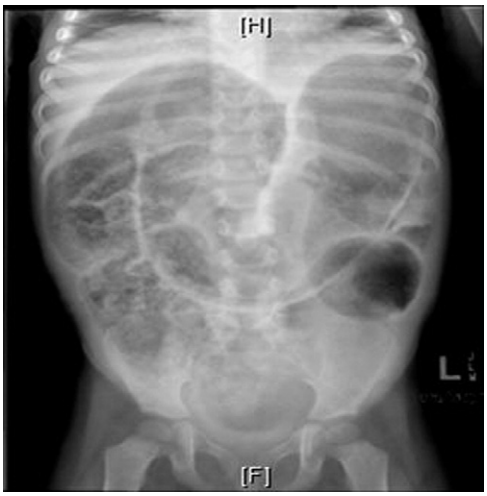


Figure 3 Abdominal radiograph of a 3 year-old child presenting with abdominal distention, fever, and vomiting 1 year after a pull through procedure. Enterocolitis was treated with irrigations and metronidazale. The patient subsequently underwent a redo pull through for persistent transition zone bowel.

administered using a large Foley catheter. The instilled saline is allowed to passively drain after each aliquot. The total amount of saline used depends on the effect of the washout and the amount of stool evacuated. Most of the irrigant returns back out of the tube, hence a large volume can be used (1000–2000 mL). The child should not be allowed to retain > 20 mL/kg of fluid. Rectal irrigations are done anywhere from 1 to 3 times a day, depending on the severity of the abdominal distention. Oral metronidazole is often added to the regimen if there is any evidence of enterocolitis, and sometimes must be administered long-term for many months.[22] After a pull-through, we are aggressive in managing these patients at the onset of abdominal distention or the presence of a dilated colon on abdominal radiographs (Figure 3) to try to avert the development of an acute episode of enterocolitis. All parents should be instructed how to administer rectal irrigations at home if the child begins to show abdominal distention. Although rarely necessary, one should always be prepared to open a proximal diverting colostomy or ileostomy in patients with persistent, severe enterocolitis not manageable with irrigations.

We then evaluate the underlying cause of the distention. Pathologically, there are 2 causes that can result in a patient having abdominal distention and recurrent enterocolitis—pull-through of an aganglionic segment or pull-through of transition zone. A rectal biopsy is required to rule out a pathologic cause. Our practice is to perform full thickness rectal biopsies with no electrocautery. The specimens are sent fresh to our pathology department. The components that we look for to exclude a pathologic reason for the abdominal distention are the presence of ganglion cells, the absence of hypertrophic never fibers, and normal acetylcholinesterase staining (Figure 4). This complication is potentially avoided with experienced pathology guidance during the initial pull-through. During frozen section analysis at the time of the initial pull-through, the pathologist must confirm normal ganglion cells and normal nerve trunks at the site of the planned anastomosis. The specimen must be full thickness because if submucosa is not analyzed, hypertrophic nerves might be missed. This recommendation may need to change the common practice of sending only muscularis, as is often done during a laparoscopic biopsy. We suspect that many cases of post pull-through patients described in the published data as having "neuronal intestinal dysplasia" may in fact have transition zone bowel.[23]

Anatomical problems resulting in enterocolitis can be related to the initial operation. Generally, anything that interferes with the normal passage of stool can result in stasis and predispose the patient to enterocolitis. Three of the causes we have seen can occur regardless of the initial procedure performed. These include stricture, usually at the anastomotic site, a retained dilated segment of colon, or a twist or kink in the pull-through segment. Each of these can result in a mechanical obstruction, and subsequent blockage of stool resulting in abdominal distention, and all can be observed on a contrast enema (Figures 5 and 6).

There are mechanical issues that are specific to the different surgical procedures. In patients who have had a previous Duhamel procedure, a megarectal pouch can cause stasis of stool.[24] The Duhamel pouch was developed in an attempt to slow colonic transit time by leaving a patch of aganglionated bowel.[25,26] Although used by some in all cases of Hirschsprung disease, it has mostly been applied to patients with total colonic aganglionosis. The pouch creates a reservoir, thus decreasing the frequency of bowel movements and increasing the amount of water absorption. However, if a pouch does not efficiently empty, a megarectal pouch can develop, leading to fecal impaction.[24] More dangerous, however, is if the fecal stasis causes bacterial overgrowth and mucosal inflammation resulting in enterocolitis. Contrast enemas in these patients often show a characteristic megarectal pouch with or without fecal impaction (Figure 7).

Similarly, children with total colonic aganglionosis who have a "colon pouch"—Martin pouch, using left colon, or

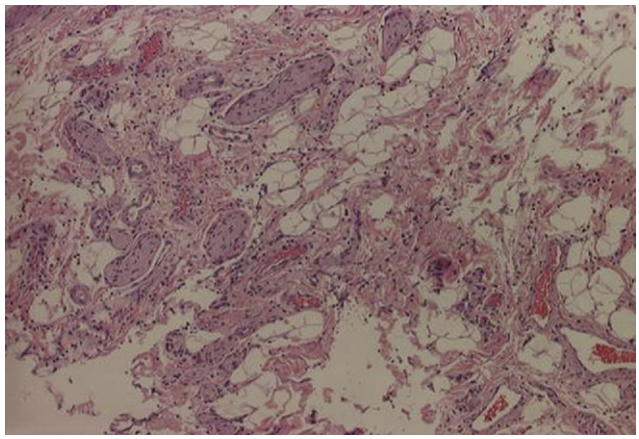


Figure 4 Biopsy specimen in a post pull-through patient demonstrating aganglionosis and the presence of hypertrophic nerves.

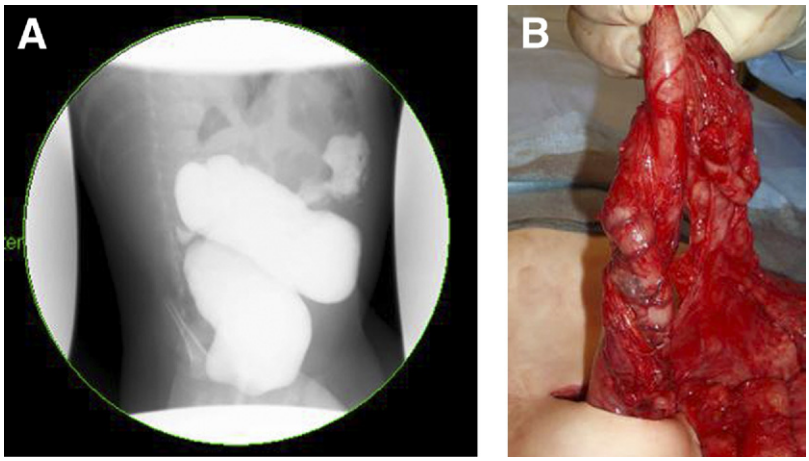


Figure 5 (A) Contrast study of a 5-month-old boy after a pull-through procedure showing a bowel obstruction. (B) He was found to have an almost 360° twist in his pull-through segment.

Kimura pouch, using right colon—are at increased risk of developing enterocolitis. The patched segment can dilate over time, resulting in fecal stasis and bacterial overgrowth.

In patients who have had a previous Soave procedure, the muscular cuff can result in a mechanical obstruction if not adequately split or if a significant length of cuff was left behind. The residual muscular cuff can cause a constricting fibrotic ring in the pelvis resulting in a functional stricture. We suspect that even though the cuff may have been split during the original operation, sometimes it can scar down or roll up and cause obstruction. Once again, proximal distention can lead to fecal stasis and an increased risk of enterocolitis. When the transanal endorectal pull-through was first introduced, a longer rectal muscular cuff was left. The trend now in those performing the transanal Soave technique is to leave only a few centimeter rectal muscular cuff before a full thickness dissection which will hopefully reduce the cuff problems observed.[27]

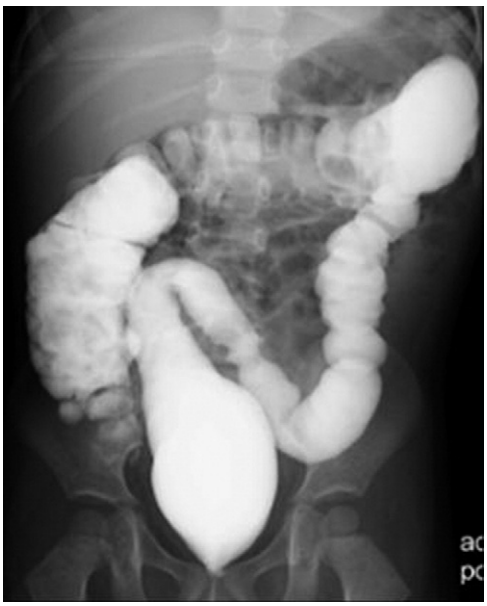


Figure 6 Contrast study of retained dilated segment of colon after a laparoscopic assisted transanal pull-through procedure. The dilated segment led to fecal stasis and enterocolitis.

A problematic rectal muscular cuff can be palpable on rectal examination. It can also be suspected on contrast enema with an increased distance between the pull-through segment and the sacrum (Figure 8).

In addition to the management of the abdominal distention and enterocolitis with irrigations and metronidazole, the mechanical problem can be corrected with a redo operation. Our approach is to perform a redo transanal resection with a Swenson technique to remove the problematic stricture, pouch, rectal muscular cuff, or distal aganglionic or transition zone bowel, with the addition of laparoscopy or laparotomy if required. If the rectum is frozen with scar, a posterior sagittal approach may be needed.[14] Particular attention is given to preserving the dentate line and avoiding excessive stretching of the sphincters.

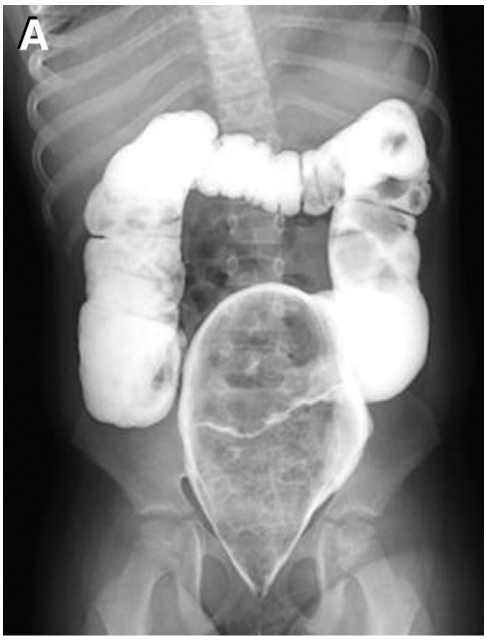
We have had the opportunity to evaluate and treat 75 patients with Hirschsprung disease who had pull-through procedures at other institutions, were referred to us with postoperative problems, and for whom we performed a reoperation. Several patients had more than one indication for a redo operation. Fifty-one have been previously reported.[14] Of the 75 patients, 29 were operated on for strictures or "kinks" resulting in a mechanical obstruction. Twenty-two patients had a megarectal pouch from a Duhamel (1 patient had a Kimura patch). Eleven patients had an obstructing Soave cuff. Five patients had an excessively dilated distal segment, with normal ganglion cells and no hypertrophic nerves. Fifteen patients had residual aganglionic segments. Eight patients had transition zone bowel pulled through that required removal. The indications for 19 of these patients were for recurrent pelvic abscesses or fistulas.

Forty of these patients had a posterior sagittal approach with or without laparotomy, because of severe scarring and fibrosis of the pelvis, or a stricture or fistula not easily

accessible from above or below. The remaining 35 patients had a transanal approach, with or without laparoscopy or laparotomy (Figure 9).

Discussion

The optimal long-term outcome for patients after surgery for Hirschsprung disease is to be fecally continent and to have normal bowel movements without abdominal distention or enterocolitis. Usually, this outcome is achieved; however, there is a small subset of patients who have difficulty after their primary operation. We believe that these patients should be evaluated with a systematic approach to determine the etiology of their problems (Figure 10).



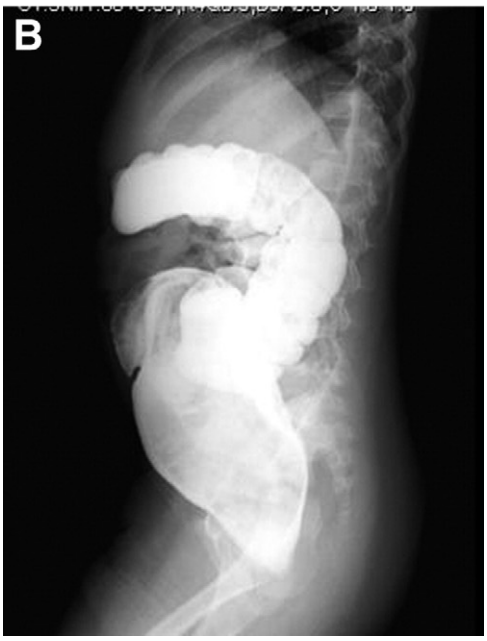


Figure 7 **(A, B)** Large Duhamel pouch causing fecal impaction.

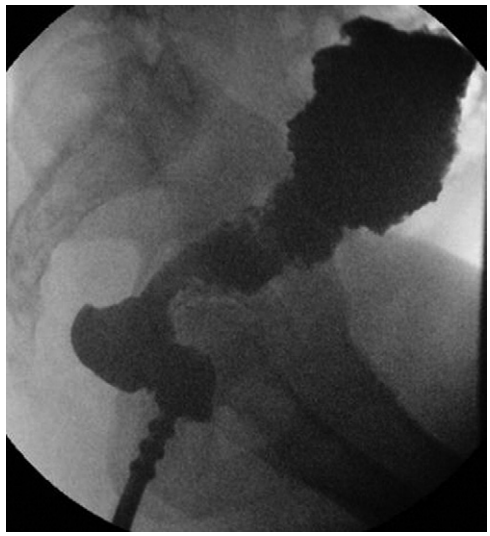


Figure 8 Problematic Soave cuff causing narrowing of the distal pull-through. Note the area of narrowing and the increased distance between sacrum and rectum, consistent with a retained and obstructing Soave cuff.

The patients can generally be classified into 2 specific groups: (1) the patient who is soiling and (2) the patient who is having repeated episodes of abdominal distention and enterocolitis. Interestingly, the same patient does not appear in both categories. We believe this is because with an open sphincter that is damaged, there is no stasis and therefore no enterocolitis; however, there is soiling.

The most important aspects of the work-up include a detailed history of the child's bowel habits, knowing the

Reoperations for Hirschsprung's Disease (75 patients*)	
Mechanical Indications	
Stricture	29
Megarectal pouch (post-Duhamel)	22
Obstructing Soave cuff	11
Dilated segment	5
Pathological Indications	
Aganglionosis	15
Transition Zone	8
Other Indications	
Fistulae (Rectocutaneous, rectourethral, rectovaginal)	12
Recurrent Pelvic Abscess	7

Note: Several patients may have more than one indication for surgery

* The first 51 patients have been reported in [14]

Figure 9 Redo operations performed for Hirschsprung disease in our institution.

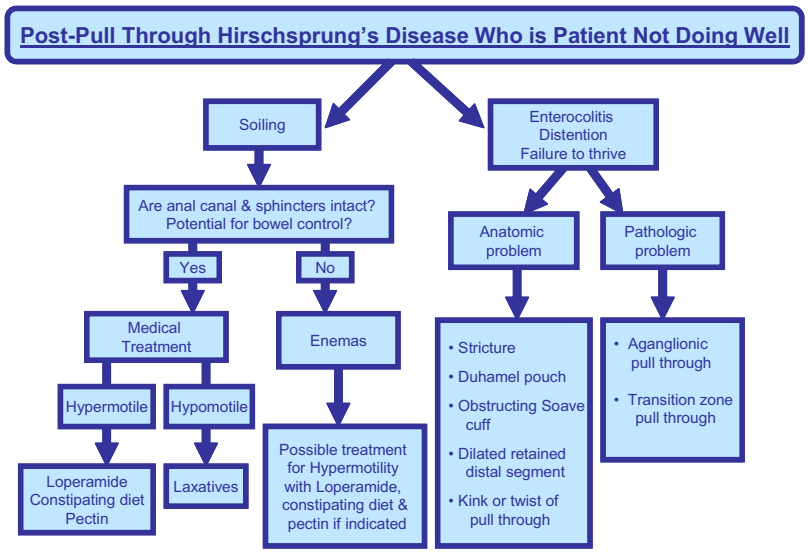


Figure 10 Evaluation and treatment algorithm for a patient who is not doing well following a primary operation for Hirschsprung disease.

original technique used for the pull-through, an examination of the anal canal and sphincters, a contrast enema to determine the anatomy of the child's colon from which its motility can be inferred, and a rectal biopsy.

Postoperative fecal incontinence in Hirschsprung disease is a devastating complication. Determining the cause of fecal incontinence is obvious in patients with an absent or damaged anal canal, but may not be known in other cases. Continence is dependent on intact sensation, voluntary sphincter control, and appropriate colonic motility. Loss of any of these 3 mechanisms can alter the patient's ability to have voluntary bowel movements.[28] Even though we may not be able to restore these patients' continence mechanism, through bowel management and the administration of daily enemas, we can have the child stop soiling in almost every case.[15] Also, we can alter the consistency of the stool and the colon's motility so the child can have better sensation and control of thicker stool, giving them the potential to have voluntary bowel movements. We can also treat constipation with laxatives if that is the cause of the pseudo-incontinence.[16]

In patients with persistent abdominal distention, constipation, and enterocolitis after operative management of Hirschsprung disease, it is important to rule out an anatomical or pathological reason for this constellation of symptoms. This can be evaluated with a contrast enema and a full thickness rectal biopsy. Several institutions use manometry in the evaluation of the motility of postoperative patients because it has been shown that constipation and bowel distention is a result of the absence of high propagating contractions.[29] From our experience, manometry does not provide any additional information in the evaluation because the contrast enema provides the needed visual evaluation of the patient's colonic motility.

Redo operations can significantly improve the abdominal distention and constipation if there is an anatomical or pathologic cause. As we have previously noted, often the redo pull-through procedure can be accomplished through a transanal approach, with or without laparoscopy or laparotomy.[14]

Although we are able to significantly change the quality of life of many of the patients with Hirschsprung disease who are not doing well after a pull-through procedure with bowel management, medications, and/or redo operations, it is the excellent performance of the primary operation that has the best opportunity to provide these patients with the ideal outcome.

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