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Reoperations in Hirschsprung disease

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Abstract

Background: We sought to identify causes of preventable complications related to operations for Hirschsprung disease.

Methods: We reviewed the cases of 51 patients with Hirschsprung disease who underwent a primary procedure elsewhere, had a complication, and were referred for reoperation.

Results: Thirty-five patients had 1 failed operation, 10 had 2, and 6 had 3. Initial operations were Soave (20), Duhamel (15), Swenson (5), transanal endorectal (4), myectomy (3), unknown (3), and laparoscopic Swenson (1). Thirty-one patients presented with a stoma. Patients without a stoma (20) had fecal impaction (8), recurrent enterocolitis (6), and fecal incontinence (6). None had both enterocolitis and incontinence. Reoperation was performed posterior sagittally (40) or transanally (5). Indications included stricture (21), megarectal Duhamel pouches (12), fistulae (11 [8 rectocutaneous, 2 rectourethral, and 1 rectovaginal]), pouchitis (2), and retained aganglionic bowel (8). After reoperation, 14 were continent, 11 had a stoma (8 permanent), 6 had voluntary bowel movements but soiled occasionally, 6 received rectal irrigations to avoid enterocolitis, 6 were incontinent but clean with bowel management, and 2 were lost to follow-up.

Conclusion: Stricture, megarectal pouch, fistula, and retained aganglionic bowel are preventable complications. Enterocolitis is partially preventable but can occur after a technically correct procedure. Fecal incontinence is a preventable complication likely because of anal canal damage.

During the last 25 years, we have had the opportunity to treat 51 patients, born with Hirschsprung disease, operated on at other institutions, who experienced significant complications that required further treatment. Because most of these complications can be considered avoidable, we decided to review these cases to determine the causes of these complications and try to elaborate recommendations to prevent them.

1. Materials and methods

A retrospective review of the medical records of these 51 patients was performed with institutional review board approval (CHMC 06-01-04). The patients and/or their families were contacted by phone, letter, or an interview in our clinic.

2. Results

The ages of the patients ranged from 1 to 23 years, with an average of 5.7 years. Thirty-four were males and 17

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Table 1 Type of initial operation

Type of initial operation	No. of patients
Soave	17
Duhamel	14
Transanal endorectal pull-through	6
Swenson	5
Unknown	3
Myotomy, myectomy	3
Soave and right colonic pouch	2
Swenson J pouch	1
Total	51

females. Thirty-five patients had undergone 1 previous operation, 10 had been subjected to 2 procedures, and 6 had 3 previous surgical interventions.

The operations initially performed on these patients included 17 Soave procedures, 14 Duhamel, 6 transanal, 5 Swenson, 3 unknown type, 3 myotomy and/or myectomy, 2 Soave with a right colonic patch, and 1 Swenson with a J pouch (Table 1).

Thirty-one patients came to us with an enterostomy and 20 without. Of these 20 patients who came with no enterostomy, 8 experienced fecal impaction, 6 severe enterocolitis, and 6 fecal incontinence. No patient had both enterocolitis and incontinence. From the group of patients who came with an enterostomy (31 patients), 20 (65%) experienced a severe stricture or acquired atresia of the attempted coloanal anastomosis. Eleven patients had a rectal fistula, and 1 of them also had a rectal stricture. The fistula went from the rectum to the skin in 8 cases, to the urethra in 2, and to the vagina in 1.

Our management included reoperation in 45 cases and bowel management for fecal incontinence alone in 6 patients. The operations included 40 approached posterior sagittally (with or without a laparotomy) and 5 performed transanally. The posterior sagittal approach (Fig. 1) was used in cases with severe scarring and fibrosis of the pelvis

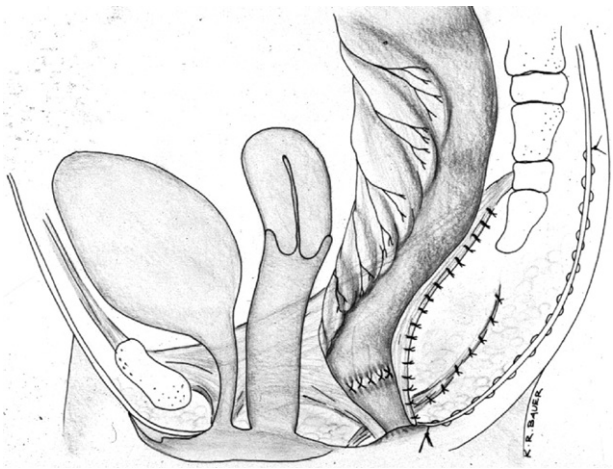

Fig. 1 Posterior sagittal transphincteric approach for reoperation of a Hirschsprung pull-through.

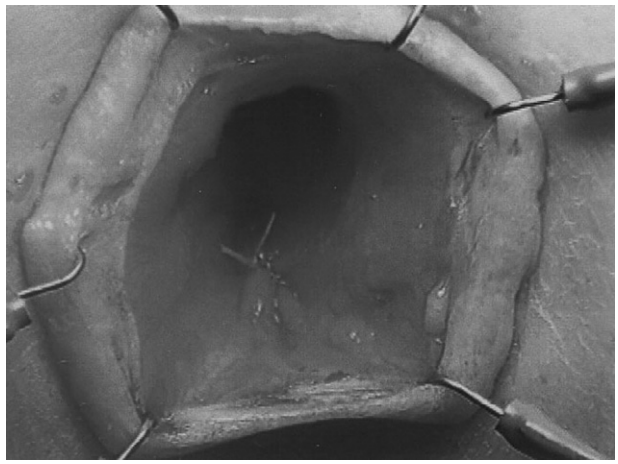

Fig. 2 Anastomosis below the dentate line.

or in those patients whose stricture or fistula was located in the pelvis at a site considered too low if approached from above or too high if approached from below. The transanal approach was used in those cases with an intact anal canal and minimal fibrosis. The 6 patients who were subjected to bowel management for the treatment of fecal incontinence were not considered good candidates for a reoperation because an examination under anesthesia showed a coloanal anastomosis performed much lower than the pectinate line (Fig. 2), which was interpreted as evidence of damage to the sensitive area of the anal canal as well as the sphincter mechanism. We therefore believed that the only treatment for those patients was bowel management.

The reoperations performed by us in these cases were indicated in 21 patients because of a stricture or acquired atresia of the rectum; in 12 patients who experienced fecal impaction and a giant megarectal pouch (because of a Duhamel type of operation); in 8 patients because a secondary rectal biopsy showed that they had a retained aganglionic segment (they underwent a redo pull-through to reach a ganglionic segment of colon); and in 11 patients because they had a fistula from the rectum to the skin, urethra, or vagina. Finally, 2 patients were reoperated on because they experienced severe, intractable pouch inflammation (both of these cases had total colonic aganglionosis) (Table 2).

Table 2 Reasons for reoperation in 45 patients

Reason for reoperation	No. of patients
Stricture, acquired atresia, and dehiscence	21
Megarectal pouch (post-Duhamel)	12
Aganglionosis on rectal biopsy	8
Cutaneous fistula	8
Urethral fistula	2
Pouchitis	2
Vaginal fistula	1
Total	54[a]

[a] Some patients had more than 1 indication.

Table 3 Follow-up results of reoperations

Follow-up results of reoperations	No. of patients
Voluntary bowel movements	14
Stoma[a]	11
On rectal irrigations	6
Voluntary bowel movements, occasional soiling	6
Fecal incontinence (on bowel management)	6
Lost to follow-up	2
Total	45

[a] Permanent in 8 patients.

We have followed up the patients 6 months to 25 years after reoperation. Fourteen of them have voluntary bowel movements and normal social lives. Eight patients have a permanent stoma and 3 a temporary one that we are planning to close. Six patients are on long-term rectal irrigations for the management of severe enterocolitis. Six patients have voluntary bowel movements and occasional soiling, which is interpreted as partial fecal incontinence. Six patients are totally incontinent but remain clean with a bowel management regimen involving a daily enema. Two patients have been lost to follow-up (Table 3).

In addition, 12 patients, including those with a stoma, have a chronic open wound of unknown origin (Fig. 3). One still has a rectovaginal fistula, 1 has an anastomotic stricture, and 1 experiences pouchitis and failure to thrive.

Of the 8 patients who have a permanent stoma, 6 have the kind of chronic open wound shown in Fig. 3. One patient has a stoma because we were not able to pull the bowel down because of a short mesentery. One patient has a stoma because of intractable enterocolitis.

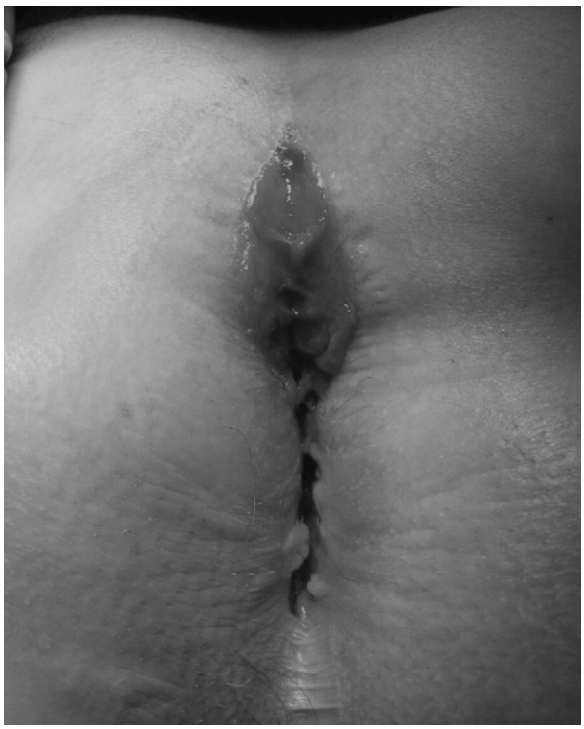

Fig. 3 Chronic open wound, fistula, and sinus.

3. Discussion

The large number of patients we saw who experienced complications post-Hirschsprung operation are not representative of the incidence of these complications in other institutions because ours is a referral center for colorectal problems in children.

Complications and sequelae related to the treatment of Hirschsprung disease can be classified as (a) preventable, (b) nonpreventable, and (c) partially preventable (Table 4).

Preventable complications include stricture and acquired atresia of the pulled-through bowel, retention of aganglionic colon or aganglionic pull-through, fistula formation, residual megarectal pouch (post-Duhamel), pouchitis, and fecal incontinence, which are reported complications [1-8].

The nonpreventable sequela that occurred in this study is enterocolitis. We, like others, consider enterocolitis a rather mysterious, unpredictable sequela of unknown origin [1,9,10,]. Interesting ideas have been described to try to explain this condition [10]. We know that there are certain factors, such as fecal stasis with or without a retained piece of aganglionic bowel or the administration of broad-spectrum antibiotics, which may aggravate the problem, but the basic pathophysiology of this condition remains unknown.

Partially preventable complications include constipation and chronic open wounds. The patients who experienced more constipation were those who underwent a Duhamel procedure. These patients were left with a rectal pouch that accumulates stool, becomes larger with time, forms a fecal impaction, and exacerbates the problem of postoperative constipation (Fig. 4). In addition, it seems that those patients who underwent a pull-through of a very dilated, normoganglionic colon will most often experience constipation. In other words, we have learned that a dilated pulled-through bowel is almost as bad as the pull-through of an aganglionic segment. On the other hand, sometimes, constipation occurs in patients subjected to a technically correct pull-through; we have no explanation for it, which is why we consider this condition partially preventable.

Table 4 Complications of Hirschsprung disease

Preventable	Nonpreventable	Partially preventable
Stricture and acquired atresia	Enterocolitis	Constipation
Aganglionic pull-through		Chronic open wound
Fistula		
Megarectal pouch		
Pouchitis		
Fecal incontinence		

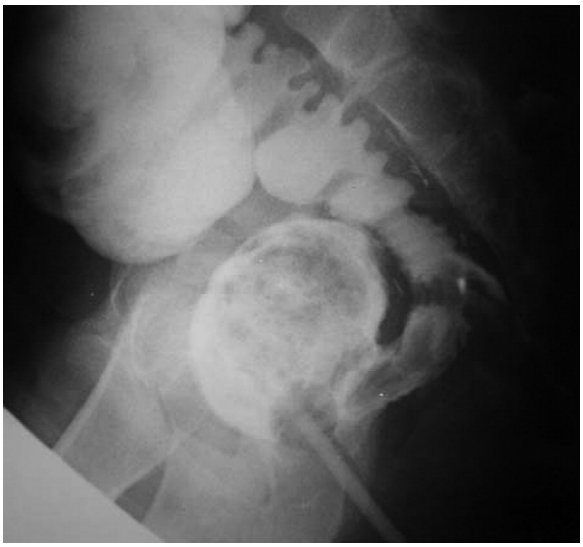

Fig. 4 Characteristic megarectal pouch with fecal impaction in a patient after Duhamel pull-through.

Chronic open wounds sometimes occurred in patients who were previously subjected to an endorectal (Soave) type of procedure. During the endorectal dissection, the surgeons may have damaged the mucosal tube and inadvertently left islets of mucosa attached to the muscular cuff. We surmise that these islets provoked collections of mucus between the muscular cuff and the bowel wall of the pulled-through colon, and eventually, these collections of mucus became infected, formed abscesses, and created chronic fistulae, which are very difficult to eradicate. However, there have been some cases of chronic open wounds for which we were unable to document the presence of these mucosal remnants. For this reason, we consider this complication partially preventable. One particular patient with a chronic open wound, after 10 years of unsuccessful treatments, demonstrated in 1 of her biopsies that she experienced granulomatous inflammatory bowel disease, a finding that has been previously described [11,12]. The wound closed after a course of medical treatment for Crohn disease. After that case, we convinced our gastroenterologists to give Infliximab to 2 of our other patients with chronic open wounds, despite not having histologic evidence of inflammatory bowel disease. Both wounds healed after several months of this treatment.

We noted cases in which the anorectal dissection was performed too low in the pelvis and the normal ganglionic bowel was anastomized below the pectinate line, which we believe explains the fecal incontinence. This is a preventable and unacceptable complication. In the literature, some of the cases of fecal incontinence related to the treatment of Hirschsprung disease have been attributed to colonic hypermotility, based on postoperative colonic manometric studies [13]. This is difficult for us to accept because individuals with an intact anal canal and sphincter mechanism may endure severe diarrhea (hypermotility) but not necessarily fecal incontinence. In addition, the authors, in their publication, did not mention whether or not the anal canal was intact. Based on our experience, we consider it an obligate step in the evaluation of patients with Hirschsprung disease with incontinence to perform an examination under anesthesia to document the integrity of the anal canal. When the coloanal anastomosis has been done below the pectinate line, we believe that the only therapeutic alternative is the implementation of a bowel management program, consisting in the administration of loperamide, a strict constipating diet, and a daily saline enema [14].

The treatment of the characteristic sequelae seen in cases subjected to Duhamel procedures (Fig. 4) consisted of the resection of the megarectal pouch and re-anastomosis of normal ganglionic bowel to the anal canal 1 cm above the pectinate line. This operation was successfully performed transanally when the patients did not have excessive scar tissue. The posterior sagittal approach was used when the patient had significant pelvic fibrosis, which was usually related to a catastrophic event (dehiscence, retraction, abscess, etc) that occurred during the original operation.

Patches or pouches [15,16] have been used in the treatment of total colonic aganglionosis. The rationale has been to try to create a reservoir with the purpose of decreasing the frequency of bowel movements and to promote water absorption. In general, we believe, as others [17], that the creation of pouches in patients with Hirschsprung disease is not advisable. It is known that stasis in Hirschsprung disease results in a proliferation of abnormal bacteria and absorption of toxins and leads to enterocolitis [18]. Pouches and patches are created with the specific purpose of slowing down fecal transit, which results in colonization of the colon with abnormal bacteria and can

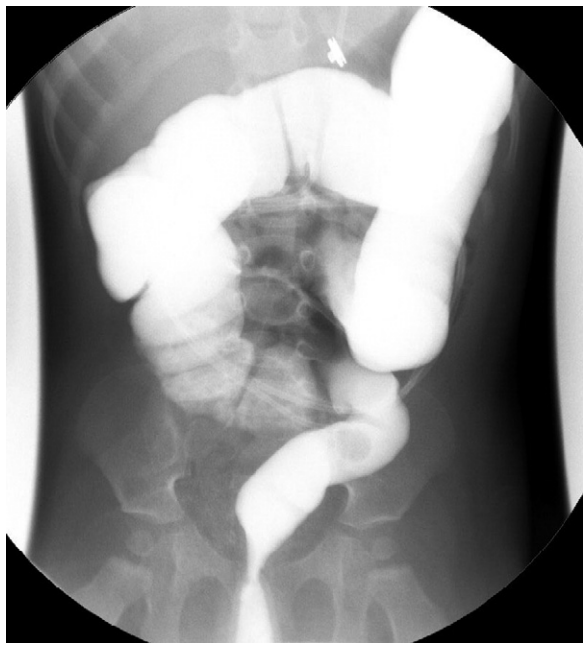

Fig. 5 Contrast study showing residual aganglionosis.

lead to severe enterocolitis. The resection of the pouches in our patients cured their symptoms.

Some of the patients who came to us with severe enterocolitis after an operation for Hirschsprung disease had a retained portion of aganglionic bowel (Fig. 5), a problem others have reported [7,19,20]. This indicates that we need more and better experienced pathologists in the field of Hirschsprung disease to operate on these patients safely.

Another group of patients with enterocolitis had normal ganglionic bowels, yet they experienced severe enterocolitis. We believe that a proactive program of postoperative rectal irrigations makes the problem of enterocolitis more manageable and allowed us to prevent some permanent stomas. Frequent irrigations, plus the administration of metronidazole in a proactive, prophylactic way postoperatively, allowed us to decrease the morbidity of these serious sequelae. These were not enemas but, rather, were irrigations done with a large Foley balloon, with a total of 20 cc/kg of saline introduced 10 to 20 cc at a time. The Foley balloon was then allowed to drop out of the body, thereby essentially washing the inside of the colon. Our routine after the pull-through was to carefully monitor the dilatation of the bowel radiologically. When indicated, we initiated a program of colonic irrigations (no enemas) 3 times per day and administration of oral metronidazole. We saw the patient every month and gradually decreased the frequency of irrigations as well as the dosage of metronidazole. We sometimes administered the metronidazole intracolonically during 1 of the day's irrigations.

Interestingly, we never saw a patient who experienced both fecal incontinence and enterocolitis. Proposed treatments for enterocolitis that are frequently unsuccessful include myotomies, forceful dilatations, and injection of botulinum toxin. What all these treatment modalities have in common is the idea of decreasing or eliminating the sphincter tone, which, taken to an extreme, will cure enterocolitis but will produce fecal incontinence.

Rectourethral and rectovaginal fistulae are truly unacceptable and preventable complications. These complications can be avoided by adherence to basic surgical principles. Professors of pediatric surgery, as well as directors of training programs, must teach the importance of these principles to avoid these complications.

The posterior sagittal approach used in the reoperation of 40 of our patients proved to be a useful surgical alternative in taking care of patients who had acquired atresias or strictures, particularly those who have been subjected to more than 1 previous operation and experienced what is known by surgeons as a *frozen pelvis*. The area of the stricture is usually located in a rather high area of the pelvis to be approached transanally and rather low to be approached abdominally (Fig. 1). However, our specific recommendation is to always leave a protective colostomy after this type of operation. Otherwise, the patient may develop a midline posterior sagittal rectocutaneous fistula postoperatively.

In 5 cases, we elected to reoperate on the patients transanally because the anatomical circumstances allowed this. The specific cases were those of patients who had a retained portion of aganglionic bowel or a dysfunctional Duhamel pouch with an otherwise nonstrictured, nonfibrotic pelvis.

One advantage we observed of the Duhamel operation is that it produces very little damage to the sphincter mechanism. We never encountered a case of rectocutaneous fistula or saw a case of real fecal incontinence associated with this type of procedure. However, the main inconvenience of the Duhamel procedure is the development of severe fecal impaction in the overdilated rectal pouch. Those pouches can be relatively easily resected transanally if a reoperation is indicated.

Even when we were able to significantly change the quality of life of many of our patients, the final results (which include the presence of a permanent stoma in 8 of our patients) indicate that once a patient with Hirschsprung disease has experienced a serious complication during the first attempt to treat the disease, the chance for that patient to experience permanent sequelae is very high. This reinforces the principle that most children with congenital conditions have a unique single opportunity to have an adequate operation that gives them good quality of life.

Acknowledgment

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Discussion

C.D. Smith, MD, MD (Charleston, SC): What role does Botox have with the patients?

Marc Levitt, MD (response): Botox works temporarily to allow the stool to eliminate. But essentially, it creates a kind of incontinence. As soon as the Botox wears off, many of their symptoms recurred. It avoids circumstances of enterocolitis because the stool left very easily. But it's not a permanent solution, and I don't really understand how it actually works. What do you do when the Botox disappears? We did not have patients that got Botox, and then their symptoms completely improved. I think it's just temporally making them incontinent for a time. But once they get the tension at high-pressure zone back, their enterocolitis and symptoms recur.

Whit Holcomb, MD (Kansas City, MO): You talked about your approach: either the transanal or the posterior sagittal approach was determined by whether or not there was a fibrotic pelvis. Is that made on a digital exam? Or how do you come to that conclusion that the pelvis is a fibrotic approach?

Marc Levitt, MD (response): When Dr Peña began, all of these reoperations were done posterior sagittally in the beginning of this series. And then, when the transanal approach became—for primary cases—more prevalent, he began to start transanally. I think it's something that you can start transanally and feel if you are able to mobilize the rectum or not. You quickly realize that as you dissect transanally, you're not getting the rectum coming towards you; it's actually disappearing into the pelvis. In most situations, the safe thing to do is to open a posterior sagittal incision so that you can mobilize the rectum laterally and fully mobilize it. So at this point, I would say it's a patient that we would start with transanally and see how we do and have the opportunity to open posterior sagittally for full mobilization. Again, to repeat, if we open a posterior sagittal incision, we always divert such a patient because we had 2 cases of rectocutaneous fistula develop after the posterior sagittal incision.

Whit Holcomb, MD (Kansas City, MO): So that means the patient is positioned prone on the operating table?

Marc Levitt, MD (response): Yes. And if they're small enough, we do a total body prep in case we need to go into the abdomen.

Whit Holcomb, MD (Kansas City, MO): The second question is, would you explain to us what your usual workup is for these patients who you're considering a reoperation?

Marc Levitt, MD (response): Most patients with Hirschsprung's disease do very well, and there's a small percentage that don't do well. You have to ask yourself, well, how are they not doing well? A common scenario is fecal incontinence, and another common scenario is recurrent enterocolitis. Our routine is to get a contrast enema in every one of those patients, which I think tells you a lot. It tells you, first of all, whether there's any stricture. It tells you whether there's a residual dilated segment. We always take such a patient to the operating room and perform an examination under anesthesia. We almost always repeat the biopsy to be absolutely certain that there's no aganglionic bowel down below. But the inspection is key. You know, obviously, if there's a stricture. And we evaluate the anal canal.

In certain circumstances, the anal canal, essentially, is absent, and the pull-through is done to the anal skin. In that case, none of those patients are continent when they've completely lost their anal canal. That will influence whether or not we do a reoperation because a reoperation in those children won't improve their fecal incontinence, if that's what they came there for. So that's a patient we subject to bowel management.

Whit Holcomb, MD (Kansas City, MO): Based on your experience with the reoperation, do you have any tips or advice on where we should start or perform our anal anastomosis in relation to the anal column?

Marc Levitt, MD (response): We try to do it at least 1.5 to 2 cm proximal to the dentate line. I think the key is to put the lone-star pins in and then actually to replace them a little bit deeper. So you really make a conscious effort at preserving a significant length of anal canal.

Dr Brian Kenney (Columbus, Ohio): Can you describe to us a little bit more about the transanal approach to these patients who have had a Duhamel? I'm not sure I understand that you're doing an endorectal dissection. How do you know when you've reached the top of the dilated pouch?

Marc Levitt, MD (response): We usually scope them first so we can see how big a pouch we're dealing with. And then, once you start with transanal dissection in Duhamels, there's very little perianal adhesions because, obviously, the anastomosis is a little bit higher than a Swenson or a Soave. So it actually is quite easy to mobilize full thickness. You end up mobilizing 2 lumens. You essentially deliver out the Duhamel pouch through a transanal approach until you get to the bowel that is not adherent to the pouch and then do a coloanal anastomosis. If you stay full thickness outside of the rectal wall, it actually mobilizes quite remarkably easy.

Brian Kenney, MD (Columbus, Ohio): Not anorectal, then? Full thickness?

Marc Levitt, MD (response): It's a full-thickness, transanal mobilization. It's essentially a Swenson with mobilizing of both the normal and the pouch together. You get the whole thing out, and then you transect and do the primary anastomosis. I think it would be difficult, if you were not full thickness, to find exactly where you need to be.

C.D. Smith, MD (Charleston, SC): Was there any patient who had an anastomosis that was too high?

Marc Levitt, MD (response): You mean a Duhamel?

C.D. Smith, MD (Charleston, SC): In a pull-through where the anastomosis had been higher, say, than 2 cm above the dentate line?

Marc Levitt, MD (response): Where we re-resected?

C.D. Smith, MD (Charleston, SC): These came for reoperation?

Marc Levitt, MD (response): Yes, and that's something I think we have a very poor understanding of. There obviously are Hirschsprung's operations that initially leave a big segment of aganglionosis, and patients do quite well, like with a Rehbein. But certain patients can't handle that even small zone of aganglionosis. We judge based on how well the patient did. If the patient had recurrent enterocolitis and had aganglionosis at the distal bowel, we would offer them a re-resection to try to get ganglionated bowel down. So yes, that can happen where the zone of aganglionosis is quite long. But that's extremely rare. More commonly, the anastomosis was done too low.