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## Hirschsprung's disease: early diagnosis and long-term outcomes

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## CHAPTER 10

# Dyssynergic defecation may play an important role in postoperative Hirschsprung's disease patients with severe persistent constipation

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# SUMMARY

## **Background**

After surgery for Hirschsprung's disease (HD) the majority of patients have satisfactory clinical outcomes. Nevertheless, a substantial number of patients remain who suffer from severe persistent constipation. Current consensus attributes these complaints to the hallmarks of HD. In non-HD patients a cause for severe constipation is dyssynergic defecation.

## **Methods**

Retrospectively, we reviewed the medical records of ten post-operative HD patients with severe persistent constipation who had undergone extensive anorectal function tests to diagnose the reason for the constipation. We analyzed the results of these tests.

## **Results**

During the last three years, ten post-operative HD patients with severe persistent constipation were given extensive anorectal function tests. All ten patients were diagnosed with dyssynergic defecation. The ages at the time of diagnosis ranged from 7 to 19 years with a median age of 12 years. Signs of an enlarged rectum were seen in all ten patients, with a maximum measured value of 845 mL.

## **Conclusions**

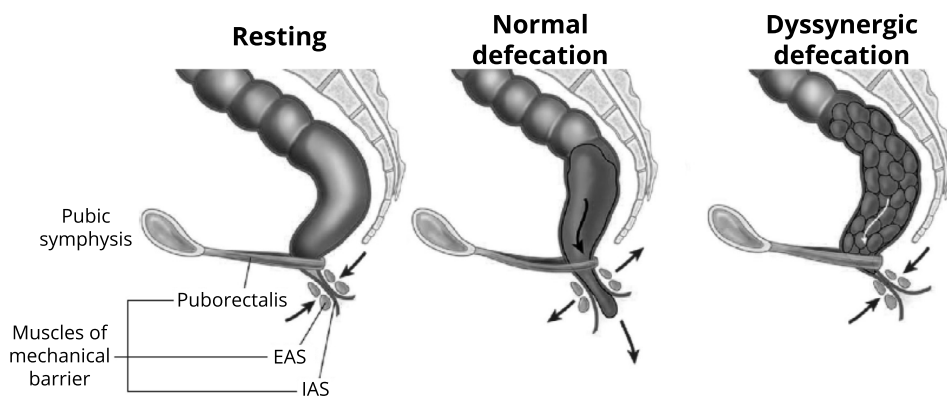
Patients with HD may also suffer from dyssynergic defecation. It is important to consider this possibility when dealing with severe persistent constipation in post-operative HD patients. Viable options for treating dyssynergic defecation are available that could prevent irreversible long-term complications.

# INTRODUCTION

Hirschsprung's disease (HD) is a birth defect characterized by aganglionosis of the distal colon. Due to obstipation proximal to the affected segment of the colon HD often presents with a failure to pass meconium during the first 24 to 48 hours after birth. Incidence numbers are estimated to be 1 in 5000 live births, with a pronounced predominance of males over females.<sup>1,2</sup>

Nowadays, after surgery for HD, the majority of patients have satisfactory clinical outcomes with mild constipation, especially after reaching adulthood.<sup>3-5</sup> Some reports, however, show a substantial number of patients with complaints including severe persistent constipation and fecal soiling.<sup>6-9</sup> Constipation in post-operative HD patients may be caused by the absence of the internal anal sphincter reflex or by a residual part of aganglionic colon remaining after surgical reconstruction. Neither of these causes, however, explains why some patients suffer from severe constipation while others have no defecation problems whatsoever.

Apart from the two causes mentioned there are other causes with good treatment options that could be responsible for the severe constipation seen in some HD patients. One is dyssynergic defecation, a disorder characterized by a paradoxical involuntary contraction of the external anal sphincter which leads to a functional neuromuscular obstruction (Figure 1).<sup>10</sup> Recently, another study showed that dyssynergic defecation is responsible for severe constipation in patients with inflammatory bowel disease.<sup>11</sup> While



**Figure 1**

A schematic representation of the normal defecation process and of dyssynergic defecation. During squeeze the external anal sphincter and the puborectal muscle are contracted and they create a mechanical barrier for stool. In normal defecation the puborectal muscle and the external anal sphincter relax, so that stool can be evacuated. In pelvic floor dyssynergia there is a paradoxical involuntary contraction of the external anal sphincter and the puborectal muscle.

the exact cause of dyssynergic defecation is unknown for the majority of the affected patients, development during childhood, pregnancy, and trauma have been identified as possible causes that might set off the defecation problems.<sup>12</sup> Most constipation complaints in HD patients are attributed to the disease itself. Nevertheless, it is possible that some HD patients have never mastered the act of defecation properly due to pre-operative obstipation and post-operative pain in the anal canal causing them to suffer from dyssynergic defecation as a result.<sup>13</sup> Thus besides being unable to relax their internal anal sphincter, HD patients might have the additional problem that their external anal sphincter contracts involuntary during defecation.

With all this in mind, we gave HD patients with severe persistent constipation a full anorectal examination, which consisted of an anal pressure profile, balloon retention test, and defecometry. We hypothesized that not all post-operative defecation complaints were attributable to HD and that dyssynergic defecation – for which viable treatment options are available – may increase the severity of constipation in these patients.

## METHODS

### **Patient selection**

Between May 2010 and May 2013, HD patients who presented with severe persistent constipation were given extensive anorectal function tests in order to diagnose the cause of their constipation. Inclusion criteria for undergoing anorectal function tests were histopathologically proven HD, proper treatment for HD, capable of understanding the tasks involved in the anorectal function tests, and severe persistent constipation that met the criteria of Rome III for functional constipation. Exclusion criteria were other evident causes of constipation in HD such as stenosis of the anastomosis or residual aganglionosis. A positive test result for dyssynergic defecation either meant inappropriate contraction of the pelvic floor or less than 20% relaxation of basal resting sphincter pressure with sufficient propulsive force during attempted defecation, as described in the Rome III criteria. Upon diagnosis, we referred patients to a specialized pelvic floor physiotherapist. We collected and reviewed the medical records and test results of these patients retrospectively.

In addition, we reviewed the patients' clinical records to collect variables such as the variant of HD, duration of constipation, applied treatment, relevant co-morbidities, episodes of Hirschsprung's associated enterocolitis, and post-operative complications.

### **Measuring equipment**

We recorded and analyzed the data with solar gastrointestinal high resolution manometry

equipment (Medical Measurement Systems, Enschede, the Netherlands), version 8.23.

Catheter 1: We used Unisensor K12981 solid-state (Boston type) circumferential catheters with an outer diameter of 12F with which we could measure circumferential pressure every 8 mm over a total length of 6.8 cm into the rectum.

Catheter 2: We used Unisensor K14204 catheters with an outer diameter of 14F with two microtip sensors to connect the rectal balloon, to inflate it, and to register the pressure inside the balloon. The solar gastrointestinal high resolution manometry equipment corrected the pressure in the rectal balloon for the resistance of the balloon itself, so that only the true pressure of the rectum was reported.

## Tests

### *The anal pressure profile*

For this test we carefully fixed Catheter 1 to the patient's buttocks near the anal canal with adhesive tape to prevent slippage during the procedure. We obtained basal anal pressure measurements while the subject was at rest while we recorded the maximum voluntary sphincter pressure during sustained and maximum pelvic floor contractions three times. We used the highest of the three values for our analysis.

### *Balloon retention test*

For this test we also carefully fixed Catheter 1 to the patient's buttocks near the anal canal with adhesive tape to prevent slippage during the procedure. Next to Catheter 1 we connected a collapsed, non-latex balloon to Catheter 2 and placed it in the rectum. While installing the catheters the patient was in the left lateral recumbent position but we administered this test with the patient sitting upright on a commode. When the patient was completely at rest we very slowly filled the balloon with water of 37°C (0.5 mL/second for children younger than 12 years or 1.0 mL/second for older children and adults). Meanwhile, we recorded the pressure in the rectal balloon and the volume inflated.

We asked the patients to retain the balloon as long as possible and to report when they experienced the first sensation (some rectal feeling), constant sensation (at home, the patient would go to the toilet), urge sensation (the patient would first go to the toilet before continuing any other activity), and the maximum tolerable sensation level. We stopped testing when the patient reached maximum tolerable sensation, *i.e.* when filling reached the limit of tolerance or maximum retainable volume if the patient had lost the balloon earlier. Then we emptied the balloon completely. This technique has been described previously.<sup>14-16</sup> It provides information about the extent to which the patient experiences rectal filling, rectal capacity, rectal compliance, and whether the anal canal responds to rectal filling by squeezing.<sup>14-16</sup>

According to our unpublished data and previous studies the normal value of the rectal volume at maximum tolerable sensation for a child of six years is 135 mL and for an adult it is 240 mL.<sup>17,18</sup>

### *Defecometry*

For this test the same types of catheter and the same patient position, *i.e.* sitting upright on a commode, were used as in the case of the balloon retention test. First, we filled the balloon with 50 mL of water of 37°C. We then asked the patient to evacuate the balloon. If the patient was unable to expel the balloon, we increased the volume into the balloon with 50 mL of water until the earlier measured urge sensation volume was reached. While the patient tried to evacuate the balloon, we measured the maximum rectal pressure, maximum anal sphincter pressure, and the time needed for evacuation. These variables provide insight into the parameters involved in the defecation process.<sup>19</sup> This test assesses whether the patient voluntarily contracts the external anal sphincter during defecation. If this is not the case, the patient suffers from dyssynergic defecation.

Earlier studies showed that normal subjects can expel a balloon filled with 100 mL of barium sulfate paste in a median of 7 seconds.<sup>20</sup> In our laboratory setting healthy subjects were able to expel the rectal balloon with 50 mL of water at body temperature. Failure to evacuate the balloon in one minute indicates an outlet obstruction which might possibly be caused by involuntary contraction of the external anal sphincter, otherwise known as dyssynergic defecation.<sup>21</sup>

### **Statistical analysis**

The data was analyzed with SPSS 20.0 for Windows (IBM SPSS Statistics, IBM Corporation, Armonk, NY). We used Q-Q plots to determine whether the values were distributed normally. Subsequently, normally distributed values were reported as means with standard deviations and abnormally distributed values were reported as medians with minimum and maximum values, as appropriate.

## RESULTS

During the three-year inclusion period we examined nine male and one female HD patient for severe constipation. The median age at the time of testing was 12 years and the range was 7 to 19 years. The mean duration of constipation was  $4.0 \pm 2.9$  years; the shortest duration was six months and the longest nine years. All ten patients had histopathologically proven HD based on an absence of ganglion cells in both submucosal and myenteric plexuses as determined by rectal suction biopsies. One patient had an ultra-

short variant of HD based on the absence of ganglion cells at 3 cm on rectal suction biopsy and the absence of the rectoanal inhibitory reflex. This patient received conservative treatment in the form of laxatives. The nine other patients all had a rectosigmoid variant of HD and had received Duhamel pull-through procedures. We examined the pathology reports of the nine patients treated surgically and found that each report mentioned distal aganglionosis with sufficient ganglion cells in the proximal anastomosed surface. Post-operative complications were limited to three patients: one case of stenosis of the

Anal pressure profile (n = 10)		Median (Range)	<b>Table 1</b> Anorectal function test results
Sphincter resting pressure (mm Hg)		70 (25 – 110)	
Maximum sphincter squeezing pressure (mm Hg)		220 (140 – 380)	
Defecometry (n = 10)		Median (Range)	
Expulsion possible (n)		6/10	
Expulsion volume (mL, n = 6)		75 (50 – 150)	
Expulsion time (s, n = 6)		10 (5 – 40)	
Maximum rectal pressure (mm Hg)		125 (70 – 185)	
Maximum anal sphincter pressure (mm Hg)		185 (125 – 285)	
Appropriate pelvic floor coordination (n)		0/10	
Balloon retention test (n = 10)		Median (Range)	
Basal rectal balloon pressure (mm Hg)		20 (10 – 25)	
First sensation	Rectal balloon pressure (mm Hg)	30 (15 – 40)	
	Rectal balloon volume (mL)	35 (10 – 295)	
Constant sensation	Rectal balloon pressure (mm Hg, n=9)	35 (25 – 45)	
	Rectal balloon volume (mL, n=9)	75 (25 – 205)	
Urge sensation	Rectal balloon pressure (mm Hg, n=9)	35 (25 – 70)	
	Rectal balloon volume (mL, n=9)	185 (70 – 565)	
Maximum tolerable volume	Rectal balloon pressure (mm Hg)	40 (25 – 80)	
	Rectal balloon volume (mL)	250 (80 – 845)	
Rectal compliance until maximum tolerable volume (mL/mm Hg)		14 (2.6 – 77.0)	

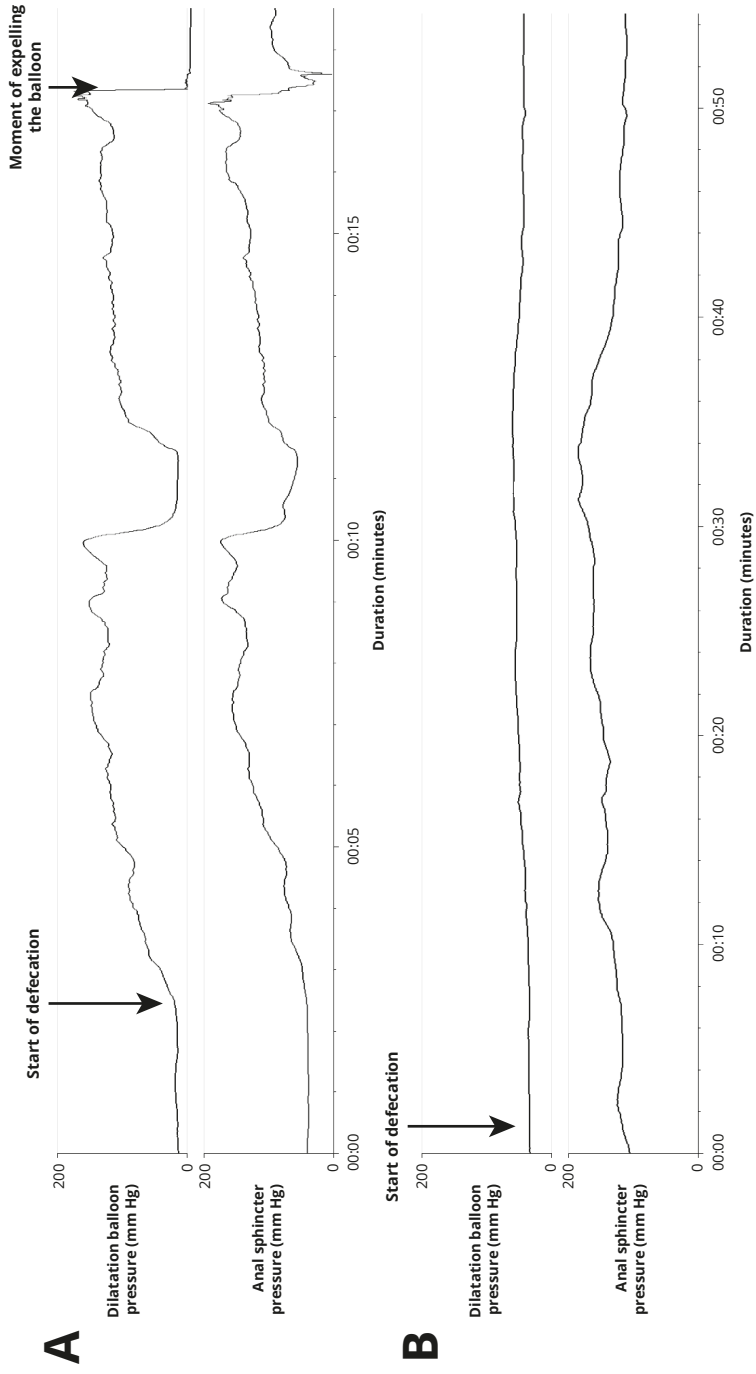


proximal anastomosis and two cases of anastomotic leakage. All three complications occurred directly following the surgical procedure and were treated subsequently. The time between the surgical corrections and subsequent complications and the anorectal function tests was 7, 9, and 17 years. The stenosis was treated with dilatation after which the symptoms decreased over time. The two cases of anastomotic leakage were treated with laparotomies combined with abdominal flushing and a temporary ileostomy in order to treat the persistent peritonitis. These two patients recovered fully during the course of several weeks. We found no statistically significant differences in the values between patients who had a post-operative complication and patients who experienced no post-operative complaints. Two patients experienced an episode of Hirschsprung's associated enterocolitis, one pre-operative and the other post-operative. All ten patients used laxatives and/or had enemas to cope with their constipation at the time the diagnoses were made. None of the patients had severe comorbidities troubling their defecation, apart from one patient who had Down's syndrome. This patient had a mild form of mental retardation and was capable of understanding the tasks involved in the anorectal function tests.

The anal pressure profile results showed an increased anal sphincter resting pressure with a median value of 70 mm Hg and a maximum squeezing pressure median of 220 mm Hg (Table 1). Furthermore, we found that in all ten patients the rectoanal inhibition reflex was absent, confirming their initial diagnosis of HD.

The defecometry test results showed that four out of ten patients completely failed to expel the balloon. And, in the event of expulsion, patients either required more volume or more time in order to succeed (Table 1 and Figure 2). Vastly increased pressures were seen in both the rectum and the anal sphincter (Table 1). This combination of sufficient propulsive force and increased anal pressure while squeezing led to the diagnosis of dyssynergic defecation. Proper coordination of defecation was absent in all ten patients we tested by means of defecometry.

The balloon retention test results showed that overall increased volumes were required to sense the balloon. We measured an increased first sensation volume of 295 mL in one patient and 150 mL in another, while normal values range from 30 mL to 50 mL depending on the patient's age. We found serious enlargement of the rectum in two patients with maximum tolerable volume values of 600 mL and 845 mL (Table 1). We found overall enlargement in eight out of ten patients (Figure 3). One patient failed to notice constant sensation and urge sensation but he did notice the maximum tolerable volume of 600 mL.

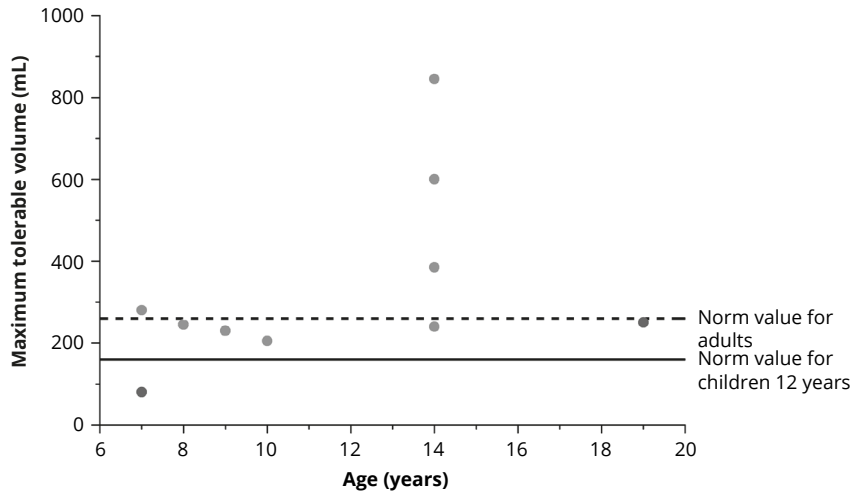


**Figure 2**

Anal pressure profiles of two patients with dyssynergic defecation.

A: The first patient was able to expel the rectal balloon despite contracting the anal sphincter.

B: The second patient failed to expel the rectal balloon due to heavy contraction of the anal sphincter.



**Figure 3**

Maximum tolerable volume measurements of all ten patients at the age of diagnosis. The black line represents the normal value for children of 12 years of age (160 mL). The dotted line represents the normal value for adults (260 mL).

## DISCUSSION

While most post-operative HD patients have good clinical outcomes and do not suffer from severe defecation complaints, a substantial number of patients seem to struggle with ongoing severe constipation.<sup>6-9</sup> One could argue that this is possibly caused by residual aganglionosis following surgical correction in patients treated for HD. Nevertheless, the appearance and number of ganglion cells in the proximal anastomosed surface was normal in all nine patients in our study who had been treated surgically. This meant that resection had been adequate. On the basis of rectal suction biopsies, the tenth patient was treated conservatively with laxatives for his ultra-short variant of HD. While this diagnosis, along with its treatment possibilities, remains undetermined,<sup>22</sup> pelvic muscle control was still completely absent in this patient. This pointed to dyssynergic defecation. In all ten patients the rectoanal inhibition reflex was absent as is seen in HD.<sup>23</sup> Consequently, all patients were, by definition, slightly constipated. Nevertheless, absence of this reflex does not explain why a substantial number of the post-operative HD patients suffer from more severe complaints than others, as the rectoanal inhibitory reflex is absent in all HD patients. We demonstrated that a piece of this complex puzzle could be dyssynergic defecation aggravating the already troublesome defecation. Following defecometry we diagnosed all ten patients in our study with dyssynergic defecation. This meant that in addition to a non-relaxing internal anal sphincter the external anal

sphincter of these patients also contracted paradoxically during defecation. While most constipation complaints in HD patients are attributed to the disease itself, it is possible that a significant number of HD patients have never mastered the act of defecation properly and, consequently, these patients have dealt with dyssynergic defecation since early childhood. While hard evidence for this theory is still lacking, the extremely enlarged rectums we found in two patients seem to indicate long-standing constipation caused by dyssynergic defecation.

Several reasons for the onset of dyssynergic defecation have been described and one of these factors could be responsible for its onset in post-operative HD patients. Firstly, Rao *et al* suggested that the onset of dyssynergic defecation symptoms during childhood, in otherwise healthy patients, may be due to faulty learning of proper defecation.<sup>12</sup> Secondly, Hyman suggested that pain in the rectum after surgery and washouts may lead to patients avoiding bowel movement as these are often associated with increased pain.<sup>13</sup> A third and final possibility might be that post-operative complications or Hirschsprung's associated enterocolitis could make patients more prone to developing dyssynergic defecation, especially since these are associated with additional treatment and sometimes even with a redo of the surgical procedure. Our results showed that three out of ten patients (30%) had a post-operative complication. Even though this is a relatively high number, the small number of patients in our study makes it difficult to statistically define post-operative complications as the pre-determining factor for dyssynergic defecation. It still remains unclear whether dyssynergic defecation in HD patients with severe complaints is caused by congenital disease, surgical correction for HD, or that it develops at a much later age regardless of the patients' medical history. Moreover, further research on a non-selected cohort of both adequately and poorly functioning HD patients is necessary to determine the exact incidence of dyssynergic defecation in the entire HD disease population.

Persistent constipation due to dyssynergic defecation was shown to have a negative influence on overall quality of life as it significantly impairs social life, sex life, work life, and family relationships.<sup>12</sup> Our data demonstrated the added risk of serious enlargement of the rectum with decreased elasticity of the rectal wall due to long-standing constipation caused by dyssynergic defecation. We found one patient who measured a maximum tolerable volume of 845 mL at the age of ten years. Absence of the rectoanal inhibitory reflex, as is seen in HD, will probably lead to a slight increase in rectal volume in all patients. Some of these values are so vastly increased, however, that they are too high even for HD. Moreover, dyssynergic defecation in HD patients may initiate a vicious circle of fear of pain during defecation and defecation avoidance behavior that may eventually cause overflow incontinence.

Nowadays, dyssynergic defecation is treated with a high fiber diet, laxatives, and biofeedback training.<sup>24</sup> Several reports already recognize the positive effects of biofeedback therapy on constipation in non-HD patients.<sup>25-29</sup> Besides improving the defecation process, biofeedback training reportedly also improves overall quality of life.<sup>30</sup> One case report showed that biofeedback training has great potential for patients with constipation and fecal seepage in the presence of HD.<sup>31</sup> In other colorectal diseases, such as inflammatory bowel diseases, dyssynergic defecation was shown to be treatable by biofeedback therapy.<sup>11</sup> This also points out the great potential of this therapy in HD patients with severe constipation. Further research on the effects of biofeedback training in this relatively young group of patients, who have dyssynergic defecation in addition to HD, is in progress. It will have to show to what extent it is possible to treat the complaints characteristic of this particular group of patients.

### **Conclusions**

It is important to consider the diagnosis of dyssynergic defecation when dealing with severe constipation in post-operative HD patients. Our results show that a number of HD patients have problems with wrongly contracting their external anal sphincters, in addition to a non-relaxing internal anal sphincter characteristic for HD. If dyssynergic defecation is diagnosed at an early age, viable treatment options are available that may prevent irreversible long-term complications, such as significant enlargement of the rectum and, eventually, overflow incontinence.

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