

Achieving Social Bowel Continence in Children With Hypermotility After Repair of Anorectal Malformations and Hirschsprung

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Contact Hours

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Abstract: Children presenting to the Pediatric Colorectal Bowel Management clinic with soiling after surgical repair of anorectal malformations or Hirschsprung can be categorized into two groups: Most are constipated and managed with laxatives and/or enemas, whereas the second subset has frequent loose stools. This second group, children with multiple loose stools, presents a unique challenge for the achievement of social bowel continence. We present three case studies illustrating various clinical presentations of incontinence with loose stools, along with dietary and pharmacologic management strategies and a treatment algorithm. Children who soil with loose stools can achieve social continence using a combination approach using toileting supports, diet manipulation, fiber supplementation, medications, and small-volume enemas.

Key Words: anorectal malformation, antidiarrheals, bowel management, Hirschsprung, hypermotility, imperforate anus

Fecal incontinence is a devastating condition that adversely affects quality of life in children, resulting in increased rates of school absence and failure, disability, and psychosocial comorbidities. Children with congenital anorectal malformations and Hirschsprung

disease are at risk of fecal incontinence, with overall incontinence rates as high as 25% in anorectal malformations (Bischoff et al., 2009; Kaul et al., 2011). Comprehensive bowel management has been shown to improve outcomes in fecal incontinence (Bischoff et al., 2009). Nearly all practitioners working in pediatric surgery will have encountered children with fecal incontinence as an outcome of colorectal repair. Most of these children are constipated and are optimized for social bowel continence with laxative or enema therapy. More rare, and often more challenging, are the children with hypermotility and frequent loose stools (see definition of hypermotility below). Social continence can be more challenging to achieve for children in the hypermotile group.

Definitions, and Inclusion and Exclusion Criteria

Many practitioners rely on colonic manometry with the measurement of high-amplitude propagating contractions to establish the diagnosis of hypermotility, but for the purpose of this article, hypermotility is defined as having greater than four bowel movements in a 24-hour period and having a nondilated, spastic, or short colon per contrast enema.

Included in this article are children with surgically repaired anorectal malformations and surgically repaired Hirschsprung disease. Excluded from this article are patients with small bowel loss resulting in short bowel syndrome, those with inflammatory bowel disease, and children on tube feedings. We also excluded children with functional constipation and fecal incontinence who do not otherwise meet the criteria of having Hirschsprung or anorectal malformations. The treatment algorithm included here may be of benefit to some of these and other populations not specifically

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included in our target population. Social bowel continence is defined as cleanliness without bowel accidents with the ability to wear regular underwear. Many children with hypermotility do not have voluntary bowel continence and thus rely on a combination of dietary manipulation, medications, and small-volume irrigations or enemas to achieve social bowel continence. The goals of bowel management in this population are defined as fewer bowel movements in a 24-hour period, decreased accidents, and improved social continence and quality of life as reported by the child and family (Rawashdeh et al., 2012). The following three case studies illustrate varying presentations of hypermotility with treatment plans to improve social continence. Please note that this is a case review only, not a study. Institutional review board approval was not obtained or required.

CASE STUDY 1

K. L. is a 10-year-old adopted Native American boy diagnosed with total colonic Hirschsprung disease in early infancy. At 1 week old, he had a diverting loop ileostomy. At the age of 5 months, he underwent appendectomy and multiple leveling biopsies with transition zone at the ileocecal valve with total colectomy with Soave pull-through of the terminal ileum (straight ileoanal anastomosis).

Additional History

K. L.'s additional history was as follows: fetal alcohol syndrome, neurodevelopmental delays, ASD (atrial septal defect) repaired at the age of 5 years, lactose intolerance, and recurrent enteritis with hospitalizations requiring intravenous antibiotics and total parenteral nutrition. With illness, he developed vomiting with abdominal distention and liquid stools greater than 10 times per day. He required multiple hospital admissions for dehydration during illness episodes. K. L. was seen in bowel management program at the age of 7 years. He was fully incontinent of stool with explosive diarrhea 6–10 times per day. He had intermittent gaseous distention.

Workup

Contrast enema was normal—no stricture or dilation. There was no significant stool burden. The workup was consistent with ileoanal anastomosis. Examination under anesthesia with majority of the dentate line was not established. The anus was in a closed position with moderate contracture found using a Pena muscle stimulator. Digital examination revealed no stricture or obstruction.

Pathologic Review

Aganglionosis was present from the distal rectum to the ascending colon. There was a normal distribution of

ganglia in the cecum and ileum; however, ganglia are abnormally large.

Recommendations

1. Small-volume saline enemas (200 ml) rectally twice a day
2. Cholestyramine 2 g orally three times a day
3. Loperamide 4 mg PO once a day (midday)
4. Loperamide 6 mg PO twice a day (a.m./p.m.)
5. Lomotil (diphenoxylate) 0.75 mg PO TID
6. Metronidazole 500 mg PO TID \times 7 days 1 week per month for overgrowth suppression
7. Skin barrier cream as needed for perianal breakdown

Follow-up

By family report, K. L. has had zero episodes of enteritis and zero hospitalizations on his current bowel program. His stools have decreased to three to four times per day. Quality of life significantly improved by family's definition.

CASE STUDY 2

A. H. is now a 14-year-old male adolescent with total colonic Hirschsprung disease. His original surgical repair included a Duhamel ileorectal pull-through at the age of 11 months. He was seen and hospitalized at the age of 9 years for recurrent symptoms of enterocolitis: abdominal pain, nausea, vomiting after pull-through, treated with hospitalization, nasogastric tube, nothing by mouth, intravenous hydration, and antibiotics. He had three hospitalizations for enterocolitis and an additional two hospitalizations for adhesive small bowel obstruction. At presentation, he was managed with daily rectal irrigations to decrease further enteritis episodes.

Workup

Examination under anesthesia revealed normal dentate line with normal anal canal. The anus was widely patent with no stenosis or stricture. The Duhamel pouch was intact. Rectal biopsy with ganglion cells was done by a pathologic review. A normal acetylcholinesterase and calretinin staining pattern was found. There were no hypertrophic nerves.

Recommendations

Because of his recurrent symptoms of enterocolitis, he underwent a revision of his pull-through to remove his Duhamel pouch and convert him to an ileoanal anastomosis. The distal 8 centimeters of the ileum underwent tapering.

1. Loperamide by mouth and an enema before bedtime consisting of 300 ml of normal saline and 5 ml of glycerin.
2. It was recommended that he adhere to a constipating diet with fiber and cholestyramine; however, A. H. did

not tolerate the fiber or cholestyramine because of texture and was not compliant with the constipating diet. Fortunately, the oral loperamide and small-volume enema at bedtime kept him clean for a long period with improved quality of life as reported by the child and his family.

Follow-up

In a short term, after revision and bowel management, he was stooling four to five times per day with control and has reported one loose stool accident occasionally at night while sleeping. He no longer needed daily irrigations. At the last visit, he has not wanted parental help with enema, so he stopped the nightly enema and continued on oral loperamide with the addition of Lomotil. Lomotil was started at 5 mg by mouth and increased to the maximum daily dose (see Figure 1). Without enemas, he experienced more stooling accidents during his sleep but stayed clean throughout the day.

The family asked about an anal irrigation system, which is an enema kit with a hand pump manufactured for independence with anal washouts. The hope was that this could provide A. H. with privacy and independence in administering his enemas without requiring further surgery (such as a Malone neoappendicostomy categorizable channel or a tube cecostomy). After discussing this with the team, the pressure of the system was tested. The consensus was, if used, to use with caution, as there are no data to support safe use in this population when irrigating the ileum rather than the colon.

Further Workup

A. H. next had a contrast study that showed inflammation of his intestine. On the basis of this and recent history of several episodes of enterocolitis and bacterial overgrowth, a 2-week trial of Proctofoam was prescribed, which did not resolve his symptoms. Next, Xifaxan 200 mg three times a day was tried in an effort to treat the inflammation and resolve symptoms. We have found this successful in other patients with similar symptoms.

CASE STUDY 3

C. C. is a 3-year-old boy adopted from China with repaired anal atresia, complicated by a “large amount” of bowel removed during original operative repair.

Additional History

Magnetic Resonance Imaging (MRI) showed a tethered cord, which required subsequent surgical tethered cord release. C. C. was fully incontinent of stool with a severe diaper rash. His mother reported, “He stools nearly constantly, sometimes liquid and other times more pasty.”

Workup

Contrast enema revealed “small segment of colon versus dilated terminal ileum: unable to differentiate.” No significant stool burden was found as per x-ray results.

Recommendations

1. Small-volume daily rectal enema (200-ml saline plus 10-ml glycerin)
2. Citrucel fiber, one tablespoon (2 g) three times per day with meals
3. Loperamide, 2 mg by mouth once a day (30 minutes before breakfast)
4. Loperamide, 1 mg by mouth twice a day (30 minutes before lunch, 30 minutes before dinner)
5. Lomotil (diphenoxylate) 1 mg by mouth twice a day
6. Diet considerations: low sugar; high in soluble fiber; avoid raw fruits and vegetables; avoid spicy or greasy foods.
7. Skin barrier cream as needed for perianal skin breakdown

Follow-up

By family report, stools have decreased to two to four times per day. Quality of life significantly improved by family's definition.

Next Steps

Per medication algorithm, increase Lomotil as needed with the goal of no stooling between enemas.

These three cases illustrate the multiple medication and enema adjustments needed to find a regimen that works for a child and his or her family, along with the complex and refractory nature of hypermotility and the range of treatments attempted to improve social continence outcomes.

MEDICATIONS

Many medications reported in the adult literature have shown effectiveness in slowing or reducing loose stools (Ehrenpreis, Chang, & Eichenwald, 2007). It is unclear if good results in adult populations translate to pediatric populations. There is a remarkable absence of pediatric studies in the literature; therefore, it is imperative to conduct more pediatric research and translate and disseminate findings. See the medication table (Table 1) for medications found to be the most useful with these cases. Thickeners such as fibers and pectin help provide bulk and substance to liquid stools, allowing for fewer stools of a denser consistency and reducing overall number of stools (Markland et al., 2015). Again citing adult literature, decreased intestinal motility, decreased frequency, and more formed stools help improve continence (Scarlett, 2004). Loperamide and diphenoxylate have both been shown to reduce diarrhea and increase anal sphincter tone (Hanauer, 2008; Sze & Hobbs,

MEDICAL MANAGEMENT ALGORITHM FOR HYPERMOTILITY

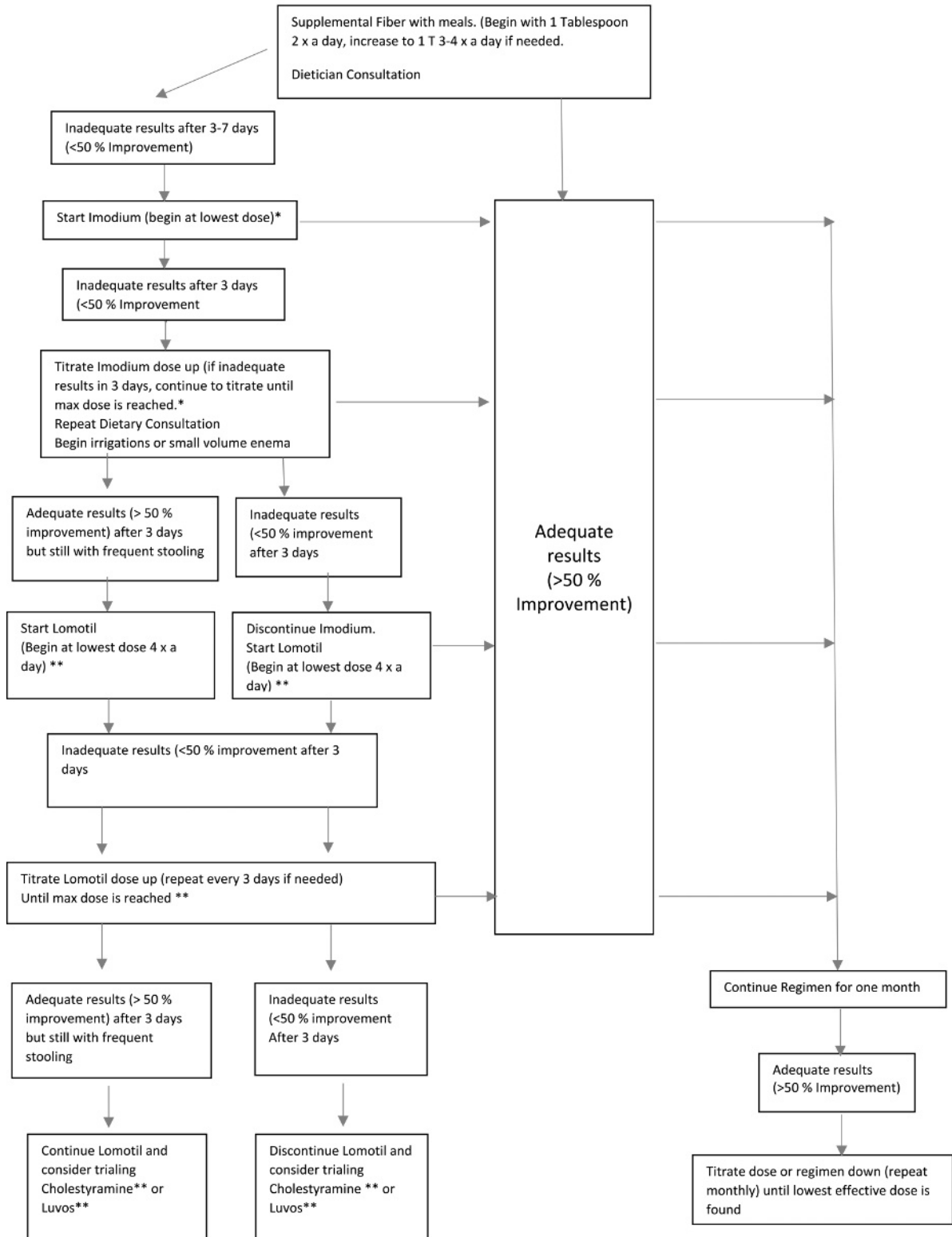


FIGURE 1. Treatment Algorithm.

Table 1: Medication Table

Medication	Mechanism of Action	Starting Dose	Titration Dose	Maximum Dose	How Supplied	Comments
Fiber supplements <input type="checkbox"/> Pectin <input type="checkbox"/> Citrucel	Reduced transit time, increased rates of defecation/stool frequency (an increase in stool weight is not a beneficial physiological effect per se; it needs to be associated with a change in defecation frequency) ^a	1 tablespoon (1 tablespoon equals 2 g) 2× a day	Increase by 1 tablespoon a day	3–4 tablespoons a day as tolerated	Pectin in powder form only Citrucel in powder form also available as a fiber gummy but usually not as effective	<ul style="list-style-type: none"> • Dissolve powder in only 2–3 oz of fluid and drink quickly before it gels. • May sprinkle on a small amount of any food (1–2 tablespoons). Make sure the entire amount of food is ingested. • Gummies have been found to be successful in a very small amount of patients and 2–3 gummies should be used for each dose. • 100% water-soluble fiber supplement is the most successful.
Imodium/Loperamide	Acts directly on circular and longitudinal intestinal muscles, through the opioid receptor, to inhibit peristalsis and prolong transit time; reduces fecal volume; increases viscosity, and diminishes fluid and electrolyte loss; shows antisecretory activity. Loperamide increases tone on the anal sphincter. ^b	0.8 mg/kg per day in 1–2 divided doses	0.8–0.24/kg per day Increase slowly by 1 mg at a time.	The maximum dose used at our institution is 0.24.	<ul style="list-style-type: none"> • 2-mg tablet • 1 mg/5-ml liquid • 1 mg/7.5-ml liquid 	<ul style="list-style-type: none"> • Take 1 hour before meals and/or at bedtime • Administration times can be tailored to when the patient's accidents most often occur. • Divided doses are recommended but can be given all at once depending on when the patient's accidents most often occur. • There have been limited studies on using higher doses: 0.5–1.0 mg/kg per day <ul style="list-style-type: none"> □ Cardiac arrhythmias noted in doses higher than recommended. • Be aware that liquid contains sugar, which can contribute to hypermotility and accidents. • Can cause drowsiness and dizziness, but these symptoms may subside once the body adjusts to the medication. • Keep hydrated.
Lomotil/diphenoxylate and atropine	Acts directly on circular and longitudinal intestinal muscles, through the opioid receptor, to inhibit peristalsis and prolong transit time; reduces fecal volume, increases viscosity, and diminishes fluid and electrolyte loss; shows antisecretory activity. Loperamide increases tone on the anal sphincter. ^b	Children 2–12 years old: 0.3–0.4 mg/kg per day in four divided doses—to start at the lowest dose, give one of the four doses per day. Adults (13 years and above): 5 mg once daily	Manufacturer's recommendations: <2 years: not recommended 2 years (11–14 kg): 1.5–3 ml four times a day 3 years (12–16 kg): 2–3 ml four times a day 4 years (14–20 kg): 2–4 ml four times a day 5 years (16–23 kg): 2.5–4.5 ml four times a day 6–8 years (17–32 kg): 2.5–5 ml four times a day 9–12 years (23–55 kg): 3.5–5 ml four times a day		5-mg tablets 1 mg/2-ml liquid	<ul style="list-style-type: none"> • Give with food. • Only liquid is recommended for children under the age of 13 years. • Be aware that the liquid formulation contains sugar and may contribute to hypermotility/accidents. • If chronic diarrhea does not improve within 10 days at maximum daily doses, diphenoxylate is not likely to be effective.

(continues)

Table 1: Medication Table, Continued

Medication	Mechanism of Action	Starting Dose	Titration Dose	Maximum Dose	How Supplied	Comments
Cholestyramine resin/Questran	Forms a nonabsorbable complex with bile acids in the intestine, releasing chloride ions in the process; inhibits enterohepatic reuptake of intestinal bile salts and thereby increases the fecal loss of bile salt-bound low-density lipoprotein cholesterol.	240 mg/kg per day In three divided doses, give one dose per day to start.	Increase frequency of starting dose to 2 × a day, 3 × a day, and then may titrate dose up as tolerated.	8 g per day	Granules 4-g packets Multidose powder 4 g/scoop	<ul style="list-style-type: none"> Mix in 2–6 oz of noncarbonated beverage, drink quickly. Do not ingest in powder form. Do not hold in mouth for a prolonged period; may cause tooth discoloration or enamel decay. Peak effect: 21 days Give with meals. May be divided in two, three, or four doses per day. Administer other drugs including vitamins or mineral supplements at least 1 hour before or at least 4–6 hours after cholestyramine. May decrease the absorption of folic acid, calcium, fat-soluble vitamins (ADEK) and iron. Therefore, baseline labs are needed and recommend monitoring while taking. May need supplementation.
Luvos	This is a pure natural product made from a stone, loess, found in the earth. Mechanism of action is unclear and not evaluated by the FDA.	1–2 teaspoons, twice daily	May use a third teaspoon every day	"Several teaspoons"	Powder (caplets)	<input type="checkbox"/> Adult dosing available only
Levsin/Hyoscyamine	Bowel antispasmodic used for reduction of symptoms in colitis and irritable bowel syndrome.	0.125 mg Half tablet twice a day	1–2 tablets every 4 hours	Maximum of 12 tablets in 24 hours	Tablets Elixir Extended-release tablets	<ul style="list-style-type: none"> Half-life: 2–3.5 hours

^aFood and Drug Administration (FDA) Dietary Fiber Guidance and Its Impact on the Food Industry. Intertek Group plc. December 2, 2016. Retrieved from <http://www.intertek.com/blog/2016-12-02-fda/>.

^bCCHMC Formulary of Medications. Lexicomp. Retrieved from http://www.crlonline.com/lco/action/doc/retrieve/docid/chicin_f/263296.

2009). Cholestyramine is also a useful adjunct for its ability to reduce stool frequency, improve anal sphincter pressures, and thicken stool (Remes-Troche, Ozturk, Philips, Stessman, & Rao, 2008).

NUTRITIONAL CONSIDERATIONS

In practice, we have found treating the child's diarrhea, reintroducing foods slowly, and increasing soluble fiber in the diet to be beneficial for children with hypermotility.

Diarrhea Recommendations

The following recommendations can help relieve diarrhea (Academy of Nutrition and Dietetics, 2017):

- Drink plenty of fluids to avoid dehydration.
- Avoid drinks that are high in sugar such as juices and sodas.
- Eat smaller, more frequent meals instead of three large meals.
- Avoid spicy foods if they make symptoms worse.
- Avoid foods high in fat, fiber, and added sugar.
- Avoid foods that are really hot or really cold.
- Avoid fried, greasy foods.
- Avoid sugar alcohols.
- Avoid raw fruits and raw vegetables.
- Avoid food and drinks with caffeine.

The BRAT diet is no longer recommended when children experience diarrhea because it is not considered a balanced diet and is low in calories (American Academy of Pediatrics Committee on Nutrition, 2014; Duro & Duggan, 2007).

Reintroducing Foods

Once diarrhea improves, slowly add foods back into the diet. Introduce one new food at a time. If stool output increases after eating a newly introduced food, temporarily eliminate that food and continue introducing new foods while monitoring for tolerance.

Increasing Soluble Fiber

Soluble fibers can cause gel formation leading to delayed gastric emptying and an increase in transit time (Groff, Gropper, & Smith, 2009). Pectin is commonly used to help manage hypermotility as it forms a viscous gel within the gastrointestinal tract causing a delay in gastric emptying. In practice, pectin is added to formula or liquids up to 3% of the total volume (Wessel & Kocoshis, 2007). Slowly increasing food sources of soluble fiber in the diet such as oats, barley, citrus fruits, apples, and legumes can also be beneficial in managing fecal incontinence (Colavita & Andy, 2016).

Everyone Tolerates Foods Differently

It is important to note that not everyone tolerates food the same way. What works for one child may not work for another. Keeping a food journal that

records what the patient eats, when they eat, and symptoms can be very helpful in determining which foods a patient tolerates and which foods cause symptoms to worsen. It is recommended to work with a registered dietitian on an individualized nutrition plan optimized for improved individual outcomes.

TREATMENT ALGORITHM NOTATION

Before Algorithm

For all patients, a water-soluble contrast enema is recommended to determine if there is stricture or any anatomical problem contributing to hypermotility. Before medical management specifically for Hirschsprung, an examination under anesthesia is recommended to confirm presence or absence of the dentate line.

After Algorithm

If at any time during treatment modalities, as outlined in the algorithm, you suspect symptoms of inflammation, consider further workup to confirm followed by treatment with anti-inflammatories if indicated. Similarly, if bacterial overgrowth is concerning, consider using an appropriate antibiotic therapy.

CONCLUSION

Hypermotility is a devastating condition, which adversely affects quality of life for the incontinent child and his or her family. The group of children with hypermotility is a smaller population of bowel management patients but can be more challenging to manage. It is important to fully evaluate the child to address underlying causes for ongoing symptoms. Each case presents with unique concerns that require individualized treatment interventions. A bowel management plan that is manageable by one family may be completely unacceptable to another family; thus, individualizing a strategy is essential. The treatment recommendations are offered as a guide when creating the care plan for the hypermotile child. An effective bowel management regimen to minimize the number of daily bowel movements and number of bowel accidents related to hypermotility is the goal to improve quality of life. This plan is enhanced using a multimodal approach with diet, fiber, and medications to achieve decreased symptoms and improved quality of life as defined by the patient and his or her family.

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