



Surgical decision-making in the management of children with intractable functional constipation: What are we doing and are we doing it right? ☆☆☆



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ABSTRACT

Background: Children with intractable functional constipation (FC) may eventually require surgery, often guided by motility testing. However, there are no evidence-based guidelines for the surgical management of intractable FC in children. **Aim:** To assess the diagnostic and surgical approach of pediatric surgeons and pediatric gastroenterologists towards children with intractable FC.

Methods: A survey was administered to physicians attending an international conference held simultaneously in Columbus (Ohio, USA) and Nijmegen (the Netherlands). The survey included 4 questions based on cases with anorectal and colonic manometry results.

Results: 74 physicians completed the questionnaire. Anorectal manometry was used by 70%; 52% of them would consider anal sphincter botulinum toxin injections for anal achalasia and 21% would use this to treat dyssynergia. Colonic manometry was used by 38%; 57% of them reported to use this to guide surgical decision-making. The surgical approach varied considerably among responders answering the case questions based on motility test results; the most commonly chosen treatments were antegrade continence enemas and anal botulinum injections. **Conclusion:** Surgical decision-making for children with intractable FC differs among physicians. There is a need for clinical guidelines regarding the role of anorectal and colonic manometry in surgical decision-making in children with intractable FC.

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Childhood constipation is a common problem in pediatric healthcare. It is characterized by infrequent bowel movements (<3 per week), hard and/or large stools, painful defecation and it is often accompanied by abdominal pain [1]. Approximately 75–90% of children with chronic constipation seen by a pediatric gastroenterologist suffer from fecal incontinence, caused by the leakage of soft stools around a large and hard fecal mass accumulated in the rectum [2]. In most cases, an organic cause for constipation is not found and affected children are diagnosed with functional constipation (FC). The reported prevalence of FC among children ranges from 0.7% to 29.6% with a mean female/male

ratio of 2.1:1 [3]. A subset of patients with FC experience severe and long-lasting symptoms that respond poorly to conventional behavioral, dietary and pharmacological management, these children are considered to have intractable FC [1]. In tertiary care centers, 50% of children referred to a pediatric gastroenterologist are still symptomatic after 5 years, and 20% still struggle with symptoms after 10 years [1]. Symptoms can even persist into adulthood despite intensive laxative treatment [4]. Persistent FC symptoms negatively affect quality of life in multiple ways (e.g., social interactions, school achievements, self-esteem) and account for significant associated healthcare costs [5–7].

Children with intractable FC may eventually require alternative therapeutic interventions including surgery. The most recent joint guidelines from the European Society of Pediatric Gastroenterology, Hepatology and Nutrition (ESPGHAN) and the North American Society of Pediatric Gastroenterology, Hepatology and Nutrition (NASPGHAN) state that colonic manometry may be indicated in children with intractable FC because evaluation of colonic motility may help guide surgical management [1]. This recommendation is mostly based on expert opinion and relies on the results of few retrospective studies that reported that normal colonic manometry predicts a successful response to

Abbreviations: ACE, antegrade continence enemas; FC, functional constipation; RAIR, rectoanal inhibitory reflex.

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antegrade continence enemas (ACE) [8] and abnormal manometry was associated with successful surgical interventions [9]. However, there are no guidelines that clearly recommend which surgical treatment should be performed based on manometry results. Without such guidelines, it is likely that the surgical approach towards children with severe FC differs among centers and among individual surgeons.

Surgery is usually considered a treatment of last resort and is generally performed with a step-up approach, beginning with the least invasive treatment and progressing to more invasive interventions only if needed. The choice of what type of surgery to perform is usually determined based on a comprehensive evaluation of the colonic and anorectal anatomy and physiology, although this evaluation may differ among centers. Generally, medical care for children with intractable FC is a joint venture, where both pediatric gastroenterology and pediatric surgery are involved. Intrasphincteric botulinum toxin injections and ACE are considered to be less invasive surgical strategies and are commonly employed in the treatment of intractable FC [10]. More invasive surgical interventions include colonic resection and diversion of the colon via an ostomy, either an ileostomy or a colostomy [10,11]. In their recent systematic review, Siminas et al. concluded that the evidence to support surgical interventions for intractable FC in children is mostly of low quality [10]. Siminas et al. provided a comprehensive overview of the literature and showed that there is no consensus regarding the diagnostic-work up that is required for surgical decision-making [10]. Therefore, the aim of this study was to assess the diagnostic and therapeutic approach of pediatric surgeons and pediatric gastroenterologists towards pediatric patients with intractable FC.

1. Material and methods

We developed a survey and administered it to physicians attending the 2015 Pediatric Colorectal, Motility and Pelvic Reconstruction Conference, held simultaneously in Columbus, Ohio (USA) and Nijmegen (the Netherlands) in November 2015. In total, 265 physicians from different specialties (pediatric and adult surgery, gastroenterology, urology and radiology) attended the conference at both locations; 147 in Columbus and 118 in Nijmegen. In Columbus, the attendees included 71 physicians from pediatric surgery (36 faculty members, 35 fellows/residents) and 29 from pediatric gastroenterology (21 faculty members, 8 fellows/residents). In Nijmegen, the distribution of physicians according to their specialties was unknown (89 faculty members, 29 fellows/residents).

The questionnaire involved 19 multiple choice questions on work experience, routine diagnostic workup in children with FC, use of non-pharmacological and pharmacological treatment for FC and use of surgery in intractable FC (Appendix). In addition, the survey included multiple choice questions on 4 theoretical cases of children with intractable FC; anorectal and colonic manometry results were given and responders were asked which surgical treatment they would choose. For each question, responders had the option to an open answer in case the multiple choice answers were insufficient.

For this study, only surveys answered by physicians from pediatric surgery and pediatric gastroenterology were included. The results are represented as percentages of the total number of responders unless otherwise specified.

2. Results

The survey was completed by 74 physicians working in pediatric surgery or gastroenterology in 16 different countries: 55 (74%) worked in pediatric surgery (29 faculty members, 23 fellows, 3 residents) and 19 (26%) in pediatric gastroenterology (14 faculty members, 5 fellows). The experience of these physicians was reported as follows: 0–5 years (43%), 5–10 years (25%), 10–15 years (8%) and > 15 years (25%). Results are presented separately for pediatric surgery (surgery) and pediatric gastroenterology (GI) in all tables.

2.1. Diagnostic work-up

Table 1 summarizes the data from the questions related to the diagnostic approach. Most responders utilized digital rectal examination in the evaluation of children with FC. Plain abdominal X-rays were obtained by the majority of physicians. In total, 62/69 of responders who used plain abdominal X-rays did not use a scoring system (e.g., Barr, Leech or Blethyn (12)) to score the radiographs. Colonic transit studies were used infrequently, whereas the use of contrast enemas was reported to be more common.

2.1.1. Anorectal manometry

Anorectal manometry was used routinely by 15 responders while 37 responders used it occasionally (Table 1). Responders who used anorectal manometry either routinely or occasionally (n = 52) utilized this test to rule out Hirschsprung's disease (65%; 83% in GI and 56% in surgery), to diagnose anal achalasia (58%; 78% in GI and 47% in surgery), to detect dyssynergia (56%; 67% in GI and 50% in surgery), to assess sphincter integrity (50%; 50% in GI and 50% in surgery) and for guidance prior to possible pelvic floor surgery (27%; 22% in GI and 29% in surgery). Out of the 52 physicians utilizing anorectal manometry, 52% (67% in GI and 44% in surgery) would consider anal sphincter botulinum toxin injections for anal achalasia and 21% (28% in GI and 18% in surgery) would use it to treat dyssynergia.

2.1.2. Colonic manometry

Colonic manometry was used routinely by 8 and occasionally by 20 responders (Table 1). Among the responders who used colonic manometry (n = 28), 61% (91% in GI and 41% in surgery) employed it to differentiate neuropathic from myopathic dysmotility, 57% (64% in GI and 53% in surgery) to guide surgical decision-making, 54% (55% in

Table 1

Diagnostic tools and frequency of use: n (%). Total number of responders: 74.

	Routinely	Occasionally	Never	Not available	Missing
Digital rectal examination	50 (68)	21 (28)	1 (1)	-	2 (3)
GI	12 (63)	7 (37)	0	-	0
Surgery	38 (69)	14 (26)	1 (2)	-	2 (4)
Abdominal X-ray (plain)	40 (54)	29 (39)	5 (7)	0	0
GI	5 (26)	12 (63)	2 (11)	0	0
Surgery	35 (64)	17 (31)	3 (6)	0	0
CTT (radiopaque markers)	10 (14)	36 (49)	19 (26)	5 (7)	4 (5)
GI	1 (5)	16 (84)	2 (11)	0	0
Surgery	9 (16)	20 (36)	17 (31)	5 (9)	4 (7)
CTT (scintigraphy)	2 (3)	7 (10)	37 (50)	20 (27)	8 (11)
GI	0	2 (11)	13 (68)	4 (21)	0
Surgery	2 (4)	5 (9)	24 (44)	16 (29)	8 (15)
Anorectal manometry	15 (20)	37 (50)	11 (15)	10 (14)	1 (1)
GI	4 (21)	14 (74)	0	1 (5)	0
Surgery	11 (20)	23 (42)	11 (20)	9 (16)	1 (2)
Colonic manometry	8 (11)	20 (27)	22 (30)	18 (24)	6 (8)
GI	3 (16)	8 (42)	4 (21)	4 (21)	0
Surgery	5 (9)	12 (22)	18 (33)	14 (26)	6 (11)
Contrast enema	26 (35)	38 (51)	7 (10)	1 (1)	2 (3)
GI	2 (11)	15 (79)	2 (11)	0	0
Surgery	24 (44)	23 (42)	5 (9)	1 (2)	2 (4)
Defecography	5 (7)	28 (38)	24 (32)	12 (16)	5 (7)
GI	0	5 (26)	10 (53)	4 (21)	0
Surgery	5 (9)	23 (42)	14 (26)	8 (15)	5 (9)
Transabdominal ultrasound	7 (10)	18 (24)	37 (50)	5 (7)	7 (10)
GI	0	1 (5)	16 (84)	2 (11)	0
Surgery	7 (13)	17 (31)	21 (38)	3 (6)	7 (13)
Transrectal ultrasound	0	10 (14)	37 (50)	10 (14)	17 (23)
GI	0	1 (5)	13 (68)	3 (16)	2 (11)
Surgery	0	9 (16)	24 (44)	7 (13)	15 (27)

Abbreviations: CTT, colonic transit time; GI, gastroenterology.

The boldface entries represent the total number of responders who preferred a certain treatment for the specific case.

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