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Enema-Induced spastic left colon syndrome: An unintended consequence of chronic enema use

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ABSTRACT

Background/purpose: Enemas have become a common practice for treating fecal incontinence and severe constipation. Several patients receiving enemas complained of severe, colicky, abdominal pain during enema administration and complained that the duration for fluid to pass was progressively increasing. Contrast studies showed a startling picture of severe right colon dilatation and a spastic, narrow, left colon. An investigation was started to seek the origin and possible management of this condition.

Methods: Medical and radiologic records were reviewed retrospectively, with emphasis on the type and ingredients of enemas used, the duration the patients had been receiving enemas, and their original diagnosis. A literature review was done on previous reports of this condition and publications related to long-term use of enemas.

Results: This series included 22 patients (average age, 19.6 years; range, 8–54) with fecal incontinence due to anorectal malformations (10 cases), myelomeningocele (5), cloaca (2), severe colonic dysmotility (2), Hirschsprung's disease (2), and sacrococcygeal teratoma (1). The average duration of enema use was 13.7 years (range, 4–45). The composition of the enemas included saline/glycerin (six cases), only saline solution (five), saline/glycerin/soap (four), plain water (three), and one case each of molasses/milk, saline/glycerin/soap/phosphate, saline/phosphate, and only phosphate. The enemas were performed in an antegrade fashion in 21 cases and rectally in 1. All patients had a dilated right colon and a narrow, spastic, left, transverse, and descending colon. Four patients underwent colonoscopy, colonic manometry, and mucosal biopsies, which did not help in explaining the etiology of the problem. In the literature, 43 reports mentioned a "long-term follow-up" for the administration of enemas, but we could not find a description of symptoms, such as in our cases.

Conclusions: An intriguing and, to our knowledge, previously unreported complication of chronic enema use is presented. We call attention to an overly concerning complication and report our findings in the hope that they will aid and stimulate more investigations into this condition. Several hypotheses to explain the cause are presented, as well as potential treatment options.

Daily enema use, administered either antegrade or retrograde, has become a common practice when treating fecal incontinence and soiling of different origins, as well as for severe colonic motility disorders.

Although decidedly effective, we have recently encountered patients whose long-term enema use seemed to be related to deleterious effects upon the colon. This phenomenon had a consistent clinical presentation, which began initially when the affected patients observed that the duration for the enema fluid to pass was progressively increasing and they complained of severe colicky abdominal pain during enema administration.

Contrast studies in these patients have shown a startling picture of severe dilatation of the right colon, followed distally by a very impressive, narrow, left portion of the transverse colon, as well as descending and rectosigmoid colon (Figs. 1–3).

After encountering enough patients with these findings, a pilot investigation was performed into the cause to exclude a random association for this intriguing complication.

Methods

The Colorectal Center at Children's Hospital Colorado maintains a database containing 196 patients receiving antegrade or retrograde enemas for the treatment of fecal incontinence and severe constipation and other colonic dysmotility disorders. Of these pa-

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Table 1
Patient's characteristics, type of enema and ingredients.

Patient's age	Original diagnosis	Years using enemas	Enema ingredients	Route of administration
Average 19.6 years Range 8–54 years	ARM 10 Myelomeningocele 5 Cloaca 2 Severe dysmotility 2 Hirschsprung's 2 Sacrococcygeal teratoma 1	Average 13.7 Range 4–45	Saline + glycerin 6 Saline 5 Saline + glycerin + soap 4 Water 3 Milk of molasses 1 Saline + glycerin + soap + Phosphate 1 Saline + phosphate 1 Phosphate 1	Antegrade 21 Rectal 1

Fig. 1. Contrast enema shows dilated right and narrow left colon.

tients, 22 have presented to our clinic complaining of increasing time to evacuate the enema fluid along with enema-induced colicky abdominal pain. For further evaluation, all patients underwent an antegrade or retrograde contrast study of the colon. Four patients underwent more extensive investigations consisting of colonic manometry and colonoscopy with biopsies for histologic study. Institutional review board approval was obtained for this study (18–1411).

The medical records of the 22 cases, were retrospectively reviewed, with emphasis on original diagnosis, age, number of years receiving enemas, and ingredients used in the enema fluid. The patient characteristics are summarized in Table 1. The average patient age was 19.6 years (range, 8–54). The primary original condition of the patients included anorectal malformation (10 cases), myelomeningocele (5), cloaca (2), severe colonic dysmotility/idiopathic constipation (2), Hirschsprung disease (2), and sacrococcygeal teratoma (1). The average duration of receiving enemas was 13.7 years (range, 4–45).

For us to know if this condition had been reported before, a literature review was performed. The search criteria included requesting publications from the last 20 years, related with: "enemas", "antegrade", "retrograde", "fecal incontinence", "intractable constipation", "colonic motility". We selected the papers specifically describing the antegrade or retrograde use of enemas for the treatment of fecal incontinence, severe intractable constipation, colonic motility disorders in children and in adults. Forty-three publications were selected for review. (1–43)

Results

The type of enemas have varied in these patients, as well as the ingredients that the patients were receiving, including saline solution/glycerin (six cases), only saline solution (five), saline solution/glycerin/soap (four), plain water (three), and one case each of molasses/milk, saline solution/glycerin/soap/phosphate, saline solution/phosphate, and only phosphate (Table 1).

The enemas were administered in an antegrade fashion through a cecostomy, continent appendicostomy, or continent neoappendicostomy in 21 cases and rectally in 1.

In all patients, the contrast study showed a conspicuously dilated right colon with massive contractions and contrast retention. Distally, there was an equally conspicuous, spastic, hypermobile, narrow, featureless, descending and rectosigmoid colon (see Figs. 1 and 2). Massive peristaltic contractions of the right colon coincided with severe episodes of colicky abdominal pain. Post-evacuation abdominal films were taken 2, 4, 6, and 24 h later, showed that the descending and rectosigmoid colon emptied rapidly, whereas the contrast remained in the dilated right colon for more than 24 h. (see Fig. 3)

Four patients accepted our suggestion of being evaluated thoroughly, and underwent a colonoscopy, suction mucosal biopsies, and colonic manometry (Table 2). One case had "changes suggestive of poor colonic motility in the left colon," one showed "diffuse disorganized peristalsis throughout," and two had "normal colonic manometry." Biopsies taken during colonoscopy showed "mild active colitis and cryptitis in the descending colon" in one case, "mild lamina propria edema and mild eosinophilia of the cecum" in one, and normal histology in the other two cases. We specifically asked our pathologists to look for the presence of fibrosis, but none was found. The colonoscopies were reported as "normal" in all four cases.

The literature review allowed us to divide our findings into four group, including:

Transanal administration of enemas in children

Six reports (total cases, 20 to 74) mentioned long-term follow-up, varying between 3 months and 8 years (8–13). The patients suffered from spina bifida and incontinence for other reasons, including anorectal malformations. A significant number of patients

Table 2
Findings on manometry, colonoscopy, and histopathology.

Patient #	Colonic Manometry	Colonoscopy	Biopsies
1	Left colon suggestive of poor motility	Normal	Descending colon with mild active colitis, mild cryptitis, no fibrosis
2	Normal	Normal	Normal, no fibrosis
3	Normal	Normal	Mild lamina propria edema, Mild eosinophilia of cecum, no fibrosis
4	Diffuse disorganized peristalsis	Normal	Normal, no fibrosis

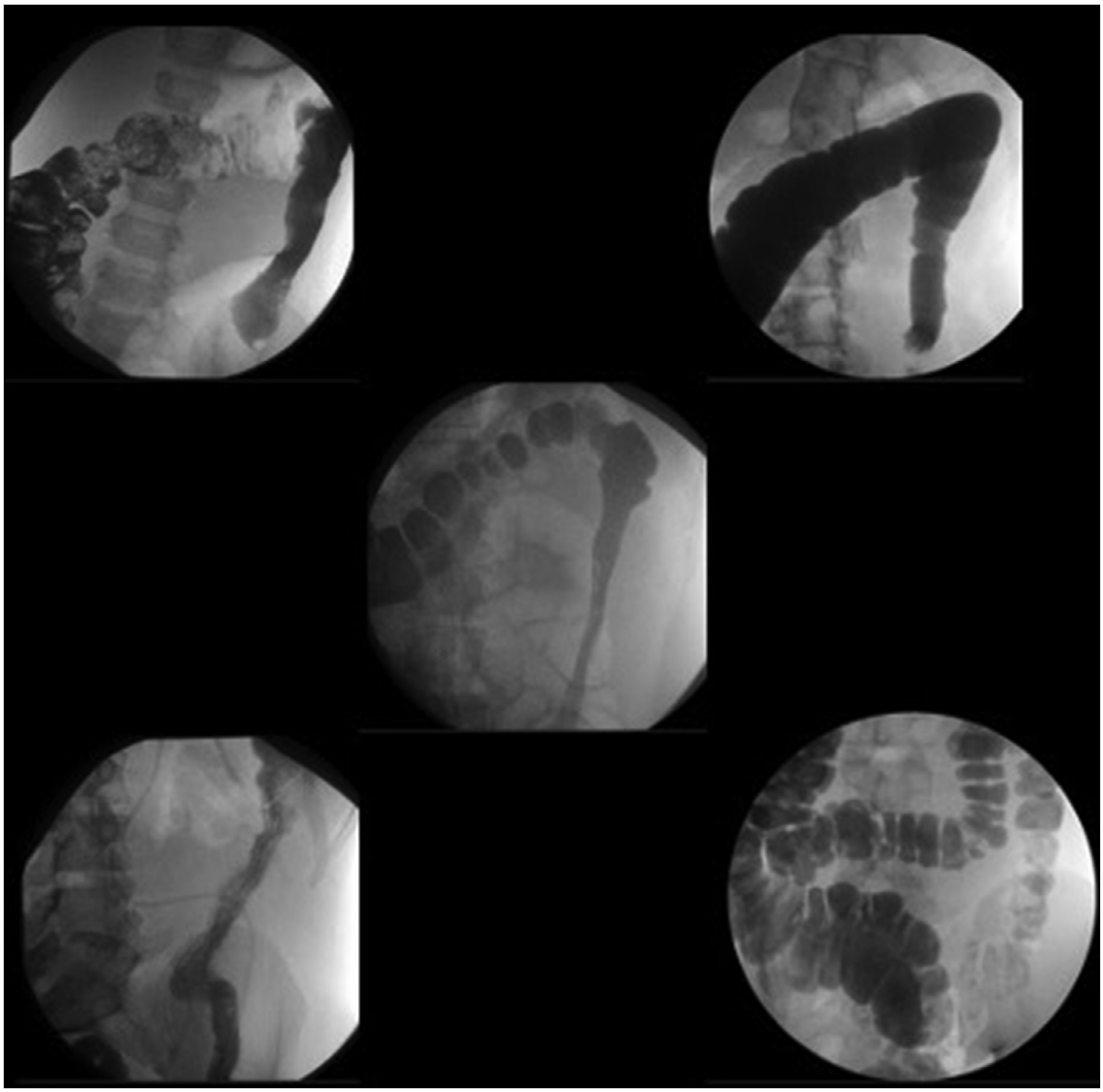

Fig. 2. Five representative contrast enemas showing dilated right and narrow, spastic left colon.

(9.5%–50%) discontinued enema administration because it was "time consuming" or "too difficult" and because it took ">60 min for the evacuation of the colon." It is reasonable to suppose that some of those patients were beginning to experience symptoms, such as those in our patients, but we cannot be certain since no mention was made of contrast studies being performed.

Antegrade administration of enemas in children

A total of 14 reports (total cases, 7 to 203) had information related to long-term follow-up, varying from 7 months to 11 years (14–28). Again, a significant number of patients decided to discontinue treatment (10%–28%) because of "slow evacuation," "pain during enema administration," "suboptimal results," "lack of effectiveness," and "failure of management." There was no mention of specific studies or investigations to determine the reasons for those complaints. Could those patients be experiencing the start of the problem that we are reporting? Perhaps, but we do not know.

Transanal administration of enemas in adults

Nine reports (total cases, 12 to 507) mentioned a "long-term follow-up," varying from 8 to 154 months (29–37). The percentage

Fig. 3. Contrast enema shows post evacuation contrast material retention in the right colon.

of patients who decided to discontinue enema administration varied from 20% to 67%. The reasons given to explain discontinuation included "abdominal pain during administration," "technical problems," and an "unsatisfactory outcome." We found no mention of a contrast enema performed to try to explain the symptoms. As expected in these reports, the percentage of patients who discontinued treatment was higher than that in the previous groups. We believe that the explanation could be because the follow-up time was longer than that in the other groups.

Antegrade administration of enemas in adults

Six reports (total cases, 27 to 426 [systematic review]) discussed "long-term follow-up," which varied from 6 to 75 months (38–43). A significant number of patients discontinued treatment (37% to 59%) because of "disappointment" with the results

Discussion

In 1987, Shandling and Gilmour (1) from Toronto, reported on the administration of enemas to treat fecal incontinence using a specially designed catheter. To our knowledge, that report started the era of bowel management, although it was not known by that name. In 1990, Malone et al. (2) described an ingenious operation to administer enemas in an antegrade fashion through the appendix. That significant contribution allowed for an improved quality of life in patients who required enemas to manage fecal incontinence and/or "intractable" constipation.

At our center, we have enthusiastically adopted the basic principles of that therapeutic modality and created a "bowel management program," which has attracted many patients and has been highly successful (3–7).

Our literature review has led us to suspect that many of the reported patients who decided to discontinue the enema administration were perhaps beginning to experience the symptoms and problems of our 22 cases. Unfortunately, none of those patients underwent a contrast study and therefore we have no evidence to support our suspicion.

Our study has two purposes. First, we are calling attention to an overly concerning and previously unreported complication associated with chronic enema use. Second, we are reporting our findings regarding this condition in the hope of aiding and stimulating more investigations into this unintended consequence.

The loss of haustral markings and the contracted appearance of the left colon seen in our patients would suggest a fibrosing pathologic process. This suggestion, however, has not been supported by the biopsies taken during the colonoscopy. Although we can only speculate on the pathophysiology of our observation, the lack of fibrosis seen in our colonic biopsies suggests that this phenomenon could be reversible. To try and confirm our speculation, we have attempted to apply the Koch's postulate of "type of approach confirms causality." We have suggested that our affected patients discontinue use of enemas for a time, after which we would like to repeat our previous studies.

We are certainly concerned regarding this, to our knowledge, unreported complication. In addition, we believe that we are looking at the "tip of the iceberg." Most of the 174 "asymptomatic" patients undergoing bowel management based on retrograde or antegrade enemas are significantly younger than the 22 symptomatic patients. We are also concerned that we may see more cases in the future.

It would be tempting to blame our observed left colon phenomenon on additives to the enema fluid, particularly phosphate. However as seen in Table 1, we cannot blame a specific ingredient as the cause of this condition. Only two of the studied patients were receiving phosphate additives to their enemas, with most using normal saline alone or glycerin and/or soap.

We are aware that our observational study has limited qualitative data and must be considered preliminary. The appearance of this syndrome has generated many unanswered questions. We are convinced that it requires further study and analysis. However, even with our limited data, we find it highly suggestive that the

"spastic left colon syndrome" occurs in patients receiving enemas over a long period of time (average 13.7 years), and that is the reason we are concerned about the great number of children receiving colonic flushes.

Unfortunately, we predict that this problem will be seen more often with time. One purpose of this study was to increase the index of suspicion in providers who prescribe enemas on a chronic basis. In addition, we hope that our findings will stimulate multi-institutional efforts of investigation to meet the challenges of this patient population.

References

[1] Shandling B, Gilmour RF. The enema continence catheter in spina bifida: successful bowel management. J Pediatr Surg 1987;22(3):271–3.
[2] Malone PS, Ransley PG, Kiely EM. Preliminary report: the antegrade continence enema. Lancet 1990:1217–18.
[3] Pena A. Bowel Management after surgery for imperforate anus. Dialog in Pediatr Urol 1996;19:12:1–8.
[4] Pena A, Guardino K, Tovilla JM, Levitt M, Rodriguez G, Torres R. Bowel management for fecal incontinence in patients with anorectal malformations. J Pediatr Surg 1998;33(1):133–7.
[5] Bischoff A, Levitt MA, Bauer C, Jackson L Holder M, Pena A. Treatment of fecal Incontinence with a Comprehensive Bowel Management Program. J Pediatr Surg 2009;44:1278–84.
[6] Bischoff A, Levitt MA, Pena A. Bowel management for the treatment of pediatric fecal incontinence. Pediatr Surg Int 2009;25(12):1027–42.
[7] Colares JHF, Purcaru M, da Silva GPF, Frota MA, da Silva CAB, Melo-Filho AAM, Bischoff A, Pena A. Impact of the bowel management program on the quality of life, in children with fecal incontinence. Pediatr Surg Int 2016;32:471–6.
[8] Liptak GS, Revell GM. Management of Bowel dysfunction in children with spinal cord disease or injury by means of the enema continence catheter. J Pediatr Feb 1992;120(2 pt 1):190–4.
[9] Sven Mattsson, Gunilla Gladh. Tap-water enema for children with myelomeningocele and neurogenic bowel dysfunction. Acta Pediatr. 2006;95(3):369–74 Mar.
[10] Jessica NG, Ford K, Dalton S, Mc Dowell S, Paul Charlesworth, Cleeve S. Pediatr Surg Int. 2015;31(8):729–34 Aug.
[11] Choi EK, Han SW, Shin SH, Ji Y, Chon JH, Im Y J. Long-term outcome of Trans-anal Irrigation for Children with Spina Bifida. Spinal Cord. 2015;53(3):216–20 Mar.
[12] King SK, Stathopoulos L, Pinnuck L, Wells J, Hutson J, Heloury Y. Retrograde continence enema in children with spina bifida: not as effective as first thought. J Pediatr Child Health. 2017;53(4):386–90 Apr.
[13] Ausili E, Marte A, Brisighelli G, Midrio P, Mosiello G, La Pergola E, Lombardi L, Iacobelli BD, Caponcelli E, Meroni M, Leva E, Rendell C. Short versus mid–long-term outcome of transanal irrigation in children with spina bifida and anorectal malformations. Child's Nervous System 2018;34:24712479.
[14] Curry JI, Osborne A, Malone PS. How to achieve a successful malone antegrade continence enema. J Pediatr Surg 1998;33(1):138–41 Jan.
[15] Shankar KR, Losty PD, Kenny SE, Booth JM, Turnock RR, Lamont GL, Rintala RJ, Lloyd DA. Functional results following the antegrade continence enema procedure. Br J Surg 1998;85(7):980–2 Jul.
[16] Marshall J, Hutson JM, Anticich N, Stanton MP. Antegrade continence enemas in the treatment of slow-transit constipation. J Pediatr Surg 2001;36(8):1227–30 Aug.
[17] Dey R, Ferguson C, Kenny SE, Shankar KR, Coldicutt P, Baillie CT, Lamont GL, Lloyd DA, Losty PD, Turnock RR. After the honeymoon—medium-term outcome of antegrade continence enema procedure. J Pediatr Surg 2003;38(1):65–8 Jan.
[18] Yardley LE, Pauniaho SL, Baillie CT Turnock RR, Coldicutt P, Lamont GL Kenny SE. After the honeymoon comes divorce: long-term use of the antegrade continence enema procedure. J Pediatr Surg 2009;44(6):1274–6 Jun.
[19] Aspirot A, Fernandez S, Cd Di Lorenzo, Skaggs B, Mousa H. Antegrade enemas for defecation disorders: do they improve the colonic motility? J Pediatr Surg 2009;44(8):1575–80 Aug.
[20] Siddiqui AA, Fishman SJ, Bauer SB, Nurko S. Long-term follow-up of patients after antegrade continence enema procedure. J Pediatr Gastroenterol Nutr 2011;52(5):574–80 May.
[21] Rodriguez L, Nurko S, Flores A. Factors associated with successful decrease and discontinuation of Antegrade Continence Enemas (ACE) in children with defecation disorders: a study evaluating the effect of ACE on colon motility. Neurogastroenterol Motil. 2013;25(2):140–e81 Feb.
[22] Bonilla SF, Flores A, Jackson CA, Chwals WJ, Orkin BA. Management of pediatric patients with refractory constipation who fail cecostomy. J Pediatr Surg 2013;48(9):1931–5 Sept.
[23] Masadeh MM, Krein M, Peterson J, Bauer M, Phearman L, Pitcher G, Liao J, Shilyansky J. Outcome of Antegrade Continent Enema (ACE) procedures in children and young adults. J Pediatr Surg 2013;48(10):2128–33 Oct.
[24] Peeraully MR, Lopes J, Wright A, Davies BW, Stewart RJ, Sing SS, More BB. Experience of the MACE procedure at a regional pediatric surgical unit: a 15-year retrospective review. Eur J Ped Surg. 2014;24(1):113–16 Feb.
[25] Randall J, Coyne P, Jaffray B. Follow-up of children undergoing antegrade continent enema: experience of over two hundred cases. J Ped Surg. 2014;9(49):1405–8 Sept.
[26] Basson S, Zani A, Mc Dowell S, Athanasakos E, Cleeve S, Phelps S, Charlesworth P. Antegrade Continence Enema (ACE): predictors of Outcome in 111 patients. Ped Surg Int. 2014;30(11):1135–41 Nov.
[27] Chong C, Featherstone N, Sharif S, Cherian A, Cuckow P, Mushtaq I, De Coppi P, Cross K, Curry J. 5 years after and ACE: what happens then? Pediatr Surg Int. 2016;32(4):397–401 Apr.
[28] Khoo AK, Auskoni E, Basson S, NG J, Cleeve S. How long will i have my ACE? the natural history of the antegrade continence enema stoma in idiopathic constipation. Pediatr Surg Int. 2017;33(11):1159–66 Nov.
[29] Gosselink MP, Darby M, Zimmerman DDE, Smits AAA, Van Kessel I, Hop WC, Briel JW, Schouten WR. Long term follow-up of retrograde colonic irrigation for defecation disturbances. Colorectal Dis. 2004;7(1):65–9 Jan.
[30] Faaborg PM, Christensen P, Kvitsau B, Buntzen S, Laurberg S, Krogh K. Long-term outcome and safety of transanal colonic irrigation for neurogenic bowel dysfunction. Spinal Cord. 2009;47(7):545–9 Jul.
[31] Christensen P, Krogh K, Buntzen S, Payandeh F, Laurberg S. Long-term Outcome and safety of transanal irrigation for constipation and fecal incontinence. Dis Colon Rectum. 2009;52(2):286–92 Feb.
[32] Faaborg PM, Christensen P, Buntzen S, Laurberg S, Krogh K. Anorectal function after long-term transanal colonic irrigation. Colorectal Dis. 2010;12(10):e314–19 Oct.
[33] Hamonet-Torny J, Bordes J, Daviet JC, Dalmay F, Joslin F, Salle JY. Long-term transanal irrigation's continuation at home. preliminary study. Ann Phys Rehabil Med. 2013;56(2):134–42 Mar.
[34] Emmett CD, Close HJ, Yianakou Y, Mason JM. Transanal irrigation therapy to treat adult chronic functional constipation: systematic review and meta-analysis (2015). BMC Gastroenterol 2015;15:139 Oct 16.
[35] Vollebregt PF, Elfrink AKE, Meijerink WJHJ, Felt-Bersma RJF. Results of long-term retrograde rectal cleansing in patients with constipation or fecal incontinence. Tech Coloproctol 2016;20(9):633–9 Sep.
[36] Bildstein C, Melchior C, Gourcerol G, Boueyre E, Bridoux V, Verin E, Leroi AM. Predictive factors for compliance with transanal irrigation for the treatment of defecation disorders. World J Gastroenterol 2017;23(11):2029–36 Mar21.
[37] Juul T, Christensen P. Prospective evaluation of transanal irrigation for fecal incontinence and constipation. Tech Coloproctol 2017;21(5):363–71 May.
[38] Lees NP, Hodson P, Hill J, Pearson RC, MacLennan I. Long-term results of the antegrade continent enema procedure for constipation in adults. Colorectal Dis 2004;6(5):362–8 Sept.
[39] Worsoe J, Christensen P, Krogh K, Buntzen S, Laurberg S. Long-term of antegrade colonic enema in adult patients: assessment of functional results. Dis Colon Rectum 2008;51(10):1523–8.
[40] Koivusalo AL, Pakarinen MP, Pauniaho S-L, Rintala RJ. Antegrade continence enema in the treatment of congenital fecal incontinence beyond childhood. Dis Colon Rectum 2008;51(11):1605–10 Nov.
[41] Patel AS, Saratzis A, Arasaradnam R, Harmston C. Use of antegrade continence enema for the treatment of fecal incontinence and functional constipation in adults: a systematic review. Dis Colon Rectum. 2015;58(10):999–1013.
[42] Chan DSY, Delicata RJ. Meta-analysis of antegrade continence enema in adults with fecal incontinence and constipation. Br J Surg 2016;103(4):322–7 Mar.
[43] Sturkenboom R, van der Wilt AA, van Kujik SMJ, Ahmad A, Janssen PT, Stassen PS, Melenhorst J, Breukink SO. Long-term outcomes of a Malone Antegrade Continence Enema (MACE) for the treatment of fecal incontinence or constipation in adults. Int J Colorectal Dis. 2018;33(10):1341 Oct.