

Treatment of constipation in adults associated with idiopathic megarectum by behavioural retraining including biofeedback

T. Mimura, T. Nicholls, J. B. Storrie and M. A. Kamm

St Mark's Hospital, London, UK

Received 29 October 2001; accepted 28 January 2002

Abstract

Background Constipation in adults associated with a grossly dilated rectum and recurrent faecal impaction, idiopathic megarectum, is rare. The aetiology of idiopathic megarectum is unknown, but may involve neuromuscular or behavioural factors. It is unknown whether the condition is reversible. This study aimed to determine the efficacy of behavioural therapy, including biofeedback, in such patients.

Methods Six patients (4 female; median age 27) with a history of rectal faecal impaction and a grossly dilated rectum on radiological examination were evaluated by structured questionnaire before, immediately after biofeedback therapy, and on follow-up. Physiological testing was performed before treatment, and 2 patients were evaluated by repeat physiological testing and contrast radiology on follow-up.

Results On median follow-up of 18 months (range 11–27), five patients felt major and one patient minor improvement in symptoms, including two with complete symptom relief. Four patients came off laxatives without recurrent faecal impaction. In the 2 studied patients rectal size did not appear to decrease.

Conclusion Behavioural retraining, including biofeedback, improved symptoms in most patients with idiopathic megarectum. In some patients symptoms completely resolved, without the need for laxatives. Although further studies are necessary in terms of both larger number of patients and longer follow-up period, behavioural treatment may be useful for such patients.

Keywords Megarectum, constipation, faecal impaction, biofeedback, behavioural retraining

Introduction

Idiopathic megarectum is characterized by a chronically dilated rectum, associated with severe constipation [1]. Recurrent faecal impaction is common and often accompanied by soiling, especially in those with an onset in childhood or adolescence [1]. Hirschsprung's disease is excluded by the presence of the recto-anal inhibitory reflex and the presence of intermuscular ganglia in the rectum.

The aetiology of this condition remains unknown, although some neuromuscular abnormalities have been reported [2–5]. Whether these are primary or secondary to long-standing distension is unknown. In some, the condition may be secondary to an abnormal defaecation mechanism, particularly pelvic floor inco-ordination, associated with a failure of toilet training during childhood

[6], leading to withholding of stool, progressive rectal distension and, ultimately, loss of rectal contractility.

The majority of patients with this disorder can be successfully managed by disimpaction followed by the use of osmotic laxatives such as magnesium sulphate [1,7]. If medical treatment fails, surgical therapy, such as the Duhamel procedure or total colectomy with ileorectal anastomosis, may be required, although the results are variable [7,8].

The condition of idiopathic megarectum differs significantly from that of idiopathic constipation, the latter involving a colon and rectum of normal diameter. Idiopathic megarectum affects males and females in late adolescence, and responds often to laxatives after disimpaction [1]. In contrast, idiopathic constipation affects predominantly women, their median age is 40, and they are often resistant to treatment with laxatives [9].

Idiopathic constipation was considered an irreversible functional disorder, requiring life-long usage of evacuants, although often with limited success. However, the

Correspondence to: Prof M. A. Kamm, St Mark's Hospital, Northwick Park, Watford Road, Harrow, Middlesex HA1 3UJ, UK.
E-mail: kamm@ic.ac.uk

advent of behavioural therapy, known as biofeedback, enabled many patients to normalize bowel function and improve symptoms without drugs. Recently the long-term efficacy of biofeedback in 100 patients with idiopathic constipation was reported. In this study, about two thirds of the patients experienced symptom improvement [10]. The use of behavioural treatment (biofeedback) has also been shown to benefit patients with constipation following hysterectomy [11], some patients with a rectocele [12,13] and patients with solitary rectal ulcer syndrome [14].

It is unknown whether constipated patients with abnormal rectal anatomy could enjoy similar benefit from biofeedback. We have hypothesized that in some patients with idiopathic megarectum there may be a reversible behavioural disorder. This study therefore aimed to evaluate the efficacy of behavioural treatment (biofeedback) in patients with idiopathic megarectum.

Patients and methods

Patients

The notes of patients classified as having idiopathic megarectum who completed biofeedback in the Physiology Unit at St. Mark's Hospital between 1996 and 1998 were reviewed. Idiopathic megarectum was defined as both having the history of faecal impaction and a grossly enlarged rectum demonstrated on radiological examinations, including a water soluble contrast (gastrografin) enema, defaecography, barium enema or plain abdominal X-ray. Hirschsprung's disease was excluded by the presence of a dilated rectum down to the pelvic floor (anal sphincter) and the presence of a rectoanal inhibitory reflex.

Eight hundred and fifty-three patients with constipation defined as infrequent bowel motions and/or impaired defaecation completed a course of biofeedback between 1996 and 1998 at our unit. Out of these, 6 patients (0.7%) fulfilled the above criteria of idiopathic megarectum and formed the basis of this study. A detailed history, anorectal physiological investigations and radiological examinations were reviewed in these 6 patients.

Data about patients and bowel function were prospectively collected before and after treatment, and on follow-up, using the same structured questionnaire.

Behavioural therapy (Biofeedback)

During behavioural therapy, each patient saw a biofeedback specialist nurse as an outpatient every two to three weeks, usually for four or five sessions.

Visual feedback of pelvic floor muscular activity was given in two patients to enable them to co-ordinate their defaecatory muscles, using an air filled-balloon and two adherent surface electrodes. In the other four patients, surface electrodes were not used, because the problem in megarectum is usually not pelvic floor inco-ordination but the loss of urge sensation. It has also been reported that behavioural therapy with or without visual feedback provides a similar outcome in patients with constipation [15]. Some patients with idiopathic megarectum did not have the sensation of full rectum with 50 ml of air, while those with idiopathic constipation normally do. Additional air was inflated only up to 100 ml in total.

Patients were also advised on normal defaecatory behaviour and bowel habits, which included (i) encouragement to visit the toilet regularly once or twice a day regardless of the feeling of urge, (ii) how to relax before attempting defaecation, (iii) appropriate posture to sit on the toilet, leaning forward with their feet slightly raised, (iv) how to brace or widen their waist for effective propulsive force and (v) how to relax rather than squeeze their defaecatory muscles.

An attempt to get the patients off laxatives, enemas and suppositories was made with more caution than in those with idiopathic constipation, in order to avoid recurrent faecal impaction.

At each biofeedback session, the therapist tried to achieve a good rapport with the patient to facilitate good understanding and collaboration. This included gaining an appreciation about the patient's personal life and psychological factors which might be relevant. When the course of biofeedback was completed patients were encouraged to continue practising the techniques they had learnt.

Follow-up

Follow-up consisted of a telephone interview by an independent person who had not been involved in the therapy. Data were obtained using a structured questionnaire containing the same questions as were recorded before and immediately after the completion of biofeedback. Bowel symptoms before biofeedback, immediately after biofeedback, and at the time of interview were assessed, including rectal faecal impaction, bowel frequency, straining at stool, digital evacuation, sense of incomplete evacuation, abdominal pain and bloating and the use of bowel evacuants such as oral laxatives, suppositories and enemas. To evaluate the subjective benefits of the therapy, patients were asked whether they felt their constipation had improved. The subjective judgement was rated as follows: not at all, to some degree, very much and cured.

At the time of the follow-up, all six patients were asked to undergo both of a gastrografin enema and a rectal sensory test to balloon distension in order to have the size of their rectum re-evaluated after biofeedback. Two of them agreed to have this re-evaluation.

Results

Patient characteristics

As is shown in Table 1, the 6 patients (4 female) had the first session of biofeedback at a median age of 27 years (range 14–48). All of them had been constipated since their early childhood. All patients had a history of faecal impaction, which had been disimpacted using enemas or under general anaesthesia. Four patients had a history of soiling.

Two patients (Patients 1 and 6) had not been diagnosed as having a megarectum until they were referred to our unit, and had been taking laxatives without adequate benefit. The other four patients were known to have a megarectum prior to referral, and were referred to our unit because their constipation could not be managed even with laxatives. One patient (Patient 5) had had an antegrade continence enema (ACE) stoma, and was dependent on this for control of constipation prior to biofeedback. None of the six patients ever had a gut-focused behavioural therapy previously, including biofeedback.

On radiological examinations, an enlarged rectum filled with formed stool was observed in all patients, of whom a gastrografin enema was performed in 3 patients; the other 3 patients had an enlarged rectum demonstrated on an abdominal X-ray. The diameter of the proximal colon appeared normal excluding megacolon, and the 6 patients were thought to have idiopathic megarectum. From their history, chronic intestinal pseudo-obstruction was not suspected, and therefore investigations to examine the upper gastrointestinal tract such as barium follow through study were not arranged in any patients except for one (Patient 3), who had a normal upper gut. These patients have been previously shown to be clinically distinguishable from patients with chronic intestinal pseudo-obstruction, and to have a normal upper gut [1]. A transit study was carried out in all the patients but one (Patient 6), and showed slow colonic transit with retention of most markers in the dilated rectum.

In the anorectal physiological studies, four patients had an abnormally increased maximum tolerated volume (MTV) and two had a normal MTV, presumably due to the impacted stool. All patients but one (Patient 3) demonstrated paradoxical pelvic floor inco-ordination on straining and all patients but two (Patients 1 and 6) were

Table 1 Characteristics of 6 patients with idiopathic megarectum and results of behavioural re-training therapy.

Patient No	Age	Sex	F/u (Months)	No of BRT sessions	Maximum tolerated volume (Normal range: 100–320 ml)	Constipation improvement		Faecal impaction		Evacuants usage		Bowel frequency* (per week)	
						After BFB	On follow-up	after BFB	after BFB	Before BFB	After BFB	Before BFB	After BFB
1	48	F	27	5	320	Very much	Cured	No	No	Oral	None	2.3	3
2	36	F	12	4	>350	Not at all	Some degree	Yes	Yes	Oral & enema	Oral	2.8	2
3	35	F	11	4	125	Some degree	Very much	Yes	Yes	Oral	Oral	1	5.5
4	19	M	11	4	>450	Some degree	Very much	No	No	Oral	Oral	1.2	5.5
5	14	F	24	5	400	Very much	Cured	No	No	Enema	Enema	7	6
6	15	M	25	3	70	Some degree	Very much	No	No	Oral	Oral	7	7

F-u: follow-up period since completing biofeedback treatment; Mo: months, BRT: Behavioural re-training therapy, Oral: oral laxatives; Bowel frequency*: with or without laxative use.

unable to evacuate a 50-ml water-filled balloon. In all patients, the rectoanal inhibitory reflex was confirmed to be present.

No patient had had previous surgery, except for one (Patient 5) who had undergone an anorectal myectomy and formation of an ACE stoma for her constipation. Three patients (Patient 2, 3 and 5) had had previous manual evacuation under general anaesthesia.

No other medical disorders were noted, including endocrinological diseases. Although no formal IQ tests or psychological profiles were performed, none of these patients were thought to be intellectually or psychologically impaired, judging from their history and the clinical impression of the treating specialist, biofeedback specialist nurses and the telephone interviewer.

Results of treatment on follow-up

The median follow-up period for these 6 patients after the completion of biofeedback was 18 months (range 11–27 months).

Bowel symptoms

As is shown in Table 1, two patients achieved complete resolution of symptoms on follow-up, and were not taking laxatives. Four were still suffering from some symptoms, although 2 of these were able to stop taking laxatives. Patient 5 was able to cease using her ACE stoma, which had been used prior to biofeedback, the device was removed, and the fistula closed spontaneously.

Although all patients had been on evacuants before biofeedback and 5 were still using them immediately after biofeedback, only 2 patients were taking oral laxatives on follow-up. Four patients who had come off evacuants on follow-up have not experienced faecal impaction after biofeedback, with 3 patients passing formed stool and variable stool consistency in one. The other 2 patients who were still on evacuants suffered from faecal impaction once after biofeedback, which required medical disimpaction. Bowel frequency was normal (between 3 times a week and 3 times a day) in all patients on follow-up.

Although most patients were able to cease laxatives and avoid recurrent impaction following biofeedback, some still had abdominal discomfort, the need to strain, and a feeling of incomplete evacuation.

Subjective improvement

As is shown in Table 1, immediately after biofeedback most patients felt that their constipation had improved, as had all patients on follow-up.

Examination, gastrografin enema and balloon distension test

Two patients (Patient 1 and 3) underwent re-evaluation of rectal size on follow-up. The rectal diameter at the pelvic brim on the gastrografin enema of Patient 1 was 7 cm both before and after biofeedback. That of Patient 3 was also unchanged, measuring 10 cm before and after biofeedback. On the balloon distension test, the maximum tolerated volume of Patient 1 remarkably decreased from 320 ml of air prior biofeedback to 190 ml after. That of Patient 3 was 125 ml before and 200 ml after biofeedback.

Discussion

Idiopathic megarectum is a rare condition, constituting a small proportion of all patients with constipation. It is characterized by a grossly enlarged rectum, episodes of recurrent faecal impaction, and passive faecal incontinence. It usually has a childhood or adolescent onset, and therefore can have significant psychological and social consequences.

Initial treatment involves disimpacting the rectum, usually followed by the use of osmotic laxatives, such as magnesium sulphate, to keep the stool semiliquid and to prevent recurrent impaction [1]. It is preferable to achieve disimpaction by the use of repeated enemas, because manual disimpaction under general anaesthesia often results in anal sphincter disruption [17]. About 75% patients are successfully managed with long-term laxatives [1]. In this study, this initial management had been carried out before the patients were referred for behavioural therapy.

In about 25% patients who cannot tolerate or do not respond to laxatives, surgical treatment can be undertaken. For megarectum and megacolon, various procedures have been performed with variable outcomes, depending on the extent and region of the dilated bowel. In those with a moderately dilated colon, a colectomy with ileorectal anastomosis seems to offer the best results, with approximately 80% achieving a successful outcome [18]. In those with an extremely dilated rectum, the most common operation has been the Duhamel procedure. In a series of 20 patients having a Duhamel operation for a grossly dilated rectum, half achieved a normal bowel frequency. However, persistence of many symptoms was common and further surgery was often required [8].

This study has demonstrated that behavioural treatment, including biofeedback, is a useful conservative treatment for some patients with idiopathic megarectum. To our knowledge this is the first study to address

the value of behavioural treatment for patients with idiopathic megarectum. Two patients had complete resolution of symptoms, and the other four felt symptomatically improved, although in two patients improvement may have related to the continuous intake of laxatives. The most convincing aspect of the response to behavioural therapy was the ability of four patients to come off laxatives, without further rectal faecal impaction, after biofeedback. These patients had experienced recurrent faecal impaction or dependence on laxatives for most of their lives. Although the number of patients is small and the follow-up period is short, this treatment considerably improved symptoms in most patients. It would seem reasonable to offer this relatively inexpensive and side-effect free treatment as the first line of therapy.

Recently we reported the efficacy of biofeedback in 100 patients with idiopathic constipation. In that study, 57 felt their constipation had improved symptomatically, and these results were maintained for up to 44 months (median 23 months) after treatment. Objective evidence for benefit included decreased laxative, suppository and enema use. Patients with slow and normal transit, males and females, and those with and without pelvic floor inco-ordination benefited equally from this therapy [10]. We have also demonstrated in 25 patients with a rectocele and impaired defaecation that this therapy leads to major symptoms relief in 16% of patients and minor symptom relief in 56% [12]. This study extends the indication for this type of therapy.

Radiologically, idiopathic megarectum is defined as a rectum larger than 6.5 cm in diameter in a lateral view of the pelvic brim [19]. The precise rectal diameter was not measured in all patients of this study. However, from the combination of their typical clinical manifestations (history and clinical findings of rectal faecal impaction and soiling, often with a rectum palpable in the abdomen) and typical radiological features, we are confident that all of these 6 patients had an idiopathic megarectum.

The size of the large rectum in the available two patients did not change on gastrografin enema before and after biofeedback. Both of them improved in symptoms with this therapy, although only one had ceased taking laxatives. This suggests that symptoms of patients with idiopathic megarectum are reversible although the enlarged structure of the rectum may not be. Longer follow-up of more patients would help determine if the rectal diameter does change.

Regarding the aetiology of idiopathic megarectum, some neuromuscular abnormalities have been reported [2–4]. Patients with idiopathic megarectum have significant thickening of their muscularis mucosa, circular

muscle and longitudinal muscle despite rectal dilatation [2]. There is also an increase in VIP and nitric oxide containing fibers in the muscularis mucosae and lamina propria and a decrease in the longitudinal muscle in rectal tissue of patients with idiopathic megarectum [3]. It is unknown whether the rectal abnormalities, including these histological findings, are a primary or secondary phenomenon. In some they may be related to an abnormal defaecation mechanism, particularly pelvic floor inco-ordination, which results from a failure of toilet training during childhood [6].

Some patients with idiopathic megarectum, especially of a child onset are mentally retarded or have an intellectual deficit [1,20]. It remains unknown whether these associations are due to shared abnormality of brain and gut neurological development, to long-term use of psychotropic drugs or to the effects of suppressed defaecation. The patients in this study appeared intellectually normal.

It is likely that patients with idiopathic megarectum have various different underlying aetiologies including neurophysiological, behavioural and psychological. Behavioural re-training therefore may be able to rectify behavioural, and possibly psychological, components. Idiopathic megarectum has been thought to be an irreversible disorder requiring life-long maintenance medical treatment [21]. Biofeedback might be able to change this view by normalizing the rectal function of the patients with idiopathic megarectum in a similar way to the normalization of colonic transit in patients with idiopathic constipation [10].

Biofeedback, in which the patient receives visual feedback of pelvic floor muscular activity, is only one component of this treatment. Other components include habit training, encouraging a patient to visit the toilet once or twice daily even when they do not feel the urge, supervision to enable the patient to come off laxatives, the co-ordination of other muscles groups, and possibly counseling and informal psychotherapy [10,16].

In principle, biofeedback specialist nurses persuade patients with idiopathic constipation to come off laxatives in order to make them responsible for their own bowel actions and restore their confidence over their bowel control. However, in patients with idiopathic megarectum, the process of stopping laxatives was slower, in order to avoid causing recurrent faecal impaction.

In conclusion, behavioural retraining improved symptoms in most patients with idiopathic megarectum. In some patients symptoms completely resolved without the further need for laxatives. Further studies with more patients and longer follow-up are now required.

References

- 1 Gattuso JM, Kamm MA. Clinical features of idiopathic megarectum and idiopathic megacolon. *Gut* 1997; **41**: 93–9.
- 2 Gattuso JM, Kamm MA, Talbot IC. Pathology of idiopathic megarectum and megacolon. *Gut* 1997; **41**: 252–7.
- 3 Gattuso JM, Hoyle CH, Milner P *et al.* Enteric innervation in idiopathic megarectum and megacolon. *Int J Colorectal Dis* 1996; **11**: 264–71.
- 4 Gattuso JM, Smith VV, Kamm MA. Altered contractile proteins and neural innervation in idiopathic megarectum and megacolon. *Histopathology* 1998; **33**: 34–8.
- 5 Gattuso JM, Debinski HS, Kangro HO *et al.* Evaluation of specific herpes DNA viruses in idiopathic megarectum and idiopathic megacolon. *Int J Colorectal Dis* 1998; **13**: 131–3.
- 6 Nixon HH. Megarectum in the older child. *Proc R Soc Med* 1967; **60**: 801–3.
- 7 Kamm MA, Stabile G. Management of idiopathic megarectum and megacolon. *Br J Surg* 1991; **78**: 899–900.
- 8 Stabile G, Kamm MA, Hawley PR *et al.* Results of the Duhamel operation in the treatment of idiopathic megarectum and megacolon. *Br J Surg* 1991; **78**: 661–3.
- 9 Preston DM, Lennard-Jones JE. Severe chronic constipation of young women: 'idiopathic slow transit constipation'. *Gut* 1986; **27**: 41–8.
- 10 Chiotakakou-Faliakou E, Kamm MA, Roy AJ *et al.* Biofeedback provides long-term benefit for patients with intractable, slow and normal transit constipation. *Gut* 1998; **42**: 517–21.
- 11 Roy AJ, Emmanuel AV, Storrice JB *et al.* Behavioural treatment (biofeedback) for constipation following hysterectomy. *Br J Surg* 2000; **87**: 100–5.
- 12 Mimura T, Roy AJ, Storrice JB *et al.* Treatment of impaired defecation associated with rectocele by behavioral retraining (biofeedback). *Dis Colon Rectum* 2000; **43**: 1267–72.
- 13 Lau CW, Heymen S, Alabaz O *et al.* Prognostic significance of rectocele, intussusception, and abnormal perineal descent in biofeedback treatment for constipated patients with paradoxical puborectalis contraction. *Dis Colon Rectum* 2000; **43**: 478–82.
- 14 Vaizey CJ, Roy AJ, Kamm MA. Prospective evaluation of the treatment of solitary rectal ulcer syndrome with biofeedback. *Gut* 1997; **41**: 817–20.
- 15 Koutsomanis D, Lennard-Jones JE, Roy AJ *et al.* Controlled randomised trial of visual biofeedback versus muscle training without a visual display for intractable constipation. *Gut* 1995; **37**: 95–9.
- 16 Storrice JB. Biofeedback: a first-line treatment for idiopathic constipation. *Br J Nursing* 1997; **6**: 152–8.
- 17 Gattuso JM, Kamm MA, Halligan SM *et al.* The anal sphincter in idiopathic megarectum. effects of manual disimpaction under general anesthetic. *Dis Colon Rectum* 1996; **39**: 435–9.
- 18 Stabile G, Kamm MA, Hawley PR *et al.* Colectomy for idiopathic megarectum and megacolon. *Gut* 1991; **32**: 1538–40.
- 19 Preston DM, Lennard-Jones JE, Thomas BM. Toward a radiological definition of idiopathic megacolon. *Gastrointest Radiol* 1985; **10**: 167–9.
- 20 Ehrenthel O. Megacolon in psychotic patients. *Gastroenterology* 1955; **29**: 285–94.
- 21 Barnes PRH, Lennard-Jones JE, Hawley PR *et al.* Hirschsprung's disease and idiopathic megarectum in adults and adolescents. *Gut* 1986; **27**: 534–41.

