

Multidisciplinary behavioural treatment of fecal incontinence and constipation after correction of anorectal malformation

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Background: Fecal incontinence and constipation are major problems after correction of anorectal malformation (ARM), caused not only by the somatic defects but also by a psychosomatic dysfunction of defecation. To better release patients from this dysfunction we offered a multidisciplinary, psycho- and physiotherapeutic therapy according to an approach developed in Nijmegen (Netherlands). We herein summarize the preliminary results to evaluate whether the approach can be adopted with similar success.

Methods: Since January 2002 multidisciplinary behavioural treatment (MBT) has been offered to children above 3 years of age and suffering from fecal incontinence and constipation after surgical correction of ARM in our department or elsewhere. Prerequisites included no anal stenosis, regulation of stool consistency, and a suitable defecation diary over 2 weeks. MBT contained regular consultations by a pediatric psychologist and a physiotherapist, teaching the child to establish a regular defecation pattern and how to push while relaxing the pelvic floor. The entry- and post-treatment situation was prospectively monitored by means of defecation and constipation scoring systems.

Results: Complete data were available in 10 patients (9 males, 1 female) with high (8 patients) and low (2) forms of anal atresia initially, who finished MBT 2-36 months ago (mean: 13 months). The average amount of stool reaching the toilet was 27% before and 90% after therapy. Clean days were absent before, reaching 3.7 days on average

after therapy. Constipation was present in 6 patients before (3 of them on enemas) and in 2 after therapy (no enemas needed). The duration of MBT was 7 months on average, range 3-23 months, with 8-9 sessions per patient, each lasting 60-90 minutes. An observation period of 7 months after treatment confirmed stable results. MBT turned out to improve body-consciousness and self-confidence.

Conclusions: MBT is effective in reducing incontinence and constipation in patients after ARM. It helps the children and their families to relieve psychosocial stress. The approach can be successfully adopted, if a team of committed specialists is available and sufficient compliance of patients and families is given.

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Introduction

Fecal incontinence and chronic constipation are frequent problems after surgical correction of anorectal malformation (ARM) in high and low types.^[1-3] This is caused not only by inborn somatic defects but frequently worsened by psychosomatic dysfunction of defecation behaviour, withholding some children from learning a normal defecation behaviour at all. Furthermore, patients may develop an aversion to the subject and to their pelvic floor, which may be aggravated by perioperative stress, painful anal manipulation such as dilatation or enema, and, finally, by shame of their incontinence. Some of these children unconsciously try to avoid defecation. An experienced team in Nijmegen recognized this problem and developed a psycho- and physiotherapeutic approach termed "multidisciplinary behavioural treatment of defecation problems" (MBT).^[4,5] We adopted this

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approach in 2002 and got evidence of its striking potential to improve the difficult situation of the patients and their social surrounding. Convinced that this interdisciplinary treatment modality is effective in any comparable setting, we herein report our initial results.

Methods

MBT has been offered to children suffering from fecal incontinence after surgical correction of anorectal malformations and having reached the developmental age of 3.5 to 4 years. When the family decides to start therapy, the pediatric surgeon has to rule out or treat a severe state of anorectal stenosis and/or constipation at first. The consistency of the stool may have to be medically corrected, making it neither too hard nor too soft. Since severe constipation worsens the incontinence (overflow incontinence^[6]), other factors such as unconscious avoidance of defecation through constipating diet or contractions of the pelvic floor while pushing for defecation have to be considered. In our patients, however, no state of serious impaction of stools or megarectum were noticed before the start of therapy.

A defecation diary over a period of two weeks will show if there is already a body rhythm to start with. Subsequently, the pediatric psychologist works in regular meetings with the child trying to relieve his frustration, fear, and reluctance to defecation. He or she also educates the parents in their role as co-therapists supporting their child to continue training steps at home. Similarly, the pediatric physiotherapist trains the child to perceive its body in a new positive way. A cornerstone in establishing productive defecation behaviour is to teach the child to push in a downward direction while simultaneously relaxing the pelvic floor. The motto is: *letting go instead of holding on*. Depending on the distance from home to hospital, the patient is treated on an in- or outpatient basis, respectively.

Each treatment session takes about one hour. A joint session of all therapists and the family will be held initially, while the parents then participate from time to time only, and if there are new home exercises to be learned. If required, they or the child may receive an individual talk with the psychologist. In the phase of MBT, we emphatically recommend to strictly avoid new anorectal trauma either through investigation or enema.

Since 2002, the MBT-team has overlooked 29 consecutive patients suffering from stool incontinence (20 of them constipated) after surgical correction of anal atresia. Ten of the 29 patients have finished MBT so far and could be evaluated. In 13 of the remaining

patients, treatment is not yet completed, and in another 6 therapy had to be stopped prematurely due to insufficient cooperation of the children (2 patients: Downs syndrome, psychiatric disease) and lacking parental compliance (4 patients).

In the evaluated group of 10 patients, 9 boys had high (8) and low (1) types of ARM and one girl had low type of ARM initially. The average age at the start of therapy was 11 years (range 6-14 years); six of the 10 patients were constipated. The median average duration of treatment was 7 months (3-23 months), with 8 to 9 sessions per therapy in intervals of several weeks. The mean follow-up after therapy was 13 months (2-36 months). The four outcome variables given by the team of Nijmegen were used to correspondingly compare the results. These variables included the fecal incontinence score according to Templeton (Table 1),^[7] the "percentage of feces in the toilet" (as estimated by the parents, practically all of them have a surprisingly exact notion in this point), the "number of days without soiling",^[5] and the degree of constipation as classified by Pena^[2] (Table 2). The data were collected through parental interviews before therapy, at its end, and at the end of the observation period.

SPSS 13.0 software package was used for data analysis. Analysis of variance was performed by the Kruskal-Wallis test and Wilcoxon's rank-sum test. Covariance analysis was used to test the impact of the duration of therapy and observation time on the therapy efficiency. A *P* value less than 0.05 was considered statistically significant. All results were considered

Table 1. Templeton-score of fecal incontinence^[7]

Quantitative assessment of fecal incontinence according to Templeton and Ditesheim		
Toilet training for stool	Successful	1
	Occasionally successful (awareness of impending stool)	0.5
	No awareness of impending stool	0
Accidents	None, or rare	1
	Three per week or less	0.5
	More than 3 per week	0
Extract underpants (or liners) needed	Never	1
	Only when having diarrhea	0.5
	Always	0
Social problems	None	1
	Infrequent odor; does not miss school (but no overnights, dates, camping, etc)	0.5
	Frequent odor affects school and play	0
Activity restrictions	None	0.5
	Avoids swimming, sports, etc	0
	No current problems	0.5
Rashes	No current problems	0.5
	Some current problems	0

Notes: Good (4-5 points), fair (2-3.5 points), poor (0-1.5 points).

as explorative because of no adjustment for multiple testing.

Results

Detailed data of the 10 patients who finished the treatment are summarized in Table 2. The figures concerning the 13 patients still on therapy are given in brackets in the following. On average, the percentage of feces in the toilet was 27% at the beginning, 85% at the end of MBT (25%/90%), and even 90% at follow-up (Fig. 1). Similarly, the number of days without soiling raised from 0 to 3.7 (2 to 3) and dropped to 3.0 at the end of observation. The fecal incontinence score ranged from 1.5 ("poor") to 3.5 ("fair") on average (2 to 3, both "fair") and did not change during the observation period (Fig. 2). Constipation was present in 6 cases before therapy, requiring diet in 2, laxatives in 1 and enema in 3. After therapy, 4 of them were free

of constipation, laxatives or diet were still required in one patient, respectively. The mean constipation-score of the 13 patients on therapy was 2 before therapy (need of laxatives) and 0 (no constipation) at the time of data collection. With regard to statistical covariance analysis, the data were of significance for the percentage of stool in the toilet before and after treatment ($P<0.05$), the effect of observation time on maximum of clean days ($P<0.05$) and the improvement of constipation before and after treatment ($P<0.05$). There was no significant difference in the effect of duration of therapy and observation time on the percentage of stool in the toilet and the degree of constipation.

Some examples may give a deeper insight into personal biographies behind the abstract figures. Patient 9 was a highly incontinent, laxative dependent 10 year old boy with a high rectourethral fistula after anal atresia (30% of the feces in the toilet, no clean days). In the past 3 years after therapy both he and his mother

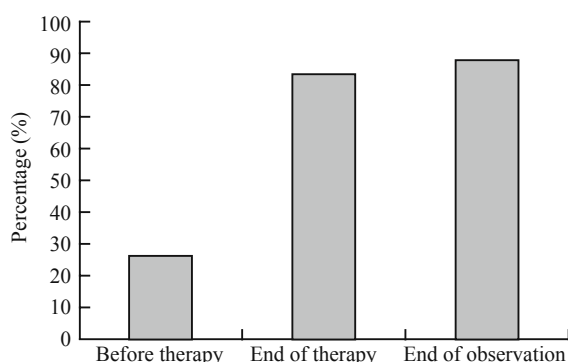


Fig. 1. Results of the 10 patients who finished MBT: average percentage of feces in the toilet.

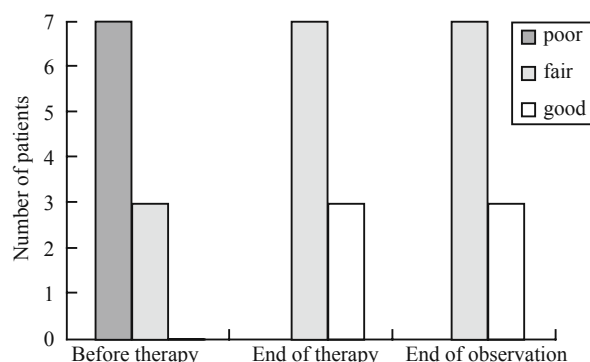


Fig. 2. Results of the 10 patients who finished MBT: Templeton continence score.

Table 2. Survey of the 10 patients who finished MBT

Number of patients	1	2	3	4	5	6	7	8	9	10
Sex	boy	boy	boy	boy	boy	boy	girl	boy	boy	boy
Type of atresia	high	high	high	high	high	high	low (vest.)	low (perin.)	high	high
Age at the start of therapy (y)	10	6	13	12	7	12	9	13	10	14
Percentage of stool in toilet before therapy	80% (b.m.)	25%	25%	60%	5%	99% (b.m.)	20%	55%	30%	95%
Percentage of stool in toilet at the end of therapy	90%	65%	80%	90%	50%	80%	99%	80%	100%	99%
Percentage of stool in toilet at the end of observation	90%	90%	90%	90%	50%	90%	99%	95%	100%	99%
Maximum of clean days before therapy	14 (b.m.)	0	0	1	0	2 (b.m.)	3	0	0	0
Maximum of clean days at the end of therapy	2-3 (b.m.)	0	7	4	0	4	7	6	90	2
Maximum of clean days at the end of observation	3 (b.m.)	2	3	4	0	4	3	6	365	7
Constipation* before therapy	3 (b.m.)	1	0	1	3	2 (b.m.)	3	0	2	0
Constipation* at the end of therapy	?†	0	0	0	0	2	0	0	1	0
Constipation* at the end of observation	?†	0	0	0	0	2	1	0	0	0
Duration of therapy (mon)	5	23	6	13	8	10	3	13	3	4
Observation time (mon)	9	17	15	11	2	4	18	19	36	9

*: constipation classified according to Pena.^[2] 0: no constipation; 1: manageable by changes in diet; 2: requires laxatives; 3: requires enema. †: two rectal irrigations are needed per week, therefore degree of constipation can not be assessed. (vest.): vestibular fistula; (perin.): perineal fistula; (b.m.): patient was under routine bowel management, meaning daily enema.

did not observe any soiling or constipation. Patient 8 was a 13-year-old boy with perineal fistula after ARM, who struggled with very loose stools despite an intake of dark chocolate and loperamide. He was deeply depressed because of his fecal incontinence and uttered suicidal fantasies. Now he has discovered a positive attitude towards life. He sees himself (according to the family) as a "whole person again" and now travels to summer camp with his peers. Recently he has received his "dream job" in the vocational training program, even after his future employer checked all of his medical records.

Discussion

Fecal incontinence is recognized to be a challenging problem after correction of ARM despite improved surgical techniques. A widely accepted explanation is the congenital defect of the continence organ itself, ie, the hypotrophy of the sphincter muscles, the lack of sensomotoric reflexes, and of snugly fitting vascular plexus. Treatment of fecal incontinence commonly aims at strengthening the sphincter muscles, either through physiotherapeutic (biofeedback) training of contractions^[8] or surgically, through Pickrell's gracilis muscle transfer.^[9] Even the implantation of an artificial sphincter has been advocated.^[10,11] However, these measures may not be satisfactory, especially in cases of high types of ARM. Therefore, a refined bowel management has been propagated^[12] and stool consistency may be influenced in either ways by medical treatment, supplemented by a food and fluid intake that meets personal requirements rather than habits.

It was a team from Nijmegen which emphatically focused on the missing development of reflexory defecation in patients with ARM that subsequently may result in a psychosomatic avoidance of passing stools.^[4,5] According to their understanding, the lack of an anal opening prevents the infant from developing a normal, reflexive or active defecation. This may be in the newborn with ARM but in a baby or child after anorectoplasty as well, either while permanently passing only small amounts of stool without filling and distension of the rectum or while withholding stool. Therefore, the prerequisite for the toilet training, normally beginning around the age of three years, is missing. Furthermore, patients after ARM may develop a psychic reluctance against defecation either as a result of recurrent manipulations on their pelvic floor, perianal sores, and painful bowel emptying or later, because of shame and social problems resulting from their fecal incontinence. We confirmed these

findings especially in patients after high ARM in that they regularly pushed away the frustrating subject of defecation. They chose a constipating diet (few meals, fibre reduced) and paradoxically contracted their pelvic floor instead of pushing and relaxing it during defecation, thus worsening their situation. As the frustrated parents often presume, the motivation of their children's behaviour is surely not laziness or disobedience but a helpless attempt to negate the unsolvable problem, so that they can turn to other psychomotor developmental tasks. The degree of psychosocial injury through incontinence is judged differently in the medical literature. Ludman and Spitz^[13] described a correlation between psychosocial adjustment and the incontinence score in a psychiatric interview and teacher's assessment of behavioural maladjustment. Ginn-Pease et al^[14] found no difference in the psychosocial adjustment of the children with ARM, compared to those with abdominal wall defects, although their ARM-patients in 47% mentioned problems of constipation and/or incontinence.

We are convinced that in a large proportion of pediatric surgical patients this psychosomatic disorder of defecation worsens incontinence and/or constipation. In this respect, any anorectal trauma needs reconsideration which may be caused by therapeutic anal manipulation such as dilatations required to prevent a child from postoperative stenosis. This is recognised to outlast up to adulthood, in that a correlation to fear, withdrawal, anxious/depressed mood, and delinquent or aggressive behaviour was detected.^[15] Some pediatric surgeons tend to wait for an automatic improvement of incontinence during puberty, which in fact occurs while the patients no longer avoid the problem trying to reach "social continence" by any means. Pena^[2,12] opposes this view. He not only introduced fundamental surgical strategies for children with ARM, but also initiated a consequent readjustment of aftercare including early and aggressive treatment of chronic constipation by means of retrograde washouts termed bowel management. The basic principle of MBT recognizes the fact that even with a weak continence organ the child can learn to pass enough stool into the toilet voluntarily, so that his fecal incontinence is reduced to a socially acceptable degree.^[4] To reach this goal, the child has to overcome his fear and avoidance of defecation and learn how to push properly while relaxing his pelvic floor. Our results indicate, similar to those from the Nijmegen group,^[5] that fecal incontinence will be reduced by MBT and that the patients develop quite a good perception of urge as a basis of voluntary defecation. Because this therapy is closely linked to the personal development of the children, we do suspend them from MBT when they are

satisfied with the degree of continence having reached. The therapy is continued when the child has gained a new level of maturity and considers his continence as no longer sufficient. Therefore, the distinction between finished and not yet finished therapies in our evaluation is in fact not as strict as it seems.

Having studied the results from Nijmegen,^[5] the advantage of MBT may be not striking with a view on their data alone: an increase from 53% to 61% of feces in the toilet and from 5.6 to 6.8 days without soiling may not show enough improvement. In some cases our figures are better, but in others they still indicate only a fair state of fecal continence, especially when the Templeton score is used for evaluation. Readjustment in view of the whole group of ARM patients remains to be seen. However, the preliminary data are outshined by changes in the personality of the children observed during therapy. They do no longer behave as a passive object, surrendering to incontinence, constipation, and enema, but become an active subject, attacking their problems and reaching acceptable results through their own resources. This considerable "side effect" is noticed by the parents as well and is one of the main achievements of MBT not evaluated by our scoring system so far. Further studies, which are under way, will additionally have to focus on this subject.

In conclusion, we recommend MBT as a basic measure in all patients after ARM suffering from incontinence by the age of 3 to 4 years when physiologic bowel control can be expected. It improves bowel rhythm and defecation habits in addition to routine treatment modalities. MBT fosters changes in the personality of the patients, giving them the best starting conditions for any of the additional therapies available, including surgical ones. However, a sufficient number and committed therapists are required for this approach as well as a durable compliance of the children and their parents.

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References

- Hassink EA, Rieu PN, Severijnen RS, Brugman-Boezeman AT, Festen C. Adults born with high anorectal atresia—how do they manage? *Dis Colon Rectum* 1996;39:695-699.
- Pena A. Anorectal malformations. *Semin Pediatr Surg* 1995;4:35-47.
- Rintala R, Mildh L, Lindahl H. Fecal continence and quality of life in adult patients with an operated low anorectal malformation. *J Pediatr Surg* 1992;27:902-905.
- van Kuyk EM, Brugman-Boezeman AT, Wissink-Essink M, Severijnen RS, Festen C, Bleijenberg G. Biopsychosocial treatment of defecation problems in children with anal atresia: a retrospective study. *Pediatr Surg Int* 2000;16:317-321.
- van Kuyk EM, Wissink-Essink M, Brugman-Boezeman AT, Oerlemans HM, Nijhuis-van der Sanden MW, Severijnen RS, et al. Multidisciplinary behavioral treatment of defecation problems: a controlled study in children with anorectal malformations. *J Pediatr Surg* 2001;36:1350-1356.
- Levitt MA, Pena A. Treatment of fecal incontinence. In: Holschneider AM, Hutson JM, eds. *Anorectal malformations in children*. Berlin: Springer, 2006: 377-383.
- Templeton JM Jr, Ditesheim JA. High imperforate anus—quantitative results of long-term fecal continence. *J Pediatr Surg* 1985;20:645-652.
- Olness K, McParland FA, Piper J. Biofeedback: a new modality in the management of children with fecal soiling. *J Pediatr* 1980;96:505-509.
- Konsten J, Baeten CG, Spaans F, Havenith MG, Soeters PB. Follow-up of anal dynamic graciloplasty for faecal continence. *World J Surg* 1993;17:404-409.
- Lehur PA, Zerbib F, Neunlist M, Glemain P, Bruley SDV. Comparison of quality of life and anorectal function after artificial sphincter implantation. *Dis Colon Rectum* 2002;45:508-513.
- Riedel JG, Festge OA. A new treatment method in severe fecal incontinence in children. *Chirurg* 1999;70:935-938.
- Pena A, Guardino K, Tovilla JM, Levitt MA, Rodriguez G, Torres R. Bowel management for faecal incontinence in patients with anorectal malformations. *J Pediatr Surg* 1998;33:133-137.
- Ludman L, Spitz L. Psychosocial adjustment of children treated for anorectal anomalies. *J Pediatr Surg* 1995;30:495-499.
- Ginn-Pease ME, King DR, Tarnowski KJ, Green L, Young G, Linscheid TR. Psychosocial adjustment and physical growth in children with imperforate anus or abdominal wall defects. *J Pediatr Surg* 1991;26:1129-1135.
- Diseth TH, Egeland T, Emblem R. Effects of anal invasive treatment and incontinence on mental health and psychosocial functioning of adolescents with Hirschsprung's disease and low anorectal anomalies. *J Pediatr Surg* 1998;33:468-475.

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