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MAIN TOPIC

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Biopsychosocial treatment of defecation problems in children with anal atresia: a retrospective study

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Abstract In a retrospective study, we examined whether multidisciplinary treatment based on a biopsychosocial approach and carried out by a pediatric surgeon, a child psychologist, and a pediatric physiotherapist is successful in reducing defecation problems (incontinence and/or constipation) in children with operated anal atresia (AA) (mean age 6.9 ± 4.01 years). A second question was whether this treatment is successful in young children aged 2–5 years. The multidisciplinary approach consisted of standard medical treatment and a behavioral program to teach children and their parents adequate defecation behavior including an adequate straining technique. Forty-three children aged 2–16 years were included: 27 boys and 16 girls with AA, of whom 26 had high or intermediate and 17 low AA. Besides continence and constipation, defecation behavior and straining technique were evaluated. The children improved significantly during treatment in all aspects of defecation. No differences in effect of treatment were found between young children (2–5 years) and older ones, so this treatment seems to be equally effective in both age groups. This study demonstrates that both somatic and behavioral factors contribute to the persistence of chronic defecation problems. It is concluded that treatment of these problems in patients with operated AA should include behavioral modification techniques.

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Introduction

Following surgical correction, a majority of children with anal atresia (AA) continue having problems with incontinence and/or chronic constipation. In the recent literature, we found that these defecation problems were more severe than was expected in childhood, adolescence, and adulthood [1–5, 7, 10, 14, 16, 17]. The level of incontinence varies from staining (mostly in low AA) to intermittent soiling or complete incontinence (mostly in high or intermediate AA). The level of constipation varies as well. According to the severity of these problems, the use of remedies varies from a simple diet or mild laxatives to the daily use of enemas or bowel irrigations, or even a stoma. The arsenal of medical treatments for defecation problems consists of dietary and toilet advice, oral or anal medication, dilatations, or surgical operations. These treatments are basically aimed at somatic factors and pay little attention to behavioral factors.

From a biopsychosocial view, we developed a multidisciplinary treatment directed at behavioral aspects that is supplementary to the medical treatment and carried out by a child psychologist, a pediatric physiotherapist, and a pediatric surgeon. This treatment is indicated for children with defecation problems that cannot be solved by standard medical treatment (SMT) alone and children who experience SMT as troublesome. We focus on teaching the child adequate defecation behavior, including an adequate defecation technique.

The aim of the present retrospective study was to answer the following questions: (1) Is multidisciplinary treatment effective in reducing defecation problems in children born with AA? (2) Is this treatment equally effective in children with low, intermediate, or high AA? and (3) Is this treatment also suitable and effective in children aged 2–5 years?

Materials and methods

The multidisciplinary treatment of 43 children (27 boys and 16 girls) with bowel problems and AA during the last decade was evaluated (Table 1). Twenty-six were treated by a child psychologist and pediatric physiotherapist together with a pediatric surgeon; 38 were treated by a child psychologist and a pediatric surgeon; and 5 were managed by a pediatric physiotherapist in co-operation with a pediatric surgeon.

Of the 43 children (aged 2–16 years, mean 6.9), 26 (60%) had a high or intermediate anorectal malformation (ARM) and 17 (40%) had a low ARM; 31 (72%) had more or less severe additional abnormalities. Nineteen (61%) (4 with low and 15 with high AA) were considered to have an additional abnormality relating to defecation (e.g., urinary tract and/or vertebral column). Twenty children (47%) were 5 years old or younger at the start of treatment.

All children were operated upon by posterior sagittal anorectoplasty according to Peña-de Vries [15] and treated by pediatric surgeons of the University Hospital Nijmegen. They were referred to the multidisciplinary team by a pediatric surgeon because of severe problems that could not be solved by SMT alone. The number of treatment sessions varied from 2 to 30 with an average of 1.4 ± 6.0. Each session lasted 45–60 min.

To evaluate the multidisciplinary treatment, the records of all children were studied. The data at the start of and following treatment were collected. The following aspects of defecation were examined: continence, constipation, defecation behavior, and, if possible, straining technique. The child psychologist scored all aspects of defecation except for straining technique, which was scored by a pediatric physiotherapist.

Continence

Continence was evaluated by the Templeton and Wingspread continence-scoring schemes.

The Templeton scheme is a quantative fecal scoring scheme that does not require a physical examination [14]. The method assigns points for the degree of awareness of impending stool, occurrence of accidental defecation, need for extra underwear or liners, social problems, restriction of physical activity, and presence of diaper rashes to a maximum of 5 points. A total score of 4 to 5 points means that the child has good continence, 1.5–3.5 points indicates fair continence, and 0–1 points indicates poor continence. Following Ong and Beasley [14], we simplified the Templeton scores by giving children with a good score (4–5) 1 point, those with a fair score (1.5–3.5) 2 points, and those with a poor score (1–0) 3 points.

The method described by Wingspread is a clinical anorectal continence-scoring scheme. A physical examination is not essential [14]. Grades of continence are divided into four main categories: ''clean'' (1 point), ''smearing'' (2 points), ''intermittent soiling'' (3 points), and ''constant soiling'' (4 points).

The continence status of each patient was not corrected for the effect of additional non-surgical therapy such as oral medications or enemas.

Constipation

The pediatric surgeon rated constipation as present or not on clinical grounds. Characteristic of the constipated children was the chronicity of their problems and the dependence upon laxatives varying from oral administration to enemas or bowel irrigation.

Defecation behavior

The psychologist rated defecation behavior on a three-point scale, from acting upon an internal or external stimulus by trying to push out the stool on the potty or toilet (1 point), not wanting to (2 points), or being in total ignorance of the task of evacuation (3 points).

Straining technique

At the start and end of treatment, the physiotherapist evaluated the straining technique on a three-point scale, from being able to strain with sufficient pressure in the right direction while completely relaxing the pelvic floor muscles (1 point) to not being able to strain sufficiently in the right direction and/or being unable to relax the pelvic floor muscles (3 points).

Treatment

The aim of the treatment was to teach the child adequate defecation behavior, including adequate technique, step by step. The pediatric surgeon, the psychologist, and the physiotherapist work closely together. For a detailed description of this treatment, we refer to van Kuyk et al. [9].

Statistical analyses

Non-parametric statistics were used (Wilcoxon, McNemar, and Mann–Whitney U test). Data are shown as means and standard deviations. A two-tailed P value less than 0.05 was considered statistically significant. The data on the various aspects were only included in the statistical analyses when they were known for both the start and end of treatment. For this reason, the numbers of patients differ on the various aspects examined.

Results

Prior to treatment, none of the children had good continence (72% poor and 28% fair) and most (86%) were soiling intermittently (28%) or constantly (58%). Almost one-half (46%) had constipation, while none of the 37 for whom it was known at both the start and end of treatment had good defecation behavior. Most (91%) of the 22 children in whom the straining technique was known at both the start and end of treatment had poor or fair technique initially.

Table 1 Summary of patient data

	Anal atresia			Low anal atresia		High anal atresia	
Age range	Total group	2–5 years	6–16 years	2–5 years	6–15 years	2–5 years	6–16 years
Number	43	20	23	9	8	11	15
Male	27 (63%)	13 (65%)	14 (61%)	3 (33%)	2 (25%)	10 (91%)	12 (80%)
Female	16 (37%)	7 (35%)	9 (39%)	6 (67%)	6 (75%)	1 (9%)	3 (20%)
Mean age	6.9 (4.01)	3.6 (.82)	9.8 (3.35)	3.4 (.73)	8.0 (2.98)	3.7 (.91)	10.8 (3.21)

During treatment, 17 children (43%) improved their continence to good and 21 (53%) to fair. More than one-half were clean (21%) or only staining (40%). Of the 18 children with constipation, more than one-half (55%) recovered. More than one-half of the children improved their defecation behavior to good (63%) and 17 (77%) gained a good straining technique. Considering the group as a whole, the children improved significantly during treatment in all aspects of defecation. The results are presented in Table 2.

Considering children with a Wingspread score of 1 (=clean) or 2 (=staining) as improved, 13 (76%) of the 17 children with low and 13 (50%) of the 26 with high AA could be considered as being successfully treated. Comparing the effect of treatment in children with high or intermediate AA with that in low AA, no significant differences were found, nor were significant differences found between the children with and without an additional congenital defect.

Comparing the results in children aged 2–5 years with those in children aged 5–16 years, no significant differences were found in any aspect of defecation. The results are presented in Table 3.

Discussion

This study suggests that teaching children with congenital anomalies adequate defecation behavior supplementary to SMT is an important contribution in reducing chronic defecation problems, not only in older children, but also in toddlers and pre-school children. By explaining the extra value of multidisciplinary treatment, the question must be raised as to why these children do not learn to be spontaneously continent and to evacuate sufficiently postoperatively. Usually the persistence of bowel problems is explained by the somatic disabilities inherent to the congenital bowel anomaly. We believe, however, that not only somatic factors, but also the interaction between somatic and psychological factors can explain why these children are at risk for developing insufficient or wrong defecation habits.

Besides the lack of a normal urge sensation and an inadequately functioning sphincter mechanism, deviating fecal consistency and diminished control over flatus can interfere with learning voluntary bowel control. Pain and unpleasant sensations in the anal area (e.g., caused by dilatations and enemas) lead to defecation avoidance behavior, including withholding and squeezing instead of relaxing and pushing out the stool. When there is no perception of an urge, there will be no stimulus to elicit defecation behavior, and as a result, learning of voluntary bowel control does not take place. Furthermore, in a child with wrong or missing defecation habits, simply giving toilet advice to the parents will not be effective, the more so due to the fact that there are often conflicts regarding defecation between parents and child. All these factors contribute to the development and persistence of vicious circles, maintaining the incontinence and constipation.

Table 2 Results of multidisciplinary treatment of children with anal atresia

	Start of treatment mean (st. dev.)	Number of patients	End of treatment mean (st.dev.)	Number of patients	Wilcoxson matched pairs[a]
Templeton score	2.7 (0.45)		1.6 (0.59)		$P = 0.00$
$n = 40$					
1 = good continence		0		17	
2 = fair continence		11		21	
3 = poor continence		29		2	
Wingspread score	3.4 (0.85)		2.2 (0.80)		$P = 0.00$
$n = 43$					
1 = clean		2		9	
2 = staining		4		17	
3 = intermittent loss		12		16	
4 = constant loss		25		1	
Constipation	1.5 (0.51)		1.2 (0.41)		$P = 0.01$
$n = 39$					
1 = no constipation		21		31	
2 = constipation		18		8	
Defecation behavior	2.8 (0.39)		1.4 (0.55)		$P = 0.00$
$n = 38$					
1 = good		0		24	
2 = fair		7		13	
3 = poor		31		1	
Straining technique	2.5 (0.67)		1.2 (0.43)		$P = 0.00$
$n = 22$					
1 = good		2		17	
2 = fair		7		5	
3 = poor		13		0	

[a] $P \leq 0.05$ statistically significant

Table 3 Effect of treatment on younger (2–5 years) compared to older children (6–16 years)

	Start of treatment mean (st. dev.)		End of treatment mean (st. dev.)		Mann–Whithney U test[a]
	2–5 years	6–16 years	2–5 years	6–16 years	Change scores (scores at the start) − (score at the end)
Templeton score	2.7 (0.46)	2.7 (0.46)	1.7 (0.57)	1.6 (0.60)	$P = 0.48$
$n = 40$					
1 = good continence	0	0	6	11	
2 = fair continence	5	6	11	10	
3 = poor continence	13	16	1	1	
Wingspread score	3.5 (0.83)	3.4 (0.88)	2.3 (0.86)	2.1 (0.76)	$P = 0.52$
$n = 43$					
1 = clean	1	1	4	5	
2 = staining	1	3	7	10	
3 = intermittent loss	6	6	8	8	
4 = constant loss	12	13	1	0	
Constipation	1.7 (0.46)	1.2 (0.44)	1.3 (0.49)	1.1 (0.30)	$P = 0.08$
$n = 39$					
1 = no constipation	5	16	12	19	
2 = constipation	13	5	6	2	
Defecation behaviour	2.9 (0.32)	2.8 (0.44)	1.5 (0.62)	1.3 (0.47)	$P = 0.74$
$n = 38$					
1 = good	0	0	10	14	
2 = fair	2	5	7	6	
3 = poor	16	15	1	0	

[a] $P \leq 0.05$ statistically significant

Contrary to our expectations, we found no differences in the effects of treatment between children with high or intermediate and low AA. Although the children with low AA had slightly better continence scores at the start and end of treatment than those with intermediate or high AA, the degree of improvement was equal.

The finding that treatment is as effective in toddlers and preschool children is very important. There are many disadvantages to not treating these children until they are at least 6 years old (e.g., as in biofeedback training): chronicity of bowel problems, increased medical consumption, as well as frequent invasive procedures (clysma, bowel irrigations, dilatation) in a sensitive region. Furthermore, intrusive procedures in this important developmental period might be especially harmful, disturbing the child's natural psychosocial development and affecting parent-child interactions [1–3]. Therefore, by treating children at as young an age as possible, we both utilize the critical period for learning new abilities and much time and effort is saved with regard to the chronicity of defecation problems. It is also easier to learn a new technique at a younger age than to change a wrong technique at an older age.

Not all children in this retrospective study benefited satisfactorily from multidisciplinary treatment. This might be due to both somatic and psychosocial factors, which, although not measured, according to our clinical experience interfere with learning voluntary bowel control. At the somatic level, the severity of the disorder as well as additional congenital defects such as urodynamic deviations and mental retardation can interfere with the necessary conditions for the learning process. Psychosocial factors interfering with teaching a child voluntary bowel control might be due to incompetence in child-rearing practices by the parents and/or family psychopathology.

Several other methodological problems are bound to this research. Because of the retrospective nature of the study, the data are incomplete and not scored blindly, making it difficult to draw firm conclusions. Because the data concerning straining technique are limited, it is especially difficult to draw conclusions concerning the role it plays in the defecation problems of these children. Prospective studies are needed. Finally, many authors [2, 4, 7] highlight the need for treatments that improve the child's continence. The same applies to the need for psychosocial support of the parents at an early stage, because of their important role in teaching the child to cope effectively with his handicap [1, 2, 11, 13]. By reducing bowel problems in the child in co-operation with the parents, this treatment is a contribution to improvement of the quality of life of these children and their parents [6, 16, 17].

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