Fecal incontinence: tests & therapy

Dobben, A.C.

Publication date
2006

Document Version
Final published version

Fecal incontinence: Tests & Therapy

This thesis was prepared at the Department of Radiology, Academic Medical Center, University of Amsterdam, the Netherlands.

This research was financially supported by ZON-MW (the Netherlands Organization for Health Research and Development): "An efficient diagnostic strategy in fecal incontinence", grant 945-01-013.
Equipment support for this research was provided by Medeco B.V., Oud Beijerland, the Netherlands; PelviTec B.V., Delft, the Netherlands; Enraf-Nonius B.V., Rotterdam, the Netherlands.
Printing of this thesis is financially supported by Medtronic B.V., Heerlen and General Electrics Medical Systems, 's-Hertogenbosch, the Netherlands.

Cover: Picture on front page by courtesy of photographer Mr. Luuc Stafleu, Amsterdam, the Netherlands.
Layout: Chris Bor, Medical Photography and Illustration, Academic Medical Center, Amsterdam, the Netherlands.
Printed by: Buijten & Schipperheijn, Amsterdam, the Netherlands.

ISBN-10: 90-9021147-0
ISBN-13: 978-90-9021147-3

Fecal incontinence: Tests & Therapy

Academisch Proefschrift

ter verkrijging van de graad van doctor
aan de Universiteit van Amsterdam
op gezag van de Rector Magnificus
prof.mr. P.F. van der Heijden
ten overstaan van een door het college voor promoties ingestelde
commissie, in het openbaar te verdedigen in de Aula der Universiteit
op donderdag 14 december 2006, te 10.00 uur

door

Annetta Christina Dobben

geboren te Groningen

Promotores:	Prof. dr. J. Stoker
	Prof. dr. P.M.M. Bossuyt
Overige leden:	Prof. dr. W.P.Th.M. Mali
	Prof. dr. P. Fockens
	Prof. dr. C.J. Mulder
	Prof. dr. J.S. Laméris
	Dr. W.A. Bemelman
	Dr. W.R. Schouten

Faculteit der Geneeskunde

Contents

General introduction and Outline of the thesis

Chapter 1

General introduction

Epidemiology

Fecal incontinence is defined as the involuntary passage of fecal material through the anal canal[1]. While not life-threatening, fecal incontinence constitutes a substantial social problem for those afflicted and a relevant public health burden. It is a disorder with a high social stigma.

International population-based studies have provided widely varying estimates of prevalence, ranging from 0.004% to 18%[2]. The incidence and prevalence of fecal incontinence in the Netherlands are not exactly known[3]. The estimated prevalence is about 100.000 subjects in the Netherlands. The true prevalence of fecal incontinence is likely underestimated due to the shame and embarrassment that individuals experience and the failure of many affected individuals to disclose this condition to their physicians[4, 5]. Prevalence increases steadily with age, from approximately 4% for incontinence in men and women aged between 40 and 49 years old, to 12% in patients aged 80 years and older. The prevalence in nursing home residents is considerably higher, approaching 47%[6]. Although the prevalence increases with age, fecal incontinence also affects younger people, in particular women as a consequence of obstetric factors.

Etiology

The anal sphincter complex consists of several pelvic floor muscles. The smooth muscle of the internal anal sphincter has autonomic innervation and contributes about 55% of the resting tone of the anal canal[2]. The external anal sphincter is a striated muscle that provides between 20 and 30% of the anal resting tone. The external anal sphincter and puborectal muscle function as one unit, bring about voluntary sphincter contraction, and normally double the sphincter pressure of the anal canal during voluntary contraction. The external anal sphincter as well as the puborectal muscle are innervated by branches of the pudendal nerve. Sensation is provided by receptors in the pelvic floor that detect rectal distension and by various receptors in the anal transition zone[2].

A number of factors are important in the maintenance of fecal continence, including stool consistency, rectal distensibility, anorectal sensation, anal sphincter integrity, and mental function. Impairment of one of these factors can cause fecal incontinence. Consequently, the cause of fecal incontinence is often multifactorial[7]. The main cause of fecal incontinence is a complicated vaginal delivery due to sphincter disruption[8]. Other causes are anorectal surgery and trauma from impalement or pelvic fracture, accounting for much of the fecal incontinence seen in men. Furthermore pudendal neuropathy, impaired anorectal sensation and rectal accommodation, or incomplete evacuation may all contribute to the pathogenesis of fecal incontinence as a result of anatomical or functional disorders[7]. Congenital malformations, such as imperforate anus, can also cause fecal incontinence.

A large number of fecal incontinent patients do not have any of these etiological factors, and are defined as having idiopathic fecal incontinence. Pelvic floor neuropathy resulting from childbirth, from excessive straining during evacuations, or in patients with a rectal prolapse are considered to contribute to this idiopathic form of fecal incontinence[1, 2].

Diagnostic work-up

Medical history and physical examination

A detailed history is essential to select and guide the diagnostic and therapeutic approach in a patient. Diaries and questionnaires are useful adjuncts in this regard. The severity of fecal incontinence can be assessed with an incontinence score. A widely used score is the Vaizey score containing several incontinence-specific items[9].

A complete physical examination will identify structural disorders (e.g. prolapse, perineal disease) and local and systemic disease processes that may affect anorectal function and dysfunction. In addition, some assessment of perineal innervation can be obtained through observation of perianal sensation and reflex contraction of the external anal sphincter to perianal stimulation and cough[10]. Digital rectal examination gives accurate information on sphincter pressures reflecting internal and external anal sphincter function but is not always reliable in detecting anal sphincter defects[2].

Anorectal function tests

Anorectal function tests can be used to explore the underlying causes of fecal incontinence. There exists considerable practice variation in terms of diagnostic tests used. Consequently, identical patients are now managed differently, based on the local availability of techniques, on personal preferences or on tradition.

Anorectal manometry evaluates the muscular contraction and relaxation of the anal sphincters by the measurement of pressures in the anal canal. Resting pressure reflects the internal anal sphincter function and squeeze pressure reflects the external anal sphincter function. In addition, the recto-anal inhibition reflex (RAIR) can be measured which reflects the inhibition of the internal anal sphincter either in response to rectal distension or during attempted defecation.

Pudendal nerve terminal motor latency testing measures the conduction of the pudendal nerves. The latency measured reflects the function of the fastest conducting nerve fibers. Consequently, a damaged nerve may nevertheless show a normal conduction time as long as some fast-conducting fibers remain[10]. In view of the lack of correlation between pudendal nerve terminal motor latencies, fiber density and outcome, the use of neurophysiological evaluation may be questioned[11].

With rectal and anal mucosal sensitivity measurement the threshold sensation of rectum and anus, respectively, can be determined. The anal and rectal sensitivity are both a summation of characteristics of the central and peripheral nerve function, submucosa and mucosa.

With rectal capacity measurement the reservoir capacity of the rectum can be determined. The reservoir capacity is crucial for normal anorectal functioning. This comprises the capacity to temporarily store faeces as well as the accurate sense of fullness of the rectum. The minimal rectal sensation perceived (sensory threshold), the volume associated with the initial urge to defecate (urge sensation), and the volume at which the patient experienced discomfort and an intense desire to defecate (the maximal tolerated volume) can be determined.

Electromyography for the detection of an external anal sphincter defect has been replaced because of the availability of other techniques, including endoanal ultrasonography and endoanal magnetic resonance (MR) imaging, as these techniques are less painful[10, 12].

Imaging techniques
Imaging can roughly be divided into imaging of the anal sphincter (endoanal ultrasonography and endoanal MR imaging) and imaging of the evacuation process (defecography).

Endoanal ultrasonography
Endoanal ultrasonography is widely available. It is the least expensive test for defining structural defects of the anal sphincter. However, the technique is operator dependent and requires both training and experience[7]. Endoanal ultrasonography provides an accurate assessment of the structural integrity of the external and internal anal sphincter and can depict scarring and loss of muscle[13]. The internal anal sphincter has clearly defined borders in contrast to the external anal sphincter[14]. Consequently, generalized external anal sphincter atrophy is difficult to appreciate because of the vague contours of the muscle ring[15].

Endoanal MR imaging
Endoanal MR imaging can depict the anal anatomy in considerable detail due to its high spatial resolution. There is a large contrast between the external anal sphincter muscle and the surrounding fat[16]. Besides defects of the external and internal anal sphincter, other forms of damage to the sphincter complex may also be encountered, such as scar tissue. A major contribution of endoanal MR imaging is the depiction of external anal sphincter atrophy and how this may adversely affect anterior anal sphincter repair[7]. Disadvantages of endoanal MR imaging are its high costs and limited availability[16].

Defecography
Defecography (evacuation proctography) involves imaging of the rectum with contrast material and observation of the process, rate, and completeness of rectal evacuation. Structural and functional alterations can also be observed[10].

The role of imaging the evacuation process by defecography in de diagnostic work-up op fecal incontinent patients has not been elucidated. As a consequence, the use of this technique is still a matter of debate as defecography is of rather limited value in incontinent patients who do not suffer from associated obstructive symptoms[15].

More recently, MR defecography has become an alternative for conventional defecography. It avoids radiation exposure and enables visualization of all organs of the pelvis at the same time. However, MR defecography is not widely available and the technique is still developing[14].

Treatment options
The treatment options for fecal incontinent patients range from conservative (dietary measures, medication, pelvic floor rehabilitation) to surgical procedures[2]. Currently, the drug therapy comprises antidiarrheal agents, bulk-forming agents, and laxatives[17]. If patients are unresponsive to these therapeutic approaches, pelvic floor rehabilitation can be the next available treatment option. Pelvic floor rehabilitation (electrical stimulation and /or pelvic floor muscle training with biofeedback) is a non-surgical treatment for fecal incontinence in which patients are trained to increase the anal sphincter contractile capacity in response to rectal distension[18].

Surgery is offered when the response to conservative therapy is not satisfactory. Surgical treatment options of fecal incontinence can be categorized into procedures that either repair or augment the native sphincter mechanism or, alternatively, those that aim at the construction of a neo-sphincter, using either autologous tissue or an artificial device. Overlapping anterior anal sphincter repair has become the operation of choice in patients with isolated anterior defects in the external anal sphincter[19]. Sacral nerve stimulation and neo-sphincter procedures are considered as treatment options when conservative treatment or anterior anal sphincter repair has failed[20].

Aim and outline of this thesis

Next to dietary measures and medication, pelvic floor rehabilitation and surgery are treatment options for fecal incontinence. The ability to identify patients with a high probability of success with treatment, distinguishing them from those with a low probability of success, would enable us to encourage the first group to undergo therapy, while withholding unnecessary treatment from the second group. In addition, such a strategy would allow us to eliminate redundant tests, thereby further increasing the efficiency of health care. Identification of patients could be made using information from medical history, physical examination, anorectal function tests and imaging techniques.

The aim of the work summarized in this thesis was to build an evidence base to develop an optimal diagnostic strategy for fecal incontinent patients. Chapter 2 and chapter 3 concern diagnostics in fecal incontinence. Several diagnostic modalities are available in the Netherlands. **Chapter 2** reports if and to what extent variation exists in the Netherlands in diagnostic work-up of patients with fecal incontinence in daily clinical practice. **Chapter 3** addresses if and how findings of anal inspection and digital rectal examination are associated with anorectal function tests and endoanal ultrasonography.

Chapter 4 and chapter 5 concern imaging in fecal incontinence. In **chapter 4** imaging of the evacuation process is studied. In this chapter we report the interobserver agreement of defecography in diagnosing enterocele, anterior rectocele, intussusception and anismus. The influence of different levels of experience on reading defecographies was also evaluated. **Chapter 5** addresses all appropriate issues of different kind of endoluminal imaging related to fecal incontinence, such as imaging protocols, pelvic floor anatomy and relevant pathology as well as a systematic approach of reading these examinations.

The remaining chapters concern treatment options. Chapter 6, chapter 7 and chapter 8 focus on pelvic floor rehabilitation. **Chapter 6** describes the outcome of pelvic floor rehabilitation in a large series of fecal incontinent patients. In addition, the outcome across a number of subgroups was compared. **Chapter 7** presents how anorectal function changes after pelvic floor rehabilitation and whether these changes are related to changes in fecal incontinence score. Furthermore, the association of changes in anorectal function with predisposing factors is described. **Chapter 8** evaluates the ability of tests to predict the outcome after pelvic floor rehabilitation.

Chapter 9 and chapter 10 focus on imaging in relation to surgery. **In chapter 9** the accuracy of endoanal MR imaging and endoanal ultrasonography for the depiction of external anal sphincter defects in fecal incontinent patients is reported, based on a prospective study using surgery as a reference standard. The study presented in **chapter 10** addressed whether findings obtained at preoperative endoluminal imaging can predict the outcome of sphincter repair. In addition, the association of postoperative endoluminal imaging findings with poor outcome is described.

Chapter 11 focuses on remaining therapy, namely containment products. A systematic review of studies on the effectiveness of anal plugs for controlling fecal incontinence is presented.

References

1. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-80
2. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *The Lancet* 2004; 13;364:621-32
3. Felt-Bersma RJ, Sloots CE. [Fecal incontinence: various causes and treatments]. *Ned Tijdschr Geneeskd* 2001;19;145:937-41
4. Mavrantonis C, Wexner SD. A clinical approach to fecal incontinence. [Review]. *Journal of Clinical Gastroenterology* 1998;27:108-21
5. Johanson JF, Lafferty J. Epidemiology of fecal incontinence: the silent affliction. *American Journal of Gastroenterology* 1996;91:33-6
6. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-85
7. Rao SS. Diagnosis and management of fecal incontinence. American College of Gastroenterology Practice Parameters Committee. *Am J Gastroenterol* 2004;99:1585-604
8. Kamm MA. Obstetric damage and faecal incontinence. *Lancet* 1994;10;344:730-3
9. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
10. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-60
11. Osterberg A, Edebol Eeg-Olofsson K, Graf W. Results of surgical treatment for faecal incontinence. *Br J Surg* 2000;87:1546-52
12. Law PJ, Kamm MA, Bartram CI. A comparison between electromyography and anal endosonography in mapping external anal sphincter defects. *Dis Colon Rectum* 1990;33:370-3
13. Law PJ, Kamm MA, Bartram CI. Anal endosonography in the investigation of faecal incontinence. *Br J Surg* 1991;78:312-4
14. Stoker J, Halligan S, Bartram CI. Pelvic floor imaging. *Radiology* 2001;218:621-41
15. Fuchsjager MH, Maier AG. Imaging fecal incontinence. *Eur J Radiol* 2003;47:108-16
16. Stoker J, Rociu E, Wiersma TG, Lameris JS. Imaging of anorectal disease. *Br J Surg* 2000;87:10-27
17. Whitehead WE, Wald, A, Norton NJ. Priorities for treatment research from different professional perspectives. *Gastroenterology* 2004;126:S180-S185
18. Chiarioni G, Bassotti G, Stegagnini S, Vantini I, Whitehead WE. Sensory retraining is key to biofeedback therapy for formed stool fecal incontinence. *Am J Gastroenterol* 2002;97:109-17
19. Baig MK, Wexner SD. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2000;87:1316-30
20. Madoff RD. Surgical treatment options for fecal incontinence. *Gastroenterology* 2004;126:S48-S54

Chapter 2

Diagnostic work-up for fecal incontinence in daily clinical practice in the Netherlands

A.C. Dobben
M.P. Terra
M. Deutekom
P.M.M. Bossuyt
R.J.F. Felt-Bersma
J. Stoker

Neth J Med. 2005 Jul-Aug;63(7):265-9

Abstract

BACKGROUND. To study variation in Dutch hospitals in applying diagnostic and treatment options for fecal incontinence.

METHODS. Surgeons, gastroenterologists, internists and gynecologists were contacted by phone or mail and requested to complete a questionnaire. The questionnaire asked for general information about patients with fecal incontinence, the use and availability of diagnostic techniques, the use of incontinence scores and therapeutic options.

RESULTS. In total 306 specialists were contacted and data were collected from 203 specialists from 86 hospitals (response rate 66%). The most frequently applied diagnostics were sigmoidoscopy (64%), endoanal ultrasonography (58%), evacuation proctography (56%) and/or anorectal manometry (51%). The choice seemed to be related to the availability of the techniques. Sigmoidoscopies were performed significantly more often in local hospitals ($p<0.001$), while in academic medical centres significantly more endoanal MR imaging examinations were conducted ($p<0.05$). The most stated treatment option was pelvic floor rehabilitation (90%), followed by dietary measures (83%), medication (71%) and surgery (68%). However, in general, combinations of treatment options were used.

CONCLUSIONS. A substantial variety exists in the diagnostic work-up of fecal incontinence. In general, at least one anorectal function test and an imaging technique are the diagnostic techniques of choice. Pelvic floor rehabilitation is the first choice in conservative treatment.

Introduction

Fecal incontinence is defined as 'the involuntary loss of flatus, liquid or solid stool that is a social or hygienic problem' [1]. The incidence and prevalence of fecal incontinence in the Netherlands are not exactly known[2]. The estimated prevalence is about 100,000 subjects in the Netherlands. The actual prevalence may be even higher due to underreporting as a consequence of the social stigma of this disorder[3].

The main causes of fecal incontinence are obstetric trauma (anal sphincter and/or pudendal nerve damage) and anorectal surgery (anal sphincter trauma) [4-6]. Apart from medical history and physical examination, there are several diagnostic techniques that can be performed: anorectal function testing, endoscopy and imaging [7-9]. Anorectal function tests comprise anorectal manometry (measurement of sphincter pressure in rest, during squeeze and straining), measurement of pudendal nerve terminal motor latency (PNTML) (to establish pudendal nerve injury), electromyography (EMG) (conventional EMG to identify the quality of sphincter tissue as well as to determine whether the muscle contracts or relaxes; single-fibre EMG to identify denervation-reinnervation potentials indicative of nerve injury), rectal capacity measurement (to detect the threshold of the first detectable sensation, sensation of urgency and maximum tolerable volume) and sensory testing (to determine the sensitivity of the anal canal and rectum)[10,11]. A sigmoidoscopy can be performed to exclude organic disease, such as a benign or malignant obstructing lesion or inflammation[10]. With imaging techniques, such as endoanal ultrasonography and magnetic resonance (MR) imaging, both internal and external anal sphincter abnormalities will be assessed[7]. Evacuation proctography (defecography) involves imaging of the rectum and observation of the process, rate, and completeness of rectal evacuation[10].

At present there is no consensus on the best diagnostic techniques for patients with fecal incontinence in the Netherlands. As a consequence of the increase in number and availability of diagnostic modalities, variation in diagnostics exists and an unambiguous strategy is lacking[10]. To assess if and to what extent variation exists in diagnostic work-up and treatment of patients with fecal incontinence in daily clinical practice in the Netherlands, we developed a survey. We restricted ourselves to an inventory of diagnostic modalities and treatment in secondary and tertiary centers.

Materials and Methods

From October 2002 to April 2004 surgeons, gastroenterologists, internists and gynecologists from all Dutch hospitals were informed about the survey by phone. If approach by phone turned out to be impossible, information was sent out by (digital) mail. For every hospital a questionnaire was sent per discipline to the most experienced specialist in the field of fecal incontinence.

The questionnaire comprised five sections. In the first, physicians were asked for general information about patients with fecal incontinence, such as how often these patients were referred to the respondent, and the age, and gender of the referred patients. In the second section information was requested about the selection of diagnostic tests which were used as routine work-up in patients with fecal incontinence. Options were anorectal function tests, endoscopy and imaging techniques (Table 1). In addition to the options for routine diagnostic techniques, in the third part questions were asked about the availability of these techniques to gain insight into where the techniques were performed. The respondent had to indicate whether the diagnostic test in question could be performed in the respondent's own hospital or if referral was needed. The fourth section was about the use of an incontinence score to determine the severity of incontinence. Respondents could choose between the Parks, Vaizey, Wexner, Pescatori, or Millar scores, and/or the American Medical System score[12]. If an incontinence score was used, the respondent was asked whether the score influenced the choice of diagnostic and therapeutic options. The final section contained questions on the therapeutic options used (conservative therapy (dietary measures, medication, pelvic floor rehabilitation), surgery or another kind of therapy).

All nonrespondents received one more reminder by phone and if necessary, a new questionnaire was sent out. If there was no response after three questionnaires had been sent out, a final nonresponse was determined. (Details of the questionnaire can be obtained from the first author)

Analyses were performed with descriptive statistics. Differences between groups were calculated with χ^2 - test. The results were statistically analyzed with SPSS 11.5. for Windows (SPSS Inc. Standard Version). We analyzed the response per specialist instead of per hospital.

Results

Response

In total 306 physicians were contacted (91 surgeons, 74 gastroenterologists, 24 internists and 117 gynecologists) from the 100 Dutch hospitals (we did not take into account categorical hospitals such as cancer institutes and outpatient clinics). The response rate was 66% (n=203) from 86 hospitals and one private clinic. Sixteen percent (n=33 questionnaires) of the response rate originated from academic medical centers. There were differences in response rate per medical specialist: the response rate of surgeons and gastroenterologists was higher (76 and 72% respectively) than that of internists and gynecologists (58 and 57% respectively).

Seventeen (29%) responding gynecologists referred their patients almost directly to another medical specialist or hospital. For the majority of physicians (75%) patients with fecal incontinence were sometimes referred, while only 12% indicated having these patients referred regularly and 3% often.

Sixteen questionnaires (8%) had to be excluded from analysis since the respondent reported no referral of patients with fecal incontinence or referred these patients immediately to another specialist. Consequently, there were 187 questionnaires remaining for analysis, from 80 different hospitals and of one private clinic.

The majority of physicians (92%) indicated that they treated their patients with fecal incontinence on an interdisciplinary and/or multidisciplinary basis.

Patients

Physicians indicated that on average 87% of the patients with fecal incontinence were female. On average almost half of these patients (47%) were more than 65 years of age. Age as well as gender was not significantly influenced by the numbers of patients referred to the physician.

Diagnostic techniques

The range of routine diagnostic techniques applied in patients with fecal incontinence varied from none to 11 examinations. On average 3.5 examinations were performed as the routine diagnostic work-up. In Table 1 the results of differences in options of diagnostic testing are shown. The majority of the respondents (64%) indicated the routine use of sigmoidoscopy. The most frequently applied imaging techniques were endoanal ultrasonography (58%) and evacuation proctography (56%). Of all anorectal function tests, anorectal manometry (51%) was most often used. The use of these techniques seems to be linked to the availability of the diagnostic techniques. The other diagnostic techniques were not performed on a regular basis.

Sigmoidoscopy and evacuation proctography were available for most of the respondents. The highest percentages of referral were for endoanal ultrasonography and anorectal manometry (32 and 39% respectively). The most commonly used combinations of diagnostic techniques were endoanal ultrasonography with anorectal manometry (41%), and sigmoïdoscopie with evacuation proctography (41%). Twelve percent of all respondents reported that they did not perform any

Table 1. Options for diagnostic techniques together with the availability of diagnostic equipment and referral of patients with fecal incontinence

Diagnostic techniques	Options for diagnostics* n (%)	Availability of diagnostics n (%)	Referral n (%)
Endoscopy			
Sigmoidoscopy	120 (64)	166 (89)	2 (1)
Anorectal function tests			
Anorectal manometry	96 (51)	71 (38)	72 (39)
Rectal capacity measurement	42 (22)	44 (24)	48 (26)
PNTML	37 (20)	47 (25)	43 (23)
Anal sensibility measurement	32 (17)	33 (18)	47 (25)
Rectal sensibility measurement	31 (17)	23 (12)	46 (25)
Conventional electromyography	26 (14)	57 (31)	26 (14)
Fine needle electromyography	6 (3)	24 (13)	22 (12)
Imaging techniques			
Endoanal ultrasonography	108 (58)	86 (46)	60 (32)
Evacuation proctography	104 (56)	136 (73)	31 (17)
Endoanal MR imaging	25 (13)	28 (15)	31 (17)
Phased array MR imaging	25 (13)	56 (30)	10 (5)
MR defecography	3 (2)	11 (6)	13 (7)

Note: * The chosen technique concerns the routine diagnostic work-up in fecal incontinent patients. The routine diagnostic work-up could be performed in its own hospital or in a referring center. PNTML = pudendal nerve terminal motor latency; MR = magnetic resonance kind of additional testing; 38% mentioned not performing any anorectal function tests and 3% reported that they did not make use of any kind of imaging technique.

When comparing the routinely performed diagnostics in academic medical centers with those performed in local hospitals, physicians in local hospitals reported significantly more use of sigmoidoscopy ($p<0.001$), while physicians in academic medical centers reported significantly more use of endoanal MR imaging examinations ($p<0.05$) (Table 2).

Incontinence score

Thirty-one percent of the respondents used an incontinence score; 13.5% indicated that they always used a score and 17.5% sometimes. A score was significantly more in use in academic medical centers compared with local hospitals ($p=0.001$) (Table 2). The most applied incontinence score was the Parks score (44%), followed by the more recently introduced Vaizey score (28%). The selection of diagnostic tests and therapeutic treatment options were influenced by an incontinence score in 6%.

Table 2. Significant differences between academic and local hospitals

	Reported use of:					
	Sigmoidoscopy		**Endoanal MRI**		**Incontinence score**	
Academic hospital	15%		29%		60%	
		p < 0.000		p<0.05		p=0.001
Local hospital	73%		11%		28%	

MRI = magnetic resonance imaging

Therapeutic treatment options

The most reported treatment option by the respondents was pelvic floor rehabilitation (90%), followed by dietary measures (83%), medication (71%) and surgery (68%). A combination of treatment options was most frequently reported. Fifty-four percent of the respondents indicated that they applied dietary measures, medication, pelvic floor rehabilitation as well as surgery as treatment options. In 7% (academic medical centers) versus 26% (local hospitals) surgery was not considered a treatment option as patients only received conservative treatment. Other therapies, such as sacral neuromodulation and anorectal or oral water enemas, were part of potential treatment options in 7% of the respondents.

Discussion

In the Netherlands the most performed diagnostics in patients with fecal incontinence are sigmoidoscopy, endoanal ultrasonography, evacuation proctography and anorectal manometry. Since sigmoidoscopy is performed to exclude local pathology such as tumors, and evacuation proctography is not a diagnostic technique specifically for fecal incontinence[7], it can be concluded that most applied diagnostic tests in patients with fecal incontinence in secondary and tertiary centers are anorectal manometry (anorectal function test) and endoanal ultrasonography (imaging technique).

Significantly more sigmoidoscopies were performed in local hospitals ($p<0.001$), while endoanal MR imaging examinations were significantly more frequent in academic medical centers ($p<0.05$). It is possible that availability does play a role, as well as the referral pattern. Almost every physician in a local hospital performs a sigmoidoscopy to exclude malignancy or proctitis, for example, while in general patients are referred to an academic medical center if comprehensive anorectal function testing and/or endoanal MR imaging is needed. There was a considerable variation in the use of the other diagnostic modalities.

Anorectal function tests

Anorectal manometry appeared to be the most commonly applied anorectal function test; it was relatively widely available and had the highest percentage of referral.

PNTML, rectal capacity measurement, and anal and rectal sensory testing were part of routine diagnostic testing to a lesser extent. Nevertheless, approximately 25% of the respondents referred their patients for these tests. It seems that these functional tests are included in the work-up when more extensive diagnostic is mandatory.

Conventional electromyography was reported to be part of the available diagnostic techniques by 31% of all respondents, but only 14% performed it as a routine procedure. Fine needle electromyography was not regarded as routine. These tests are not considered to have any substantial value and to be outdated. The performance of EMG for the detection of an external anal sphincter defect has been replaced by the availability of other techniques, such as endoanal ultrasonography or MR imaging[10,13]. For establishing pudendal nerve injury, PNTML measurement will be performed when considered appropriate[7]. The technique has been suggested for distinguishing between muscle weakness caused by pudendal nerve injury and muscle weakness caused by muscle injury in patients with fecal incontinence, but has a poor correlation with clinical symptoms and histological findings. Therefore, the clinical usefulness is controversial[10].

Imaging techniques

Endoanal and phased-array MR imaging are part of the routine diagnostic work-up for more than 10% of respondents. Endoanal ultrasonography and endoanal MR imaging are comparable techniques for evaluating external anal sphincter abnormalities. For evaluation of the internal anal sphincter complex, there is no consensus about the most accurate technique[8,14,15]. However, the sensitivity and specificity for identifying external anal sphincter atrophy with MR imaging is higher than for endoanal ultrasonography[8].

MR defecography is hardly available. Besides, the accuracy and reproducibility of conventional defecography is not (yet) established and the technique is still in development[14].

Incontinence score

Several incontinence scores have been developed[7,8,12]. Nevertheless, it appears that these scores are rarely used in daily practice. This is probably because the registration of scoring is often a complex matter and the consequences of use, other than for scientific research, have not been clearly pointed out. This study showed that scoring systems according to Parks and Vaizey are the most applied scores in the Netherlands for patients with fecal incontinence. Possible explanations are that the score according to Parks is the most uncomplicated one and the score according to Vaizey is the most complete scoring system[12].

Treatment options

A combination of treatments was predominantly reported, comprising various conservative treatment options (pelvic floor rehabilitation, dietary measures, medication), if necessary complemented with surgery. Of all therapeutic options, pelvic floor rehabilitation was the most widely applied (90%). According to Kamm pelvic floor rehabilitation and surgery are the two most utilised treatment options if dietary measures and/or medication fail[16]. However, in this study the respondents reported that they more often used pelvic floor rehabilitation as initial therapy than other conservative measures. Nevertheless, we must consider that previous conservative treatment may have been prescribed elsewhere by others.

Limitations

Potential limitations of this study should be taken into account. One limitation was that the majority of the respondents reported a relatively infrequent referral of patients with fecal incontinence, which was defined as a range from 1 to 24 patients a year. Because of the wide range, it is possible that differences exist in selected diagnostic and therapeutic options between physicians with one to five referrals a year compared with those with 20 to 24 referrals on a yearly basis.

In some of the participating hospitals this questionnaire was completed by several medical specialties while in others it was completed by only one specialty. Since this questionnaire was completed for the greater part by different medical specialties divergently, we assume it is justified considering that all hospitals have the same weighting.

This study shows that substantial variety exists in the diagnostic work-up for fecal incontinence. In general, at least one anorectal function test and an imaging technique are the diagnostic techniques of choice. Besides, there are differences in work-up between local hospitals and academic medical centers, partly related to the availability of equipment.

In the literature, guidelines for the evaluation of fecal incontinence are described[10,17-19]. In summary they all recommend, next to a detailed clinical assessment, appropriate physiological and imaging tests of the anorectum. These three sources of information are complementary. The anorectal physiology testing of choise in the presented guidelines were anorectal manometry and endoanal ultrasonography, conform the results of our study. Furthermore, between guidelines there was variation concerning the remaining diagnostic modalities.

To reduce variability we encourage developing guidelines for the diagnostic work-up of fecal incontinence in the Netherlands. We recommend that the scope of the guidelines is aimed at simplification of the diagnostic path in patients with fecal incontinence, based on scientific evidence. We want to emphasise the importance of evidence-based guidelines to reduce inadequate use as well as both overuse and underuse. As a consequence, an efficient diagnostic work-up in patients with fecal incontinence can be developed.

Acknowledgement

The authors would like to thank Bart Verwer and Martijn Kross for their help in collecting the data for this questionnaire.

References

1. Cardozo L, Khoury S, Weiri A (eds) Proceedings of the Second International Consultation on Incontinence. 1-3 July 2001. Plymouth: Health Publication Ltd, 2001
2. Felt-Bersma RJF, Sloots CEJ. Fecal incontinence: various causes and treatments [Review]. *Ned Tijdschr Geneeskd* 2001;145:937-41
3. Talley NJ, O'Keefe EA, Zinsmeister AR, Melton JL. Prevalence of gastrointestinal symptoms in the elderly: a population based study. *Gastroenterology* 1992;102:895-901
4. Kamm MA. Obstetric damage and faecal incontinence. *Lancet* 1994;344:730-3
5. Snooks SJ, Henry MM, Swash M. Faecal incontinence due to external anal sphincter division in childbirth is associated with damage to the innervation of the pelvic floor musculature: a double pathology. *Br J Obstet Gynaecol* 1985;92:824-8
6. Toglia MR. Pathophysiology of anorectal dysfunction. *Obstet Gynecol Clin North Am* 1998;25:771-81
7. Madoff RD, Parker SC, Varma MG, Lowry AC. Fecal incontinence in adults. Lancet 2004;364:621-32
8. Barucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-85
9. Hinninghofen H, Enck P. Fecal incontinence: evaluation and treatment. *Gastroenterol Clin North Am* 2003;685-706
10. American Gastroenterological Association. American Gastroenterological Association Medical Position Statement on Anorectal TestingTechniques. *Gastroenterology* 1999;116:732-60
11. Felt-Bersma RJF, Klinkenberg-Knol EC, Meuwissen SGM. Anorectal Function Investigations in incontinent and continent patients. *Dis Colom Rectum* 1990;33:479-86
12. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
13. Law PJ, Kamm MA, Bartram CI. A comparison between electromyography and anal endosonography in mapping external anal sphincter defects. *Dis Col Rectum* 1990;33:370-3
14. Stoker J, Halligan S, Bartram CI. Pelvic Floor Imaging. *Radiology* 2001;218:621-41
15. Malouf AJ, Williams AB, Halligan S, Bartram CI, Dhillon S, Kamm MA. Prospective assessment of accuracy of endoanal MR imaging and endosonography in patients with fecal incontinence. *Am J Roentgenol* 2000;175:741-5
16. Kamm MA. Faecal incontinence. *BMJ* 1998;316:528-32
17. Vaizey CJ, Kamm MA. Prospective assessment of the clinical value of anorectal investigations. *Digestion* 2000;61:207-14
18. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-80
19. Rao SS. Diagnosis and management of fecal incontinence. *Am J Gastroenterol* 2004;99:1585-604

Chapter 3

Anal inspection and digital rectal examination compared to anorectal function tests and endoanal ultrasonography in evaluating fecal incontinence

Annette C. Dobben
Maaike P. Terra
Marije Deutekom
Michael Gerhards
A. Bart Bijnen
Richelle J.F. Felt-Bersma
Lucas W.M. Janssen
Patrick M. M. Bossuyt
Jaap Stoker

Int J Colorect Dis; accepted

Abstract

BACKGROUND. Anal inspection and digital rectal examination are routinely performed in fecal incontinent patients but it is not clear to what extent they contribute to the diagnostic work-up. We examined if and how findings of anal inspection and rectal examination are associated with anorectal function tests and endoanal ultrasonography.

METHODS. A cohort of fecal incontinent patients (n=312, 90% females; mean age 59) prospectively underwent anal inspection and rectal examination. Findings were compared with results of anorectal function tests and endoanal ultrasonography.

RESULTS. Absent, decreased and normal resting and squeeze pressures at rectal examination correlated to some extent with mean (±SD) manometric findings: mean resting pressure 41.3 (±20), 43.8 (±20) and 61.6 (±23) Hg ($p<0.001$); incremental squeeze pressure 20.6 (±20), 38.4 (±31) and 62.4 (±34) Hg ($p<0.001$)). External anal sphincter defects at rectal examination were confirmed with endoanal ultrasonography for defects <90 degrees in 36% (37/103); for defects between 90-150 degrees in 61% (20/33); for defects between 150-270 degrees in 100% (6/6). Patients with anal scar tissue at anal inspection had lower incremental squeeze pressures ($p=0.04$); patients with a gaping anus had lower resting pressures ($p=0.013$) at anorectal manometry. All other findings were not related to any anorectal function test or endoanal ultrasonography.

CONCLUSIONS. Anal inspection and digital rectal examination can give accurate information about internal and external anal sphincter function but are inaccurate for determining external anal sphincter defects <90 degrees. Therefore, a sufficient diagnostic work-up should comprise at least rectal examination, anal inspection and endoanal ultrasonography.

Introduction

Fecal incontinence is a complex and challenging problem of diverse etiology[1]. It is relatively common and substantially impairs quality of life[2]. Its estimated prevalence in community-dwelling adults varies from two to 24 percent but the true prevalence of fecal incontinence is likely underestimated due to the shame and embarrassment that individuals experience and the failure of many affected individuals to disclose this condition to their physicians[3-7].

The diagnostic work-up for fecal incontinent patients in general comprises anal inspection, digital rectal examination, anorectal function tests and imaging[8-14]. Many anorectal function techniques are available and have contributed to understanding the pathophysiology of fecal incontinence [15, 16]. Digital rectal examination, as well as comprehensive anal inspection, forms the basis for making a diagnosis in fecal incontinent patients. If necessary, additive tests can be called for. Anorectal manometry is used commonly to quantify resting pressure (mainly internal anal sphincter), and squeeze pressure generated by the external anal sphincter (EAS) [14, 17]. Anal sensation measurement is less frequently used to determine the threshold of the sensitivity of the anal canal[18]. Endoanal ultrasonography is used to identify specific sphincter defects, particularly those amendable to surgical repair[17].

It is not clear to what extent digital rectal examination and anal inspection contribute to the diagnostic work-up in patients with fecal incontinence. Studies that compared digital rectal examination with endoanal ultrasonography showed that careful digital rectal examination detected some but not all of the EAS defects[19-21]. Studies that evaluated the ability of digital rectal examination to determine sphincter function have produced contradictory results[22-32]. It is uncertain how accurate particular findings are at digital rectal examination and to what extent observations at anal inspection have clinical implications.

In this study we wanted to investigate the usefulness and limitations of anal inspection and digital rectal examination compared to anorectal function tests and endoanal ultrasonography. We wanted to determine, in a large group of patients with fecal incontinence due to mixed etiology, the contribution of anal inspection and digital rectal examination in the diagnostic work-up. We therefore compared the results of anal inspection and digital rectal examination to the outcome of several anorectal function tests and endoanal ultrasonography in fecal incontinent patients.

Materials and Methods

Patients

This study was performed as part of a study evaluating pelvic floor rehabilitation in a large cohort of fecal incontinent patients, conducted between December 2001 and April 2005 in 15 medical centers in the Netherlands. Details of that study are reported elsewhere[33]. The study was approved by the medical ethics committee of all participating centers and all included patients had signed informed consent.

Inclusion criteria were the existence of fecal incontinence complaints for six months or more, a Vaizey incontinence score of at least 12[34], and failure of conservative treatment, based on dietary recommendations and/or antidiarrhetics. Excluded were patients with an age below 18, patients diagnosed less than two years ago with an anorectal tumor and patients with a previous ileoanal or coloanal anastomosis. As this study investigated the treatment effect of pelvic floor rehabilitation, patients with chronic diarrhoea (always fluid stools, three or more times a day), overflow incontinence, proctitis, soiling and rectal prolapse were also excluded from participation.

The collected patient information included patients' age, gender, severity and nature of fecal incontinence, and possible predisposing factors for fecal incontinence.

Anal inspection and digital rectal examination

Included patients underwent anal inspection and digital rectal examination. Anal inspection (including perianal sensation) and digital rectal examination were performed by one of the participating clinicians with the patient in the left lateral position with adequate illumination. Perianal sensation was assessed by stroking the perianal skin with a cotton bud in each of the perianal quadrants. Digital rectal examination was performed by inserting a lubricated, gloved index finger into the rectum to assess the presence or absence of an EAS defect and to determine resting and incremental squeeze pressure. Either resting pressure or incremental squeeze pressure were scored as 'normal', 'decreased' or 'absent' at the discretion of the investigator. In two centers the inspection and examination was done by residents. In the remaining centers the assessments were done by staff (colorectal) surgeons, gynecologists or a gastroenterologist.

Anorectal Function Tests

Anorectal function tests were performed by physicians or specialized technicians of all participating hospitals. The tests were performed with patients in left lateral position and flexed hips to 90 degrees.

Anorectal manometry took place according to the solid-state or water perfused technique without or with sleeve. The solid-state method or water-perfusion method without sleeve was performed by means of a pull-through technique. The catheter (Konigsberg Instrument Inc., Pasadena, CA; Medtronic, Skolvunde, Denmark; Dentsleeve Pty Ltd, Parkside, Australia) was inserted in the anal canal and the (mean) maximal resting pressure (mmHg) was measured. Subsequently, the (mean) maximal squeeze pressure (mmHg) was determined by asking patients to squeeze three times during 10 seconds with one-minute intervals. An average maximal squeeze pressure was calculated. Further, the difference (mmHg) between anal and rectal pressure during straining and coughing was assessed.

With anal sensation measurement the threshold sensation was determined. A ring electrode was positioned into the mid-anal canal. A connected stimulation electrode (Dantec Keypoint, Skovlunde, Denmark) mounted on a Foley urine catheter was applied and the current was increased gradually (up to a maximum of 20 mAmp), until patients reported some sensation. To determine the threshold, the lowest of three consecutive sensations was used.

Endoanal ultrasonography

Endoanal ultrasonography was performed to define anatomic defects of the EAS, with an ultrasound scanner with a radial endoscopic probe (7.5 or 10 MHz transducer)[14]. The

endoscopic probe was introduced into the anus to the anorectal verge and slowly withdrawn. A defect of the EAS was defined as a discontinuity of the muscle ring (anatomic defect) and/or characterized by loss of the normal architecture, with an area of amorphous texture that usually has low reflectiveness (functional defect, scar tissue)[35]. Localization and size of the defect were noted in degrees as well as longitudinally (proximal, distal or full length).

Statistical Considerations

We compared findings at anal inspection and digital rectal examination to the results of additional diagnostic tests: anorectal manometry, anal sensation measurement, and endoanal ultrasonography. We used analysis of variance to investigate differences in manometric measurements between patients with normal, decreased or absent resting and squeeze pressures at digital rectal examination. To test differences between the methods of anorectal manometry used and differences between staff clinicians and residents we performed stratified analysis of variance. For post-hoc analyses we used Bonferonni corrections.

To evaluate the discriminatory power of determining normal and abnormal resting and squeeze pressures, we calculated the area under respective receiver operating characteristic (ROC) curves, assuming a binormal distribution. The area under these curves was calculated. It can be interpreted as a measure of discrimination, i.e. as the probability that a random patient with an abnormal resting pressure, determined at rectal examination, will have a lower resting pressure at anorectal manometry compared to a patient with a normal resting pressure at digital rectal examination. Patients with decreased and absent pressures were classified as 'abnormal' resting or squeeze pressures.

To compare anal inspection and digital rectal examination with anal sensation, and endoanal ultrasonography we used the independent Student's t test or McNemar test statistics. To investigate differences between the size and diastases of EAS defects the Fisher's exact test and χ^2-test were used[36].

For all statistical tests p-values below 0.05 were considered to indicate statistical significance. We used SPSS for Windows (version 11.5, 2002) to perform statistical analysis of our data. All data were checked by double data-entry for validation.

Results

Between December 2001 and April 2005, 323 consenting eligible patients were included in this study. We could not use the data of 11 patients because they dropped out of the study for various reasons. Consequently, 312 cases remained for analysis, of which 280 were female (90%).

The median duration of fecal incontinence was 5 years (range 0.5 to 57). Clinical characteristics of these patients are shown in Table 1. Predisposing conditions for fecal incontinence are summarized in Table 2. Results from anal inspection, digital rectal examination, anorectal manometry, anal sensation measurement, and endoanal ultrasonography are shown in Table 3.

Anorectal manometry was performed with three different techniques, but there were no significant differences in results between techniques and findings from anal inspection or digital

Table 1. Clinical characteristics of patients

Baseline characteristics (n=312)	n (%)
Age	59 (± 13)*
Vaizey incontinence score	18 (± 3)*
Female	280 (90)
Passive incontinence	10 (3)
Urge incontinence	118 (40)
Combined passive and urge	170 (57)

Note: * denotes mean (± SD)

Table 2. Predisposing conditions

Anatomic	n (%)			Functional	n (%)
Congenital	1 (0.3)	**Colorectal surgery**		Neurological disorders	44 (14)
		Ileocoecal resection	4 (1)	Metabolic disorders	43 (14)
Obstetric injury		Colectomy	3 (1)	Inflammatory bowel diseases	9 (3)
Vaginal deliveries	260 (96)	Sigmoid resection	5 (2)		
median deliveries	2 (range 0-10)	Rectopexy	11 (4)	Systemic disorders	13 (4)
Breech delivery	24 (9)				
Long labor	78 (29)	**Anal surgery**			
High birth weight infant	83 (31)	Haemorroidectomy	28 (9)		
Forceps delivery	23 (9)	Sphincterotomy	4 (1)		
Vacuum pump delivery	29 (11)	Sphincter repair	22 (7)		
Episiotomy	151 (57)	Fistel operation	16 (5)		
Rupture	138 (52)	Lord procedure	7 (2)		
suture childbed	104 (39)	Remaining	16 (5)		
suture surgery	34 (13)				
		Urological surgery			
Gynecological surgery		Cystectomy (Bricker)	2 (0.6)		
Hysterectomy	114 (37)	Burch	29 (9)		
Tension-free vaginal tape	2 (0.6)	Remaining	32 (10)		
Sacropexy	6 (2)				
Remaining	28 (9)				

Note: one condition is not restricted to one patient; per patient various conditions could be present in the medical history.

rectal examination (data not shown). Neither were there differences in the results obtained by the residents versus those of the staff clinicians (data not shown).

Anal inspection in relation to anorectal function tests

There was no significant difference in the threshold for anal sensation between patients with and those without an intact sensibility (5.6 (±6) mAmp versus 4.6 (±6) mAmp, p= 0.289; t = 1.061; df = 293). Patients with anal scar tissue had significantly lower incremental squeeze pressures at

Table 3. Data from anal inspection, digital rectal examination, anorectal function tests and endoanal ultrasonography (n=312)

Anal inspection	**n (%)**
Anal scar	168 (57)
Dermatitis	36 (12)
Gaping anus	40 (14)
Keyhole deformity	17 (6)
Intact sensibility	248 (84)
Sphincter reflex	195 (66)
Presence of fecal matter	67 (23)
Digital rectal examination	
Resting pressure	
absent	11 (4)
decreased	197 (66)
normal	88 (30)
Squeeze pressure	
absent	63 (21)
decreased	198 (67)
normal	35 (12)
EAS defect	97 (33)
Anorectal function techniques	
Anorectal manometry	
Resting pressure (mmHg)	48.2 (± 22)*
Incremental squeeze pressure (mmHg)	38 (± 32)*
Anal sensation measurement	7.6 (± 5)*
Endoanal ultrasonography	
EAS defect < 90 degrees	103 (73)
EAS defect >90 < 150 degrees	33 (23)
EAS defect > 150 < 270 degrees	6 (4)

Note: * denotes mean (± SD); EAS defect = External Anal Sphincter defect. We could not retrieve complete information of all items for every patient.

anorectal manometry compared to patients without anal scar tissue (34 (±30) versus 43 (±36) mmHg, p= 0.039; t = -2.075; df = 235). Patients with a gaping anus, detected at anal inspection, had significantly lower resting pressures at anorectal manometry compared to patients without a gaping anus (40 (±23) versus 50 (±22) mmHg, p= 0.013; t = -2.496; df = 237).

We could not find any significant difference in anorectal function testing for patients with dermatitis, keyhole deformity, sphincter reflex or presence of fecal matter.

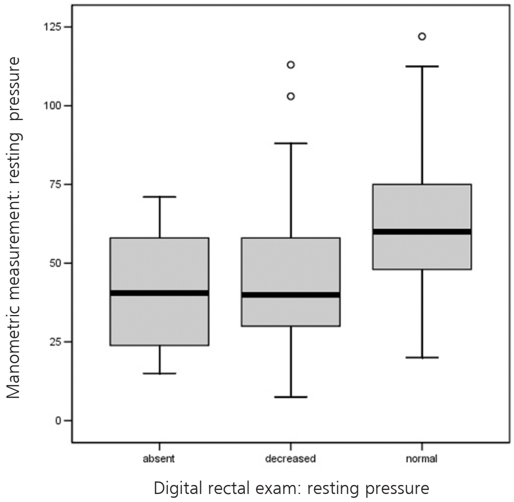


Figure 1a. Resting pressure at digital rectal examination in relation to manometric measurements

Note: Thick bar denotes the mean, the box denotes the interquartile range and the error bars denotes the total range. Individual points are outliers.
Exam = Examination

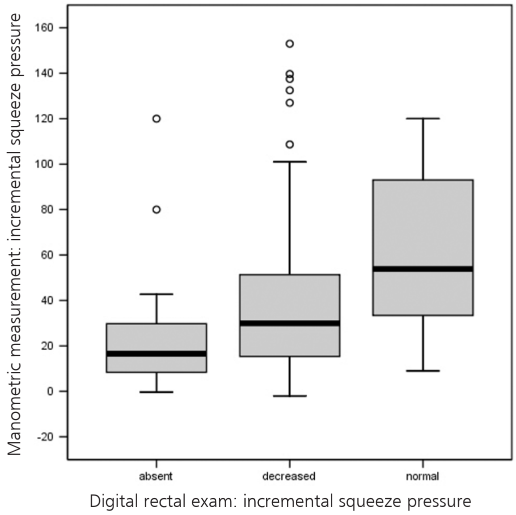


Figure 1b. Incremental squeeze pressure at digital rectal examination in relation to manometric measurements

Note: Thick bar denotes the mean, the box denotes the interquartile range and the error bars denotes the total range. Individual points are outliers.
Exam = Examination

Digital rectal examination in relation to anorectal function tests and endoanal ultrasonography

Absent, decreased and normal resting pressure as assessed at digital rectal examination corresponded to some extent with manometric findings. The mean resting pressure in these three groups was 41.3 (±20), 43.8 (±20) and 61.6 (±23) Hg, respectively ($p<0.001$; $F = 17.439$; df = 238). Post hoc testing showed a significant difference between normal versus decreased resting pressure ($p= 0.03$) and between normal versus absent resting pressure ($p< 0.001$;) (Figure 1a). Discrimination between patients with normal versus abnormal resting pressure was estimated fair (area under the ROC curve was 0.72 (95% confidence interval (CI) 0.64 to 0.79).

Absent, decreased and normal squeeze pressure as assessed at digital rectal examination corresponded to a certain extent with manometric findings. The mean incremental squeeze pressure in these three groups was 20.6 (±20), 38.4 (±31) and 62.4 (±34) Hg, respectively

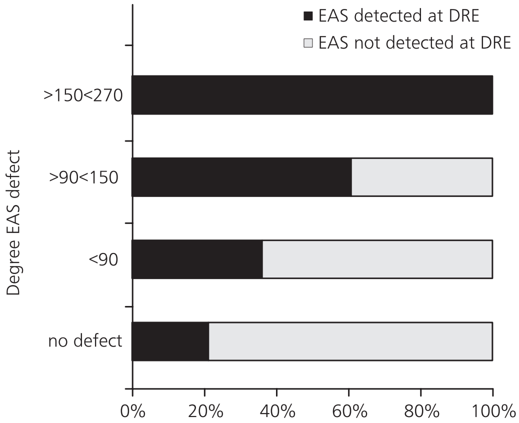


Figure 2. Degrees of EAS defect at endoanal ultrasonography in relation to digital rectal examination.

Note: EAS defect= external anal sphincter defect; DRE= digital rectal examination

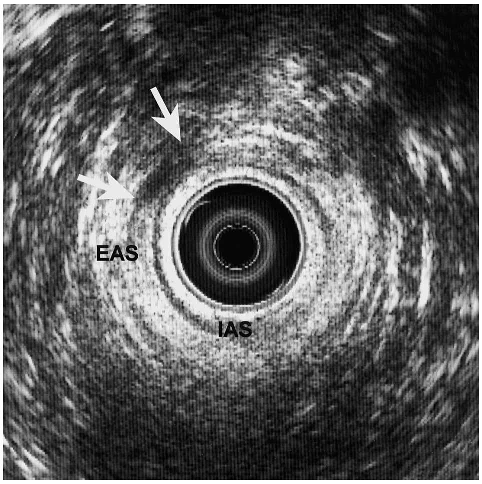


Figure 3. Endoanal ultrasonography of a 38 year old female with a complicated vaginal delivery in medical history. The image demonstrates disruption of the external anal sphincter muscles in the distal anal canal of 30 degrees (white arrows). The top of the figure is anterior. At digital rectal examination no EAS defect was detected.

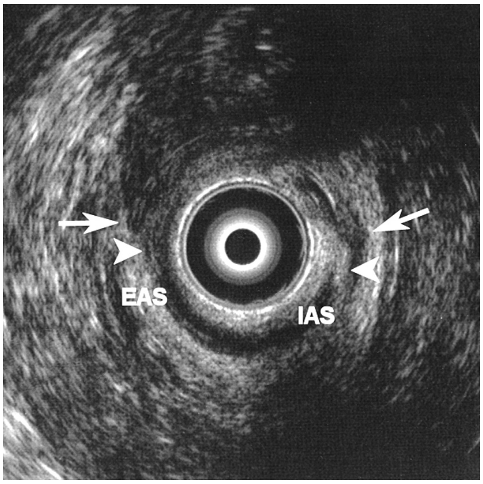


Figure 4. Endoanal ultrasonography of a 55 year-old female with a complicated vaginal delivery in the past and a large external defect at digital rectal examination. This finding was confirmed at endoanal ultrasonography, revealing disruption of the full length of the external anal sphincter muscles over 180 degrees (white arrows). Arrow heads show a large defect of the internal anal sphincter. The top of the figure is anterior.

($p<0.001$; F = 17.977; df = 238) (Figure 1b). Post hoc testing showed a significant difference in incremental squeeze pressure between all groups. Discrimination between patients with normal versus abnormal squeeze pressure was estimated fair (area under the ROC curve was 0.75 (95% CI 0.66 to 0.85).

EAS defects, as diagnosed at digital rectal examination, irrespective of the size, differed significantly from the findings at endoanal ultrasonography (Mc Nemar $p<0.001$). The sensitivity of digital rectal examination was 44% (95% C.I. 36% - 87%), with a specificity of 79% (95% C.I. 71% - 86%). The depicted EAS defects at endoanal ultrasonography were of different sizes. There were 103 EAS defects < 90 degrees, defined as small EAS defects; 33 EAS defects between 90 and 150 degrees, defined as moderate EAS defects and six EAS defects between 150 and 270 degrees, defined as large EAS defects. When we compared the different sizes of EAS defects depicted at endoanal ultrasonography to the findings from digital rectal examination, we found that the latter correctly identified small EAS defects in 37 patients (true positive rate 36%; false negative rate 64% (Figure 2 en 3); moderate EAS defects in 20 patients (true positive rate 61%; false negative rate 39% (Figure 2), and large EAS defects in all patients (Figure 2 and 4).

We could not retrieve data of the length of the EAS defect for all patients. In patients where disruption of the full length of the EAS muscles was depicted at endoanal ultrasonography, rectal digital examination detected small EAS defects in 40% (17/42); moderate EAS defects in 80% (12/15) and large EAS defects in 100% (5/5). Overall, it was easier to detect an EAS defect at digital rectal examination when the disruption was of full length compared to partial EAS defects ($p = 0.028$; $\chi^2 = 5.365$; df = 1). Stratified by the length of the EAS defect, there was no significant difference in diagnosing small EAS defects of partial or full length. For moderate EAS defects more defects of full length were diagnosed ($p = 0.043$; $\chi^2 = 5.105$; df = 1). Large EAS defects were scored correctly.

Discussion

The results of this study show that resting pressure and squeeze pressure, determined at digital rectal examination are to some extent related to anorectal manometry findings. EAS defects diagnosed at digital rectal examination differed significantly from the EAS defects depicted at endoanal ultrasonography. However, stratified by the size of the EAS defect, it was demonstrated that especially small defects are difficult to detect at digital rectal examination (false negative rate 64%). Patients in which anal scar tissue was observed at anal inspection had lower incremental squeeze pressures; patients in which a gaping anus was observed had lower resting pressures.

A number of potential limitations to this study should be addressed. The clinicians that performed anal inspection and digital rectal examination were unblinded to patients' medical history. Since clinicians were aware of risk factors with respect to anorectal lesions, the measurements might have been influenced by information bias.

Patients with anal abnormalities, with or without complaints, can have disturbed anal sensitivity[37]. The anal canal is richly innervated by receptors that can subserve sensations of touch, pain and temperature. Both the perianal skin and the anal canal are richly innervated by sensory nerves containing non-myelinated and myelinated fibers[38]. Impairment of perianal sensation suggest peripheral neuropathy or more central lesions[12]. We could not find a relation between intact sensibility at digital rectal examination and anal sensation threshold. In fact, anal sensation threshold for patients with absent sensibility at digital rectal examination did not differ from patients with intact sensibility. In our study these two measurements were not performed concurrently, but at different points in time. Either test gave qualitative information about anal sensation. Only assessment of qualitative information might have introduced bias in outcome.

Besides, anal sensation is maximal in the region of the anal valves, which might result in different awareness of sensation at either performed test[39].

Anal inspection and digital rectal examination contribute to making the right diagnosis in the following clinical circumstances. Diagnoses of value at anal inspection can be the detection of anal scar tissue and / or a gaping anus. However, both anal scar tissue and a gaping anus are observations that can not explicitly being validated or verified. As the results of this study show that both parameters indicate lower squeeze and / or resting pressures, the observation of anal scar tissue and / or a gaping anus might alert the clinician to identify external and / or internal anal sphincter weakness[25].

Previous studies[22, 23, 28] have reported that squeeze pressure is more correlated to digital rectal examination than resting pressure. Our study showed that both resting and squeeze pressures are to some extent related to anorectal manometry findings. Resting and squeeze pressure can be quantified at anorectal manometry to define functional sphincter weakness. However, either for resting as for squeeze pressure there is a substantial overlap in what can be considered as normal or abnormal values. This study showed a fair discrimination between normal and abnormal resting and squeeze pressures values. Normal anal canal pressures vary according to sex, age, and techniques used[14]. The wide variation in normal values makes comparisons between patients and healthy subjects difficult[1]. Therefore, the advantage anorectal manometry can offer is quantification, for example for considering surgery. Suitable candidates for surgery are patients with a major EAS defect; for patients with sufficient sphincter function, surgery is of less relevance[3, 42].

The assessment of EAS defects is not reliable at digital rectal examination since small defects (< 90 degrees) are hard to detect. The fact that there is no difference established in diagnosing either small defects of partial and full length, underscores the difficulty of accurate sensing. The difference in circumferential detection sensitivity makes comparison between digital rectal examination and endoanal ultrasonography difficult. EAS defects are more easily palpable at the anterior site in females but only palpable by complete tears in males. This fact contributes to differences in diagnosing EAS defects at digital rectal examination versus depiction of EAS defects at endoanal ultrasonography. To our knowledge, no other studies that compared digital rectal examination with endoanal ultrasonography[19, 22] made a differentiation between the sizes of the EAS defects. Unfortunately, we examined for defects > 90 degrees, the accuracy of the diagnosis increased. Therefore, we recommend when an EAS defect has been depicted to describe the size of the defect, since therapeutic consequences are related to the size. For detecting internal anal sphincter defects, digital rectal examination is not an appropriate tool, while endoanal ultrasonography is able to depict (unsuspected) internal anal sphincter defects[21, 22].

With this study we have demonstrated that internal and external anal sphincter weakness, observed during anal inspection or diagnosed at digital rectal examination, are well but not perfectly related to findings at anorectal manometry. Compared to endoanal ultrasonography, only major sphincter defects could be diagnosed accurately at digital rectal examination. Therefore, in daily clinical practice a sufficient diagnostic work-up to determine sphincter integrity and function should comprise anal inspection, digital rectal examination as well as endoanal ultrasonography. Additional physiological tests may be needed when conservative therapeutic measures fail and invasive treatment options are needed.

References

1. Osterberg A, Edebol Eeg-Olofsson K, Graf W. Results of surgical treatment for faecal incontinence. *Br J Surg* 2000;87:1546-1552
2. Bharucha AE, Fletcher JG, Harper CM, Hough D, Daube JR, Stevens C, Seide B, et al. Relationship between symptoms and disordered continence mechanisms in women with idiopathic faecal incontinence. *Gut* 2005;54:546-555
3. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *The Lancet* 2004;364:621-632
4. Mavrantonis C, Wexner SD. A clinical approach to fecal incontinence. [Review]. *Journal of Clinical Gastroenterology* 1998;27:108-121
5. Prather CM. Physiologic variables that predict the outcome of treatment for fecal incontinence. *Gastroenterology* 2004;126:S135-S140
6. Chiarioni G, Scattolini C, Bonfante F, Vantini I. Liquid stool incontinence with severe urgency: anorectal function and effective biofeedback treatment. *Gut* 1993;34:1576-1580
7. MacMillan A. Prevalence of fecal incontinence in community-dwelling adults: a systematic review of the literature'. 47 ed. *Dis Colon Rectum* 2004:1341-1349
8. Rudolph W, Galandiuk S. A practical guide to the diagnosis and management of fecal incontinence. *Mayo Clin Proc* 2002;77:271-275
9. Kouraklis G, Andromanakos N. Evaluating patients with anorectal incontinence. *Surg Today* 2004;34:304-312
10. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-1685
11. Bharucha AE. Outcome measures for fecal incontinence: anorectal structure and function. *Gastroenterology* 2004;126:S90-S98
12. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-1880
13. Rao SS. Diagnosis and management of fecal incontinence. American College of Gastroenterology Practice Parameters Committee. *Am J Gastroenterol* 2004;99:1585-1604
14. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-760
15. Rao SS, Patel RS. How useful are manometric tests of anorectal function in the management of defecation disorders? *Am J Gastroenterol* 1997;92:469-475
16. Liberman H, Faria J, Ternent CA, Blatchford GJ, Christensen MA, Thorson AG. A prospective evaluation of the value of anorectal physiology in the management of fecal incontinence. *Dis Colon Rectum* 2001;44:1567-1574
17. Salvioli B, Bharucha AE, Rath-Harvey D, Pemberton JH, Phillips SF. Rectal compliance, capacity, and rectoanal sensation in fecal incontinence. *Am J Gastroenterol* 2001;96:2158-2168
18. Dobben AC, Terra MP, Deutekom M, Bossuyt PM, Felt-Bersma RJ, Stoker J.Diagnostic work up of faecal incontinence in daily clinical practice in the Netherlands. *The Neth J Medicine* 2005;63:265-9
19. Nielsen MB, Hauge C, Rasmussen OO, Pedersen JF, Christiansen J. Anal endosonographic findings in the follow-up of primarily sutured sphincteric ruptures. *Br J Surg* 1992;79:104-106
20. Sultan AH, Kamm MA, Talbot IC, Nicholls RJ, Bartram CI. Anal endosonography for identifying external sphincter defects confirmed histologically. *Br J Surg* 1994;81:463-465

21. Romano G, Rotondano G, Esposito P, Pellecchia L, Novi A. External anal sphincter defects: Correlation between pre- operative anal endosonography and intraoperative findings. *British Journal of Radiology* 1996;69:6-9

22. Keating JP, Stewart PJ, Eyers AA, Warner D, Bokey EL. Are special investigations of value in the management of patients with fecal incontinence? *Dis Colon Rectum* 1997;40:896-901

23. Hallan RI, Marzouk DEMM, Waldron DJ, Womack NR, Williams NS. Comparison of digital and manometric assessment of anal- sphincter function. *Br J Surg* 1989;76:973-975

24. Eckardt VF, Kanzler G. How reliable is digital examination for the evaluation of anal sphincter tone? *Int J Colorectal Dis* 1993;8:95-97

25. Hill J, Corson RJ, Brandon H, Redford J, Faragher EB, Kiff ES. History and examination in the assessment of patients with idiopathic fecal incontinence. *Dis Colon Rectum* 1994;37:473-477

26. Wexner SD, Jorge JM. Colorectal physiological tests: use or abuse of technology? *Eur J Surg* 1994;160:167-174

27. Buch E, Alos R, Solana A, Roig JV, Fernandez C, Diaz F. Can digital examination substitute anorectal manometry for the evaluation of anal canal pressures? *Revista Espanola de Enfermedades Digestivas* 1998;90:90-93

28. Felt-Bersma RJ, Klinkenberg-Knol EC, Meuwissen SG. Investigation of anorectal function. *Br J Surg* 1988;75:53-55

29. Fink RL, Roberts LJ, Scott M. The role of manometry, electromyography and radiology in the assessment of faecal incontinence. *Aust N Z J Surg* 1992;62:951-958

30. Cook TA, Mortensen NJM. Management of faecal incontinence following obstetric injury. *Br J Surg* 1998;85:293-299

31. Herbst F, Teleky B. Alteration of maximum anal resting pressure by digital rectal examination prior to manometry: analysis of agreement between repeat measurements. *Int J Colorectal Dis* 1994;9:207-210

32. Kaushal JN, Goldner F. Validation of the digital rectal examination as an estimate of anal sphincter squeeze pressure. *Am J Gastroenterol* 1991;86:886-887

33. Terra MP, Dobben AC, Berghmans B, Deutekom M, Baeten CG, Janssen LW et al. Electrical stimulation and pelvic floor muscle training with biofeedback in patients with fecal incontinence: a cohort study of 281 patients**.** *Dis Colon Rectum* 2006;49:1149-1159

34. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80

35. Bartram CI, DeLancy J.O.L., (eds). Imaging pelvic floor disorders. Berlin Heidelberg New York: Springer-Verlag, 2003

36. Altman DG. Practical statistics for medical research. Boca Ration, Florida: CRC Press LLC, 1999

37. Felt-Bersma RJ, Poen AC, Cuesta MA, Meuwissen SG. Anal sensitivity test: what does it measure and do we need it? Cause or derivative of anorectal complaints. *Dis Colon Rectum* 1997;40:811-816

38. Rao SS. Manometric evaluation of defecation disorders: Part II. Fecal incontinence. *Gastroenterologist* 1997;5:99-111

39. Jorge JM, Wexner SD. Etiology and management of fecal incontinence. *Dis Colon Rectum* 1993;36:77-97

40. Azpiroz F, Enck P, Whitehead WE. Anorectal functional testing: review of collective experience. *Am J Gastroenterol* 2002;97:232-240

41. Felt-Bersma RJF, Klinkenberg-Knol EC, Meuwissen SGM. Anorectal function investigations in incontinent and continent patients - differences and discriminatory value. *Dis Colon Rectum* 1990;33:479-486

42. Felt-Bersma RJ, van Baren R, Koorevaar M, Strijers RL, Cuesta MA. Unsuspected sphincter defects shown by anal endosonography after anorectal surgery. A prospective study. *Dis Colon Rectum* 1995;38:249-253

Chapter 4

Prospective assessment of interobserver agreement for defecography in fecal incontinence

Annette C. Dobben
Tjeerd G. Wiersma
Lucas W. M. Janssen
Rien de Vos
Maaike P. Terra
Cor G.M.I. Baeten
Jaap Stoker

Am J Roentgenol. 2005 Nov;185(5):1166-72

Abstract

OBJECTIVE. The primary aim of our study was to determine the interobserver agreement of defecography in diagnosing enterocele, anterior rectocele, intussusception, and anismus in fecal incontinent patients. The subsidiary aim was to evaluate the influence of level of experience on interpreting defecography.

SUBJECTS AND METHODS. Defecography was performed in 105 consecutive fecal incontinent patients. Observers were classified by level of experience and their findings were compared with the findings of an expert radiologist. The quality of the expert radiologist's findings was evaluated by an intraobserver agreement procedure.

RESULTS. Intraobserver agreement was good to very good except for anismus: incomplete evacuation after 30 sec (κ0.55) and puborectalis impression (κ0.54). Interobserver agreement for enterocele and rectocele was good (κ0.66 for both) and for intussusception, fair (κ0.29). Interobserver agreement for anismus: incomplete evacuation after 30 sec was moderate (κ0.47), and for anismus: puborectalis impression was fair (κ0.24). Agreement in grading of enterocele and rectocele was good (κ0.64 and 0.72, respectively) and for intussusception, fair (κ0.39). Agreement separated by experience level was very good for rectocele (κ0.83) and grading of rectoceles (κ0.83) and moderate for intussusception (κ0.44) at the most experienced level. For enterocele and grading, experience level did not influence the reproducibility.

CONCLUSION. Reproducibility for enterocele, anterior rectocele, and severity grading is good, but for intussusception is fair to moderate. For anismus, the diagnosis of incomplete evacuation after 30 sec is more reproducible than puborectalis impression. The level of experience seems to play a role in diagnosing anterior rectocele and its grading and in diagnosing intussusception.

Introduction

Defecography (evacuation proctography) is used in the diagnostic work-up of patients with fecal incontinence, although the role of defecography in such a work-up has not been elucidated. Hinninghofen and Enck[1] report that because of the high radiation exposure, defecography can be recommended in the diagnostic work-up only if functional obstruction of the passage of stool cannot be excluded otherwise (e.g., by endoscopy or anorectal manometry). Other authors underscore the importance of the role of defecography for accurate diagnosing of intussusceptions and anterior rectoceles[2,3] in determining the cause of outlet obstruction symptoms in patients with combined fecal incontinence[4]. In a suggested work-up of fecal incontinent patients by Felt-Bersma and Cuesta[5], defecography is the only diagnostic procedure to detect an intussusception. To our knowledge, no data are available on the reproducibility of this technique in diagnosing enterocele, anterior rectocele, and intussusception, including their grading, or in diagnosing anismus in a large group of fecal incontinent patients. Reproducibility studies have been performed in healthy volunteers[6], in patients with disordered defecation[7,8], in patients with constipation[9], and in patients with a variety of clinical defecation disturbances[10–12]. In all these patient groups, abnormalities comparable to abnormalities in a fecal incontinence group were evaluated. However, the patient groups were small, or the differences in grading were not assessed. Moreover, the influence of experience in interpreting defecography on reproducibility has not been described. Furthermore, there might be differences in incidences of abnormalities in a fecal incontinent patient group compared with other patient groups[4]. Therefore, in this study we examine whether a high degree of consensus can be achieved with defecography, specifically for fecal incontinent patients with enterocele, anterior rectocele, intussusception, or anismus[13] (incomplete evacuation after 30 sec or nonrelaxing puborectalis muscle during evacuation) between experts and observers with different levels of experience. The primary aim of the study is to determine the interobserver agreement of defecography in diagnosing enterocele, anterior rectocele, intussusception, and anismus (incomplete evacuation after 30 sec or nonrelaxing puborectalis muscle during evacuation) in fecal incontinent patients. The secondary aim is to evaluate the influence of experience on interpreting defecography.

Subjects and Methods

In a large cohort study, defecography was performed in three academic medical centers and recorded on videotape (VHS) from January 2002 until November 2003. In this cohort study, we focused on the question of interobserver agreement in defecography results between experts and observers with different levels of experience, particularly with regard to agreement in diagnostic interpretation in fecal incontinent patients. The observers were radiologists (staff and resident radiologists) except for one who was a colorectal surgeon. With respect to validity, there is no reference standard to which defecography can be compared[7]. To examine the degree of consensus, observers were classified by level of experience and were compared with an expert radiologist with experience of more than 1,000 defecography assessments. The quality of the findings of the expert radiologist was evaluated by an intraobserver agreement procedure after a time interval of at least 8 weeks.

Patient Selection

Patients were those who were referred for their fecal incontinence to one of the three centers. In most patients, standardized conservative treatment—including dietary advice and antidiarrheals—had failed. Included were all men who were referred, and only women of at least 45 years, or women younger than 45 years after sterilization, who were fecally incontinent for more than 6 months with an incontinence score of 12 or greater according to the scoring system of Vaizey et al.[14]. The age restriction for women was applied because of the radiation dose. The radiation dose for defecography is approximately 2–3 mSv and therefore within World Health Organization limits for research purposes. Female patients younger than 45 years without sterilization were also excluded except on clinical indication (e.g., symptoms of prolapse) because of the radiation dose exposure. Excluded were patients younger than 18 years; patients diagnosed less than 2 years ago with an anorectal tumor; and patients with chronic diarrhea (always fluid stools, 3 times a day), overflow incontinence, proctitis, soiling, previous ilioanal or coloanal anastomosis; and patients with an anorectal prolapse. Although intussusception grade 3 (for definitions of grades, see under Evaluation Criteria) or enterocele grade 3 is only seen in anorectal prolapse and may lead to fecal incontinence[3], patients with these disorders were not entered in our study. These exclusion criteria were used because this study was part of a larger cohort study concerning the diagnostic work-up in patients with fecal incontinence in relation to the two most commonly used treatment techniques (pelvic floor rehabilitation and anterior anal repair). For example, incontinence caused by rectal prolapse requires a different surgical treatment[15].

The cohort study was approved by the medical ethics committee of the participating institutions, and all patients signed informed consent forms. No additional approval or informed consent was required for this study.

Study Design

Data from 105 consecutive patients were collected, with 35 patients per center. In only one medical center, in addition to videotape recordings, radiographs were obtained during rest, squeeze, straining, and evacuation. Clinical assessments were made on the basis of a standardized scoring form. The form included the following items: Was an enterocele present? If yes, enter grade 1, 2, or 3. Was an anterior rectocele present? If yes, enter grade 1, 2, or 3. Was an intussusception present? If yes, enter grade 1, 2, or 3. In anismus, Was evacuation complete after 30 sec? Was puborectalis muscle nonrelaxing during evacuation?

Other abnormalities such as cystocele were scored as well, but these concerned a variety of lesions with low prevalence. Therefore, these findings were disregarded in the analyses.

Evaluation Criteria

Enterocele was defined as a herniation of a peritoneal sac with extension of the small bowel along the ventral rectal wall and between the rectum and the vagina[16] (Fig. 1).

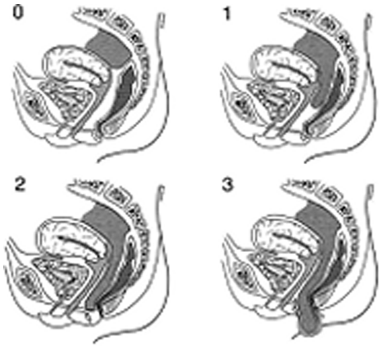


Figure 1. Grading system for enterocele. 0 = no enterocele; 1 = extension distal half of the vagina; 2 = reaching down to the perineum; 3 = protruding out of the anal canal. Patients with grade 3 enterocele were excluded from this study.
Reproduced with permission. Permission is granted by John Wiley & Sons Ltd on behalf of the BJSS Ltd.© British Journal of Surgery Society Ltd. Stoker et al: Imaging of anorectal disease, British Journal of Surgery 2000 Jan; 87(1):10-27. Review.

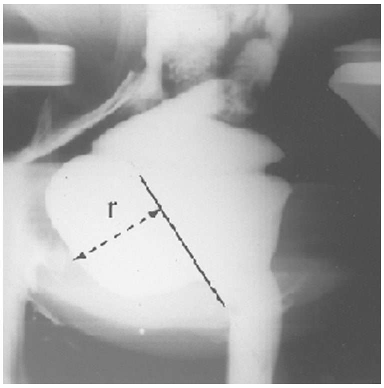


Figure 2. Grading system for anterior rectocele. 0 = no rectocele; 1 = rectocele < 2 cm; 2 = 2 cm ≤ rectocele < 4 cm; 3 = rectocele ≥ 4 cm.
Reprinted with permission from the American Journal of Roentgenology: AJR 2000 Jan;174(1):81-8.

Rectocele was defined as an outward bulge of the (usually anterior) rectal wall[16]. The presence of an anterior rectocele was measured perpendicularly to the extrapolated line of the rectal wall illustrated in Figure 2, using the maximum depth of the bulge[17] (Fig. 2).

Anterior rectoceles were scored as follows: grade 0 = no rectocele; grade 1 = rectocele smaller than 2 cm; grade 2 = 2 to less than 4 cm; grade 3 = rectocele 4 cm or greater.

Intussusception was defined as a circular invagination of the proximal rectal wall during defecation[16] (Fig. 3). Grade 1 is a minimal infolding of part of the rectal wall or circumferential infolding that remains entirely intrarectal. Grade 2 is scored when the leading edge of the intussusception is intraanal, into the orifice of the anal canal.

Anismus was defined as incomplete evacuation after 30 sec or a nonrelaxing puborectalis muscle during evacuation[13, 18] (Fig. 4). The rectum below the main rectal fold should empty in less than 30 sec; retention proximally was not significant. We defined complete obliteration of the puborectalis impression as total relaxation of the puborectalis muscle during attempted evacuation; incomplete obliteration was defined as paradoxical contraction of or inability to relax the puborectalis muscle during attempted evacuation, leading to poor rectal emptying.

Interobserver and Intraobserver Reliability

In the three academic medical centers, the first assessment of defecography was performed by 24 observers with different levels of experience. To evaluate if there was a difference in assessment by level of experience, all observers were classified into experience categories. Categories of assessment were 1–10 defecography examinations with supervision; 11–50 defecography

Figure 3. Grading system voor intussusception. 0 = no intussusception; 1 = intra rectal intussusception (a minimal infolding of part of the rectal wall or circumferential infolding which remains entirely intrarectal); 2 = intra anal intussusception (when the leading edge of the intussusception is intra-anal, into the orifice of the anal canal); 3 = extra anal intussusception (protruding out of the anal canal). Patients with rectal prolapse (grade 3 intussusception) were excluded from this study.
Reproduced with permission. Permission is granted by John Wiley & Sons Ltd on behalf of the BJSS Ltd.© British Journal of Surgery Society Ltd. Stoker et al: Imaging of anorectal disease, British Journal of Surgery 2000 Jan; 87(1):10-27. Review.

Figure 4. Anismus defined as incomplete evacuation after 30 seconds and / or non-relaxing musculus puborectalis during evacuation. The rectum below the main rectal fold should empty in less than 30 seconds; retention proximally was not significant. We defined complete obliteration of the puborectalis impression as total relaxation of the puborectalis muscle during attempted evacuation; incomplete obliteration was defined as paradoxal contraction of or inability to relax the puborectalis muscle during attempted evacuation leading to poor rectal emptying.
Reproduced with permission. Permission is granted by John Wiley & Sons Ltd on behalf of the BJSS Ltd.© British Journal of Surgery Society Ltd. Stoker et al: Imaging of anorectal disease, British Journal of Surgery 2000 Jan; 87(1):10-27. Review.

examinations with supervision; 11–50 defecography examinations without supervision; 51–100 defecography examinations with supervision; 51–100 defecography examinations without supervision; 101–250 defecography examinations with supervision; more than 250 defecography examinations; and more than 1,000 defecography examinations. Supervision was provided by radiologists with experience in at least 100 defecography examinations. In subsequent assessment by the expert radiologist, the videotapes were presented randomly. All collected defecography data were observed independently and blinded for results. To consider reproducibility, the second assessment of all data was made on the basis of the same standardized scoring form. Only clinical data considered relevant were provided: these included sex, date of birth, diagnosis of fecal incontinence, and previous operations. The second reviewer (i.e., the expert) was not involved in the primary interpretations. To determine the reliability of the interpretations of this observer, an intraobserver assessment was performed after at least 8 weeks of 30 patients (10 patients per center) who were representative of the disease spectrum of the interobserver study. The second reviewer was blinded to the results of the first assessment and to previous interpretations by others, and to the selection criteria of the defecography examination included in the intraobserver assessment study. To avoid recall bias and effects of ordering, the videotapes were randomly presented. All assessments were done under the same conditions: the same videotape recorder was used (type TVR 4510, Grundig) in the same room and during the same working hours.

Defecography Procedure

The standard procedure in all three participating centers was as follows: One to four hours before the examination, the patient was instructed to take an oral contrast medium (a barium sulfate suspension diluted in water [Micropaque, Guerbet; or Polibar, EZ-EM]). In two centers, a Microlax (sodium lauryl sulfoacetate–sodium citrate–sorbitol, Pharmacia) microenema was administered 20–120 min before the start of the procedure. The investigation started with the patient in the left decubital position. A thick barium paste (200–300 mL of barium sulfate prepared by the hospital pharmacy or Evacu-Paste [EZ-EM]) was placed in the rectum by means of an injection pistol. In only one center, Liquid Polibar (EZ-EM), combined with a mix of Metamucil (psyllium, Procter & Gamble), was used. For opacifying the vagina in women, 30 mL of amidotrizoic acid 50% gel solution and barium sulfate solution (produced by the hospital pharmacy or by EZ-EM) was introduced with a syringe, with a pediatric enema tip attached to it, into the vagina. The perineum was visualized by using a catheter. Subsequently, the entire imaging table was tilted upright 90° and the patient was seated on a radiolucent, especially developed defecography chair. The dynamics of defecation were recorded on videotape (type TVR 4510, Grundig). Videotape recordings (and radiographs) were made first, with the patient at rest, when the pelvic floor muscles were shown to be completely relaxed; next, during instructed straining, at maximum distention of the anal canal; and finally, during squeezing at the end of defecation, when the patient was asked to contract the pelvic floor muscles maximally. The procedure was terminated when rectal evacuation stopped or after several attempts of straining at defecation. The complete procedure took approximately 15 min (room time).

To evaluate whether diagnostic judgment could have been influenced by technical differences between hospitals, a scoring form was developed. The form included seven items: screen acquisition, whether analog or digital; presence of time registration; imaging quality; presence of recordings at rest, during straining, and during squeezing; use of vaginal tampon; use of contrast medium (product, application, volume); and use of catheter marked in centimeters for quantification of abnormalities, corrected for magnification.

Statistical Analysis

Intra- and interobserver agreement was quantified using Cohen's kappa statistic for nominal data (software program SPSS [version 11.5, Statistical Package for the Social Sciences] for Windows [Microsoft]). For ordinal data, a weighted kappa statistic was used (software program StatXact, version 3.0 for Windows, Cytel Software). We considered a kappa value of 0.8 satisfactory and used a cutoff value of 0.6 because a lower kappa value was not acceptable. To calculate the sample size to a kappa value of 0.8 (distance from kappa to limit 0.2) with a confidence interval of 95%, a one-tailed tested minimum of 25 patients per center was needed. To increase statistical power, we chose to include 35 patients per center. Kappa values range from 0 to 1, where 0–0.2 is considered poor, 0.2–0.4 is fair, 0.4–0.6 is moderate, 0.6–0.8 is good, and greater than 0.8 is very good agreement[19].

Results

Defecography results of 105 consecutive, consenting patients (15 men, 90 women) with an average age of 58.5 years (range, 24–78 years) were studied. The prevalence of abnormalities and the grading of enterocele, anterior rectocele, and intussusception as determined by the second reviewer (the expert radiologist) are shown in Table 1 (for interobserver assessment) and in Table 2 (for intraobserver assessment).

TABLE 1 Prevalence of abnormalities in the study population used for interobserver assessment (N = 105)

	Frequency N = 105	Percentage %
Enterocele	9	8.6
Grade 1	2	1.9
Grade 2	7	6.7
Grade 3	0	0
Anterior rectocele	56	53.3
Grade 1	34	32.4
Grade 2	13	12.4
Grade 3	9	8.6
Intussusception	11	10.5
Grade 1	2	1.9
Grade 2	8	7.6
Grade 3	1	1.0
Anismus: incomplete evacuation after 30 seconds	38	36.2
Anismus: non-relaxing puborectalis muscle during evacuation	20	19

Intraobserver Agreement

The intraobserver agreement, calculated to assess the quality of the findings of the expert radiologist, was very good for the assessment of enterocele (κ0.84; 95% confidence interval [CI], 0.51–1), good for anterior rectocele (κ0.73; CI, 0.48–0.98) and intussusception (κ0.71; CI, 0.41–1), and moderate for anismus: incomplete evacuation after 30 sec (κ0.55; CI, 0.27–0.83) and anismus: nonrelaxing puborectalis muscle during evacuation (κ0.54; CI, 0.16–0.91).

The intraobserver agreement for the grading of enterocele, anterior rectocele, and intussusception was very good (for grading of enterocele, weighted κ0.96; CI, 0.86–1) or good (for grading of anterior rectocele, weighted κ0.74; CI, 0.60–0.88; and for intussusception, κ0.74; CI, 0.47–1).

Level of Experience

To evaluate whether differences existed in assessment by level of experience, all radiologists were classified into experience categories. When analyzing the data, we reduced the experience

TABLE 2 Prevalence of abnormalities in the study population used for intra-observer assessment (N = 30)

	Frequency N = 30	Percentage %
Enterocele	3	10
Grade 1	0	0
Grade 2	3	10
Grade 3	0	0
Anterior rectocele	17	56.7
Grade 1	12	40
Grade 2	5	16.7
Grade 3	0	0
Intussusception	5	16.7
Grade 1	1	3.3
Grade 2	3	10
Grade 3	1	3.3
Anismus: incomplete evacuation after 30 seconds	13	43.3
Anismus: non-relaxing puborectalis muscle during evacuation	7	23.3

groups from seven to three categories to create comparable groups in size and in prevalence of abnormalities. Therefore, the presented analyses are derived from the following groups: little experience, *n* = 39 assessments (experience, < 100 assessments with supervision); moderate experience, *n* = 31 assessments (experience, < 1,000 assessments without supervision); and very experienced, *n* = 35 assessments (experience, > 1,000 assessments).

Interobserver Agreement

Interobserver agreement for enterocele was good (κ0.66; CI, 0.43–0.89). The results separated by level of experience show that experience did not play a role in diagnosing enteroceles. The interobserver agreement for anterior rectocele was also good (κ0.66; CI, 0.51–0.81). Only when the experience of the observer was more than 1,000 did the agreement become very good (κ0.83; CI, 0.65–1). For intussusception, the interobserver agreement was fair (κ0.29; CI, 0.11–0.46). Only when the experience rose over 1,000 did the agreement become moderate (κ0.44; CI, 0.16–0.72). Interobserver agreement for anismus: incomplete evacuation after 30 sec was moderate (κ0.47; CI, 0.30–0.64). No relationship was seen between the level of experience and the level of agreement. For nonrelaxing puborectalis muscle, the interobserver agreement was fair (κ0.24; CI, 0.01–0.47), as well as for the different levels of experience. Results for agreement in the grading of enterocele and anterior rectocele were good (weighted κ0.64; CI, 0.41–0.87; and weighted κ0.72; CI, 0.61–0.83, respectively) and for intussusception, fair (weighted κ0.39; CI, 0.16–0.61). The results separated by observers' level of experience for the difference in grading of anterior rectocele showed good agreement (weighted κ0.69; CI, 0.47–0.9) at the little-experience level versus very good agreement (weighted κ0.83; CI, 0.74–0.93) at the most experienced level. The results for level of experience for the differences in grading of enterocele and intussusception were not calculated because of too-low prevalences.

Technical Differences

Concerning the influence of technical differences in diagnostic interpretation between hospitals, the conclusions were as follows: In only one center, clock time was registered on the videotape images, facilitating reproducibility studies. To register whether evacuation was after 30 sec, the reproducibility in this center was moderate (κ0.54; CI, 0.26–0.82). Centers with no registration of time on the videotape images showed fair agreement (κ0.39; CI, 0.12–0.67; and κ0.35; CI, 0–0.72).

In one center, the use of vaginal contrast material was limited to a small volume. An important reason for vaginal opacification is to detect enteroceles[20]. The reproducibility for enteroceles in this center was moderate (κ0.44; CI, 0–0.89) versus good (κ0.72; CI, 0.36–1) and very good (κ0.84; CI, 0.54–1) in the other centers with an adequate volume of the vaginal contrast medium. To measure the size of a rectocele, a catheter marked in centimeters can be helpful.

In this study, in only one center the perineum was visualized using a catheter marked in centimeters. No statistical significance was found between the measurement of the severity of the rectocele in the centers without a magnification factor and the measurement in the one center with a magnification factor. The results of this study indicate that centimeter marking does not contribute to an increase in agreement.

Discussion

The results of this study show that the reproducibility of diagnosing enterocele and anterior rectocele and their grading in defecography studies in patients with fecal incontinence are good. Diagnosing intussusception and its severity seems far more difficult. Concerning anismus in fecal incontinent patients, the diagnosis "incomplete evacuation after 30 sec" is more reliable than the diagnosis of nonrelaxing puborectalis muscle during evacuation. In this study, the level of experience in diagnostic interpretation of defecography studies did not seem to play a role in diagnosing enterocele and anismus. The latter concerns both incomplete evacuation after 30 sec and nonrelaxing puborectalis muscle during evacuation. The level of experience seems to play a role in diagnosing anterior rectocele and its severity and in diagnosing intussusception. Although in this study the reproducibility of enterocele, anterior rectocele, intussusception, and anismus (incomplete evacuation after 30 sec or puborectalis impression during evacuation) was studied in a population of fecal incontinent patients, the results may be of interest for other patient groups. Concerning the influence of technical differences between hospitals in diagnostic interpretation, differences were found in the presence of time registration, the use of contrast medium, and the use of a catheter marked in centimeters, but all were statistically not significant.

Limitations

No reference standard is available for defecography. To calculate interobserver variability, we chose an expert radiologist with a high level of experience (> 1,000 interpretations). To increase reliability, we calculated stability in scoring using an intraobserver procedure. This study shows that the chosen expert radiologist is very reliable for diagnosing abnormalities such as

enterocele, anterior rectocele, and intussusception and their severity. However, for both aspects of anismus, the reliability of our expert radiologist appeared moderate. This study was a cohort study concerning the diagnostic work-up in patients with fecal incontinence in relation to pelvic floor rehabilitation and anterior anal repair. Patients with a rectal prolapse reported at medical history or found at physical examination were therefore excluded. Consequently, our patient population had a low prevalence of rectal prolapse. Only one rectal prolapse was not mentioned at medical history or detected at physical examination, and this is most likely related to the fact that a rectal prolapse may be transient and difficult to reproduce[4].

To our knowledge, no data are available about the reproducibility of diagnosing enteroceles and their corresponding grading or about diagnosing anismus on defecography. Few data are available about observer agreement for puborectalis impression, anterior rectoceles and their corresponding grading, and intussusception. Concerning the latter, the data give conflicting results[7, 8]. No reproducibility data of the differences in grading intussusception are available in the literature.

In our study, we did not specifically investigate a learning curve. By classifying the observers by their level of experience, we intended to study whether a little or a lot of experience may influence reproducibility.

Enterocele

In our study, the reproducibility of enterocele and its corresponding grading were good. The prevalence of enteroceles was low (8.6%) in our fecal incontinent study population. Not uncommonly, an enterocele accompanies a deep rectal intussusception or rectal prolapse[21]. Possibly, if we had not excluded rectal prolapse, the prevalence of enteroceles in our study population would have been higher.

Anterior Rectocele

Klauser et al.[7], in a group of 100 patients with defecation disorders, reported good total agreement, which is comparable to our data. However, no kappa values are available. Müller-Lissner et al.[8] reported observer agreement with kappa values of more than 0.4 in another group of 14 selected defecography studies from patients with disordered defecation. Exact interobserver agreements were not given. The prevalence of anterior rectoceles in our study group is comparable to that in other studies[8, 17]. Small rectoceles (2 cm) are not of clinical relevance[9, 18, 22]. A small rectocele may be found in 25–77% of asymptomatic control subjects[2]. Failure to recognize the correct grading of such abnormalities can easily lead to over- or underdiagnosis and inappropriate treatment.

Intussusception

Our data show, for both intussusception and its corresponding grade, fair agreement at a low prevalence (10.5%). The reasons for fair agreement are, among others, as follows. First, our reviewers had little experience in diagnosing intussusception. Second, examinations were interpreted incorrectly with respect to intussusception; a minimal infolding that disappears after the bolus has passed is probably caused by a transient prolapse of the rectal wall and should not be considered pathologic [4]. Third, an intussusception may appear at the end of the evacuation; when the examination is not completely videorecorded, it is difficult to clearly diagnose the presence or absence of an intussusception. And fourth, there is seldom doubt about complete extraanal prolapse (intussusception grade 3); however, lesser grades of prolapse are more difficult to diagnose, and prolapse of the anal canal cushions and variations of fold patterns caused by some degree of asymmetry of the rectum during its emptying, may confuse the interpretation [2].

When an intraanal intussusception occurs (grade 2), difficulty in diagnosis arises in the way the rectum collapses. The collapse is not uniform, and it is easy to make false assumptions from viewing its collapse in only one plane. Usually an intussusception appears when the proximal part of the rectum telescopes into the distal part or even further (extraanal). As a result, the proximal part tapers off. Suspicion of an S-form rectum occurs when the proximal part remains circular. In that case it may be possible that the proximal and distal rectum shifted parallel to each other. In the lateral view, the rectum is compressed by the pelvic contents as it empties and flattened out, so that the folds are not seen unless viewed in the anteroposterior plane[18]. In our study, two patients were suspected of having an S-form rectum as diagnosed by the second observer (expert radiologist). There was disagreement with the first interpretation in which both patients were scored with the presence of intussusception and no additional anteroposterior view was made. Therefore, this pitfall could not be confirmed on an anteroposterior view.

Anismus

Although the exact incidence of anismus is unknown, it is a finding in constipated and outlet-obstructed patients[23] and apparently represents an important etiologic factor[2]. However, Voderholzer et al.[24] showed that paradoxical sphincter contraction is a common finding in healthy control subjects and in patients with chronic constipation and fecal incontinence. Our data show prevalences of 36.2% for incomplete evacuation and 19% for puborectalis muscle impression. In our study, both intra- and interobserver agreements for anismus, incomplete evacuation after 30 sec, were moderate. With regard to incomplete evacuation[7] or incomplete rectal emptying [6, 8, 9] in populations of asymptomatic subjects, reports of disordered defecation and constipation in the literature confirm the variability of our findings.

Only Pfeifer et al. [9] showed inter- and intraobserver data of puborectalis impression. However, no kappa values are available and the agreement rates were low. Our data showed a fair interobserver agreement and a moderate intraobserver agreement for puborectalis impression. Emptying after 30 sec was shown to be the best indicator of anismus, with a positive predictive value of 90%[13, 18]. Therefore, we defined anismus in our study as incomplete evacuation after 30 sec or a nonrelaxing puborectalis muscle during evacuation.

We did not differentiate the primarily functional from the anatomic causes for incomplete evacuation [23, 25]. Neither did we distinguish other causes of delayed emptying. Consequently, data of incomplete evacuation by a nonrelaxing puborectalis muscle could not be distinguished from incomplete evacuation caused by a rectocele or by other factors (e.g., patient felt very uncomfortable, could not defecate because of lack of privacy, or was not instructed adequately). The fact that the observers were not able to distinguish this has led to a wider interpretation by observers of the idea of "anismus," probably with an overestimation of its prevalence. We assume that because of the absence of explicit diagnostic criteria, moderate intraobserver agreement and fair to moderate interobserver agreement were to be expected.

Conclusion

We conclude that the reproducibility for enterocele and anterior rectocele and their grading is good and can be valuable in the diagnostic work-up. With regard to intussusception, agreement is fair and becomes moderate only when the observer is very experienced. In the diagnostic work-up, an observer who is highly experienced in diagnosing intussusceptions is needed. For anismus, the diagnosis of incomplete evacuation after 30 sec is more reproducible than puborectalis impression. The level of experience seems to play a role in diagnosing anterior rectocele and its grading and in diagnosing intussusception.

References

1. Hinninghofen H, Enck P. Fecal incontinence: evaluation and treatment. *Gastroenterol Clin North Am* 2003;32:685-706
2. Jorge JM, Habr-Gama A, Wexner SD. Clinical applications and techniques of cinedefecography. *Am J Surg* 2001;182:93-101
3. Fuchsjäger MH, Maier AG. Imaging fecal incontinence. *Eur J Radiol* 2003;47:108-116
4. Wiersma TG, Mulder CJ, Reeders JW. Dynamic rectal examination: its significant clinical value. *Endoscopy* 1997;29:462-471
5. Felt-Bersma RJF, Cuesta MA. Fecal incontinence: which test and which treatment. *Neth J Med* 1994;44:182-188
6. Goei R, van Engelshoven J, Schouten H, Baeten C, Stassen C. Anorectal function: defecographic measurement in asymptomatic subjects. *Radiology* 1989;173:137-141
7. Klauser AG, Ting KH, Mangel E, Eibl-Eibesfeldt B, Müller-Lissner SA. Interobserver agreement in defecography. *Dis Colon Rectum* 1994;37:1310-1316
8. Müller-Lissner SA, Bartolo DC, Christiansen J, et al. Interobserver agreement in defecography: an international study. *Z Gastroenterol* 1998;36:273-279
9. Pfeifer J, Oliveira L, Park UC, Gonzalez A, Agachan F, Wexner SD. Are interpretations of video defecographies reliable and reproducible? *Int J Colorectal Dis* 1997;12:67-72
10. Choi JS, Wexner SD, Nam YS, et al. Intraobserver and interobserver measurements of the anorectal angle and perineal descent in defecography. *Dis Colon Rectum* 2000;43:1121-1126
11. Ferrante SL, Perry RE, Schreiman JS, Cheng SC, Frick MP. The reproducibility of measuring the anorectal angle in defecography. *Dis Colon Rectum* 1991;34:51-55
12. Yang X, Partanen K, Farin P, Ji H, Soimakallio S. Reproducibility of five anorectal morphologic measurements in defecography. *Acad Radiol* 1994;1:224-228
13. Halligan S, Malouf A, Bartram CI, Marshall M, Hollings N, Kamm MA. Predictive value of impaired evacuation at proctography in diagnosing anismus. *AJR* 2001;177:633-636
14. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
15. Silvis R, Gooszen HG, Kahraman T, et al. Novel approach to combined defaecation and micturition disorders with rectovaginovesicopexy. *Br J Surg* 1998;85:813-817
16. Stoker J, Rociu E, Wiersma TG, Lameris JS. Imaging of anorectal disease. *Br J Surg* 2000;87:10-27
17. Wiersma TG. *Dynamic rectal examination: clinicoradiological correlation* [dissertation]. Utrecht, The Netherlands: University of Utrecht, 1994
18. Bartram C. Dynamic evaluation of the anorectum. *Radiol Clin North Am* 2003; 41:425-441
19. Altman DG. *Practical statistics for medical research*. Boca Ration, FL: CRC Press LLC,1999
20. Halligan S. Commentary: imaging of anorectal function. *Br J Radiol* 1996;69:985-988
21. Kelvin FM, Maglinte DD. Dynamic evaluation of female pelvic organ prolapse by extended proctography. *Radiol Clin North Am* 2003;41:395-407
22. Delemarre JB, Kruyt RH, Doornbos J, et al. Anterior rectocele: assessment with radiographic defecography, dynamic magnetic resonance imaging, and physical examination. *Dis Colon Rectum* 1994;37:249-259
23. Halligan S, Bartram CI, Park HJ, Kamm MA. Proctographic features of anismus. *Radiology* 1995;197:679-682

24. Voderholzer WA, Neuhaus DA, Klauser AG, Tzavella K, Müller-Lissner SA, Schindlbeck NE. Paradoxical sphincter contraction is rarely indicative of anismus. *Gut* 1997;41:258-262

25. Janssen LW, Van Dijke CF. Selection criteria for anterior rectal wall repair in symptomatic rectocele and anterior rectal wall prolapse. *Dis Colon Rectum* 1994;37:1100-1107

26. Kelvin FM, Maglinte DDT, Hale DS, Benson JT. Female pelvic organ prolapse: a comparison of triphasic dynamic MR imaging and triphasic fluoroscopic cystocolpoproctography. *AJR* 2000;174:81-88

Endoluminal imaging in fecal incontinence

Chapter 5

Annette C. Dobben
Richelle J.F. Felt-Bersma
Fiebo J.W. ten Kate
Jaap Stoker

Submitted

Abstract

Fecal incontinence is a disabling disorder and can have a substantial impact on daily life. Physical examination, anorectal function tests and endoluminal imaging can be used to confirm the diagnosis and to clarify the anatomy and function of the anorectal region. The ability to image the sphincter muscles accurately has radically altered our understanding of the pathogenesis of fecal incontinence. Currently, the most well-known endoluminal techniques are endoanal ultrasonography and endoanal magnetic resonance (MR) imaging. For endoanal ultrasonography radial probes with a 360° view are preferable as they are not limiting the field of view. For endoanal MR imaging T2-weighted sequences are recommended as they result in optimal contrast difference.

The present consensus in diagnosing disorders of the external anal sphincter is that endoanal ultrasonography and endoanal MR imaging are equivalent. For internal anal sphincter disorders there is still no consensus about the preferred technique of choice. In the selection of patients for anterior anal sphincter repair, the advantage of endoanal MR imaging is the ability to measure sphincter thickness and atrophy. Atrophy of the external anal sphincter is a negative predictor for surgical outcome. In the post surgical work-up endoanal ultrasonography is useful to depict residual sphincter defects as a cause of failure of the repair.

In the past few years three-dimensional endoanal ultrasonography and external phased array MR imaging made their advent in the field of fecal incontinence with promising results. Imaging of the sphincter complex has increased our understanding resulting in a more adequate evaluation of fecal incontinence.

Introduction

Fecal incontinence is a complex and challenging problem of diverse etiology[1]. It is relatively common and substantially impairs quality of life[2]. International population-based studies have provided widely varying estimates of prevalence, ranging from 0.004% to 18%[3]. The true prevalence of fecal incontinence is likely underestimated due to the shame and embarrassment that individuals experience and the failure of many affected individuals to disclose this condition to their physicians.

Fecal incontinence can be classified into three specific types: urge incontinence, defined as the unwanted loss of stool despite active attempts to inhibit defecation; passive incontinence, defined as unwanted loss of stool without the patient awareness, or a combination of urge and passive incontinence[4]. Urge incontinence is mainly related to an external anal sphincter insufficiency and passive incontinence to an internal anal sphincter insufficiency. External anal sphincter insufficiency is caused by a defect of the muscle or by damage to the pudendal nerve, resulting in atrophy of the external anal sphincter with subsequent weakness. Childbirth is the main cause of fecal incontinence in women. Both the muscle itself as well as the pudendal nerve can be damaged during delivery.

Sometimes the internal anal sphincter can also be damaged in combination with the external anal sphincter when a large rupture of the anal sphincter occurs. Isolated internal anal sphincter defects are mainly caused by anorectal surgery (anal sphincter trauma)[5].

Imaging techniques with endoluminal devices allow a good visualization of the anal sphincter complex. At present the most well-known endoluminal techniques are conventional endoanal ultrasonography (US) and endoanal magnetic resonance imaging (MR imaging). In the past few years investigative imaging tools like three-dimensional endoanal US and external phased array MR imaging made their advent in the field of fecal incontinence with promising results.

Since endoluminal imaging has taken up a prominent place in the evaluation of fecal incontinence we would like to demonstrate the techniques of endanal US and endoanal MR imaging, the anatomy and pathology of anal sphincter muscles, the role of endoanal imaging in the work-up and treatment of fecal incontinence and discuss new developments like three-dimensional endoanal ultrasonography and the use of external phased array MR imaging.

Imaging techniques

Endoanal US protocol

Ultrasonography apparatus and probes

There are several types of ultrasonography probes that have been developed. Radial probes, suitable for the anal sphincter with a 360° view are developed as well as linear and curved array probes with a limited field.

A hard water filled cone over the transducer is necessary to image the anal canal. Degassed water should be used, to avoid air artifacts. Several industries provide ultrasonography machines. The rigid rotating endoprobes with a 360° view are preferable. Rigid mechanical probes are provided by Bruel & Kjaer Medical (Herlev, Denmark), with a focal range of 5-16 MHz and 360°

image, and Aloka (Tokyo, Japan) (7.5-12.5 MHz and 270° image). The flexible endoscopic Olympus radial scanner (Tokyo, Japan) (7.5-12 MHz) has a 360° image. Flexible endoscopic sector scanners are from Pentax / Hitachi (Tokyo, Japan) (5 and 7.5 MHz and 100° image) and Olympus (Tokyo, Japan) (7.5 MHz and 180° image).

Bruel & Kjaer Medical has also developed software to construct three-dimensional images.

Performance conventional endoanal US

Patients can be in the left lateral position, supine or prone. The first is more comfortable for the patient. Slight deformation can occur in the lateral position concerning left and right differences according to gravity, but that is only of importance when thickness is measured. A digital rectal examination is mandatory to be informed about possible abnormalities (stenosis, painfull lesion, tumor). The rigid probe is covered for hygienic reasons with a condom filled with ultrasonography gel to ensure a good acoustic contact, followed by covering the outside of the probe with an anaesthetic gel. Then, the probe is gently introduced into the rectum, following the anorectal angle. Landmarks are the prostate, the vagina and the puborectal muscle. Finally, the probe is slowly withdrawn until all levels, perpendicularly to the anal canal, are scanned. Images are obtained at proximal, middle and distal levels.

Three-dimensional endoanal US

Three-dimensional endoanal ultrasonography is performed in the same way as the conventional ultrasonography concerning preparation and introduction. The difference is that a puller with a pre-set speed pulls the rotating probe (external puller) or the ultrasonography head (internal puller) slowly caudally, so that at pre-set times an image is made throughout the anal canal. At present a three-dimensional endoanal ultrasonography is provided by Bruel & Kjaer Medical. We perform endoanal ultrasonography using a three-dimensional diagnostic ultrasonography system (Falcon 2101, Bruel & Kjaer Medical, Herlev, Denmark) with a 5-16 MHz rotating endoscopic probe (type 2050). This probe has an internal puller, within filled with fluid. The external diameter is 1.7 cm.

After performance, the images need to be reconstructed to three-dimensional images by computer software (Life Imaging system 2000, L3Di, version 3.5.5, Bruel & Kjaer Medical), resulting in the possibility to view the images from every possible angle. In addition, to obtain length and subsequently volume measurements. Other software packages for other systems also exist (3D echotech, GE medical systems, Milwaukee WI, USA; Vaytek Inc., Fairfiled, Iowa, USA).

MR imaging protocol

Endolumired coils

There are some differences in design and diameter of the endoluminal coils used for endoluminal imaging. In some institutions a rectal coil with balloon is used, but more optimal results are obtained with a dedicated anal coil without balloon. Cylindrical saddle geometry receiver coil, rectangular receive-only coil as well as phased array geometry coil are used, where our experience concerns the latter two coils [6]. With these coils high spatial resolution MR images can be obtained. The diameter of the coils available ranges from 7-17 mm, while the coil holder will add approximately 2 mm to the outer diameter. A larger diameter coil (e.g. 17 mm) can be recommended as this will result in a more uniform signal intensity of the anal sphincter muscles, while the diameter will not be a disadvantage in fecal incontinent patient. The length of the coil we use is 8 cm, while the coil holder length is 10 cm.

Performance

The endoanal coil is covered with a condom and after application of a lubricant (we use ultrasonography gel) inserted in the anal canal in a left lateral position. After positioning of the endoanal coil the patient turns in supine position and supportive pads are used. To reduce motion artifacts the patient is asked to fast for four hours and attention should be paid to patient comfort during the examination. We use bowel relaxants (1 ml butylscopolamine bromide, Buscopan, 20 mg/ml, Boehringer Ingelheim, Germany – which is not approved in the USA – or 1 mg of glucagon hydrochloride, Glucagen, Bagsvaerd, Denmark) to reduce bowel peristalsis. A bowel relaxant can be beneficial, but it is not scientifically proven.

The patient should be informed not the squeeze the coil and to relax their anal sphincter and pelvic floor muscles.

Endoanal MR imaging

The optimal imaging protocol for endoluminal MR imaging in fecal incontinent patients is not established. Based on our experience in over 1000 patients, we use only two T2-weighted turbo spin-echo sequences. T2-weighted sequences result in optimal contrast difference between the anal sphincters and the surrounding structures when relative limited T2-weighting is used. The use of T1-weighted sequences without intravenous contrast medium is not beneficial (sphincters are hardly to discern), while the use of intravenous contrast medium has not been demonstrated to be superior to T2-weighted sequences. The following T2 weighted fast spin echo sequences are recommended for use at 1.5T: TR 2500 - 3500 ms, TE 70 - 90 ms, echo train length 10, field of view 10 x 10 cm (axial) and 16 x 16 cm (coronal), imaging matrix 256 x 512, 3 mm slice thickness, 0.3 mm interslice gap and 2 excitations. Axial images and coronal images are used with slice orientation perpendicular and parallel to the anal sphincter and endoanal coil to reduce partial volume effects. The field of view encloses the whole anal sphincter.

External Phased array MR imaging

Standard external phased array coils can be used, with a scan protocol comparable to the endoanal protocol, except for adjustments to increase local signal-to-noise by increasing field of view, slice thickness and / or number of excitations. As in endoluminal imaging axial and coronal sequences are orientated perpendicular and parallel to the anal sphincter.

Anatomy anal sphincter

The anal sphincter surrounds the anal canal and is composed of several cylindric layers[7]. The innermost layer of the anal sphincter is the subepithelium that seals off the anal canal (anal cushions). The next layer is the cylindrical smooth muscle of the internal anal sphincter, supplied by autonomic nerves. The internal anal sphincter is approximately 2.9 mm thick on endoluminal imaging[8]. On MR imaging with T2 weighted sequences in the axial plane the internal anal sphincter appears as a relatively hyperintense circular structure with a homogeneous uniform architecture (Fig 1).

The internal anal sphincter is the terminal continuation of the circular smooth muscle of the rectum and often separated from the longitudinal muscle. The latter is a fibroelastic structure coursing through the fat containing intersphincteric space and forming a network. The longitudinal muscle layer is seen as a relatively hypointense layer within the hyperintense intersphincteric space (Fig 2).

The outermost layer comprises striated muscle with inferiorly the external anal sphincter and superiorly the puborectal muscle. The external anal sphincter is a muscle under voluntary control. The height of the external anal sphincter anteriorly is approximately 14 mm in women and 27 mm in men[8]. The thickness of the external anal sphincter is 4 mm on endoanal MR imaging. The external anal sphincter extends approximately 1 cm beyond the internal anal sphincter. The external anal sphincter is demonstrated as a clearly defined ring of hypointense signal intensity (Fig 3).

The puborectal muscle (also named pubovisceral muscle) is part of the levator ani muscle which also includes the levator plate at the anorectal junction. The puborectal muscle is a sling-like muscle and closely aligned to the deep part of the external anal sphincter. On endoanal MR imaging the puborectal muscle is approximately 28 mm high and 5.6 mm thick. Also the puborectalis muscle and the levator ani muscle have a relative hypointense signal intensity[9] (Fig 4). The latter can be easily evaluated in the coronal plane (Fig 5).

The sphincter complex is embedded in the fat containing ischioanal space, which is relative hyperintense (Fig 4).

On conventional endoanal US the internal anal sphincter appears as a well-defined, low reflective ring, usually symmetric in thickness (Fig 6)[10]. The longitudinal muscle in the upper anal canal can be seen as low reflective bundles and in the lower canal as more reflective bundles. The puborectal muscle is identified as a medium reflective (mixed echogenic) sling-like structure. The external anal sphincter muscle is closely related to the puborectal muscle dorsally, appears as a ring-like structure of the same mixed echogenicity (Fig 6).

Pathology anal sphincter complex

Anal sphincter defects and scar tissue

Endoanal MR imaging and endoanal US

A defect of the anal sphincter at endoluminal imaging commonly is defined as a discontinuity of the muscle ring (anatomic defect) and / or is recognized by a hypo-intense deformation (endoanal MR imaging) or hypo-reflectivity (endoanal US) of the normal pattern of the muscle layer due to replacement of muscle cells by fibrous tissue (functional defect, scar tissue)[10, 11]. With endoluminal imaging, predominantly endoanal MR imaging, it is possible to distinguish an anal sphincter defect (Fig 7) from scarring (Fig 8).

Since distinction between either pathologic conditions has not been demonstrated to be of clinical relevance yet, in our institutions both conditions are classified in one definition.

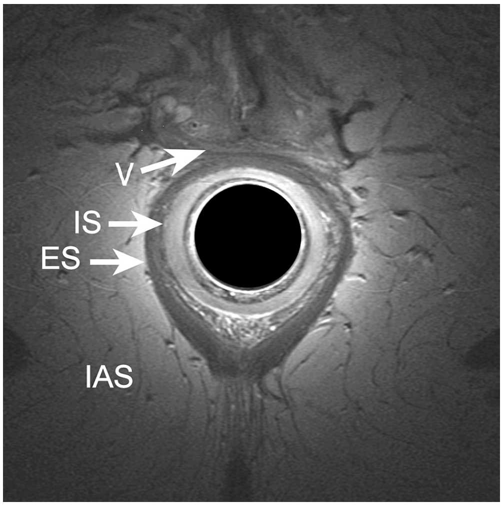


Figure 1. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the mid-anal canal shows normal anatomy of a 35-years-old-female. The external anal sphincter (ES) is the outer part of the sphincter ring at this level and relative hypointense; the inner part of the sphincter ring comprises the internal anal sphincter (IS) and is shown as relative hyperintense.
IAS = ischioanal space, V = vaginal introitus with bulbospongiose muscle

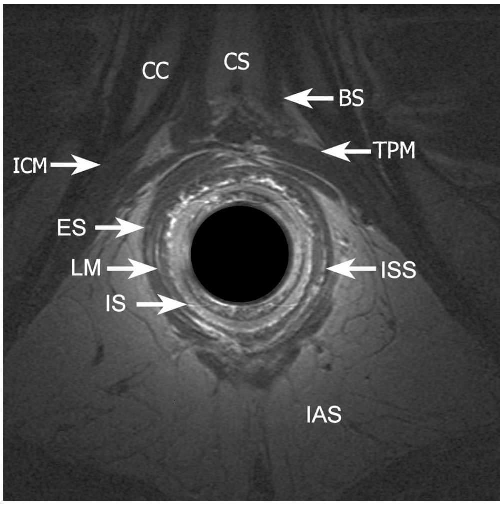


Figure 2. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the mid-anal canal shows normal anatomy in a 52-years-old man. The longitudinal muscle (LM) is clearly demonstrated within the intersphincteric space (ISS) between the internal (IS) and external (ES) anal sphincters. IAS = ischioanal space, CC = corpus cavernosum, CS = corpus spongiosum, ICM = ischiocavernosum muscle, TPM = transverse perineal muscle, BS = bulbospongiosus

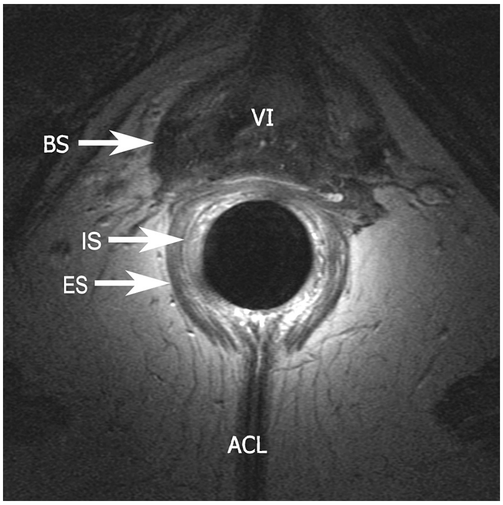


Figure 3. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the distal anal canal in a 57-years-old woman shows normal anatomy of the external anal sphincter (ES) which is relative hypointense. IS = lower edge internal anal sphincter, VI = vaginal introitus, BS = bulbospongiosus, ACL = anococcygeal ligament

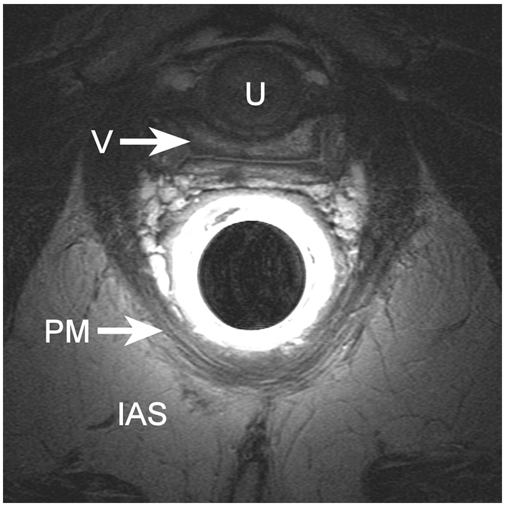


Figure 4. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the proximal anal canal in a 68-years-old woman shows a normal puborectal muscle (PM). The puborectal muscle is a relative hypointense, sling-like muscle. IAS = ischioanal space, V = vagina, U = urethra

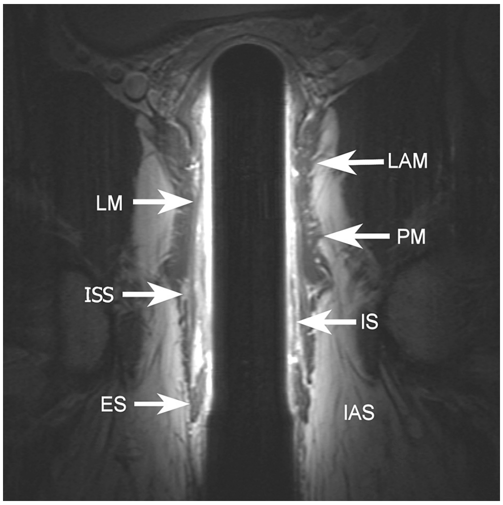


Figure 5. Coronal endoanal T2- weighted fast spin-echo (2500/70) MR image through the anal canal shows normal anatomy of the sphincter complex in a 52-years-old man with relative hypointense external anal sphincter (ES) and relative hyperintense internal anal sphincter (IS). IAS = ischioanal space, LM = longitudinal muscle, ISS = inter sphincteric space, PM = puborectal muscle, LAM = levator ani muscle

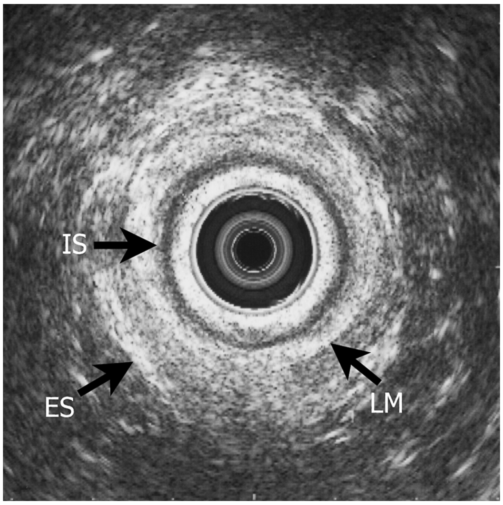


Figure 6. Transverse endoanal ultrasonography at the mid-anal canal obtained from a 65-years-old man demonstrating normal anatomy at the mid-anal canal of the internal anal sphincter (IS), longitudinal muscle (LM), and external anal sphincter (ES). The top of the figure is anterior.

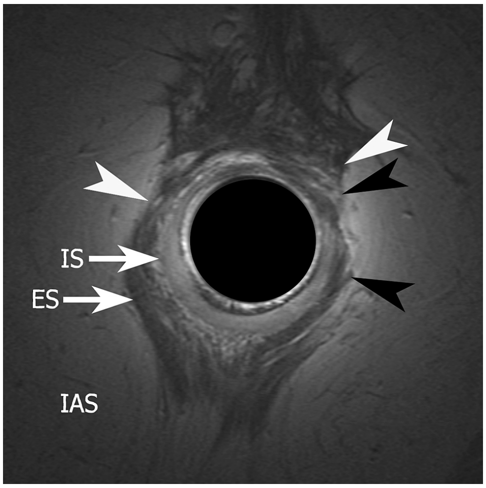


Figure 7. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from a 53-years-old fecal incontinent woman after a complicated vaginal delivery (breech delivery and episiotomy) shows a defect of the external anal sphincter (ES) which is demonstrated by a discontinuity of the anterior outer sphincter ring (black arrowheads) and scar tissue (white arrowheads). IS = internal anal sphincter, IAS = ischioanal space

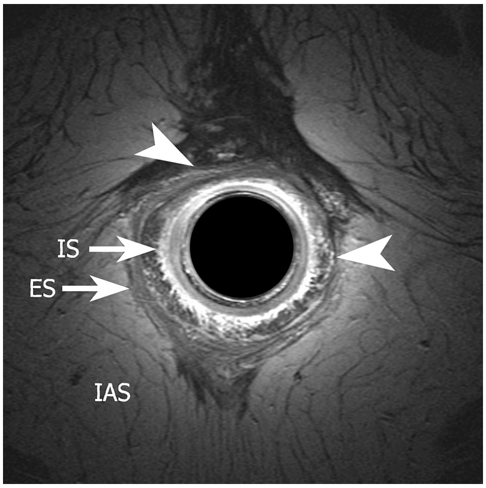


Figure 8. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from a 53-years-old fecal incontinent woman after a complicated vaginal delivery (long labor, assisted delivery, rupture) shows scar tissue of the external anal sphincter (ES) (arrowheads), which is demonstrated by a hypointense anterior outer sphincter ring which lacks the normal ES architecture (see posteriorly and see Figs. 1 and 2). IS = internal anal sphincter, IAS = ischioanal space

The comparison between endoanal US and endoanal MR imaging in the depiction of sphincter lesions has been described in the literature. Malouf and colleagues[12] concluded in a prospective study of 52 patients with an expert panel as reference standard, that endoanal US and endoanal MR imaging are equivalent in diagnosing external anal sphincter injury, but MR imaging is inferior in diagnosing internal anal sphincter injury.

A retrospective study study performed by Rociu et al[13] in 22 patients with surgery as reference standard emphasizes the precise description of the extent and structure of complex sphincter lesions and superiority in clinical decision-making by endoanal MR imaging compared to endoanal US. This, as MR imaging provides higher spatial resolution and better inherent image contrast for lesion characteristics. A recent study in 36 patients[14] showed that both imaging techniques can be considered equal concerning the depiction of external anal sphincter defects and therefore either technique is useful in the selection of patients as candidates for surgery. Resuming, the present consensus in diagnosing disorders of the external anal sphincter is that endoanal US and endoanal MR imaging are equivalent[12] (Fig 9[a,b],10 [a,b]). The advantages of endoanal US are its availability, limited costs and more widely available experience[15]. In contrast, endoanal MR imaging may allow for a clear visualization of the external anal sphincter as there is large contrast difference between the external anal sphincter muscle and the surrounding fat. For postoperative assessment endoanal US can reveal residual defects that are relevant in terms of fecal incontinence. Vaizey and Bartram[16] showed that defects depicted on endoanal US were associated with a poor clinical outcome after anterior anal sphincter repair. At present, the association of post surgical scarring with clinical outcome has not been thoroughly investigated (Fig 9[c]). The role of endoanal MR

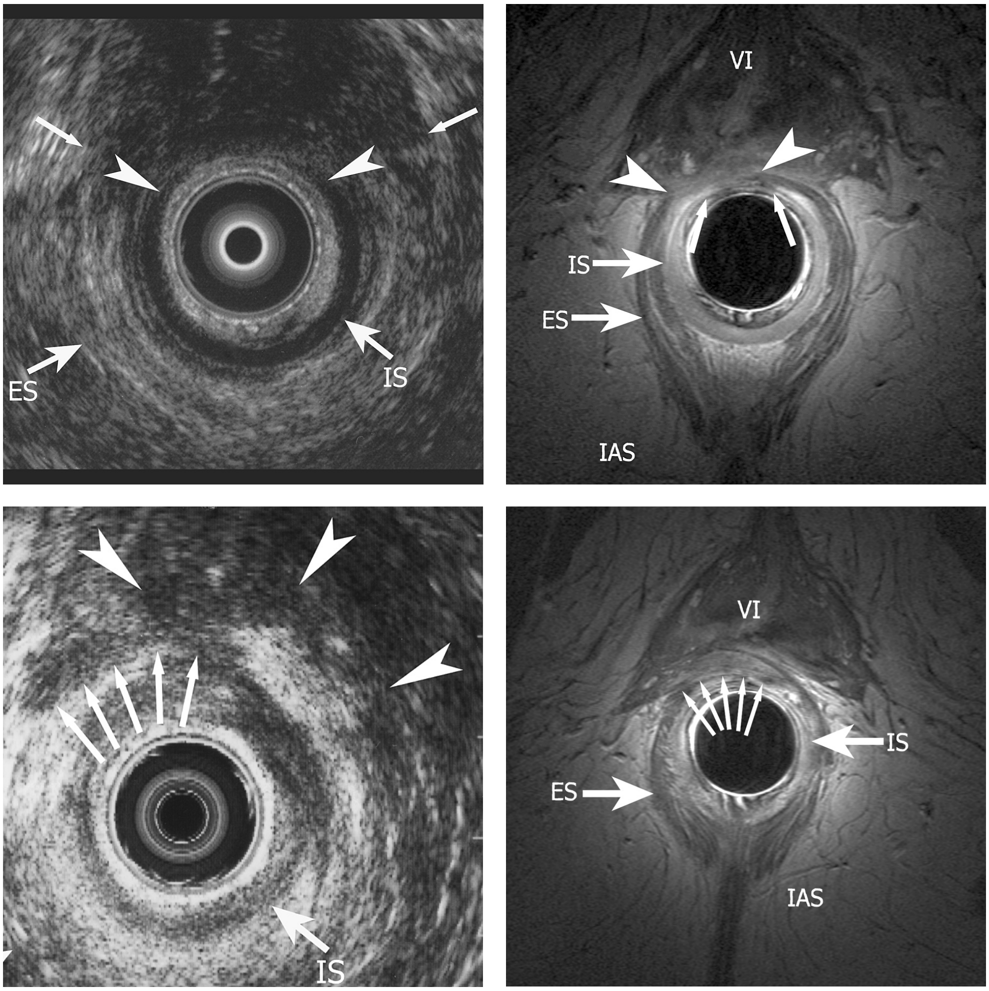


Figure 9[a], 9[b], 9[c], 9[d]. (**9[a]**) Transverse endoanal ultrasonography at the mid-anal canal obtained from a 53-years-old woman after a complicated vaginal delivery (rupture) demonstrating an external (area of amorphous texture; thin arrows) and internal (discontinuity of the sphincter ring; arrow heads) anal sphincter defect from 10 – 2 o'clock. The top of the figure is anterior. (**9[b]**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from the same patient showing a defect of the anterior external anal sphincter (ES) (arrowheads) and the internal anal sphincter (thin arrows) from 11 – 1 o'clock by discontinuity of the outer and inner sphincter ring. The high intrinsic contrast resolution makes delineation of the external anal sphincter boundaries clearly visible. Due to severe fecal incontinence complaints, this patient underwent surgery. (**9[c]**) Transverse endoanal US obtained from the same patient after anterior anal sphincter repair demonstrating a sphincter overlap (thin arrows) at the mid-anal canal. Overlap is difficult to visualize owing to the low soft-tissue contrast resolution. An area with scar tissue (arrow heads) is depicted. The top of the figure is anterior. (**9[d]**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from the same patient after anterior anal sphincter repair showing a clear overlap of both external anal sphincter ends, left over right, (thin arrows). Continuity of the sphincter ring has been restored. Although appearances after anterior anal sphincter repair at endoluminal imaging show overlap of both sphincter ends, surgery failed for this patient as the patient was still fecal incontinent. IS = internal anal sphincter, IAS = ischioanal space, VI= vaginal introitus

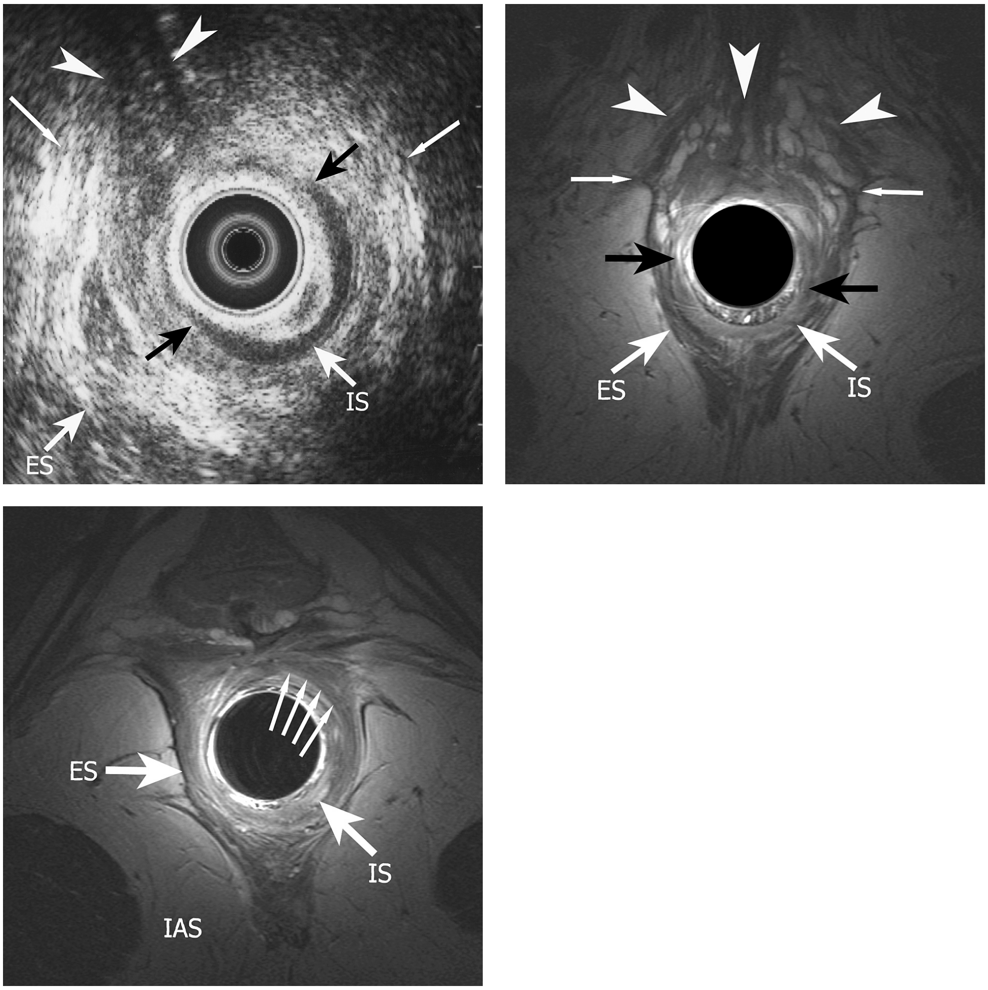


Figure 10^a, 10^b, 10^c. (**10^a**) Transverse endoanal ultrasonography at the mid-anal canal obtained from a 31-years-old woman after a complicated vaginal delivery (breech delivery, rupture) showing an external anal sphincter defect (thin arrows) and scar tissue (arrowheads). The top of the figure is anterior. An internal anal sphincter (IS) defect is depicted from 8 – 2 o' clock (black arrows). IAS = ischioanal space (**10^b**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from the same patient before surgery shows a defect (thin arrows) and scar tissue (arrowheads) anterior to the external anal sphincter (ES) demonstrated by discontinuity of the sphincter ring, very low signal intensity and disordered architecture. Also an anterior internal anal sphincter (IS) defect is depicted (black arrows), identifiable by the discontinuity of the anterior part of the internal sphincter. Patient underwent anterior anal sphincter repair since she suffered from severe fecal incontinence. (**10^c**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained after surgery from the same patient. Overlap of the anterior anal sphincter (thin arrows) is depicted. Much less disordered architecture is demonstrated (compare to Fig 10^b). The patient improved substantially.

imaging in the evaluation of sphincter repair is not studied yet (Fig 9^d, 10^c). Although internal anal sphincter disorders are adequate depicted at either endoanal US and endoanal MR imaging, there is still no consensus about the preferred technique of choice[12, 15].

Puborectal muscle lesions (Fig 11) are relatively uncommon in fecal incontinent patients and are generally seen in patients with major pelvic trauma. Defects of the puborectal muscle are primarily depicted in combination with internal and or external anal sphincter lesions.

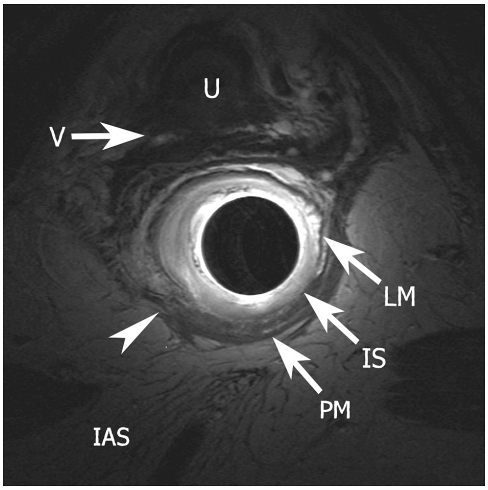


Figure 11. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image obtained from a 66-years-old fecal incontinent woman after a complicated vaginal delivery (high birth weight, long labor, episiotomy) showing a defect of the puborectal muscle (PM) from 8 – 11 o' clock (arrow head; compare to normal left side). IAS = ischioanal space, IS = internal anal sphincter, LM = longitudinal muscle, V = vagina, U = urethra

Anal sphincter atrophy

Endoanal MR imaging and comparison with histology

Atrophy of the external anal sphincter, puborectal muscle or levator ani muscle at endoanal MR imaging is characterized by severe thinning of the muscle fibers and / or hyperintense sphincter tissue. The latter represent the replacement of muscle fibers by fat[11]. Atrophy of the internal anal sphincter is characterized by diffuse muscle thinning (muscle thickness less than 2 mm)[11]. The coronal imaging plane is optimal for the evaluation of atrophy of the external anal sphincter (Fig 12b, 13b); atrophy of the internal anal sphincter is most easily appreciated at an axial image. Endoanal MR imaging is able to accurate depict atrophy of the anal sphincter (Fig 12[a,b], Fig 13[a,b]) and can differentiate between moderate (< 50 % thinning of the external anal sphincter and / or replacement of external anal sphincter muscle by fat) and severe atrophy (≥ 50% thinning of the external anal sphincter and / or replacement of the external anal sphincter muscle by fat)[17]. In a study of Briel and collegues[18] MR imaging correctly identified sphincter morphology in 23 of 25 cases (92%). In detecting sphincter atrophy endoanal MR imaging showed 89% sensitivity and 94% specificity.

Severe generalized external anal sphincter atrophy at endoanal MR imaging negatively affects continence after anterior anal repair[18]. Therefore, it is advisable using endoanal MR imaging in the preoperative work-up to select patients as candidates for surgery.

Microscopic histopathological investigation confirms findings of external anal sphincter atrophy revealed by MR imaging. Histological, the external anal sphincter is considered to be atrophied when the striated muscle tissue exhibits diminished volume, in association with replacement by fatty tissue (Fig 14, 15[a,b,c])[18].

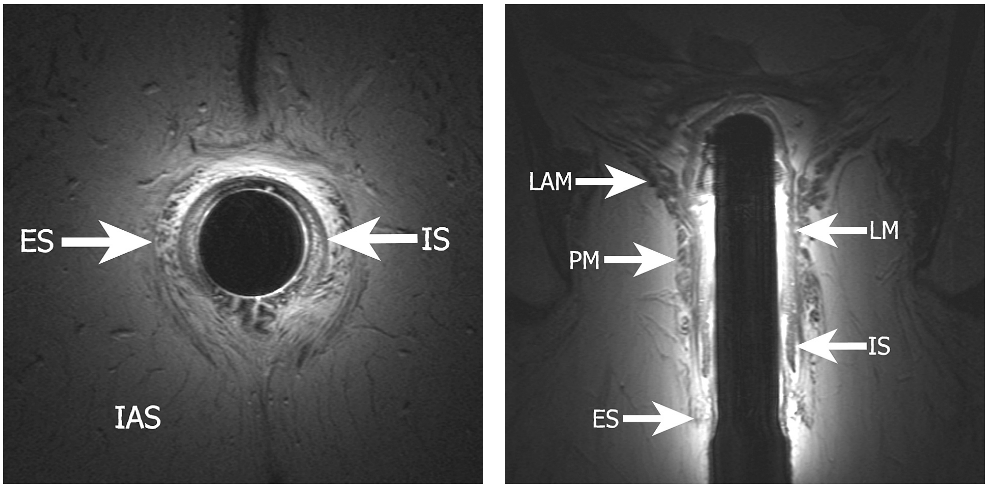


Figure 12[a], 12[b]. (**12[a]**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image and (**12[b]**) coronal endoanal T2- weighted fast spin-echo (2500/70) MR image showing severe thinning of the external anal sphincter (ES) and diffuse replacement by fat in a 69-years-old fecal incontinent woman with no risk factors for pudendal nerve damage in the past (compare to Figs. 1, 2 and 5). The puborectal muscle and levator ani muscle are relatively spared. IS = internal anal sphincter, IAS = ischioanal space, PM = puborectal muscle, LAM = levator ani muscle, LM = longitudinal muscle

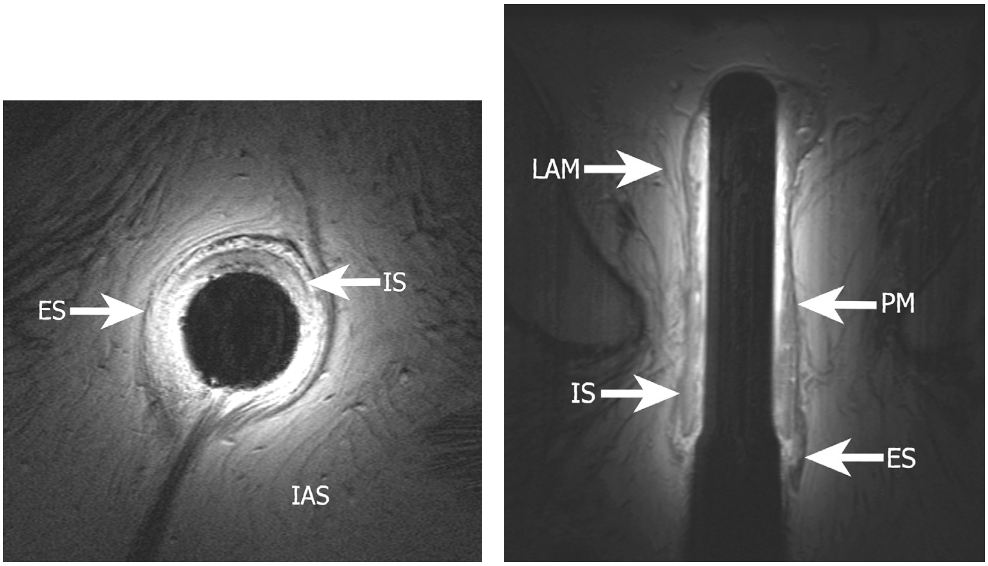


Figure 13[a], 13[b]. (**13[a]**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image showing severe thinning of the external anal sphincter (ES) and diffuse replacement by fat in a 46-years-old man with fecal incontinence with a neurological disorder (spinal) in the past (compare to Figs. 1 and 2). At this level the anterior inferior edge of the internal anal sphincter (IS) is just visible. (**13[b]**) Coronal endoanal T2- weighted fast spin-echo (2500/70) MR image obtained of the same patient shows severe thinning of the external anal sphincter, puborectal muscle (PM) and levator ani muscle (LAM) (compare to Fig 5). IAS = ischioanal space

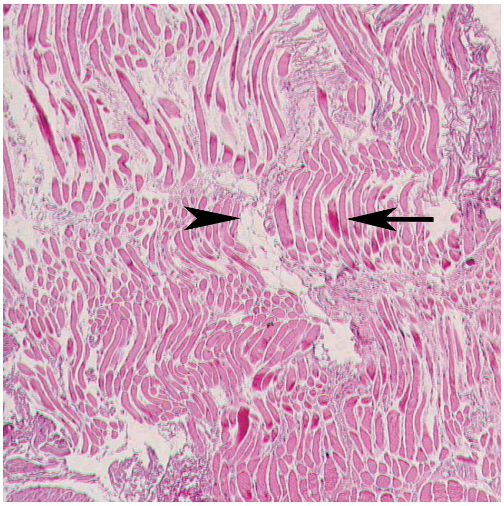


Figure 14. Biopsy specimen of normal external anal sphincter showing myocytes (arrow). Minimal fat tissue (arrow head) is visible. (Haematoxilin, Azofloxin; 16x)

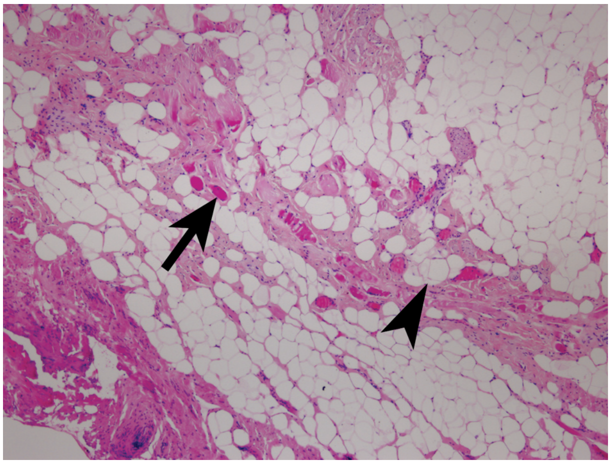
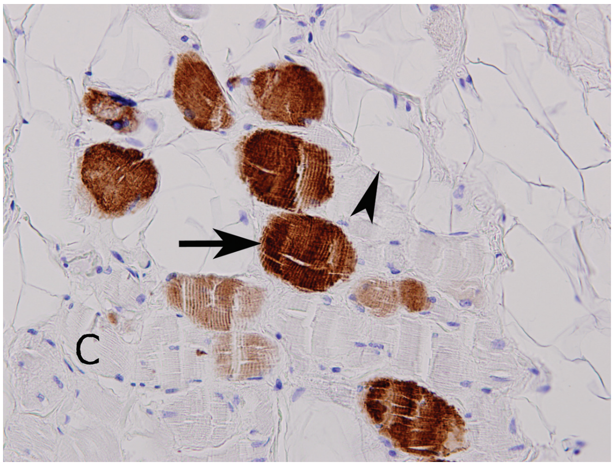
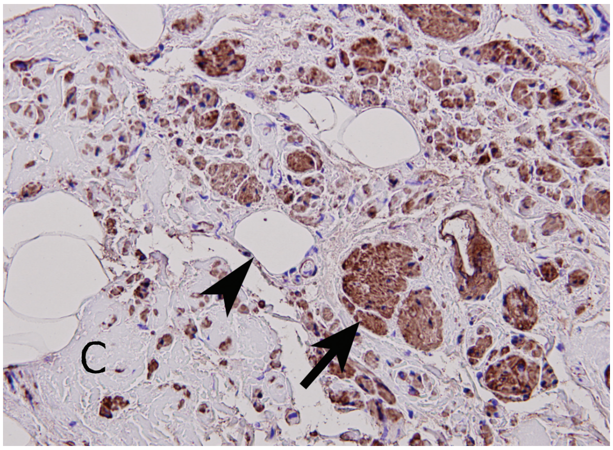


Figure 15[a], 15[b], 15[c]. (**15[a]**) Anus wall biopsy of moderate atrophic external anal sphincter muscle comprising striated muscle (arrow) with replacement by fat tissue (arrow head) from a 61-years-old fecal incontinent woman after a complicated vaginal delivery (breech delivery, heavy child, rupture). (Haematoxilin, Eosin; 16x) On endoanal MR imaging moderate atrophy of the external anal sphincter was depicted (Figure 20[a]).



(**15[b]**) Anus wall biopsy of the anal sphincter muscle obtained from the same patient shows a closer, more detailed view of atrophy of the external anal sphincter, demonstrated by striated muscle (arrow) which is characterized by finger-print structure. Replacement of muscle by fat tissue (arrow head) is demonstrated. C = connective tissue. (Immunostain with antibodies against desmin; 80x)



(**15c**) Anus wall biopsy of the internal anal sphincter obtained from the same patient shows mild atrophy. Smooth muscle (arrow) is surrounded by connective tissue (C) and replacement of muscle by fat tissue (arrow head) is demonstrated. (Immunostain with antibodies against smooth muscle antigen; 16x)

The role of imaging in work-up and therapy of fecal incontinence

Continence is a complex function of multiple anatomic, physiologic, and psychological factors. A systematic evaluation of the patient should reveal the underlying pathophysiology, leading to appropriate therapy. The diagnostic work-up for fecal incontinent patients in general comprises anorectal inspection, digital rectal examination, anorectal function testing and imaging techniques[19].

With digital rectal examination the presence or absence of an external anal sphincter defect can be assessed and resting and incremental squeeze pressure can be determined qualitatively. However, compared to endoanal US, only major sphincter defects can be diagnosed accurately. Furthermore, internal and external anal sphincter weaknesses assessed at digital rectal examination are well but not perfectly related to findings at anorectal manometry.

Anorectal manometry evaluates quantitatively the muscular contraction and relaxation of the anal sphincters by the measurement of pressures in the anal canal. The mean value of the resting pressure is measured as well as the average maximum squeeze pressure.

With anal mucosal sensitivity measurement the threshold sensation of the anus can be determined. Anal sensitivity is a summation of characteristics of the central and peripheral nerve function, and mucosa.

With rectal capacity measurement the reservoir capacity of the rectum can be determined. The reservoir capacity is crucial for normal anorectal functioning. This comprises the capacity to temporarily store faeces as well as the accurate sense of fullness of the rectum.

Pudendal nerve terminal motor latency testing measures the conduction of the pudendal nerve. This technique can help to distinguish whether a weak external anal sphincter muscle results from either nerve or muscle injury.

As anorectal function tests give insight in the functional aspects of the anorectal region, imaging techniques are able to visualize the anatomy and pathology of the anal sphincter muscles. Therefore, endoanal imaging techniques have a central position in the work-up of fecal incontinence and operate as complementary to anorectal function tests. The ability using either endoanal US or endoanal MR imaging has altered our understanding of the pathogenesis of fecal incontinence and has the potential to guide evaluation and management[20]. Endoluminal imaging is useful in the pre-operative work-up to select patients as candidates for surgery and in the postoperative work-up to evaluate the outcome of surgery. Preoperatively, both endoanal MR imaging and endoanal US can reveal external anal sphincter defects to select patients for surgery. Additionally, endoanal MR imaging can depict atrophy of the anal sphincter which is important as patients with extreme external anal sphincter atrophy often have a poor outcome after anterior anal sphincter repair. Postoperatively, endoanal US can reveal persistent defects of the external anal sphincter resulting in failure of the repair[21]. The role of endoanal MR imaging after surgery has not been established yet. Short term results of sphincter repair are contradictory, and recent data suggest that results following sphincter repair deteriorate with time[20]. At present it is not clear if endoanal MR imaging can play a role in identifying etiology factors that are of importance as potential causes of early failure of the repair.

New developments

External phased array MR imaging and three-dimensional endoanal US

Recently, the use of external phased array MR imaging and three-dimensional endoanal US have been studied in the evaluation of fecal incontinence.

External phased array MR imaging provides detailed imaging of the anal sphincter and lower pelvic region without the use of an endoluminal coil (Fig 16[a,b], 17[a,b], 18). Although imaging with an endoluminal coil is able to accurate demonstrate the anal sphincters due to the high spatial resolution[15], endoluminal techniques are primarily used at specialized centers as a dedicated device is necessary. In addition, the introduction of the endoluminal probe or coil leads to discomfort. These two disadvantages of endoluminal techniques could be overcome with the use of external phased array coils. Recent studies showed that external phased array MR imaging is comparable to endoanal MR imaging in the depiction of clinically relevant anal sphincter defects[22] (Fig 19[a,b], 20[a,b,c]) and in the depiction of sphincter atrophy[17] (Fig 21[a,b]), when sufficient experience is available.

With the introduction of three-dimensional endoanal US multiplanar endosonography of the anal canal can be provided (Fig 22, 23). Conventional US is limited to viewing the anal canal in the axial plane, with no capability for longitudinal imaging or measurement. Three-dimensional endoanal US produces a three-dimensional volume that may be reviewed and used to perform measurements in any plane. The depiction of sphincter defects is equal for either conventional or three-dimensional endoanal US in the axial plane (Fig 24, 25), but longitudinal sphincter measurements can be obtained with three-dimensional endoanal US as pictured in Figure 22. With three-dimensional endoanal US it has been shown that in fecal incontinent females with an external anal sphincter defect the anterior sphincter length is shorter and anterior external anal sphincter thickness is smaller[23].

Measurements with three-dimensional endoanal US studies in young controls concerning thicknesses of various layers have been compared to endoanal MR imaging to determine equivalent depth axial images. Good intraobserver and interobserver correlation has been

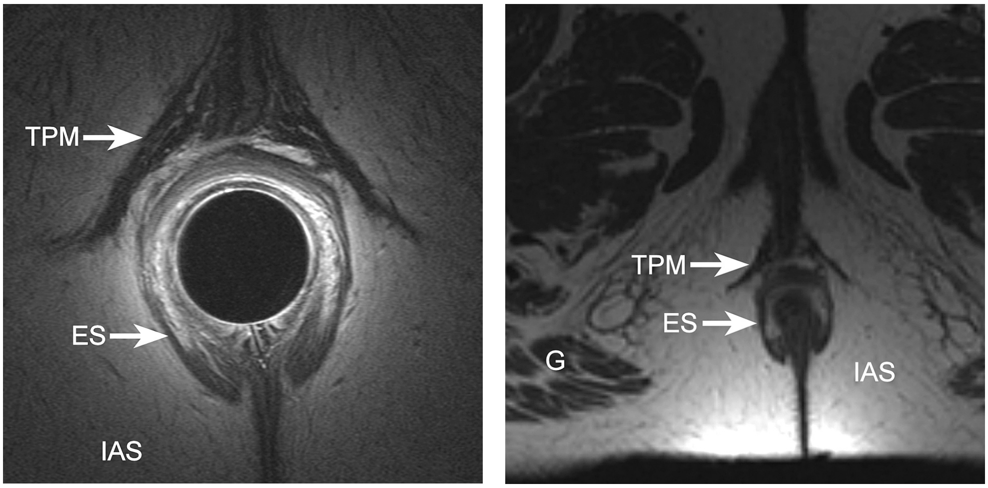


Figure 16a, 16b. (**16a**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image shows the lower part of a normal external anal sphincter (ES) at the distal anal canal in a 56-years-old woman. The external anal sphincter is the hypointense outermost sphincter muscle. (**16b**) Transverse T2- weighted fast spin-echo (2500/70) external phased array MR image of the same patient on the same level. The external anal sphincter is less detailed demonstrated with external phased array MR imaging compared to endoanal MR imaging, but can be readily identified. TPM = transverse perineal muscle, IAS = ischioanal space, GM = gluteus muscle

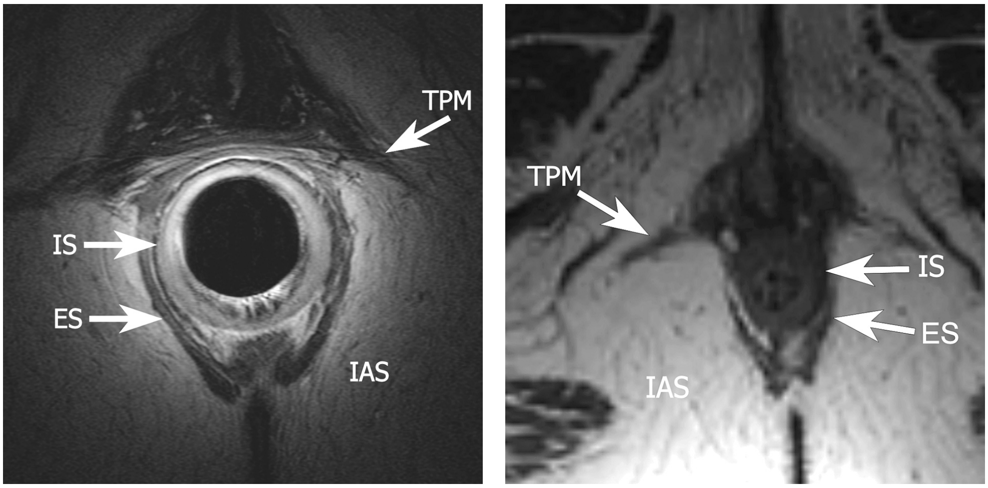


Figure 17a, 17b. (**17a**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image demonstrates a hyperintense internal sphincter. (**17b**) Transverse T2- weighted fast spin-echo (2500/70) external phased array MR image obtained from the same patient at the same level shows the internal anal sphincter (IS) as a homogeneous isointense to hypointense circular band surrounding the anal canal. The difference of the internal anal sphincter signal intensity with the external phased array coil is most likely related to the higher spatial resolution of the endoanal examination. Figure 17 concerns the same patient as Figure 16. ES = external anal sphincter, TPM = transverse perineal muscle, IAS = ischioanal space

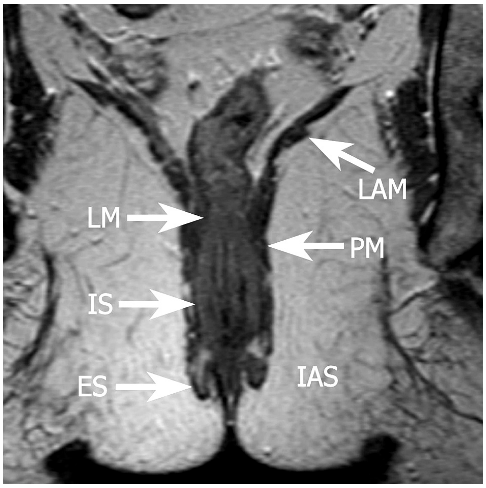


Figure 18. Coronal T2- weighted fast spin-echo (2500/70) external phased array MR image through the anal canal shows normal anatomy of the sphincter complex. It is difficult to make a reliable differentiation between the longitudinal muscle (LM) and external anal sphincter (ES) muscle as either are hypointense. The internal anal sphincter (IS) is isointense to hypointense. Figure 18 concerns the same patient as Figure 16. IAS = ischioanal space, PM = puborectal muscle, LAM = levator ani muscle

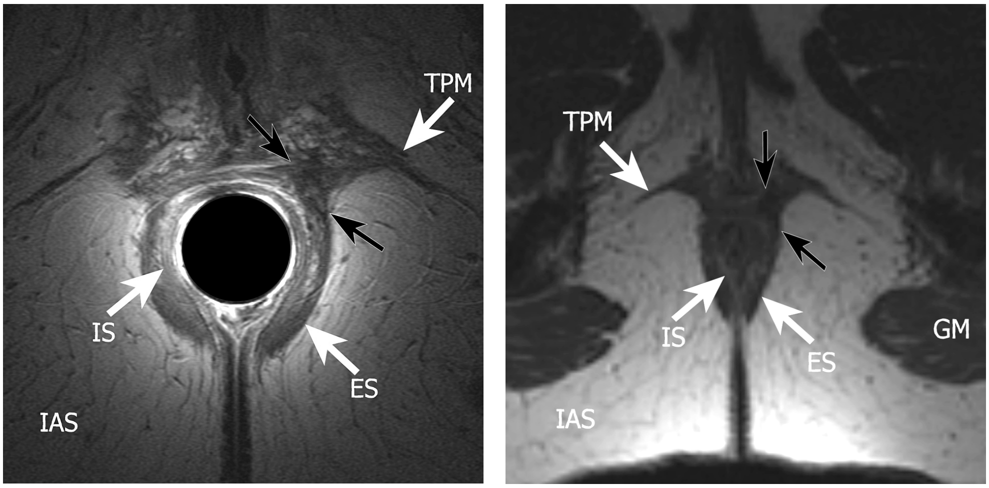


Figure 19a, 19b. (**19a**) Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image from a 42-years-old fecal incontinent woman after a complicated vaginal delivery (long labor, assisted delivery, rupture) shows scar tissue of the external anal sphincter (ES) from 1 – 2 o' clock (open arrows). (**19b**) Transverse T2- weighted fast spin-echo (2500/70) external phased array MR image of the same patient on the same level. Compare the structure of scar tissue with Figure 19a. Note that scar tissue is more hypointense with distorted and asymmetric architecture on endoanal MR imaging. IS = lower part of the internal anal sphincter, TPM = transverse perineal muscle, IAS = ischioanal space, GM = gluteus muscle

shown[10]. Therefore, it may be concluded, amongst experienced observers, that three-dimensional endoanal ultrasonography provides reliable measurements of layer thickness.

The depiction of sphincter atrophy has not been thoroughly evaluated yet. In a recent study with 18 patients three-dimensional endoanal US and endoanal MR imaging showed no difference in the assessment of external anal sphincter atrophy, but there was a substantial difference in grading[24]. Another study with 18 fecal incontinent patients show that volume measurements have been disappointing and reproducibility of volume measurements is moderate[25]. This most

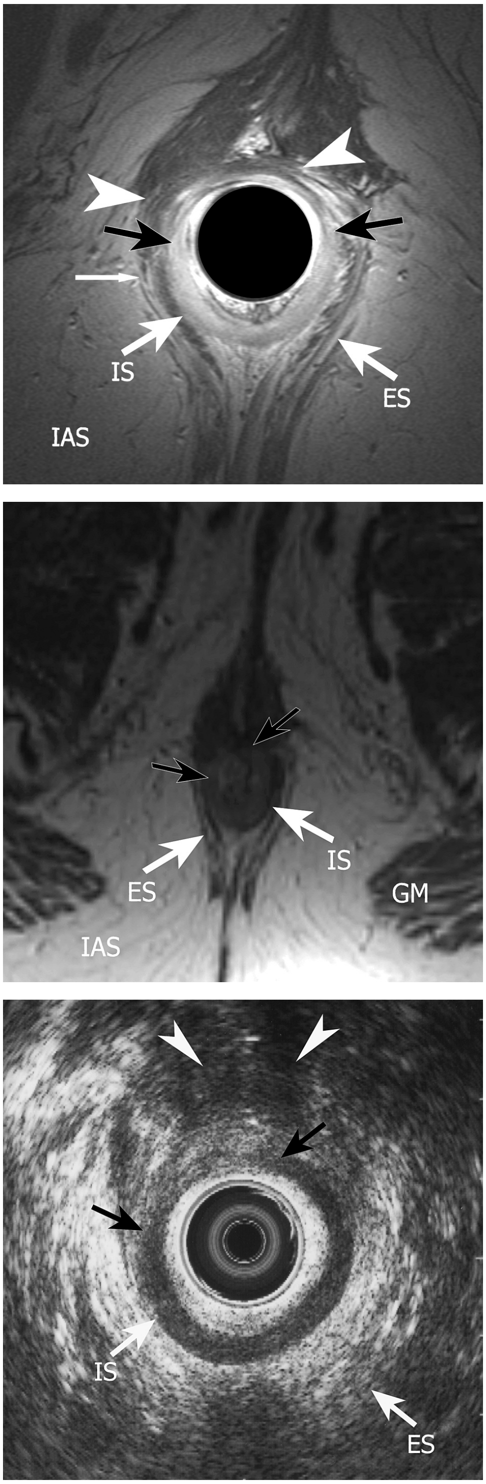


Figure 20[a], 20[b], 20[c].(**20[a]**) Transverse endoanal T2-weighted fast spin-echo (2500/70) MR image and (**20[b]**) transverse T2- weighted fast spin-echo (2500/70) external phased array MR image of a 61-years-old fecal incontinent woman after a complicated vaginal delivery (breech delivery, heavy child, rupture) shows an internal anal sphincter defect (black arrows). On endoanal MR imaging also scar tissue of the external anal sphincter (ES) is visible from 10 – 1 o'clock (arrow heads) and moderate generalized atrophy (thin arrow) is depicted. Histopathology is demonstrated in Figure 15. (**20[c]**) Transverse endoanal ultrasonography at the distal anal canal obtained from the same patient demonstrating an internal anal sphincter defect (black arrows) and an area of low reflectiveness and scar tissue of the external anal sphincter (arrow heads). IS = internal anal sphincter, ES = external anal sphincter, IAS = ischioanal space, GM = gluteus muscle

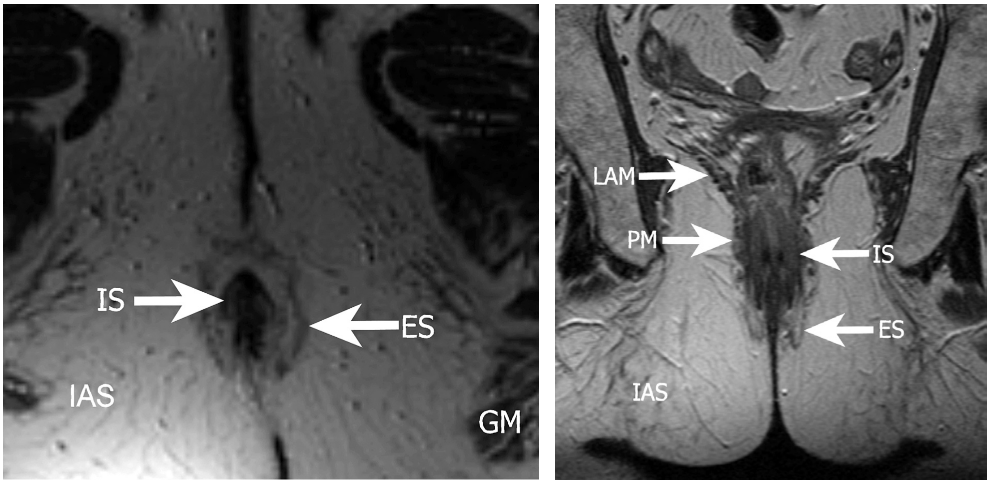


Figure 21^{a}, 21^{b}. (**21^{a}**) Transverse T2- weighted fast spin-echo (2500/70) external phased array MR image and (**21^{b}**) coronal T2- weighted fast spin-echo (2500/70) external phased array MR image showing severe thinning of the external anal sphincter (ES) and diffuse replacement by fat in a 69-years-old fecal incontinent woman with no risk factors for pudendal nerve damage in the past. IS = internal anal sphincter, IAS = ischioanal space, PM = puborectal muscle, LAM = levator ani muscle, GM = gluteus muscle

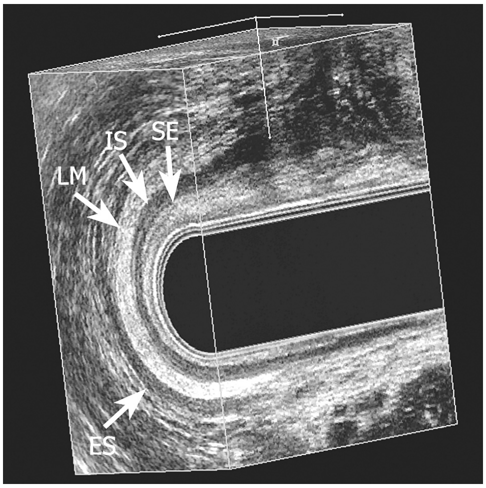


Figure 22. Three-dimensional endoanal ultrasonography, lateral view, from just below the mid-anal canal to above the puborectal muscle obtained from a 60-years-old man demonstrating normal anatomy of the four-layer structure of the sphincter complex. The top of the figure is anterior. SE = subepithelial tissues, IS = internal anal sphincter, LM = longitudinal muscle, ES = external anal sphincter

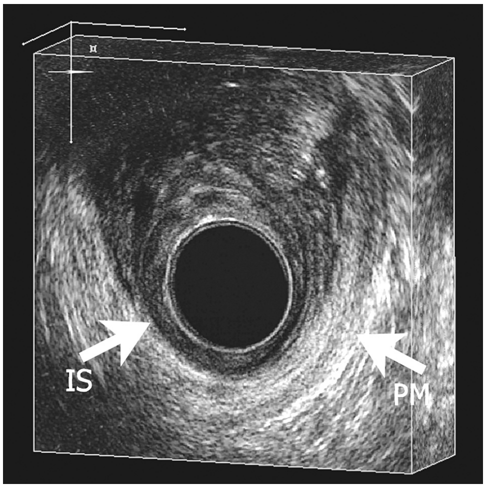


Figure 23. Three-dimensional endoanal ultrasonography at the proximal anal canal from a 60-years-old man demonstrating normal anatomy of the puborectal muscle (PM), visualized as a sling-like structure. The top of the figure is anterior. IS = internal anal sphincter

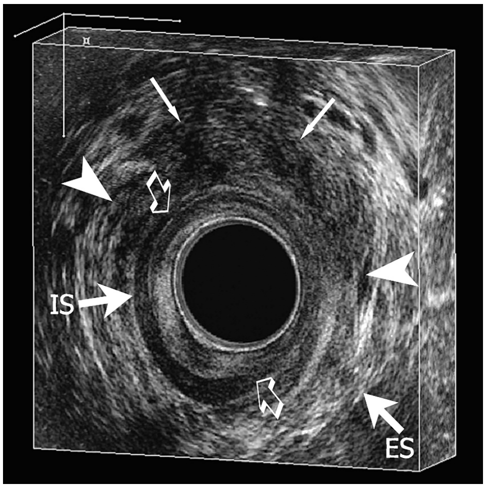


Figure 24. Three-dimensional endoanal ultrasonography at the mid-anal canal obtained from a 37-years-old woman demonstrating a large obstetric tear involving both the internal (open arrows) and external (arrow heads) anal sphincter after a complicated vaginal delivery visualized by discontinuity of the inner and outer sphincter ring. Scar tissue (thin arrows) of the external anal sphincter is depicted anteriorly by segments of very low reflectivity. The top of the figure is anterior.

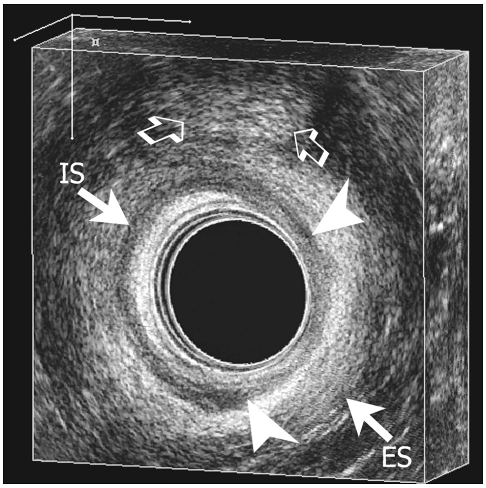


Figure 25. Three-dimensional endoanal ultrasonography at the mid-anal canal obtained from a 57-years-old woman demonstrating a large internal anal sphincter defect from 2 – 5 o'clock (arrow heads) and a small anterior external anal sphincter defect from 11 – 13 o'clock (open arrows) after a lateral internal sphincterotomy (LIS) and complains of soiling. The top of the figure is anterior.

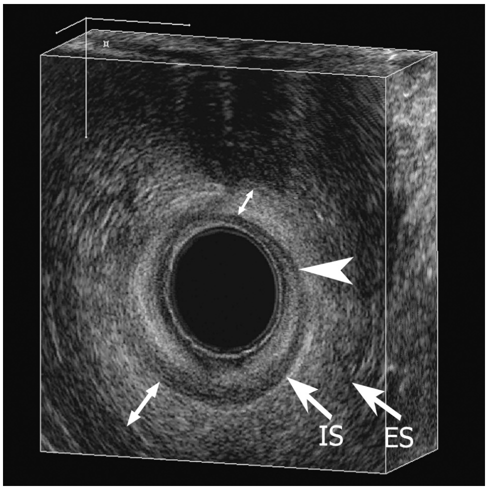


Figure 26. Three-dimensional endoanal ultrasonography at the mid-anal canal obtained from a 65-years-old woman demonstrating moderate atrophy of the external (ES) anal sphincter after a history of constipation and fecal incontinence. Compare the anterior double arrow with posterior. Also some atrophy at the internal anal sphincter is depicted (arrow head). The top of the figure is anterior.

likely can be attributed to the difficult delineation of the external anal sphincter. Fat replacement within the atrophied muscle causes loss of the normal muscle/fat interface border at the outer margin of the external anal sphincter. The outer border of the external anal sphincter is then not defined, and thickness can not be accurately measured (Fig 26)[26].

The most impressive feature of three-dimensional endoanal US is the easiness of viewing the anal sphincter in all different angles and therefore might obtain a better view and insight in the local pathology at endoanal US.

Conclusions

Endoluminal imaging has a prominent role in the diagnostic work-up and evaluation of therapy in fecal incontinence. Supplementary to anorectal function tests, the anatomy and pathology of the anal sphincter complex can be visualized with endoluminal imaging. For the depiction of external anal sphincter defects both endoanal MR imaging and endoanal ultrasonography are useful, whereas endoanal US is cheaper and more operator dependent and endoanal MR imaging is, in contrast to endoanal US, restricted to specialized centers. In the selection of patients for anterior anal sphincter repair, the advantage of endoanal MR imaging is the ability to measure sphincter thickness and atrophy, although with endoanal US an impression about atrophy can also be obtained. In the post surgical work-up endoanal ultrasonography is useful to depict residual sphincter defects as a cause of failure of the repair. The role of postoperative performed endoanal MR imaging in the evaluation of sphincter repair is yet unknown.

New developments have made their advent in the evaluation of sphincter integrity, like external phased array MR imaging and three-dimensional endoanal ultrasonography. Clinical relevant defects and generalized atrophy of the sphincter muscles can be assessed with either endoanal MR imaging or external phased array MR imaging, providing that sufficient experience is available. Three-dimensional endoanal US and conventional endoanal US are equal in the depiction of sphincter defects in the axial plane, but the advantage of multiplanar viewing with three-dimensional endoanal US made sphincter assessment easier and might result in improved diagnostic confidence. For the evaluation of external anal sphincter atrophy with endoanal US more research is needed.

In conclusion, endoluminal imaging has increased our understanding of the sphincter complex resulting in a more adequate evaluation of fecal incontinence.

References

1. Osterberg A, Edebol Eeg-Olofsson K, Graf W. Results of surgical treatment for faecal incontinence. *Br J Surg* 2000;87:1546-52
2. Bharucha AE, Fletcher JG, Harper CM, Hough D, Daube JR, Stevens C, Seide B, Riederer SJ, Zinsmeister AR. Relationship between symptoms and disordered continence mechanisms in women with idiopathic faecal incontinence. *Gut* 2005;54:546-55
3. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *The Lancet* 2004;13;364:621-32
4. Cheetham MJ, Malouf AJ, Kamm MA. Fecal incontinence. *Gastroenterol Clin North Am* 2001;30:115-30
5. Kamm MA. Obstetric damage and faecal incontinence. *The Lancet* 1994;10;344:730-3
6. Stoker J, Rociu E, Zwamborn AW, Schouten WR, Lameris JS. Endoluminal MR imaging of the rectum and anus: technique, applications, and pitfalls. *Radiographics* 1999;19:383-98
7. Stoker J. The anatomy of the pelvic floor and sphincters. In: Bartram CI, DeLancey JOL, Halligan S, Kelvin FM, Stoker J (eds). Imaging pelvic floor disorders. Berlin Heidelberg New York: Springer-Verlag, 2003;1-26
8. Rociu E, Stoker J, Eijkemans MJC, Lameris JS. Normal anal sphincter anatomy and age- and sex-related variations at high-spatial-resolution endoanal MR imaging. *Radiology* 2000;217:395-401
9. Terra MP, Stoker J. Fecal incontinence: Endoanal ultrasonography and MR imaging. In: Santoro GA, Di Falco G (eds). Benign anorectal diseases: diagnosis with endoanal and endorectal ultrasonography and new treatment options. Italia: Springer-Verlag, 2006;115-126
10. Bartram CI. Ultrasound. In: Bartram CI, DeLancey JOL, Halligan S, Kelvin FM, Stoker J (eds). Imaging pelvic floor disorders. Berlin Heidelberg New York: Springer-Verlag, 2003;69-80
11. Rociu E, Stoker J, Zwamborn AW, Lameris JS. Endoanal MR imaging of the anal sphincter in fecal incontinence. *Radiographics* 1999;19:S171-S177
12. Malouf AJ, Williams AB, Halligan S, Bartram CI, Dhillon S, Kamm MA. Prospective assessment of accuracy of endoanal MR imaging and endosonography in patients with fecal incontinence. *Am J Roentgenol* 2000;175:741-5
13. Rociu E, Stoker J, Eijkemans MJ, Schouten WR, Lameris JS. Fecal incontinence: endoanal US versus endoanal MR imaging. *Radiology* 1999;212:453-8
14. Dobben AC, Terra MP, Slors JF, Deutekom M, Gerhards MF, Beets-Tan RG, Bossuyt PM, Stoker J. Comparison of endoanal magnetic resonance imaging and endoanal ultrasonography in the depiction of external anal sphincter defects in fecal incontinent patients . *Radiology.* In press
15. Stoker J, Rociu E, Wiersma TG, Lameris JS. Imaging of anorectal disease. *Br J Surg* 2000;87:10-27
16. Vaizey CJ, Bartram C. Surgical mangement in fecal incontinence. In: Bartram CI, DeLancey JOL, Halligan S, Kelvin FM, Stoker J (eds). Imaging pelvic floor disorders. Berlin Heidelberg New York: Springer-Verlag, 2003;165-170
17. Terra MP, Beets-Tan RG, Van der Hulst VP, Deutekom M, Dijkgraaf MG, Bossuyt PM, Dobben AC, Baeten CG, Stoker J. MR imaging in evaluating atrophy of the external anal sphincter in patients with fecal incontinence. *Am J Roentgenol.* In press
18. Briel JW, Zimmerman DD, Schouten WR. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2001;88:729-30
19. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-60

20. Baig MK, Wexner SD. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2000;87:1316-30

21. Felt-Bersma RJF, Cuesta MA, Koorevaar M. Anal sphincter repair improves anorectal function and endosonographic image - A prospective clinical study. *Dis Colon Rectum* 1996;39:878-85

22. Terra MP, Beets-Tan RG, Van der Hulst VP, et al. Anal Sphincter Defects in Patients with Fecal Incontinence: Endoanal versus External Phased-Array MR Imaging. *Radiology* 2005;236:886-95

23. West RL, Felt-Bersma RJ, Hansen BE, Schouten WR, Kuipers EJ. Volume measurements of the anal sphincter complex in healthy controls and fecal-incontinent patients with a three-dimensional reconstruction of endoanal ultrasonography images. *Dis Colon Rectum* 2005;48:540-8

24. Cazemier M, Terra MP, Stoker J, et al. Atrophy and defects detection of the external anal sphincter: comparison between three-dimensional anal endosonography and endoanal magnetic resonance imaging. *Dis Colon Rectum* 2006;49:20-7

25. West RL, Dwarkasing S, Briel JW, et al. Can three-dimensional endoanal ultrasonography detect external anal sphincter atrophy? A comparison with endoanal magnetic resonance imaging. *Int J Colorectal Dis* 2005;20:328-33

26. Fuchsjager MH, Maier AG. Imaging fecal incontinence. *Eur J Radiol* 2003;47:108-16

Chapter 6

Electrical stimulation and pelvic floor muscle training with biofeedback in patients with fecal incontinence: a cohort study of 281 patients

Maaike P. Terra
Annette C. Dobben
Bary Berghmans
Marije Deutekom
Cor G.M.I. Baeten
Lucas W.M. Janssen
Guy E.E. Boeckxstaens
Alexander F. Engel
Richelle J.F. Felt-Bersma
J. Frederik M. Slors
Michael F. Gerhards
A. Bart Bijnen
Ellen Everhardt
W. Ruud Schouten
Patrick M.M. Bossuyt
Jaap Stoker

Dis Colon Rectum 2006 Aug;49(8):1149-59

Abstract

PURPOSE. Pelvic floor rehabilitation is an appealing treatment for patients with fecal incontinence but reported results vary. This study was designed to assess the outcome of pelvic floor rehabilitation in a large series of consecutive patients with fecal incontinence caused by different etiologies.

METHODS. A total of 281 patients (252 females) were included. Data about medical history, anorectal manometry, rectal capacity measurement, and endoanal ultrasonography were collected. Subgroups of patients were defined by anal sphincter complex integrity, and nature and possible underlying causes of fecal incontinence. Subsequently patients were referred for pelvic floor rehabilitation, comprising nine sessions of electrical stimulation and pelvic floor muscle training with biofeedback. Pelvic floor rehabilitation outcome was documented with Vaizey score, anorectal manometry, and rectal capacity measurement findings.

RESULTS. Vaizey score improved from baseline in 143 of 239 patients (60 percent), remained unchanged in 56 patients (23 percent), and deteriorated in 40 patients (17 percent). Mean Vaizey score reduced with 3.2 points ($P < 0.001$). A Vaizey score reduction of ≥ 50 percent was observed in 32 patients (13 percent). Mean squeeze pressure (+5.1 mmHg; $P = 0.04$) and maximal tolerated volume (+11 ml; $P = 0.01$) improved from baseline. Resting pressure ($P = 0.22$), sensory threshold ($P = 0.52$), and urge sensation ($P = 0.06$) remained unchanged. Subgroup analyses did not show substantial differences in effects of pelvic floor rehabilitation between subgroups.

CONCLUSIONS. Pelvic floor rehabilitation leads overall to a modest improvement in severity of fecal incontinence, squeeze pressure, and maximal tolerated volume. Only in a few patients, a substantial improvement of the baseline Vaizey score was observed. Further studies are needed to identify patients who most likely will benefit from pelvic floor rehabilitation.

Introduction

Fecal incontinence is a common disorder[1] and can be defined as the involuntary loss of flatus, liquid or solid stool, which is a social or hygienic problem[2]. Fecal incontinence is primarily caused by anal sphincter defects, neuropathy, reduced rectal capacity and compliance, reduced fecal consistency, or a combination of these causes[3].

Pelvic floor rehabilitation, pelvic floor muscle training combined with biofeedback[4, 5] and electrical stimulation[6], is a commonly used therapy and several studies have evaluated its outcome in patients with fecal incontinence. Interpretation and comparison of reported results are difficult, because these studies varied in patient selection, sample size, methodology, biofeedback, and electrical stimulation techniques used, as well as in outcome measures, criteria for success, and length of follow-up period[3, 5–7]. The reported success rates after pelvic floor muscle training with biofeedback vary from 0 to 100 percent[8].

6

Studies evaluating the outcome of electrical stimulation in isolation or in combination with pelvic floor muscle training with biofeedback are rare but have shown promising results[9–12]. Because pelvic floor rehabilitation is simple, inexpensive, and mostly without unfavorable physical side effects, it is an appealing conservative treatment option in patients with fecal incontinence[3,13]. This study was designed to assess the outcome of a standardized pelvic floor rehabilitation program, comprising nine sessions of electrical stimulation and pelvic floor muscle training with biofeedback, in a large population of consecutive patients with fecal incontinence caused by different etiologies. In addition, the outcome across a number of clinical subgroups was compared.

Patients and Methods

Patients

This prospective cohort study was performed between December 2001 and March 2005 in 16 medical centers in the Netherlands. The Medical Ethics Committees of all centers had approved the study.

We invited consecutive patients with fecal incontinence. Inclusion criteria were fecal incontinence complaints for at least six months, Vaizey score of at least[12, 14] and failure of conservative treatment, including dietary measures and medication. Patients younger than aged 18 years, patients diagnosed with an anorectal tumor, chronic diarrhea (always fluid stools, 3 or more times per week), overflow incontinence, proctitis, soiling, previous ileoanal or coloanal anastomosis, and/or rectal prolapse were excluded, as were patients who had received pelvic floor rehabilitation in the previous six months or who were considered to be cognitively unable to undergo pelvic floor rehabilitation.

Diagnostic Tests

Eligible patients who signed informed consent were included. Information about medical history, including duration, nature, severity, and possible underlying causes for fecal incontinence, was obtained by physicians.

Nature of fecal incontinence was classified as passive incontinence (defecation loss without the patient's knowledge), urge incontinence (lack to defer defecation for 15 minutes), or a combination of passive and urge incontinence[15,16]. Severity of fecal incontinence was assessed with the grading system of Vaizey[14], ranging from 0 (complete continence) to 24 (complete incontinence). Possible underlying causes for fecal incontinence were divided in two main groups: anatomic disorders vs functional disorders.

Anorectal manometry took place according to the solidstate or water-perfused technique with or without sleeve[17]. The solid-state method or water-perfusion method without sleeve was performed by means of a pull-through technique. The (mean) maximal resting pressure (normal range, 35–70 mmHg) was measured[17]. Subsequently, the (mean) maximum squeeze pressure (normal range, 140–220 mmHg) was determined by asking patients to squeeze three times during ten seconds with one-minute intervals[17]. An average maximum squeeze pressure was calculated. Rectal capacity measurement was performed by use of a balloon catheter or a barostat[17]. The minimal rectal sensation perceived (sensory threshold; normal range, 10–30 ml), the volume associated with the initial urge to defecate (urge sensation; normal range, 60–150 ml), and the volume at which the patientexperienced discomfort and an intense desire to defecate (maximal tolerated volume; normal range, 150–250 ml) were determined[17].

Endoanal ultrasonography was performed with an ultrasound scanner with a radial endoscopic probe (7.5- or 10-MHz transducer)[17]. The integrity of the anal sphincter complex was assessed. A defect of the internal or external anal sphincter was defined by a discontinuity of the muscle ring (anatomic defect) and / or characterized by loss of the normal architecture, with an area of amorphous texture that usually has low reflectiveness (functional defect, scar tissue). A defect was noted when the axially extent comprised at least one hour (i.e., 30 degrees) of the circumference of the sphincteric ring and the longitudinally extent included at least the proximal, middle, distal, or a combination of different levels of the anal canal.

Anorectal manometry, rectal capacity measurement, and endoanal ultrasonography were performed according to a standard procedure by physicians or technicians with at least ten years of experience in performing and evaluating the specific tests. In two hospitals, the tests were performed by residents with approximately one year of experience in performing and evaluating the tests, supervised by a physician with at least ten years of experience with anorectal manometry, rectal capacity measurement, and endoanal ultrasonography.

Pelvic Floor Rehabilitation

After diagnostic testing, patients were referred for pelvic floor rehabilitation. Nationwide, 71 specialized pelvic physiotherapists, trained and instructed in auniform way, contributed to this study. Pelvic floor rehabilitation was administered according to a standardized protocol, which had been developed by clinicians and physiotherapists specialized in the field of pelvic floor disorders. Patients underwent weekly sessions of a minimal of 35 minutes for 9 weeks. Details about the training program and at home exercises were explained to the patient.

Physiotherapists were aware of details about medical history but were blinded for findings at anorectal manometry, rectal capacity measurement, and endoanal ultrasonography. During the sessions, physiotherapists obtained data by performing digital rectal examination, rectal balloon training, and electromyography. Treatment targets and program were formulated based on these

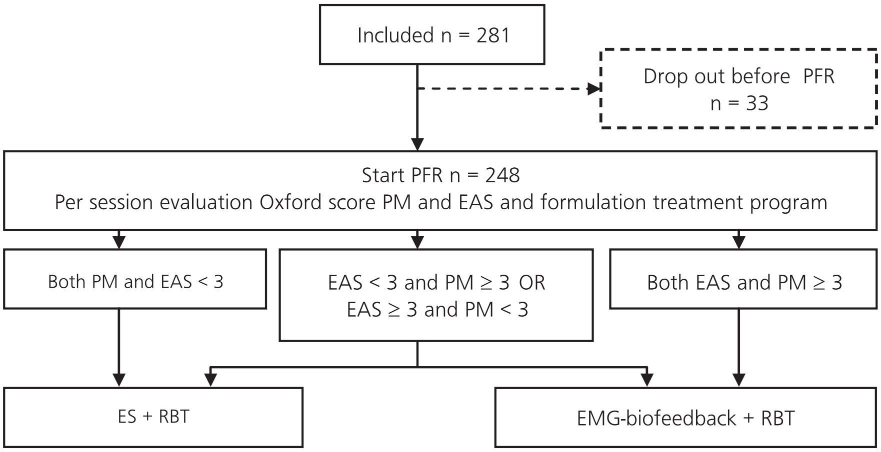


Figure 1. Patient flow in the study. The Oxford score reflects the strength of the puborectal muscles and external anal sphincter muscle. The Oxford score ranges from 0 to 5 with 0 indicating no muscle contraction, 1 vibrating contraction, 2 weak contraction, 3 average contraction, 4 good contraction and 5 strong contraction. PFR = pelvic floor rehabilitation, EAS = external anal sphincter; PM = puborectal muscle, ES = electrical stimulation, EMG = electromyographic feedback, RBT = rectal balloon training

data. Each subsequent session included rectal balloon training, electrical stimulation, and / or electromyographic feedback (Fig. 1).

Digital Rectal Examination

External anal sphincter and puborectal muscle strength was assessed according to the Oxford score[18, 19], ranging from 0 (no muscle contraction) to 5 (strong contraction). Endurance of submaximal strength and exhaustion of these muscles also were determined.

Electrical Stimulation

Electrical stimulation was offered only to patients with a poorly functioning external anal sphincter and/or puborectal muscle (Oxford score <3) and performed at 50 Hz with biphasic pulse duration of 200 microseconds using an intra-anal electrode (NEEN, PelviTec BV, Delft, the Netherlands) and a muscle stimulator (Myomed 932, Enraf-Nonius NV, Delft, the Netherlands). Electrical stimulation was given during 20 minutes in 13-second cycles, comprising 5 seconds activity and 8 seconds rest. Electrical stimulation was offered until patients were able to develop independently enough (Oxford score ≥3) external anal sphincter and puborectal muscle strength.

Biofeedback

Rectal balloon training was offered to all patients. A rectal balloon (Medeco BV, Oud Beijerland, The Netherlands) attached to a 60 mlsyringe was introduced in the rectum and slowly inflated with air. Sensory threshold, urge sensation, and maximal tolerated volume were assessed[17].
Patients with an insensitive rectum were taught to perceive smaller volumes of distention until a normal level of sensory threshold was reached or further improvement could not be expected. In patients with a hypersensitive rectum, stepwise relaxation and suppression of urgency feeling

in response to rectal filling were learned until a normal level of urge sensation was reached or the urge did not diminish with time or was uncomfortable. Patients with weakness of the pelvic floor muscles learned to contract these muscles immediately and strongly in response to rectal filling. The physiotherapist gave auditory feedback to the patient.

Electromyographic Feedback

Electromyographic feedback was offered to all patients with an average functioning external anal sphincter and/or puborectal muscle (Oxford score ≥3) and performed with the same device and electrode as used for electrical stimulation. Activity of the external anal sphincter and puborectal muscle in response to rectal filling, during rest, maximal contraction, and after maximal contraction were registered and presented to the patient.

Contraction capacities, including duration, relaxation, timing, and coordination of the pelvic floor muscles, were trained. Auditory feedback was given to stimulate correct performance of contractions. Patients were instructed and encouraged to perform a home maintenance program consisting of the above mentioned contraction exercises.

Any protocol violations or study-related adverse events were noted. Three months after pelvic floor rehabilitation, patients were referred back to the physician, who obtained data from medical history, anorectal manometry, and rectal capacity measurement to evaluate the effects of pelvic floor rehabilitation.

Statistical Analysis

To assess outcome of pelvic floor rehabilitation differences in Vaizey score, anorectal manometry findings and rectal capacity measurement findings between baseline and after therapy were calculated and tested for significance using t-test statistics for paired data. A reduction in Vaizey score < 50 percent was defined as a slight improvement and a reduction of ≥50 percent as a substantial improvement. Pearson's correlation coefficient was computed to assess the ssociation between change in Vaizey score after therapy and baseline Vaizey score.

We also evaluated the results in clinical subgroups, defined by anal sphincter complex integrity at endoanal ultrasonography, and by nature and possible underlying causes of fecal incontinence. Analysis of variance was used to study the existence of significant differences in mean change of outcome measure between clinical subgroups. Whenever significant differences between means were found, Bonferroni corrections were used to determine which means differed.

Student's t-test for paired data and chi-squared statistics were used to test for significant differences between pelvic floor rehabilitation data obtained at the initial and final session.
For all statistical tests, P values < 0.05 were considered to represent statistical significance.

Results

Patients

We included 281 patients (252 females; mean age, 59 years (standard deviation (SD) ±13). The median duration of fecal incontinence was five (range, 0.5 –57) years. The mean Vaizey score was 18 (SD ±3.1). Table 1 shows the possible underlying causes for fecal incontinence.

Table 1. Possible underlying causes of fecal incontinence in 29 males and 252 females

Characteristics	n (%)
Anatomical	
Anatomical congenital	
Spina bifida	1 (0.4%)
Anatomical traumatic	
Complicated vaginal delivery	208 (74%)
Anorectal surgery	60 (21%)
Colorectal surgery	27 (9.6%)
Gynaecological surgery	110 (39%)
Urological surgery	49 (17%)
Functional	
Neurological	
Cerebral disorders	9 (3.2%)
Spinal cord disorders / surgery	26 (9.2%)
Miscellaneous	6 (2.1%)
Metabolic	
Diabetes Mellitus	26 (9.2%)
Inflammatory bowel diseases	
Crohn's disease	2 (0.7%)
Systemic diseases	
Connective tissue disease	13 (4.6%)

Mean resting and squeeze pressure were 48 mmHg (SD ±22) and 87 mmHg (SD ±39), respectively. Mean sensory threshold, urge sensation, and maximal tolerated volume were 50 ml (SD ±33), 93 ml (SD ±49), and 156 ml (SD ±68), respectively. Available details about the respective subgroups are shown in Table 2.

Pelvic Floor Rehabilitation

Figure 1 shows the flow of patients in the study. Of 281 patients, 248 (88 percent) started with pelvic floor rehabilitation. Thirty-three patients (12 percent) did not start the pelvic floor rehabilitation program for different reasons, including concomitant disease (n = 9), lack of time or motivation (n = 9), spontaneous improvement of fecal incontinence (n= 2), or unknown reasons (n = 13).

The mean number of weeks between inclusion and first treatment session was 13 (SD ±10). The mean number sessions with electrical stimulation, rectal balloon training, and electromyographic feedback was 1.8 (SD ±2.9), 7.2 (SD ±1.4), and 6.9 (SD ±1.8), respectively. The mean duration between first and final treatment session was 10 weeks (SD ±5). Details of diagnostic pelvic floor rehabilitation data are shown in Table 3.

In some patients, one or more planned training sessions with electrical stimulation, rectal balloon training, and / or electromyographic feedback had to be cancelled because of patient-

Table 2. Mean change of Vaizey score, findings from anal manometry and findings from rectal capacity measurement after pelvic floor rehabilitation in clinical subgroups of patients

Baseline characteristics	Vaizey score		Resting pressure		Squeeze pressure	
	Mean change	p	Mean change	p	Mean change	p
Endoanal sonography						
No defect (n = 94)	-3.4	0.84	3.6	0.22	4.9	0.77
Solitary defect IAS (n = 10)	-2.9		-8.9		-6.4	
Solitary defect EAS (n = 79)	-2.6		1.7		7.1	
Defect IAS and EAS (n = 67)	-3.3		-0.8		4	
Nature incontinence						
Passive incontinence (n = 9)	-4.4	0.15	1.8	0.03 *	7.6	0.82
Urge incontinence (n = 108)	-3.9		3.5		7.5	
Combined incontinence (n = 154)	-2.6		-0.6		4.3	
Possible underlying causes						
None (n = 30)	-4.5	0.41	-2.9	0.32	-9.6	0.47
Only anatomical (n = 166)	-3.1		2.7		6.7	
Only functional (n = 15)	-1.4		3.1		3.3	
Anatomical and Functional (n = 70)	-3.7		-0.9		4.9	

Notes: Mean change = data after therapy minus data before therapy; IAS = internal anal sphincter; EAS = external anal sphincter; PM = puborectal muscle; combined incontinence = passive and urge incontinence. *Significant difference in mean change of resting pressure between patients with passive and those with combined incontinence.

specific (n = 24), physiotherapist-specific (lack of time, n = 12; other, n= 82), or technical-specific (n = 8) reasons.

In 35 patients, electrical stimulation, rectal balloon training, and/or electromyographic feedback sessions did not take place according the protocol.

In 13 patients, the rectal balloon was damaged and in 8 patients another intra-anal electrode than prescribed by protocol was used during one or more sessions. Adverse events comprised pain (n = 3), rectal bleeding (n = 4), and latex allergy (n = 1).

Outcome of Pelvic Floor Rehabilitation

Mean number of weeks between the last treatment session and evaluation of outcome was 11 (SD ±10). Three patients dropped out after therapy (lack of time or motivation, n = 2; concomitant disease,n = 1). Vaizey scores at baseline and after therapy were available in 239 patients (85 percent).

Figure 2 shows the changes from baseline. The Vaizey score improved from baseline in 143 patients (60 percent), remained unchanged in 56 (23 percent), and deteriorated in 40 others (17 percent). Vaizey score improvement was slight in 111 patients (47 percent) and substantial in 32 patients (13 percent). Mean Vaizey score after pelvic floor rehabilitation was 15 (SD ±5.4), an average change of -3.2 points (95 percent confidence interval (CI), -2.6 to -3.9; $p < 0.001$). Change in Vaizey score was not strongly associated with baseline Vaizey score ($r = 0.2$; $p = 0.001$).

	Sensory threshold		Urge sensation		Maximal tolerated volume	
	Mean change	p	Mean change	p	Mean change	p
	4.1	0.72	6.8	0.52	18	0.4
	-4.8		-3.1		18	
	-2		-0.14		-0.12	
	2.4		12		12	
	-32	0.05	-13	0.57	1.2	0.57
	-1		5.8		17	
	3.8		7		8	
	24	0.25	24	0.72	18	0.4
	2		7.9		17	
	-18		-18		0.12	
	-2.4		-0.3		12	

6

Table 3. Mean change in diagnostic pelvic floor rehabilitation data

Characteristics	First session mean (± SD)	Final session mean (± SD)	Mean change	p
Digital rectal examination				
Oxford score PM	2.9 (± 1)	3.6 (± 1)	0.7	< 0.001
Oxford score EAS	2.5 (± 1)	3.1 (± 1.1)	0.6	< 0.001
Endurance submaximal PM strength (sec)	13 (± 10)	19 (± 10)	6.4	< 0.001
Exhaustion PM (n)	3.8 (± 1.5)	4.8 (± 1.7)	1	< 0.001
Endurance submaximal EAS strength (sec)	11 (± 9.2)	15 (± 11)	3.9	< 0.001
Exhaustion EAS (n)	3.5 (± 1.7)	4.2 (± 1.6)	0.7	< 0.001
Rectal capacity measurement				
Sensory threshold (ml)	31 (± 22)	34 (± 28)	1.8	0.35
Urge sensation (ml)	81 (± 51)	91 (± 37)	12	< 0.001
Maximal tolerated volume (ml)	173 (± 72)	213 (± 57)	44	< 0.001
Electromyography				
Resting tone PM and EAS before activity (μV)	6.9 (± 5.1)	5.7 (± 3.8)	-1.2	< 0.001
Activity PM and EAS at maximal contraction (μV)	28 (± 17)	34 (± 19)	5.8	< 0.001
Resting tone PM and EAS after maximal contraction (μV)	6 (± 4.6)	4.7 (± 3.9)	-1.2	< 0.001

Notes: Mean change = data after therapy minus data before therapy; PM = puborectal muscle; EAS = external anal sphincter

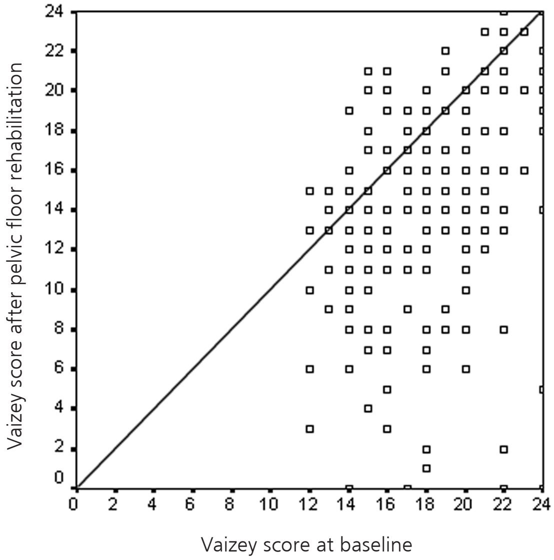


Figure 2. Vaizey score at baseline and after pelvic floor rehabilitation. The line reflects no change of the Vaizey score compared to baseline. The blocks under this line represent the patients in which the Vaizey score improved. In the majority of patients the Vaizey score decreased after therapy.

Baseline and follow-up anorectal manometry findings were available in 217 patients (77 percent). Mean resting pressure increased from baseline in 114 patients (53 percent), remained unchanged in 11 (5 percent), and decreased in 92 patients (42 percent). Mean squeeze pressure increased from baseline in 121 patients (56 percent), remained unchanged in 3 (1 percent), and decreased in 93 patients (43 percent). Mean resting and squeeze pressure after therapy were 50 mmHg (SD ±24) and 91 mmHg (SD ±44), respectively, a mean increase of 1.6 mmHg (95 percent CI, -1 to 4.1; $p = 0.22$) and 5.1 mmHg (95 percent CI, 0.3 –10; $p = 0.04$). Baseline and follow-up sensory threshold, urge sensation, and maximal tolerated volume were available in respectively 204 (73 percent), 207 (74 percent), and 207 (74 percent) patients. After pelvic floor rehabilitation, mean sensory threshold increased in 93 patients (45 percent), remained unchanged in 22 (11 percent), and decreased in 89 patients (44 percent). Mean urge sensation increased from baseline in 111 patients (54 percent), remained unchanged in 19 (9 percent), and decreased in 77 others (37 percent). Mean maximal tolerated volume increased from baseline in 112 patients (54 percent), remained unchanged in 17 (8 percent), and decreased in 78 patients (38 percent). Mean sensory threshold, urge sensation, and maximal tolerated volume after treatment were 53 ml (SD ±37), 102 ml (SD ±53), and 170 ml (SD ±68), respectively, a mean increase of 1.6 ml (95 percent CI, -3.4 to 6.7;$p = 0.52$), 6 ml (95 percent CI, -0.3 to 12; $p = 0.06$), and 11 ml (95 percent CI, 2.7– 20; $p = 0.01$).

Significant differences in change from baseline in Vaizey score and findings from anorectal manometry and rectal capacity measurement in any of the subgroups are demonstrated in Table 2.

Discussion

This study demonstrates that a pelvic floor rehabilitation program comprising electrical stimulation and different elements of pelvic floor muscle training with biofeedback leads to a slight improvement from baseline in the severity of fecal incontinence in a majority of patients with fecal incontinence. Only a few of the studied patients demonstrated a substantial improvement after treatment. The significant improvement from baseline in mean Vaizey score was modest and was accompanied by a significant but small increase in average squeeze pressure and maximal tolerated volume, and without a significant change in mean resting pressure, sensory threshold, and urge sensation. Subgroup analyses showed that the changes in Vaizey score, squeeze pressure, sensory threshold, urge sensation, and maximal tolerated volume were irrespective of anal sphincter complex integrity and the nature and possible underlying causes of fecal incontinence. A significant difference in change of resting pressure was only found between patients with passive incontinence and those with combined incontinence.

A number of potential limitations of this study should be taken into account. The observed changes in Vaizey score, findings from anorectal manometry, and rectal capacity measurement findings after therapy in our study cannot exclusively be attributed to the pelvic floor rehabilitation program, because this study was not designed as a randomized, clinical trial. The effects of patient expectations, placebo response, and natural course cannot be completely excluded[20]. We performed a prospective cohort study because we wanted to evaluate the effects of a pelvic floor rehabilitation program in a large patient group of patients with fecal incontinence caused by mixed etiologies and compare the results in clinical subgroups. As a result of the design of this study, changes in outcome cannot be ascribed to specific elements of the pelvic rehabilitation program used in this study.

6

In the literature, several outcome measurements have been used to evaluate the success of pelvic floor rehabilitation. We decided to use the Vaizey score as our primary outcome measure, because in clinical practice this score is a widely used summary score containing important incontinence-specific items[14]. A previous study demonstrated that this scoring system is reproducible and correlates well with physician's clinical impression[14, 21]. Studies of our group have shown that higher Vaizey scores are associated with more reported problems in general health domains[22] and that changes in Vaizey score reflect patient's subjective perception of relief[23]. The latter study revealed that after therapy the mean Vaizey score was one point lower for patients who rated their situation as worse or equal, four points for patients who reported their situation to be better, and nine points lower in patients who rated their situation much better. The Wexner incontinence score, another widely used incontinence score, also is reproducible and correlates well with the physician's clinical impression[14, 21]. We found similar for the Vaizey incontinence score a significant average improvement compared with baseline for the Wexner incontinence score in the total patient group and no significant difference in change of Wexner score between subgroups (data not shown). As Bharucha[24] has emphasized the importance of using objective outcome measures in therapeutic trials, we incorporated anorectal manometry and rectal capacity measurement as additional outcome measures.

We are aware of the susceptibility of the Vaizey score to recall bias yet. Naliboff[25] reported that a single retrospective rating of "usual" symptom intensity may be a good reflection of the average symptom intensity for chronic pain. Pamuk et al[26] have shown in a constipation study that the results concerning the presence of constipation were highly compatible between self-reported constipation and diary-based constipation. We expect the frequency of reported symptoms in the Vaizey score to reflect the actual frequency of symptoms. Diaries may be less affected by recall bias, because they can capture experiences close to the time of occurrence. Unfortunately, a recent study has demonstrated that the compliance rate of paper diaries is low and that hoarding is a frequently encountered problem in using paper diaries[27]. Self-assessment questionnaires may be preferable to questionnaires by an interviewer (e.g., Vaizey score) for assessing the level of complaints because they reduce the likelihood of socially desirable or biased answers[28]. Bakx and colleagues[28] have developed a specific colorectal functional outcome (COREFO) questionnaire; however, this questionnaire was not available at the time this study was initiated. The treatment program in this study was customized on a per patient basis by using a patient's Oxford score to guide therapy.

We are aware of the subjective component in the Oxford score, but a previous study showed good agreement between digital assessment of pelvic floor muscle strength and vaginal perineometry[18]. Patients participating in this study live widespread over the Netherlands. To diminish patient discomfort and to increase compliance,a large number of physiotherapists were selected to participate in our study. The high number of involved physiotherapists may have increased the variability in the Oxford score.

There is no consensus in the literature about the optimal number and duration of pelvic floor rehabilitation sessions[3]. We limited therapy to nine sessions because most patients in the Netherlands have reimbursement for this maximum number only. It is unknown whether this limitation prevented some patients from obtaining adequate improvement.

Perfect treatment requires both a cooperative patient and a skilled and motivated physiotherapist[29, 30]. Physiotherapists participating in this study were experienced in performing pelvic floor rehabilitation and uniformly trained and instructed. Minor protocol violations were observed, which may have decreased somewhat the homogeneity of the collected data.

Comparisons between our findings and those from other studies are difficult, because previous studies varied in patient selection, design, outcome measures, as well as in the biofeedback or electrical stimulation techniques used [3, 5–7]. Other studies also have reported a significant improvement in severity of fecal incontinence [9, 20, 31] a significant improvement in squeeze pressure[10, 30–34] and no significant change in resting pressure [32, 34].

In contrast to previous studies, we did not observe a significant change in sensory threshold and urge sensation [34, 35]. Several authors emphasized the importance of patient selection and reported that pelvic floor rehabilitation is less effective in patients with neurogenic fecal incontinence [36] and passive incontinence[12], but we could not find a strong basis for these arguments. Norton and colleagues found better results in patients with an intact anal sphincter complex[12]. We found as earlier studies[37, 38] that effects were not significantly different between clinical subgroups based on anal sphincter complex integrity.

Pelvic floor rehabilitation is designed to strengthen the voluntary muscles and to enhance anorectal sensory perception and compliance. In addition to an increase of squeeze pressure, one expects an improvement to discriminate and respond to smaller rectal volumes in patients with a hyposensitive rectum, and a tolerance of larger volumes and urge resistance in patients with a hypersensitive rectum. We observed a significant increase in squeeze pressure and maximal tolerated volume; however, the effects were small and might be of no clinical relevance. Furthermore, no significant changes in sensory threshold and urge sensation were observed. The improvement of the majority of the diagnostic pelvic floor rehabilitation data also was limited. The fact that we were unable to demonstrate a substantial improvement of trained parameters could be explained by the complex underlying pathophysiology in the participating patients, because the mean Vaizey score was relatively high, indicating moderate-to-severe fecal incontinence complaints. Another explanation could be that our study group consisted of patients with a wide spectrum of predisposing causes. The limited number of pelvic floor rehabilitation session also may have contributed.

Because physiologic data did not improve substantially in our study, it is puzzling and unclear what exactly caused symptom improvement. As a result of the complex and multifactorial pathogenesis of fecal incontinence, we can only hypothesize which factors contributed to improvement.

Conclusions

This study demonstrated that a pelvic floor muscle rehabilitation program can provide relief, with a substantial improvement of fecal incontinence complaints in a minority of patients only. The observed changes in Vaizey score, squeeze pressure, and maximal tolerated volume after treatment were significant but small. Because subgroup analyses were unable to identify patients who were most likely to benefit from pelvic floor rehabilitation, evaluating predictors of response to select those patients is a topic for future research.

Acknowledgement

The authors thank the following participating investigators of our study group for scientific advice and collecting data: J. B. V. M. Delemarre[a]; E. van der Harst, P. P. L. O. Coene; E. J. Spillenaar-Bilgen; V. H. van der Vaart; W. F. van Tets; J. J. G. M. Gerritsen; J. W. de Bruijne; M. G. W. Dijkgraaf; R. G. H. Beets-Tan; V. P. M. van der Hulst; T. G. Wiersma; M. N. J. M. Wasser; A. B. Huisman; J.A. de Priester; A. C. Sikkenk; T. D.Witkamp.
[a]Deceased.

References

1. Macmillan AK, Merrie AE, Marshall RJ, et al. The prevalence of fecal incontinence in community dwelling adults: a systematic review of the literature. *Dis Colon Rectum* 2004;47:1341-9
2. Cardozo L, Khoury S, Weiri A (eds) Proceedings of the Second International Consultation on Incontinence. 1-3 July 2001. Plymouth: Health Publication Ltd, 2001
3. Madoff RD, Parker SC, Varma MG, et al. Faecal incontinence in adults. *Lancet* 2004;364:621-32
4. Engel BT, Nikoomanesh P, Schuster MM. Operant conditioning of rectosphincteric responses in the treatment of fecal incontinence. *N Engl J Med* 1974;290:646-9
5. Norton C. Behavioral management of fecal incontinence in adults. *Gastroenterology* 2004;126:S64-70
6. Hosker G, Norton C, Brazzelli M. Electrical stimulation for faecal incontinence in adults. *Cochrane Database Syst Rev* 2000;2:CD001310
7. Norton C, Hosker G, Brazzelli M. Biofeedback and/or sphincter exercises for the treatment of faecal incontinence in adults. *Cochrane Database Syst Rev* 2000;2:CD002111
8. Norton C, Kamm MA. Anal sphincter biofeedback and pelvic floor exercises for faecal incontinence in adults: a systematic review. *Aliment Pharmacol Ther* 2001;15:1147-54
9. Beddy P, Neary P, Eguare EI, et al. Electromyographic biofeedback can improve subjective and objective measures of fecal incontinence in the short term. *J Gastrointest Surg* 2004;8:64-72
10. Fynes MM, Marshall K, Cassidy M, et al. A prospective, randomized study comparing the effect of augmented biofeedback with sensory biofeedback alone on fecal incontinence after obstetric trauma. *Dis Colon Rectum* 1999;42:753-8
11. Lorenz EP, Wondzinski A. Results of conservative and surgical therapy of anal incontinence. 1974 to 1992 patient sample. *Zentralbl Chir* 1996;121:669-75
12. Norton C, Kamm MA. Outcome of biofeedback for faecal incontinence. *Br J Surg* 1999;86:1159-63
13. Jorge JM, Habr-Gama A, Wexner SD. Biofeedback therapy in the colon and rectal practice. *Appl Psychophysiol Biofeedback* 2003;28:47-61
14. Vaizey CJ, Carapeti E, Cahill JA, et al. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
15. Rao SS. Pathophysiology of adult fecal incontinence. *Gastroenterology* 2004;126:S14-22
16. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-80
17. Diamant NE, Kamm MA, Wald A, et al. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-60
18. Isherwood PJ, Rane A. Comparative assessment of pelvic floor strength using a perineometer and digital examination. *BJOG* 2000;107:1007-11
19. Laycock J, Jerwood D. Pelvic floor muscle assessment: The PERFECT scheme. *Physiotherapy.* 2001;87:631-42
20. Norton C, Chelvanayagam S, Wilson-Barnett J, et al. Randomized controlled trial of biofeedback for fecal incontinence. *Gastroenterology* 2003;125:1320-9
21. Baxter NN, Rothenberger DA, Lowry AC. Measuring fecal incontinence. *Dis Colon Rectum* 2003;46:1591-605
22. Deutekom M, Terra MP, Dobben AC, et al. Impact of faecal incontinence severity on health domains. *Colorectal Dis* 2005;7:263-9

23. Deutekom M, Terra MP, Dobben AC, et al. Selecting an outcome measure for evaluating treatment in fecal incontinence. *Dis Colon Rectum* 2005;48:2294-301

24. Bharucha AE. Outcome measures for fecal incontinence: anorectal structure and function. *Gastroenterology* 2004;126:S90-8

25. Naliboff BD. Choosing outcome variables: global assessment and diaries. *Gastroenterology* 2004;126: S129-34

26. Pamuk ON, Pamuk GE, Celik AF. Revalidation of description of constipation in terms of recall bias and visual scale analog questionnaire. *J Gastroenterol Hepatol* 2003;18:1417-22

27. Stone AA, Shiffman S, Schwartz JE, et al. Patient noncompliance with paper diaries. *BMJ* 2002;324:1193-4

28. Bakx R, Sprangers MA, Oort FJ, et al. Development and validation of a colorectal functional outcome questionnaire. *Int J Colorectal Dis* 2005;20:126-36

29. Ryn AK, Morren GL, Hallbook O, et al. Long-term results of electromyographic biofeedback training for fecal incontinence. *Dis Colon Rectum* 2000;43:1262-6

30. Keck JO, Staniunas RJ, Coller JA, et al. Biofeedback training is useful in fecal incontinence but disappointing in constipation. *Dis Colon Rectum* 1994;37:1271-6

31. Mahony RT, Malone PA, Nalty J, et al. Randomized clinical trial of intra-anal electromyographic biofeedback physiotherapy with intra-anal electromyographic biofeedback augmented with electrical stimulation of the anal sphincter in the early treatment of postpartum fecal incontinence. *Am J Obstet Gynecol* 2004;191:885-90

32. Chiarioni G, Bassotti G, Stegagnini S, et al. Sensory retraining is key to biofeedback therapy for formed stool fecal incontinence. *Am J Gastroenterol* 2002;97:109-17

33. Patankar SK, Ferrara A, Larach SW, et al. Electromyographic assessment of biofeedback training for fecal incontinence and chronic constipation. *Dis Colon Rectum* 1997;40:907-11

34. Rao SS, Welcher KD, Happel J. Can biofeedback therapy improve anorectal function in fecal incontinence? *Am J Gastroenterol* 1996;91:2360-6

35. Glia A, Gylin M, Akerlund JE, et al. Biofeedback training in patients with fecal incontinence. *Dis Colon Rectum* 1998;41:359-64

36. Van Tets WF, Kuijpers JH, Bleijenberg G. Biofeedback treatment is ineffective in neurogenic fecal incontinence. *Dis Colon Rectum* 1996;39:992-4

37. Leroi AM, Dorival MP, Lecouturier MF, et al. Pudendal neuropathy and severity of incontinence but not presence of an anal sphincter defect may determine the response to biofeedback therapy in fecal incontinence. *Dis Colon Rectum* 1999;42:762-9

38. Rieger NA, Wattchow DA, Sarre RG, et al. Prospective trial of pelvic floor retraining in patients with fecal incontinence. *Dis Colon Rectum* 1997;40:821-6

Functional changes after pelvic floor rehabilitation in fecal incontinence

Annette C. Dobben
Maaike P. Terra
Bary Berghmans
Marije Deutekom
Guy E. E. Boeckxstaens
Lucas W. M. Janssen
Patrick M. M. Bossuyt
Jaap Stoker

Int J Colorectal Dis. 2006 Sep;21(6):515-21

Chapter 7

Abstract

BACKGROUND. Pelvic floor rehabilitation is a common treatment option in patients with fecal incontinence. Although pelvic floor rehabilitation may result in relief of symptoms, to what extent improvement is associated with changes in anorectal function is still unclear.

AIM. The aim of the present study was to investigate prospectively how anorectal function changes with pelvic floor rehabilitation and whether these changes are related to changes in fecal incontinence score.

METHODS. Consenting consecutive patients (n=266) with fecal incontinence (91% women; mean age, 59 years) underwent anorectal manometry, anal and rectal mucosal sensitivity measurements, and rectal capacity measurement at baseline and after nine sessions of standardized pelvic floor pelvic floor rehabilitation. These findings were compared with changes in Vaizey incontinence score.

RESULTS. On follow-up 3 months after pelvic floor rehabilitation, squeeze pressure (p=0.028), as well as urge sensation threshold (p=0.046) and maximum tolerable volume (p=0.018), had increased significantly. The extent of improvement was not related to age, duration of fecal incontinence, menopause, and endoultrasonography findings. All other anorectal functions did not change. An improvement in the Vaizey score was moderately correlated with an increase in incremental squeeze pressure (r=0.14, p=0.04) and a decrease in anal mucosal sensitivity threshold (r=0.20, p=0.01).

CONCLUSIONS. Pelvic floor rehabilitation improves squeeze pressure, urge sensation, and maximum tolerable volume. However, improved anorectal function does not always result in a decrease in fecal incontinence complaints.

Introduction

Fecal incontinence is defined as "the involuntary loss of flatus, liquid or solid stool that is a social or hygienic problem" [1]. Reported prevalence values range from 1.4% in the general population (defined as soiling of underwear, outer clothing, furnishing, or bedding for several times a month or more often)[2] to 46% in institutionalized elderly people (defined as at least one incontinent episode per week)[3, 4]. The real prevalence is possibly even higher than reported [5], as anal incontinence is associated with high social stigma and, therefore, people do not easily seek help for this disorder because of embarrassment[6, 7]. Fecal incontinence has been classified clinically into three specific types: urge incontinence (unwanted loss of stool despite active attempts to inhibit defecation), passive incontinence (unwanted loss of stool without patient awareness), or a combination of urge and passive incontinence[8].

Initial treatment options for fecal incontinence are conservative and include medication and dietary measures. In case of failure, pelvic floor pelvic floor rehabilitation is an additional conservative therapy[9]. A combination of pelvic floor muscle exercises and anal sphincter exercises with biofeedback, originally based on the principles of operant conditioning, is a common clinical treatment for fecal incontinence, although reports on the contribution of biofeedback to subjective and objective improvements have shown contradictory results[10–15]. Some have suggested that sphincter exercises with biofeedback therapy have a therapeutic effect[10]. Others have referred to the effects of expectancy (placebo response) [16] and the crucial role of patient motivation and patient–therapist interaction[10].

To what extent biofeedback influences objective measures is unclear. Objective changes can be evaluated with anorectal function testing, including anorectal manometry, anal and rectal mucosal sensitivity measurements, and rectal capacity measurement [17–20]. Bharucha[21] has summarized the results of 19 studies on the effects of biofeedback therapy on anal sphincter pressures and rectal sensation in adults with fecal incontinence. Although resting and squeeze pressures increased by varying degrees after biofeedback in some studies, the magnitude of improvement was relatively small and did not correlate with symptom improvement. However, the patient groups studied were quite small and differed in patient characteristics as well as in duration of biofeedback therapy. Therefore, we extensively documented various anorectal function changes 3 months after standardized pelvic floor rehabilitation in a large group of selected patients with fecal incontinence. Pelvic floor rehabilitation consisted of pelvic floor muscle training combined with biofeedback and electrical stimulation. We also examined whether anorectal function changes are associated with age, duration of fecal incontinence, gender, menopause, imaging findings, or other predisposing factors. Furthermore, we evaluated whether such changes are related to changes in incontinence severity score.

Materials and Methods

Patients

This study, which was conducted between December 2001 and March 2005 in 16 medical centers in the Netherlands, evaluated pelvic floor rehabilitation performed on a large group of fecal incontinent patients. This study was approved by the medical ethics committees of all participating centers. Inclusion criteria were as follows: existence of fecal incontinence complaints for 6 months or more, a Vaizey incontinence score of at least 12[22], and failure of conservative treatment, based on dietary recommendations and/or antidiarrheals. Criteria for exclusion of patients were as follows: age below 18 years, diagnosis of anorectal tumor less than 2 years ago, and previous ileoanal or coloanal anastomosis. As this study investigated any treatment effect of pelvic floor rehabilitation, patients with chronic diarrhea (always fluid stools, three or more times a day), overflow incontinence, proctitis, soiling (leakage of fecal material out of the anus after normal defecation, leading to perineal eczema), and rectal prolapse were also excluded. Besides, we chose not to include patients who could not understand the requirements for participation in this study and those who were unlikely to cooperate. Consenting consecutive patients were included.

Evaluation of the severity of symptoms as well as possible predisposing factors for fecal incontinence was performed by a participating clinician. The severity of fecal incontinence was assessed according to the grading system of Vaizey et al.[22]. This grading system contains items on the type (gas, fluid, and solid) and frequency of fecal incontinence and additional items addressing alterations in lifestyle, the need to wear a pad or a plug, use of constipating medications, and the presence of urge incontinence. The total score on the Vaizey scale ranges from 0 (complete continence) to 24 (complete incontinence).

Anorectal function tests

All diagnostic tests were performed by specialized physicians or technicians in seven medical centers out of all participating hospitals. Prior to testing, patients received standard written information concerning the tests. All tests were performed in the left lateral position, with hips flexed to 90°.

Anorectal manometry was performed according to solidstate technique or water perfusion technique, depending on the preference or experience of the hospital. The solidstate technique was performed in three centers, using a Konigsberg catheter (Konigsberg Instrument, Inc., Pasadena, CA) connected to a computer-assisted polygraph (Synectics Medical, Stockholm, Sweden). The water perfusion technique was performed in the remaining centers: without sleeve in three centers, using a Zinetics Catheter (Medtronic, Skolvunde, Denmark) connected to a perfusion pump (Dentsleeve Pty Ltd, Adelaide, Australia), or with sleeve in one center, using a multilumen waterperfused sleeve catheter assembly and a terminal inflatable balloon (Dentsleeve Pty Ltd, Parkside, Australia) connected to a polygraph (Synectics Medical). With the solid-state method or with the four-channel water perfusion method without sleeve, maximum anal sphincter pressure was determined by means of a pullthrough technique. Next, the position of the microtransducer was placed at the level of the maximum pressure zone. With the water perfusion technique with sleeve (which was positioned in the anal canal), recordings were made with a 4-cm-long sleeve and five radially distributed side holes. Each side hole was perfused with degassed water at a rate of 0.3 ml/min, and intraluminal pressures were sensed by external transducers. After insertion of the catheter, the recordings were allowed to stabilize in the anal canal for 5 min, after which the mean value of the resting pressure was measured for 1 min. To measure external anal sphincter function, patients were instructed to maximally squeeze on three occasions, at 1 min intervals. The average maximum squeeze pressure (mmHg) and the mean incremental squeeze pressure (maximal squeeze pressure minus resting pressure; mmHg) were calculated. In addition, patients were asked to maximally strain on three occasions, constantly at 1 min intervals. For consistency, the third maximal straining was used for analyses.

Defecation index was calculated as the ratio of rectal pressure to residual anal pressure[23]. Subsequently, the reflectory external anal sphincter function was measured by asking the patient to cough. The difference between rectal pressure and anal pressure was determined (mmHg).

To assess rectal–anal inhibitory reflex (RAIR), the terminal balloon was inflated with increasing volumes (10–50 ml). RAIR was defined as "present" when a balloon volume $\leq$50 ml induced a reduction of internal anal sphincter pressure of at least 10 mmHg for 5 s. If an inflation of more than 50 ml was needed to reduce the internal anal sphincter pressure, RAIR was defined as "absent."

With rectal and anal mucosal sensitivity measurements, the threshold sensations of the rectum and the anus, respectively, were determined. A ring electrode (Dantec Keypoint, Skovlunde, Denmark) was inserted into the rectum approximately 10 cm above the anal verge to measure rectal mucosal sensitivity. This stimulation electrode was mounted on a Foley catheter, and the current was increased gradually (up to a maximum of 20 mA) until patients reported some sensation. To determine the threshold, the lowest of three consecutive sensations was used. The electrode was then positioned into the middle anal canal, and the same procedure was performed in the anus to estimate anal mucosal sensitivity threshold.

7

With rectal capacity measurement, the reservoir capacity of the rectum can be determined. This comprises the capacity to temporarily store feces as well as the accurate sense of fullness of the rectum. The capacity measurement of the rectum was performed by introducing a single-use urinary catheter (female catheter, 14 Ch) with a latex balloon tied to the end, which was then covered with a lubricant and connected to a 50 ml syringe. The balloon catheter was inflated with air with slow increments of 50 ml until the maximum tolerable volume was reached. The minimal rectal sensation perceived (sensory threshold), the volume associated with the initial urge to defecate (urge sensation), and the volume at which the patient experienced discomfort and an intense desire to defecate (the maximum tolerable volume) were determined. Only in one center was rectal capacity measurement performed with a barostat procedure. A compliant polyethylene barostat bag was fastened to one side of a polyvinyl catheter. The catheter was linked to a strain gauge and a computer-controlled air injection system (G&J Electronics Inc., Ontario, Canada). With the patient in the left lateral position, the bag was inserted into the rectum, 10 cm from the anal canal. Before each measurement, approximately 100 ml of air was injected and aspirated from the bag to unfold it. Next, the bag was inflated with air to a selected pressure plateau (range, 0–60 mmHg). Subjects were instructed to report the times when they experienced the first sensation, urge, and maximum tolerable volume.

To define the anatomic defects of the external and/or internal anal sphincter, endoanal ultrasonography was performed using an scanner with a radial endoscopic probe (a 7.5- or 10-MHz transducer)[17]. The endoscopic probe was introduced into the anus up to the anorectal verge and then slowly withdrawn. The integrity of the external and internal anal sphincters was assessed.

After assessment of fecal incontinence severity and performance of anorectal function tests, the patients were referred for pelvic floor rehabilitation.

Pelvic floor rehabilitation

Pelvic floor rehabilitation was administered by 71 specialized pelvic physiotherapists according to a standardized protocol. Patients underwent nine sessions on a weekly basis. Specialized pelvic floor rehabilitation comprised rectal balloon training and electromyographic feedback, unless puborectal muscle strength and/or external anal sphincter muscle strength was <3, according to the Oxford scoring system[24, 25]. The Oxford score ranges from 0 (no muscle contraction) to 5 (strong contraction). When the Oxford score of the puborectal muscle strength and/or the external anal sphincter muscle strength was <3, patients received electrical stimulation and rectal balloon training. Electrical stimulation was offered until patients were able to independently develop enough puborectal muscle strength and external anal sphincter strength (i.e., muscle strength ≥3). Rectal balloon training was performed with a condom attached to a 50 ml syringe, which was introduced into the rectum and slowly inflated with air. Patients with an insensitive rectum or a hypersensitive rectum were taught to perceive smaller or larger volumes of distension, respectively. Sensory threshold, urge sensation, and maximum tolerable volume were assessed during each session. Electromyographic feedback was performed with an intra-anal electrode (NEEN; PelviTec BV, Delft, the Netherlands) attached to a monitor (Myomed 932; Enraf- Nonius NV, Delft, the Netherlands). Patients were trained on contraction, including duration, relaxation, timing, and coordination of pelvic floor muscles. The patients were instructed and encouraged to perform standardized exercises at home.

Approximately 3 months after pelvic floor rehabilitation, the severity of fecal incontinence was determined by means of the Vaizey score, and anorectal function tests were repeated according to the procedure performed at baseline.

Statistical considerations

Changes from baseline in average anorectal function after pelvic floor rehabilitation were calculated and tested for significance using Student’s t test for paired data. To investigate differences between anorectal manometric techniques, we used analysis of variance. For post hoc analyses, we used Bonferonni corrections.

We used Pearson’s correlation coefficients to compare changes in anorectal function after treatment with changes in Vaizey score from baseline. We compared changes in anorectal function in subgroups defined by age (<35, 35–<50, 50–65, and >65 years), duration of fecal incontinence (≤5 years and >5 years), anal sphincter defects, and other predisposing factors using analysis of variance. To assess changes in RAIR, urge incontinence, and passive incontinence before and after pelvic floor rehabilitation, we used the McNemar test.

For all statistical tests, $p<0.05$ was considered to indicate statistical significance. We used SPSS for Windows (version 11.5, 2002) to perform statistical analyses of our data.
All data were checked by double data entry for validation.

Results

Between December 2001 and March 2005, 323 consenting eligible patients were included in this study. We had to exclude 57 patients from data analysis due to inadequate follow-up period

Table 1. Clinical characteristics of patients of enrolled patients

Patient characteristics	
Age (mean) (SD)	59 (13)
Vaizey incontinence score (mean) (SD)	18 (3)
Passive incontinence (%)	10 (4)
Urge incontinence (%)	102 (40)
Combined urge and passive incontinence (%)	143 (56)
Duration of fecal incontinence (median years) (range)	5 (0.5-57)
Female (%)	241 (91)
Menopausal women (%)	179 (74)
Findings at endoanal ultrasonography	
No defect (%)	91 (38)
EAS defect (%)	76 (32)
IAS defect (%)	9 (4)
Combined EAS and IAS defect (%)	62 (26)
Predisposing conditions	
No evidently predisposing condition (%)	26 (10)
Anatomic congenital (%)	1 (0.4)
Anatomic traumatic (%)	
Obstetric injury	199 (75)
Colorectal surgery	74 (28)
Gynecological surgery	104 (39)
Urological surgery	51 (19)
Functional (%)	
Neurological	34 (13)
Metabolic	34 (13)
Inflammatory bowel diseases	9 (3)
Systemic disorders	10 (4)

Note: Predisposing conditions are not mutually exclusive. Per patient various conditions may exist.

(n=16) or dropout (n=41), leaving 266 cases available for analysis, of which 241 were women (91%). The demographic data and clinical details of these patients are shown in Table 1.

Changes in anorectal function after specialized pelvic floor rehabilitation

Anorectal manometry was performed using three different techniques, whereas rectal capacity measurement was performed using two different techniques. There were no significant differences between techniques in the changes of anorectal function (data not shown).

Changes after treatment are summarized in Table 2. The mean (±SD) squeeze pressure increased significantly from 85.3 (±39) to 91.2 (±44) mmHg (p=0.028). The resting pressure and the incremental squeeze pressure did not change significantly after pelvic floor rehabilitation. No differences in rectoanal coordination were found. RAIR was present in 206 (89%) patients before and after therapy.

Both anal and rectal mucosal sensitivity thresholds did not change. The mean (±SD) urge sensation increased from 94 (±50) to 100.4 (±50) ml (p=0.046), and the mean (±SD) maximum tolerable volume increased from 158.3 (±70) to 169 (±66) ml (p=0.018). Sensory threshold did not change after therapy.

Patient age, duration of fecal incontinence, menopause, endoanal ultrasonography findings, and other predisposing conditions were not associated with anorectal function changes. The

Table 2. Changes in anorectal function after physiotherapy

Anorectal Manometry (mmHg)	**Overall (n=248) Baseline mean (SD)**	**After therapy mean (SD)**	**mean change (95%CI)**	**p***	**r****	**P*****
Resting pressure	48.5 (22)	50.8 (24)	2.3 (−0.4 to 5.1)	0.09	0	0.9
Squeeze pressure	85.3 (39)	91.2 (44)	5.9 (0.7 to 11)	**0.03**	-0.12	0.1
Incremental squeeze pressure	36.9 (34)	40.4 (35)	3.5 (-0.7 to 7.9)	0.1	0.14	**0.04**
Coughing rectal - anal differences	20.3 (40)	23.9 (39)	3.6 (-2.6 to 9.9)	0.26	0.04	0.8
Defecation index	1.0 (0.8)	0.9 (0.8)	-0.1 (-0.27 to 0.07)	0.26	0	0.9
Sensitivity Measurement (mAmp)						
Anal sensitivity	7.8 (5.7)	7.2 (4.9)	-0.6 (-1.5 to 0.3)	0.2	0.20	**0.01**
Rectal sensitivity	28 (18)	26.2 (16)	-1.8 (-4 to 0.4)	0.1	0.04	0.6
Rectal Capacity Measurement (ml)						
Sensory threshold	50.7 (34)	52.7 (34)	2 (-2.9 to 6.9)	0.4	0.1	0.1
Urge	94 (50)	100.4 (50)	6.4 (0.13 to 12.7)	**0.05**	0.06	0.4
Maximum tolerated volume	158.3 (70)	169 (66)	10.7 (1.8 to 19.5)	**0.02**	0.02	0.8

Note:* p-values calculated for change in function (paired data).
** correlation between changes in Vaizey incontinence score and changes in anorectal function.
*** p-values of correlated data.

sensory threshold increased significantly more in men than in women (mean change (±SD), 22 (±50) vs 0.13 (±34) ml; p=0.014) after therapy.

In patients without passive incontinence, both incremental squeeze pressure (mean difference (±SD), 4 (±29) vs 21 (±49) mmHg; p=0.018) and rectoanal coordination (mean difference (±SD), 5 (±39) vs 25 (±53); p=0.04) improved significantly when they were instructed to cough.

Passive incontinent patients had a significant decrease in the anal mucosal sensitivity threshold after therapy, compared to patients without passive incontinence (mean change (±SD), 5.6 (±6) vs 0.3 (±6) mA; p=0.036).

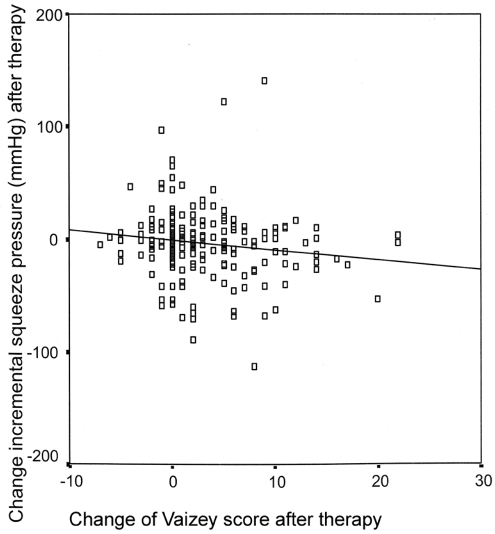


Figure 1. Change in Vaizey score compared to change in incremental squeeze pressure after pelvic floor rehabilitation

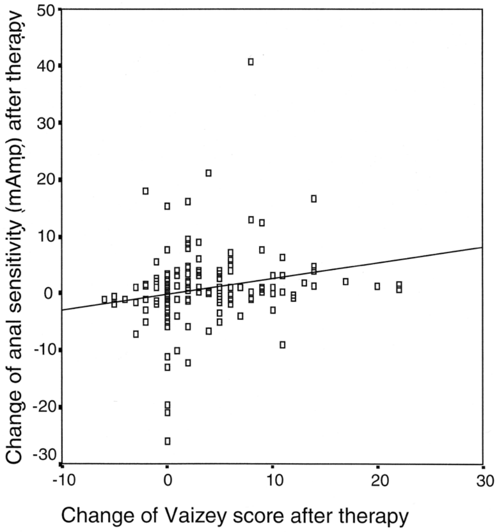


Figure 2. Change of Vaizey score compared to change in anal mucosal sensitivity after pelvic floor rehabilitation

Clinical outcome

Approximately 3 months after treatment (median number weeks, 10), the average Vaizey score had decreased from 18 to 15 ($p<0.001$). Urge incontinence was observed in 66 patients (27%) after pelvic floor rehabilitation, compared to 102 patients (40%) at baseline ($p<0.001$). The frequencies of passive incontinence and combined passive/urge incontinence had not changed (data not shown). A reduction of the Vaizey score by at least 50% was obtained by 34 patients (14%).

Relationship between clinical outcome and changes in anorectal function

Improvement in the Vaizey score was significantly correlated with an increase in incremental squeeze pressure ($r=0.14$, $p=0.04$) (Fig. 1) and a decrease in anal mucosal sensitivity threshold ($r=0.20$, $p=0.01$) (Fig. 2). Both correlations are rather low, explaining less than 2% of the variance. More details on changes in anorectal function and their relationship with clinical outcome are found in Table 2.

Discussion

This study shows that pelvic floor rehabilitation produces significant but small changes in squeeze pressure, urge sensation, and maximum tolerable volume, but not in other anorectal functions. Improvement in the Vaizey score is associated with incremental squeeze pressure and anal mucosal sensitivity measurement.

In this study, we extensively evaluated a large group of fecal incontinent patients with mixed etiology. To our knowledge, anorectal function tests in such a large group of fecal incontinent patients have not been evaluated before. We did not use a control group and were therefore not able to evaluate specific therapy-related effects. Whether or not (and to what extent) specific and nonspecific effects influence outcome can only be speculated. Furthermore, the large sample size of our cohort might explain the statistically significant difference.

One hypothesis is that the incontinence score is a subjective outcome measure, whereas changes in function are objective measures. Several incontinence scores have been developed[6, 19, 22]. However, the Vaizey scoring system is the most complete[22]. A previous study demonstrated that this scoring system was reproducible and correlated well with physicians' clinical impression[22, 26]. Despite this fact, subjective elements might have introduced bias into the outcome.

We decided to work with specialized physiotherapists only. These physiotherapists were trained before the start of the study and performed their treatment according to a standardized protocol to which they adhered. Tailoring the number of sessions to patients' requirements may result in more pronounced effects[9, 12].

In this study, the squeeze pressure improved whereas the resting pressure did not change. This implies that the therapy improved the contractile force of the striated muscles of the external anal sphincter, but had little effect on the involuntary smooth muscles of the internal anal sphincter[6].

To our knowledge, this is the first study to evaluate changes in anal and rectal mucosal sensitivity after pelvic floor rehabilitation. Rao[27] noticed the importance of quantitative assessment of anal perception using electrical stimulation, as sampling of rectal contents in the anal mucosa may play an important role in maintaining continence. Felt-Bersma et al.[28] showed that the anal mucosal sensitivity threshold in fecal incontinent patients was significantly higher than that of controls, but they concluded that anal sensitivity measurement was not a crucial tool in daily clinical management. Our study shows no change in anal and rectal mucosal sensitivity threshold after therapy. The correlation between an improvement in Vaizey score and a decrease in the anal mucosal sensitivity threshold was, although significant, weak and unlikely to be of clinical relevance. The correlation between a decrease in Vaizey score and an increase in incremental squeeze pressure was even smaller. Consequently, it is difficult to explain the statistically significant difference in Vaizey score after therapy by changes in function only.

We found that the urge sensation threshold increased significantly. Our study population is composed mainly of patients with urge incontinence (40%) or with combined passive/urge incontinence (56%). An increase in the urge sensation threshold might explain the significant improvement in urge incontinence ($p<0.001$), as these patients have a hypersensitive rectum and have learned to suppress feelings of urgency in response to rectal filling up to the normal level of urge sensation.

Prather[29] concludes that a lower maximum tolerable volume is a predictor of poor outcome. Our study showed a significant improvement in the maximum tolerable volume, but we found no significant correlation between fecal incontinence score and the maximum tolerable volume. Hence, it seems that an improved maximum tolerable volume does not obviously result in a decrease in fecal incontinence complaints.

Differences in age, duration of fecal incontinence, menopause, and endoanal ultrasonography findings did not modify anorectal function changes. Patients with passive incontinence appeared to have no improvement in incremental squeeze pressure; thus, squeeze training seemed to be less effective in this group.

In this study, we evaluated only the short-term outcomes of pelvic floor rehabilitation. We realized that beneficial effects may not persist or that physiological changes may take longer to become apparent and that, therefore, long-term evaluation is required. We found improved squeeze pressure, urge sensation, and maximum tolerable volume. An improved squeeze pressure is in line with findings in other studies[11, 12, 14]. The clinical significance of urge sensation and maximum tolerable volume is less well-established[17].

The most pertinent issue is whether improvement in anorectal function leads to improvement in fecal continence. The results of this study show that such an association exists, but that it is rather weak, partially due to the relatively small size of the average improvement. We therefore conclude that, after a short-term follow-up, nine sessions of standardized pelvic floor rehabilitation will result in minor improvement in the fecal incontinence score and in anorectal function for most patients.

References

1. Cardozo L, Khoury S, Weiri A (eds) Proceedings of the Second International Consultation on Incontinence. 1-3 July 2001. Plymouth: Health Publication Ltd, 2001
2. Perry S, Shaw C, McGrother C et al. Prevalence of faecal incontinence in adults aged 40 years or more living in the community. *Gut* 2002;50:480-484
3. Borrie MJ, Davidson HA. Incontinence in institutions: costs and contributing factors. *CMAJ* 1992;147:322-328
4. Johanson JF, Irizarry F, Doughty A. Risk factors for fecal incontinence in a nursing home population. *J Clin Gastroenterol* 1997;24:156-160
5. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *Lancet* 2004;364(9434):621–632
6. Mavrantonis C, Wexner SD. A clinical approach to fecal incontinence. *J Clin Gastroenterol* 1998;27:108-121
7. Jorge JM, Wexner SD. Etiology and management of fecal incontinence. *Dis Colon Rectum* 1993;36:77-97
8. Cheetham MJ, Malouf AJ, Kamm MA. Fecal incontinence. *Gastroenterol Clin North Am* 2001;30:115-130
9. Kamm MA. Faecal incontinence. *BMJ* 1998;316:528-532
10. Norton C, Hosker G, Brazzelli M. Biofeedback and/or sphincter exercises for the treatment of faecal incontinence in adults. *Cochrane Database Syst Rev* 2000(2):CD002111
11. Rao SS, Welcher KD, Happel J. Can biofeedback therapy improve anorectal function in fecal incontinence? *Am J Gastroenterol* 1996;91:2360-2366
12. Ozturk R et al. Long-term outcome and objective changes of anorectal function after biofeedback therapy for faecal incontinence. *Aliment Pharmacol Ther* 2004;20:667-674
13. Sangwan YP, Coller JA, Barrett RC et al. Can manometric parameters predict response to biofeedback therapy in fecal incontinence? *Dis Colon Rectum* 1995;38:1021-1025
14. Martinez-Puente Mdel C, Pascual-Montero JA, Garcia-Olmo D. Customized biofeedback therapy improves results in fecal incontinence. *Int J Colorectal Dis* 2004;19:210-214
15. Chiarioni G, Scattolini C, Bonfante F, Vantini I. Liquid stool incontinence with severe urgency: anorectal function and effective biofeedback treatment. *Gut* 1993;34:1576-1580
16. Chiarioni G, Bassotti G, Stegagnini S et al. Sensory retraining is key to biofeedback therapy for formed stool fecal incontinence. *Am J Gastroenterol* 2002;97(1):109-117
17. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-760
18. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. Am *J Gastroenterol* 2000;95:1873-1880
19. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-1685
20. Rao SS, Patel RS. How useful are manometric tests of anorectal function in the management of defecation disorders? *Am J Gastroenterol* 1997;92:469-475
21. Bharucha AE. Outcome measures for fecal incontinence: anorectal structure and function. *Gastroenterology* 2004;126(Suppl 1):S90-S98
22. Vaizey CJ et al. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80

23. Rao SS, Welcher KD, Leistikow JS. Obstructive defecation: a failure of rectoanal coordination. *Am J Gastroenterol* 1998;93:1042-1050

24. Isherwood PJ, Rane A. Comparative assessment of pelvic floor strength using a perineometer and digital examination. *BJOG* 2000;107:1007-1011

25. Laycock J, Jerwood D. Pelvic floor muscle assessment: the perfect scheme. *Physiotherapy* 2001;87:631-642

26. Baxter NN, Rothenberger DA, Lowry AC. Measuring fecal incontinence. *Dis Colon Rectum* 2003;46:1591-1605

27. Rao SSC. Diagnosis and management of fecal incontinence. American College of Gastroenterology Practice Parameters Committee. *Am J Gastroenterol* 2004;99:1585-1604

28. Felt-Bersma RJ, Poen AC, Cuesta MA, Meuwissen SG. Anal sensitivity test: what does it measure and do we need it? Cause or derivative of anorectal complaints. *Dis Colon Rectum* 1997;40:811-816

29. Prather CM. Predictors of outcome in treatment trials. *Gastroenterology* 2004;126:S135-S140

Chapter 8

Can the outcome of pelvic floor rehabilitation in patients with fecal incontinence be predicted?

Maaike P. Terra
Marije Deutekom
Annette C. Dobben
Cor G.M.I. Baeten
Lucas W.M. Janssen
Guy E.E. Boeckxstaens
Alexander F. Engel
Richelle J.F. Felt-Bersma
J. Frederik M. Slors
Michael F. Gerhards
A. Bart Bijnen
Ellen Everhardt
W. Ruud Schouten
Bary Berghmans
Patrick M.M. Bossuyt
Jaap Stoker

Submitted

Abstract

PURPOSE. Pelvic floor rehabilitation does not provide the same degree of relief in all fecal incontinent patients. We aimed at studying prospectively the ability of tests to predict the outcome of pelvic floor rehabilitation in patients with fecal incontinence.

MATERIALS AND METHODS. 250 consecutive patients (228 women) underwent medical history and a standardized series of tests, including physical examination, anorectal manometry, pudendal nerve latency testing, anal sensitivity testing, rectal capacity measurement, defecography, endoanal ultrasonography, and endoanal MR imaging. Subsequently, patients were referred for pelvic floor rehabilitation. Outcome of pelvic floor rehabilitation was quantified by the Vaizey incontinence score. Linear regression analyses were used to identify candidate predictors and to construct a multivariable prediction model for the post-treatment Vaizey score.

RESULTS. After pelvic floor rehabilitation the mean baseline Vaizey score (18; SD ± 3) was reduced with 3.2 points ($p < 0.001$). In addition to the baseline Vaizey score, six elements from medical history were significantly associated with the post-treatment Vaizey score (R^2:0.20). The predictive value was significantly but marginally improved by adding the following test results: perineal and/or perianal scar tissue (physical examination), maximal squeeze pressure (anorectal manometry) and internal anal sphincter atrophy (endoanal MR imaging) (R^2:0.23; $p = 0.02$).

CONCLUSION. Additional tests have a limited role in predicting success of pelvic floor rehabilitation in patients with fecal incontinence.

Introduction

Fecal incontinence is a common[1, 2] disabling condition that affects the lifestyle of patients[3]. Continence is a multi-factorial mechanism, requiring an intact chain of anatomical structures and physiological mechanisms[4]. Fecal incontinence is primarily caused by anal sphincter defects, neuropathy, reduced rectal capacity and compliance, or a combination of these factors[4].

There is a wide variety of treatment options available for patients with fecal incontinence ranging from conservative therapy (dietary measures (fibres, avoidance of foods that cause diarrhoea or urgency), medical treatment (anti-diarrhoeas medications, bulking agents), pelvic floor rehabilitation) to surgical intervention[4]. Biofeedback and electrical stimulation are both pelvic floor rehabilitation techniques commonly used in patients with fecal incontinence. The outcome of these treatment modalities alone or in combination has been extensively evaluated, leading to a wide range of reported success rates[5-8]. Some of this variability can be explained by between study differences in patient selection, methodology, biofeedback and/or electrical stimulation techniques used, outcome measurements, criteria for success and duration of follow-up[4, 9-11]. A recent study evaluating pelvic floor rehabilitation (pelvic floor muscle training with biofeedback and electrical stimulation) in a large population with fecal incontinence due to different etiologies, demonstrated that pelvic floor rehabilitation provides a “slight” relief of fecal incontinence complaints (a reduction in Vaizey score of < 50%) in the majority of patients and a “substantial” relief (a reduction in Vaizey score of ≥ 50%) in a minority only[12]. Identification of factors predictive of the response to pelvic floor rehabilitation would be helpful in selecting patients for pelvic floor rehabilitation and counselling patients on the likely outcome of pelvic floor rehabilitation[13, 14]. To select patients who may benefit from pelvic floor rehabilitation an accurate evaluation of the underlying pathophysiology and an understanding of the likely cause of fecal incontinence are crucial[13, 14].

Additional to medical history, several tests can be used to assess patients with fecal incontinence, including physical examination, anorectal function tests and imaging techniques[4, 15, 16]. Up till now, there is no consensus regarding which tests should be performed in patients with fecal incontinence and what their utility is in selecting patients for pelvic floor rehabilitation[13, 14, 17].

The purpose of this study was to prospectively determine the value of tests, in isolation and in combination, to predict the outcome of pelvic floor rehabilitation in a large series of patients with fecal incontinence due to different etiologies.

Materials and Methods

Patients

This prospective study was performed between December 2001 and April 2005 in 16 medical centers in the Netherlands. The Medical Ethics Committees of all hospitals approved the study.

Consecutive patients with fecal incontinence were invited. Inclusion criteria were fecal incontinence complaints for at least six months, a Vaizey incontinence score of at least 12[18] and failure of conservative treatment (including diet measurements and medication). Patients under eighteen, patients diagnosed with an anorectal tumor, patients with chronic diarrhoea (always fluid stools, three or more times a week), overflow incontinence, proctitis, soiling (leakage of fecal material out of the anus after normal defecation often leading to perineal eczema), previous ileoanal or coloanal anastomosis and rectal prolapse (intussusception grade three, circular invagination of the proximal rectal wall during defecation extending beyond the anal opening) were excluded, as were patients who had received pelvic floor rehabilitation in the previous six months. Patients who were considered to be unable to undergo pelvic floor rehabilitation because of limited comprehension or intellectual capacity were also excluded.

Eligible patients were asked for signed informed consent. Participating patients underwent medical history and a standardized series of tests, consisting of physical examination, a set of anorectal function tests and imaging techniques. As not all participating centers were well equipped to perform each of the anorectal function tests and/or imaging techniques, patients from these centers were referred to one of the other participating centers to undergo the specific examinations. After testing all patients were referred for a standardized pelvic floor rehabilitation program. The outcome of pelvic floor rehabilitation will be reported elsewhere. This study focuses on the predictive value of medical history and additional tests.

Medical history

Medical history was obtained by physicians and included duration, type, and degree of fecal incontinence, as well as bowel habits and likely underlying causes for fecal incontinence. All participating physicians used the same structured forms to obtain information from medical history. The type of incontinence was divided in passive incontinence (defecation loss without the patient's knowledge) and urge incontinence (unwanted loss of stool despite active attempts to inhibit defecation)[14, 19]. Degree of fecal incontinence was assessed according to the grading system of Vaizey[18]. This grading system contains several quantitative and qualitative incontinence-specific items and the total score ranges from 0 (complete continence) to 24 (complete incontinence). Bowel habits comprised frequency of defecation (≤ 7/week or > 7/week), stool consistency (thin, soft mushy, solid, firm, varying) and sensation of incomplete evacuation (< 1/week or ≥ 1/week). The likely underlying causes for fecal incontinence were divided in relevant subgroups reflecting the whole spectrum of causes of fecal incontinence (Table 1).

Additional tests

All tests were performed by specialized physicians or technicians according to a standard procedure that had been established during joined meetings of the research group members of all participating hospitals.

Physical examination

Physical examination comprised inspection of the perineum and perianal area for presence of scar tissue and digital rectal examination[16]. Digital rectal examination assessed the resting pressure and squeeze pressure (inadequate (absent or decreased) or adequate (normal)) of the anal sphincter complex, as well as the presence of an anal sphincter defect.

Table 1. Baseline characteristics and candidate predictors from medical history

Medical history		ß	p
Vaizey score at baseline (points) (± SD) *	18 (± 3)	0.61	.00 *
Gender (female) *	228 (91%)	-1.3	0.16 *
Age (y) (± SD)	59 (± 13)	-0.03	0.23
Duration of fecal incontinence (y) (± SD) *	8 (± 9)	-0.05	0.19 *
Presence of urge incontinence	241 (96%)	1.67	0.35
Presence of passive incontinence *	145 (58%)	1.54	0.02 *
Frequency defecation (< 7 times/week)	189 (76%)	0.44	0.57
Sensation of incomplete evacuation (≥ 1/week) *	142 (57%)	1.11	0.10 *
Thin stool consistency *	9 (4%)	4.31	0.01 *
Soft mushy stool consistency	79 (32%)	0.16	0.82
Solid stool consistency *	57 (23%)	-1.2	0.12 *
Firm stool consistency	9 (4%)	1.55	0.37
Varying stool consistency	90 (36%)	-0.21	0.75
Rupture after vaginal delivery repaired at childbed *	84 (34%)	1.75	0.01 *
Rupture after vaginal delivery repaired at operating room	31 (12%)	-0.38	0.7
Any obstetric risk factor (e.g., high-birth-weight infant, long second stage of labour, instrumental delivery)	190 (76%)	-2.58	0.74
Any ano-and colorectal risk factor (e.g., surgery for anal fistulas, anal fissures, hemicolectomy)	69 (28%)	0.48	0.52
Any gynecological risk factor (e.g., hysterectomy) *	99 (40%)	1.03	0.12 *
Any urological risk factor (e.g., Burch operation)	47 (19%)	0.77	0.36
Any neurological risk factor (e.g., cerebral and spinal cord disorders)	32 (13%)	-0.54	0.58
Any metabolic risk factor (e.g., diabetes mellitus, thyroid disorders)	33 (13%)	0.44	0.65
Any fecal consistency risk factor (e.g., diverticulitis)	9 (4%)	1.53	0.39

Note: Unless otherwise indicated, data are the number of patients, * indicates p-value below 0.2 (i.e., candidate predictor).

8

Anorectal function tests

Anorectal manometry

Anorectal manometry took place according to the solid-state or water perfused technique without or with sleeve[16]. The solid-state method or water-perfusion method without sleeve was performed by means of a pull-through technique. The catheter (Konigsberg Instrument Inc., Pasadena, CA; Medtronic, Skolvunde, Denmark; Dentsleeve Pty Ltd, Parkside, Australia) was inserted in the anal canal and the (mean) maximal resting pressure (mmHg) was measured. Subsequently, the (mean) maximal squeeze pressure (mmHg) was determined by asking patients to squeeze three times during 10 seconds with one-minute intervals. An average maximal squeeze pressure was calculated. Further, the difference (mmHg) between anal and rectal pressure during straining and coughing was assessed.

Pudendal nerve terminal motor latency testing

Pudendal nerve terminal motor latency testing assessed at the right and the left side the pudendal nerve terminal motor latency using a St. Mark's Hospital electrode (Dantec; Skovlunde, Denmark)[20]. The pudendal nerve was stimulated on each side and the time needed for the external anal sphincter to contract after stimulation was measured. Latencies longer than 2.2 milliseconds were classified as pathologic.

Anal sensitivity testing

Anal sensitivity testing was performed with a stimulation electrode (Dantec Keypoint, Skovlunde, Denmark) mounted on a Foley Ch 12 catheter[16]. The anal sensation was measured by positioning the electrode into the mid-anal canal and gradually increasing the current (up to a maximum of 20 mAmp), until patients reported some sensation. To determine the threshold for anal sensation (mAmp) the lowest of three following measurements was used.

Rectal capacity measurement

The capacity measurement of the rectum was performed by using a balloon attached on a Foley Ch 14 catheter or a barostat[16]. The balloon catheter was introduced in the rectum and slowly inflated with air. The minimal rectal sensation perceived (sensory threshold), the volume associated with the initial urge to defecate (urge sensation) and the volume at which the patient experienced discomfort or pain and an intense desire to defecate (the maximal tolerated volume) were determined.

Imaging techniques

Defecography

Defecography was performed with contrast medium in rectum, small bowel and in females the vagina[21, 22]. The dynamics of defecation were evaluated. The presence of an intussusception (intussusception grade one or two, intrarectal or intra-anal circular invagination of the proximal rectal wall during defecation), anterior rectocele (outward bulge of the anterior rectal wall), enterocele (prolapse of the small bowel into the rectogenital space), sigmoidocele (prolapse of the sigmoid colon into the rectogenital space), or peritoneocele (prolapse of peritoneal fat or fluid into the rectogenital space) was assessed[21, 22].

Endoanal ultrasonography

Endoanal ultrasonography was performed with an ultrasound scanner (Bruel and Kjaer, Gentfofte, Denmark; Multiview Aloka, Tokyo, Japan) with radial endoscopic probe and a 7.5- or 10-MHz transducer[23, 24]. The endoscopic probe was introduced into the anus to the level of the anorectal verge and slowly withdrawn. The presence of an internal and/or external anal sphincter defect was assessed. A defect of the internal or external anal sphincter was defined by a discontinuity of the muscle ring and/or characterized by loss of the normal architecture, with an area of amorphous texture that usually has low reflectiveness[23, 24].

Endoanal MR imaging

Endoanal MR imaging was performed at a 1.0- or 1.5-T MR unit (Philips Gyroscan ACS-NT, Philips Medical Systems, Best, the Netherlands; General Electric Horizon Echospeed, General

Electric, Milwaukee, Ill) with a dedicated endoanal coil[23-25]. The endoanal coil was inserted in the anal canal and the presence of defects of the internal and external anal sphincter was assessed, as was the presence of internal and external anal sphincter atrophy. A defect of the internal or external anal sphincter was defined as a discontinuity of the muscle and/or a hypointense deformation of the normal pattern of the muscle layer due to replacement of muscle cells by fibrous tissue[25]. External anal sphincter atrophy was defined as diffuse thinning of the muscle and/or replacement of muscle fibres by fat and internal anal sphincter atrophy as diffuse muscle thinning (< 2 mm)[25].

Pelvic floor rehabilitation

Pelvic floor rehabilitation was administered by specialized pelvic physiotherapists according to a standardized protocol, which was compounded by clinicians and physiotherapists specialized in the field of pelvic floor disorders. Participating physiotherapists were uniformly trained and instructed to perform the treatment protocol adequately. Patients underwent weekly 35 minutes sessions for nine weeks. During the sessions physiotherapists obtained data by performing digital rectal examination, rectal balloon training and electromyography. Treatment targets and program were formulated based on these data. The pelvic floor rehabilitation program comprised rectal balloon training, electrical stimulation and/or electromyographic feedback. Electrical stimulation was offered only to patients with a poorly functioning external anal sphincter and/or puborectal muscle (Oxford score < 3). The Oxford score reflects the strength of the puborectal muscles and external anal sphincter muscle and ranges from 0 (no muscle contraction) to 5 (strong contraction)[26, 27]. Rectal balloon training was offered to all patients. Patients with an insensitive or hypersensitive rectum were respectively taught to perceive smaller or larger volumes of distension. Electromyographic feedback was offered to all patients with an average functioning external anal sphincter and/or puborectal muscle (Oxford score ≥ 3). Contraction capacities including duration, relaxation, timing and coordination of the pelvic floor muscles were trained. An extensive explanation and description of the pelvic floor rehabilitation program and specific treatment targets have been reported elsewhere[12]. Outcome of pelvic floor rehabilitation was assessed three months after completing the pelvic floor rehabilitation program, using the Vaizey score.

Statistical analysis

This study aimed to identify elements from patient's medical history, physical examination, anorectal function tests, and imaging tests that could predict the Vaizey score after treatment. First, the assumption of linearity between the continuous variables and the change in Vaizey score was studied, using visual inspection and spline functions. If necessary, the continuous variables were transformed to better approach linearity. Then linear regression analyses of the post-treatment Vaizey score were used to identify candidate predictor variables, using the baseline Vaizey score as a covariate in all of the models. Since the aim of this analysis is prediction, a liberal p-value ($p < 0.2$) was chosen to select candidate predictors[28].

Subsequently, multivariable linear regression analysis with a stepwise backwards selection procedure was used to construct prediction models for the post-treatment Vaizey score. The initial prediction model (model 1) included elements from medical history only. Subsequently, to calculate the added value of elements derived from tests, above the variables identified from medical history, different models were built. Separate models calculated the added value of the candidate predictors derived from physical examination (model 2), anorectal function tests (model 3), and imaging techniques (model 4), each time using a stepwise backwards selection procedure ($p < 0.2$).

A final model (model 5) was built combining both the predictors from medical history and the predictors from all additional tests. The total proportion explained variance (R^2) explained by this final model was examined. The R^2 takes value in the 0 to 1 range, with values closer to 1 indicating a better fit.

Results

Patients

In total 287 patients were included. Thirty-seven patients (13%) dropped out before or during the pelvic floor rehabilitation program, and baseline and follow-up Vaizey score were available for 250 patients (87%). Their mean age was 59 years (SD ± 13); 228 (91%) were female and 22 (9%) were male. The median duration of fecal incontinence was 5 years (interquartile range 2 to 10). The mean Vaizey incontinence score at baseline was 18 (SD ± 3). Mean Vaizey score after pelvic floor rehabilitation was 15 (SD ± 5), an average reduction of 3.2 points (95% CI: -2.6 to -3.9; $p < 0.001$).

Candidate predictors

Baseline characteristics obtained from medical history, physical examination, anorectal function tests and imaging techniques are summarized in Tables 1 and 2. None of the variables appeared to have a non-linear relation with the post-treatment Vaizey score.

The results from the regression analyses to identify candidate predictor variables are shown in Tables 1 and 2. Higher baseline Vaizey scores were significantly associated with higher post-treatment Vaizey scores, i.e., a worse outcome of pelvic floor rehabilitation ($p < 0.001$).

The following elements from medical history were also significantly associated ($p < 0.2$) with worse treatment outcome: presence of passive incontinence, thin stool consistency, sensation of incomplete evacuation, primary repair of a rupture after vaginal delivery at childbed, and any gynecological risk factor. Female gender, a longer duration of fecal incontinence, and the presence of solid stool consistency were significantly associated with a better treatment outcome (Table 1).

Inadequate maximal squeeze pressure, perineal and/or perianal scar tissue and presence of an anal sphincter defect were the only candidate predictors identified from physical examination (Table 2). All these variables were negatively associated with treatment outcome.

Of the anorectal function tests results, only a higher resting pressure and maximal squeeze pressure at anorectal manometry were positive predictive of outcome after pelvic floor rehabilitation (Table 2). A defect of the external anal sphincter and atrophy of the internal anal sphincter depicted at endoanal MR imaging, were found to have a significantly negative association with treatment outcome (Table 2). Data obtained at defecography or endoanal ultrasonography were not associated with the post-treatment Vaizey score.

Table 2. Baseline characteristics and candidate predictors from additional tests

Additional tests		ß	p
Physical examination			
Squeeze pressure (inadequate) *	208 (88%)	1.34	0.19 *
Resting pressure (inadequate)	166 (70%)	0.03	0.97
Perineal and/or perianal scar tissue *	138 (59%)	1.5	0.03 *
Defect anal sphincter complex *	80 (34%)	1.05	0.14 *
Anorectal functional tests			
Resting pressure (mmHg) (± SD) *	49 (± 23)	-0.03	0.04 *
Maximal squeeze pressure (mmHg) (± SD) *	87 (± 40)	-0.02	0.006 *
Difference anal-rectal pressure, coughing (mmHg) (± SD)	20 (± 38)	0	0.78
Difference anal-rectal pressure, straining (mmHg) (± SD)	8 (± 31)	0.01	0.54
Sensory threshold (ml) (± SD)	49 (± 33)	0.01	0.34
Urge sensation (ml) (± SD)	92 (± 49)	0	0.46
Maximal tolerable volume (ml) (± SD)	156 (± 68)	0	0.95
Pathological pudendal nerve latency right side	83 (38%)	0.06	0.93
Pathological pudendal nerve latency left side	85 (39%)	-0.39	0.57
Threshold anal sensation (mAmp) (± SD)	7.6 (± 6)	0.01	0.88
Defecography			
Presence of anterior rectocele	52 (27%)	-0.3	0.71
Presence of entero-, sigmo-, or peritoneocele	39 (21%)	0.21	0.82
Presence of intussusception	74 (39%)	0.68	0.35
Endoanal sonography			
Presence of EAS defect	136 (58%)	0.32	0.63
Presence of IAS defect	68 (29%)	0	0.99
Endoanal MR imaging			
Presence of EAS defect *	88 (46%)	1.1	0.14 *
Presence of IAS defect	71 (37%)	0.46	0.57
Presence of EAS atrophy	127 (66%)	-0.06	0.95
Presence of IAS atrophy *	34 (18%)	1.39	0.16 *

Note: Unless otherwise indicated, data are the number of patients, * indicates p-value below 0.2 (i.e., candidate predictor), EAS = external anal sphincter, IAS = internal anal sphincter

Multivariable analyses of response to treatment

After identifying predictor variables we investigated their pattern of missingness. Data were complete for almost 96% data points, and we were able to complete the dataset using multiple imputations based on correlations.

The initial multivariable model included all candidate predictors identified at medical history. In the backward elimination procedure two variables had a p-value above 0.2 and were therefore removed from the model: gender and any gynecological risk factor. The remaining model (model 1 at Table 3) had a total R^2 of 0.20.

Table 3. Several prediction models for the post-treatment Vaizey score

Medical history and tests	Model 1		Model 2		Model 3	
	ß	p	ß	p	ß	p
Physical examination						
Squeeze pressure (inadequate)						
Defect anal sphincter complex						
Perineal and / or perianal scar tissue *			0.94	0.15		
Anal manometry						
Resting pressure (mmHg)						
Maximal squeeze pressure (mmHg) *					-0.19	0.03
Endoanal MRI						
Presence of EAS defect						
Presence of IAS atrophy *						
Medical history						
Vaizey score at baseline *	0.51	0	0.51	0	0.49	0
Gender						
Duration of fecal incontinence *	-0.06	0.14	-0.06	0.11	-0.06	0.11
Presence of passive incontinence *	1.5	0.02	0.94	0.15	1.14	0.09
Sensation of incomplete evacuation (≥ 1/week) *	0.83	0.19	0.78	0.22	0.96	0.13
Thin stool consistency *	3.43	0.04	3.26	0.06	3.73	0.03
Solid stool consistency *	-0.99	0.19	-1.06	0.16	-0.78	0.31
Rupture after vaginal delivery repaired at childbed *	2.07	0	1.85	0.01	1.73	0.01
Any gynecological risk factors						
Constant	**4.2**		**3.8**		**6.5**	
R^2 of model	**0.2**		**0.21**		**0.22**	
Significance of change†			**0.15**		**0.03**	

In the second multivariable analysis we added the candidate predictors from physical examination to model 1 and applied a backwards elimination strategy. In addition to the variables from medical history, only perineal and/or perianal scar tissue was significantly associated with the Vaizey score after therapy; inadequate squeeze pressure and presence of an anal sphincter defect were not. Adding perineal and/or perianal scar tissue marginally increased the total R^2 to 0.21 (model 2 in Table 2; p = 0.15).

In a similar way, we looked at the candidate predictors from anorectal manometry. Only maximal squeeze pressure was significantly associated with the post-treatment Vaizey score in this multivariable model and increased the R^2 to 0.22 (model 3 in Table 2; p = 0.03).

Of the candidate predictors from endoanal MR imaging, only internal anal sphincter atrophy remained in model 4. This model resulted in a R^2 of 0.21 (model 4 at Table 2; p = 0.16).

	Model 4		Model 5	
	ß	p	ß	p
			0.8	0.22
			-0.02	0.02
	1.16	0.08	1.59	0.06
	0.53	0	0.51	0
	-0.06	0.08	-0.06	0.07
	1.55	0.02	1.16	0.08
	0.8	0.21	0.88	0.16
	3.18	0.06	3.25	0.06
	-1.09	0.15	-0.94	0.22
	2.16	0	1.63	0.02
	3.7		**5.6**	
	0.21		**0.23**	
	0.16		**0.02**	

Note: * indicates in multivariable analysis p-value below 0.2 (i.e., predictor), † indicates compared to model 1
(only parameters from medical history), EAS = external anal sphincter, IAS = internal anal sphincter

8

The final multivariable model (model 5) contained, in addition to the Vaizey score at baseline, six patient characteristics (passive incontinence, thin stool consistency, solid stool consistency, duration of fecal incontinence, sensation of incomplete evacuation and primary repair of a rupture after vaginal delivery at childbed) and three test variables (perineal and/or perianal scar tissue, maximal squeeze pressure and internal anal sphincter atrophy). A higher Vaizey score at baseline, passive incontinence, thin stool consistency, sensation of incomplete evacuation, primary repair of a rupture after vaginal delivery at childbed, perineal and/or perianal scar tissue, and atrophy of the internal anal sphincter were associated with poor response, whereas a longer duration of fecal incontinence, solid stool consistency and a higher maximal squeeze pressure were related with better response. This complete model resulted in a R^2 of 0.23 (model 5 in Table 3; p=0.02).

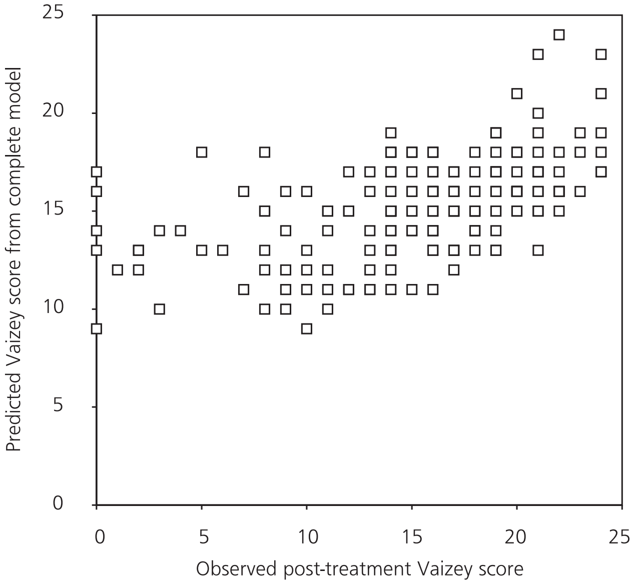


Figure 1. Association between the predicted post-treatment Vaizey score based on medical history and additional tests and the observed post-treatment Vaizey score

Figure 1 shows the association between the predicted Vaizey score based on the complete model (model 5) and the observed post-treatment Vaizey score. On basis of the final model it is impossible to accurately determine the post-treatment score of an individual patient. Particularly in patients with a low observed Vaizey score after treatment the model tends to overestimate the predicted post-treatment Vaizey score.

Discussion

This study demonstrates that tests have a limited role in predicting outcome of pelvic floor rehabilitation in patients with fecal incontinence due to mixed etiologies. We found a number of elements from medical history to be associated with the post-treatment Vaizey score, including duration of fecal incontinence, passive incontinence, thin or solid stool consistency, sensation of incomplete evacuation, primary repair of a rupture after vaginal delivery at childbed, and baseline Vaizey score. Adding test parameters from physical examination (perineal and/or perianal scar tissue), anorectal manometry (maximal squeeze pressure) and endoanal MR imaging (internal anal sphincter atrophy) marginally improved the predictive value to outcome, but overall no accurate assessment of the post-treatment Vaizey score in an individual patient was possible, especially not in those patients with a low observed Vaizey score.

A number of potential limitations should be taken into account. Some patients groups were excluded for this study as in these patients a specific disorder was held responsible for the fecal incontinence complaints. These patients needed treatment for that disorder and would not a priori be eligible for pelvic floor rehabilitation. Consequently, the study population of this study does not represent the full spectrum of fecal incontinence complaints and, therefore, the observed results cannot be unconditionally generalized to all patients with fecal incontinence.

The majority of participating patients was female (91%), an imbalance that is not due to a form of selection bias but is inherent to the aetiology of fecal incontinence, as obstetric damage of the anal sphincter complex proved to be a major cause of fecal incontinence[29]. To assess the outcome of pelvic floor rehabilitation we used the Vaizey score, as this score is a widely used score containing important incontinence-specific items like frequency and type of fecal incontinence, alteration in life style and pad and/or medication use. The Vaizey score has proved to be reproducible and previous studies have demonstrated an association between this scoring system, physicians' clinical impression and patients' subjective perception of relief[18, 30, 31].

The additional tests, although performed according to standard procedures, were performed by different specialized physicians or technicians, and the equipment used was not identical for all tests. This goes hand in hand with the multicenter design of our study, reflecting daily clinical practice. Due to the design of our study the observed changes in Vaizey score after treatment cannot exclusively be attributed to the pelvic floor rehabilitation program, as this study was not randomized with a parallel control group receiving no treatment. The cohort study design was selected as we wanted to evaluate the value of tests in predicting the outcome of pelvic floor rehabilitation in a large group of patients with fecal incontinence due to mixed etiologies, as worldwide this was, till now, not well established[13, 14, 17].

In contrast to other studies[32, 33] this study found that symptom severity and duration of fecal incontinence could predict outcome after pelvic floor rehabilitation to some extent. The observation that a longer duration was related to better outcome is not completely clear, but the fact that patients with a higher Vaizey score, indicating more severe fecal incontinence, were less likely to respond to pelvic floor rehabilitation could be explained by the fact that in these patients the underlying pathophysiology will be more extensive than in patients with a lower score. We did not find an association between outcome and age or gender, while another study had reported that patients under age 55 had a negative response to treatment[34]. The fact that thin stool consistency was related to poor outcome and solid stool consistency to good response, confirms the importance of stool consistency, additionally to normal anorectal function, in maintaining continence[14, 15]. Previous studies have reported that pelvic floor rehabilitation was less effective in patients with neurogenic fecal incontinence[35] and more effective in patients with fecal incontinence due to anal surgery or trauma[33], but we found only in patients with a primary repair of a rupture at childbed after vaginal delivery and scar tissue of the perineum and/or perianal area a worse outcome of pelvic floor rehabilitation. The sensation of incomplete rectal evacuation might for instance be related to the presence of an anterior rectocele or irritable bowel syndrome, but neither information from defecography nor from rectal capacity measurement was related to outcome. The latter is in contrast with other studies, which reported improved outcomes in patients with the ability to sense rectal distension[36, 37].

Unlike previous studies[32, 37, 38] this study showed that baseline maximal squeeze pressure was related to outcome. Pelvic floor rehabilitation aims to reinforce the external anal sphincter and its effects may be more pronounced in patients with a reasonable pre-treatment maximal squeeze pressure, reflecting external anal sphincter function.

The unfavorable outcome of pelvic floor rehabilitation in patients with internal anal sphincter atrophy might be explained by the fact that the internal anal sphincter is the main factor responsible for maintaining continence at rest, and its function is not trained by pelvic floor rehabilitation. Potentially the same explanation holds for the worse outcome in patients with passive incontinence, which is thought to be related to internal anal sphincter dysfunction[39]. Norton et al found better results after pelvic floor rehabilitation in patients with an intact anal sphincter complex[8] but we demonstrated, as did earlier studies[32, 40] that the presence of an internal and/or external anal sphincter defect was not important for predicting outcome following pelvic floor rehabilitation, just like the presence of external anal sphincter atrophy.

Diagnostic tests are used to gain information about the underlying pathophysiology of fecal incontinence[4, 15, 16]. Fecal incontinence is a multifactorial disorder and results of different tests should be combined to achieve a clear impression about the aetiology. Substantial variation exists between institutions and clinicians in the interpretation of test results and their management consequences[17]. Although this study has shown that some elements from medical history and additional test variables were predictive to response after pelvic floor rehabilitation, the overall predictive value of the multivariable model was limited. On basis of additional tests to assess fecal incontinence patients can not be informed on the likely outcome of pelvic floor rehabilitation. This suggests that additional tests are not strictly essential before referring patients for pelvic floor rehabilitation and that these tests might be more helpful in selecting patients for surgical treatment options.

Acknowledgements

The authors thank the following participating investigators of our study group for scientific advice and collecting data: J.B.V.M. Delemarre[a], E. van der Harst, P.P.L.O. Coene, E.J. Spillenaar-Bilgen, C.H. van der Vaart, W.F. van Tets, J.J.G.M. Gerritsen, J.W. de Bruijne, M.G.W. Dijkgraaf, R.G.H. Beets-Tan, V.P.M. van der Hulst, T.G. Wiersma, M.N.J.M. Wasser, A.B. Huisman, J.A. de Priester, A. Sikkenk, T.D. Witkamp.
[a]Deceased.

References

1. Perry S, Shaw C, McGrother C, Matthews RJ, Assassa RP, Dallosso H, Williams K, Brittain KR, Azam U, Clarke M, Jagger C, Mayne C, Castleden CM. Prevalence of faecal incontinence in adults aged 40 years or more living in the community. *Gut* 2002;50:480-484
2. Thomas TM, Egan M, Walgrove A, Meade TW. The prevalence of faecal and double incontinence. *Community Med* 1984;6:216-220
3. Deutekom M, Terra MP, Dobben AC, Dijkgraaf MG, Baeten CG, Stoker J, Bossuyt PM. Impact of faecal incontinence severity on health domains. *Colorectal Dis* 2005;7:263-269
4. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *Lancet* 2004; 364:621-632
5. Beddy P, Neary P, Eguare EI, McCollum R, Crosbie J, Conlon KC, Keane FB. Electromyographic biofeedback can improve subjective and objective measures of fecal incontinence in the short term. *J Gastrointest Surg* 2004;8:64-72
6. Fynes MM, Marshall K, Cassidy M, Behan M, Walsh D, O'Connell PR, O'Herlihy C. A prospective, randomized study comparing the effect of augmented biofeedback with sensory biofeedback alone on fecal incontinence after obstetric trauma. *Dis Colon Rectum* 1999;42:753-758
7. Lorenz EP, Wondzinski A. Results of conservative and surgical therapy of anal incontinence. 1974 to 1992 patient sample. *Zentralbl Chir* 1996;121:669-675
8. Norton C, Kamm MA. Outcome of biofeedback for faecal incontinence. *Br J Surg* 1999;86:1159-1163
9. Hosker G, Norton C, Brazzelli M. Electrical stimulation for faecal incontinence in adults. *Cochrane Database Syst Rev* 2000;CD001310
10. Norton C, Hosker G, Brazzelli M. Biofeedback and/or sphincter exercises for the treatment of faecal incontinence in adults. *Cochrane Database Syst Rev* 2000;CD002111
11. Norton C. Behavioral management of fecal incontinence in adults. *Gastroenterology* 2004; 126: S64-S70
12. Terra MP, Dobben AC, Berghmans B, Deutekom M, Baeten CG, Janssen LW, Boeckxstaens GE, Engel AF, Felt-Bersma RJ, Slors JF, Gerhards MF, Bijnen AB, Everhardt E, Schouten WR, Bossuyt PM, Stoker J. Electrical stimulation and pelvic floor muscle training with biofeedback in patients with fecal incontinence: a cohort study of 281 patients. *Dis Colon Rectum* 2006;49:1149-1159
13. Prather CM. Physiologic variables that predict the outcome of treatment for fecal incontinence. *Gastroenterology* 2004;126:S135-S140
14. Rao SS. Pathophysiology of adult fecal incontinence. *Gastroenterology* 2004;126:S14-S22
15. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-1685
16. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-760
17. Whitehead WE, Wald A, Norton NJ. Priorities for treatment research from different professional perspectives. *Gastroenterology* 2004;126:S180-S185
18. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
19. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-1880
20. Kiff ES, Swash M. Slowed conduction in the pudendal nerves in idiopathic (neurogenic) faecal incontinence. *Br J Surg* 1984;71:614-616

21. Jorge JM, Habr-Gama A, Wexner SD. Clinical applications and techniques of cinedefecography. *Am J Surg* 2001;182:93-101

22. Wiersma TG, Mulder CJ, Reeders JW, Tytgat GN, Van Waes PF. Dynamic rectal examination (defecography). *Baillieres Clin Gastroenterol* 1994;8:729-741

23. Stoker J, Halligan S, Bartram CI. Pelvic floor imaging. *Radiology* 2001;218:621-641

24. Stoker J, Bartram CI, Halligan S. Imaging of the posterior pelvic floor. *Eur Radiol* 2002;12:779-788

25. Rociu E, Stoker J, Zwamborn AW, Lameris JS. Endoanal MR imaging of the anal sphincter in fecal incontinence. *Radiographics* 1999;19:S171-S177

26. Isherwood PJ, Rane A. Comparative assessment of pelvic floor strength using a perineometer and digital examination. *BJOG* 2000;107:1007-1011

27. Laycock J, Jerwood D. Pelvic floor muscle assessment: The PERFECT scheme. *Physiotherapy* 2001;87:631-642

28. Spiegelhalter DJ. Probabilistic prediction in patient management and clinical trials. *Stat Med* 1986;5:421-433

29. Henry MM. Pathogenesis and management of fecal incontinence in the adult. *Gastroenterol Clin North Am* 1987;16:35-45

30. Deutekom M, Terra MP, Dobben AC, Dijkgraaf MG, Felt-Bersma RJF, Stoker J, Bossuyt PM. Selecting an outcome measure for evaluating treatment in fecal incontinence. *Dis Colon Rectum* 2005;48:2294-301

31. Baxter NN, Rothenberger DA, Lowry AC. Measuring fecal incontinence. *Dis Colon Rectum* 2003; 46:1591-1605

32. Rieger NA, Wattchow DA, Sarre RG, Cooper SJ, Rich CA, Saccone GT, Schloithe AC, Toouli J, McCall JL. Prospective trial of pelvic floor retraining in patients with fecal incontinence. *Dis Colon Rectum* 1997;40:821-826

33. Cerulli MA, Nikoomanesh P, Schuster MM. Progress in biofeedback conditioning for fecal incontinence. *Gastroenterology* 1979;76:742-746

34. Fernandez-Fraga X, Azpiroz F, Aparici A, Casaus M, Malagelada JR. Predictors of response to biofeedback treatment in anal incontinence. *Dis Colon Rectum* 2003;46:1218-1225

35. Van Tets WF, Kuijpers JH, Bleijenberg G. Biofeedback treatment is ineffective in neurogenic fecal incontinence. *Dis Colon Rectum* 1996;39:992-994

36. Chiarioni G, Bassotti G, Stegagnini S, Vantini I, Whitehead WE. Sensory retraining is key to biofeedback therapy for formed stool fecal incontinence. *Am J Gastroenterol* 2002;97:109-117

37. Wald A. Biofeedback therapy for fecal incontinence. *Ann Intern Med* 1981;95:146-149

38. Sangwan YP, Coller JA, Barrett RC, Roberts PL, Murray JJ, Schoetz DJ, Jr. Can manometric parameters predict response to biofeedback therapy in fecal incontinence? *Dis Colon Rectum* 1995;38:1021-1025

39. Vaizey CJ, Kamm MA, Bartram CI. Primary degeneration of the internal anal sphincter as a cause of passive faecal incontinence. *Lancet* 1997;349:612-615

40. Leroi AM, Dorival MP, Lecouturier MF, Saiter C, Welter ML, Touchais JY, Denis P. Pudendal neuropathy and severity of incontinence but not presence of an anal sphincter defect may determine the response to biofeedback therapy in fecal incontinence. *Dis Colon Rectum* 1999; 42:762-769

Chapter 9

Comparison of endoanal magnetic resonance imaging and endoanal ultrasonography in the depiction of external anal sphincter defects in fecal incontinent patients

Annette C. Dobben
Maaike P. Terra
J. Frederik M. Slors
Marije Deutekom
Michael F. Gerhards
Regina G.H. Beets-Tan
Patrick M. M. Bossuyt
Jaap Stoker

Radiology, accepted

Abstract

PURPOSE. To prospectively compare in a multicenter study the agreement between endoanal magnetic resonance imaging and endoanal ultrasonography for depicting external anal sphincter defects in fecal incontinent patients.

MATERIALS AND METHODS. The study was approved by the medical ethics committee of all participating centers. Consenting fecal incontinent patients from 13 different hospitals were examined with endoanal magnetic resonance (MR) imaging and endoanal ultrasonography. Patients with an anterior external anal sphincter defect, depicted on endoanal MR imaging and / or endoanal ultrasonography, underwent anal sphincter repair. The operative findings were used as reference standard in the determination of anterior external anal sphincter defects. Cohen's kappa statistic and McNemar test were used to calculate agreement and differences between diagnostic techniques.

RESULTS. The agreement between endoanal MR imaging and endoanal ultrasonography in 237 patients (90% females; mean age 59) was fair for the depiction of sphincter defects (kappa 0.24, 95% CI 0.12 – 0.36). At surgery external anal sphincter defects were depicted in 86% of patients (31/36). There was no significant difference between MR imaging and ultrasonography in the depiction of sphincter defects (p=0.23). Sensitivity and positive predictive value of endoanal MRI were 81% and 89% respectively, versus 90% and 85% at endoanal ultrasonography.

CONCLUSIONS. For the selection of patients for anal sphincter repair both endoanal MR imaging and endoanal ultrasonography are sensitive tools in the preoperative assessment and both techniques can be used to depict surgically repairable anterior EAS defects.

Introduction

Apart from medical history and physical examination, the evaluation of fecal incontinence may require anorectal functional tests and imaging [1]. The ability to visualize the anatomy and pathology of the anal sphincter muscles using either endoanal ultrasonography (US) or endoanal magnetic resonance (MR) imaging has altered our understanding of the pathogenesis of fecal incontinence and has the potential to guide further evaluation and management[2].

Currently, endoanal US is the preferred diagnostic technique to select patients for surgery[3, 4]. The advantages of endoanal US are its availability, limited costs and more widely available experience[5]. In contrast endoanal (MR imaging may allow for a clear visualization of the external anal sphincter as there is large contrast difference between the external anal sphincter muscle and the surrounding fat and endoanal MR imaging is able to detect external anal sphincter atrophy[5].

Previous studies of these imaging techniques have concluded that they should be considered comparable in the selection of patients for surgery[3, 6-13], but all of these studies were single center studies. Thus, the purpose of our study was to prospectively compare in a multicenter study the agreement between endoanal magnetic resonance imaging and endoanal ultrasonography for depicting external anal sphincter defects in fecal incontinent patients.

Materials and Methods

Study Design

The study was approved by the medical ethics committee of all participating centers. All included patients that entered the study signed informed consent.

Between December 2001 and May 2005 consecutive patients with fecal incontinence were included in 13 medical centers in the Netherlands. Details of the study design are reported elsewhere[14].

Eligible patients were all referred for standardized, specialized pelvic floor rehabilitation after a standardized diagnostic work-up, including imaging. If pelvic floor rehabilitation failed, overlapping anterior anal sphincter repair was considered as the next available treatment option for patients with an external anal sphincter (EAS) defect. An EAS defect was defined as a solitary external anal sphincter defect of more than thirty degrees of the circumference of the sphincteric ring, detected at endoanal MR imaging and / or endoanal ultrasonography. Excluded from overlapping anterior anal sphincter repair were patients with severe generalized external anal sphincter atrophy, defined as extensive thinning of the EAS muscle or diffuse replacement of EAS muscle by fat[15], detected on endoanal MR imaging.

Clinical assessment

Evaluation of the severity of symptoms as well as a detailed medical history was obtained by one of the 13 participating clinicians. The severity of fecal incontinence was assessed according to the grading system of Vaizey[16].

Imaging tests

Since imaging modalities were not available in all of the 13 participating centers both endoanal US and endoanal MR imaging were performed in seven centers. Consequently, certain patients had their exams in another center than that from which they originated. All endoanal US, except for one, were performed by six clinicians with experience ranged from 10 to 14 years. In one center the endoanal US was performed by a technician with 10 years of experience. All endoanal MR imaging were performed by technicians with experience ranged from one to five years. For logistic reasons endoanal US was performed prior to endoanal MR imaging.

Endoanal ultrasonography

Endoanal ultrasonography was performed with an ultrasonography scanner (Bruel and Kjaer, Gentfofte, Denmark; Multiview Aloka, Tokyo, Japan) with a radial endoscopic probe (7.5 or 10 MHz transducer) and a sonolucent plastic cone while the patient laid in the left lateral position with their knees bent at 90 [2, 11, 17]. The endoscopic probe was introduced into the anal canal, positioned at the upper aspect of the puborectalis sling, and slowly withdrawn until all levels, perpendicularly to the anal canal, were scanned.

Endoanal MR Imaging

Endoanal MR imaging was performed at a 1 T or 1.5 T MR unit (Philips Gyroscan ACS-NT, Philips Medical Systems, Best, the Netherlands; General Electric Horizon Echospeed, General Electric, Milwaukee, Ill) with a dedicated endoanal coil with a diameter of 19 mm[11, 15, 18]. All patients were asked to fast four hours prior to the MR imaging examination to reduce artifacts from bowel peristalsis. Bowel relaxants were used in one of the institutions (one ml of butylscopolamine bromide (Buscopan, 20 mg/ml; Boehringer Ingelheim, Germany or one mg glucagon hydrochloride (Glucagen, Bagsvaerd, Denmark). The endoanal coil was covered with a condom and after application of lubricant inserted in the anal canal in a left lateral position. After positioning of the endoanal coil the patients turned in supine position and supportive pads were used to stabilize the coil.

Scan parameters were optimized for the MR imaging machines used based on extensive previous experience. The following T2 weighted fast spin echo sequences were used according to a standardized imaging protocol that had been established during joined meetings: TR 2500 - 3500 ms, TE 70 – 90 ms, echo train length 10, field of view 10 x 10 cm (axial) and 16 x 16 cm (coronal), imaging matrix 256 x 512, 3 mm slice thickness, 0.3 mm interslice gap and 2 excitations. Axial images and coronal images with slice orientation perpendicular and parallel to the anal sphincter and endoanal coil were performed.

Image analysis

The images were analyzed separate from the imaging session. Endoanal US images were analyzed on a personal computer to capture the series of the endoanal US images. The endoanal MR image analysis were performed by using a workstation viewing software (IMPAX SP4 SU4 DS3000, AGFA, Mortsel, Belgium or Easy Vision Workstation, Philips Medical System, Best, the Netherlands).

An EAS defect at endoanal US was defined as a discontinuity of the muscle ring (anatomic defect) and/ or characterized by loss of the normal architecture, with an area of amorphous texture that usually has low reflectiveness (functional defect, scar tissue)[4]. A defect of the EAS at endoanal MR imaging was defined as a discontinuity of the muscle ring (anatomic defect) and / or recognized by a hypointense deformation of the normal pattern of the muscle layer due to replacement of muscle cells by fibrous tissue (functional defect, scar tissue)[15].

The endoanal US images were scored by anyone of the six observers out of seven centers where the imaging was performed (2 gastroenterologists and 4 surgeons who all are experts in the field with a considerable amount of experience in reading endoanal US images (10 to 14 years)).

The endoanal MR images were scored by anyone of the three observers (R.G.H.B.T. and J.S.), also out of the participating centers, and all experienced radiologists in evaluating abdominal MR imaging (eight to 12 years).

Both endoanal US and endoanal MR imaging were evaluated separately. Observers were blinded to the findings of the other technique and the medical history of the patients except for age, gender and the presence of fecal incontinence.

Anterior Anal Repair

The decision to perform surgery was made by a participating surgeon (J.F.M.S. and M.F.G.) based on imaging findings (e.g. extent of the EAS lesion at endoanal imaging and / or degree of sphincter atrophy) and complementary clinical information (e.g. the severity of fecal incontinence, the willingness of the patient to undergo surgery) and findings from anorectal physiology testing.

Overlapping anterior anal sphincter repair was performed as previously described[19, 20] in eight participating centers by anyone of eight experienced colorectal surgeon (six to 25 years). Operative findings were recorded and used as reference standard.

Statistical Considerations

Patient groups were compared with respect to their characteristics with analysis of variance and χ^2-test.

The depiction of EAS defects at endoanal MR imaging was compared with endoanal US. To calculate the agreement between both diagnostic techniques we used Cohen's kappa statistic with 95% confidence intervals (CI). The agreement was considered poor (≤0.20), fair (0.21-0.40), moderate (0.41-0.60), good (0.61-0.80) or very good (>0.80)[21].

The findings of EAS defects determined at surgery were compared with endoanal MR imaging and endoanal US. To assess if significant differences exist between both diagnostic techniques, the McNemar test was used. Sensitivity and positive predictive values with 95% CI were calculated for the depiction of EAS defects with surgery as reference standard and the imaging techniques as index tests.

For all statistical tests p-values below 0.05 were considered to indicate statistical significance. SPSS for Windows (version 11.5, 2002) was used to perform statistical analysis of our data.

Results

Imaging data from 237 patients were collected of which 214 patients (90%) were female. Their mean age was 58.6 years (± SD 13). Clinical characteristics are summarized in Table 1. The mean (± SD) interval between the performance of endoanal MR imaging and endoanal US was 3 (± 36) days. A study specific flow diagram (Figure 1) visually reveals the procedures used to sample patients and to obtain data.

Table 1. Clinical characteristics for all patients in the main cohort (n= 237)

Characteristics		
Female (%)		214 (90)
Mean age (SD)		59 (13)
Median during of fecal incontinence (range)		5 (0.5-57)
Mean Vaizey incontinence score (SD)		18.2 (2.9)
Obstetric history	Parous (%)	191 (89)
	Median deliveries (range)	2 (1-10)
Obstetric risk factors	Breech delivery (%)	17 (9)
	Long labor (%)	51 (27)
	Child > 8 pounds (%)	55 (29)
	Forceps delivery (%)	8 (4)
	Vacuum pump delivery (%)	20 (10)
	Episiotomy (%)	114 (60)
	Rupture (%)	111 (58)
Previous anal surgery	Haemorroidectomy (%)	21 (9)
	Sphincterotomy (%)	3 (1)
	Sphincter repair (%)	18 (8)
	Fistel operation (%)	10 (4)
	Lord procedure (%)	5 (2)
	Remaining (%)	10 (4)

Note: US = ultrasonography; MRI = magnetic resonance imaging

Endoanal MR imaging versus endoanal US

An EAS defect was depicted in 31 patients on only endoanal MR imaging; in 60 patients on only endoanal US and in 77 patients on both modalities (Table 2). The characteristics of the patient groups showed close resemblance. Patients where only an EAS defect was depicted on endoanal US were significant older than patients with an EAS defect depicted on both modalities. The agreement between endoanal MR imaging and endoanal US for mapping EAS defects was fair (Kappa 0.24; 95% CI 0.12 – 0.36) (Table 3). Most defects were scored as located at the anterior side or anterior-lateral side of the EAS at either technique (at endoanal MR imaging in 104/108 (96%) patients; at endoanal US in 130/137 (95%) patients).

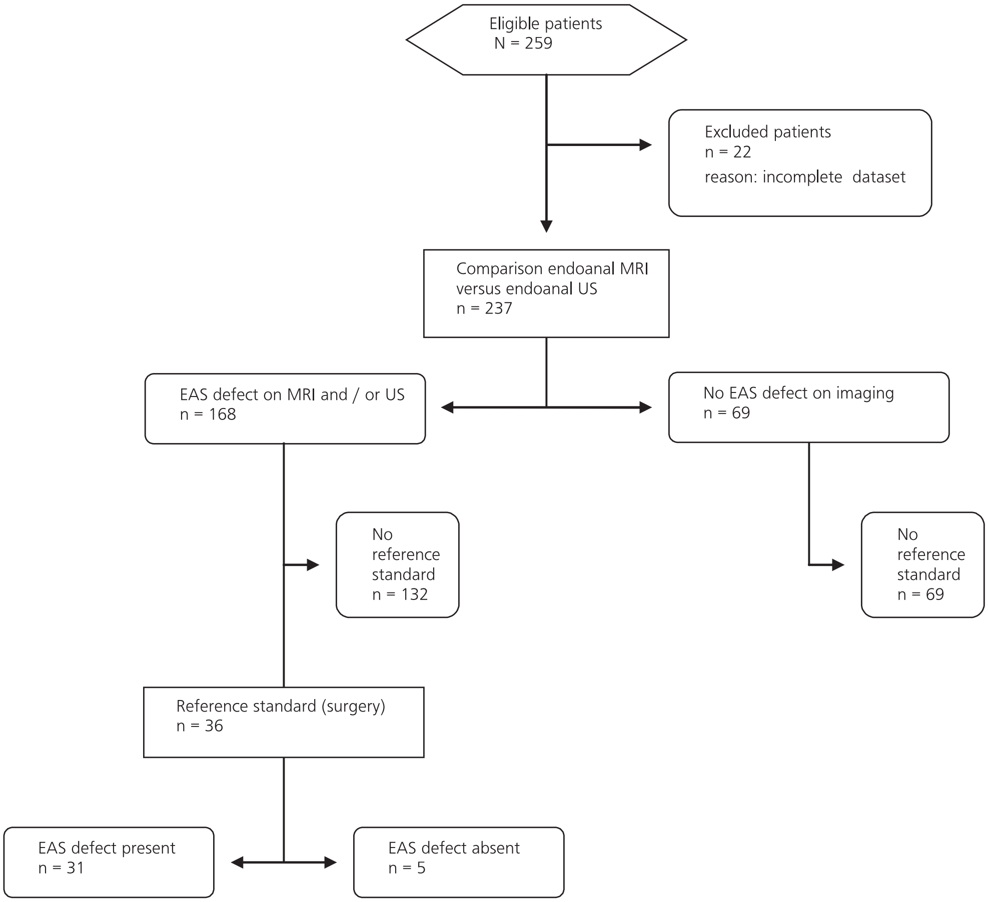


Figure 1. Flow diagram that summarizes patient sampling. MRI = Magnetic Resonance Imaging; US = Ultrasonography; EAS = External Anal Sphincter

Validity to detect EAS defects of imaging tests versus surgery

In total there were 71% (168/237) with an EAS defect depicted at either endoanal MR imaging and / or endoanal US. Twenty-one percent (36/168) of these patients underwent an anterior anal repair. Main reasons for patients not to be operated included the following: 1) Another intervention than anterior anal repair (e.g. sacral nerve stimulation) was performed (n= 44); 2) Refusal of the patient or clinician to operate for miscellaneous reasons (n=31); 3) Sufficient improvement by pelvic floor rehabilitation (n= 18); 4) Reason unknown (n=15); 5) Severe generalized atrophy was depicted on endoanal MR imaging and therefore surgery was not recommended (n=12); 6) Patient dropped out the study before surgery (n=7); 7) Patient is still on waiting list to be operated (n=5).

The mean interval between imaging tests and surgery was 10 months (range seven to seventeen months). Data was collected from 36 patients who underwent anterior anal repair

Table 2. Clinical data from patients with an EAS defect depicted on endoanal US and / or endoanal MRI (n = 237)

		Only EAS defect on Endoanal US (n = 60)	Only EAS defect on Endoanal MRI (n = 31)	EAS defect on both imaging modalities (n = 77)	P value
Female (%)		57 (95)	28 (89)	74 (96)	0.5
Mean age (SD)		61 (12)	58 (14)	54 (14)	**0.02****
Median during of fecal incontinence (range)		4.5 (0.5-57)	5.5 (1.5-44)	6 (0.5-40)	0.4
Mean Vaizey incontinence score (SD)		18.4 (3)	17.4 (3)	18.4 (3)	0.2
Obstetric history	Parous (%)*	47 (82)	24 (86)	70 (95)	0.08
	Median deliveries (range)	2 (1-7)	3 (1-5)	2 (1-5)	0.7
Only obstetric risk factors (%)		31 (55)	20 (67)	48 (64)	0.2
Only previous anal surgery (%)		4 (7)	3 (10)	6 (8)	
Obstetric risk factors combined with anal surgery (%)		12 (22)	2 (7)	17 (23)	
No evident obstetric risk factors or anal surgery (%)		9 (16)	5 (16)	4 (5)	

Note: EAS = external anal sphincter; US = ultrasonography; MRI = magnetic resonance imaging
* concerns only females.
** significant difference between EAS defect on endoanal US versus both modalities.

Table 3. Agreement between endoanal US and endoanal MRI for the presence of EAS defects in 237 patients

		Endoanal US		
		yes	*no*	*total*
Endoanal MRI	*yes*	**77**	31	108
	no	60	**69**	129
	total	137	100	237

Note: US = ultrasonography; MRI = magnetic resonance imaging; EAS = external anal sphincter
Bold numbers explain the numbers of patients where agreement was reached for either US and MRI.

of which 34 patients (94%) were female. Their mean age was 51 years (± SD 12.5). Clinical characteristics are summarized in Table 4.

Data of surgery and endoanal MR imaging versus surgery and endoanal US (Table 5) showed that surgery detected in 86% (31/36) an anterior EAS defects and could not find an anterior EAS defect in five patients (14%). There was no significant difference in the depiction of EAS defects between endoanal MR imaging and endoanal US (Mc Nemar p= 0.23) (Figures 2 and 3). The sensitivity of detecting EAS defects was 81% (25/31; 95% CI 0.67 - 0.95) at endoanal MR imaging versus 90% (28/31; 95% CI 0.80 - 1) at endoanal US. Positive predictive value for detecting EAS

Table 4. Clinical characteristics for patients studied in the AAR cohort (n= 36)

Characteristics		
Female (%)		34 (94)
Mean age (SD)		51 (12.5)
Median during of fecal incontinence (range)		6.5 (0.5-40)
Mean Vaizey incontinence score (SD)		18 (3.4)
Obstetric history*	Parous (%)	31 (91)
	Median deliveries (range)	2 (1-5)
Obstetric risk factors**	Breech delivery (%)	5 (16)
	Long labor (%)	7 (23)
	High birth weight infant (%)	10 (32)
	Forceps delivery (%)	2 (6)
	Vacuum pump delivery (%)	3 (10)
	Episiotomy (%)	16 (52)
	Rupture (%)	24 (77)
Previous anal surgery	Haemorroidectomy (%)	5 (14)
	Sphincterotomy (%)	1 (3)
	Sphincter repair (%)	4 (11)
	Fistel operation (%)	4 (11)
	Remaining (%)	2 (6)

Note: More than one covariate of the variabeles 'obstetric risk factors' and 'previous anal surgery' can be related to one patient.
AAR = anterior anal repair
* Concerns only females.
** Concerns only parous women.

Table 5. Comparison of endoanal US and endoanal MRI versus surgery in the depiction of EAS defects (n=36)

		Surgery		
		yes	*no*	*total*
Endoanal US	*yes*	**28**	5	33
	no	3	**0**	3
	total	31	5	36
Endoanal MRI	*yes*	**25**	3	28
	no	6	**2**	8
	total	31	5	36

Note: EAS = external anal sphincter; US = ultasound; MRI = magnetic resonance imaging

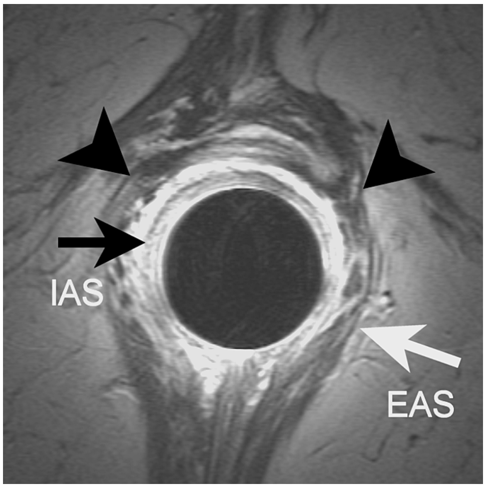


Figure 2a. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image showing scar tissue (arrow heads) at the anterior EAS at the distal anal canal in a 56-year-old woman with a complicated vaginal delivery, hysterectomy and Lord-procedure in the past. This finding was confirmed at anal sphincter repair revealing an anterior EAS defect. IAS=lower edge internal anal sphincter, EAS= external anal sphincter

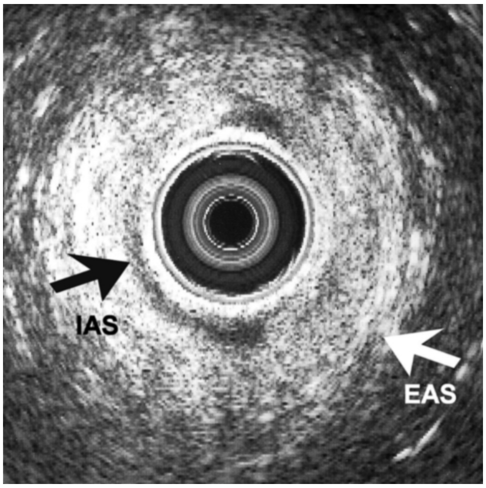


Figure 2b. Transverse endoanal ultrasonography obtained at the distal anal canal from the same patient as in Figure 2a. Anal sphincters were diagnosed as intact. The top of the figure is anterior. IAS = lower edge internal anal sphincter, EAS = external anal sphincter

Table 6. Results from surgery compared to endoanal US and endoanal MRI

	Diagnosis surgery	Diagnosis at endoanal US	Confirmed by surgery	Diagnosis at endoanal MRI	Confirmed by surgery
External anal sphincter					
Defect	31/36	33/36	28	28/36	25
Location defect					
Anterior	31	32	26	28	25
Posterior	0	1	0	0	0
Defect accompanied by atrophy	4	0	0	14	2
No defect	5	3	0	8	2
Atrophy	3	1	0	5	1

Note: US = ultrasonography; MRI = magnetic resonance imaging

defects was 89% at endoanal MR imaging (25/28) and 85% at endoanal US (28/33). Complete agreement between both techniques for anterior EAS defects was 69% (25/36). We could not calculate specificity and negative predictive values since one of the cells contained zero patients.

Details of the findings at surgery compared to endoanal US and endoanal MR imaging (Table 6) showed four of the detected EAS defects were accompanied by thinning of the EAS due to generalized EAS atrophy. The depiction of atrophy was overlooked in almost all cases except for

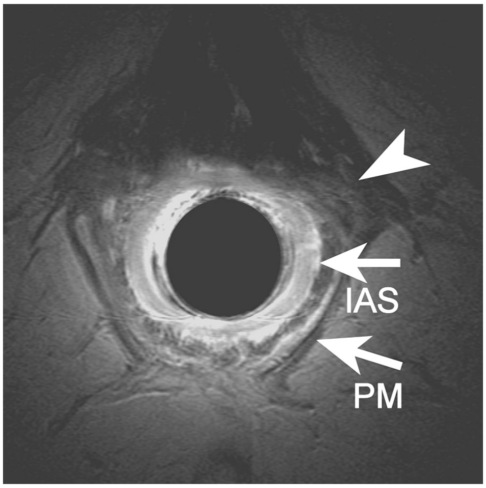


Figure 3a. Transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the proximal anal canal in a 69-year-old woman with a complicated vaginal delivery and hysterectomy in the past. Anal sphincters were diagnosed as intact. Retrospectively, slight asymmetry of the structures (arrow head) left anterolateral to the internal anal sphincter as compared to the right anterolateral site can be seen; most likely representing the external anal sphincter defect diagnosed at surgery. IAS = internal anal sphincter, PM = lower edge puborectal muscle

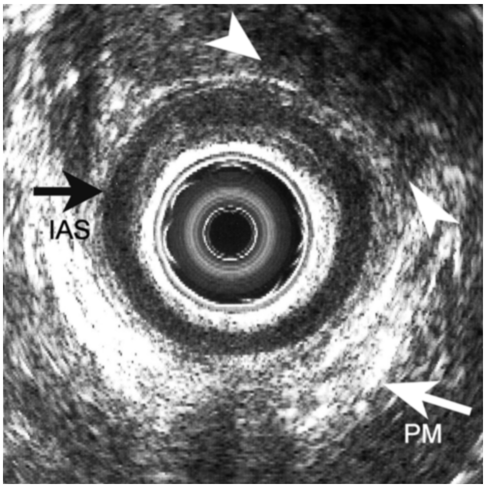


Figure 3b. Transverse endoanal ultrasonography obtained from the same patient as in Figure 3a showing an EAS defect at upper edge (arrow heads) at the proximal anal canal. This finding was confirmed at anal sphincter repair, revealing an anterior EAS defect. The top of the figure is anterior. IAS = internal anal sphincter, PM = lower edge puborectal muscle

one at endoanal US, while endoanal MR imaging diagnosed more patients with atrophy than was found at surgery. Endoanal MR imaging depicted in five patients generalized atrophy which was confirmed by surgery for one patient. Endoanal MR imaging depicted in 14 patients an EAS defect accompanied by atrophy, which was confirmed by surgery in two patients.

Discussion

When validating our findings at endoanal MR imaging and endoanal US with the findings at anterior anal repair, the results of our study show that both diagnostic techniques are sensitive for depicting anterior EAS defects that are amendable to surgery. Furthermore, both techniques can be used as adequate tools in the positive prediction of EAS defects in fecal incontinent patients. Specificity and negative predictive values are low or could not be calculated. This most likely is caused by a low prevalence of negative data as a consequence of the fact that only patients with an EAS defect depicted on imaging were referred for surgery.

Our study shows that the agreement between endoanal MR imaging and endoanal US is fair for the depiction of EAS defects. Despite the fact that all observers can be qualified as 'experienced', the variety in evaluation of EAS defects is substantial. Earlier studies demonstrated that both imaging techniques are accurate in mapping defects of the external anal sphincter[15, 22-27]. DeSouza and co-authors[23] concluded that endoanal MR imaging correctly diagnosed sphincter tears in seven patients, all validated by surgery. Others showed[25-27], by confirming the defect at surgery, that endoanal US accurately detected sphincter lesions in fecal incontinent patients. Unfortunately, those studies consisted of small patient groups and only compared one imaging technique with surgery. To our knowledge, there are two comparative studies published on endoanal MR imaging versus endoanal US in the depiction of EAS defects in a population of fecal incontinent patients[3, 10]. A prospective study of 52 fecal incontinent patients found complete agreement between endoanal MR imaging and endoanal US and the final diagnosis in 62%[10]. A retrospective study of 22 fecal incontinent patients demonstrated a fair agreement in diagnosing external anal sphincter damage between endoanal MR imaging and endoanal US (Kappa 0.38)[3]. This is concordant with the findings of our prospective comparative study. The reliability of our findings might have been influenced by the variety between various observers from different centers. However, despite the fact that the two quoted comparative studies are single center studies, the results of our multicenter study does do not differ substantially. We also found a fair agreement (kappa 0.24) between both imaging techniques in a large cohort of fecal incontinent patients. Furthermore, the results of our multicenter study give a better reflection of daily clinical practice, and are therefore easier applicable to external validity.

A number of potential limitations to our study should be addressed. A major limitation is partial verification bias, since we do not know the surgical findings in the non-operated patient group. This may lead to overestimation of sensitivity and underestimation of specificity. Potential true or false negatives therefore can not be calculated.

Also, in our study only endoluminal imaging of EAS lesions were evaluated. These results were compared with findings at surgical anterior anal repair. An anterior EAS defect can be considered generally as a surgically remedial tear. The EAS defects in our study population were mainly located at the anterior side of the sphincter complex. This confirmed our expectations, since the majority of our cohort consists of females with one or more obstetric risk factor(s)[3]. We did not include internal anal sphincter findings in our comparative study as for isolated internal anal sphincter damage there is no surgical option available[10], except for injectable silicone biomaterial implants (a new experimental therapy which is still under investigation)[28].

In the selection of candidates for surgery previous studies have shown that endoanal MR imaging is an accurate diagnostic technique in depicting EAS atrophy, contrary to endoanal US [7, 17, 29-34]. The accurate demonstration of the EAS at endoanal MR imaging, especially of its borders and fat content, facilitates the evaluation of atrophy. EAS atrophy is characterized by generalized thinning of its muscle fibers and /or fatty replacement [31]. External anal sphincter atrophy negatively affects continence after anterior anal repair[29, 35]. Although AAR seems to confer substantial benefits on these patients[36], short term results vary in the literature and are contradictory[9, 37-41]. To prevent unnecessary surgery, endoanal MR imaging seems a useful diagnostic technique in the preoperative assessment. Unfortunately, our study showed that endoanal MR imaging depicted 14 patients with an EAS defect accompanied by EAS atrophy which was confirmed by surgery for only two patients. This implies that it may not have been reasonable to exclude patients on the basis of MR atrophy. However, since surgery is able to determine generalized sphincter thinning rather than fatty infiltration, histology is needed to confirm the latter. So, histology is the reference standard for EAS atrophy. In our study no histology has been performed. Consequently, patients with EAS atrophy characterized by fatty replacement could not be assessed at surgery. So, it can be hypothesized that in reality more patients than recorded are affected with EAS atrophy as was suggested at MR imaging.

We have demonstrated a fair agreement between endoanal MR imaging and endoanal US in a large cohort of fecal incontinent patients. We were only able to validate our findings in a subgroup of patients that underwent surgery. The selection of this subgroup is based on diagnostic imaging. We do not know to what extent a certain preference for one of the imaging modalities might have played a role in decision making by the clinician. Endoanal US is widely available, contrary to endoanal MR imaging. The use of the latter has been restricted to specialized centers, because the required endoanal coil is not yet available with every MR machine [6]. Therefore, it is possible that experience with one technique had influenced the selection process for surgery.

When validating our results in a small cohort against surgery, we can conclude that both imaging techniques can be considered as useful in the selection of patients for surgery, where endoanal MR imaging is able to depict EAS atrophy, which is associated with a poor outcome of AAR. However, in contrast to endoanal US, MR imaging is restricted to specialized centers. Therefore, the technique of choice in clinical decision making shall depend on the infrastructure of the center. The technique of choice in clinical decision making may depend on the infrastructure of the center.

References

1. Dobben AC, Terra MP, Deutekom M, Bossuyt PM, Felt-Bersma RJ, Stoker J. Diagnostic work up of faecal incontinence in daily clinical practice in the Netherlands. *The Neth J Med* 2005; 63:265-269
2. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-60
3. Rociu E, Stoker J, Eijkemans MJ, Schouten WR, Lameris JS. Fecal incontinence: endoanal US versus endoanal MR imaging. *Radiology* 1999;212:453-458
4. Bartram CI. Ultrasound. In: Imaging pelvic floor disorders. Bartram CI, DeLancy JOL, Halligan S, Kelvin FM, Stoker J (eds). Springer-Verlag Berlin Heidelberg New York 2003;69-80
5. Stoker J, Rociu E, Wiersma TG, Lameris JS. Imaging of anorectal disease. *Br J Surg* 2000; 87:10-27
6. Beets-Tan RG, Morren GL, Beets GL, Kessels AG, el Naggar K, Lemaire E, Baeten CG, van Engelshoven JM. Measurement of anal sphincter muscles: endoanal US, endoanal MR imaging, or phased-array MR imaging? A study with healthy volunteers. *Radiology* 2001;220:81-89
7. Stoker J, Hussain SM, Lameris JS. Endoanal magnetic resonance imaging versus endosonography. *Radiol Med* (Torino) 1996;92:738-741
8. Fletcher JG, Busse RF, Riederer SJ, Hough D, Gluecker T, Harper CM, Bharucha AE. Magnetic resonance imaging of anatomic and dynamic defects of the pelvic floor in defecatory disorders. *Am J Gastroenterol* 2003;98:399-411
9. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003; 124:1672-1685
10. Malouf AJ, Williams AB, Halligan S, Bartram CI, Dhillon S, Kamm MA. Prospective assessment of accuracy of endoanal MR imaging and endosonography in patients with fecal incontinence. *Am J Roentgenol* 2000;175:741-745
11. Stoker J, Halligan S, Bartram CI. Pelvic floor imaging. *Radiology* 2001;218:621-641
12. Frudinger A, Halligan S, Bartram CI, Price AB, Kamm MA, Winter R. Female anal sphincter: Age-related differences in asymptomatic volunteers with high-frequency endoanal US. *Radiology* 2002;224:417-423
13. Christensen AF, Nyhuus B, Nielsen MB, Christensen H. Three-dimensional anal endosonography may improve diagnostic confidence of detecting damage to the anal sphincter complex. *Br J Radiol* 2005;78:308-311
14. Terra MP, Dobben AC, Berghmans B, Deutekom M, Baeten CG, Janssen LW et al. Electrical stimulation and pelvic floor muscle training with biofeedback in patients with fecal incontinence: a cohort study of 281 patients. *Dis Colon Rectum* 2006;49:1149-1159
15. Rociu E, Stoker J, Zwamborn AW, Lameris JS. Endoanal MR imaging of the anal sphincter in fecal incontinence. *Radiographics* 1999;19:S171-S177
16. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80
17. Stoker J, Rociu E, Zwamborn AW, Schouten WR, Lameris JS. Endoluminal MR imaging of the rectum and anus: technique, applications, and pitfalls. *Radiographics* 1999;19:383-398
18. Stoker J, Rociu E. Endoluminal MR imaging of anorectal diseases. *J Magn Reson Imaging* 1999;9:631-634
19. Briel JW, de Boer LM, Hop WC, Schouten WR. Clinical outcome of anterior overlapping external anal sphincter repair with internal anal sphincter imbrication. *Dis Colon Rectum* 1998; 41:209-214
20. Slade MS, Goldberg SM, Schottler JL, Balcos EG, Christenson CE. Sphincteroplasty for acquired anal incontinence. *Dis Colon Rectum* 1977;20:33-35

21. Altman DG. Practical statistics for medical research. Boca Ration, Florida: CRC Press LLC; 1999

22. deSouza NM, Williams AD, Gilderdale DJ. High-resolution magnetic resonance imaging of the anal sphincter using a dedicated endoanal receiver coil. *Eur Radiol* 1999;9:436-443

23. deSouza NM, Hall AS, Puni R, Gilderdale DJ, Young IR, Kmiot WA. High resolution magnetic resonance imaging of the anal sphincter using a dedicated endoanal coil. Comparison of magnetic resonance imaging with surgical findings. *Dis Colon Rectum* 1996;39:926-934

24. Law PJ, Kamm MA, Bartram CI. Anal endosonography in the investigation of faecal incontinence. *Br J Surg* 1991;78:312-314

25. Cuesta MA, Meijer S, Derksen EJ, Boutkan H, Meuwissen SG. Anal sphincter imaging in fecal incontinence using endosonography. *Dis Colon Rectum* 1992;35:59-63

26. Meyenberger C, Bertschinger P, Zala GF, Buchmann P. Anal sphincter defects in fecal incontinence: Correlation between endosonography and surgery. *Endoscopy* 1996;28:217-224

27. Deen KI, Kumar D, Williams JG, Olliff J, Keighley MR. Anal sphincter defects. Correlation between endoanal ultrasound and surgery. *Ann Surg* 1993;218:201-205

28. Kenefick NJ, Vaizey CJ, Malouf AJ, Norton CS, Marshall M, Kamm MA. Injectable silicone biomaterial for faecal incontinence due to internal anal sphincter dysfunction. *Gut* 2002; 51:225-228

29. Briel JW, Stoker J, Rociu E, Lameris JS, Hop WCJ, Schouten WR. External anal sphincter atrophy on endoanal magnetic resonance imaging adversely affects continence after sphincteroplasty. *Br J Surg* 1999;86:1322-1327

30. Briel JW, Zimmerman DD, Stoker J, Rociu E, Lameris JS, Mooi WJ, Schouten WR. Relationship between sphincter morphology on endoanal MRI and histopathological aspects of the external anal sphincter. *Int J Colorectal Dis* 2000;15:87-90

31. Stoker J, Bartram CI, Halligan S. Imaging of the posterior pelvic floor. *Eur Radiol* 2002; 12:779-788

32. deSouza NM, Puni R, Kmiot WA, Bartram CI, Hall AS, Bydder GM. MRI of the anal sphincter. *J Comput Assist Tomogr* 1995;19:745-751

33. Enck P, Heyer T, Gantke B, Schmidt WU, Schafer R, Frieling T, Haussinger D. How reproducible are measures of the anal sphincter muscle diameter by endoanal ultrasound? *Am J Gastroenterol* 1997;92:293-296

34. Rociu E, Stoker J, Eijkemans MJC, Lameris JS. Normal anal sphincter anatomy and age- and sex-related variations at high-spatial-resolution endoanal MR imaging. *Radiology* 2000; 217:395-401

35. Briel JW, Zimmerman DD, Schouten WR. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2001;88:729-730

36. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *The Lancet* 2004; 364:621-632

37. Baig MK, Wexner SD. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2000;87:1316-1330

38. Wexner SD, Marchetti F, Jagelman DG. The role of sphincteroplasty for fecal incontinence reevaluated: a prospective physiologic and functional review. *Dis Colon Rectum* 1991;34:22-30

39. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-1880

40. Jorge JM, Wexner SD. Etiology and management of fecal incontinence. *Dis Colon Rectum* 1993;36:77-97

41. Rao SS. Diagnosis and management of fecal incontinence. American College of Gastroenterology Practice Parameters Committee. *Am J Gastroenterol* 2004;99:1585-1604

Chapter 10

The role of endoluminal imaging in clinical outcome of overlapping anterior anal sphincter repair in fecal incontinent patients

Annette C. Dobben
Maaike P. Terra
Marije Deutekom
J. Frederik M. Slors
Lucas W.M. Janssen
Patrick M. M. Bossuyt
Jaap Stoker

Submitted

Abstract

BACKGROUND. Anterior sphincter repair has become the operation of choice in incontinent patients with external anal sphincter (EAS) defects but not all patients benefit from surgery. The aim was to investigate if endoluminal imaging can identify determinants in outcome of sphincter repair.

METHODS. Pre- and postoperative Vaizey incontinence score was evaluated and endoanal magnetic resonance imaging (MRI) and endoanal ultrasonography were performed. We evaluated the association between preoperatively assessed EAS measurements with outcome as well as postoperatively depicted residual defects, atrophy, tissue at overlap, and sphincter overlap with outcome.

RESULTS. After surgery the mean Vaizey score in 30 patients (97% females; mean age 50) had improved from 18 to 13 ($p<0.001$).

MRI demonstrated that baseline measurement of increased EAS thickness correlated with a better outcome ($r=0.42$; $p=0.032$). Patients with a visible overlap and less than 20% fat tissue had a better outcome than patients with non-visible, fatty overlap (decrease in Vaizey score 7 versus 2 points; $p=0.037$).

Ultrasonography showed that patients with a persistent EAS defect had a worse outcome compared to patients without an EAS defect (17 versus 10; $p=0.003$).

CONCLUSIONS. MRI was useful in determining EAS thickness and structure and ultrasonography was effective in depicting residual EAS defects.

Introduction

Initial treatment options for fecal incontinence are conservative and include medication and / or dietary measures. In case of failure, pelvic floor rehabilitation is an additional conservative therapy[1]. If conservative treatment fails, sphincter repair can be an option. Anterior overlapping sphincter repair (sphincteroplasty) has become the operation of choice in fecal incontinent patients with anterior defects of the external anal sphincter (EAS) muscle, particularly in patients with postobstetric trauma[2]. Overlapping repair seems to confer substantial benefits on these patients[3, 4] but reported short term results vary [2] while recent data suggest that the effects of sphincteroplasty deteriorate with time[5-9].

It is unclear why some patients do not benefit from surgery. Hypotheses have pointed to post surgical breakdown of the repair, scarring, and pudendal neuropathy, related either to the initial injury or to the subsequent repair as well as to the role of aging[10]. The relationship between pudendal neuropathy as measured by prolonged latency at pudendal nerve motor latency testing and the outcome of anal sphincter repair is still controversial[10]. Ternent and colleagues[11] showed that the size of an EAS sphincter defect, postoperatively determined by endoanal ultrasonography (US), significantly correlated with change in continence following sphincter repair. Other studies showed that postoperatively persistent EAS defects depicted on endoanal US were associated with a poor clinical outcome after anterior anal sphincter repair[12-15] and patients may undergo a second or even third sphincter repair[16]. A study with magnetic resonance (MR) imaging[17] found that extreme atrophy of the external anal sphincter is a predictor for poor outcome of anterior anal sphincter repair. Furthermore, endoanal MR imaging is able to depict EAS atrophy rather than assuming atrophy by pudendal nerve latencies[18-20].

Anorectal function tests can give us only partial understanding of the origin of sphincter repair failures. Therefore, we wanted to further investigate the role of endoluminal imaging. We accordingly set up a study to investigate prospectively if preoperative endoluminal imaging can identify factors that can predict the outcome of sphincter repair, and if postoperative endoluminal imaging findings are associated with poor outcome. We submitted patients to endoanal MR imaging as well as endoanal US before and after surgery and evaluated the association between EAS measurements, atrophy, residual defects, sphincter overlap, tissue at overlap, and the improvement, or the lack thereof, after repair.

Materials and Methods

Study Design

This study was designed within a large cohort study evaluating the effects of pelvic floor rehabilitation in patients with fecal incontinence due to mixed etiology. Details of that study are reported elsewhere[21]. The cohort study was conducted between December 2001 and May 2005 in 8 medical centers (blinded for review). The study had been approved by the medical ethics committee of all participating centers. Informed consent was obtained from all included patients.

Inclusion criteria were the existence of fecal incontinence complaints for six months or more, a Vaizey incontinence score of at least 12[22], and failure of conservative treatment, based on standardized, specialized pelvic floor rehabilitation, dietary recommendations and/or antidiarrhetics. Excluded were patients with an age below 18, patients diagnosed less than two years ago with an anorectal tumor and patients with a previous ileoanal or coloanal anastomosis. To investigate the treatment effect of pelvic floor rehabilitation, patients with chronic diarrhea (always fluid stools, three or more times a day), overflow incontinence, proctitis, soiling (leakage of fecal material out of the anus after normal defecation leading to perineal eczema), and rectal prolapse were also excluded from participation. The presence of a rectal prolapse was determined by evacuation proctography.

Consecutive patients were included. In patients with a relevant external anal sphincter defect in whom pelvic floor rehabilitation had failed as an initial therapy, overlapping anterior anal sphincter repair was considered as the next available treatment option. A relevant EAS defect was defined as an external anal sphincter defect in the axial plane of more than one hour, depicted at endoanal MR imaging and / or endoanal sonography. Excluded from surgery were patients with extreme generalized external anal sphincter atrophy detected on endoanal MR imaging [17, 19].

Imaging tests

Endoanal sonography

Endoanal sonography was performed with an ultrasound scanner (Bruel and Kjaer, Gentfofte, Denmark; Multiview Aloka, Tokyo, Japan) with a radial endoscopic probe (7.5 or 10 MHz transducer) and a sonolucent plastic cone while the patient laid in the left lateral position with their knees bent at 90 [16, 23, 24]. The endoscopic probe was introduced into the anal canal, positioned at the upper aspect of the puborectalis sling, and slowly withdrawn until all levels, perpendicularly to the anal canal, were scanned.

The images were evaluated by anyone of four observers out of different hospitals who all were experts in the field with a considerable amount of experience in reading endoanal US images (10 to 14 years). Preoperatively, the presence of an EAS defect was assessed as well as scarring[15]. Postoperatively, a residual EAS defect was defined as a complete hypoechogenic gap in the region of the repair, with separated fibers of the EAS and no evidence of overlapping sphincters[11, 14] and / or no decrease of the EAS defect compared to the defect depicted at baseline.

Pre- and postoperatively, the extent of a defect was axially indicated in hours (1-12 hours, with 12 o'clock anterior, three o'clock left lateral, six o'clock posterior and nine o'clock right lateral) as was atrophy of the EAS. Atrophy was judged on its reflection of the outer interface and length.

The pre surgical and post surgical endoanal US was evaluated unblinded to patient outcome.

Endoanal MR imaging

Endoanal MR imaging was performed at a 1 T or 1.5 T MR unit (Philips Gyroscan ACS-NT, Philips Medical Systems, Best, the Netherlands; General Electric Horizon Echospeed, General Electric, Milwaukee, Ill) with a rectangular receive-only coil with a diameter of 19 mm[16, 20, 25, 26].

Scan parameters were optimized for the MR imaging machines used based on extensive previous experience. The following T2 weighted fast spin echo sequences were used according to a standardized imaging protocol that had been established during joined meetings: TR 2500 - 3500 ms, TE 70 – 90 ms, echo train length 10, field of view 10 x 10 cm (axial) and 16 x 16 cm (coronal), imaging matrix 256 x 512, 3 mm slice thickness, 0.3 mm interslice gap and 2 excitations. Axial images and coronal images with slice orientation perpendicular and parallel to the anal sphincter and endoanal coil were performed.

Since this study concerns a pilot study identifying factors playing a role in surgical outcome, image analysis were done by just one reader who is a highly experienced radiologist (blinded for review) and is the most experienced reader available in evaluating abdominal MR imaging (12 years) and endoanal MR imaging (approximately 1000 examinations).

Preoperatively, the presence of EAS defect and EAS atrophy were scored according to previously described definitions[25, 27]. Pre- and postoperatively, the extent of a defect was axially indicated in hours (1-12 hours, with 12 o'clock anterior, three o'clock left lateral, six o'clock posterior and nine o'clock right lateral).

Postoperatively, the presence of EAS defects was assessed, as were the presence of EAS atrophy and the extent of sphincter overlap. A residual defect was defined as a full thickness discontinuity of more than half of the anterior external sphincter and / or no decrease of the EAS defect compared to the defect depicted at baseline.

EAS overlap was defined as visible overlapping edges of both EAS ends and measured in hours. As we hypothesized that replacement of EAS muscle by fat or scar tissue might influence clinical outcome, the post surgical EAS structure was evaluated qualitatively by scoring percentages (by units of 10) of muscle, fat and scar tissue at the level of the anterior sphincter overlap. Muscle was defined as tissue with low signal intensity and a structured orientation of fibers. Fat has high signal intensity, while scar tissue has very low signal intensity and disordered architecture[25].

To determine if EAS thickness (normal value is 4 mm on endoluminal imaging)[28] is associated with clinical outcome, measurements (mm) were performed pre- and post surgery anteriorly at 12 o'clock position and right lateral on nine o'clock, at the level of one cm superior to the anal verge. Nine o'clock was chosen as most anterior EAS defects do not extend to nine o'clock. The anterior post surgical measurement included the sphincter overlap. Longitudinal measurements (mm) of the anterior sphincter length were also obtained (normal value is approximately 27 mm, while in women approximately 15 mm on endoluminal imaging)[28]. The radiologist evaluating endoanal MR images pre-operatively and postoperatively was unblinded to patient outcome.

Overlapping anterior anal sphincter repair

The decision to perform surgery was made by a participating surgeon based on imaging findings (e.g. extent of the EAS lesion at endoanal imaging and / or degree of sphincter atrophy) and complementary clinical information (e.g. clinical examination and the severity of fecal incontinence) and findings from anorectal function testing. Overlapping anterior anal sphincter repair was performed as previously described[29, 30] in eight participating centers by one of eight experienced colorectal surgeons (six to 25 years).

After surgery the severity of fecal incontinence was determined with the Vaizey incontinence score and endoluminal imaging was repeated, similar to the procedures at baseline.

Statistical Considerations

Changes in Vaizey incontinence scores after surgery from baseline were tested for significance using Wilcoxon's test for paired data.

To evaluate the association of factors with the outcome of surgery the Mann-Whitney test was used.

Spearman's correlation coefficients were used to evaluate the association between the size of an EAS defect and the change in Vaizey incontinence score before and after surgery. Also, Spearman's correlation coefficient was used to determine any correlation between EAS measurements (thickness and length) before surgery and outcome of surgery, and between post surgery EAS tissue structure (fat, scar or muscle) and outcome of surgery at endoanal MR imaging.

For all statistical tests p-values below 0.05 were considered to indicate statistical significance. We used SPSS for Windows (version 11.5, 2002) to perform statistical analysis of our data.

Results

In this study 30 fecal incontinent patients could be included, of which 29 patients (97%) were female. Their mean age was 50 years (± SD 12). Clinical characteristics are summarized in Table 1. Before surgery, all patients had an anterior EAS defect depicted on solely endoanal US (n=6) or endoanal MR imaging (n=4), or on both imaging modalities (n=20).

The median interval between clinical assessment, preoperative imaging and surgery was 10 months (range 5 to 21). The follow up period for clinical assessment ranged from one to 20 months (median 4.7 months) and for imaging from three to 18 months (median 4.5 months) after surgery.

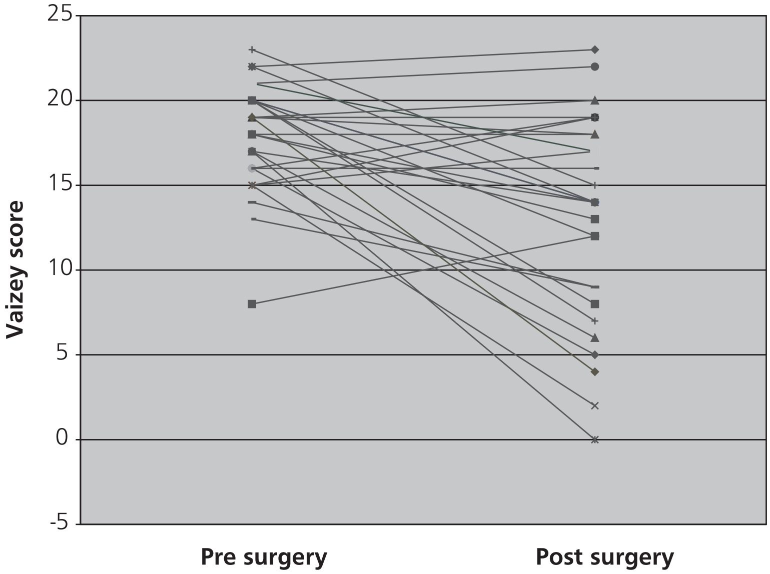


Figure 1. Ladder graph presenting the individual changes in Vaizey score before and after surgery (mean change is a decrease of five points). Each patient is represented by a different symbol.

Table 1. Baseline characteristics (n= 30)

Characteristics		
Female (%)		29 (97)
Mean age (SD)		50 (132
Median during of fecal incontinence (range)		6.5 (0.5-22)
Mean Vaizey incontinence score (SD)		18 (3)
Passive incontinence		2 (7)
Urge incontinence		10 (33)
Combined passive and urge incontinence		18 (60)
Obstetric history*	Parous (%)	28 (97)
	Median deliveries (range)	2 (1-5)
Obstetric risk factors**	Breech delivery (%)	4 (14)
	Long labor (%)	5 (18)
	High birth weight infant (%)	11 (39)
	Forceps delivery (%)	1 (4)
	Vacuum pump delivery (%)	3 (11)
	Episiotomy (%)	15 (54)
	Rupture (%)	23 (82)
Previous anal surgery	Haemorroidectomy (%)	5 (17)
	Sphincterotomy (%)	1 (3)
	Sphincter repair (%)	4 (11)
	Fistel operation (%)	2 (7)
	Remaining (%)	1 (3)

Notes: More than one covariate of the variabeles 'obstetric risk factors' and 'previous anal surgery' can be related to one patient. We could not retrieve complete information of all items for every patient.
* Concerns only females
** Concerns only parous women

Clinical outcome

The mean (±SD) Vaizey score changed significantly after surgery, from 18 (±3) to 13 (±6) ($p<0.001$), resulting in a mean improvement of 25%. In 10 patients (33.3%) the Vaizey score improved less than 5% or deteriorated; in 13 patients (43.3%) the Vaizey score improved 5 to 50%; in seven patients (23.3%) it had improved more than 50% (Figure 1). The latter were all patients without previous anorectal surgery in their medical history in contrast to the other patients with minor (7 out of 13 patients) or no improvement (3 out of 10 patients).

Patients younger than 65 years had a better clinical outcome than patients of 65 years or older ($p<0.034$). The mean (±SD) Vaizey score of the younger patients improved 5 (±6) points (29%) compared to a deterioration of 2 (±2) points (11%) in the older patient subgroup.

Table 2a. Findings before surgery on endoanal ultrasonography

	n	Vaizey at baseline mean (SD)	Vaizey after surgery mean (SD)	Difference mean (SD)	P value
Size EAS defect					
1 - 3 hours	15	18 (3)	14 (6)	4 (6)	0.788
4 - 8 hours	9	17 (2)	13 (6)	4 (5)	

Note: EAS = external anal sphincter

Table 2b. Findings before surgery on endoanal MR imaging

	n	Vaizey at baseline mean (SD)	Vaizey after surgery mean (SD)	Difference mean (SD)	P value
Size EAS defect					
1 - 3 hours	11	18 (2)	11 (7)	7 (7)	0.191
4 - 8 hours	13	18 (4)	14 (5)	4 (5)	
EAS atrophy					
yes	15	18 (3)	15 (4)	3 (6)	0.145
no	15	18 (3)	12 (7)	6 (6)	
EAS atrophy					
mild	10	17 (4)	15 (5)	2 (6)	0.460
moderate	5	18 (2)	14 (6)	4 (6)	

Note: EAS = external anal sphincter

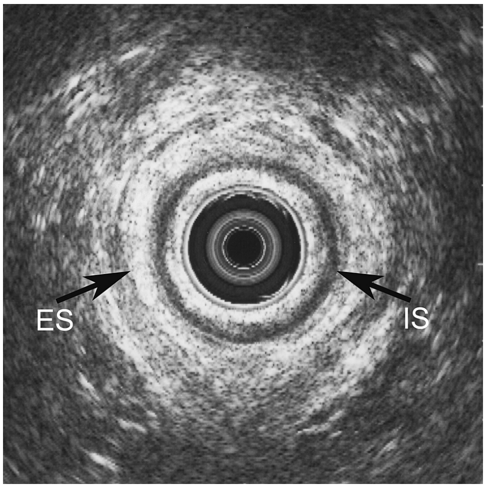


Figure 2a. Transverse endoanal ultrasonography obtained from a 65-years-old man demonstrating normal anatomy at the mid-anal canal of the internal anal sphincter (IS) and external anal sphincter (ES). The top of the figure is anterior.

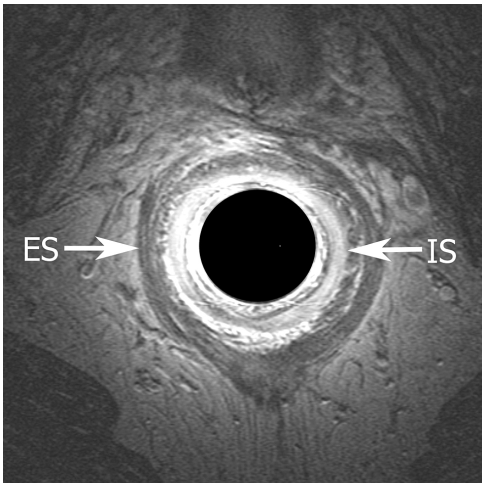


Figure 2b. Transverse endoanal T2-weighted fast spin-echo (2500/70) MR image obtained from a 78-year-old man demonstrating normal anatomy at the mid-anal canal of the internal anal sphincter (IS) and external anal sphincter (ES). The top of the figure is anterior.

Imaging

The size of the EAS defect as preoperatively assessed on both endoanal US (p= 0. 0.788) and endoanal MR imaging (p= 0.191) was not associated with clinical outcome (Table 2[a] and 2[b]). Normal anatomy of the sphincter complex is pictured in Figure 2[a] and 2[b].

Endoanal US

In 11 patients endoanal US depicted an EAS defect postoperatively. The postoperative Vaizey score was significantly worse in patients with a postoperative depicted EAS defect compared to patients without an EAS defect (17 (±4) versus 10 (±6); p= 0.003). The size of a residual EAS defect post surgery was not significantly associated with the change in incontinence score (p= 0.553) (Table 4[a]). There was no EAS atrophy depicted on either pre-surgical or post-surgical assessments at endoanal US.

Table 3. External anal sphincter measurements

	Baseline	After Surgery	Difference	P value
	mean (SD)	mean (SD)	mean (95% CI)	
Sphincter thickness anterior	3.2 (1.6)*	5.8 (2.1)**	2.6 (1.6 - 3.5)	**<0.001**
Sphincter thickness at nine hours	4.1 (1.2)	3.3 (1.2)	-0.8 (-1.5 - -0.2)	**0.018**
Sphincter height	10 (5.3)	15.1(5.7)	5.1 (2.2 - 8)	**0.003**
At the level of sphincter overlap:		median (range)		
Fat %		10 (0 - 70)		
Scar %		35 (0 - 100)		
Muscle %		45 (0-100)		

Note:*Sphincter thickness before surgery was measured at the anterior side at 12 hours. In case of an anterior sphincter defect measurement was done at 3 hours .
**Sphincter thickness after surgery was measured at the anterior side at 12 hours, including the overlap.

Table 4a. Findings after surgery on endoanal ultrasonography

	Endoanal ultrasonography				
	n	**Vaizey at baseline mean (SD)**	**Vaizey after surgery mean (SD)**	**Difference mean (SD)**	**P value**
Size EAS defect					
1 - 3 hours	9	18.5 (3)	18 (3)	0.5 (4)	0.553
4 - 6 hours	2	18 (1)	16.5 (3.5)	1.5 (2)	
Residual EAS defect					
yes	11	18 (2)	17 (4)	1 (3.5)	**0.003**
no	15	18 (4)	10 (6)	8 (6)	

Note: EAS = external anal sphincter.

10

Endoanal MR imaging

We had to exclude post surgery MR data of one patient from analysis because of extensive susceptibility artefacts.

There was no significant difference in change of Vaizey score between patients with and without generalized EAS atrophy (p=0.145; Table 2[b]) and between patients with mild and moderate EAS atrophy depicted at baseline (p=0.460; Table 2[b]).

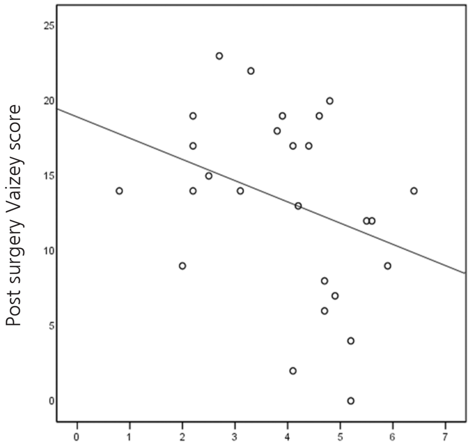


Figure 3. Correlation between external anal sphincter thickness at baseline and outcome of surgery determined by the Vaizey score. Note: EAS = external anal sphincter

Table 4b. Findings after surgery on endoanal MR imaging

Endoanal MR imaging					
	n	**Vaizey at baseline mean (SD)**	**Vaizey after surgery mean (SD)**	**Difference mean (SD)**	**P value**
Size EAS defect					
1 - 3 hours	13	18 (2)	12 (7)	6 (6.5)	
4 - 6 hours	0	0	0	0	
Residual EAS defect					
yes	13	18 (2)	12 (7)	6 (6.5)	0.536
no	14	19 (3)	15 (6)	4 (5.5)	
EAS atrophy					
yes	12	18 (2)	14.5 (5)	3.5 (6)	0.256
no	14	19 (3)	12 (7)	7 (6)	
Sphincter overlap					
yes	19	18.5 (3)	13 (6)	5.5 (6)	0.463
no	6	18.5 (3)	15 (6)	3.5 (6)	

Note: EAS = external anal sphincter

EAS thickness and height had changed significantly from baseline after surgery (Table 3). Increased EAS thickness on nine hours was significantly correlated with a better surgical outcome (r= 0.42; p= 0.032; Figure 3). All other sphincter measurements were not associated with surgical outcome.

Postoperatively, there was no significant difference in clinical outcome between patients with and without a persistent EAS defect (p= 0.536), or EAS atrophy (p= 0.256) (Table 4[b]) detected at MR imaging. No significant difference could be found between patients with visible anterior sphincter overlap (Figure 4[a] and 4[b]) compared to patients in whom a sphincter overlap could not be depicted (p= 0.463).

There was no significant correlation between the amount of fat (r= 0.03), scar (r= 0.13) or muscle (r= 0.04) depicted at endoanal MR imaging and surgical outcome as measured with the Vaizey score, unless sphincter overlap was interacted with fat. Patients with a clear visible overlap and less than 20% fat tissue (n= 15) have a significantly better surgical outcome than patients (n= 10) with a fatty anterior EAS in whom overlap could not be visualised (mean (±SD) decrease in Vaizey score was 7 (±6) versus 2 (±5) points; p= 0.037).

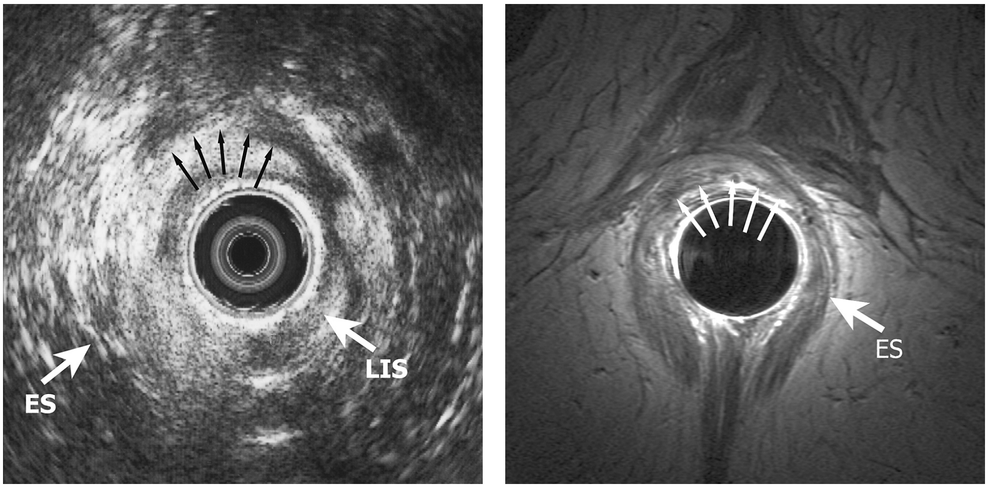


Figure 4a and 4b. (**4a**) Transverse endoanal ultrasonography and (**4b**) transverse endoanal T2- weighted fast spin-echo (2500/70) MR image at the mid - distal anal canal obtained from a 53-years-old woman after a complicated vaginal delivery (rupture) demonstrating sphincter overlap (thin arrows) of both external anal sphincter ends, left over right, after anterior anal sphincter repair and continuity of the sphincter ring has been restored. Although appearances at endoluminal imaging show overlap, surgery failed for this patient as the patient was still fecal incontinent.
The top of either figure is anterior. LIS = lower edge internal anal sphincter, ES = external anal sphincter.

Discussion

This study shows that patients with a residual EAS defect depicted on endoanal US had a significantly worse clinical outcome at a short term follow up after overlapping anterior anal sphincter repair. As in many other studies a persistent EAS defect on endoanal US was associated with poor outcome [31, 32]. In contrast to earlier studies[11, 33] we could not find a relation between

the size of an EAS defect postoperatively, depicted on endoanal US, and the change of Vaizey fecal incontinent score after surgery.
We have tried to identify determinants that can be visualized with endoluminal imaging that play a role in the outcome of surgical sphincter repair. This study demonstrates that preoperative increased sphincter thickness as measured on endoanal MR imaging is correlated with a better clinical outcome. In former studies it has been established that extreme generalized atrophy (thinning of the EAS muscle fibers and /or fatty replacement) is a negative predictor for surgical outcome[17, 18]. In this study EAS thickness also played a role in success or failure of the sphincter repair.

There are potential limitations of this study that should be considered. The depiction of residual EAS defects on endoanal MR imaging was insufficient. This can be explained by the lack of experience in evaluating the post surgical anatomy with endoanal MR imaging. Furthermore, there are no fixed diagnostic criteria for endoanal MR imaging to evaluate the post surgical status. To distinguish a residual defect in an area of post surgical scar tissue is probably far more difficult than expected.

Clinical outcome was studied in terms of changes in the Vaizey incontinence score, which is a subjective outcome measuring instrument. Grading of fecal incontinence is difficult and several incontinence scores have been developed[22, 34, 35]. The Vaizey score is the most complete scoring system[22, 36]. A previous study demonstrated that this scoring system is reproducible and correlated well with physician's clinical impression[22, 37]. Despite this fact, subjective elements may have introduced bias or imprecision in outcome.

As this study was a pilot study the patient group studied was small. Due to the limited number of patients, we have to be careful in drawing conclusions.

Both the data of MR images and US images were assessed by a single radiologist. Consequently, no reproducibility data is available yet.

Until now denervation of the EAS muscle postoperatively has been determined by pudendal nerve terminal motor latencies (PNTML)[11, 17, 38-42]. In view of the lack of correlation between PNTML, fiber density and outcome, the use of neurophysiological evaluation may be questioned[43]. A consequence of prolonged pudendal nerve latencies is EAS atrophy. Endoanal MR imaging is able to accurately depict EAS atrophy. The fact that we could not find a relation between postoperative depicted atrophy and surgical failure was in contradiction with our prior expectations. It is plausible that the exclusion from surgery of patients with extreme generalized atrophy has narrowed the disease spectrum to such an extent that it is difficult to show any relation.

This study showed that advanced age (over 65 years) was associated with a poor outcome. The associations between age and the results of sphincter repair have not been elucidated yet[2]. Although in the literature there is a trend towards younger patients with a favorable outcome, this study states that only when patients are older than 65 years there is a significant difference in outcome[44-47].

In conclusion we have demonstrated that preoperatively performed endoluminal imaging might function as a potential predictor of surgical outcome and that certain postoperative findings at endoluminal imaging are associated with poor surgical outcome. Endoanal MR imaging is predominantly useful in the determination of EAS thickness and structure and endoanal US is predominantly effective in the postoperative assessment for depiction of residual EAS defects. Further research is needed to answer remaining questions about failure of anterior anal sphincter repair.

References

1. Kamm MA. Faecal incontinence. *BMJ* 1998;316(7130):528-532
2. Baig MK, Wexner SD. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2000;87:1316-1330
3. Madoff RD, Parker SC, Varma MG, Lowry AC. Faecal incontinence in adults. *The Lancet* 2004; 364:621-632
4. Barisic G, Krivokapic Z, Markovic V, Popovic M, Saranovic D, Marsavelska A. The role of overlapping sphincteroplasty in traumatic fecal incontinence. *Acta Chir Iugosl* 2000;47(4 Suppl 1):37-41
5. Barisic GI, Krivokapic ZV, Markovic VA, Popovic MA. Outcome of overlapping anal sphincter repair after 3 months and after a mean of 80 months. *Int J Colorectal Dis* 2006;21:52-56
6. Halverson AL, Hull TL. Long-term outcome of overlapping anal sphincter repair. *Dis Colon Rectum* 2002;45:345-348
7. Karoui S, Leroi AM, Koning E, Menard JF, Michot F, Denis P. Results of sphincteroplasty in 86 patients with anal incontinence. *Dis Colon Rectum* 2000;43:813-820
8. Malouf AJ, Norton CS, Engel AF, Nicholls RJ, Kamm MA. Long-term results of overlapping anterior anal-sphincter repair for obstetric trauma. *The Lancet* 2000;355:260-265
9. Bravo GA, Madoff RD, Lowry AC, Parker SC, Buie WD, Baxter NN. Long-term results of anterior sphincteroplasty. *Dis Colon Rectum* 2004;47:727-731
10. Madoff RD. Surgical treatment options for fecal incontinence. *Gastroenterology* 2004;126(1 Suppl 1): S48-S54
11. Ternent CA, Shashidharan M, Blatchford GJ, Christensen MA, Thorson AG, Sentovich SM. Transanal ultrasound and anorectal physiology findings affecting continence after sphincteroplasty. *Dis Colon Rectum* 1997;40:462-467
12. Savoye-Collet C, Savoye G, Koning E, Thoumas D, Michot F, Denis P et al. Anal endosonography after sphincter repair: specific patterns related to clinical outcome. *Abdominal Imaging* 1999;24:569-573
13. Vaizey CJ, Norton C, Thornton MJ, Nicholls RJ, Kamm MA. Long-term results of repeat anterior anal sphincter repair. *Dis Colon Rectum* 2004;47:858-863
14. Pinedo G, Vaizey CJ, Nicholls RJ, Roach R, Halligan S, Kamm MA. Results of repeat anal sphincter repair. *Br J Surg* 1999;86:66-69
15. Bartram CI. Ultrasound. In: Bartram CI, DeLancy JOL, Halligan S, Kelvin FM, Stoker J (eds). Imaging pelvic floor disorders. Berlin Heidelberg New York: Springer-Verlag, 2003;69-80
16. Stoker J, Halligan S, Bartram CI. Pelvic floor imaging. *Radiology* 2001;218:621-641
17. Briel JW, Stoker J, Rociu E, Lameris JS, Hop WCJ, Schouten WR. External anal sphincter atrophy on endoanal magnetic resonance imaging adversely affects continence after sphincteroplasty. *Br J Surg* 1999;86:1322-1327
18. Briel JW, Zimmerman DD, Stoker J, Rociu E, Lameris JS, Mooi WJ et al. Relationship between sphincter morphology on endoanal MRI and histopathological aspects of the external anal sphincter. *Int J Colorectal Dis* 2000;15:87-90
19. Briel JW, Zimmerman DD, Schouten WR. Factors predictive of outcome after surgery for faecal incontinence. *Br J Surg* 2001;88:729-730
20. Stoker J, Rociu E. Endoluminal MR imaging of anorectal diseases. *J Magn Reson Imaging* 1999;9:631-634

21. Terra MP, Dobben AC, Berghmans B, Deutekom M, Baeten CG, Janssen LW et al. Electrical stimulation and pelvic floor muscle training with biofeedback in patients with fecal incontinence: a cohort study of 281 patients. *Dis Colon Rectum* 2006;49:1149-1159

22. Vaizey CJ, Carapeti E, Cahill JA, Kamm MA. Prospective comparison of faecal incontinence grading systems. *Gut* 1999;44:77-80

23. Stoker J, Rociu E, Zwamborn AW, Schouten WR, Lameris JS. Endoluminal MR imaging of the rectum and anus: technique, applications, and pitfalls. *Radiographics* 1999;19:383-398

24. Diamant NE, Kamm MA, Wald A, Whitehead WE. AGA technical review on anorectal testing techniques. *Gastroenterology* 1999;116:735-760

25. Rociu E, Stoker J, Zwamborn AW, Lameris JS. Endoanal MR imaging of the anal sphincter in fecal incontinence. *Radiographics* 1999;19:S171-S177

26. Terra MP, Beets-Tan RG, van der Hulst VP, Dijkgraaf MG, Bossuyt PM, Dobben AC et al. Anal Sphincter Defects in Patients with Fecal Incontinence: Endoanal versus External Phased-Array MR Imaging. *Radiology* 2005;236:886-895

27. Stoker J, Bartram CI, Halligan S. Imaging of the posterior pelvic floor. *Eur Radiol* 2002; 12:779-788

28. Rociu E, Stoker J, Eijkemans MJC, Lameris JS. Normal anal sphincter anatomy and age- and sex-related variations at high-spatial-resolution endoanal MR imaging. *Radiology* 2000; 217:395-401

29. Briel JW, de Boer LM, Hop WC, Schouten WR. Clinical outcome of anterior overlapping external anal sphincter repair with internal anal sphincter imbrication. *Dis Colon Rectum* 1998; 41:209-214

30. Slade MS, Goldberg SM, Schottler JL, Balcos EG, Christenson CE. Sphincteroplasty for acquired anal incontinence. *Dis Colon Rectum* 1977;20:33-35

31. Felt-Bersma RJF, Cuesta MA, Koorevaar M. Anal sphincter repair improves anorectal function and endosonographic image - A prospective clinical study. *Dis Colon Rectum* 1996;39:878-885

32. Felt-Bersma RJ, van Baren R, Koorevaar M, Strijers RL, Cuesta MA. Unsuspected sphincter defects shown by anal endosonography after anorectal surgery. A prospective study. *Dis Colon Rectum* 1995;38:249-253

33. Nielsen MB, Dammegaard L, Pedersen JF. Endosonographic assessment of the anal sphincter after surgical reconstruction. *Dis Colon Rectum* 1994;37:434-438

34. Mavrantonis C, Wexner SD. A clinical approach to fecal incontinence. [Review]. *Journal of Clinical Gastroenterology* 1998;27:108-121

35. Bharucha AE. Fecal incontinence. *Gastroenterology* 2003;124:1672-1685

36. Malouf AJ, Chambers MG, Kamm MA. Clinical and economic evaluation of surgical treatments for faecal incontinence. [Review]. *Br J Surg* 2001;88:1029-1036

37. Baxter NN, Rothenberger DA, Lowry AC. Measuring fecal incontinence. *Dis Colon Rectum* 2003;46:1591-1605

38. Wexner SD, Marchetti F, Jagelman DG. The role of sphincteroplasty for fecal incontinence reevaluated: a prospective physiologic and functional review. *Dis Colon Rectum* 1991; 34(1):22-30

39. Tjandra JJ, Han WR, Goh J, Carey M, Dwyer P. Direct repair vs. overlapping sphincter repair: a randomized, controlled trial. *Dis Colon Rectum* 2003;46:937-942

40. Gilliland R, Altomare DF, Moreira H, Jr., Oliveira L, Gilliland JE, Wexner SD. Pudendal neuropathy is predictive of failure following anterior overlapping sphincteroplasty. *Dis Colon Rectum* 1998;41:1516-1522

41. Oliveira L, Pfeifer J, Wexner SD. Physiological and clinical outcome of anterior sphincteroplasty. *Br J Surg* 1996;83:502-505

42. Londono-Schimmer EE, Garcia-Duperly R, Nicholls RJ, Ritchie JK, Hawley PR, Thomson JP. Overlapping anal sphincter repair for faecal incontinence due to sphincter trauma: five year follow-up functional results. *Int J Colorectal Dis* 1994;9:110-113

43. Osterberg A, Edebol Eeg-Olofsson K, Graf W. Results of surgical treatment for faecal incontinence. *Br J Surg* 2000;87:1546-1552

44. Rasmussen OO, Colstrup H, Lose G, Christiansen J. A technique for the dynamic assessment of anal sphincter function. *Int J Colorectal Dis* 1990;5:135-141

45. Nikiteas N, Korsgen S, Kumar D, Keighley MR. Audit of sphincter repair. Factors associated with poor outcome. *Dis Colon Rectum* 1996;39:1164-1170

46. Osterberg A, Graf W, Holmberg A, Pahlman L, Ljung A, Hakelius L. Long-term results of anterior levatorplasty for fecal incontinence - A retrospective study. *Dis Colon Rectum* 1996;39:671-674

47. Sitzler PJ, Thomson JP. Overlap repair of damaged anal sphincter. A single surgeon's series. *Dis Colon Rectum* 1996;39:1356-1360

Chapter 11

Plugs for containing fecal incontinence

Marije Deutekom
Annette C. Dobben

Cochrane Database of Systematic Reviews 2005;(3):CD005086

Abstract

BACKGROUND. Fecal incontinence is a distressing disorder with high social stigma. Not all people with fecal incontinence can be cured with conservative or surgical treatment and they may need to rely on containment products, such as anal plugs.
The objective was to assess the performance of different types of anal plugs for containment of fecal incontinence.

METHODS.
Search strategy. We searched the Cochrane Incontinence Group Specialized Register (searched 22 November 2004), MEDLINE (January 1966 to November 2004), CINAHL (January 1982 to November Week 3 2004), EMBASE (January 1996 to 2004 Week 47), INVERT (Dutch nursing database) (January 1993 to November 2004) and Web of Science (January 1988 to November 2004). Reference lists of identified trials were searched and plugs manufacturers were contacted for trials. No language or other limitations were imposed.

Selection criteria. Types of studies: This review was limited to randomized and quasi-randomized controlled trials (including crossovers) of anal plug use for the management of fecal incontinence.
Types of participants: Children and adults with fecal incontinence.
Types of interventions: Any type of anal plug. Comparison interventions might include no treatment, conservative (physical) treatments, nutritional interventions, surgery, pads and other types or sizes of plugs.

Data collection and analysis. Two reviewers independently assessed methodological quality and extracted data from the included trials. Authors of all included trials were contacted for clarification concerning methodological issues.

RESULTS. Four studies with a total of 136 participants were included. Two studies compared the use of plugs versus no plugs, one study compared two sizes of the same brand of plug, and one study compared two brands of plugs. In all included studies there was considerable dropout (in total 48 (35%) dropped out before the end of the study) for varying reasons. Data presented are thus subject to potential bias.
'Pseudo-continence' was, however, achieved by some of those who continued to use plugs, at least in the short-term. In a comparison of two different types of plug, plug loss was less often reported and overall satisfaction was greater during use of polyurethane plugs than polyvinyl-alcohol plugs.

CONCLUSION. The available data were limited and incomplete, and not all pre-specified outcomes could be evaluated. Consequently, only tentative conclusions are possible. The available data suggest that anal plugs can be difficult to tolerate. However, if they are tolerated they can be helpful in preventing incontinence. Plugs could then be useful in a selected group of people either as a substitute for other forms of management or as an adjuvant treatment option. Plugs come in different designs and sizes; the review showed that the selection of the type of plug can impact on its performance.

Introduction

Fecal incontinence is defined as the involuntary passage of fecal material through the anal canal[1]. The reported prevalence values range from 1.4% in the general population (defined as soiling of underwear, outer clothing, furnishing, or bedding several times a month or more often)[2] to 46% in institutionalized elderly people (defined as at least one incontinent episode per week)[3]. It is possible that the real prevalence is even higher than reported as fecal incontinence is associated with high social stigma and people are reluctant to seek help for this disorder because of embarrassment[4, 5].

The causes for fecal incontinence are diverse. In most cases a combination of factors leads to incontinence. Frequently cited causes are injuries during childbirth and prior anorectal surgery[6, 7]. But many other causes have been described, including loose stool, intestinal hurry, and neurological disease or injury.

Treatments available range from conservative therapy, such as dietary recommendations and anti-diarrhoeal medication, to surgical treatment by either sphincter repair, dynamic graciloplasty, artificial anal sphincter implantation, or sacral nerve stimulation[4, 8]. Nowadays, the most common treatments are pelvic floor muscle training - with or without biofeedback - and anterior anal sphincter repair[6]. The reported success rates with these forms of treatment vary, but it is recognized that none of the treatments will resolve the incontinence problems in all patients.

Where incontinence persists despite active treatment there may be no option other than containment. Brazzelli et al[9] have reviewed the use of pads for the containment of anal and urinary incontinence. Problems when using pads for fecal incontinence are that the odour from the anal leakage is difficult to control and extensive use of pads can result in skin condition problems. A possible way to avoid these problems is the use of an anal plug (sometimes called 'tampon'): a device specially developed for containing fecal incontinence.

Different types of anal plugs are known, all aiming to block the loss of stool. They were first used in patients suffering from fecal incontinence due to major neurological problems, such as caused by spina bifida[10]. Nowadays, plugs are also sometimes used by patients with fecal incontinence who do not have an underlying neurological condition.

At this point it is unclear how effective anal plugs are in controlling stool loss in patients with fecal incontinence (with or without neurological impairments) and whether some types of anal plugs are more effective than others. This review aims to bring together in a systematic way the best available evidence to address these issues.

Objectives

To assess the performance of different types of anal plugs for containment of fecal incontinence.

The following comparisons were considered:

1. anal plugs versus no plugs
2. one type of anal plug versus another
3. anal plugs versus any other treatment

Methods

Criteria for considering studies for this review

Types of studies

This review was limited to randomized and quasi-randomized controlled trials (including crossovers) of anal plug use for the management of fecal incontinence.

Types of participants

All patients (children and adults) with fecal incontinence.

Types of intervention

Studies investigating the relative performance of anal plugs. Potential comparison interventions include no treatment, conservative (physical) treatments, nutritional interventions, surgery, pads, and other types or sizes of plugs.

Types of outcome measures

1. Patient symptoms
   frequency of incontinence of stool or flatus (diary or self-report)
   degree of incontinence (e.g. stool weight)
   incontinence score
   episodes of anal urgency
2. Physical measures
   achievement of pseudo-continence (continence only while wearing a plug)
   wearing time and frequency of use
   leakage rate
   odor control
3. Patient satisfaction
   satisfaction with incontinence controlling capacity
   tolerability of plug (including persistence in using the plug)
   comfort of plug in use
   comfort of plug removal/ease of removal
   feeling of cleanness
4. Health status measures
   impact of incontinence on health status, social life, and quality of life
5. Costs
6. Other outcomes
   non pre-specified outcomes later judged important when performing the review

Search methods for identification of studies

We formulated a comprehensive and exhaustive search strategy in an attempt to identify all relevant studies regardless of language or publication status (published, unpublished, in press, and in progress).

This review has drawn on the search strategy developed for the Cochrane Incontinence Group. Relevant trials were identified from the Group's specialized register of trials, which is described under the Cochrane Incontinence Group's details in *The Cochrane Library*. The register contains trials identified from MEDLINE, CINAHL, The Cochrane Central Register of Controlled Trials (CENTRAL), and hand searching of journals and conference proceedings.

For this review the authors performed the following additional searches. All searches were carried out on 26 November 2004. The following electronic bibliographic databases were searched: MEDLINE (January 1966 to November 2004); CINAHL (January 1982 to November Week 3 2004); EMBASE (January 1996 to 2004 Week 47); INVERT (Dutch nursing database - Index van de Nederlandstalige Verpleegkundige Tijdschriftliteratuur) (January 1993 to November 2004); andWeb of Science (January 1988 to November 2004).

The following search terms were used:

1.tampon*
2.plug*
3.incontinen*
4.stool*
5.faec*
6.fecal incontinence/ [mesh]
7.atus [mesh] OR anal [mesh]
8.anus OR anal
9.(1 or 2) AND (3 or 4 or 5 or 6 or 7 or 8)
* = truncation symbol

Additionally all reference lists of identified trials were searched. We contacted two manufacturers that marketed plugs to ask for details of unpublished or ongoing trials. We did not impose any language or other limitations on the searches.

Methods of the review

Study selection

Two reviewers assessed the title and abstract of references identified by the search strategy. The full reports of all potentially eligible randomized and quasi-randomized controlled trials were then obtained for further assessment of eligibility. Any disagreements were resolved by discussion. Studies were only included if they were randomized or quasi-randomized trials.

Methodological quality assessment

The quality of eligible trials was assessed independently by the two reviewers using a pre-defined quality assessment form (see details under the Incontinence Group in *The Cochrane Library*). Reviewers were not blind to author, institution or journal. Disagreements between reviewers were resolved by discussion. Studies were not excluded from the review on the basis of methodological quality.

Data abstraction

Relevant data regarding inclusion criteria (study design, participants, interventions and outcomes), quality criteria (randomization and blinding), and results were extracted independently by the two reviewers using a data abstraction form adapted from the form designed by the Dutch Cochrane Center. In cases where insufficient data were reported authors were contacted for further information (such as method of randomization, statistical methods).

Data analysis

Data were analyzed using the MetaView statistical software in Review Manager (RevMan 4.2.5). For dichotomous variables, relative risks RR and 95% confidence intervals (CI) were derived for each outcome. It was not possible to combine data from the included studies as outcomes and type of comparisons varied. We instead present a qualitative synthesis of the results of the primary studies.

Description of studies

The search strategy identified 13 potentially eligible studies. When full citations were obtained nine studies could not be included: seven were patient series, one was a case study, and one study was excluded as we understood from the author that this paper did not report a randomized trial. Thus in total, four studies met our inclusion criteria[10-13]. Two of these studies were derived from the specialized trials register of the incontinence group[11, 12]. One was derived by the additional searches performed by one of the authors[10]. The final trial was obtained by contacting an anal plug manufacturer[13].

The reports of two of the included trials had not been published at the time of finishing the review[12, 13]. We received permission from the authors to use their data in our review.

The total number of participants across the trials was 136. For a detailed description of individual studies please refer to the Table 1.

Design

Three studies used a randomized crossover designs[10, 11, 13] and one was a parallel group randomized controlled trial[12].

Sample size

Sample sizes were 16[13], 34[10], 38[11], and 48[12].

Diagnosis

All studies included patients with fecal incontinence. One study included patients who were partially continent or incontinent following imperforate anus repair[11]. One study included children who had fecal incontinence due to a high type imperforate anus and children with spina bifida[13]. One study included children (greater than 4 years) and young adults (16-45 years) who were incontinent due to congenital or acquired neurogenic disorders[12], and one included adult outpatients after failure of previous treatment[10].

Location/setting

One trial was carried out in Scotland and participants were identified primarily by hospital specialists from Paediatric Surgery or Gastroenterology in Aberdeen, Inverness and Glasgow[12]. One trial was carried out in Germany in a hospital for Paediatric Surgery[11], one in Belgium at the departments of Paediatrics and Urology in an academic medical center[13], and one in England in a specialist colorectal hospital where patients received an individual instruction with a nurse specialist[10].

Interventions

The four identified trials made the following comparisons:

1. anal plug versus no plug[12, 13]
2-I. one type of anal plug versus another: comparison of two sizes of the same type of plug (poly-urethane anal plug)[10]
2-II. one type of anal plug versus another: comparison of two different types of plugs (poly-urethane anal plug versus polyvinylalcohol plug)[11]

Length of treatment

Three trials lasted between four and six weeks[10, 11, 13]. One trial lasted one year[12].

Outcomes

Common reported outcomes were frequency of incontinent episodes (effectiveness of treatment), satisfaction and tolerance.

Methodological quality

Potential for selection bias at trial entry

In all crossover trials the order of the intervention was randomized [10, 11, 13]. In none of these studies were details provided concerning the methods used for randomization and concealment. In the parallel group randomized controlled trial the participants were randomly allocated to the intervention or control group[12]. Randomization was performed using pre-determined codes.

Potential for bias at time of treatment or outcome assessment

As the studies included in this review investigated anal plugs it is difficult to blind patients and staff to intervention. In two studies the use of plugs was compared to a control intervention in which patients did not receive any treatment[12, 13]. Blinding was impossible. In the remaining two randomized crossover studies two types or two sizes of plugs were compared[10, 11]. Both studies did not report any blinding.

Potential for bias in trial analysis

In three studies the number and reasons for patient dropouts were clearly described[10, 11, 13].
In one study[10] 23 of the 34 (68%) patients did not start or dropped out. Reasons why patients dropped out were: they disliked the idea and did not start the study (n=4), they failed to attend the clinic (n=2), they dropped out because of discomfort after trying the first plug (n=8); and dropped out after trying one size of plug refusing to try the second one (n=9).

In one study [11] 15 of the 38 patients (39%) dropped out before the end of the study. Two patients liked the first tested product and ended participation, six patients found the smallest available size of the products, tested first, too big, two patients reported discomfort, one patient constantly lost one of the products and four patients failed to complete the protocol for non-plug related reasons.

In one study[13] 4 of the 16 patients dropped out (25%). Reported reasons for this were discomfort and pain (n=2) and losing the plug (n=2).

In one study[12] 6 patients dropped out from the 48 included, but did not report any reasons for this. All those who dropped out were in the intervention group.

In only one of the studies was an intention to treat analysis performed[13]. In none of the trials was there a description whether data-analysis was performed blindly.

Results

Three randomized crossover trials and one randomized controlled trial were included in this review. As the reported outcome measures varied amongst trials, a quantitative synthesis of the results was not feasible. Unfortunately the data from the randomized crossover studies were not presented in a form suitable for inclusion in the formal analysis.

Comparison 1: anal plugs versus no plugs

Two of the included studies compared the use of anal plugs with standard treatment[12, 13]. In both studies patients were allowed to choose their preferred size of plug. In both trials a choice could be made between small or larger Coloplast plugs.

Patient symptoms

Pseudo continence was reported in six out of 16 patients in the treatment period in the crossover study, and in none of the 16 during the control period. Patients achieving pseudo continence were reported to show greater satisfaction with treatment during plug use (no further data provided by the author) than when not using a plug[13]. Three of the 16 patients (two with anal atresia and one with spina bifida) continued using the plug after the study. Neither stool frequency nor stool consistency was affected by use of the plug (no further data provided by the author). In the parallel group trial, clinically derived condition-specific measures (such as protection, rash/skin problems, and unpleasant odor) tended to favor the plugs group for all patients (adults and children)[12], although no difference was statistically significant, confidence intervals were wide, and dropout rates were considerable.

Patient satisfaction

In the randomized crossover trial[13] four patients did not complete the treatment period due to discomfort and pain (n=2; anal atresia) and losing of the plug (n=2; spina bifida). The plug was thus not tolerated in four out of 16 patients. All patients tolerated the control period.

Health status measures

Bond and colleagues[12] reported data for adults on changes in general health (3/14 versus 0/5; RR: 2.63; 95% 0.16 to 43.63), bodily pain (6/15 versus 3/5; RR: 0.67; 95% 0.26 to 1.72) and various measures of well being (derived from SF-36). Confidence intervals were all wide, reflecting the small numbers studied.

Costs

Little or no evidence was obtained that the plug led to significant reductions in the overall costs of care[12].

Comparison 2-I: one type of anal plug versus another (comparison of two sizes)

One study[10] compared two sizes of a plug in a randomized crossover design. Due to the high dropout in this study and the incomplete data, no results concerning the comparison are available.

Comparison 2-II: one type of anal plug versus another (comparison of two types)

One study[11] compared two types of plugs in a randomized crossover design: Polyurethane anal plug (Conveen, Coloplast) (PU plug) versus EFF-EFF polyvinyl-alcohol plug (Med. SSE-System) (PVA plug).

Patient symptoms

The absence of soiling episodes was reported in 15 (65%) patients when using the PU plug and by 14 (60%) patients when using the PVA plug.

Patient satisfaction

Feelings of security were reported by 16 patients (69%) while using the PU plug and by 10 patients (43%) when using the PVA plug. Loss of plug was reported by 7 patients (30%) with the PU plug and by 15 patient 65% with the PVA plug. Inconvenience was reported by 9 patients (39%) when using the PU plug and by 16 (69%) patients when using the PVA plug. Overall satisfaction, defined as patients' opinion that the plug was good to very good, was reported more often for the PU plug (n=17) than for the PVA plug (n=8). 14 patients preferred the PU plug, 5 patients preferred the PVA plug, and 4 patients reported no preference.

No data were available for the other pre-specified outcomes.

Comparison 3: anal plug versus any other treatment

No eligible trials were found.

Discussion

This review of anal plugs for the containment of fecal incontinence was limited by the small quantity of eligible studies and participants and the fact that combining data was either impossible or inappropriate. Only one parallel group randomized controlled trial that compared the use of plugs with no intervention could be included[12]. The other three included trials were randomized crossover studies. These also reported dropouts and are further limited by not allowing longer-term acceptability rates to be assessed. One such trial compared plug use versus no intervention[13]. The other two studies compared two types of plugs. Two sizes of the same plug were investigated in one study[10] and two brands of plugs were compared in the other study[11].

Reported outcome measures varied. Two studies reported patient symptoms[11, 13]; one study reported physical measures[12]; two studies reported patient satisfaction[11, 13] and one study reported the outcome of health status measures and costs[12]. There are other variables that may influence the successful wearing of anal plugs, such as leakage rate, skin problems, odor, wearing time, frequency of use, age, social environment and patient characteristics. Unfortunately there were insufficient data in the included studies to take these factors in consideration in our review.

Participant groups also varied between the studies. The participants in two studies[11, 13] suffered from fecal incontinence due to congenital diseases. These patients are a minority in the total population of patients with fecal incontinence.

Due to the diversity in comparisons, outcome measures and type of participants we were not able to perform a quantitative synthesis of the data but described the data per comparison. This does not allow us to state firm and precise conclusions and emphasizes the need for further research.

The methodological quality of the four included trials was generally poor. Inclusion and exclusion criteria were given in two studies[12, 13], and one study described only inclusion criteria[11]. However, none of the studies provided outcomes related to severity and / or frequency of fecal incontinence.

Concealment of allocation was performed in only one study[12]. Due to the nature of the intervention of the studies it appeared to be impossible to blind the patients or outcome assessors. Only one trial reported that the researcher who was responsible for the inclusion of patients was securely blinded to the randomization process[12].

Incompleteness of follow up occurred in most trials, caused by selective withdrawal of patients, to a large extent related to intolerance or dissatisfaction with the intervention. An intention to treat analysis could be performed in only one trial[13]. In the study comparing two sizes of anal plugs the high rate of dropout meant that it was not possible to extract data[10]. However, it did not appear that there was a difference in dropout rates between studies or patient groups. Most trials studied small patient groups, limiting the power to detect differences between groups.

One trial excluded data from patients who reported that they did not have difficulties with these particular outcomes before or after the intervention[12]. Thus, Bond only reported comparisons between the intervention group and control group when the outcome troubled the patient and it was reported as the same, improved or worse. The results presented in this review included data from all the patients. Consequently, our results differ from those published by the author.

Authors' conclusions

Implications for practice

This review has focused on the performance of anal plugs for containment of fecal incontinence. The available data were limited and incomplete and not all pre-specified outcomes could be evaluated. Consequently, we can only draw tentative conclusions.

The available data suggest that anal plugs can be difficult to tolerate. For those who do persist, the limited evidence available suggests that plugs may be helpful in alleviating the problems caused by incontinence. In a minority of people with fecal incontinence, plugs could be useful either as a substitute for other forms of management or as an adjuvant treatment option. Plugs come in different designs and sizes; the review showed that the selection of the type of plug can impact on its performance. Specifically, a polyurethane plug performed better than a polyvinyl-alcohol plug in one trial.

Implications for research

This review has illustrated how difficult it is to undertake rigorous evaluations of devices for incontinence. We were only able to extract data from three trials, of which two trials presented small patients groups. Crossover designs are attractive for chronic conditions like intractable fecal incontinence, but the trials reviewed showed high dropout rates (to the extent that in one trial no useful information was generated), and anyway do not allow longer-term performance to be evaluated. Whether or not people persist with using a plug is likely to be a good indication of its value. However, the single parallel group randomized trial that could potentially address this issue illustrated the difficulties of using this design: recruitment proved difficult, and there were more dropouts in the group allocated plugs. This could, therefore, be a scenario where high quality observational studies could provide useful information. The strength of a randomized controlled trial, however, is that it allows a more reliable assessment of what would happen without the use of plugs; this would clearly be preferable if problems with compliance and retention in the study could be resolved.

There are lessons to be learnt for the design and conduct of future trials. Better reporting of study methods and detailed descriptions of interventions is a necessity. Future studies should describe their procedure for blinding, especially in studies where it is quite difficult, if not impossible, to maintain true blinding. Patients will be aware of wearing a plug, and this may affect outcome. While tolerance is a key point in the willingness to use an anal plug, the reasons for patient withdrawal and dropout must be specified and the long-term effects need to be evaluated. Both intention-to-treat analysis and per-protocol analysis should be performed. Sensitivity analyses should explore sensible assumptions about those participants with missing data, for example due to dropout.

When evaluating different plugs, investigators should take into account costs and quality of life, as well as other, specific advantages and disadvantages of plug use. To avoid bias from socially desired answers from participants, advantages and disadvantages of plug use should be elicited by an independent researcher.

Table 1. Characteristics of included studies

Study	Methods	Participants	Interventions
Bond 2005	Randomized controlled trial (2:1)	48 patients (28 childeren; age >4 years and 20 adults; age 16-45 years)	Poly-urethane anal plug (Conveen, Coloplast; 2 sizes) No intervention
Norton 2001	Randomized cross-over trial	Adult out-patients (n=34) attending a specialist colorectal hospital after failure of previous treatment	Poly-urethane anal plug (Conveen, Coloplast) 37 mm diameter when open Poly-urethane anal plug (Conveen, Coloplast) 45 mm diameter when open
Pfrommer 2000	Randomized cross-over trial	38 partially continent or incontinent patients following imperforate anus repair (age > 6-15)	Poly-urethane anal plug (Conveen, Coloplast) (size closed / open diameter of 14.5/38 mm or 15.5/45 mm) EFF-EFF polyvinyl-alcohol plug (Med. SSE-System) (diameters ranging from 15 mm to 38 mm. Used size dependent on anal canal diameter)
Van Winckel 2005	Randomized cross-over trial	7 patients (age 4 to 12; 3 girls) with high type of imperforate anus and 9 patients with spina bifida (age 6 to 13; 2 girls)	Poly-urethane anal plug (Conveen, Coloplast) (size 12 or 13 mm; depending on preference. No intervention

Outcomes	Notes
Functional Status II-R-SF-36 Patient Generated Index of Quality of Life Care Generated Index of Quality of Life Dartmouth COOP Charts Condition-specific measure developed for research Qualitative data Health service utilization data Costs data Evaluation of education package	32 (16 children and 15 adults) allocated plugs; 16 (11 childeren and 5 adults) allocated to no plugs. 6 participants did not complete the trial (5 in plug group and 1 in control group)
Comfort of inserting plug Comfort of plug in use Comfort of taking plug out Capacity of controlling fecal leakage Preferences Patient characteristics which predict when the plug will help the most	Of the 34 patients offered the plug 4 refused as they disliked the idea; 2 failed to attend; 8 dropped out after trying first plug, because of discomfort and 9 dropped out after trying one size of plug refusing to try the second size. 11 patients completed the protocol
Stool consistence Awareness of repletion Effectiveness of treatment Feeling of security Loss of plug Inconvenience Overall satisfaction	Drop-out: 2 patients liked first tested product 6 patients found the smallest available size of the products tested first too big 2 patients reported discomfort 1 patient constantly lost one of the products 4 patients failed to complete the protocol for non-plug related reasons
Number of stools Number of soiling episodes Number of diapers or pads used Number of plugs Satisfaction	2 patients (with imperforate anus) dropped out due to discomfort and pain 2 patients (with spina bifida) dropped out because of losing the plug

Acknowledgements

We are grateful to all members of the Cochrane Incontinence Group in Aberdeen (especially to June Cody and Sheila Wallace) for their assistance with the review. Thanks are also due to all authors and to suppliers of anal plugs for their cooperation. Finally we would like to thank Christine Norton for her useful comments on the protocol.

References

1. Soffer EE, Hull T. Fecal incontinence: a practical approach to evaluation and treatment. *Am J Gastroenterol* 2000;95:1873-1880
2. Perry S, Shaw C, McGrother C, Matthews RJ, Assassa RP, Dallosso H, et al. Prevalence of faecal incontinence in adults aged 40 years or more living in the community. *Gut 2002*;50:480-4
3. Borrie MJ, Davidson HA. Incontinence in institutions: costs and contributing factors. *Canadian Medical Association Journal* 1992;147:322-8
4. Jorge JM, Wexner SD. Etiology and management of fecal incontinence. *Dis Colon Rectum* 1993;36:77-97
5. Mavrantonis C, Wexner SD. A clinical approach to fecal incontinence.*Journal of Clinical Gastroenterology* 1998;27:108-21
6. Kamm MA. Faecal incontinence. *BMJ* 1998;316:528-32
7. Toglia MR. Pathophysiology of anorectal dysfunction. *Obstetrics and Gynecology Clinics of North America* 1998;25:771-81
8. Matzel KE, Bittorf B, Stadelmaier U, Hohenberger W. Sacral nerve stimulation in the treatment of faecal incontinence. *Chirurg* 2003;74:26-32
9. Brazzelli M, Shirran E, Vale L. Absorbent products for containing urinary and/or fecal incontinence in adults. *Journal of Wound, Ostomy and Continence Nursing* 2002;29:45-54
10. Norton C, Kamm MA. Anal plug for faecal incontinence. *Colorectal Disease* 2001;3:323-7
11. Pfrommer W, Holschneider AM, Lofer N, Schauff B, Ure BM. A new polyurethane anal plug in the treatment of incontinence after anal atresia repair. *European Journal of Pediatric Surgery* 2000;10:186-90
12. Bond C. The anal plug: an evaluation of a novel management option for faecal incontinence. Final report to Chief Scientist Office, Scottish Executive Health Department, Edinburgh 2005
13. Van Winckel M. Clinical evaluation of a new anal medical device to achieve faecal continence in spina bifida and anal atresia patients, 2005

Summary & Implications

Chapter 12

Summary

The major aim of the work in this thesis was to collect evidence for building an optimal diagnostic strategy for fecal incontinent patients. To reach this goal various topics were studied.

At present there is no consensus concerning the best diagnostic techniques for fecal incontinent patients in the Netherlands. In **chapter 2** we describe the variation in the Netherlands in the diagnostic work-up and treatment of patients with fecal incontinence. The results were based on a survey. The questionnaire comprised five sections: general information, selection of diagnostic tests, availability of diagnostic techniques, use of an incontinence score, and therapeutic options. In total 306 physicians were contacted from all 100 Dutch hospitals. The majority of the respondents reported the routine use of sigmoidoscopy. The imaging techniques that were most often applied were endoanal ultrasonography and defecography. Of all anorectal function tests, anorectal manometry was most frequently used. For most of the respondents sigmoidoscopy and defecography were available. The highest percentages of referral concerned endoanal ultrasonography and anorectal manometry. The Parks score was the most frequently used incontinence score was, followed by the more recently introduced Vaizey score. The preferred treatment option was pelvic floor rehabilitation, followed by dietary measures, medication and surgery.

The diagnostic work-up for fecal incontinent patients in general comprises anal inspection, digital rectal examination, anorectal function tests and imaging. It is not clear to what extent digital rectal examination and anal inspection contribute to the diagnostic work-up. In **chapter 3** we report a study in 312 fecal incontinent patients to determine their diagnostic yield. Patients underwent anal inspection and digital rectal examination where after findings were compared with results of anorectal manometry, anal sensation measurement, and endoanal ultrasonography. Absent, decreased and normal resting and squeeze pressures at digital rectal examination correlated to some extent with manometric findings. External anal sphincter defects at digital rectal examination were confirmed with endoanal ultrasonography for defects smaller than 90 degrees in 36%, for defects between 90 and 150 degrees in 61%, and for defects between 150 and 270 degrees in 100%. Patients with anal scar tissue at anal inspection had lower incremental squeeze pressures and patients with a gaping anus had lower resting pressures at anorectal manometry.

With reproducibility studies the extent of agreement in assessment can be determined. The primary aim of the study presented in **chapter 4** was to determine the interobserver agreement of defecography in diagnosing enterocele, anterior rectocele, intussusception and anismus in fecal incontinent patients. The subsidiary aim was to evaluate the influence of different levels of experience on the assessments. Defecographies were performed in 105 consecutive fecal incontinent patients. The observers were classified by different levels of experience and were compared to the findings from an expert radiologist. The quality of the expert radiologist was evaluated by an intra-observer agreement procedure.

The intra-observer agreement was good to very good, except for anismus. The interobserver agreement for enterocele and rectocele were good, and fair for intussusception. Incomplete evacuation for more than 30 seconds appeared to have a moderate interobserver agreement,

and a fair interobserver agreement was observed for puborectalis impression. The interobserver agreement in grading enterocele and rectocele was good, and reproducibility was fair for intussusception. The agreement, stratified by experience levels, was very good for rectocele and its grading and moderate for intussusception at the most experienced level. For enterocele and its grading different experience levels did not influence the reproducibility.

An "educational exhibit" is a type of paper that introduces the reader to a specific topic. **Chapter 5** describes a series of issues related to endoluminal imaging in fecal incontinent patients, including practical aspects. The present consensus in diagnosing disorders of the external anal sphincter stipulates that endoanal ultrasonography and endoanal MR imaging are equivalent. In the selection of patients for anterior anal sphincter repair, the principal advantage of endoanal MR imaging is its ability to determine external sphincter atrophy. Atrophy of the external anal sphincter is a negative predictor for surgical outcome. In the post surgical work-up endoanal ultrasonography is useful to depict residual sphincter defects as a cause of failure of the repair.

The use of external phased array MR imaging and three-dimensional endoanal ultrasonography has recently been studied in the evaluation of fecal incontinence with favorable results.

Initial treatment options for fecal incontinence are conservative and include medication and dietary measures. In case of failure, pelvic floor rehabilitation is an additional conservative therapy, but evidence justifying this primary role is sparse. In **chapter 6** we report the outcome of standardized pelvic floor rehabilitation in 281 fecal incontinent patients. The treatment program comprised nine sessions of electrical stimulation and / or pelvic floor muscle training with biofeedback. The outcome of treatment was evaluated by the Vaizey score, anorectal manometry and rectal capacity measurement. After therapy the mean Vaizey score was significantly lower, indicating improvement ($p< 0.001$). However, only 13% of the patients had improved substantially (≥ 50% reduction in Vaizey score). The mean squeeze pressure and maximal tolerated volume changed significantly, but resting pressure, sensory threshold and urge sensation did not. Subgroup analyses did not show substantial differences in effects of pelvic floor rehabilitation.

Chapter 7 contains a study of anorectal function changes after pelvic floor rehabilitation. We extensively documented functional changes in 266 fecal incontinent patients and evaluated whether these changes, if any, were associated with changes in fecal incontinence score. Patients underwent anorectal manometry, anal and rectal mucosal sensitivity measurement, and rectal capacity measurement at baseline and after nine sessions of standardized pelvic floor rehabilitation. After therapy the squeeze pressure had increased significantly, as well as the urge sensation threshold, and maximum tolerated volume. The extent of improvement was not associated with age, duration of fecal incontinence, menopause or endoanal ultrasonography findings. All other anorectal functions had not changed. Improvement of the Vaizey incontinence score was weakly associated with changes in incremental squeeze pressure and in anal mucosal sensitivity.

Pelvic floor rehabilitation does not provide the same degree of relief in all fecal incontinent patients. If diagnostic tests could be used to predict the Vaizey score after treatment, we would be able to withhold pelvic floor rehabilitation from patients wit a low chance of success. The study in 250 patients in **chapter 8** was designed to identify elements from patient's medical history, physical examination, anorectal function tests, and imaging tests that could predict the Vaizey score after treatment. Linear regression analysis of the post-treatment Vaizey score was used to identify candidate predictor variables and to construct a multivariable prediction model. The initial prediction model included elements from medical history only. Subsequently, to calculate the added value of elements derived from tests above the variables identified from medical history, different models were built. A final model was built combining both the predictors from medical history and the predictors from all additional tests. This complete model resulted in a R^2 of 0.23 (p=0.02), which indicates that it can only play a limited role in predicting success of pelvic floor rehabilitation.

Currently both endoanal ultrasonography and endoanal MR imaging are to be considered as adequate tools for the depiction of external anal sphincter defects. Only a few comparative studies of these techniques have been published. In the study reported in chapter 9 endoanal ultrasonography and endoanal MR imaging were prospectively compared with respect to their ability for depicting external anal sphincter defects. The study was performed in a cohort of 237 fecal incontinent patients and operative findings were used as reference standard in a sub cohort consisting of 36 patients. The agreement between endoanal ultrasonography and endoanal MR imaging in the main cohort was fair for the depiction of external sphincter defects. There was no significant difference between endoanal ultrasonography and endoanal MR imaging in the depiction of external sphincter defects in the sub cohort. Sensitivity and positive predictive value at endoanal ultrasonography were 90% and 85% versus 81% and 89% at endoanal MR imaging respectively. Thus both techniques can be used to depict surgically remedial anterior external anal sphincter defects.

Anterior sphincter repair has become the operation of choice in fecal incontinent patients with defects of the anterior external anal sphincter, but not all patients benefit from surgery. Since anorectal function tests can give us only partial understanding of the origin of sphincter repair failures, we investigated the role of endoluminal imaging in identifying factors that are associated with the outcome of sphincter repair. The study is reported in chapter 10. A series of 30 patients were submitted to endoanal MR imaging as well as endoanal ultrasonography before and after surgery. Increased external anal sphincter thickness on nine hours, as measured by endoanal MR imaging, was significantly correlated with a better surgical outcome. Surgical outcome did not differ between patients with and without a persistent external anal sphincter defect or external anal sphincter atrophy depicted on endoanal MR imaging. Patients with a clearly visible overlap and less than 20% fat tissue at the level of anterior sphincter overlap at MR had a significantly better surgical outcome than patients with a fatty anterior external anal sphincter in which overlap could not be visualised. Patients with a persistent external anal sphincter defect depicted on endoanal ultrasonography had a less favorable outcome compared to patients without an external anal sphincter defect.

The most comprehensive scientific evidence can be obtained by means of a systemic review of the literature, in which all of the studies are included, their validity is evaluated, and more precise summary estimates of the treatment effect can be obtained through meta-analysis. A systematic review of the effectiveness of the use of anal plugs in fecal incontinent patients is reported in **chapter 11**. Four studies with a total of 136 participants were included. Two studies compared the use of plugs versus no plugs, one study compared two sizes of plugs, and one study compared two brands of plugs. There was a considerable dropout rate in the included studies, mounting up to 48 patients (35%), affecting the validity and generalizability of the results. Data presented were thus subject to potential bias. The available data suggest that anal plugs are difficult to tolerate, but if tolerated can be helpful in alleviating incontinence.

Implications for clinical practice and research

Our main research question was whether diagnostic tests can be used to identify patients that will benefit from pelvic floor rehabilitation and anterior anal sphincter repair. To answer this question we have studied a cohort of patients with moderate to severe fecal incontinence in order to evaluate the effects of pelvic floor rehabilitation. Unfortunately, pre-selection by clinicians limited the size of the group of patients who subsequently received anterior anal sphincter repair and affected the generalizability of the results obtained for the second part of our objective. Consequently, developing a diagnostic strategy was limited to the treatment with pelvic floor rehabilitation.

The diagnostic strategy for the treatment with pelvic floor rehabilitation in fecal incontinent patients has ultimately resulted in using no diagnostic tests at all, as additional tests played only a limited role in the selection of patients. Since we are not able to predict which patients will benefit from pelvic floor rehabilitation, guidance by means of anorectal function and imaging tests is not necessary before commencing therapy. We therefore recommend that all patients with moderate to severe fecal incontinence should be treated with pelvic floor rehabilitation provided that serious pathology has been excluded. This implies that patients who are seeking help for their incontinence complaints can be referred by their general practitioner for pelvic floor rehabilitation instead of first undergoing a series of tests in (different) hospitals. They should be aware that the overall changes in Vaizey score are moderate and that only a small percentage of patients will be treated successfully. The absence of a need for additional testing in these patients before they start pelvic floor rehabilitation may reduce the large practice variability that we observed in the Netherlands.

If pelvic floor rehabilitation does not lead to sufficient improvement, patients can be referred for further testing to guide advanced treatment. When a surgical approach is considered, one can recommend anorectal manometry, endoanal ultrasonography and / or endoanal MR imaging. Anorectal manometry can offer quantification of resting pressure and squeeze pressure. Both endoanal ultrasonography and endoanal MR imaging can be used to depict surgically remedial anterior external anal sphincter defects. However, endoanal ultrasonography is to be recommended since this technique is widely available and is the least expensive test for defining structural defects of the anal sphincter. Endoanal MR imaging is only available at a limited number of centers. Endoanal MR imaging can be recommended for the depiction of external anal sphincter atrophy when anterior anal sphincter repair is considered.

We were not able to collect sufficient evidence for building a diagnostic strategy for the treatment with anterior anal sphincter repair. Such a strategy would be a welcome addition for other surgical interventions or investigative therapies, such as Sacral Nerve Stimulation (SNS). This implies that the need for evidence for the selection of patients who will benefit from surgery or SNS persists. For the evaluation of such therapies a study design with sufficient diagnostic tests at baseline is helpful in predicting the success of treatment.

It can be expected that in the near future more patients will seek help for fecal incontinence and that the need for adequate testing and effective therapy will increase even more. The mean age of the patients in the studies reported in the present thesis was 59 years and the main cause of fecal incontinence was a complicated vaginal delivery. Accordingly, our current patient group consisted mainly of elderly females, from a generation that may have been reluctant to discuss their complaints, for various reasons including shame. Future generations may be more ready to ask for help, breaching the taboo of fecal incontinence, and starting to increase the need for therapy. This will most likely encourage health care professionals to develop efficient and effective diagnostic and therapeutic strategies in their evidenced-based practice.

The main cause of fecal incontinence was obstetric trauma. This implies that if we were able to identify obstetric risk factors we could reduce the numbers of fecal incontinent patients. So far we can neither predict nor remedy the risk for obstetric trauma. Many women with sphincter injuries do not develop incontinence until later in life suggesting fecal incontinence is a cumulative, multifactorial process. Therefore a topic for etiology studies could be the identification of obstetric risk factors in relation to other risk factors for fecal incontinence.

Based on the results of this thesis and future expectations, we recommend the exclusive use of only evidenced-based tests in the work-up of fecal incontinent patients. To reach that goal, the evidence base for selecting the right tests has to be expanded in future research.

Samenvatting & Implicaties

Samenvatting

Fecale incontinentie wordt gedefinieerd als het ongewenst verlies van gas, dunne of vaste ontlasting door het anale kanaal. Ondanks het feit dat de aandoening niet levensbedreigend is, leidt het toch tot substantiële sociale problemen en wordt fecale incontinentie gekenmerkt door een sfeer van taboe. Naar schatting is de prevalentie in Nederland ongeveer 100.000 personen. De belangrijkste oorzaak voor fecale incontinentie is een gecompliceerde partus (vrouwen) en anorectale chirurgie (mannen).

De diagnostische work-up bestaat, naast anamnese en lichamelijk onderzoek, uit anorectale functietesten en beeldvormende technieken. Initiële therapeutische maatregelen zijn doorgaans conservatief (dieetaanpassingen, medicatie, bekkenbodemtherapie) en bij onvoldoende resultaat invasief (chirurgische interventies, bv. sfincterplastiek). Gezien de substantiële variatie in toegepaste diagnostische testen, was het voornaamste doel van dit proefschrift om een efficiënte diagnostische strategie te ontwikkelen voor patiënten met matig tot ernstige fecale incontinentie, resulterend in een predictiemodel waarmee patiënten met een hoge respectievelijk lage kans op behandelsucces kunnen worden geïdentificeerd. Tevens zijn in dit proefschrift de uitkomsten van diverse subdoelen uiteengezet, beginnend met algemene diagnostiek bij fecale incontinentie. Vervolgens werden verschillende beeldvormde technieken bestudeerd en tot slot zijn diverse behandelingsmogelijkheden geëvalueerd.

Aangezien er op dit moment in Nederland geen consensus is aangaande de diagnostische work-up bij fecale incontinentie, is in **hoofdstuk 2** de variatie in diagnostiek en behandeling beschreven aan de hand van de uitkomsten van een enquête, gehouden in alle nederlandse ziekenhuizen. De meerderheid van de respondenten gebruikte een sigmoïdoscopie routinematig. Meest toegepaste beeldvormende technieken waren endoanale echografie en defecografie. Manometrie was het meest toegepaste anorectale functieonderzoek. De Parks score was de meest gebruikte fecale incontinentiescore gevolgd door de meer recent geïntroduceerde Vaizey score. De meest toegepaste behandelingsvorm was bekkenbodemtherapie, gevolgd door dieetmaatregelen en medicatie en als laatste chirurgie.

In **hoofdstuk 3** is beschreven in hoeverre rectaal toucher en anale inspectie een bijdrage leveren aan de diagnostische work-up bij 312 fecaal incontinente patiënten. Nadat rectaal toucher en anale inspectie waren uitgevoerd, werden de resultaten vergeleken met bevindingen van anorectale manometrie, anale sensitiviteitsmeting en endoanale echografie.

Een afwezige, verlaagde en normale rustdruk en knijpkracht bij rectaal toucher correleerde enigszins met manometrische bevindingen. De bevindingen bij rectaal toucher van externe anale sfincter defecten kleiner dan 90 graden kwamen in 36% overeen met endoanale echografie, in 61% voor defecten tussen 90–150 graden en in 100% voor defecten tussen 150–270 graden. Patiënten met anaal litteken weefsel gediagnosticeerd bij anale inspectie hadden een lagere incrementele knijpkracht en patiënten met een openstaande anus hadden een lagere rustdruk bij anorectale manometrie.

In **hoofdstuk 4** zijn de resultaten weergegeven van een reproduceerbaarheidstudie aangaande het defecogram bij 105 patiënten. Het primaire doel was om de interobserver overeenkomst te bepalen voor het diagnosticeren van een enterocele, anterior rectocele, intussusceptie en anismus.

Het secundaire doel was het evalueren van de invloed van verschillende ervaringsniveaus op de beoordeling.

De beoordelaars werden geclassificeerd in verschillende ervaringsniveaus en hun bevindingen werden vergeleken met de bevindingen van een expert radioloog. De kwaliteit van deze expert werd geëvalueerd met behulp van een intraobserver overeenkomst procedure.

De intraobserver overeenkomst was goed tot zeer goed, behalve voor anismus. De interobserver overeenkomst voor enterocele en rectocele was goed en matig voor intussusceptie. Incomplete evacuatie voor meer dan 30 seconden had een redelijke interobserver overeenkomst en puborectalis impressie had een matige overeenkomst. De overeenkomt in gradering van enterocele en rectocele was goed en matig voor intussusceptie. De overeenkomst, gestratificeerd naar ervaringsniveaus, was zeer goed voor rectocele, inclusief de gradering, en redelijk voor intussusceptie op het meest ervaren niveau. Verschil in ervaringsniveaus speelde geen rol bij enterocele inclusief de gradering.

In **hoofdstuk 5** is een reeks van onderwijskundige en pragmatische aspecten weergegeven, allen gerelateerd aan endoluminale beeldvorming bij fecale incontinentie. Aan de hand van beeldmateriaal is het volgende gedemonstreerd: de huidige consensus in diagnosticeren van afwijkingen van de externe anale sfincter is dat endoanale echografie en endoanale MRI vergelijkbaar zijn. Echter, endoanale MRI heeft het voordeel dat het bij de selectie van patiënten voor sfincterplastiek atrofie van de externe anale sfincter kan aantonen. Atrofie is een negatieve predictor voor de uitkomst van chirurgie. In de postchirurgische diagnostiek is endoanale echografie bruikbaar om residuale sfincter defecten aan te tonen als een mogelijke oorzaak voor het chirurgisch falen. Het gebruiken van externe phased array MRI en driedimensionale echografie voor de evaluatie van fecale incontinentie zijn recent geïntroduceerd en laten gunstige resultaten zien.

De behandelingsopties voor fecale incontinentie zijn in eerste instantie conservatief, zoals dieetmaatregelen en medicatie. Bij onvoldoende resultaat is bekkenbodemtherapie een andere toegepaste conservatieve behandelingsoptie, maar bewijs voor deze behandeling is tot op heden zeer schaars. In **hoofdstuk 6** wordt de uitkomst gerapporteerd van gestandaardiseerde bekkenbodemtherapie bij 281 patiënten. Het behandelingsprogramma omvatte 9 sessies van bekkenbodem-spiertraining met biofeedback en/of elektrische stimulatie. De behandelingsuitkomst werd geëvalueerd aan de hand van de Vaizey score, anorectale manometrie en rectale capaciteit. Na therapie bleek de Vaizey score significant lager ($p< 0.001$), wat verbetering inhield. Echter, maar 13% verbeterde substantieel (meer dan 50% reductie van de Vaizey score). De gemiddelde knijpkracht en het maximaal tolereerbare volume veranderden significant, maar overige anorectale functies niet. Subgroep analyse liet geen substantiële veranderingen in effect zien.

In **hoofdstuk 7** is onderzocht in hoeverre anorectale functies veranderen door behandeling met bekkenbodemtherapie bij 266 patiënten. Tevens werd onderzocht of de mate van verandering was geassocieerd met verandering in Vaizey score. Patiënten ondergingen voor en na bekkenbodemtherapie diverse functietesten, te weten: anorectale manometrie, anale sensitiviteitsmeting en rectale capaciteitsmeting. Na therapie nam de knijpkracht significant toe evenals de urge sensatie en het maximaal tolereerbare volume. De mate van verandering was niet geassocieerd met leeftijd, duur van fecale incontinentie, menopauze of bevindingen op endoanale echografie. Verbetering van de Vaizey score bleek zwak geassocieerd met veranderingen in incrementele knijpkracht en in anale sensitiviteit.

Bekkenbodemtherapie was niet bij alle patiënten succesvol. Indien de Vaizey score na therapie kan worden voorspeld met informatie van diagnostische testen, kunnen patiënten worden geïdentificeerd met een hoge respectievelijk lage kans op behandelsucces. Hiermee zou onnodige behandeling kunnen worden voorkomen. In **hoofdstuk 8** is beschreven hoe bij 250 patiënten elementen van anamnese, lichamelijk onderzoek, anorectale functietesten en beeldvormende testen werden geïdentificeerd die de Vaizey score na therapie kunnen voorspellen. Door middel van lineaire regressie analyse werden kandidaatpredictoren geïdentificeerd en werden verschillende predictiemodellen geconstrueerd. Het eerste predictiemodel omvatte alleen elementen van de anamnese. Vervolgens werden elementen van verschillende testen toegevoegd en werden verschillende modellen ontwikkeld. Het finale model omvatte alle predictoren van anamnese inclusief de predictoren van de diverse testen. Dit complete model resulteerde in een R^2 van 0.23 (p= 0.02). Hieruit volgt dat additionele testen een zeer beperkte rol spelen in het voorspellen van succes van bekkenbodemtherapie.

In **hoofdstuk 9** zijn endoanale echografie en endoanale MRI prospectief vergeleken met betrekking tot het diagnosticeren van externe anale sfincterdefecten in een cohort van 237 patiënten. In een subcohort van 36 patiënten werden operatieve bevindingen gebruikt als referentiestandaard. De overeenkomst tussen endoanale echografie en endoanale MRI was zwak voor het aantonen van sfincterdefecten in het grote cohort. Er was geen significant verschil tussen endoanale echografie en endoanale MRI in het subcohort voor het aantonen van sfincterdefecten. Sensitiviteit en positief predictieve waarde van endoanale echografie waren 90% en 85% versus 81% en 89% met endoanale MRI. Geconcludeerd kan worden dat beide beeldvormende technieken kunnen worden gebruikt voor het aantonen van chirurgisch te repareren externe anale sfincterdefecten.

Sfincterplastiek is de meest gangbare operatie bij patiënten met een anterior defect van de externe anale sfincter, maar niet alle patiënten hebben baat bij deze ingreep. Aangezien anorectale functietesten maar gedeeltelijk inzicht kunnen geven in het falen van de plastiek, is in **hoofdstuk 10** bij 30 patiënten onderzocht welke rol endoluminale beeldvorming heeft bij het identificeren van factoren die zijn geassocieerd met de uitkomst van chirurgie. Alle patiënten ondergingen een endoanale MRI en endoanale echografie voor en na operatie. Een dikkere externe anale sfincter (op 9 uur gemeten) op MRI was significant geassocieerd met een betere chirurgische uitkomst. Er was geen verschil in chirurgische uitkomst tussen patiënten met en zonder een residuaal sfincterdefect en atrofie gediagnosticeerd op endoanale MRI. Patiënten met een duidelijk zichtbare sfincteroverlap en minder dan 20% vet, ter hoogte van de overlap, hadden een significant betere chirurgische uitkomst dan patiënten met een vette sfincter waarbij overlap niet zichtbaar was op MRI. Patiënten met een residuaal sfincter defect op endoanale echografie hadden een significant slechtere uitkomst dan patiënten zonder een residuaal sfincterdefect.

In **hoofdstuk 11** zijn de resultaten weergegeven van een systematische review naar de effectiviteit van het gebruik van anale pluggen bij fecale incontinentie. Vier studies werden geïncludeerd met in totaal 136 patiënten. Twee studies vergeleken het gebruik van een plug versus geen plug, 1 studie vergeleek het gebruik van 2 maten pluggen en 1 studie vergeleek 2 types pluggen. Er was een aanzienlijk uitvalspercentage van de participerende patiënten (48 patiënten (35%) in de geïncludeerde studies wat de validiteit en generaliseerbaarheid van de studieresultaten negatief beïnvloedde. De beschikbare data suggereren dat anale pluggen lastig te verdragen zijn, maar indien ze wel verdragen worden, kan het gebruik van pluggen bijdragen tot het verminderen van fecale incontinentie.

Klinische implicaties

De belangrijkste implicaties zijn gerelateerd aan het primaire doel van dit proefschrift, namelijk het ontwikkelen van een diagnostische strategie gebaseerd op de uitkomsten van bekkenbodemtherapie en sfincterplastiek.

Uit dit proefschrift blijkt dat bekkenbodemtherapie bij veel patiënten met matig tot ernstige chronische fecale incontinentie enigszins verbetering geeft van de fecale incontinentieklachten maar slechts bij een klein aantal patiënten een substantiële verbetering. Aanvullende testen zoals anorectale functietesten en (endoanale) beeldvorming kunnen maar zeer beperkt de uitkomst van bekkenbodemtherapie bij deze patiëntenpopulatie voorspellen. Derhalve kan een huisarts een patiënt direct voor bekkenbodemtherapie doorverwijzen, mits serieuze pathologie is uitgesloten. Patiënten zonder afdoende verbetering van fecale incontinentie door bekkenbodemtherapie kunnen naar een specialist worden verwezen voor verdere diagnostiek en behandeling.

Door het kleine aantal geopereerde patiënten was een nauwkeurige evaluatie van de waarde van testen voor het voorspellen van de uitkomst van sfincterplastiek niet mogelijk. Hierdoor kunnen er geen uitspraken worden gedaan aangaande een diagnostisch pad in relatie tot chirurgie.

List of publications

Dobben AC, Terra MP, Deutekom M, Bossuyt PM, Felt-Bersma RJ, Stoker J. Diagnostic work up of faecal incontinence in daily clinical practice in the Netherlands. Netherlands journal of Medicine 2005; 63(7):265-9.

Dobben AC, Wiersma TD, Janssen LW, de Vos R, Terra MP, Baeten CG, Stoker S. Prospective assessment of interobserver agreement for defecography in fecal incontinence. Am J Roentgenol 2005; 185(5):1166-72.

Dobben AC, Terra MP, Berghmans B, Deutekom M, Boeckxstaens GE, Janssen LW, Bossuyt PM, Stoker J. Functional changes after physiotherapy in fecal incontinence. Int J Colorectal Dis. 2006 Sep;21(6):515-21.

Dobben AC, Terra MP, Slors JFM, Deutekom M, Gerhards MF, Beets-Tan RGH, Bossuyt PMM, Stoker J. Comparison of endoanal magnetic resonance imaging versus endoanal ultrasonography in the depiction of external anal sphincter defects in fecal incontinent patients. Radiology; accepted.

Dobben AC, Terra MP, Deutekom M, Gerhards M, Bijnen AB, Felt-Bersma RJ, Janssen, LW, Bossuyt PM, Stoker J. Anal inspection and digital rectal examination in fecal incontinent patients. Int J Colorectal Dis; accepted.

Dobben AC, Terra MP, Deutekom M, Slors JFM, Janssen LWM, Bossuyt PMM, Stoker J. The role of endoluminal imaging in clinical outcome of overlapping anterior anal sphincter repair in fecal incontinent patients. Submitted.

Dobben AC, Felt-Bersma RJF, Ten Kate JW, Stoker J. Endoluminal imaging in fecal incontinence. Submitted.

Deutekom M, **Dobben AC**, Dijkgraaf MG, Terra MP, Stoker J, Bossuyt PM. Costs of outpatients with fecal incontinence. Scandinavian Journal of Gastroenterology 2005; 40(5):552-8.

Deutekom M, Terra MP, **Dobben AC**, Dijkgraaf M, Baeten CG, Stoker J, Bossuyt PM. Impact of faecal incontinence severity on health domains. Colorectal Disease 2005; 7(3):263-9

Deutekom M, Terra MP, **Dobben AC**, Dijkgraaf MG, Felt-Bersma RJ, Stoker J, Bossuyt PM. Selecting an outcome measure for evaluating treatment in faecal incontinence. Dis Colon Rectum 2005; 48(12):2294-301.

Deutekom M, Terra MP, Dijkgraaf MG, **Dobben AC**, Stoker J, Boeckxstaens GE, Bossuyt PM. Patients' perception of tests in the assessment of faecal incontinence. Br J Radiol. 2006 Feb;79(938):94-100.

Deutekom M, **Dobben AC**, Terra M, Engel A, Stoker J, Bossuyt P, Boeckxstaens G Clinical presentation of faecal incontinence and anorectal function, what is the relationship? Am J Gastroenterology; accepted.

Deutekom M, **Dobben A**. Plugs for containing faecal incontinence. Cochrane Database of Systematic Reviews 2005 (3): CD005086.

Terra MP, Beets-Tan RG, van Der Hulst VP, Dijkgraaf MG, Bossuyt PM, **Dobben AC**, Baeten CG, Stoker J. Anal sphincter defects in patients with fecal incontinence: endoanal versus external phased-array MR imaging. Radiology, 2005; 236(3):886-95.

Terra MP, Beets-Tan RG, Van der Hulst VP, Deutekom M, Dijkgraaf MG, Bossuyt PM, **Dobben AC**, Baeten CG, Stoker J. MR imaging in evaluating atrophy of the external anal sphincter in patients with fecal incontinence. Am J Roentgenol. 2006 Oct;187(4):991-9.

Terra MP, **Dobben AC**, Berghmans B, Deutekom M, Baeten CG, Janssen LW, Boeckxstaens GE, Engel AF, Felt-Bersma RJ, Slors JF, Gerhards MF, Bijnen AB, Everhardt E, Schouten WR, Bossuyt PM, Stoker J. Electrical stimulation and pelvic floor muscle training with biofeedback in patients with faecal incontinence: a cohort study of 281 patients. Dis Colon Rectum. 2006 Aug;49(8):1149-59.

Terra MP, Deutekom M, Beets-Tan RG, Engel AF, Janssen LW, Boeckxstaens GE, **Dobben AC**, Baeten CG, De Priester JA, Bossuyt PM, Stoker J. Relation between external anal sphincter atrophy at endoanal MRI and clinical, functional, and anatomical characteristics in patients with fecal incontinence. Dis Colon Rectum. 2006 May;49(5):668-78.

Terra MP, Deutekom M, **Dobben AC**, Baeten CG, Janssen LW, Boeckxstaens GE, Engel AF, Felt-Bersma RJ, Slors JF, Gerhards MF, Bijnen AB, Everhardt E, Schouten WR, Bossuyt PM, Stoker J. Can the outcome of pelvic floor rehabilitation in patients with faecal incontinence be predicted? Submitted.

Terra MP, Beets-Tan RGH, Vervoorn I; Deutekom M, Wasser MNJM, Witkamp TD, **Dobben AC,** Baeten CGMI, Bossuyt PMM, Stoker J. Pelvic floor muscles lesions at endoanal MR imaging in patients with fecal incontinence. Submitted.

Dankwoord

Heerlijk! Het boekje is, ondanks 3 verhuizingen, emigratie naar Brabant, huwelijk, zwangerschap en bevalling, afgerond. Ik ben erg dankbaar voor de mensen die mij hierbij direct of indirect hebben geholpen of gesteund.

Prof. dr. J. Stoker, beste Jaap. Ik heb het als zeer prettig ervaren om onder jouw begeleiding deze eindstreep te halen. Je bent in staat om, ondanks je drukke schema, steeds weer tijd te maken voor het lezen en corrigeren van stukken, bedenken van oplossingen en bediscussiëren van nieuwe ideeën en hypotheses. Dat was voor mij een erg productieve werkwijze.

Daarnaast heb jij je als mens en professional op heel effectieve wijze ingezet tijdens ziekte in mijn familie, wat ik zeer heb gewaardeerd.

Prof. dr. P.M.M. Bossuyt, beste Patrick. Jouw grondige methodologische kennis en inzichten hebben mij geholpen om mijn publicaties steeds weer te verbeteren en naar een hoger niveau te tillen. Door je vermogen om dit onderzoek in breder perspectief te bekijken, heb je me geleerd om vanuit diverse invalshoeken onderzoeksvragen te beantwoorden en de noodzakelijke nuanceringen aan te brengen. Mede door het vertrouwen dat jij gaf in een succesvolle afronding, heb jij me gestimuleerd om met enthousiasme aan mijn boekje te werken en was je voor mij een plezierige begeleider.

Prof. dr. J.S.Laméris. Ik wil u bedanken voor het feit dat ik in de gelegenheid ben gesteld om als onderzoeker te gaan werken op de afdeling radiologie. De mogelijkheden die ik op uw afdeling heb gekregen om mijzelf te ontwikkelen, lagen ver boven de verwachtingen die ik had toen ik in 1999 in dienst trad. Ik waardeer het dat u aan mijn promotiecommissie wil deel nemen.

De overige leden van de promotiecommissie prof. dr. P. Fockens, prof. dr. W.P.Th.M. Mali, prof. dr. C.J. Mulder, dr. W.A. Bemelman en dr. W.R. Schouten wil ik bedanken voor de tijd die u heeft genomen om dit manuscript te beoordelen.

Prof. dr. J.A. Reekers, Prof. dr. C. van Kuijk, beste Jim, beste Kees. Jim, jij hebt me zonder veel omhaal van woorden het AMC binnengeloodst en me voorgesteld aan Kees. Kees, het door jou geschetste functieprofiel leek mij dusdanig aantrekkelijk dat ik eigenlijk direct heb ingestemd met een baan op de afdeling radiologie. Van deze keuze heb ik geen moment spijt gehad.

De FIT- promovendi dr. Maaike Terra en dr. Marije Baart de la Faille-Deutekom.

Lieve Maaike, ik ben trots op jou. Je hebt de afgelopen jaren van intensieve samenwerking laten zien dat je een enorme doorzetter bent en volhoudt, zelfs in soms heel lastige omstandigheden. In maart mocht ik tijdens jouw promotie één van jouw paranimfen zijn en nu is het vice versa. Je hebt het onderzoekstraject glansrijk doorlopen en bent inmiddels begonnen als assistent radiologie. Dit betekent dat wij helaas niet meer samenwerken. Ik mis ons lachen, kleppen, discussiëren en (te) luide kamerbrede overleg, maar gelukkig kunnen we hier buiten werk gewoon mee doorgaan.

Lieve Marije, jij de eerste FIT-promovenda, ik de laatste. Het was fijn om zo'n KEB-er als jij in het FIT-team te hebben om de statistische ingewikkeldheden op begrijpbare wijze gepresenteerd te krijgen. Na je huwelijk, promotie en reis naar Zuid-Amerika, ben je weer teruggekomen op de

KEB om me bij te staan bij de laatste FIT-loodjes. Nu rest ons nog slechts een drankje te drinken in en op jouw nieuwe Amsterdamse stulpje.

Sabine Kuhlman. Sabine, dank je wel voor de administratieve en logistieke ondersteuning die je, al dan niet voor de FIT-studie, hebt gegeven. Ook Marion de Jong-Putto, Inge Vervoorn en Jascha Mooij wil ik bedanken voor de inzet die jullie hebben getoond bij de omvangijke klus van dataverzameling en -invoer.

Tevens wil ik de vele participanten uit de diverse ziekenhuizen bedanken voor hun deelname aan de FIT-studie.

In het bijzonder wil ik prof. dr. C.G.M.I. Baeten, prof. dr. G.E.E. Boeckxstaens en dr. L.W.M. Janssen bedanken voor de forse aantallen geïncludeerde patiënten.

Dr. R.G.H. Beets-Tan, dr. R.J.F. Felt-Bersma, dr. J.F.M. Slors en dr. Tj. G. Wiersma. Beste Regina, Richelle, Frederik en Tjeerd, ik ben jullie zeer erkentelijk voor de geleverde bijdragen aan mijn publicaties.

Dr. B. Berghmans. Beste Bary, samen met de deelnemende bekkentherapeuten, heb je een belangrijke prestatie geleverd voor de FIT-studie. Het fysiotherapeutische deel van de studie is onder jouw leiding succesvol afgerond. Ik wil jou, en alle bekkentherapeuten die zich hiervoor hebben ingezet, zeer bedanken.

Tevens wil ik prof.dr. A.B.Bijnen, dr. J.W. De Bruijne, dr. P.P.L.O. Coene, dr. J.B.V.M. Delemarre (†), dr. M.G.W. Dijkgraaf, dr. A.F. Engel, dr. E. Everhardt, dr. M.F. Gerhards, dr. J.J.G.M. Gerritsen, dr. E. van der Harst, dr. A.B. Huisman, dr. V.P.M. van der Hulst, dr. J.A. de Priester, dr. A. Sikkenk, dr. E.J. Spillenaar Bilgen, dr. W.F. van Tets, dr. C.H. van der Vaart, dr. M.N.J.M. Wasser en dr. T.D. Witkamp bedanken voor de patiënteninclusie, dataverzameling, input voor publicaties en overige medewerking aan dit omvangrijke onderzoek.

In de diverse ziekenhuizen hebben veel medewerkers zich op verschillende wijze ingezet waardoor patiëntenzorg samen met uitvoering van onderzoek kon worden gerealiseerd. Dank daarvoor, in het bijzonder Elly, Jack, Raschel, Sandra, Shanta, Bram, Cynthia, Hellen, Olle, Ramona, Tamira, Jac Oors, Hilda, Jacueline, Anneke, Rachel, José, Huub, Jarno en Sacha.

En natuurlijk de onderzoekers van de radiologie, al of niet vertrokken van G1 naar C1. Als één van de eerste bewoners van de onderzoeksgang G1 (samen met (inmiddels dr.) Ro) was ik getuige van een ware onderzoekers-explosie en ontpopte G1 zich tot de gezelligste onderzoeksgang van het AMC, met niemand minder dan: Maartje (succes op 2 maart!), Nicole, Wouter, Shandra, Jasper, Karin, Adrienne, Ayso, Sebastiaan, Anneke, Marjolein en Jochem. Allemaal nog heel veel succes op het onderzoekers-pad!

En er bestaat natuurlijk geen onderzoek zonder onze IT. Jan, JP, Martin en Onno, heel veel dank voor al jullie computer-geduld!

Wil, Franca, David, Inge, Jelle, Anke, Jos, Anneke, Hans, Jo, Teun, Ulricke, Machiel, Lot, Bien, Li en Corientje. Lieve vriendjes en vriendinnetjes, ik wil jullie bedanken voor trouwe vriendschap die hele drukke en hectische tijden overwint.

Lieve Kees, Elisabeth, Melchior en Annemiek, dank voor jullie medeleven en belangstelling.

Lieve Jan, Paul, Leeyanne, Rob en Mathieu, dank voor jullie vertrouwen en steun. Familie is er altijd; door dik en dun.

Lieve papa en mama, jullie zijn de grootste supporters van mijn leven.

Frederik en Elisa, het is heerlijk om met jullie te leven. Lieve Frederik, jij bent voor mij een onbetaalbare verrijking. Lieve La, jij bent de allerliefste baby van de hele wereld.

Curriculum Vitae

Annette de Bruijne- Dobben werd geboren in Groningen op 26 oktober 1966. In 1984 behaalde zij haar h.a.vo. diploma aan het Farel College te Amersfoort. Vier jaar later in 1988 sloot zij de h.b.o.-v. af in Zwolle met de afstudeerdifferentiatie 'Werklastmeting in een algemeen ziekenhuis', waarmee de interesse voor de uitvoering van onderzoek was geboren. Eerst echter is zij gaan reizen en heeft gewerkt en gewoond in Soroka Medical Center, Beersheva, Israël. Teruggekeerd in Nederland heeft zij in het VU-Medisch Centrum gewerkt als verpleegkundige op chirurgie en pulmonologie / oncologie. Vanaf 1994 – 1996 heeft zij gewerkt als intensive care verpleegkundige, eveneens in het VU-MC. Vervolgens waren onderzoek en onderwijs aspecten die steeds meer haar interesse kregen. Tijdens haar functie als site manager van Site Management Organisation 'Good Clinical Practice' in Amsterdam, volgde zij de docentencursus, werkte mee aan de ontwikkeling van de post-h.b.o. opleiding voor researchverpleegkundigen in Rotterdam en gaf les aan diezelfde opleiding. Tijdens één van de detacheringsprojecten vanuit 'Good Clinical Practice' werd zij benaderd door de afdeling radiologie van het Academisch Medisch Centrum in Amsterdam om te gaan werken als researchcoördinator, het opzetten van de post-h.b.o. opleiding voor radiologisch laboranten en later, na het succesvol afronden van de Master of Science studie 'Evidenced Based Practice' aan de Universiteit van Amsterdam, als promovenda van de studie 'An efficient diagnostic strategy in fecal incontinence'.
Annette is getrouwd met Frederik en moeder van Elisa.