PELVIC PHYSIOTHERAPY IN FAECAL INCONTINENCE

Pelvic physiotherapy in faecal incontinence
Esther M.J. Bols

ISBN: 978 94 6159 050 3
Cover design: Mischa Visser
Lay-out: Yvonne Leenders
Printed by: Datawyse - Universitaire Pers Maastricht

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Pelvic physiotherapy in faecal incontinence

PROEFSCHRIFT

Ter verkrijging van de graad van doctor
aan de Universiteit Maastricht,
op gezag van de Rector Magnificus,
Prof. mr. G.P.M.F. Mols,
volgens het besluit van het College van Decanen,
in het openbaar te verdedigen
op woensdag 15 juni 2011 om 14:00 uur

door

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The studies presented in this thesis were conducted at the Department of Epidemiology, CAPHRI, the School for Public Health and Primary Care, Maastricht University. CAPHRI is part of the Netherlands School of Primary Care Research (CaRe), which has been acknowledged since 1995 by the Royal Netherlands Academy of Arts and Sciences (KNAW).

Financial support by the Dutch Association for Physical Therapy for Pelvic Floor Disorders and Pre- and Postnatal Healthcare (NVFB) and the Royal Dutch Society for Physical Therapy (KNGF) for the publication of this thesis is gratefully acknowledged.

Financial support for the printing of this thesis was kindly provided by the Department of Epidemiology (Maastricht University).

'Life is like a box of chocolates...you never know what you're gonna get!'

Aan mijn lieve ouders,

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CHAPTER 1

GENERAL INTRODUCTION AND OUTLINE OF THE THESIS

GENERAL INTRODUCTION

'Je Zal Het Maar Hebben' ('What if it happened to you?') is a Dutch television programme which vividly discusses several disorders that are extremely embarrassing, may cause withdrawal from social activities and reduce quality of life. When the subject of 'faecal incontinence' was discussed, the presenter was standing in a sewerage system (BNN, episode 'Shit happens', July 21, 2003).

I have frequently found that many people do not know what faecal incontinence is, or are unaware how many people are affected by it. Faecal incontinence is the complaint of involuntary loss of faeces (including liquid and solid stools).¹ Anal incontinence is mostly used to include faeces or flatus. Terms like 'the unvoiced symptom' or 'the silent affliction' typically describe and illustrate the enormous embarrassment felt by patients affected by it.² As was illustrated by the location where the TV presenter was standing, faecal incontinence is associated with negative concepts like dirt, disgust and smelliness. Figures on help-seeking behaviour confirm the idea of suffering in silence, since they show that only one third of patients have the courage to consult a physician.³ The reasons for not seeking help are unclear but may relate to social class, mild symptoms, lack of bother or embarrassment, or a perceived lack of effective treatment. Cross-sectional studies have shown that about 0.4–18% of people suffer from faecal incontinence (incontinence for liquid and solid stools), although these rates are likely to be underestimated.^{4,5} Coping techniques used by patients include the use of incontinence absorption materials, never straying far from a toilet, avoiding social contacts, only leaving home after defecation, using perfume to disguise bad smells and making noises at moments they expect to lose gas or stool. From this perspective, there is enough reason to characterise faecal incontinence as an example of the kind of embarrassing disorders featured in the 'What if it happened to you' programme.

The ageing of the population will make faecal incontinence an increasingly important socioeconomic problem in the coming decades, especially since it leads to reduced quality of life and premature institutionalisation, which has important cost implications.⁶ It is not, as is often incorrectly thought, only elderly and/or institutionalised people who are affected. Younger people are affected as well, especially postpartum women with obstetric risk. However, conflicting results about the contributions of maternal, obstetric and foetal characteristics to postpartum faecal incontinence hamper the prevention and management of this problem.

There are numerous methods to treat patients with faecal incontinence, which can be classified as non-surgical or surgical. Mild incontinence is mostly treated successfully with conservative methods like constipating medication, dietary changes, lifestyle advice or toilet behaviour. If these fail, or in the case of more severe faecal incontinence, the next option is usually pelvic physiotherapy, and failure of pelvic physiotherapy is then often a criterion for proceeding to surgical management.⁷ Depending on the un-

derlying cause and severity, surgical treatment may offer equal or sometimes greater benefit, but also involves a greater risk of complications.

Pelvic physiotherapy was first introduced in the 1970s and is regarded as safe, inexpensive and well-tolerated.^{8,9} The first publications reported on the use of biofeedback (element of pelvic physiotherapy), and these were followed by the first randomised clinical trial, published in 1984.¹⁰ Since then, about 16 randomised clinical trials have studied the effects of pelvic physiotherapy for faecal incontinence, the majority having been done after 2000.¹¹⁻²⁶ The differences between these trials in terms of study methods, patient groups, duration of follow-up, and methodological weaknesses hamper the interpretation of the findings.²⁷ In particular, no decisive conclusions have been drawn about the effectiveness of different methods of biofeedback, the use of biofeedback compared with exercises alone, or biofeedback combined with other interventions.^{7,28,29} The lack of high-quality research in the field of pelvic physiotherapy may have several consequences. First, insurance companies or other third-party health care funders do not reimburse physiotherapy treatment for patients with faecal incontinence, because it remains unproven whether it is effective and cost-effective, and this may limit access to valuable treatment. Second, referring physicians need to keep abreast of the options offered by physiotherapeutic diagnostics and intervention. Lack of recommendations indicating which elements of pelvic physiotherapy treatment are effective hampers early clinical decision-making, efficient use of resources, efficient referral and adequate cooperation between physicians and physiotherapists. In view of the lack of knowledge about effective modalities of physiotherapy treatment in faecal incontinence, we undertook a randomised controlled trial which included adult patients who had had moderate to severe faecal incontinence complaints for at least six months.

The design of studies evaluating effectiveness of treatment of faecal incontinence is problematic as no well-validated outcome measures are available. Frequently used outcome measures to evaluate the severity of faecal incontinence and the quality of life of the patients, such as the Vaizey score, Wexner score, and Fecal Incontinence Quality of Life Scale, have not been thoroughly evaluated in terms of several psychometric properties. In particular, there is a lack of information on the clinically meaningful interpretation of changes in the outcomes. This field needs further exploration in order to optimise sample size calculations, evaluation and interpretation of outcomes. Finally, patient selection on the basis of predictive factors will help to improve the management of patients with faecal incontinence. So far, it is unclear which patients are likely to benefit from physiotherapy treatment, which is relevant for medical decision making and patient counselling. Therefore, we aimed to identify patient characteristics predicting a favourable outcome of pelvic physiotherapy treatment.

AIM AND OUTLINE OF THE THESIS

The aim of this thesis is to present new evidence concerning different aspects of the management of patients with faecal incontinence, in order to enable physicians and physiotherapists to provide competent individual counselling and treatment to all patients who suffer from it. Although interest in the subject of faecal incontinence has increased in recent years, there is still a long way to go to optimise the diagnostic and treatment process for these patients. The aim of the work summarised in this thesis was (1) to examine the state of the art for various aspects of the management of patients with faecal incontinence, (2) to evaluate which delivery-related factors are important in the development of faecal incontinence in a frequently affected young patient group, i.e. postpartum women, (3) to assess psychometric properties and minimally important change for the Vaizey score, Wexner score and Fecal Incontinence Quality of Life Scale for clinical use in patients with faecal incontinence, (4) to assess the effect of adding rectal balloon training to pelvic floor muscle training in patients with faecal incontinence, and (5) to identify patient characteristics predicting a favourable outcome of physiotherapy treatment.

Chapter 2 presents a survey of the state of the art. Its aim is to give an overview of aetiological factors for faecal incontinence, the diagnostic work-up, reported outcome measures and the conservative and surgical treatment options for faecally incontinent patients, including their effectiveness.

The literature review shows that vaginal delivery has been identified as one of the major causes of faecal incontinence in young women. Delivery may be followed by sphincter defects and/or pudendal neuropathy due to excessive straining. Many women present with faecal incontinence symptoms immediately after delivery or later in life, presumably because the cumulative effects of multiple deliveries, progressive neuropathy, ageing and the menopause are too much for the compensatory mechanisms. The literature overview yielded no definitive conclusions as to which maternal, obstetric or foetal characteristics contributed most to postpartum faecal incontinence.

Chapter 3 therefore presents the results of a systematic review to assess the aetiological factors for postpartum faecal incontinence.

The literature review also showed that several multi-item scoring systems have emerged, the Vaizey score being a frequently reported outcome. So far, inconclusive results have been presented on the psychometric properties for evaluative purposes following physiotherapy treatment. Accordingly, **chapter 4** explores the psychometric properties of the Vaizey score, giving special attention to the minimally important change.

The next two chapters describe a randomised controlled trial assessing the effectiveness of rectal balloon training combined with pelvic floor muscle training, compared to pelvic floor muscle training alone in patients with faecal incontinence. **Chapter 5** describes the design of the randomised controlled trial, while **chapter 6** presents its

results in terms of short-term effectiveness of rectal balloon training as an add-on therapy to pelvic floor muscle training.

The remaining chapters report on secondary analyses of data from the randomised controlled trial. **Chapter 7** addresses the baseline characteristics which are important in predicting a favourable or unfavourable outcome of physiotherapy treatment in patients with faecal incontinence, while **chapter 8** critically evaluates the Vaizey score, Wexner score and Fecal Incontinence Quality of Life Scale for clinical use in these patients. The results of the psychometric evaluation as well as estimates of the minimally important change are presented.

Finally, **chapter 9** summarises the main findings reported in this thesis, and discusses methodological aspects of the different studies, as well as implications for clinical practice and for future research.

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CHAPTER 2

PHYSIOTHERAPY AND SURGERY IN FAECAL INCONTINENCE: AN OVERVIEW

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Published in: Physical Therapy Reviews 2008;13(2):1-20

ABSTRACT

Faecal incontinence (FI) is a major healthcare problem that is particularly embarrassing and affects about 2 to 24% of the adult population. Apart from being a social burden to the patient, FI is also a major economic burden. Several diagnostic measures are available to assess the nature and severity of FI, although guidance for conservative or surgical therapy is not always obvious. Conservative treatment consists of dietary and pharmacological treatment, bowel management, use of products and appliances and physiotherapeutic interventions. Physiotherapy is often tried before surgical treatment because it is safe and inexpensive; it is also possible to employ other treatments after its use. Promising results of physiotherapy in FI have been published, although its effectiveness should be further elucidated. Surgical treatment aims at correcting the anal sphincters and pelvic floor, using encirclement procedures and sacral nerve stimulation. When the outcomes of both conservative and surgical treatment are unsatisfactory, colostomy is the only remaining solution.

INTRODUCTION

Faecal incontinence (FI) is a major health care problem that is particularly embarrassing and affects about 2 to 24% of the adult population.¹ FI can be defined as the recurrent involuntary excretion of faeces in inappropriate places or at inappropriate times.² FI covers a wide spectrum of symptoms, from involuntary but recognised passage of gas, liquid, or solid stool (urge incontinence) to unrecognised anal leakage of mucus, fluid, or stool (passive incontinence).³ FI is mostly associated with older people, as a natural aspect of ageing. However, younger people are often affected as well, resulting in difficulties participating in school, work or social life. It is not hard to imagine what it is like to experience loss of faeces in the middle of a shop, workplace, bus or school; often, it is a problem that the person affected finds difficult to explain to others, such as family or a partner. The implications of FI to the affected individual are huge: staying at home nearby a toilet, having to avoid social contact including relationships or sexual contact, having feelings of depression and low self-confidence.⁴ A lot of these implications are due to the unpredictable character of FI and the fear of odour.

Apart from being a social burden to the patient, FI is also a major economic burden. Direct medical costs are those incurred by obtaining medical care for prevention, diagnostics, therapeutics, rehabilitation and care. Direct non-medical costs are those associated with travel/time for the purpose of obtaining health care and extra costs, like absorbent material or skincare products. Indirect non-medical costs are costs associated with production losses associated with paid and unpaid work.⁵ Limited data are available on healthcare costs for FI.^{3,6} Recently, total costs in the Netherlands were estimated at €2169 per faecally incontinent patient a year.⁵ Production losses of paid and unpaid work accounted for more than half of the total costs, costs of health care visits for almost a fifth and costs associated with protective material for only one tenth, while incontinence medication formed only 5% of the total costs.⁵

Estimates of FI prevalence and associated healthcare costs are limited by the different definitions of FI. Estimates of its prevalence vary greatly and depend on the clinical setting and on how FI is defined based on severity and frequency.⁷⁻⁹ The reported prevalence ranges from 2 to 24% in people living in the community to 47% in the institutionalised elderly.^{1,2,4,8,10-12} In the Netherlands, the estimated prevalence of the disorder is approximately 80,000 patients, which represents 0.5% of the total population.¹³ This estimate is based on selling figures for diapers and probably represents an underestimation of the total problem. In the elderly (≥ 60 years) living at home, the prevalence of FI in the Netherlands is estimated to be 6%; in the same population the prevalence of double incontinence is estimated to be 3%.¹⁴

The proportion of the population aged 65 years and older is increasing; this will result in a higher prevalence of FI in the next 15 years. In 2005, 13.8% of the inhabitants living in the Netherlands were aged 65 years and over (2.3 million); this percentage is expected to be 15.1% in 2010, 19.0% in 2020 and 22.3% in 2030.¹⁵

The actual prevalence of FI is likely to be higher due to frequent underreporting. Underreporting of FI is common because patients are often embarrassed about visiting physicians, or unaware of the possible treatment options. It is estimated that, for all ages combined, the prevalence of clinically significant faecal or double incontinence in the population as a whole is twice that identified by surveying the population known to the various health and social services.²

Nowadays, a wide range of treatment and diagnostic options is available, patient outcome has improved, and increasing awareness is generated from media and in the community. More patients are becoming aware that FI is a common, treatable disorder, more patients consult their doctor, and an increasing number of patients are referred for diagnosis and treatment.¹⁶

This literature review aims to give an overview of the development of FI, the diagnostic measures and the conservative and surgical treatment options for faecally incontinent patients, including their effectiveness. The review elaborates on certain topics:

(1) a strength and weakness analysis of several FI severity scores, including their psychometric properties, (2) an assessment of the psychometric properties of several condition-specific and generic quality of life instruments in FI, (3) an assessment of the methodological quality of published (randomised) controlled trials of physiotherapy in FI, using the Delphi checklist, and (4) the introduction of a new FI severity score, the PREFAB-score, based on the well-known PRAFAB-score in the urinary incontinence field.

The search methods started with a literature search in the Cochrane Library for relevant systematic reviews within the scope of this review, using the MeSH term faecal incontinence. Five relevant systematic reviews were retrieved. Furthermore, PubMed was searched for relevant reviews and trials, using faecal incontinence as MeSH term and f(a)ecal incontinence as free text words. Time constraints were applied to the database search; only articles since 1st January 1980 were included. Finally, text books on FI were consulted and the reference list of relevant articles were checked for useful articles.

For the assessment of the methodological quality of published (randomised) controlled trials of physiotherapy in FI, the following electronic databases were searched for trials: The Cochrane Central Register of Controlled Trials, Medline, EMBASE, CINAHL and the Physiotherapy Evidence Database (PEDro). The search terms f(a)ecal incontinence were used solitary or in combination with the search terms representing the interventions: physiotherapy, physical therapy techniques, rectal balloon (training), electrical stimulation, electric stimulation (therapy), pelvic floor (muscle exercises), (anal) sphincter exercises, exercise (therapy), Kegel exercises, levator ani and biofeedback. This search yielded 14 trials.

Anatomy and pathophysiology

Continence depends on several anatomic and physiologic entities: anal sphincter function, pelvic floor function, rectal distensibility, anorectal sensation, anorectal reflexes,

intact nervous system, mental function, stool volume, stool consistency, and colonic transit. Deficiency of one or more of these factors can lead to incontinence.¹⁷

The anal sphincter muscles are located in the distal part of the anal canal, which arises from the sigmoid colon and rectum. The anal sphincter mechanism involves the internal anal sphincter, external anal sphincter and puborectalis muscle. The internal anal sphincter is a circular smooth muscle layer under involuntary control and is mainly contracted in rest. This sphincter represents 80% of the basal resting pressure.^{17,18}

Internal anal sphincter dysfunction is often associated with faecal seepage (passive FI). Besides during childbirth, the sphincter can be damaged by anorectal surgery (sphincterotomy or fistulotomy), anal stretch¹⁹ and primary degeneration.²⁰

The external anal sphincter is a striated muscle, innervated by the pudendal nerve (S2–S4) and comprises of three parts: a subcutaneous, a superficial and a deep part. In rest, the external sphincter is sub-maximally contracted and only modestly contributes to basal pressure. Basal pressure normally redoubles in case of voluntary sphincter contraction.²¹ Besides this contraction, a sudden rise in intra-abdominal pressure initiates a spinal reflex which causes the external sphincter to contract. In addition to both sphincters, the haemorrhoidal plexus also contributes to basal pressure (15%).

The puborectalis muscle is part of the levator ani muscle, joined with the pubococcygeus and ileococcygeus muscle. The puborectalis muscle is anatomically and functionally closely related to the external anal sphincter. The puborectalis muscle forms a muscular sling around the anorectal junction and creates an angulation between the anal canal and rectum due to its attachment anterior to the pubic bone. At rest, the anorectal angle is 90°, increasing during straining and defecation to about 135°, which facilitates the passage of stool.¹⁸ The puborectalis muscle sling and anorectal angle seem to contribute to the maintenance of continence, although the extent of their contribution is uncertain.¹⁷

In response to rectal distension the external sphincter contracts, coinciding with reflex inhibition of the internal anal sphincter.²² Further accommodation of the rectum occurs when defecation is not appropriate and the internal sphincter regains its tone. Consequently, the possibility to postpone defecation is dependent on rectum distensibility, reservoir function and efficiency of the anal sphincter mechanism. Adequate rectal sensation is necessary to notice rectal contents. Presence of a small amount results in some relaxation of the internal anal sphincter and the anal mucosa is able to discriminate gas, solid or liquid faeces.²³ Rectal sensation is often impaired in patients with diabetes, spinal disease or constipation.¹⁷

Voluntary relaxation of the external anal sphincter and puborectal sling causes opening of the anal canal. Straining further decreases anal pressure due to relaxation of the internal and external sphincter, and defecation is possible.

Aetiological factors

Many aetiological factors can be associated with FI.^{24,25} The most important causes of FI are summarised in Table 2.1. The different aetiologies are based on motoric, sensory or reservoir problems; often, a multifactorial aetiology is present.^{26,27}

Table 2.1 The aetiology of faecal incontinence

Trauma	Congenital abnormalities	Neurological	Colorectal causes
Obstetric trauma Direct trauma Surgery	Imperforate anus Hirschsprung's disease Spina bifida	<i>Cerebral:</i> Stroke Parkinson's disease Dementia Tumour Psychological causes Brain injury <i>Spinal:</i> Spinal cord injury Paraplegia Cauda equina syndrome Multiple Sclerosis <i>Peripheral:</i> Pudendopathy Diabetes mellitus	Atrophy due to ageing Inflammatory bowel disease Impaction Procidentia Diarrhoea Fistulae Rectal prolapse/rectocele /enterocele

Obstetric trauma

Obstetric trauma is one of the major causes of FI in women. After delivery, a sphincter defect and/or pudendal neuropathy following excessive straining can occur. Endo-sonography has revealed that 35% of women after delivery had a defect of the external anal sphincter, the internal anal sphincter, or both.²⁸ Risk factors for a ruptured sphincter are vaginal birth, midline episiotomy, forceps delivery, vacuum-assisted delivery, induced labour, high birth weight, and epidural anaesthesia.^{24,29} The reported frequency of incontinence of stool in primiparous women ranges from 2 to 6%, and incontinence of either stool or flatus from 13 to 25%. After several perineal lacerations the rate of anal incontinence increases towards 17 to 62%.²⁹ Many women present symptoms later in life, presumably because cumulative effects of multiple deliveries, progressive neuropathy, ageing and the menopause overcome compensatory mechanisms.^{29,30}

In an evaluation concerning the effect of ageing on anal canal pressure, McHugh et al. found that ageing in women was associated with a consistent reduction in resting anal canal pressures and maximum squeeze pressure.³¹ The resting anal pressure in women was reduced by 25% in the sixth decade and by 49% in the eighth decade compared with the third decade. Maximum squeeze pressure was reduced by only 6% by the sixth decade and by 37% in the eighth decade compared with women in the third decade.

Other aetiological factors

Several colorectal, urological or gynaecological interventions can cause FI as well. The surgical procedures most related to FI are sphincterotomy for anal fissure, sphincter dilatation, haemorrhoidectomy, fistulotomy, and ileal pouch reconstruction.¹⁷

Children who are born with congenital abnormalities, such as imperforate anus or Hirschprung's disease (encopresis), often experience lifelong problems with incomplete evacuation of faeces and soiling despite anatomical correction.¹⁰

Specific neurological diseases associated with FI include diabetes, multiple sclerosis, Parkinson's disease, stroke, and spinal cord injury. Denervation or neuropathy is frequently present in patients with diabetes, vaginal delivery, descending perineum syndrome, chronic straining at stool and rectal prolapse.⁷

FI disproportionately affects individuals with physical and mental disabilities, especially in nursing homes. Chassagne et al. identified five characteristics associated with the development of FI in the institutionalised elderly: a history of urinary incontinence, poor mobility, poor cognitive function, age older than 70 years and neurological disease.^{32,33} In these homes, the association between urinary incontinence and FI is also well known. This so-called 'double incontinence' can be explained by the same underlying causes of poor mobility and cognitive impairment.³²

Demented residents are at high risk of developing faecal impaction because of neglect of the call to stool, impaired awareness of rectal fullness and coexistent limited mobility.³⁴ Moreover, an association was found between poor health and patients with long-lasting FI.³²

FI is often related to irritable bowel syndrome and constipation, which is thought to be more prevalent amongst women.⁶ The difference in prevalence of FI between men and women narrows with age. At the age of 70 years, men experience FI as often as do women.³⁵

Symptoms associated with FI are sometimes related to other causes, known as pseudo incontinence: perianal leakage of material other than stool (due to fistulas, prolapsing haemorrhoids, anorectal neoplasms, sexually transmitted diseases and poor hygiene) and frequent defecation movements and urge sensations without loss of anal contents (due to inflammatory bowel disease, pelvic irradiation, irritable bowel syndrome, and low anterior resection of the rectum). Careful diagnostics should differentiate between pseudo incontinence and clinically important FI.¹⁷

Diagnostic procedures

To determine the nature and seriousness of FI, different objective diagnostic procedures are available. A medical history and physical examination precede the additional diagnostic investigations of the rectum, anus and pelvic floor. Ultimately, a functional or anatomic diagnosis is made. The outcome of diagnostics guides decision-making concerning therapy and, at the same time, provides objective baseline data to evaluate the selected treatment.

Medical history

The following topics should be addressed during the medical history: start of FI, frequency and constitution of lost faeces, accompanying blood or mucus loss. The presence of constipation, incomplete evacuation, soiling, diarrhoea, prolapse, urinary incontinence, pain during defecation and urge incontinence (inability to defer defecation for 15 min) should be checked. Furthermore, the use of pads or specific medication concerning FI is checked. Inquiry on some of these aspects can be obtained by a (stool) diary.

The obstetric history should also establish: number of deliveries, duration birth, weight of the child, forceps or vacuum delivery, episiotomy or sphincter rupture. Previous intestinal, gynaecological, urological or colorectal surgeries should be reported and the occurrence of other surgeries, inflammatory diseases, neurological diseases or diabetes should be verified. Finally, the physician must specifically ask in what way the patient is socially disabled.^{17,24}

Physical examination

The perianal area should be inspected for presence of faecal matter, scars, fistulas, dermatitis, gapping anus, keyhole deformity, haemorrhoids and skin tags. Scars can indicate previous episiotomies or perianal lacerations; a gapping anus can indicate major loss of sphincter function. A gapping anus is often associated with rectal prolapse.³³ Deformities in the anal region may be due to previous haemorrhoidectomy, fistulotomy or fissurectomy. Sometimes, chronic skin irritation is present. In addition, the patient should be asked to strain to demonstrate mucosal or rectal prolapse, perineal descent, or paradoxal straining.

Rectal toucher is used to assess perianal sensation, anal pressure at rest, pressure during contraction and during straining. Sphincter defects can be located and palpated, especially during contraction. In women, the presence of a rectocele can be established. Vaginal examination can give a good impression of the pelvic floor musculature and may detect abnormalities of the recto-vaginal wall.^{17,24}

Anorectal manometry

Anorectal manometry determines resting pressure of the anal sphincter, squeeze pressure (peak force of external anal sphincter and puborectalis muscle; normally 2 to 3 times resting pressure), pressure during straining and anal canal length.²¹ There is a reasonable correlation between pressures felt at rectal examination and measured by anal manometry.³⁶ Furthermore, anal and rectal pressures are determined during coughing, a Valsalva manoeuvre (blowing on hand, blowing a balloon) and straining. For that purpose, a probe is introduced into the rectum and withdrawn into the anal canal in small increments (station pull through) or withdrawn continuously (continuous pull through). Nowadays, different types of probes and pressure-recording devices are available. Normally, a manometric probe is used with multiple pressure sensors and a balloon.³⁶ Measurement of anal canal pressure can determine if anal canal pressure is

insufficient, but cannot determine whether this is due to a sphincter defect or neuropathy. Neuropathy is differentiated by pudendal nerve terminal motor latency (PNTML) and a sphincter defect is detected by anal endosonography and/or endoluminal MRI.³⁷

The range of normal pressure values is large due to the influence of gender and age, amongst other factors.³⁸ Patients with FI have lower pressures than healthy controls, but there is a considerable overlap of values between the two groups. Therefore, anal manometry has limited diagnostic value, but more evaluative value. Anyhow, values obtained should be interpreted in relation to other findings.^{24,36}

Anorectal sensation

Anal sensibility is a summation of properties of central and peripheral nerve function, submucosa and mucosa. It is unknown whether the actual stimulus for the induction of perception is tension or elongation of the gut wall, or reflex contraction of the gut wall or the distortion of the mesentery and adjacent structures.³⁹ An altered anal sensibility is often present in patients with FI. To measure anal sensibility, an electrode is placed in the anal canal and gradually an increasing electrical current (mA) is applied until the threshold of sensation is reached (block wave stimulus). Anal sensibility is diminished in almost all patients with anorectal disease, but the clinical relevance of this measurement is questionable.^{24,40}

Rectal sensation and compliance evaluation involves the measurement of volume and pressure during inflation of a balloon in the rectum with air or water. Both volume (V) and pressure (P) are measured during first sensation (± 10 – 20 mL), urge to defecate (± 150 mL) and maximal tolerable volume (± 250 – 300 mL). Rectal compliance can be calculated from these measures (dV/dP , normally between 4 and 10).²¹ Again, a large range of normal values is observed. Only very small maximal tolerable volumes have clinical relevance (< 60 mL). Rectal balloon expulsion can also be used to evaluate recto-anal inhibition reflex (RAIR). Impaired or absent recto-anal reflex suggests either afferent or efferent neuronal injury.^{7,41}

Neurophysiological testing

Pudendus nerve terminal motor latency (PNTML) measures the conduction time of the right and left pudendal nerve (normally 1.5–2 ms). A stimulating electrode is mounted on the index finger of the glove of the examiner and a contraction is recorded in an electrode at the base of the index finger. Long latencies are associated with neuropathy or traction injury to the nerve. Pudendal neuropathy is present in the majority of faecally incontinent patients. Uncertainty exists on the accuracy of PNTML and its predictive value on outcome.³³

Electromyography (EMG), using a concentric needle electrode or a surface electrode, can demonstrate functional muscle tissue in the anus by quantifying motor unit potentials indicative for denervation and reinnervation. Neurogenic (pudendal) damage can cause an increased fibre density, which increases the number of action potentials

derived from one motor unit. Using single fibre EMG, it is possible to record the activity of single muscle fibers.^{42,43}

Anal endosonography

Anal endosonography provides an anatomical image of the anal sphincters and pelvic floor. This technique gives an instant excellent image of the condition of the sphincters and establishes the size of a possible internal or external sphincter defect. Several ultrasound devices are available; a 360° rotating ultrasound head is preferable. Scanning is carried out with a 7.5 or 10 MHz ultrasound head.

Defects of both sphincters can be demonstrated with good reproducibility. While it is not always possible to palpate a defect, this measure can assist in detecting weakened or scarred sphincters.^{7,24}

Endoanal Magnetic Resonance Imaging (MRI)

MRI, like anal endosonography, can give a good impression of the anatomy of the anus and pelvic floor. Both MRI and anal endosonography can adequately demonstrate sphincter defects and can give an adequate impression of external sphincter function. Imaging can be achieved using conventional methods or a coil. MRI plays a major role in establishing atrophy of the external anal sphincter by demonstrating the amount of fat present and revealing the thickness and surface area of the external anal sphincter. This estimate is useful for predicting success of sphincter repair. In addition, MRI is preferred in cases with high anorectal malformations (anorectal atresia and related urogenital abnormalities), since anal endosonography does not provide adequate imaging of the pelvic floor above the level of the puborectalis muscle. However, anal endosonography is often the preferred investigation as it is easy to perform, cheap and readily available.^{7,24,41}

Defecography (evacuation proctography)

Defecography is a dynamic radiological study of attempted defecation.⁴² Thickened barium contrast is introduced into the rectum and while the patient is seated on a commode, X-ray images or a videotape of the pelvis can be recorded. Abnormalities, such as a rectocele and rectal intussusception (incomplete rectal prolapse), can then be detected during rest, straining and squeezing.^{44,45} Because such abnormalities can also be found in normal subjects, defecography is of uncertain value in the evaluation of a patient with FI. In demonstrated rectal prolapse, rectopexy can be offered with expectation of a good clinical result.

Severity scores

Besides an anatomical and functional diagnosis, it is of interest to the clinician to get an impression of the severity of FI independent of direct clinical examination. Assessment can be done at any time and provides a cost-effective means of monitoring the patient's status.⁴⁶

The severity of FI is not only dependent on the consistency, frequency, amount and moment of stool loss, but also on factors like use of coping mechanisms and altered lifestyle.⁴⁷⁻⁴⁹ Several severity scores have been developed to get a quick impression of the severity and evaluate progress during or after therapy (Table 2.2 and Box 2.1).

The most important differences are the domains of severity to be assessed, the use of weighting and the range of the summary scores. Scoring systems should ideally be reliable, valid and responsive (sensitive to change).⁶⁰ To assess responsiveness it is important to determine what constitutes a clinically important change, and the minimum level of change which must occur before change is detected by the instrument.

Some scoring systems are simple and easy to use, but they lack discriminative power and responsiveness given the small number of grading categories.^{50-52,61} Other scoring systems award identical scores for different degrees of dysfunction (incontinent to gas, liquid or solid stool). This may not reflect the real subjective experiences of an individual, since a summary score can be made up in different ways. In this case, weighted scales are preferred, although weights are often arbitrarily chosen and Vaizey et al. found that The American Medical Systems score was not superior to the Vaizey score.⁵⁷

Other scores proposed incorporating pad use and lifestyle alteration.^{11,43,53,56,57} However, these items do not necessarily reflect severity of FI. Lifestyle alteration might be influenced by emotional status, coping skills or concomitant disease. Pad use might be influenced by patients' insecurity or the presence of urinary incontinence. Therefore, lifestyle alteration and pad use are found to be more associated with quality of life in FI than severity of FI, which are two different concepts.⁶⁰

In an attempt to incorporate amount of faeces loss in a severity score, the PREFAB-score was developed. Amount of faeces loss determines to a great extent social impact and severity of FI. The PREFAB-score is a modified version of the widely used and accepted PRAFAB-score in the urinary incontinence field.⁴⁷ The score has proven to be reliable, valid and responsive.⁶² Since most of the PRAFAB-score, except for the 'amount' item, is applicable to faecally incontinent patients, it can be adapted to assess faecally incontinent patients as well. The feasibility and validity of the PREFAB-score is currently being evaluated in clinical practice.

Future research in the field of self-reported severity measures should concentrate on the need for weighting and the use of coping mechanisms as an indicator of severity.

Table 2.2 Self-reported severity measures

	Domains of severity	Weighted	Range	Strength ²	Weakness ³	Reliability	Validity	Sensitivity to change ⁴
Grading scales								
Bule ⁵⁰	2,4	No	Excellent–Poor	1	1,2,3			
Parks ⁵¹	1	No	Grade 1–4	1	1,2,3			
Williams scale ⁵²	1,2	No	Grade 1–5	1	1,2,3,5			
Summary scales								
Hull ⁵³	2,4,5	No	0–36	1,2	1,2			
Jorge/Wexner ⁴³	1,2,4,5	No	0–20	1,2	1,2,4	0.75 ^b		0.87
Lunniss ⁵⁴	1,2,6	No	0–13	1,2	1,2,4			
Pescatori ⁵⁵	1,2	No	0–6	1,2	1,2,3	0.58 ^b		0.87
PREFAB ^a	2,3,4,5	No	6–20	1,2	2			
Rockwood ⁴⁶	1,2	Externally developed weights	Patient rate: 0–61 Surgeon rate: 0–57	2	1,4	0.75 ^c	Convergent ^e	
The American Medical Systems score ⁴⁶	1,2,5	Weighted	0–120	2	1,4	0.84 ^b		0.86
Vaizey ⁵⁷	1,2,4,5,6	No	0–24	1,2	1,2,4	0.87 ^b 0.88 ^d	Convergent ^f	0.94
Vioili ⁵⁸	1,2	Self-determined weights	0–20	1,2	1,4			

¹Domains of severity: 1=consistency of stool loss; 2=frequency of stool loss; 3=amount of stool loss; 4=use of coping mechanisms (wearing pad or diaper); 5=impact or changes in life; 6=assessment of urgency.

²Strength: 1=easy to fill out; 2=summary score.

³Weakness: 1=does not take into account amount of stool loss; 2=all items are equally important in determining severity; 3=does not take into account that a patient can be incontinent for more than one faecal consistency; 4=does not take into account that a patient sometimes does not have a certain faecal consistency, so you cannot score all items; 5= vague quantifiers, like occasional.

⁴Correlation (Pearson *r*) between incontinence grading system and the investigators' assessment of improvement.

^aThe PREFAB-score is a modified version of the widely used and accepted PRAFAB-score in the urinary incontinence field. The feasibility and validity of the PREFAB-score is currently evaluated in clinical practice.

^bTest-retest reliability: intraclass correlation coefficient (ICC).

^cTest-retest: Pearson *r*, telephone administrations.

^dInter-observer reliability: intraclass correlation coefficient (ICC).

^eSignificant correlation of the surgeon and patient severity scores with three out of four domains of the Fecal Incontinence Quality of Life Scale.

^fSignificant correlation with clinical assessment: 0.79.

Box 2.1 Definitions of reported validity items⁵⁹

Content validity	The extent to which the concepts of interest are comprehensively represented by the items in the questionnaire
Face validity	Face validity pertains to whether the test measures at first sight what it is assumed to measure ('looks valid' to the examinees who take it, the administrative personnel who decide on its use, and other technically untrained observers)
Criterion validity	The extent to which scores on a particular instrument relate to a gold standard (positive rating: correlation with gold standard is ≥ 0.70)
Predictive validity	The extent to which a measure predicts scores on some criterion measure (disease or health status)
Construct - convergent validity	The extent to which scores on a particular instrument relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured
Construct - discriminant validity	The extent to which scores on a particular instrument do not relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured
Reproducibility	The degree to which repeated measurements in stable persons (test-retest) provide similar answers
Internal consistency	Extent to which items in a questionnaire (sub)scale are correlated (homogeneous), thus measuring the same concept (positive rating: Cronbach's alpha, 0.70–0.95)
Sensitivity to change	The ability of a questionnaire to detect clinically important changes over time, even if these changes are small

Quality of life scales

In addition to generic quality of life scales, like the SF-36, condition-specific quality of life scales have been developed, like the Fecal Incontinence Quality of Life Questionnaire (FIQL), the Gastrointestinal Quality of Life Index (GIQLI) and the Manchester Health Questionnaire.^{63,64} The FIQL objectively measures the physical, psychological, and social impact of FI on the patient's lifestyle. Psychometric evaluation showed reliable and valid measurement results.^{60,64,65} The FIQL is more responsive, but the GIQLI is useful in comparing different gastrointestinal conditions.

Generic scales miss the responsiveness (questions) for specific conditions, but can often adequately assess gross changes. In addition, generic instruments allow comparisons between disease groups and between studies.⁶³ Table 2.3 summarises the psychometric properties of several condition-specific and generic instruments.

CONSERVATIVE TREATMENT

Treatment of faecally incontinent patients consists of conservative as well as surgical interventions. Conservative interventions consist of dietary adaptations, medication, bowel management, absorbent materials, and physiotherapy (Tables 2.4 and 2.5).

Dietary and pharmacologic treatment

Initially, most patients with FI are treated by dietary or medical means. Medical management is mostly directed at altering stool consistency, stool volume, and colon transit time. A high fibre diet, sufficient fluid intake and stool-bulking agents might improve stool volume and consistency. In addition, dietary adaptations can improve regularisation of the timing of defecation.

Table 2.3 Psychometric properties of condition-specific and generic instruments^a

	Reliability	Validity	Sensitivity to change
Condition-specific instruments			
FIQL ⁶⁴	-Test-retest: scores do not differ statistically, no reliability coefficient -Internal consistency ^b : 0.8–0.96	-Convergent ^d : 0.28–0.65 (compared to SF-36) -Discriminant: scores differ significantly from controls	?
GIQLI ⁶⁶	-Test-retest ^c : 0.92 (CI: 0.77–0.97) -Internal consistency (Cronbach's alpha): >0.90	-Convergent: Pearson <i>r</i> with QL Index: 0.53 Pearson <i>r</i> with the Affect Balance Scale: 0.42 -Discriminant: mean score normal individuals significantly higher -Discriminant: mean score individuals mobile in the community, confined to home and for bedridden patients significantly different	GIQLI before and after surgery (2 FUs) is significantly different
Manchester Health Questionnaire ⁶⁷	-Test-retest ^d : 0.81–0.93 -Internal consistency ^b : 0.73–0.91	-Convergent ^d : 0.30–0.65 (compared to 2 items of symptom inventory) -Criterion (compared to SF-36): –0.35 to –0.77 -Content: +	?
Modified Manchester Health Questionnaire ⁶⁸	-Test-retest ^{d,e} : 0.55–0.98 -Internal consistency ^b : 0.79–0.92	-Convergent ^d : 0.93	?
Generic instruments			
MOS (SF-36) ^{f,69,70}	-Test-retest: + -Internal consistency: +	-Content: + -Concurrent: + -Criterion: + -Construct: + -Predictive: +	+
MOS (SF-20) ⁷¹	-Internal consistency ^b : 0.81–0.88	-item convergent validity ^d : 0.45–0.79 -item discriminant validity ^{d,g} : +	?
EuroQol ^{72,73}	?	-Construct: +	?

CI, confidence interval; FU, follow-up; QL, Quality of Life.

^a+ = positive rating for validity item; ? = unknown/insufficient information.^bCronbach's alpha per domain.^cIntraclass correlation coefficient.^dPearson correlation per domain.^eTelephone-administered.^fPsychometric properties are evaluated in numerous publications.^gEach item correlates significantly higher with its hypothesised scale than with other scales.

Table 2.4 Conservative therapies for faecal incontinence

A. Dietary
1. Fibre
2. Fluid intake
3. Stool-bulking agents
B. Pharmacologic
1. Motility-altering agents
2. Stool-altering agents (volume, consistency)
C. Bowel management
1. Retrograde/antegrade colonic irrigation
2. Enema use
D. Products and appliances
1. Pads and pants
2. Anal plug
E. Physiotherapeutic interventions
1. Pelvic floor muscle and sphincter training
2. Electrical stimulation
3. Biofeedback
4. Rectal balloon training

Table 2.5 Therapeutic options and goals regarding conservative treatment of faecal incontinence

Therapy	Goal
A. Dietary	Altering stool consistency Altering stool volume Altering transit time Regularisation of timing of defecation
B. Pharmacologic	Altering stool consistency Altering stool volume Altering transit time Regularisation of timing of defecation
C. Bowel management	Colonic cleansing
D. Products and appliances	Prevention of faecal passage or soiling
E.1 Pelvic floor muscle training	Improve coordination of muscle contractions Improve muscular strength Increase awareness and isolated contraction of pelvic floor muscles and anal sphincter
E.2. Electrical stimulation	Increase awareness and isolated contraction of pelvic floor muscles and anal sphincter
E.3 Biofeedback	Information on activity anal sphincters and puborectalis muscle (EMG-BF) Increase awareness and isolated contraction of anal sphincter and pelvic floor muscles (EMG-BF). Accent on timing and coordination.
E.4 Rectal balloon training	Decrease sensory threshold in case of passive FI (P-BF) Increase sensory threshold in case of urge FI (P-BF) Improve rectoanal inhibitory reflex (P-BF)

EMG-BF, electromyographic biofeedback using intra-anal electromyographic sensor/probe or perianal surface electromyographic electrodes; P-BF, pressure biofeedback using rectal balloon.

Bowel management

Retrograde colonic irrigation can be considered before proceeding to surgery or as an additional therapy after failed surgery.⁷⁴ The use of retrograde colonic irrigation in the morning is effective: it keeps the rectum empty and can diminish the chance of FI during the day. Cleansing enemas are used in patients with overflow incontinence for disimpaction. In this case, enemas alter the consistency of stool and provoke the desire to defecate.

There is not much known about the success rate of retrograde colonic irrigation. Only a few studies have been published to date. Retrograde irrigation has been reported to

be minimally invasive, easy to learn and safe, with only minor side-effects.⁷⁵ Colonic irrigation can be performed in different ways: by retrograde rectal irrigation, through a colostomy, or by antegrade irrigation with a Malone antegrade colonic enema (MACE), a transverse or sigmoid colostomy (ACE).⁷⁴

Dietary treatment, pharmacologic treatment and bowel management usually make the patient feel more secure.

Anal plug

An anal plug, designed to block the loss of stool, consists of a cup-shaped foam plug, with a gauze string for removal.⁷⁶ Patients with abnormal anorectal sensitivity, in particular, achieve continence with its use. Furthermore, the odour of lost stool can be controlled. However, achievement of continence is only possible if the plug is supported by a functioning pelvic floor. Provocation of local irritation and a desire to defecate are also disadvantageous features. Accordingly, the anal plug seems to be an option for incidental rather than continuous use in selected patient groups.^{24,76,77}

Physiotherapeutic interventions

Physiotherapeutic interventions are non-invasive and inexpensive, require no sophisticated equipment and have hardly any side-effects. Moreover, all other therapeutic options are still available while physiotherapeutic interventions are being used.⁷⁸ For these reasons, physiotherapy is often tried before surgical treatment. Physiotherapeutic management of FI includes pelvic floor muscle training (PFMT), biofeedback (BF) (including rectal balloon training (RBT)) and electrical stimulation (ES) and is offered by pelvic physiotherapists. Often, one or more physiotherapeutic treatments are combined, depending on the underlying cause of FI.

Patients with such an embarrassing condition like FI must be treated carefully. Therefore, prior to physiotherapeutic interventions, explanation and information should be given about the disorder. One determinant of success of physiotherapy is a reliable relationship with the therapist and motivation of both the therapist and the patient.⁷⁹ After diagnostic assessment, manually directed techniques or specifically designed equipments are used to treat the motoric, sensory or coordination component of the disorder. The chosen patient-specific treatment protocol is often directed by the PERFECT-assessment.⁸⁰ Table 2.6 explains each of the characters of the PERFECT-acronym. The power represents the strength of the pelvic floor muscles and is measured using the modified Oxford grading system (Table 2.6). This Oxford scale is internationally accepted and is the most frequently used muscle grading method, however, the inter-observer variability has been reported to be high.⁸¹ Assessment of endurance strength and peak force strength helps to prescribe an appropriate exercise program. Patients graded 0, 1 or 2 on the modified Oxford scale seem to be more suitable for rectal balloon training and/or electrical stimulation, instead of pelvic floor muscle exercises.¹⁸

Table 2.6 The PERFECT-assessment scheme and modified Oxford scale⁸⁰

P	Power (pressure)
	0 No muscle contraction
	1 Flickering contraction
	2 Weak contraction
	3 Moderate contraction
	4 Good contraction
	5 Strong contraction
E	Endurance
R	Repetitions
F	Number of fast maximum, one-second contractions
E	Elevation of the pelvic floor
C	Cocontractions
T	Timing and coordination

Pelvic floor muscle and sphincter training

The pelvic floor muscles support the abdominal organs and work tonically and reflexly to maintain continence. Approximately 70% slow-twitch and 30% fast-twitch fibres are present in the pelvic floor muscles to serve this purpose.⁸⁰ Patients suffering from urinary or faecal incontinence often show weakened muscle function. In 1948 Arnold Kegel was the first person to develop exercises aimed originally at controlling continence in postpartum women.⁸² PFMT aims to restore the muscular strength, coordination and timing of contractions.

Exercises consist of selective voluntary contractions and relaxations of pelvic floor muscles and anal sphincter with a repetitive character. Exercises can activate latent motor units to the point that the muscle becomes functional again.⁸³ Exercises in a progressive resistance program adhere to the basic muscle training principles. The principle of overload is based on stimulation of the muscle beyond its normal level of performance. To evaluate this principle the PERFECT-assessment is necessary prior to training. The principle of specificity refers to training a muscle in the way the muscle needs to be used. Exercises are adapted to slow-twitch fibres (endurance exercises) and fast-twitch fibres (power and speed exercises). The principles of maintenance and reversibility alert the patient to train regularly, sometimes for life. Inactivity will convert the muscle to its pretrained status and symptoms can occur again.⁸⁴ Awareness of the different muscles involved in maintaining continence is necessary to be sure of avoidance of cocontractions of surrounding muscles (abdominals, buttocks, thighs and back) and ensure activation of the relevant muscles. Sometimes, when patients find and use the relevant muscles at the appropriate time, symptoms can reduce at once.⁸⁵ Ultimately, exercises should be practised in different starting positions, from lying and sitting to standing, simulating every-day situations as much as possible.¹⁸

Electrical stimulation

The use of electrical stimulation (ES) for the treatment of urinary and faecal incontinence spans a 35-year period with apparent success.^{83,86} ES is achieved by applying an electrical current, thereby passively stimulating the pelvic floor muscles, sphincters and accompanying nerve structures. The purpose of ES is to re-educate weakened and poorly functioning pelvic floor muscles by means of increasing awareness and isolated contraction of the stimulated structures.⁸⁶ ES is often used as an adjunct to pelvic floor muscle exercises and biofeedback therapy, to assist with identification and isolation of pelvic floor muscles and to increase contraction strength.

In ES, the number of motor units recruited is dependent on a number of factors. These include the parameters of the electrical stimulus, impedance (resistance to the flow of the current) and the size and orientation of the electrodes. The electrodes should be placed as close as possible to the pelvic floor muscles. The electrical stimulus should be stimulating enough to depolarise the nerve, but uncomfortable sensations should be avoided.⁸⁷

The precise mechanisms by which ES can restore faecal control are not well understood: in the case of optimising the gracilis neosphincter, a transformation of fast-twitch muscle fibres to less-fatigable slow-twitch muscle fibres is hypothesised, including increases in capillary density. Some studies have shown an increase in axonal budding following denervation.⁸⁷ Changes in muscle fibre diameter may be important as well. Besides physiological and metabolic changes, enhanced awareness of the anal sphincter is essential.⁸⁷ A possible working mechanism of neuromodulation through efferent or afferent nerve stimulation needs to be further investigated.

Contra-indications for ES are anal infections, rectal bleeding, complete denervation of the pelvic floor (will not respond), swollen/painful haemorrhoids, deficient sensation, atrophy of mucosa, six week period after surgery, pacemaker, dementia, pregnancy and pain during palpation.⁸⁸

Biofeedback and rectal balloon training

Haskell and Rovner made the first attempt at biofeedback for anal incontinence in 1967.⁸³ They reported the successful use of electrodes and electromyography in 71% of patients treated for anal incontinence. Cerulli was the first to use biofeedback by way of insertion of three balloons: one intrarectal (to allow rectal distension) and two intra-anal (to record internal and external sphincter contractions separately).⁸⁹

Many authors consider biofeedback to be a useful adjunct to PFMT or ES in patients with FI. Biofeedback is a technique that monitors biological signals and electrically amplifies these to provide feedback to the patient. Biofeedback intends to control physiological processes that are normally under involuntary control.

Biofeedback was originally described as an operant conditioning therapy.⁹⁰ Positive confirmation will encourage the performed response, in contrary to negative confirmation. Biofeedback is effective for a variety of medical conditions: headache, urinary incontinence and chronic pain. Engel et al. applied operant conditioning of recto-

sphincteric responses in the treatment of FI.⁹⁰ Originally, the aim was to counteract internal sphincter relaxation due to rectal distension by forceful contraction of the external anal sphincter with minimal sensory delay. Nowadays, three modalities of biofeedback in the treatment of FI can be recognised:⁹¹

1. An intra-anal electromyographic (EMG) sensor, an anal manometric probe (measuring intra-anal pressure change), or perianal surface EMG electrode is used to inform the patient about the activity of the pelvic floor muscles by way of a visual display and/or an auditory signal. The patient attempts to aim the response to the ideal response. The goal of this treatment modality is to create awareness of the squeezing musculature and strengthen it without rectal distension. In addition, the correct muscle response and progress of the patient can be demonstrated. Training can focus on endurance force (submaximal contraction sustained for prolonged time) or increase of squeeze amplitude (peak force). The exercises are based on 'Kegel' exercises, originally used for urinary incontinence.⁹²

2. The second modality involves the use of a manometric rectal balloon (rectal balloon training). The rectal balloon is filled with air to imitate rectal contents. The patient with an elevated sensory threshold is trained to discriminate smaller rectal volumes, resulting in an earlier warning from stool entering the rectum and earlier external sphincter response to counteract reflex inhibition of the internal sphincter.⁷⁸ However, progressive distension of the rectal balloon is used in patients with a hypersensitive rectum to resist urge feelings.

3. The third modality is a three-balloon system (a balloon-tipped water perfused catheter or a Schuster-type three-balloon probe) used to train a forceful external anal sphincter contraction after a stimulus of rectal distension.^{49,90} In this way, external sphincter contraction counteracts relaxation of the internal anal sphincter due to rectal distension. This treatment was originally described by Engel et al.^{90,93} Some authors feel that sensory delay is an important factor in FI.⁷⁸

Overall, biofeedback gives feedback about the possibility, degree and quality of contracting and relaxing the pelvic floor, and gives feedback on the coordination between rectal distension and the contracting anal closing system.⁸³ Patients most likely to benefit from biofeedback include those who have motivation, intact cognitive skills, some rectal sensation, and nearly intact sphincters and innervation.^{43,94,95} It is reported that patients with neurological deficits (diabetes, spina bifida, multiple sclerosis) are less likely to be treated successfully.⁹⁴

Even though continence is achieved after physiotherapeutic treatment, the recto-sphincteric reflexes sometimes remain abnormal, suggesting that the external sphincter response to rectal distension is an unreliable predictor of treatment outcome.⁹³

Effectiveness of physiotherapeutic interventions

Randomised controlled trials are thought to be the fundamental paradigm of clinical research and represent the strongest scientific proof of effectiveness of a particular intervention.⁹⁶ Evaluating effectiveness of physiotherapeutic interventions has been impeded by lack of standardisation, a large variety of outcome measures and low methodological quality.⁹¹

Methodological quality of published trials

Table 2.7 gives an overview of the methodological quality of published (randomised) controlled clinical trials, randomised cross-over designs and single case experiments concerning effectiveness of physiotherapy in FI. To assess the methodological quality, the Delphi checklist with nine pre-described items is used (Table 2.8). This Delphi list contains criteria for internal validity, external validity and statistical analysis.⁹⁷

Only four out of 14 trials scored higher than the cut-off point of four points (out of a possible 9). Single or double blinding was often impossible due to the study design or the double role of therapist and assessor of data, resulting in a lower attained maximum score. The majority of trials did not mention concealment of randomisation. Statements about similarity of intervention groups at baseline, blinding and analysis on an intention-to-treat basis were also insufficient in most studies.

Table 2.7 Methodological quality of published (randomised) controlled trials

Author	Publication type	D1	D2	D3	D4	D5	D6	D7	D8	D9	Total ^a
Norton ⁹⁸	RCT	+	+	+	+	±	±	+	+	+	7
Solomon ⁹⁹	RCT	+	+	+	+	+	-	-	+	+	7
Mahony ¹⁰⁰	RCT	+	+	+	+	+	-	-	+	-	6
Norton ⁷⁹	RCT	+	+	+	+	±	-	-	+	+	6
Fynes ¹⁰¹	RCT	+	±	+	-	+	-	-	+	-	4
Heymen ⁹⁴	RCT	+	±	±	+	±	-	-	+	-	3
Latimer ⁹³	SCE	+	±	±	-	±	±	-	+	+	3
Guillemot ¹⁰²	CCT	-	-	+	-	±	-	-	+	-	2
Illycky ¹⁰³	RCT	+	±	±	+	±	-	-	-	-	2
Miner ⁷⁸	RCO	+	±	±	-	±	-	±	+	±	2
Suhr ¹⁰⁴	CCT	-	-	±	+	±	-	-	+	±	2
Enck ¹⁰⁵	CCT	-	-	-	-	-	-	-	+	-	1
Sprakel ¹⁰⁶	CCT	-	-	-	+	-	-	-	-	-	1
Whitehead ¹⁰⁷	CCT	-	-	±	-	±	±	-	+	±	1

RCT, randomised clinical trial; SCE, single case experiments; CCT, controlled clinical trial; RCO, randomised cross over design.

^ayes (+)=1 point; no (-)/don't know (±)=0 points.

Table 2.8 Delphi list

Criteria		
D1	Was a method of randomisation performed?	Yes/no/don't know
D2	Was the treatment allocation concealed?	Yes/no/don't know
D3	Groups are similar at baseline regarding most important prognostic indicators?	Yes/no/don't know
D4	Were eligibility criteria specified?	Yes/no/don't know
D5	Was the outcome assessor blinded?	Yes/no/don't know
D6	Was the care provider blinded?	Yes/no/don't know
D7	Was the patient blinded?	Yes/no/don't know
D8	Were point estimates and measures of variability presented for the primary outcome measures?	Yes/no/don't know
D9	Did the analysis include an intention-to-treat analysis?	Yes/no/don't know

Internal validity items: D1, D2, D3, D5, D6, D7, D9; External validity items: D4; Statistical items: D8.

Evidence-based effectiveness of physiotherapeutic interventions

PFMT has been proven effective in the treatment of stress urinary incontinence.¹⁰⁸

Since the anal sphincter and puborectalis muscle form a part of the same pelvic floor as the closing urethral system, expectations are raised for the same positive results of pelvic floor muscle re-educative techniques in FI. Wilson et al. concluded that PFMT, properly taught, is still the mainstay of physiotherapy.¹⁰⁹

Success rates of physiotherapy in FI are generally based on numerous uncontrolled trials, mainly focusing on BF therapy. There are over 60 uncontrolled trial reports in the literature on the use of BF for the management of FI.¹¹⁰ Some authors conclude that BF is the treatment of choice for FI on the basis of these observational studies¹¹¹ and controlled clinical trials.^{102,105} An overall cure and improvement rate of 72% has been reported.¹¹² However, the results of a Cochrane review on the effects of BF and/or PFMT for the treatment of FI in adults were based on only eleven randomised controlled trials and showed that some elements of BF therapy and sphincter exercises might have a therapeutic effect, but this is not certain.¹¹⁰

Moreover, it was suggested that RBT improved continence more than sham training. This is in agreement with other authors who consider lowering the threshold volume for discrimination of rectal distension an important factor in the success of BF therapy.^{78,93,113,114}

In an attempt to assess the effect of the different BF components (balloon/pressure BF, electromyographic BF) in the management of FI, two complicated cross-over trials have been performed.^{78,93} Group comparisons were impossible due to the small sample in one trial and the single case experiments in the other.

A second Cochrane review evaluated ES in adult patients with FI.⁸⁶ Insufficient data was available to allow reliable conclusions on the effect of ES in the management of FI.⁸⁶ A recently published randomised controlled trial examined whether anal ES, using an anal probe electrode in the absence of any adjunctive exercises or advice, would improve symptoms of FI and anal sphincter pressures when compared with 'sham' ES. Patients rated that their bowel control had improved to a modest extent. However, there was no statistically significant difference detected between the groups, suggesting that 1 Hz was as effective as 35 Hz. This raises the possibility that the main effect is

not sphincter contraction but sensitisation of the patient to the anal area, or simply the effect of intervening per se.⁹⁸ This result is in agreement with Mahony et al.¹⁰⁰ who concluded that the addition of ES did not enhance symptomatic outcome, and Suhr et al., who concluded that ES is inferior to pressure BF.¹⁰⁴ In contrast, Sprakel et al. found ES to be effective in the conservative treatment of anorectal incontinence.¹⁰⁶

Finally, the results of a recent large cohort study on ES and PFMT with BF for patients with severe FI indicated that pelvic floor muscle rehabilitation leads to a modest improvement in severity of FI, squeeze pressure and maximal tolerated volume.¹¹⁵ Although uncertainty exists on the effectiveness of physiotherapeutic interventions in FI, results are promising.¹¹²

Which patients benefit from physiotherapy?

Amongst most studies, authors try to find patient characteristics that respond best to physiotherapy treatment. Manometric data appears to be no reliable predictor of outcome.^{93,105} Patients can improve sensory threshold, anal canal pressures or recto-sphincteric response to rectal distension, without an accompanying decrease in severity of FI. On the other hand, patients can experience fewer symptoms, without changes in manometric data. Unlike these contrary findings there are some accepted predictors.

Physiotherapy seems to be most beneficial in case of a sphincter deficit, which causes both abnormal lower and upper anal canal pressures and abnormal squeeze pressures.¹⁰² Decreased anal canal pressures can be influenced by strengthening the external anal sphincter and puborectalis muscle, which are both under voluntary control. Poor outcome is more common in patients with a major sphincter defect or trauma of the anal sphincter after surgery.^{102,104,106} Other conditions associated with poor response include severe organic lesions, low anterior resection, keyhole deformity and irritable bowel syndrome. In patients with neurological involvement (diabetes, spina bifida, multiple sclerosis), suboptimal results can be expected as rectal sensation is largely disturbed or even absent.^{94,116} Prather identified duration of FI, pudendal nerve terminal motor latency, patient age, symptom severity, pre-treatment anal sphincter pressures and anal ultrasonography as factors which were not important in predicting outcome following biofeedback retraining.¹¹⁶

According to Sprakel, all aetiological variations are seen as indications for electrical stimulation treatment, given the absence of differences between posttraumatic/post-operative or idiopathic origin of the insufficiency of the sphincter mechanism.¹⁰⁶ Finally, motivation and cognitive skills are associated with achieving best results.^{116,117}

SURGICAL TREATMENT

When conservative treatment fails, surgical treatment is considered. There is little evidence to either support or refute surgical interventions for FI.¹¹⁸ Surgical procedures involve interventions to anatomically and mechanically correct the pelvic floor or anal sphincters (Table 2.9). Mostly, defects result from obstetric trauma, surgery for anal fissures, haemorrhoids or fistulas. To determine which surgical treatment is most suitable, causal determinants of anatomic or functional nature need to be analysed. Underlying causes of soiling or diarrhoea need to be excluded and treated specifically.²⁴

Table 2.9 Surgical interventions for faecal incontinence

-
A. Surgery of the anal sphincters/pelvic floor
1. Direct apposition (classical repair)
2. Plication or reefing
a. anterior - preanal repair
b. posterior - postanal repair
c. Combined (total pelvic floor repair)
3. Overlapping
a. external sphincter only
b. combined with internal sphincter
B. Encirclement procedures
1. Silicone material
a. Injectable biomaterials
2. Neosphincter procedures
a. Skeletal muscle flaps (musculoplasties)
- Gluteoplasty
- (Dynamic) graciloplasty
b. Artificial bowel sphincter
C. Sacral nerve stimulation (SNS)
D. Colostomy
-

A. Surgery of the anal sphincters/pelvic floor

1. Direct apposition

Both ends of the ruptured sphincter are sutured, without performing a levatorplasty and overlapping sphincteroplasty. This procedure is often used by obstetricians at the time of injury after delivery.¹¹⁹ Reported satisfactory results vary between 47–100%.¹²⁰

2. Plication or reefing

a. Anterior repair/ preanal repair (anterior levatorplasty and sphincteroplasty)

Presence of a sphincter rupture, most being observed ventrally after obstetric trauma, allows for an anterior repair. Scar tissue is removed and direct apposition, plication or reefing is used to restore the anal sphincter. This procedure does not seem widely distributed and has a success and improvement rate of respectively 46% and 71%.¹¹⁹

b. Postanal repair

Presence of motoric and severe neurogenic dysfunction allows for a postanal repair. Improved continence was thought to be achieved by plication of the m. puborectalis, m. pubococcygeus, m. iliococcygeus, and external anal sphincter to restore the anorectal angle and by elongating the functional length of the anal canal.¹²⁰ Long-term

success rate of persistent improvement is 27–50%, although complete continence is rarely reached.^{24,119}

c. Combined (total pelvic floor repair)

Total pelvic floor repair involves a combination of postanal repair with anterior levatorplasty and sphincteroplasty.

3. Overlapping

a. Overlapping sphincteroplasty

Overlapping sphincteroplasty is mostly the procedure of choice in case of an isolated anal sphincter defect as seen years after the injury.¹²¹ Continuity of the sphincter in these patients is only maintained by scar tissue. Reported improvement in the short term is 60–90% of patients. However, results deteriorate with time and continence is rarely seen.¹¹⁹

b. Internal sphincteroplasty

Direct surgical repair of the internal anal sphincter (IAS) can be done in case of an isolated defect in the internal sphincter. Less information is available on its effectiveness.²⁴

B. Encirclement procedures

1. Silicone material

a. Injectable biomaterials

A weak or disrupted internal anal sphincter can cause passive FI. Use of a bulking agent such as loperamide or codeine phosphate provides only a modest response and is poorly tolerated by patients in the long term.^{19,122} Injection of silicone-based biomaterial either around the IAS or into a defect is a simple surgical solution for those who fail to respond to conservative treatment.¹²³ Earlier uncontrolled trials reported injection of autologous fat,^{124,125} injection of polytetrafluoroethylene (Teflon)¹²⁶ and injection of glutaraldehyde cross-linked collagen around the IAS.¹²⁷ These techniques have no current place in the management of FI, due to poor results and migration or degradation problems. Silicone injections (Macroplastique) have been used in the treatment of urinary stress incontinence to augment the bladder neck and increase urethral resistance.¹²⁸ Currently, the injectable silicone biomaterial, PTP™ (Bioplastique) is applied in patients with FI.¹²⁸ Animal studies show that after injection the carrier gel is excreted over a three day period, leaving the non-absorbable non-biodegradable silicone particles. The gel is then slowly replaced by a fibrin and proto-collagen matrix that surrounds the silicone particles, which at six weeks becomes stabilised with collagen fibres. The bulking particles content constitutes approximately one-third of the final volume.^{19,123} The particles seem to stay in place and do not give embolisation.¹⁹

Tjandra et al. performed the largest study so far on the use of injectable silicone material in FI. It was shown that in the medium term the treatment is safe and significantly improves outcome.¹⁹ Injection through endoanal ultrasound guidance showed better improvement in faecal continence than guidance by palpation.

There is no general agreement on injection technique.^{19,128} Under local anaesthesia, trans-sphincteric injections of the bulking agent can be performed just above the dentate line (avoiding sensitive anal lining) and below the puborectalis ring into the submucosa, as Kenefick et al. did.¹²³ This technique differed from the one used by Tjandra et al.; that is, injection through the anal skin into the inter-sphincteric space. Trans-sphincteric injections seem to avoid back-leakage of the product and injection through the anal skin is less likely to cause infections, erosions of implants, or anal pain.

2. Neosphincter procedures

Patients who fail to respond to all other medical, conservative and surgical treatments are considered end-stage patients and are suitable candidates for a neosphincter procedure. These procedures include unilateral and bilateral musculoplasties (non-stimulated and stimulated graciloplasty, gluteoplasty) and artificial bowel sphincter in cases of functional deficits with a definable muscular defect.

a. Musculoplasties

Musculoplasties are applied to create a new sphincter, thereby replacing a non-functional anal sphincter. Musculoplasties come up for discussion in cases where the anal sphincter has been denervated or damaged beyond the scope of direct repair and in cases of primary central cord disorders.

Muscles, such as the m. gluteus maximus (inferior gluteal nerve L5–S2) or m. gracilis (obturator nerve, L2–L4), are suitable because of their ample blood and nerve supply and proximity to the anus. Encirclement of the anal outlet will not improve control if a patient is experiencing reservoir problems due to e.g. inflammatory disease.¹²⁹ In patients with impaired sensitivity, good closure of the anal canal can be achieved, but the inability to sense extensive filling can lead to overflow incontinence.¹³⁰

Non-stimulated gluteoplasty

The inferior portion of the origin of each m. gluteus maximus (4–5 cm) is detached from the sacrum and coccygeus, bifurcated, and tunnelled subcutaneously to encircle the anus. The ends are sutured to form two opposite slings of voluntary muscles.¹²⁹

Graciloplasty and dynamic graciloplasty (DGP)

Graciloplasty was originally introduced by Pickrell et al. in 1952.¹³¹ In this procedure, the gracilis muscle with intact innervation and vascularisation was detached from the tuberositas tibiae, mobilised to surround the anus and fixed to the opposite ischial tuberosity. Transposition is possible as the m. gracilis is not essential with regard to

functioning of the leg. Early reported results were disappointing owing to the solitary voluntary control of this transposed muscle.^{130,132} Continence was entirely dependent on forceful contractions, which were only briefly sustained. From this point of view, electrical stimulation of the m. gracilis muscle was introduced, encouraged by the knowledge gathered for cardiomyoplasty.¹³³ Feasibility of the combination of muscle transposition and stimulation (DGP) was demonstrated by Baeten et al. in a case report.¹³⁴ Leads are implanted in the muscle and connected by way of subcutaneous tunnelling to a pulse generator, positioned subcutaneously in the abdominal wall. Before continuous stimulation is applied, the muscle is trained according to a stimulation protocol lasting approximately eight weeks.¹³⁰ Electrical stimulation brings about a transformation from fatigue-prone to fatigue-resistant muscle fibres. The alteration of morphological, physiological and biochemical characteristics result in a sustained contraction of the transposed muscle. In this way, an imitation of a normal anal function is demonstrated, which enables the patient to maintain permanent continence for a prolonged period of time.¹³² An external magnet can switch off the continuous stimulation of the gracilis muscle, resulting in muscle relaxation and defecation at a suitable moment. After defecation, the same magnet activates the stimulator. Replacement of the battery of the stimulator is required after about seven years, and involves a relatively uncomplicated procedure using local anaesthesia. A recently published multicenter study¹³⁵ demonstrated long-term efficacy of dynamic graciloplasty in the majority of patients with end-stage FI, in terms of functional and quality-of-life measures.

Geerdes et al. presented emerged complications in a series of 67 patients with a mean follow-up of 2.7 years after implantation of the neurostimulator.²⁸ The technique was successful in 52 patients (78%), whereas 22% failures occurred. These failures resulted from complications concerning technical problems (perforation of anal canal, insufficient contraction of distal part of gracilis, non-stimulatable muscle, transient leg oedema, loose wrap, device- or lead related problems), infections (usually at the side of implantation of the neurostimulator or leads), and problems attributable to an abnormal physiology of the muscle or an anorectal functional imbalance (severe constipation, overflow incontinence, soiling, non-distending rectum, strong peristalsis). Sometimes no clear reasons are indicated for failure of dynamic graciloplasty. Of course, adjustments of technique and care are considered in case of difficulties, like changing antibiotic regime.

However, despite complications, this procedure offers the possibility of preventing permanent colostomy in patients experiencing severe FI.^{130,132,136,137} Previous to surgery, good selection of patients is essential to prevent disappointment. Patients familiar with impaired sensation and/or motility problems due to congenital cause or degeneration do not seem to benefit from this technique.²⁸ Baeten et al. reported that patients with incontinence resulting from a trauma were more likely to achieve continence after the procedure (92%; n=24) than patients with pudendal nerve lesions (64%; n=14), caudal lesions (50%; n=2), or anal atresia (50%; n=12), probably because

of lack of sensibility.¹³⁰ Wexner et al. evaluated 47 prognostic factors for correlation with outcome. None was significant, except for duration of incontinent symptoms.¹³⁵ According to Pearl et al., the long gracilis tendon is relatively avascular and seems to behave more like a Thiersch wire than a dynamic sphincter mechanism.¹²⁹ To prevent ischemia, a vascular delay procedure was introduced, including ligation of the minor vascular pedicles, five weeks before transposition of the muscle.⁵² But Geerdes et al. argued that ligation of the distal vascular pedicles during mobilising did not impair blood flow in the distal part of the gracilis muscle or affect functional outcome.¹³⁸ Unlike imposed stress factors after tenotomy (atrophy and reduction in maximum power, impaired venous or lymph drainage, increase of connective tissue after loss of normal resting muscle tone and curved position), the adapted muscle functions were forceful enough to close the anal canal in a dynamic way. The most beneficial approach for muscle conditioning seems to result from delayed muscle stimulation (six weeks) instead of vascular delay or immediate stimulation.¹³⁹

b. Artificial bowel sphincter (ABS)

An alternative to musculoplasties is the artificial bowel sphincter. The ABS was adapted from the artificial urinary sphincter (AMS 800®), which was introduced in 1972 by American Medical Systems.⁶⁵ The ABS consists of an inflatable cuff that is positioned around the anus and is connected by a subcutaneous tube to a pump. This pump is implanted in the scrotum or labium major and can be unlocked when defecation is desirable. The fluid is then pumped from the cuff towards a pressure regulating balloon that is placed in the preperitoneal plane underneath the m. rectus abdominis.¹³ The ABS gives an adequate closure of the anal canal and is described to give a successful outcome in 85% of the cases with a functioning device. A major drawback is the high infection rate and the risk of erosion through the anal canal.^{65,140}

C. Sacral Nerve Stimulation (SNS)

A functional deficit of the anal sphincter muscles without a definable defect allows appliance of electrical stimulation to the sacral nerves.¹⁴¹ Peripheral nerves S2–S4 supply the striated pelvic floor, urinary, and anal sphincter muscles. Needles are inserted into the sacral foramina of S2, S3, and S4, which enable proximity to the dual sacral spinal nerves before formation of the sacral plexus. Stimulation of each sacral nerve corresponds with a typical movement of foot, heel, perineum or levator ani. Appliance of a current and gradually increasing amplitude determines the optimal motoric or sensory response. The most efficient position is chosen for temporary chronic stimulation. When satisfying results are achieved, permanent implantation is realised. A foramen electrode with a connecting lead is attached to the perist of the chosen sacral level. After subcutaneous tunnelling, the stimulating device is implanted in a subcutaneous pocket in the ipsilateral lower abdominal wall. Once implanted, the parameters of the pulse generator can be modified by telemetry and an external

magnet can be used to switch the impulse generator on and off in case of desired defecation.¹⁴²

The advantage of SNS is the prediction of effect after permanent implantation by the experimental phase. In this test phase, the function can be tested with an external stimulator. In addition, this intervention's fewer technical demands, less invasive nature, and relatively short educational course are preferable.²⁵ Disadvantages are breakage of leads, dislocation/migration, infections and pain at the site of implantation. The exact mechanism of SNS is still unclear.¹⁴³

During SNS, both efferent and afferent fibres in the sacral nerve are stimulated. Some report a motoric effect, being an increase in resting and/or squeeze sphincter pressure; others report modulation of sacral reflexes by sensible stimulation, effecting rectal sensitivity, wall tension or compliance.¹⁴⁴ A placebo effect of SNS seems to be excluded, although double blind studies to verify this are difficult to carry out given the typical tingling sensation around the anus during stimulation.^{145,146}

Electrical stimulation of the sacral spinal nerves has already been proven to be effective in the urological field.¹⁴¹ Firstly, in 1995, Matzel et al. reported two successful interventions in faecally incontinent patients; one patient improved from gross incontinence to minor soiling.¹⁴² The first Dutch experiences concerning SNS were published in 2002.²⁵ 82% of the patients showed a ≥ 50 % improvement in continence after test stimulation. Permanent implantation was performed in 71%. Mean follow-up of six months showed maintenance of effect. In 2004, a larger trial followed, which presented similar results and maintenance of effects after a one-year follow-up.¹⁴⁴ Patients after low anterior resection and patients with a partial spinal cord injury seemed to benefit minimally from SNS, given the possible extensive damage to nerve supply innervating rectum and pelvic floor.

D. Colostomy

If all other therapeutic options are unsuccessful, a permanent stoma is the only alternative to manage the incontinence problems. At least, quality of life is improved and the patient is relieved from further humiliation and social isolation.

CONCLUSIONS

From this review it is clear that FI is a common and treatable disorder. Outcome of diagnostic tests should be interpreted in relation to each other to assess the nature and severity of FI. In the management of FI physiotherapy should be tried first before proceeding to surgery. Physiotherapy shows promising results, although its effectiveness has to be further elucidated. Surgery is considered when conservative treatment is not helpful. Depending on the nature of the anatomic or functional deficit, the most suitable surgical intervention is determined. The surgical interventions have varying improvement and complications rates.

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CHAPTER 3

A SYSTEMATIC REVIEW OF AETIOLOGICAL FACTORS FOR POSTPARTUM FAECAL INCONTINENCE

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Published in: Acta Obstetrica et Gynecologica 2010;89:302-314

ABSTRACT

Background

Conflicting results are reported about the contribution of maternal, obstetric and foetal characteristics to postpartum faecal incontinence (FI), which is hampering prevention and management of FI.

Objective

To perform a systematic review identifying delivery-related aetiological factors for postpartum FI.

Search strategy

Literature searches of PubMed, EMBASE, CINAHL, DocOnline and reference lists from 1980 up to 2009 were conducted.

Selection criteria

Prospective cohort studies evaluating maternal, obstetric or foetal risk factors for postpartum FI, with a follow-up period of at least three months, were assessed. We reviewed full reports in English, German or Dutch, with anal incontinence, faecal incontinence, flatus incontinence, soiling, urgency and FI severity scores as reported outcomes.

Data collection and analysis

Data on study characteristics, methodological quality and outcome were extracted from 31 studies according to a standardised protocol. Clinical and methodological sources of heterogeneity permitted only a qualitative analysis.

Main results

A third- or fourth-degree sphincter rupture was the only aetiological factor strongly (anal incontinence) or moderately (flatus incontinence) associated with postpartum FI. No association with other postulated risk factors was found, e.g. birth weight or instrumental delivery. The potential co-existence of different risk factors impedes the interpretation of the influence of a single delivery-related risk factor.

Conclusions

This systematic review, including only longitudinal studies and recognising the importance of separating results for different outcomes, identifies that a third- or fourth-degree sphincter rupture is the only factor, which is strongly (anal incontinence) or moderately (flatus incontinence) associated with postpartum FI.

INTRODUCTION

Faecal incontinence (FI) affects activities of daily life and has been recognised as a distressing disorder. The prevalence of FI in the adult population ranges from 2 to 24%, depending upon the definition of FI used.¹ There is no consensus on methods of classifying the symptoms and causes of FI.² Classification by symptom is commonly used, for example, involuntary but recognised passage of flatus, liquid or solid stool (urge incontinence), or unrecognised anal leakage (passive incontinence).^{2,3} Others use classification by the character of the leakage: solid, liquid or flatus incontinence. Within the latter classification, the term anal incontinence (AI) is mostly used to include flatus incontinence. Other possible classifications are by patient group or by presumed primary underlying cause.²

Although FI is a common disorder, barely one third of patients report their problem to a physician or specialist, probably due to embarrassment or lack of knowledge about treatment options.^{4,5} As continence depends on several anatomical and physiological entities, deficiency of even one of these factors can result in incontinence.⁶

Although several risk factors have been reported to influence the continence mechanism and are associated with FI,⁷ delivery is considered the most important risk factor for FI in adult women,^{5,8-10} with reported prevalence rates of 10–27% for incontinence of faeces and 18–25% for flatus, at six weeks postpartum. At six months postpartum, the prevalence rates are 8–17% and 18–27%, respectively, depending on mode of delivery and perineal injury.¹¹ Postpartum FI is often associated with anal sphincter injury. The reported incidence of anal sphincter injury diagnosed by endo-anal ultrasonography in primiparous women is 26.9%, decreasing to 8.5% after subsequent vaginal deliveries.¹² However, the presence of anal sphincter injury does not always correlate with symptoms.¹³ Oberwalder et al. estimated that the probability of FI being associated with an anal sphincter defect was 76.8–82.8%.¹² Although defects are not always associated with postpartum AI, such defects are more likely to lead to symptoms in the long-term.¹⁴

Several authors have reported aetiological factors for postpartum FI based on consensus, others on review or experience.¹⁵⁻¹⁷ However, studies vary widely due to lack of consistency in the definitions of FI or to differences in methods of assessment (questionnaire/interview), in study design, in classification, in follow-up and in study population (primiparous vs. multiparous women). The conflicting results that authors have reported about the contributions of maternal, obstetric and foetal characteristics to postpartum FI are hampering prevention and management of FI.

There have been two systematic reviews on mode of delivery and postpartum FI. One was by Pretlove et al., examining mode of delivery and predisposition to AI in the first year postpartum with separate analyses for solid and liquid stool incontinence with or without flatus.¹⁸ When symptoms of solid, liquid or flatus incontinence were assessed, they found these to be associated with mode of delivery. These trends were no longer statistically significant when considering only symptoms of solid and liquid stool. The

other review by Nelson et al. was on caesarean section in the prevention of AI, for which they found no evidence.¹⁹ To date, there has been no systematic assessment, based on only longitudinal studies, recognising the importance of separating results for different outcomes and including a broad range of aetiological factors predisposing to postpartum FI. We therefore conducted this systematic review on delivery-related risk factors for postpartum FI.

MATERIAL AND METHODS

Data sources and searches

The following electronic databases were searched for relevant articles: PubMed (1980–November 2009), EMBASE (1989–November 2009), CINAHL (1982–November 2009) and the DocOnline database of the Dutch Institute of Allied Health Care (up to November 2009). All reference lists of included studies were screened for unidentified articles.

The keywords (MeSH and free text words) for searching each database and representing impairment were fecal (or faecal) incontinence in the title or in the abstract. Keywords used to represent aetiology terms were aetiology, aetiologic(al) factor(s), aetiol*(truncated), risk assessment and risk factor(s) in the title or in the abstract.

Type of study and study population

This systematic review includes prospective cohort studies, since this is considered the optimal design for assessing risk factors.¹⁵ Only studies describing singleton deliveries were considered. The final review includes only full reports in English, German, or Dutch, with a consistent follow-up period of at least three months. Aetiological factors of interest were maternal, obstetric and foetal risk factors for postpartum FI. Studies only addressing diagnostic-related risk factors (manometry, pudendus latency, or sensation thresholds) for postpartum FI were excluded because of the poor correlation between diagnostic measures and FI symptoms.¹⁶ If studies included diagnostic-related risk factors, only the maternal, obstetric and foetal risk factors were used. Studies evaluating anal sphincter rupture confirmed by endosonography were included, as this is currently the most accurate method to evaluate anal sphincter damage.^{8,12}

Outcome measures

The outcomes of interest were AI, FI, flatus incontinence, soiling, urgency and FI severity scores. Taking the classification by the character of the leakage into account, AI was defined as incontinence for flatus, liquid or solid stool. FI was defined as incontinence for liquid or solid stool. These definitions were based on the National Institute for Health and Clinical Excellence guideline.² To avoid terminological confusion, the terms AI and FI are used throughout this review as defined here.

Furthermore, studies had to present the association (crude or adjusted odds ratio/relative risk with corresponding confidence interval or correlation) or values of statistical

significance (P-value) of at least one aetiological factor with the outcome of interest. If necessary, an association measure or confidence interval was calculated from the data.

Data extraction

One reviewer (E.B.) examined all the citations and abstracts retrieved by the electronic searches. Reports of potentially relevant studies were retrieved in full text if the study met our inclusion criteria or in case of uncertainty. Two reviewers (E.B./R.B.) applied the selection criteria to the publications and three pairs of reviewers (E.B./E.H., E.B./B.B., E.B./R.B.) extracted study methods and study outcome from the included studies. Any difference of opinion was resolved by discussion and final consensus. Reviewers were not blinded regarding authors, institutions or journals.

Methodological quality assessment

Three pairs of reviewers (E.B./E.H., E.B./B.B., E.B./R.B.) independently assessed the methodological quality of identified studies, using a 14-item criteria list adapted from Kuijpers et al. (Table 3.1 and Appendix 3.1).²⁰ Disagreement among reviewers was resolved by discussion, with one reviewer (R.B.) making the final decision in case of persistent disagreement. The criteria list covered items of internal validity (items C–J and M), generalisability (items A,B,K and L) and precision (item N). A positive score indicated a well-described and well-performed item. A negative score indicated that the item was well described but not well performed, or the item was unclear due to insufficient information in the article. The total score was computed by counting all positively rated items, and ranged between 0 and 14.

A study was judged to be of 'high quality' if the quality score was nine points or more (>60% of the maximum attainable score). Studies with a lower score were judged to be of 'low quality'.

Data analyses

Depending on the homogeneity in study methods and study outcome, statistical pooling was considered. As pooling was inappropriate, a qualitative analysis was performed. Evidence for associations between each of the aetiological factors and outcome was summarised in a level of evidence table, based on Kuijpers et al. (Table 3.2).²⁰ The levels of evidence took the number of cohort studies assessing an association, consistent findings and methodological quality into account. Two sensitivity analyses were conducted to assess whether exclusion of studies with only univariable results and exclusion of those with a low methodological quality led to different conclusions.

Table 3.1 Criteria list for the assessment of the methodological quality of prospective cohort studies (score 0–14)

Study population		
A.	Description of inclusion and exclusion criteria	+/-
B.	Description of study population	+/-
Response		
C.	Response $\geq 75\%$	+/-
D.	Information about non-responders versus responders	+/-
Follow-up (extent and length)		
E.	Follow-up of at least six months	+/-
F.	Loss to follow-up/dropouts $< 20\%$	+/-
G.	Information completers versus loss to follow-up/dropouts	+/-
Outcome		
H.	Standardised assessment of relevant outcome criteria	+/-
I.	Valid measurements or attempts to validate measurements	+/-
Aetiological factors		
J.	Standardised assessment of patient characteristics and potential aetiological factor(s)	+/-
Data presentation		
K.	Frequencies of most important outcome measures	+/-
L.	Frequencies of most important aetiological factors	+/-
Analysis		
M.	Analysis controlled for confounding or effect modification	+/-
N.	Number of cases in final multivariable model at least ten times the number of independent variables in the analysis	+/-

Table 3.2 Levels of evidence

Strong	Consistent findings ($\geq 75\%$) in at least two high quality cohort studies
Moderate	Consistent findings ($\geq 75\%$) in one high quality cohort study and at least one low quality cohort study
Weak	Findings of one high quality cohort study or consistent findings ($\geq 75\%$) in at least three or more low quality cohort studies
Inconclusive	Inconsistent findings irrespective of study quality, or less than three low quality cohort studies available

RESULTS

Study selection

The electronic search yielded 5,508 citations, and reference checking yielded three additional citations (Figure 3.1). After a first screening and after removing duplicate abstracts, 140 titles remained. A second screening identified 92 titles that focused specifically on delivery-related risk factors and these were retrieved in full for further analysis. One author²¹ was contacted to obtain his manuscript, three authors were contacted for specification of the study design and four for specification of the comparisons made in the analysis for modes of delivery. Finally, 31 studies were suitable for methodological quality assessment, 28 (90.3%) of which were published in the last ten years. Three pairs of studies that were not considered independent, as they were based on the same cohort but different follow-up periods,²²⁻²⁸ were assessed separately for their methodological quality. In the qualitative analysis, however, they counted as one cohort, unless the same risk factors were analysed with no contradictory results. Two other studies^{9,11} were considered independent, as they concerned different analyses within the same cohort.

Figure 3.1 Study selection.

Methodological quality

The results of the methodological quality assessment are presented in Table 3.3. The quality scores ranged from 5 to 13 points on a 0–14 point scale, and 11 of the 31 studies were judged to be of low quality. Items C, D, G and N were insufficiently reported in most studies.

Study characteristics and outcome

Table 3.4 summarises the methods used in the cohort studies. A total of 14 cohort studies included primiparous women only and 16 included both primiparous and multiparous women. The case definition mostly incorporated a definition of FI or AI. While 18 cohort studies included fewer than 500 participants, 10 included between 500 and 1,000 participants and 3 included over 1,000 participants. Follow-up ranged from three months to ten years.

Appendix 3.2 summarises the study outcomes, including the method of outcome assessment and the presence or absence of an association in univariable or multivariable analysis. Only significant associations (crude or adjusted odds ratio/relative risk with corresponding confidence interval, or correlation, or P-value) are presented. A total of 12 cohort studies performed a multivariable analysis, 8 both a univariable and a multivariable analysis and 11 only a univariable analysis.

Table 3.3 Results of the methodological quality assessment of the included cohort studies

First author PY	A	B	C	D	E	F	G	H	I	J	K	L	M	N	Quality score	Score (%)
Baydock 2009	+	+	-	-	-	+	-	+	-	+	+	+	-	-	7	50.0
Borello-France 2006	+	+	-	-	+	-	+	+	+	+	+	+	+	+	11	78.6
Brincat 2009	+	+	-	-	+	-	-	+	+	+	+	-	-	-	7	50.0
Burgio 2007	+	+	-	-	+	+	-	+	+	+	+	-	-	+	10	71.4
Chailha 1999	+	+	-	-	-	+	+	+	-	+	+	+	+	-	9	64.3
Chiarelli 2003	+	+	+	+	+	-	-	+	+	+	+	+	+	-	11	78.6
Damon 2005	+	+	+	-	+	-	-	+	+	+	+	+	+	-	10	71.4
Eason 2002	+	+	-	-	-	-	-	+	+	+	+	+	+	+	8	57.1
Faltin 2000	+	+	-	+	-	+	-	+	+	+	+	+	+	+	10	71.4
Faltin 2001	+	+	-	+	+	S ⁺	-	+	+	+	+	+	-	-	10 ^a	71.4
Farrell 2001	+	+	+	-	+	L ⁻	-	+	-	+	+	-	-	-	9	64.3
Fornell 1996	+	+	-	-	+	+	-	+	-	+	+	+	-	-	8	57.1
Fornell 2005	+	+	-	-	+	-	-	+	-	+	+	+	-	-	7	50.0
Fruelinger 2008	+	-	+	-	+	-	-	+	-	+	+	+	+	-	6	42.9
Groutz 1999	-	+	-	-	+	-	-	+	-	+	-	+	+	+	9	64.3
Leeman 2007	+	+	-	-	+	-	+	+	+	+	-	+	-	-	5	35.7
Liebling 2004	+	+	+	+	+	S ⁺	+	+	+	+	+	+	+	-	9	64.3
MacArthur 1997						L ⁻	+	+	+	+	+	+	+	-	13	92.9
MacArthur 2001	+	-	-	-	+	-	+	+	-	+	+	+	-	-	12	85.7
MacArthur 2005	+	+	-	-	-	-	+	+	-	+	+	+	+	-	7 ^a	50.0
Mahony 2007	+	+	-	-	+	-	+	+	-	+	+	+	+	+	9 ^a	64.3
Mahony 2008	+	+	-	-	-	-	-	+	-	+	+	+	+	+	10 ^a	71.4
Meyer 2000	+	+	-	+	-	+	+	+	-	+	+	+	+	-	8	57.1
Nordenstam 2009	+	+	-	-	+	-	-	+	-	+	+	+	-	-	9	64.3
Pollack 2004	+	+	+	-	+	-	-	+	+	+	+	+	+	-	7	50.0
Serati 2008	+	+	+	+	+	+	-	+	+	+	+	+	+	-	10	71.4
Starck 2006	+	+	+	+	+	+	-	+	+	+	+	+	+	-	10	71.4
Tetzschner 1996	+	+	-	-	+	+	+	+	-	+	+	+	+	-	11	78.6
Tjandra 2008	+	+	-	-	+	+	-	+	-	+	-	-	+	+	12	85.7
Van Brummen 2006	+	+	-	-	+	+	-	+	+	+	+	+	+	-	7	50.0
Zetterström 1999	+	+	+	+	+	S ⁺	-	+	+	+	+	+	+	+	10	71.4
Total	30	28	10	7	23	11 ^b	10	31	13	31	29	28	20	9	12	85.7

PY, publication year; S, short-term follow-up; L, long-term follow-up.

^aStudy assumed to be prospective; not certain as author did not respond.

^bThree studies with different results for short- and long-term follow-up were excluded.

Table 3.4 Study methods of the included cohort studies

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases) Response at baseline (%)	Dropout	FU postpartum (range): numbers analysed (%)
Baydock 2009	Primiparous and multiparous women SVD, FD, VE Singleton vaginal delivery	Leakage of liquid or solid stool at least once every two weeks.	N=717 (23) Response= ?	4 m: 11.9%	4 m: 632 (88.1%)
Borello-France 2006	Primiparous women Single child ≥ 37 w of gestation 1 st cohort: anal sphincter tear 2 nd cohort: SVD without tear 3 rd cohort: CS Ability to participate in telephone interviews	FI: any frequency of involuntary leakage of mucus, liquid stool, or solid stool on the FISI. Flatus I: any frequency of involuntary leakage of gas on the FISI. Urgency: 'how often do you have a strong desire to move your bowels, which makes you rush to the toilet?'	N=921 (FI 91; flatus I 161; urgency 210) Response= ?	6 m: 18%	6 m: 759 (82%)
Brincat 2009	Primiparous women during pregnancy with expected singleton birth and vaginal delivery	'Have you lost control over your stools or bowel movements, that is, have you had any bowel accidents?'	N=240 (8) Response= ?	1 y: 37.1%	1 y: 151 (62.9%)
Burgio 2007	Primiparous women Single child ≥ 37 w of gestation 1 st cohort: anal sphincter tear 2 nd cohort: SVD without tear 3 rd cohort: CS Ability to do telephone interviews	FI: involuntary leakage of mucus, liquid stool, or solid stool on the FISI. Definition of FI does not include gas, but FISI computation does.	N=921 (91) ^a Response= ?	6 m: 18%	6 m: 759 (82%)
Chalilha 1999	Primiparous consecutive women SVD, FD, VE, CS Singleton pregnancy in the 3 rd trimester English speaking	FI: incontinence of flatus, liquid, solid stool and soiling of underwear. Faecal urgency: inability to defer defecation for longer than 5 min.	N=549 (37) Response= ?	3 m: 0%	3 m: 549 (100%)

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases) Response at baseline (%)	Dropout	FU postpartum (range): numbers analysed (%)
Chiarelli 2003	Primiparous and multiparous women Women with forceps/vacuum-assisted delivery and/or high birth weight of infant (≥4000 g)	Frank FI: 'Do you ever accidentally pass solid/liquid bowel motions into your underwear?' Precursor symptoms: (1) Flatus I: inability to hold wind when you really want to, (2) Soiling: noticing soiling from back passage on underwear, (3) Urgency: inability to put off going to toilet for 15 min	N=720 (FI 43; precursors 285) Response= 54.3%	12 m: 21.1%	12 m: 568 (78.9%)
Damon 2005	Primiparous consecutive women SVD of newborn in cephalic position	AI	N=197 (11) Response= ?	75 m: 72.6%	75 m (71-79): 54 (27.4%)
Eason 2002	Primiparous and multiparous women participating in trial of prenatal perineal massage SVD, FD, VE, CS	Involuntary loss of stool or passage of flatus	N=1198 (252) Response= ?	3 m: 20.8%	3 m: 949 (79.2%)
Faltin 2000	Primiparous women SVD Absence of anal sphincter tears	Incontinence to flatus, liquid, or solid stools	N=150 (22) Response= 30.9%	3 m: 4%	3 m: 144 (96%)
Faltin 2001	Primiparous women >37 w of gestation SVD, FD, VE	AI: any incontinence to flatus, liquid or solid stools since delivery and symptoms still present at three months postpartum with at least monthly events	N=100 (3 m: 16; 30 m: 11) Response= 8.9%	3 m: 8% 30 m: 23%	3 m: 92 (92%) 30 m: 77 (77%)

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases) Response at baseline (%)	Dropout	FU postpartum (range): numbers analysed (%)
Farrell 2001	Primiparous women SVD, FD, VE, CS	'Do you accidentally lose flatus or stool from the rectum?'	N=593 (FI 21; flatus I 70) Response= 85.9%	6 m: 18.4%	6 m: FI 470 6 m: flatus I 315
Fornell 1996	Consecutive primiparous and multiparous women SVD Women with and without primarily sutured anal sphincter rupture	Subjective severity of flatus I, liquid stool I, soiling and solid stool I (1=severe problems, 6=no problems)	N=82 (?) Response= ?	6 m: ?	6 m: 82
Fornell 2005	Consecutive primiparous and multiparous women SVD Women with and without primarily sutured anal sphincter rupture	Subjective severity of flatus, liquid and solid stool I	N=82 (?) Response= ?	10 y: 28%	10 y: 59 (72%)
Frudinger 2008	Primiparous women during 3 rd trimester Low-risk singleton pregnancy	Modified Wexner score	N=134 (59) Response= 95%	10 y: 20.1%	10 y: 107 (79.9%)
Groutz 1999	Unselected consecutive primiparous and multiparous women SVD, FD, VE, CS	AI: any involuntary leakage of solid or liquid faeces or gas	N=300 (21) Response= ?	3 m: ?	3 m: 300 1 y: only symptomatic women; 19 (90.5%)
Leeman 2007	Primiparous and multiparous women SVD Women with (un)sutured 2 nd degree lacerations and intact perineum ≥18 y Speaking/writing English or Spanish	AI: a score >0 on the Fecal Incontinence Scale FI: loss of loose or formed stool; a score >3 on the Fecal Incontinence Scale	N=215 (AI 49; FI 13) Response= ?	3 m: 20%	3 m: 172 (80%)

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases) Response at baseline (%)	Dropout	FU postpartum (range): numbers analysed (%)
Liebling 2004	Primiparous and multiparous women Women with an operative delivery in surgery during the 2 nd stage of labour Gestation >37 w Live born, singleton and cephalic pregnancy	Loss of control of bowels Increased passage of flatus	N=393 (FI 7; flatus 156) Response= 100%	1 y: 20.1%	1 y: 314 (79.9%)
MacArthur 1997	Primiparous and multiparous women SVD, FD, VE, CS	Frank I: 'no warning that she needed to go' Soiling or staining of underwear Urgency: felt the need to go but could not hold on	N=906 (36) Response= ?	45 w: ?	45 w (SD 10.1 w): 877
MacArthur 2001	Primiparous and multiparous women SVD, FD, VE, CS	'Do you ever lose control of wind or bowel motions from your back passage in between visits to toilet?'	N=10989 (699) ^b Response= ?	3 m: ? ^b	3 m: 7275 ^b
MacArthur 2005	Primiparous and multiparous women SVD, FD, VE, CS	FI: loss of control of bowel motions with any degree of frequency reported at both 3 m and 6 y	N=10989 (413) ^b Response= ?	6 y: 47.8% ^b	6 y (5.0-7.2): 4110 (52.2%) ^b
Mahony 2007	Primiparous and multiparous women Women following primary repair of a first recognised obstetrical anal sphincter injury	Presence of flatus I, faecal soiling, frank FI, faecal urgency and impact on daily activities (modified Wexner score)	N=500 (61 moderate; 22 severe) Response= ?	?	3 m: 500
Mahony 2008	Secondgravid women FD of first infant	Presence of flatus I, faecal soiling, frank FI, faecal urgency and impact on daily activities (modified Wexner score)	N=52 (5) Response= 27.2%	3 m: 3.8%	3 m: 50 (96.2%)
Meyer 2000	Primiparous women FD, SVD	Flatus I, liquid stool I, solid stool I	N=151 (5) Response= ?	10 m: 29.1%	10 m: 107 (70.9%)

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases)	Dropout	FU postpartum (range): numbers analysed (%)
Nordenstam 2009	Primiparous women SVD Swedish speaking	'Do you experience involuntary loss of intestinal gas/loss of stools?' 'Do you experience leakage of fluid around the anal opening?'	N=304 (74) Response= 98%	10 y: 24%	10 y: 231 (76%)
Pollack 2004	Primiparous women, Swedish speaking SVD	Incontinence to solid stool, loose stool or involuntary flatus	N=309 (85) Response= 88.5%	5 m + 9 m + 5 y: 21.7%	5 m + 9 m + 5 y: 242 (78.3%)
Serati 2008	Primiparous and multiparous women Every parity, age and gestational week at delivery	AI (gas, liquid and solid stool I)	N=521 (6 m: 24; 1 y: 23) Response= 76.4%	6 m + 1 y: 15.6%	6 m + 1 y: 336 (84.4%)
Starck 2006	Primiparous and multiparous women Women following primary suture of a 3 rd or 4 th degree perineal tear	Presence of gas I, liquid stool I, solid stool I, wears pad and lifestyle alteration (Wexner score)	N=61 (1 y: 22; 4 y: 25) Response= 88.4%	1 y + 4 y: 32.8%	1 y + 4 y: 41 (67.2%)
Tetzschner 1996	Primiparous and multiparous women Consecutive women with obstetric anal sphincter rupture	Involuntary loss of flatus or incontinent to fluid or solid faeces	N=94 (16) Response= ?	3 m: ?	3 m: 94
Tjandra 2008	Primiparous and multiparous women Women following primary repair of 3 rd or 4 th degree tear sustained during SVD	Wexner score	N=121 (25) Response= ?	3 m: 5.8%	3 m: 114 (94.2%)
Van Brummen 2006	Primiparous women with low-risk pregnancy between 12-18 w gestation Good Dutch language skills	'Do you experience incontinence for liquid or solid stool?' (FI) 'Do you experience incontinence for gas' (flatus I)	N=524 (3 m: flatus I 146; FI 26 / 12 m: flatus I 124; FI 13) Response= 55%	3 m: 12.8% 12 m: 22.3%	3 m: 457 (87.2%) 12 m: 407 (77.7%)

First author PY	Recruitment	Case definition	Number (n) of participants in cohort (number of cases)	Dropout	FU postpartum (range): numbers analysed (%)
Zetterström 1999	Primiparous women, Swedish speaking SVD	'Do you have involuntary leakage of intestinal gas?' 'Do you have involuntary leakage of stools?'	N=309 (5 m: 75; 9 m: 74) Response= 88.5%	5 m: 7.1% 9 m: 10%	5 m: 287 (92.9%) 9 m: 278 (90.0%)

■

PY, publication year; FU, follow-up; SVD, spontaneous vaginal delivery; FD, forceps delivery; VE, vacuum extraction; ?, unknown; m, months; w, weeks; CS, caesarean section; FI, faecal incontinence; I, incontinence; FISl, Fecal Incontinence Severity Index; y, year(s); g, grams; AI, anal incontinence; SD, standard deviation.

^aCases derived from paper by Borello-France 2006.

^bNumbers based on all women, but study outcome presented from only primiparous women.

Levels of evidence

Since the clinical and methodological heterogeneity of the reviewed studies precluded statistical pooling, a qualitative analysis was conducted. Appendix 3.3 presents a qualitative summary of all aetiological factors and their associations, both divided into cohort studies with quality scores $>60\%$ and $\leq 60\%$. Levels of evidence were assigned for each outcome within the 81 different aetiological factors examined. If possible, the outcome 'any incontinence' was assessed. Table 3.5 shows all aetiological factors with strong or moderate levels of evidence. A cross mark in the table indicates the outcome measure(s) for which strong or moderate evidence is available regarding an association or no association with the given aetiological factor. Since aetiological factors with weak or inconclusive evidence demonstrate conflicting evidence, the discussion focuses on factors with strong or moderate levels of evidence.

Third- or fourth-degree sphincter rupture is the only aetiological factor that was strongly (AI) or moderately (flatus incontinence) associated with postpartum FI, whereas no moderate or strong evidence of an association was found for any of the remaining factors.

Levels of evidence after sensitivity analyses

Appendix 3.4 shows the changes in the levels of evidence for aetiological factors and the corresponding outcomes after exclusion of studies with low methodological quality and of studies with only univariable results. As studies with univariable results mostly had low methodological quality, the two analyses showed very similar results. Most results changed from moderate to weak evidence because of a decrease in power (change from two to one cohort per outcome). In these cases, the highest obtainable level of evidence was weak. Furthermore, excluding cohort studies often left no cohort for the particular aetiological factor and outcome. An analysis of the aetiological factors whose level of evidence changed by at least two steps showed that all observed changes were supported by other outcomes for the aetiological factor concerned, except for the aetiological factors education and forceps delivery compared to spontaneous vaginal delivery (outcome 'any incontinence').

Table 3.5 Summary of aetiological factors for postpartum faecal incontinence with strong or moderate levels of evidence

Subgroup	Aetiological factor	Prevalence association	S/M ^a	Any I	AI	FI	Flatus I	(Mod) Wexner ^b
Maternal	Marital status	-	S	x		x		
	Obesity/BMI	-	S		x		x	
	Maternal height	-	S		x			
	Smoking	-	S	x			x	
Obstetric	Age at first birth	-	S		x			
	Age, primiparae and multiparae	-	S	x				
	Maternal weight	-	M	x				
	Gestational age	-	S/M	x (S)	x (M)			
	Parity	-	S	x	x			
	FD vs. SVD	-	M		x	x		
	VE vs. SVD	-	S	x	x	x		
	CS vs. SVD	-	S	x	x	x		
	ID vs. SVD	-	S		x	x		
	EM vs. SVD	-	M			x	x	
	ID vs. CS	-	S/M	x (S)	x (M)	x (S)		
	FD vs. VE	-	S/M	x (S)	x (M)			
	Duration 1 st stage of labour	-	S	x			x	
	Duration 2 nd stage of labour-passive	-	M				x	
	Duration 2 nd stage of labour-active	-	M	x			x	
	Total duration of 2 nd stage	-	S	x	x	x		
	Total duration of labour	-	S	x	x			
	Intact perineum	-	S	x				
	1 st -2 nd degree sphincter rupture	-	S	x	x	x		
	1 st -3 rd degree sphincter rupture	-	S/M	x (S)		x (M)	x (S)	
	1 st -4 th degree sphincter rupture	-	M	x				
	3 rd /4 th degree sphincter rupture	+	S/M		x (S)		x (M)	
		-	S					x
	Type of sphincter defect - EAS	-	M	x				
	Type of sphincter defect - EAS + IAS	-	S	x		x		
	Episiotomy	-	S	x				
	Foetal presentation at delivery	-	S	x				
	External fundal pressure	-	S		x			
	Induced onset of labour	-	S	x				
	Oxytocin stimulation	-	S	x				
	Pudendal block	-	M		x			
	Anaesthesia: epidural	-	S/M	x (S)	x (S)		x (M)	
	Analgesia: general	-	S	x				
	Marked abdominal striae	-	S	x				

Table 3.5 continued

Subgroup	Aetiological factor	Presence association	S/M ^a	Any I	AI	FI	Flatus I	(Mod) Wexner ^b
Foetal	Birth weight	-	S	x	x	x	x	
	Head circumference	-	S/M	x (S)	x (M)	x (S)	x (M)	

I, incontinence; AI, anal incontinence; FI, faecal incontinence; BMI, body mass index; FD, forceps delivery; SVD, spontaneous vaginal delivery; VE, vacuum extraction; CS, caesarean section; ID, instrumental delivery; EM, emergency caesarean section; EAS, external anal sphincter; IAS, internal anal sphincter.

^aStrong or moderate level of evidence.

^b(Modified) Wexner score.

DISCUSSION

This systematic review based on 31 cohort studies provides evidence that third- or fourth-degree sphincter rupture (either clinically obvious or detected by endosonography) is strongly (AI) or moderately (flatus incontinence) associated with postpartum FI. Besides anal sphincter injury, other potential risk factors for postpartum FI, such as age, instrumental delivery, birth weight, prolonged labour, epidural anaesthesia, obesity, and parity, have been addressed in mainly population-based surveys and reviews.^{16,17,29,30} Moreover, a consensus report considered routine episiotomy to be the most easily preventable risk factor for FI.¹⁵ Despite these reports, the present systematic review found no strong or moderate evidence for an association with other postulated risk factors, which thus do not appear to be markers of postpartum FI. This evidence is provided by a systematic search of prospective cohort studies only, which is considered the optimal design for assessing risk factors.

The following arguments might explain why our results present no association for several risk factors, although these are commonly expected: first, some risk factors are thought to be associated with sphincter rupture and therefore with FI. However, the risk factors for a sphincter rupture are not automatically the same as for postpartum FI. Given the main causes of a rupture (first delivery, instrumental delivery, occiput posterior position, high birth weight, prior tear, epidural use or episiotomy), a sphincter rupture can in this reasoning act like an intermediary factor for postnatal FI.^{11,29,30} Strong evidence that third- or fourth-degree sphincter rupture is positively associated with postpartum FI does therefore not necessarily confirm cause and effect. So, the interpretation of cause-consequence relations remains difficult even in prospective designs.

Second, the potential co-existence of different risk factors or (hidden) protective factors conceals the influence of a single delivery-related risk factor. For example, the main cause of perineal damage can be an obstetric indication (e.g., prolonged second stage) that necessitates the use of forceps delivery or vacuum extraction. Or a more carefully assisted delivery in case of a higher birth weight (based on unawareness or according to a certain protocol which can differ between hospitals). Irrespective of the different pathways by which anal sphincter rupture may be associated to postpartum FI (intermediary or multifactorial), the most important point is that this factor is a marker predisposing to postpartum FI.¹¹

Third, several methodological shortcomings (univariable analysis, chosen cut-off points etc.) can under- or overestimate the reported risks in the separate studies.

Fourth, the majority of included studies had a follow-up of no longer than one year. It is likely that sphincter ruptures lead more often to FI later in life, presumably because cumulative effects of multiple deliveries, progressive neuropathy, aging and the menopause overcome compensatory mechanisms.

Finally, some studies included in their multivariable analysis only those variables that were statistically significant after a univariable analysis. This reduces the number of

variables in the multivariable model, which is necessary due to the low number of cases in most studies (to obtain a stable model, the events per independent variable in the model should be at least ten). However, a non-significant association in a univariable analysis can change if controlled for confounding or effect modification.

A comparison of all included studies shows a number of clinical and methodological sources of heterogeneity. First, some studies evaluated only primiparous women or vaginal deliveries, whereas others included multiparous women also or evaluated high-risk deliveries. Second, different definitions of FI were used, and FI was assessed by using different measurement tools and often with unknown psychometric properties. Third, studies took different aetiological factors and confounders into account and differed in terms of follow-up periods. Methodological quality was also a matter of concern in many studies. Therefore, levels of evidence were assigned to each aetiological factor for the associated outcome, instead of a pooled analysis.

Risk factors were analysed in relation to different outcomes, as was recommended in the National Institutes of Health consensus, because different risk factors may exist for different outcomes.^{15,31} However, considering each individual outcome for an aetiological factor means that less power is available to achieve strong evidence. Therefore, all outcomes were combined in the level of evidence for 'any incontinence', to make it more likely that a sufficient number of studies with consistent results would be found.

Some methodological aspects may have influenced the results. First, we present univariable results when multivariable results were not available. Since this can introduce bias, we performed a sensitivity analysis to determine the influence of removing cohort studies with univariable results only. As the resulting changes in the aetiological factors are supported by other outcomes, the cohort studies with low methodological quality or only univariable results probably did not greatly influence our conclusions. Second, publication bias can result from the omission of relevant studies contained in unknown databases or in another language. Third, some criteria in the list for methodological quality assessment have cut-off points. A study with a minimum follow-up of six months was judged to be of higher quality because we believe that a shorter period is insufficient for perineal healing and recovery of function. Likewise, the response rate, the dropout rate and the definition of a high-quality cohort study were arbitrarily chosen and difficult to define, but in agreement with other authors.^{20,32,33} All methodological quality criteria were assigned equal weights. Future studies are necessary to provide evidence for a different classification. Fourth, 20 of the 31 (64.5%) studies recruited women after delivery, which might introduce bias since the nature of the delivery can be a reason to encourage women to enter the study.¹⁸ Fifth, differences in the prevalence rates of postpartum incontinence and related risk estimates can result from different definitions of FI, from using different measurement tools with often unknown psychometric properties, or from different postpartum follow-up periods.¹⁶ Finally, long-term results might be influenced by the cumulative effect of aging, additional deliveries, menopause and progression of neuropathy.^{17,25,34} Unfortunately, the wide range of follow-up periods

precludes clear conclusions on risk factors for short- and long-term follow-up, since separate analysis would decrease the power in assessing levels of evidence per outcome. In regard to these limitations, the conclusions should be interpreted with caution.

A third- or fourth-degree sphincter rupture after delivery is a primary marker of postpartum FI. Unfortunately, not all of these injuries are recognised at the time of delivery.²³ Moreover, women are often inadequately informed about whether they have sustained sphincter laceration during delivery and antenatal women are inadequately informed about the risks of an anal sphincter injury.^{35,36} Although anatomical defects after delivery do not always result in anorectal symptoms, women with a third- or fourth-degree tear should be told that they have an increased risk for postnatal FI. As it is an embarrassing condition, physicians should make enquiries about this condition at postnatal follow-up. Moreover, these women should be monitored carefully and support should be provided if necessary since this may prevent further cases of postpartum FI or deterioration of existing cases.^{37,38} Faltin et al. even demonstrated that the detection and repair of occult defects halved the prevalence of FI.³⁹

To achieve a reduction in postpartum FI, we need to know more about the aetiological pathways for postpartum incontinence. Future research should focus on these mechanisms and on comparing delivery modes in large prospective studies, with sufficient methodological quality, conducted with several populations, using multivariable analyses and sufficiently long follow-up periods. Finally, research should include aetiological factors that require further analysis, because they show inconclusive or weak evidence or because they have rarely been studied and may be potential risk factors (like tissue atrophy or nerve injury).⁴⁰

CONCLUSIONS

The influence of each individual aetiological factor is difficult to interpret. The results of this systematic review, including only longitudinal studies and recognising the importance of separating results for different outcomes, show that the only marker strongly (AI) or moderately (flatus incontinence) associated with postpartum FI is a third- or fourth-degree sphincter rupture.

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APPENDICES

Appendix 3.1 Operationalisation of 14-item criteria list

- +: Item is described and well performed (1 point).
 - : Item is described and not well performed, or unclear (insufficient information in the article to answer the question), or otherwise (0 points).
- Study population
- A. Positive if criteria were formulated for at least previous delivery and nature of delivery (vaginal, caesarean, instrumental, or high-risk delivery (e.g. high birth weight/anal sphincter tear)).
 - B. Positive if the most relevant baseline characteristics of the study population in relation to the research question were described.
- Response
- C. Positive if the response rate of the sampling frame was $\geq 75\%$.
 - D. Positive if information regarding patient/disease characteristics of responders and non-responders was provided to assess whether selective response could be excluded.
- Follow-up (extent and length)
- E. Positive if the follow-up period was at least six months and consistent for each participant.
 - F. Positive if the total number of loss to follow-up/dropouts of consenting participants was $< 20\%$.
 - G. Positive if demographic/clinical information regarding completers versus those lost to follow-up/dropouts (such as age and other potential aetiological factors) was provided at the main moment of outcome measurement (as clearly specified by the author) to assess whether selective loss to follow-up/dropouts could be excluded.
- Outcome
- H. Positive if standardised questionnaires or objective measurements were used at each follow-up moment.
 - I. Positive if measurements were used which have been previously validated in peer-reviewed articles or have been recommended in guidelines, or attempts were described to validate the instrument sufficiently according to the reviewer.
- Aetiological factors
- J. Positive, if standardised questionnaires or objective measurements were used at baseline.
- Data presentation
- K. Positive if frequency, percentage, mean, or median (interquartile range) and standard deviation or confidence interval were reported for the most important outcome measures.
 - L. Positive if frequency, percentage, mean, or median (interquartile range) and standard deviation or confidence interval were reported for the most important aetiological factors.
- Analysis
- M. Positive if the analysis was controlled for confounding or effect modification, including a description of potential confounders or effect modifiers.
 - N. Positive if the number of cases in the final multivariable model was at least ten times the number of independent variables included in the analysis.
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Appendix 3.2 Study outcome of the included cohort studies

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Baydock 2009	Telephone interview			Maternal age Birth weight Parity Weight gain Length of 2 nd stage of labour Postpartum UTI FD vs. SVD ^a VE vs. SVD ^a 1 st –4 th degree tear Epidural use Episiotomy			
Borello-France 2006	Telephone interview				FI: ST vs. SVD Flatus I: ST vs. SVD Flatus I: SVD vs. CS Urgency: ST vs. SVD Urgency: SVD vs. CS	OR=1.9 (1.2-3.2) OR=1.7 (1.1-2.7) OR=0.5 (0.2-1.0) OR=1.5 (1.0-2.2) OR=0.53 (0.3-0.9)	FI: SVD vs. CS
Brincat 2009	Questionnaire	Education	P=0.001	BMI Age Ethnicity, white Income, US\$ Baby weight Head circumference FD vs. SVD VE vs. SVD CS vs. SVD ID vs. SVD FD vs. CS VE vs. CS ID vs. CS FD vs. VE 1 st –3 rd degree tear Episiotomy Position of foetal head			

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Burgio 2007	Telephone interview				STC: Caucasian Antenatal UI 4 th vs. 3 rd degree tear Older age Predelivery BMI VC: UI during pregnancy	OR=6.1 (1.3-29.4) OR=2.2 (1.1-4.3) OR=2.0 (1.0-4.0) OR=1.6 (1.2-2.1) OR=1.3 (1.0-1.7) P<0.05	STC/VC/CC: Black Asian Other ethnicities Hispanic Education Marital status Head circumference Birth weight Duration of 2 nd stage of labour Labour augmentation Type of analgesia ID vs. SVD ^a Shoulder dystocia Episiotomy STC/CC: UI during pregnancy VC/CC: Caucasian Antenatal UI Older age Predelivery BMI
Chailha 1999	Interview in person/telephone with symptom questionnaire	Flatus I: High joint-mobility scores	P=0.021	Flatus I + FUR: Smoking Ethnicity Antenatal BMI Striae Hernia Varicose veins Haemorrhoids Family history of incontinence or prolapse FUR: High joint-mobility scores			Flatus I + FUR: Augmentation Epidural SVD vs. ID SVD vs. CS ID vs. CS 1 st -3 rd degree tear 1 st stage of labour Active 2 nd stage of labour Passive 2 nd stage of labour Birth weight Head circumference

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Chiarelli 2003	Telephone interview	Precursors: Multiparity	OR=1.8 (1.3-2.6)		FI: UI 12 m >35 y Joint hypermobility Constipation Precursors: UI 12 m Flow-stopping inability Constipation Multiparity	OR=6.0 (2.4-15.3) OR=4.3 (1.6-12.0) OR=2.8 (1.1-7.2) OR=2.5 (1.1-5.5) OR=1.9 (1.2-3.0) OR=2.2 (1.4-3.4) OR=1.9 (1.2-3.0) OR=1.7 (1.0-2.8)	FI + precursors: UI before pregnancy UI during pregnancy UI immediately postpartum Overweight 1 st -2 nd degree tear Marked abdominal striae FD vs. VE FI: Flow stopping inability Multiparity
Damon 2005	Postal questionnaire	UI AS defect + AS defect >60° PP AI	OR=7.8 (1.7-34.6) ^b OR=11.7 (2.5-54.0) ^b OR=50.4 (5.0-508.4) ^b OR=6.3 (1.4-28.6) ^b	Parity BMI FD Baby weight Combined EAS/IAS defect	Anal sphincter defect index delivery	OR=10.5 (2.1-52.4)	Parity BMI PP AI FD Baby weight Precursors: Aged >35 y Joint hypermobility
Eason 2002	Postal questionnaire	FI: Pregnancy UI Perineal massage Birth weight ≥4000 g Episiotomy 2 nd degree tear 3 rd or 4 th degree tear Flatus I FD vs. SVD CS vs. SVD Length of 2 nd stage 0.5-0.9 h Length of 2 nd stage 1.0-1.4 h Episiotomy without extension into anal sphincter	OR=2.5 (1.0-5.8) OR=0.5 (0.1-1.6) OR=2.0 (0.8-4.7) OR=2.2 (1.0-4.6) OR=0.6 (0.2-2.2) OR=2.8 (0.8-9.6) OR=2.7 (2.0-3.6) OR=1.3 (0.4-4.1) OR=0.5 (0.1-2.3) OR=1.3 (0.5-3.2) OR=0.5 (0.1-2.0) OR=1.6 (0.6-4.4)	FI/flatus I: Age BMI Weight gain during pregnancy Parity New UI during pregnancy Epidural anaesthesia 1 st degree tear Natural hair colour Stretch marks FI: VE vs. SVD Length of 2 nd stage ≥1.5 h	AI: FD vs. SVD 3 rd or 4 th degree tear	ARR=1.4 (1.0-2.1) ARR=2.1 (1.4-3.1)	Birth weight VE vs. SVD CS vs. SVD Episiotomy Length of 2 nd stage of labour Perineal massage Episiotomy without extension into anal sphincter

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Eason 2002 continued		Flatus I: Prepregnancy UI VE vs. SVD FD vs. SVD 2 nd degree tear 3 rd or 4 th degree tear Episiotomy without extension into anal sphincter	OR=1.5 (1.2-2.0) OR=1.3 (0.9-1.9) OR=1.6 (1.2-2.2) OR=1.4 (1.0-2.0) OR=2.2 (1.4-3.4) OR=1.3 (0.9-1.9)	Flatus I: Birth weight ≥4000 g Episiotomy CS vs. SVD Perineal massage Length of 2 nd stage of labour			
Faltin 2000	Postal questionnaire	Birth weight FI during pregnancy	P=0.008 P=0.047	Maternal age Height BMI Epidural analgesia Duration of 2 nd stage labour ID vs. SVD Intact perineum Episiotomy FI before pregnancy	Birth weight >3500 g FI during pregnancy 3 rd -4 th degree sphincter tear	OR=6.7 (2.2-20.5) OR=5.9 (1.1-31.0) OR=8.8 (2.9-26.5)	Maternal age Height BMI Epidural analgesia Duration 2 nd stage of labour ID vs. SVD Intact perineum Episiotomy FI before pregnancy
Faltin 2001	Postal questionnaire	3 m: Clinical sphincter tear Endosonographic tear Episiotomy 30 m: Clinical sphincter tear Endosonographic tear	RR=4.3 (1.9-9.7) OR=7.7 (1.6-37.0) ^b P=0.009 RR=3.4 (1.2-9.8) OR=5.7 (1.1-28.5) ^b	3 m + 30 m: Age at first delivery Birth weight Epidural analgesia ID vs. SVD Intact perineum 30 m: Episiotomy 2 nd delivery			
Farrell 2001	Postal questionnaire	6 m flatus I: FD vs. SVD FD vs. CS EM vs. EL ≥3 rd degree tear Weight gain FD vs. SVD Duration of labour More complex repair of perineum	RR=2.5 (1.6-4.0) RR=2.5 (1.4-4.5) P=sign P=0.02 P=0.02 OR=3.2 (1.4-7.5) OR=1.1 (1.0-1.1) P=0.04	6 m flatus I + FI: SVD vs. CS Duration of 2 nd stage Duration of passive stage Duration of active stage Duration of head engaged Birth weight Maternal weight Maternal age Duration of epidural			

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Farrell 2001 continued		6 m FI: Head circumference FD vs. CS	P=0.02 RR=3.6 (1.0-13.4)	BMI Type of perineal repair Failure to perform Kegel's exercises before or during pregnancy 6 m flatus I: EM vs. SVD Head circumference 6 m FI: EM vs. EL FD vs. SVD Labour time Weight gain Episiotomy ≥3 rd degree tear			
Fornell 1996	Written questionnaire	3 rd -4 th degree sphincter rupture	P<0.0001 (flatus I) P<0.05 (liquid stool I) P<0.0001 (solling) P<0.001 (solid stool I)				
Fornell 2005	Postal questionnaire	Flatus I: 3 rd -4 th degree sphincter tear Multiparity	P=0.0001 P=0.03	Solid stool I: 3 rd -4 th degree sphincter tear			
		Liquid stool I: 3 rd -4 th degree sphincter tear	P=0.01	Liquid + solid stool I: Multiparity			3 rd -4 th degree sphincter tear
Frunder 2008	Modified Wexner score						
Groutz 1999	Telephone interview	3 m: 1 st stage of labour Passive 2 nd stage of labour Active 2 nd stage of labour FD vs. SVD ID vs. SVD VE vs. SVD Episiotomy	P=0.001 P=0.01 P=0.04 OR=12.6 (1.0-151.6) OR=8.6 (3.3-22.4) OR=8.4 (3.2-22.3) OR=5.2 (1.5-18.1)	3 m: FD vs. CS ID vs. CS VE vs. CS SVD vs. CS FD vs. VE Gestational age Epidural analgesia Narcotics analgesia Birth weight Apgar score			

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association AI + FI: 2 nd degree laceration	Multivariable Association	Association measure (95% CI)	No association
Leeman 2007	Postal questionnaire						
Liebling 2004	Postal questionnaire						ID vs. CS CS after instrumental vs. immediate CS
MacArthur 1997	Home interview	VE vs. SVD ^a FD vs. SVD ^a EM vs. SVD ^a ID vs. SVD ^a ID vs. CS ^a VE vs. EM ^a VE vs. CS ^a	OR=8.7 (2.6-29.4) OR=2.4 (1.0-5.7) OR=1.7 (1.1-4.4) OR=3.2 (2.1-6.8) OR=2.9 (1.1-7.9) OR=5.1 (1.3-20.3) OR=8.0 (2.0-31.8)	Parity Age ≥ 30 y Induced onset of labour CS vs. SVD FD vs. VE FD vs. EM FD vs. CS Episiotomy 1 st degree tear $\geq 2^{\text{nd}}$ degree Episiotomy and tear Intact perineum 1 st stage of labour ≥ 10 h 2 nd stage of labour ≥ 2 h Active 2 nd stage of labour ≥ 2 h Birth weight ≥ 4000 g Head circumference ≥ 36 cm			
MacArthur 2001	Postal questionnaire				Primiparae FI: FD vs. SVD CS vs. SVD Maternal age 30-34 y Maternal age 35+ Asian ethnic group BMI not known	Primiparae FI flatus I: VE vs. SVD OR= 1.9 (1.3-2.9) OR= 0.6 (0.4-1.0) Breech delivery vs. SVD OR= 1.7 (1.2-2.4) Induced onset of labour OR= 1.8 (1.0-2.9) Intact perineum OR= 3.2 (2.0-5.0) Laceration (1 st -3 rd degree tear) OR=1.5 (1.0-2.2) Episiotomy Duration of 2 nd stage	

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
MacArthur 2001 continued					Primiparae flatus I: FD vs. SVD Maternal age 30-34 y Maternal age 35+ Asian ethnic group	Independent associations remained, but with smaller ORs	Birth weight Maternal age 25-29 y Pre-pregnancy BMI All women FI: Parity Primiparae flatus I: CS vs. SVD BMI not known
MacArthur 2005	Postal questionnaire				Primiparae FI 6 y: FD vs. SVD Maternal age 25-29 y Maternal age 30-34 y Maternal age 35+ Asian ethnic group	OR=2.5 (1.4-4.6) OR=2.6 (1.1-6.4) OR=6.0 (2.4-14.8) OR=6.0 (1.9-19.5) OR=13.5 (6.9-30.8)	Primiparae FI 6 y: VE vs. SVD CS vs. SVD Parity Induced onset of labour Intact perineum Episiotomy Perineal laceration Birth weight Duration of 2 nd stage labour Pre-pregnancy BMI
Mahony 2007	Modified Wexner score	Age 30-34 y Age ≥35 Parity =2 Parity ≥3 ID vs. SVD Ultrasound evidence of partial IAS defect Ultrasound evidence of full thickness IAS defect	OR=1.5 OR=4.6 OR=2.5 OR=3.8 OR=4.2 OR=2.9 OR=7.1		ID vs. SVD IAS defect thickness (partial >1 quadrant)	OR=3.1 (1.2-7.9) OR=5.1 (1.5-22.9)	Age Parity IAS defect thickness (partial < 1 quadrant) EAS defect
Mahony 2008	Modified Wexner score			3 rd -4 th degree tear			
Meyer 2000	Interview with questionnaire			FD vs. SVD			
Nordenstam 2009	Postal questionnaire				3 rd -4 th degree tear 2 nd stage of labour >1 h	RR=2.9 (1.1-7.7) RR=3.2 (1.2-9.2)	Age Subsequent deliveries Post-term pregnancy ID vs. SVD Upright delivery High birth weight/macrosomia

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Tetzschner 1996	Interview at assessment						3 m ² : Head circumference Baby weight Pudendal nerve block Length of 2 nd stage of labour 3 ^{rd-4} th degree tear VE vs. SVD Parity
Tjandra 2008	Wexner score						Age Parity Foetal presentation Birth weight FD vs. SVD VE vs. SVD FD vs. VD ID vs. SVD Episiotomy 3 ^{rd-4} th degree tear Type/extent of defect
Van Brummen 2006	Postal Defecation Distress Inventory	Flatus I: BMI Presence of symptoms at 12 w gestation FI: Birth weight Presence of symptoms at 12 w gestation 3 ^{rd-4} th degree tear	P=0.024 OR=6.3 (3.9-10.0) P=0.043 OR=22.8 (6.9-75.0) OR=15.17 (1.8-127.2)	FI + flatus I: Maternal age 1 ^{st-2} nd degree tear Marital state Educational level Chronic disease Medication use Duration 2 nd stage of labour Alcohol use Foetal presentation Smoking ID vs. SVD ^a VE vs. SVD ^a FD vs. SVD ^a CS vs. SVD ^a EL vs. SVD ^a EM vs. SVD ^a Gestational age	Flatus I: BMI Symptoms at 12 w gestation FI: 3 ^{rd-4} th degree tear	P=0.008 P=0.000 P=0.000	FI + flatus I: Maternal age 1 ^{st-2} nd degree tear Marital state Educational level Chronic disease Medication use Duration of 2 nd stage of labour Alcohol use Foetal presentation Smoking ID vs. SVD VE vs. SVD FD vs. SVD CS vs. SVD EL vs. SVD EM vs. SVD Birth weight Gestational age

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Van Brummen 2006 continued				Fl: BMI at 12 m postpartum			Fl: BMI at 12 m postpartum Presence of symptoms at 12 w gestation
				Flatus I: 3 rd -4 th degree tear Birth weight			Flatus I: 3 rd -4 th degree tear Birth weight
Zetterström 1999	Postal questionnaire	5 m: Maternal age Duration of labour >12 h 2 nd stage of labour >1 h Use of oxytocin Fundal pressure to assist ID vs. SVD 3 rd -4 th degree tear	OR=3.4 (1.6-7.1) OR=2.6 (1.4-5.1) OR=2.7 (1.4-5.0) OR=2.1 (1.02-4.2) OR=2.3 (1.3-4.4) OR=4.9 (2.2-10.8) OR=3.8 (1.8-8.1)	5 m + 9 m: Maternal height Maternal weight - 1 st trimester Maternal weight - at birth Cervical dilatation 1 st stage of labour Gestational age Medical complications during pregnancy Induction of labour DP - kneeling DP - standing DP - sitting DP - lateral	5 m: Maternal age 2 nd stage of labour >1 h ID vs. SVD 3 rd -4 th degree tear 9 m: Maternal age 3 rd -4 th degree tear	OR= 2.7 (1.2-6.0) OR= 2.2 (1.1-4.2) OR= 3.1 (1.3-7.4) OR= 3.0 (1.3-6.7) OR= 3.0 (1.4-6.4) OR= 2.4 (1.04-4.9)	5 m: Duration of labour >12 h Use of oxytocin Fundal pressure to assist delivery 9 m: Lithotomy position
		9 m: Maternal age Lithotomy position 3 rd -4 th degree tear	OR=3.1 (1.5-6.6) OR=3.2 (1.2-8.5) OR=2.4 (1.1-5.2)				

First author PY	Method of outcome assessment	Univariable Association	Association measure (95% CI)	No association	Multivariable Association	Association measure (95% CI)	No association
Zetterström 1999 continued				Presentation of foetus Analgesia Nitrous oxide (N ₂ O) Epidural anaesthesia Paracervical block Pudendal block Morphine Episiotomy Baby weight Baby length Head circumference 5 m: Lithotomy position 9 m: Duration of labour >12 h 2 nd stage of labour >1 h Use of oxytocin Fundal pressure to assist ID vs. SVD			

PY, publication year; CI, confidence interval; UTI, urinary tract infection; FD, forceps delivery; vs., versus; SVD, spontaneous vaginal delivery; VE, vacuum extraction; w, weeks; FI, faecal incontinence; ST, sphincter tear; OR, odds ratio; I, incontinence; m, months; CS, caesarean section; BMI, body mass index; ID, instrumental delivery; STC, sphincter tear cohort; VC, vaginal control group; CC, caesarean control cohort; UI, urinary incontinence; FUR, faecal urgency; y, years; AS, anal sphincter; PP, postpartum; AI, anal incontinence; ARR, adjusted relative risk; h, hours; g, grams; RR, relative risk; EM, emergency caesarean section; EL, elective caesarean section; sign, significant; cm, centimeter; EAS, external anal sphincter; IAS, internal anal sphincter; DP, delivery position.

^aComparisons for mode of delivery are assumed, as author did not respond.

^bCalculated from table (ORs).

^cOnly results of primiparae at index delivery extracted (multiparae follow-up too inconsistent).

^dResults 2–4 y follow-up not extracted because of inconsistent follow-up.

Appendix 3.3 Levels of evidence for aetiological factors for postpartum FI

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Socio-demographics							
Education ^{9,38,41}	Any incontinence	Inconclusive (+/-)	3	0	1	2	0
	FI	Inconclusive (+/-)	3	0	1	2	0
	Flatus I	Weak (-)	1			1	0
Ethnicity ⁴²	Any incontinence	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Inconclusive (+/-)	2	1	0	1	0
	FI	Inconclusive (+/-)	2	1	0	1	0
Asian ethnic group ^{25,26,42}	Any incontinence	Inconclusive (+/-)	2	1	0	1	0
	FI	Inconclusive (+/-)	2	1	0	1	0
	Flatus I	Weak (+)	1	1 (STC) ^b	0		
	Any incontinence	Inconclusive (+/-)	2			1 (VC/CC) ^b	1
	FI	Inconclusive (+/-)	2			1	0
Caucasian ^{9,41}	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Black ⁹	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Hispanic ⁹	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Other ⁹	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Marital status ^{9,38}	Any incontinence	Strong (-)	2			2	0
	FI	Strong (-)	2			2	0
	Urgency	Strong (-)	2			2	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Maternal height ^{26,43}	Any incontinence	Strong (-)	2 (3 FU)			1,1 ^c	0
	FI	Strong (-)	2 (3 FU)			1,1 ^c	0
	Urgency	Strong (-)	2 (3 FU)			1,1 ^c	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Income, US\$ ⁴¹							
Incontinence	Any incontinence	Strong (-)	1			1	0
	FI	Inconclusive (-)	1			1	0
Urinary incontinence (UI) ^{9,14,31,44}							
Before pregnancy	Any incontinence	Inconclusive (+/-)	3	1 (STC) ^b	1	1,1 (VC/CC) ^b	0
	FI	Inconclusive (+/-)	3	1 (STC) ^b	1	1,1 (VC/CC) ^b	0
	Urgency	Inconclusive (+)	1	0			
During pregnancy	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
Immediately after pregnancy	Any incontinence	Inconclusive (+/-)	3	1 (VC) ^b	0	1,1 (STC/CC) ^b	0
	FI	Inconclusive (+/-)	3	1 (VC) ^b	0	1,1 (STC/CC) ^b	0
	Urgency	Inconclusive (-)	1			0	
Flow stopping inability ³¹	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
Flatus I ⁴⁴	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Delivery characteristics							
Maternal age							
At first birth ^{9,21-26,38,41,43,45}	Any incontinence	Inconclusive (+/-)	8	2,1 (STO) ^{31,1} (30-34y, 35+) ³²	0	3,1 (VOC) ^{31,1} (25-29y) ³²	3
	AI	Strong (-)	4 (5 FU)	1	0	2,1 ^c	0
	FI	Inconclusive (+/-)	3	1,1 (STO) ^{31,1} (30-34y, 35+) ³²	0	1,1 (VOC) ^{31,1} (25-29y) ³²	2
	Flatus I	Inconclusive (+/-)	6	1 ^c (30-34y, 35+) ³²	0	1,1 (25-29y) ³²	1
Primiparae and multiparae ^{5,13,31,44,46-48}							
	Any incontinence	Strong (-)	6	1 (FI) ³¹	0	2,1 (prec) ^{31,1}	4
	AI	Weak (-)	1 (2 FU)	1 (>35 y)	0	1 ^c	0
	FI	Inconclusive (+/-)	3	1 (>35 y)	0	1,1 (≥30 y)	1
	Flatus	Inconclusive (-)	1	0	0	0	1
	Precursors	Weak (-)	1	1 (>35 y)	0	1 (>35 y)	0
	Flushing/urgency	Inconclusive (-)	1	0	0	0	1 (≥30 y)
	Wexner score	Weak (-)	1	0	0	1	0
	Mod Wexner score	Inconclusive (-)	1	0	0	0	1
Maternal weight - first trimester²⁴							
Maternal weight - at birth ^{21,24}	AI	Weak (-)	1 (2 FU)	1 ^c	0	1 ^c	1
	Any incontinence	Moderate (-)	2	0	0	0	0
	AI	Weak (-)	1 (2 FU)	1 ^c	0	1 ^c	0
	FI	Inconclusive (-)	1	0	0	0	1
	Flatus I	Inconclusive (-)	1	0	0	0	1
Weight gain^{27,44,48}							
	Any incontinence	Inconclusive (+/-)	3	0	1 (fla) ^b	0	2,1 (FI) ^b
	AI	Weak (-)	3	0	0	0	3
	FI	Inconclusive (+/-)	2	0	1	0	1
Gestational age^{10,23,24,38}							
	Any incontinence	Strong (-)	3	0	0	2	1
	AI	Moderate (-)	2	0	0	1	1
	FI	Weak (-)	1	0	0	1	0
	Flatus I	Weak (-)	1	0	0	1	0
Parity^{5,13,14,22,23,25,26,28,31,34,44,47}							
	Any incontinence	Strong (-)	14	1,1 (prec) ³¹	1 (fla) ³²	6,1 (FI) ³¹	5,1 (liq+solid) ³²
	AI	Strong (-)	6 (7 FU)	1	0	3,1 ^c	2
	FI	Strong (-)	4	0	1	2	1
	Flatus I	Inconclusive (+/-)	1	0	1	0	1
	Liquid stool I	Inconclusive (-)	1	0	0	0	1
	Solid stool I	Inconclusive (-)	1	0	0	0	1
	Precursors	Weak (+)	1	1	0	0	1
	Flushing/urgency	Inconclusive (-)	1	0	0	0	1
	Wexner score	Weak (-)	1	1	0	1	0
	Mod Wexner score	Inconclusive (-)	1	0	0	0	1
Cervical dilation on admission²⁴							
	AI	Weak (-)	1	0	0	1	0
FD vs. SVD^{5,10,14,21,25,26,38,41,44,46,48,49}							
	Any incontinence	Inconclusive (+/-)	11	1	3,1 (fla) ^b	3	3,1 (FI) ^b
	AI	Inconclusive (+/-)	4	0	2	1	1
	FI	Moderate (-)	5	1	0	1	3
	Flatus I	Inconclusive (+/-)	3	1	1	1	0
	Flushing/urgency	Inconclusive (+)	1	0	1	0	0
	Wexner score	Weak (-)	1	1	1	1	0

Aetiological factors ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
VE vs. SVD ^{5,10,25,26,34,38,44,46}	Any incontinence	Strong (-)	9	0	2	3	4
	AI	Inconclusive (+/-)	3	0	1	0	2
	FI	Strong (-)	5	0	1	2	2
	Flatus I	Strong (-)	2	0	1	2	0
	FI/soiling/urgency	Inconclusive (+)	1	0	1	2	0
	Wexner score	Weak (-)	1				
CS vs. SVD ^{5,10,11,21,25,38,41,42,44}	Any incontinence	Strong (-)	10	1 ^{b1, 1b2}	0	1	0
	AI	Inconclusive (-)	2			4, 1 ^{b1, 1b2}	5
	FI	Strong (-)	6	1 ^{b1d}	0	0	2
	Flatus I	Strong (-)	5	1 ^{b2}	0	2, 1 ^{b2}	2
	Urgency	Inconclusive (+/-)	2	1 ^{b2}	0	2, 1 ^{b1}	1
	FI/soiling/urgency	Inconclusive (-)	1			1	0
ID vs. SVD ^{5,8,10,13,22,24,38,41,43,45,46}	Any incontinence	Inconclusive (+/-)	12	1 (FU=1)	3	7, 1 (FU=2)	1
	AI	Inconclusive (+/-)	5 (7 FU)	1 (FU=1) ^b	1	2, 1 ^{c, 1} (FU=2) ^b	0
	FI	Strong (-)	3			2	1
	Flatus I	Strong (-)	2			2	0
	Urgency	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (+)	1	0	1	1	0
Breech delivery vs. SVD ^{25,48}	Wexner score	Weak (-)	1	0	1	1	0
	Mod Wexner score	Inconclusive (+)	1				
	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
EL vs. SVD ³⁸	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Inconclusive (+/-)	3	0	1	1	1
	FI	Weak (-)	1			1	0
	Flatus I	Moderate (-)	2			1	1
EM vs. SVD ^{521,38}	Any incontinence	Inconclusive (+)	1	0	1	1	0
	FI	Weak (-)	1			1	0
	Flatus I	Moderate (-)	1			1	1
	FI/soiling/urgency	Inconclusive (+)	1			1	0
	Any incontinence	Weak (-)	4	0	1	0	3
	AI	Inconclusive (-)	1			0	1
FD vs. CS ^{5,10,21,41}	Any incontinence	Inconclusive (+/-)	2	0	1	0	1
	FI	Inconclusive (+)	1	0	1	0	1
	Flatus I	Inconclusive (+)	1	0	1	0	1
	FI/soiling/urgency	Inconclusive (-)	1			0	1
	Any incontinence	Inconclusive (+/-)	3	0	1	0	2
	AI	Inconclusive (-)	1			0	1
VE vs. CS ^{5,10,41}	Any incontinence	Inconclusive (-)	1			0	1
	FI	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1
	FI/soiling/urgency	Inconclusive (+)	1	0	1	0	1
	Any incontinence	Strong (-)	5	0	1	2	2
	AI	Inconclusive (-)	1			0	1
ID vs. CS ^{5,10,41,42,50}	Any incontinence	Strong (-)	1			0	1
	FI	Moderate (-)	2			1	1
	Flatus I	Strong (-)	2			2	0
	Urgency	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (+)	1	0	1	1	0
	Any incontinence	Inconclusive (+)	1			1	0

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
CS after instrumental vs. immediate CS ⁵⁰	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (-)	1			0	1
EM vs. FD ⁵	FI/soiling/urgency	Inconclusive (+)	1		1 ^d		
EM vs. VE ⁵	FI/soiling/urgency	Inconclusive (+/-)	1				
EM vs. EL ²¹	Any incontinence	Inconclusive (-)	1		1 (fla)	0	1 (FI)
	FI	Inconclusive (-)	1			0	
	Flatus I	Inconclusive (+)	1		1		
	FI/soiling/urgency	Inconclusive (-)	1			1	0
FD vs. VE ^{5,10,31,41,46}	Any incontinence	Strong (-)	5			2	3
	AI	Inconclusive (-)	1			0	1
	FI	Moderate (-)	2			1	1
	Precursors	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (-)	1			1	1
	Wexner score	Weak (-)	1			1	0
	FI/soiling/urgency	Weak (-)	1			1	0
Duration of stage of labour	Any incontinence	Strong (-)	4		1	2	1
	AI	Inconclusive (+/-)	2 (3 FU)		1	1 ^c	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (-)	1			0	1 (≥ 10 h)
	Any incontinence	Inconclusive (+/-)	3		1	0	1
	AI	Inconclusive (+)	1		1	1	1
	FI	Inconclusive (-)	1			0	1
	Flatus I	Moderate (-)	2			1	1
	Urgency	Weak (-)	1			1	1
	Any incontinence	Moderate (-)	4		1	1	0
	AI	Inconclusive (+)	1		1	1	2
	FI	Inconclusive (-)	1			0	1
	Flatus I	Moderate (-)	2			1	1
	Urgency	Weak (-)	1			1	0
	FI/soiling/urgency	Inconclusive (-)	1			0	1 (≥ 2 h)
	Any incontinence	Strong (-)	12		0	6, 1 (FU=2) ^b	5
	AI	Strong (-)	6 (8 FU)		0	1, 1 ^c , 1 (FU=2)(>1h) ^b , 1 (>1h)	1 (≥ 0.5 h), 1
	FI	Strong (-)	5			2, 1 (≥ 1 h)	2
	Flatus I	Strong (-)	3			1, 1 (≥ 1 h)	1
Total duration of labour ^{21,23,24,47}	FI/soiling/urgency	Inconclusive (-)	1			0	1 (≥ 2 h)
	Any incontinence	Inconclusive (+/-)	3		1 (fla) ^b	2	1 (FI) ^b
	AI	Strong (-)	2 (3 FU)			0	0
	FI	Inconclusive (-)	1			1, 1 (>12 h)	1
Duration of head engaged ²¹	Flatus I	Inconclusive (+)	1		1	0	1
	Any incontinence	Inconclusive (-)	1			0	1
	FI	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Sphincter rupture ^b Intact perineum 5,25,26,43,45,46	Any incontinence	Strong (-)	5			4	1
	AI	Strong (-)	2 (3 FU)			1,1 ^c	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Fl/soiling/urgency	Inconclusive (-)	1			0	1
1 st degree 5,44	Wexner score	Weak (-)	1			1	0
	Any incontinence	Inconclusive (-)	2			0	2
	AI	Inconclusive (-)	1			0	1
	Fl/soiling/urgency	Inconclusive (-)	1			0	1
	Any incontinence	Inconclusive (-)	2			0	2
2 nd degree 44	AI	Inconclusive (-)	1			0	1
Sutured 51	Any incontinence	Weak (+)	1			1	0
	AI	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
	AI	Weak (-)	1			1	0
Unsutured 51	FI	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
	AI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Fl/soiling/urgency	Weak (-)	1			1	0
≥2 nd degree 5	Any incontinence	Inconclusive (-)	1			0	1
1 st –2 nd degree 3,1,38	Strong (-)	Strong (-)	2			2	0
	Strong (-)	Strong (-)	2			2	0
	Weak (-)	Weak (-)	1			1	0
	Weak (-)	Weak (-)	1			1	0
	Precursors	Weak (-)	1			1	0
1 st –3 rd degree 25,26,41,42	Any incontinence	Strong (-)	3			2	1
	FI	Moderate (-)	2			1	1
	Flatus I	Strong (-)	2			2	0
	Urgency	Strong (-)	1			1	0
	Any incontinence	Weak (-)	2			1	0
1 st –4 th degree 47,48	AI	Moderate (-)	1 (2 FU)			1	0
	FI	Weak (-)	1			1 ^c	0
	Any incontinence	Weak (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	0
	Urgency	Inconclusive (-)	1			0	1
3 rd degree/4 th degree 11,13,14,21–24,27,28,34,38,43–46,52,53	Any incontinence	Strong (+)	12	5,1 (F) ^{p1}	2,1 (fla) ^{p2}	3,1 (fla) ^{p1}	2,1 (F) ^{p2}
	AI	Strong (+)	6 (7 FU)	3,1 ^c	1	0	1
	FI	Inconclusive (+/-)	3	2	0	0	1
	Flatus I	Inconclusive (+/-)	4	1	2	1	0
	Liquid stool	Moderate (+)	1	0	1	0	0
	Solid stool	Inconclusive (+)	2	0	1	0	1
	Urgency	Inconclusive (+/-)	1	1	0	0	0
	Soiling	Weak (+)	1	0	1	1	0
	Wexner score	Inconclusive (+)	1				
	Mod Wexner score	Weak (-)	2			1	0
4 th degree vs. 3 rd degree 9	FI	Strong (-)	1	1 (STC)	0	2	0
	Wexner score	Weak (+)	1 (2 FU)	1 ^c	0		
	Defect score 54	Weak (+)	1 (2 FU)				

Aetiological factors ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Type of sphincter defect							
IAS ^{13,46}	Any incontinence	Inconclusive (+/-)	2	0	1	1	0
	Wexner score	Weak (-)	1			1	0
	Mod Wexner score	Inconclusive (+)	1		1		
	Any incontinence	Moderate (-)	2			1	1
	Wexner score	Weak (-)	1			1	0
EAS ^{13,46}	Any incontinence	Inconclusive (-)	1			0	1
	Mod Wexner score	Strong (-)	2			2	0
	Any incontinence	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0
Combined ^{14,46}							
Extent of sphincter defect	Any incontinence	Weak (-)	1			1	0
	Mod Wexner score	Weak (-)	1			0	1
	Wexner score	Inconclusive (+)	1	0	1	1	0
	Mod Wexner score	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0
Epistomy ^{5,3,10,21,23,26,41,43,48}	Any incontinence	Weak (+)	1	1	0		
	AI	Strong (-)	13	0	1	7	5
	AI	Inconclusive (+/-)	6 (8 FU)	0	1,1 (FU=1) ^b	2,1,1 (FU=2) ^b	1
	Flatus I	Strong (-)	5			2	3
	Flatus I	Weak (-)	1			1	0
Epistomy and tear ⁵	Any incontinence	Inconclusive (-)	1			0	1
	AI	Weak (-)	1			1	0
	Flatus I	Inconclusive (-)	1			0	1
	Flatus I	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0
Epistomy without extension into anal sphincter ⁴⁴	Any incontinence	Inconclusive (-)	1			0	1
	AI	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (+/-)	1	0	1	0	1
	Flatus I	Inconclusive (-)	1			0	1
	Wexner score	Weak (-)	1			1	0
Foetal presentation at delivery ^{24,38,46}	Any incontinence	Strong (-)	3			3	0
	AI	Weak (-)	1 (2 FU)			1 ^c	0
	Flatus I	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0
External fundal pressure ^{23,24}	Any incontinence	Strong (-)	2 (3 FU)			1,1 ^c	0
	AI	Strong (-)	17			9	7
	AI	Strong (-)	8 (10 FU)	1 (>3500 g)	0	2,2 ^c	2,1 (≥4000 g)
	Flatus I	Strong (-)	6			3	3
	Flatus I	Strong (-)	4			3	1
Birth weight ^{5,10,11,14,21,26,34,38,41,48}	Any incontinence	Weak (-)	1			1	0
	AI	Inconclusive (-)	1			0	1
	Flatus I	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0
Baby length ²⁴	Any incontinence	Weak (-)	1 (2 FU)			1 ^c	0
	AI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Wexner score	Weak (-)	1			1	0

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Head circumference 5,9,21,23,24,31,34,41,42	Any incontinence	Strong (-)	8	0	1 (F) ^b	4	3,1 (fla) ^b
	AI	Moderate (-)	2			1	1
	FI	Strong (-)	4	0	1	2	1
	Flatus I	Moderate (-)	2			1	1
	Urgency	Weak (-)	1			1	0
	Flailing/urgency	Inconclusive (-)	1			0	1 (≥36 cm)
	Precursors	Inconclusive (-)	1			1	0
Induced onset of labour ^{5,24,26}	Any incontinence	Strong (-)	3			2	1
	AI	Weak (-)	1 (2 FU)			1 ^c	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Flailing/urgency	Inconclusive (-)	1			0	1
Oxytocin stimulation ^{9,23,24,42}	Any incontinence	Strong (-)	3			3	0
	AI	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
Pudendal block ^{23,24,34}	AI	Moderate (-)	2			1	1
Paracervical block ^{23,24}	AI	Weak (-)	1			1	0
Anaesthesia	Any incontinence	Strong (-)	8			5	3
Epidural ^{10,23,24,42-45,47,48}	AI	Inconclusive (-)	5 (7 FU)			2 ^c	1
	FI	Moderate (-)	2			0	2
	Flatus I	Weak (-)	1			1	1
	Urgency	Weak (-)	1 (2 FU)			1 ^c	0
Nitrous oxide ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0
Analgesia	Any incontinence	Strong (-)	2			2	0
General ^{9,24}	AI	Weak (-)	1 (2 FU)			1 ^c	0
	FI	Weak (-)	1			1	0
	AI	Inconclusive (-)	1			0	1
Narcotics ¹⁰	AI	Inconclusive (-)	1 (2 FU)			1 ^c	0
Morphine ²⁴	AI	Weak (-)	1 (2 FU)			0	1
Duration of epidural ²¹	Any incontinence	Inconclusive (-)	1			0	1
	FI	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1
Early postpartum FI ^{14,23,34}	AI	Inconclusive (+/-)	3 (4 FU)	1 ^c	1	1	0
AI during pregnancy ^{38,43}	Any incontinence	Inconclusive (+/-)	2	1,1 (fla) ^b	0	1 (F) ^b	0
	AI	Weak (+)	1	1	0	1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (+)	1	1	0	1	0
AI before pregnancy ⁴³	AI	Weak (-)	1			1	0
Upright birth position ^{22,23}	AI	Weak (-)	1			1	0
Delivery position-kneeling ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0
Delivery position-standing ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0
Delivery position-sitting ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0
Delivery position-lateral ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Lithotomy position ²⁴	AI	Weak (-)	1 (2FU)			1 ^c	0
Marked abdominal striae ^{3,142}	Any incontinence	Strong (-)	2			2	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Precursors	Weak (-)	1			1	0
Perineal massage ⁴⁴	AI	Inconclusive (-)	1			0	1
Medical complications during Pregnancy ²⁴	AI	Weak (-)	1 (2 FU)			1 ^c	0
Shoulder dystocia ⁹	FI	Weak (-)	1			1	0
Apgar score ¹⁰	AI	Inconclusive (-)	1			0	1
Position of foetal head ⁴¹	FI	Inconclusive (-)	1			0	1
General factors							
Medication use ³⁵	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Inconclusive (+/-)	11	1,1 (Q NR) ^{31,1} , 1 (fla) ³²	0	5,1 (Q) ^{31,1} , 1 (F) ³²	3
	AI	Strong (-)	2			2	0
Obesity/BMI ^{3,14,21,25,26,31,38,41,44}	FI	Inconclusive (+/-)	7	1 (STO) ^{33,1} (Q NR) ³¹	0	1,1 (Q) ^{31,1} , 1 ^{32,1} (V/C/C) ³³	3
	Flatus I	Strong (-)	5	1 ³²	0	1,1 (Q) ^{31,1} , 1 (Q NR) ³¹	2
	Urgency	Weak (-)	1			1	0
	Precursors	Weak (-)	1			1	0
	Any incontinence	Weak (+)	1	1	0	1	0
Constipation ³¹	FI	Weak (+)	1	1	0	1	0
	Precursors	Weak (+)	1	1	0	1	0
	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
Smoking ^{35,42}	Any incontinence	Strong (-)	2			2	0
	FI	Weak (-)	1			1	0
	Flatus I	Strong (-)	2			2	0
	Urgency	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
Alcohol use ³⁵	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
	FI	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
Joint hypermobility ^{31,42}	Any incontinence	Inconclusive (+/-)	2	1, 1 (F) ³¹	0	1 (prec) ³¹	0
	FI	Weak (+)	1	1	0		
	Flatus I	Weak (+)	1	1	0		
	Urgency	Weak (+)	1	1	0		
	Precursors	Weak (-)	1			1	0
Hernia ⁴²	Any incontinence	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
Varicose veins ⁴²	Urgency	Weak (-)	1			1	0
	Any incontinence	Weak (-)	1			1	0

Aetiological factor ^a	Outcome	Level of evidence (+/-)	Cohorts assessed	Significant association QS > 60%	Significant association QS ≤ 60%	No association QS > 60%	No association QS ≤ 60%
Family history of incontinence /prolapse ⁴²	Any incontinence	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
Haemorrhoids ⁴²	Any incontinence	Weak (-)	1			1	0
	Flatus I	Weak (-)	1			1	0
	Urgency	Weak (-)	1			1	0
Failure to perform Kegel exercises before or during pregnancy ²¹	Any incontinence	Inconclusive (-)	1			0	1
	Fl	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1
Natural hair colour ⁴⁴	Any incontinence	Inconclusive (-)	1			0	1
	Fl	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1
Stretch marker ⁴⁴	Any incontinence	Inconclusive (-)	1			0	1
	Fl	Inconclusive (-)	1			0	1
	Flatus I	Inconclusive (-)	1			0	1
Post-term pregnancy ²²	AI	Weak (-)	1			1	0
Postpartum urinary tract infection ⁴⁵	Fl	Inconclusive (-)	1			0	1

^a, presence of association: -, no association; QS, quality score; Fl, faecal incontinence; I, incontinence; STC, sphincter tear cohort; VC, vaginal control group; CC, caesarean control cohort; AI, anal incontinence; FU, follow-up; UI, urinary incontinence; m, months; y, years; prec, precursors (flatus I, soiling and urgency); fla, flatus incontinence; liq, liquid stool incontinence; solid, solid stool incontinence; FD, forceps delivery; vs., versus; SVD, spontaneous vaginal delivery; VE, vacuum extraction; CS, caesarean section; ID, instrumental delivery; EL, elective caesarean section; EM, emergency caesarean section; h, hours; cm, centimeter; Mod, modified; STC, sphincter tear cohort; IAS, internal anal sphincter; EAS, external anal sphincter; g, grams; BMI, body mass index; Q (NR), quartiles (no response category).

^bMultivariable analyses results are presented. If not available, univariable results are presented.

^cCorresponding studies within one aetiological factor.

^dIncludes two follow-up moments.

^eProtective aetiological factor.

^fFirst-degree tear: a perineal laceration extending through the vaginal mucosa and perineal skin only, second-degree tear: laceration extending into the perineal muscles, third-degree tear: laceration involving the external anal sphincter, fourth-degree tear: laceration affecting both the anal sphincter and the anorectal mucosa.

Appendix 3.4 Change in level of evidence after removal of studies with low methodological quality or only univariable results

Aetiological factor	Outcome	Removal of studies with low methodological quality	Removal of studies with only univariable results
Education	FI	Inconclusive (+/-) → strong (-)	-
Income, US\$	FI	a	-
UI before	Flatus I	a	-
UI during	Flatus I	a	-
Flatus I	FI	a	-
Maternal age primiparous and multiparous	FI/soiling/urgency	a	-
	Mod Wexner score	a	-
	Flatus I	a	-
	FI	Inconclusive (+/-) → weak (+)	-
	Any	Strong (-) → inconclusive (+/-)	-
Maternal weight at birth	Flatus I	a	-
	FI	a	-
	Any	Moderate (-) → weak (-) ^b	-
Weight gain	Flatus I	a	-
	FI	a	-
	Any	a	-
Gestational age	AI	Moderate (-) → weak (-) ^b	-
	Flatus I	a	-
	Liquid stool	a	-
	Solid stool	a	-
	FI/soiling/urgency	a	-
	AI	Strong (-) → inconclusive (+/-)	-
	Any	Inconclusive (+/-) → weak (-)	-
FD vs. SVD	AI	Inconclusive (+/-) → weak (-)	Inconclusive (+/-) → inconclusive (+/-)
	FI	Moderate (-) → inconclusive (+/-)	-
	Any	Inconclusive (+/-) → strong (-)	Inconclusive (+/-) → inconclusive (+/-)
VE vs. SVD	AI	a	-
	FI	a	-
	FI/soiling/urgency	a	-
CS vs. SVD	AI	a	Inconclusive (-) → inconclusive (-)
	FI/soiling/urgency	a	-
	Any	Strong (-) → inconclusive (+/-)	-
TD vs. SVD	AI	Inconclusive (+/-) → strong (-)	-
	FI/soiling/urgency	a	-
	Mod Wexner score	a	Inconclusive (+) → inconclusive (+)
EM vs. SVD	Any	Inconclusive (+/-) → strong (-)	-
	Flatus I	Moderate (-) → weak (-) ^b	-
	FI/soiling/urgency	a	-
FD vs. CS	Any	Inconclusive (+/-) → weak (-)	-
	AI	a	-
	FI	a	-
	Flatus I	a	-
	FI/soiling/urgency	a	-
	Any	a	-

Aetiological factor	Outcome	Removal of studies with low methodological quality	Removal of studies with only univariable results
VE vs. CS	AI	a	-
	FI/soiling/urgency	a	-
	Any	a	-
ID vs. CS	AI	a	-
	FI	Moderate (-) → weak (-)	-
	FI/soiling/urgency	a	-
EM vs. FD	FI/soiling/urgency	a	-
EM vs. VE	FI/soiling/urgency	a	-
EM vs. EL	FI	a	-
	Flatus I	a	-
	Any	a	-
FD vs. VE	AI	a	-
	FI	Moderate (-) → weak (-)	-
	FI/soiling/urgency	a	-
Duration 1 st stage of labour	AI	Inconclusive (+/-) → weak (-) ^b	-
Duration 2 nd stage-passive	FI/soiling/urgency	a	-
	AI	a	-
	FI	a	-
Duration 2 nd stage-active	Flatus I	Moderate (-) → weak (-) ^b	-
	Any	Inconclusive (+/-) → weak (-)	-
	AI	a	-
	FI	a	-
	Flatus I	Moderate (-) → weak (-) ^b	-
	FI/soiling/urgency	a	-
Duration 2 nd stage	Any	Moderate (-) → weak (-)	-
	FI/soiling/urgency	a	-
	FI	a	-
Duration of head engaged	Flatus	a	-
	Any	Inconclusive (+/-) → strong (-)	-
	FI	a	-
	Flatus I	a	-
Intact perineum	Any	a	-
	AI	Strong (-) → strong (-)	Strong (-) → weak (-) ^b
	FI/soiling/urgency	a	-
1 st degree	AI	a	-
2 nd degree	FI/soiling/urgency	a	-
	AI	Inconclusive (-) → inconclusive (-)	-
	FI	a	-
2 nd degree sutured	AI	Weak (-) → weak (-)	*
	FI	Weak (-) → weak (-)	*
	AI	Weak (-) → weak (-)	*
2 nd degree unsutured	AI	Weak (-) → weak (-)	*
	FI	Weak (-) → weak (-)	*
	AI	Weak (-) → weak (-)	*
≥2 nd degree	FI/soiling/urgency	a	-
1 st -3 rd degree	FI	Moderate (-) → weak (-)	-
1 st -4 th degree	FI	a	-

Aetiological factor	Outcome	Removal of studies with low methodological quality	Removal of studies with only univariable results
3 rd /4 th degree	FI	Inconclusive (+/-) → strong (+)	-
	Flatus I	Moderate (+) → inconclusive (+/-)	-
	Solid I	a	-
	Liquid I	a	-
	Soling	a	-
	Mod Wexner score	Strong (-) → strong (-)	Strong (-) → weak (-) ^b
TIAS defect	Any incontinence	Inconclusive (+/-) → weak (-)	Inconclusive (+/-) → inconclusive (+/-)
	Mod Wexner score	a	Inconclusive (+) → inconclusive (+)
EAS defect	Any incontinence	Moderate (-) → weak (-) ^b	Moderate (-) → moderate (-)
	Mod Wexner score	a	Inconclusive (-) → inconclusive (-)
Episiotomy	AI	Inconclusive (+/-) → strong (-)	-
	FI/soling/urgency	a	-
Episiotomy and tear	FI/soling/urgency	a	-
Episiotomy without extension into anal sphincter	AI	a	-
More complex repair of perineum	FI	a	-
	Flatus I	a	-
	Any	a	-
Birth weight	FI/soling/urgency	a	-
Head circumference	AI	Moderate (-) → weak (-) ^b	-
	Flatus I	Moderate (-) → weak (-) ^b	-
	FI/soling/urgency	a	-
Induced onset of labour	FI/soling/urgency	a	-
Epidural anaesthesia	FI	a	-
	Flatus I	Moderate (-) → weak (-) ^b	-
	AI	Moderate (-) → weak (-) ^b	-
Pudendal block	AI	a	-
Narcotics	AI	a	-
Duration of epidural	Any	a	-
	FI	a	-
	Flatus I	a	-
Perineal massage	AI	a	-
	AI	a	-
Appar score	FI	a	-
Position of foetal head	Flatus I	Strong (-) → inconclusive (+/-)	-
Obesity	Any	a	-
Failure Kegel exercises	FI	a	-
	Flatus I	a	-
Natural hair colour	Any	a	-
	FI	a	-
	Flatus I	a	-
Stretch markers	Any	a	-
	FI	a	-
	Flatus I	a	-
Postpartum UTI	FI	a	-

FI, faecal incontinence; →, changes into; UI, urinary incontinence; I, incontinence; -, unchanged compared to first sensitivity analysis; Mod, modified; FD, forceps delivery; SVD, spontaneous vaginal delivery; VL, vacuum extraction; CS, caesarean section; ID, instrumental delivery; EL, emergency caesarean section; EI, elective caesarean section; IAS, internal anal sphincter; EAS, external anal sphincter; UTI, urinary tract infection.

^aNo cohort study left for this aetiological factor and outcome. ^bChange from two to one cohort per outcome.

CHAPTER 4

INCONCLUSIVE PSYCHOMETRIC PROPERTIES OF THE VAIZEY SCORE IN FAECALLY INCONTINENT PATIENTS: A PROSPECTIVE COHORT STUDY

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Published in: Neurourology and Urodynamics 2010;29(3):370-7

ABSTRACT

Aim

To determine the psychometric properties of the total Vaizey score and its individual items.

Methods

The study was conducted as part of a prospective cohort study assessing the outcome of pelvic floor rehabilitation in patients with faecal incontinence.

Results

One hundred ninety-four patients were analysed, 53 of whom provided data on the global perceived effect (GPE) score. Pelvic floor rehabilitation resulted in a significant reduction of the total Vaizey score and most individual items. The total Vaizey score changed in agreement with the GPE scores. The total Vaizey score was responsive, but some individual items yielded inconsistent results for three different measures. The test-retest reliability was adequate or excellent for six individual items and the total Vaizey score. The internal consistency was low for the total Vaizey score at baseline, in contrast to the follow-up and change scores. The estimates for the minimally important change (MIC) and smallest detectable change yielded moderately consistent results. An MIC of -5 points seemed preferable and yielded the lowest misclassification rate.

Conclusions

More research is required to confirm conclusions on the psychometric properties of the total Vaizey score and its individual items, and to justify its use in research and routine clinical practice.

INTRODUCTION

Faecal incontinence (FI) is a frequently unrecognised and underestimated health condition, with an estimated prevalence of 2–24%. Prevalence rates vary depending on the population and on the definition of FI that is used.¹ FI can be defined as the recurrent involuntary excretion of faeces in inappropriate places or at inappropriate times.² FI covers a wide spectrum from involuntary but recognised passage of gas, liquid, or solid stool (urge incontinence) to unrecognised anal leakage of mucus, fluid, or stool (passive incontinence). Temporary or permanent FI affects activities of daily life and has been recognised as having a psychosocial impact.

Several severity scores have been developed to capture the consequences of FI. The Vaizey score is a widely used severity score and is reported to be a reproducible score, which correlates highly with physicians' clinical impression.^{3,4} It has also been demonstrated that higher Vaizey scores are associated with more self-reported problems in general health domains⁴ and that changes in Vaizey score reflect patients' subjective perception of relief.^{5,6}

Although the Vaizey score is not a frequently used outcome measure in physiotherapy trials, it has recently been used as evaluation measure in two studies assessing the outcome of a pelvic floor rehabilitation (PFR) program in patients with FI.^{6,7} However, changes in the Vaizey score are difficult to interpret and there is a lack of data to determine the minimally important change (MIC) for use in research and routine clinical practice. Determining the MIC in physiotherapy trials would facilitate future sample size calculations and help define successful outcome. To date, the Vaizey score has not been thoroughly evaluated in terms of several psychometric properties. Therefore, we made an attempt to assess the test-retest reliability, internal consistency and responsiveness of the Vaizey score, and to define the MIC and the smallest detectable change (SDC).

MATERIALS AND METHODS

Study design and subjects

The study was conducted as part of a large diagnostic cohort study whose methods have been described in detail elsewhere.⁷ Between December 2001 and April 2004, consenting eligible patients with FI visiting one of 16 participating Dutch hospitals were consecutively included. For the purpose of this study, patients were only included when complete baseline and follow-up (FU) Vaizey scores and complete results of a stool and drinking diary were available. Patients received a standardised PFR program with weekly sessions of at least 35 minutes' duration for nine weeks. PFR consisted of rectal balloon training and either electrical stimulation or EMG-biofeedback, depending on the Oxford score of the external anal sphincter and puborectalis muscle preceding treatment.⁸

The Vaizey score was used to evaluate the PFR program and reflected the severity of FI, ranging from 0 (complete continence) to 24 (complete incontinence) (Table 4.1). Assessments took place at baseline and approximately three months after PFR. The global perceived effect (GPE) score was obtained by asking 79 patients who had recently completed PFR to rate their present situation relative to their level of incontinence before treatment.⁵ The GPE score consisted of the following scale: (1) much better, (2) better, (3) no change, (4) worse, and (5) much worse. Although the GPE score is not a ‘gold standard’, it has been advocated as an alternative offering a reasonable estimate of true change. A sample size of at least 50 patients was considered to be sufficient for the assessment of floor or ceiling effects, test-retest reliability, the MIC and the SDC. However, for the assessment of the internal consistency a sample size of at least 100 patients is required and the required sample size for the assessment of the responsiveness is unclear.⁹

Table 4.1 Faecal incontinence score according to Vaizey et al.

Incontinent	Never	Rarely	Sometimes	Weekly	Daily
Solid stool	0	1	2	3	4
Liquid stool	0	1	2	3	4
Gas	0	1	2	3	4
Alteration in lifestyle	0	1	2	3	4
				No	Yes
Need to wear a pad or plug				0	2
Taking constipating medication				0	2
Lack of ability to defer defecation for 15 minutes				0	4

Minimum score=0=perfect continence; maximum score=24=totally incontinent.
Never: no episodes in the past four weeks; rarely: 1 episode in the past four weeks; sometimes: >1 episode in the past four weeks, but <1 a week; weekly: 1 or more episodes a week but <1 a day; daily: 1 or more episodes a day.

Psychometric properties and statistical analyses

Vaizey score distributional characteristics

Demographic and clinical characteristics of all participants were determined at baseline. The Vaizey change scores of participants who completed the diary were compared with those of non-participants, to check for selection bias. Additionally, participants who completed the GPE score were compared for the same characteristic with participants who did not complete the score.

Vaizey scores at baseline and at three months FU were calculated for each Vaizey item and the total Vaizey score. Four individual items were measured on a five-point scale, ranging from 0 to 4, corresponding to never, rarely, sometimes, weekly and daily. The individual items of ‘urge’ and ‘pad use’ had five frequencies, but only two values (never=0 (no); rarely to daily=4 (yes) for urge; rarely to daily=2 (yes) for pad use (Table 4.1). Medication use was rated on a dichotomous scale (yes/no). The marginal homogeneity test, the McNemar test, and the Wilcoxon signed rank test were used to assess significant changes in total Vaizey score and the individual items.

We explored differences in Vaizey change scores for three subtypes of FI: passive incontinence, urge incontinence and mixed incontinence with a frequency of at least once per four weeks. Passive incontinence was defined as defecation without the patient being aware of it. Urge incontinence was defined as the inability to defer defecation for more than 15 minutes.⁷

Ceiling and floor effects

Ceiling and floor effects were considered to be present if $\geq 15\%$ of responders had the highest and lowest possible scores, respectively, on an individual item and the total Vaizey score.⁹ The presence of such effects may affect content validity, reliability, and responsiveness.

Test-retest reliability and internal consistency

The test-retest reliability was assessed using the sample of stable patients who rated themselves as 'unchanged' on the GPE score. The weighted kappa and marginal homogeneity test were used to assess the test-retest reliability. Values for weighted Cohen's kappa coefficients (identical to the ICC_{agreement}) were rated as: poor (≤ 0.40), adequate (0.41–0.74) and excellent (≥ 0.75). The internal consistency was evaluated by computing Cronbach's alpha, corrected item-total, and Cronbach's alpha if item were deleted, for the baseline, FU and Vaizey change scores. The internal consistency was rated positive when Cronbach's alpha was > 0.7 (adequate) and ≥ 0.8 (excellent), when the 'corrected item-total' was in between 0.30 and 0.90, and when 'Cronbach's alpha if item were deleted' did not exceed the overall Cronbach's alpha. A positive rating indicates that all items evaluate different aspects of the same construct.

Responsiveness

Responsiveness refers to the ability of an outcome measure to detect clinically important changes over time, even if these changes are small. The mean Vaizey change was calculated for each GPE category. The Wilcoxon signed rank test, the effect size (ES) and the standardised response mean (SRM) were determined to evaluate the significance of change and to determine the magnitude of the instrument's responsiveness. The greater an effect size, the greater an instrument's responsiveness. The effect sizes were considered moderate when the absolute values were > 0.50 and large when > 0.80 . The same analysis was done for the total sample, for each individual item as well as for the total Vaizey score.

The Spearman correlation coefficient was calculated to assess the correlation between the changes in the total and individual Vaizey item scores and the GPE score. Spearman's correlations were considered excellent with absolute values ≥ 0.60 , adequate with absolute values between 0.31 and 0.59 and poor with absolute values ≤ 0.30 .

Minimally important change

The MIC in the Vaizey score was visualised according to the anchor-based MIC distribution. This method combines the advantages of the anchor-based and distribution-based approaches, and visualises the total amount of misclassification.¹⁰ The anchor-based approach uses an external criterion to determine the MIC, while the distribution-based approach includes a measure of variability.

Three methods were used to estimate the MIC:

1. An anchor-based method proposed by Jaeschke et al.¹¹ This approach uses the GPE score as an external criterion (anchor). The method calculates the mean change in Vaizey scores of patients who rated themselves as 'better' minus the mean score of the patients who were 'unchanged'. The category of patients who rated themselves as 'better' on the GPE score was defined as showing a minimally important improvement, thereby separating the successfully recovered (better, much better) from the unsuccessfully recovered patients (no change, worse and much worse) on the anchor.
2. Another anchor-based method was based on the Receiver Operator Curve (ROC) analysis, identifying the exact cut-off values for the Vaizey change scores which yield the smallest total proportion of misclassification.¹² This means that the proportion of patients correctly classified as showing important improvement both in terms of the GPE score and the Vaizey change score should be as high as possible (sensitivity), as should the proportion of patients who are correctly classified as not showing important improvement (specificity). We assumed that false-negative results were just as undesirable as false-positive results. The area under the curve (AUC) was considered excellent with values ≥ 0.90 , adequate with values between 0.70–0.89 and poor with values < 0.70 .
3. The 95% limit cut-off point was based on the distributional characteristics of patients who are 'unchanged' according to the anchor ($\text{mean}_{\text{change}} + 1.645 \times \text{SD}_{\text{change}}$). In addition to the 95% limit cut-off point, the variability of the Vaizey change score was expressed as the standard error of measurement (SEM). The SEM was calculated as $\text{SD}_{\text{baseline}} \times [\sqrt{1-r}]$. The test-retest reliability was used as a reliability parameter. Finally, since the MIC should be detectable beyond the measurement error, we calculated the smallest detectable change (SDC). The SDC shows which Vaizey changes fall outside the measurement error, and was calculated as $1.645 \times \sqrt{2} \times \text{SEM}$.

Statistical analyses were performed with SPSS version 15.0 (SPSS®, Inc., Chicago, IL). A two-tailed P-value of 0.05 was considered to indicate statistical significance.

RESULTS

Participants

A total of 264 patients completed the PFR program, 66 of whom completed the GPE score. We identified 194 patients with complete diary and Vaizey score data; interview data (GPE) were available for 53 of them.

The mean Vaizey change score of non-participants ($n=45$) did not differ from that of participants ($n=194$) who completed the diary ($P=0.219$), nor did it differ between participants who completed the GPE score ($n=53$) and participants ($n=141$) who did not do so ($P=0.561$).

The mean age of the 194 patients was 59 years ($SD=12.1$), in a predominantly female population (90.2%). The majority had experienced symptoms for more than five years. The nature of incontinence was mainly mixed FI, with only five patients having passive FI. In many cases, no specific origin of the FI symptoms could be assigned. Baseline demographic data and clinical characteristics are presented in Table 4.2.

Table 4.2 Baseline demographic and clinical characteristics: % (No.) unless otherwise stated

	Total (n=194)	
	%	n
Age (years) (mean; SD)	59.0 (12.1)	
<45	12.9	25
45-54	18.5	36
>54	68.6	133
Sex		
Female	90.2	175
Male	9.8	19
Onset of FI symptoms (month) (mean; SD)	104.5 (112.4)	
<1	7.3	14
1-5	42.7	83
>5	48.5	94
Unknown	1.5	3
Parity (mean; SD)	2.4 (1.2)	
0	5.2	10
1	10.8	21
2	37.6	73
≥3	35.5	69
Vaizey score (mean; SD)		
Baseline	18.2 (2.8)	
After PFR	15.2 (5.0)	
Average change	-3.0 (4.9)	
Nature of incontinence		
Passive FI	2.6	5
Urge FI	37.1	72
Mixed FI	56.2	109
None	0.5	1
Missing	3.6	7
Origin		
Surgery	18.0	35
Accident	0.5	1
Delivery	19.6	38
Peri-menopausal	2.1	4
Psychological trauma	0.5	1
Other	6.7	13
Unclear	52.6	102
Previous gynaecological surgery		
Hysterectomy	39.4	69
Current constipating medication (yes)	28.4	55

Table 4.2 continued

	Total (n=194)	
	%	n
Postmenopausal (yes)	68.0	132
Co-morbid conditions (yes)	27.8	54
Cardiovascular problems	17.0	33
Diabetes mellitus	7.2	14
Lung problems	6.2	12
COPD	3.1	6
Asthma	2.1	4
Other	1.0	2

SD, standard deviation; PFR, pelvic floor rehabilitation.

Vaizey score distributional characteristics

The Vaizey distributional characteristics are presented in Table 4.3. The total Vaizey score fell from 18.20 (SD=2.78) at baseline to 15.19 (SD=5.04) at FU. Apart from pad and medication use, all individual items and the total Vaizey score showed a significant reduction.

Although no floor or ceiling effects were observed for the total Vaizey score (Table 4.3), all individual items showed a strong tendency to cluster in the worst frequency category (ranging from never to daily). Floor effects were limited to the individual items of ‘pad use’, ‘solid FI’ and ‘medication use’. However, medication use has only two response categories, whereas the remaining individual items have five, and the total baseline Vaizey score ranges from 12 to 24. The more response categories, the lower the probability of ceiling and floor effects.

Separation of Vaizey score characteristics in terms of type of incontinence (passive FI, urge FI and mixed FI) showed that most patients experienced mixed FI and only five had passive FI (Table 4.2). The greatest changes in the mean of the individual items and total Vaizey scores were observed for urge FI, except for the individual Vaizey items of ‘solid FI’ and ‘adjustment’. These items showed higher values for passive FI, although these estimates were less reliable due to the low number of patients.

Test-retest reliability

The weighted kappa assessment of the test-retest reliability of the total Vaizey score was performed on the unchanged group and was adequate with 0.55 (Table 4.4). Most subscale items also had adequate estimates, except for urge (poor) and medication (excellent). In view of the range of total Vaizey scores, which varied from 12 to 24 at baseline and from 0 to 24 at FU, the test-retest reliability can be interpreted as adequate, which was supported by the non-significant P-values of the marginal homogeneity test.

Table 4.3 Distributional characteristics, variability and comparison of Vaizey scores between baseline (T0) and follow-up (T1)

Vaizey item	Baseline mean (SD) (n=194)	Median (min-max)	Ceiling ^a %	Floor ^b %	FU		T1-T0 mean change (SD)	P-value marginal homogeneity test
					mean (SD) (n=194)	Median (min-max)		
Urge	4.30 (1.07)	5 (1-5)	59.8	3.1	3.69 (1.47)	4 (1-5)	-0.61 (1.31)	0.00
Pads	4.16 (1.48)	5 (1-5)	69.1	16.0	4.03 (1.58)	5 (1-5)	-0.14 (1.48)	0.19
Flatul	4.56 (0.70)	5 (1-5)	66.0	0.5	4.04 (1.04)	4 (1-5)	-0.53 (1.16)	0.00
Liquid	4.27 (0.88)	4 (1-5)	49.0	2.1	3.57 (1.21)	4 (1-5)	-0.70 (1.24)	0.00
Solid	3.12 (1.48)	3 (1-5)	20.6	25.3	2.61 (1.40)	3 (1-5)	-0.52 (1.36)	0.00
Adjustment	4.05 (1.30)	5 (1-5)	55.7	8.8	3.34 (1.56)	4 (1-5)	-0.71 (1.48)	0.00
Medication	1.31 (0.47)	1 (1-2) ^c	68.6	31.4	1.28 (0.45)	1 (1-2)	-0.03 (0.41)	0.38 ^d
Total score	18.20 (2.78)	18 (12-24)	4.1	2.6	15.19 (5.04)	16 (0-24)	-3.01 (4.85)	0.00 ^e

SD, standard deviation; FU, follow-up.

^a Ceiling effect: defined as ≥15% of patients in the worst (highest) outcome in terms of item/total score.^b Floor effect: defined as ≥15% of patients in the most desirable (lowest) outcome in terms of item/total score.^c Coded as follows: (1)=no, (2)=yes.^d McNemar test for nominal data.^e Wilcoxon signed rank test (no difference in significance between Wilcoxon and paired t-test on the total Vaizey score).**Table 4.4** Test-retest reliability between baseline and follow-up Vaizey scores, and internal consistency measures

Vaizey item	Weighted Kappa ^a (95% CI) (n=30)	Mean (SD) (n=30)	P-value marginal homogeneity test		Cronbach's alpha ^b (n=194)	Corrected item-total ^c (n=194)	Cronbach's alpha if item deleted (n=194)
Urge	0.20 (-0.13-0.51)	-0.50 (1.38)	0.06	0.25	0.47	0.59	0.19
Pads	0.64 (0.37-0.81)	-0.37 (1.03)	0.06	0.06	0.34	0.31	0.33
Flatul	0.52 (0.20-0.74)	-0.03 (0.72)	0.80	0.41	0.52	0.52	0.15
Liquid	0.53 (0.22-0.74)	-0.17 (0.91)	0.32	0.52	0.73	0.71	0.05
Solid	0.64 (0.38-0.81)	-0.27 (1.20)	0.23	0.17	0.51	0.38	0.23
Adjustment	0.73 (0.51-0.86)	0.03 (1.16)	0.87	-0.21	0.32	0.31	0.49
Medication	0.76 (0.56-0.88)	0.03 (0.32) ^c	1.00 ^d	-0.06	0.05	-0.10	0.33
Total score	0.55 (0.25-0.75)	-0.77 (3.36)	0.46 ^e				0.73
Overall				0.30	0.71	0.69	0.30
							0.71

^a Weighted kappa coefficient is equal to ICC_{agreement}.^b Vaizey score at baseline, follow-up and Vaizey change scores.^c Coded as follows: (1)=no, (2)=yes.^d McNemar test.^e Wilcoxon signed rank test (small difference in significance between Wilcoxon and paired t-test (P=0.22) on the total Vaizey score).

Internal consistency

The internal consistency was adequate for the total Vaizey scores at FU and Vaizey change scores (0.71 and 0.69, respectively), whereas the total baseline Vaizey score was poor (0.30) (Table 4.4).

At baseline, the subscale items of 'urge FI' 'pad use', 'solid stool incontinence', 'adjustment', and 'medication use' seem to measure a different construct from the other items of the scale (corrected item-total correlation <0.30). FU and Vaizey change scores showed values below 0.30 for medication use.

In view of 'Cronbach's alpha if item were deleted', estimates at baseline indicated that the items of 'pad use', 'adjustment', and 'medication use' seem to measure a different concept, as they exceed the overall Cronbach's alpha. Medication use differed from the remaining items in terms of FU and Vaizey change scores.

Responsiveness

Table 4.5 presents the mean changes in the Vaizey score at FU, for each of the five GPE categories. Eight patients (15.1%) rated themselves as 'much better', 11 (20.8%) as 'better' and 30 (56.6%) as 'unchanged'. The categories 'worse' and 'much worse' were reported by only three and one patients, respectively (7.5%). As a result of the low number of patients who were deteriorated after PFR, the MIC for those who were deteriorated was not determined.

Whether the change scores corresponded to the reported GPE category was evaluated as follows:

1. The group using the categories 'better' and 'much better' showed higher mean change scores than the unimproved group (unchanged, worse and much worse), which was reflected by significant changes in the total Vaizey score. The unchanged group showed no significant change, as expected. Given the small number of patients who deteriorated (n=4), the estimates of these Vaizey changes were less reliable.
2. Spearman's correlation between the GPE score and changes in the Vaizey score was -0.55 ($P<0.01$), indicating adequate longitudinal validity.
3. The unchanged group had an ES lower than 0.50, as expected. Contrary to our expectations, effect sizes and SRMs did not increase with the magnitude of subjective improvement.

Table 4.5 Patient's perception of change in terms of global perceived effect (GPE), and mean changes and percentage (%) of change in the Vaizey score at follow-up

GPE score (n=53)	Baseline mean (SD)	FU mean (SD)	n (%)	Change ^a mean (SD)	%	ES ^b	SRM ^c	P-value Wilcoxon signed rank test ^d
1. Much better	17.50 (3.63)	10.63 (5.18)	8 (15.1)	-6.88 (5.0)	-39.3	-1.9	-1.4	0.01
2. Better	17.36 (2.50)	11.91 (3.75)	11 (20.8)	-5.45 (3.39)	-31.4	-2.2	-1.6	0.01
3. Unchanged	18.03 (2.90)	17.27 (4.10)	30 (56.6)	-0.77 (3.36)	-4.3	-0.3	-0.2	0.46
4. Worse	17.33 (0.58)	16.00 (4.58)	3 (5.7)	-1.33 (4.16)	-7.7	-2.3	-0.3	0.66
5. Much worse	17.00	17.00	1 (1.9)	0	0.0	–	–	–
Total score	17.75 (2.79)	15.08 (4.96)	53 (100)	-2.68 (4.38) ^e	-15.1	-1.0	-0.6	0.00
Spearman's <i>r</i>				-0.55 ^e				
Improved	17.42 (2.93)	11.37 (4.32)	19 (35.8)	-6.05 (4.08)	-34.7	-2.1	-1.5	0.00
Not improved	17.94 (2.73)	17.15 (4.02)	34 (64.2)	-0.79 (3.32)	-4.4	-0.3	-0.2	0.38

SD, standard deviation; FU, follow-up.

^amin-max=0-24; Range=24, [Change score / Baseline score x 100%].^bES, effect size; mean change score per category divided by the standard deviation of baseline score.^cSRM, standardised response mean; mean change score per category divided by the standard deviation of the change scores.^dNo differences in significance between Wilcoxon and paired t-tests, except for GPE=3 (P=0.22) and the 'not improved' group (P=0.17).^eSpearman's correlation coefficient between the changes in the total Vaizey score and patient's perceived benefit assessed by the GPE.

Whether the total Vaizey score and individual Vaizey items were responsive to PFR was interpreted by means of Table 4.6. Neither pad use and nor medication use changed significantly (P=0.19 and 0.38, respectively), and both showed small effect sizes and small Spearman correlation coefficients. Although the individual item of 'solid stool incontinence' demonstrated the largest change over time (24.5%), the corresponding effect size was not very large. The individual item of 'urge' showed a significant decrease in Vaizey score, but the Spearman correlation coefficient ($r=-0.009$) did not correspond to this finding.

Table 4.6 Responsiveness of the Vaizey score between baseline (T0) and follow-up (T1)

	T0 mean (SD) (n=194)		Median (min-max) (n=194)		T1 mean (SD) (n=194)		Median (min-max) (n=194)		Change mean (SD) ^a (n=194)		P-value ^b (n=194)		Effect sizes ES / SRM ^c (n=194)		r_{spearman} ^d (n=53)	
Urge	4.30 (1.07)		5 (1-5)		3.69 (1.47)		4 (1-5)		-0.61 (1.31)		0.00		-0.6		-0.009 (P=0.952)	
Pads	4.16 (1.48)		5 (1-5)		4.03 (1.58)		5 (1-5)		-0.14 (1.48)		0.19		-0.1		0.004 (P=0.978)	
Flatulence	4.56 (0.70)		5 (1-5)		4.04 (1.04)		4 (1-5)		-0.53 (1.16)		0.00		-0.8		-0.369*	
Liquid	4.27 (0.88)		4 (1-5)		3.57 (1.21)		4 (1-5)		-0.70 (1.24)		0.00		-0.8		-0.459*	
Solid	3.12 (1.48)		3 (1-5)		2.61 (1.40)		3 (1-5)		-0.52 (1.36)		0.00		-0.4		-0.360*	
Adjustment	4.05 (1.30)		5 (1-5)		3.34 (1.56)		4 (1-5)		-0.71 (1.48)		0.00		-0.5		-0.457*	
Medication	1.31 (0.47)		1 (1-2) ^e		1.28 (0.45)		1 (1-2)		-0.03 (0.41)		0.38 ^f		-0.1		0.045 (P=0.747)	
Total score	18.20 (2.78)		18 (12-24)		15.19 (5.04)		16 (0-24)		-3.01 (4.85)		0.00 ^g		-1.1		-0.549*	

^aCorrected for the minimum GPE score of item scores of 1 point (min-max=1-5; range 4); [Change score / Baseline score-1] x 100%].

^bMarginal homogeneity test.

^cES=mean change score per category divided by the standard deviation of the baseline score. SRM=mean change score per category divided by the standard deviation of the change scores.

^dSpearman correlation coefficients between the changes in the total and individual Vaizey item scores and patient's perceived benefit assessed by the GPE.

^eCoded as follows: (1)=no, (2)=yes.

^fMcNemar test for nominal data.

^gWilcoxon signed rank test (no difference in significance between Wilcoxon and paired t-test on the total Vaizey score).

*Significant at the 0.01 level (two-tailed).

Minimally important change

The GPE score was considered suitable for use as an anchor, as it correlated sufficiently with the Vaizey change scores ($r=-0.55$). The estimates for the MIC and SDC yielded moderately consistent results (Table 4.7). However, adding (1-sensitivity) and (1-specificity) to calculate the overall misclassification by the ROC method resulted in very similar values when comparing MICs of -1, -2 and -5 (0.41, 0.37, and 0.41, respectively). However, an MIC of -5 is at least higher than the SDC and therefore a statistically reliable change beyond measurement error. In addition, an MIC of -5 yielded the lowest absolute number of misclassified patients ($n=10$), with five patients being misclassified who were not importantly improved ($n=34$) on the anchor, against five who were importantly improved ($n=19$) on the anchor. MICs of -1 and -2 yielded a total of 13 and 11 misclassified patients, respectively. We therefore preferred an MIC value of -5. The AUC of the ROC analysis was considered adequate (0.848).

Table 4.7 Minimally important change according to three methods and smallest detectable change ($n=53$)

	Mean change [abs]
1. Change method ¹¹	-4.70 [-5]
2. ROC optimal cut-off point	-2.00/-5.00 [-2/-5]
3. 95% limit cut-off point	-6.30 [-7]
SDC	-4.39 [-5]
Range (min-max)	-4.30 (-2.00 to -6.30) [-2 to -7]

Abs, absolute values; SDC, smallest detectable change.

Figure 4.1 shows the overall misclassification for importantly and not importantly improved persons resulting from different methods to estimate MIC and SDC.

Figure 4.1 Distribution of Vaizey change scores of patients (percentages) reporting an important improvement compared to those not importantly improved on the anchor ($n=53$). Change in Vaizey score: +, indicating improvement; -, indicating deterioration.

DISCUSSION

Just as previously found for surgical treatment, the Vaizey score also proves sensitive to change following PFR.³ Exceptions are some of the individual Vaizey items, viz. pad use and medication use. These items may correlate more with the patient's self-confidence and therefore may not change as rapidly as the other items. The effect sizes suggest that the frequency of solid stool incontinence is not as responsive to PFR as that of liquid stool or flatus incontinence ($ES < 0.5$). Incontinence for solid stool is probably less susceptible to improvement by PFR from a clinical and physiological point of view, as it is considered worse compared with incontinence for liquid stool or flatus, although this is not necessarily the case from a patient's point of view. The individual Vaizey item of 'urge' showed a significantly decreased score, although this was not reflected in the Spearman correlation coefficient ($r = -0.009$). Including non-responsive items in a severity scale may cause underestimation of the effectiveness of the intervention program. Future research should focus on the responsiveness of both the individual items and the total Vaizey score.

As expected, patients with urge FI showed greater improvement in terms of the individual items and the total Vaizey score than those with mixed FI, presumably due to the more complex character of mixed FI. Estimates for passive FI were only based on five patients, which made a comparison with the other two types of FI unreliable. Future research should elucidate which type of FI is most responsive to PFR, to achieve both easier selection of patients suitable for PFR and more realistic expectations from PFR.

Responses for the individual Vaizey items tended to cluster in the worst frequency. This may be associated with the inclusion of moderately to severely faecally incontinent patients (with a baseline Vaizey score of at least 12). Moreover, a higher baseline Vaizey score offers more room for improvement, which corresponds with our finding of only four deteriorated patients (7.5%). A stratified analysis was considered to determine the influence of the baseline Vaizey score on the MIC, but this analysis was abandoned due to the low number of patients with GPE data in the 'unchanged' and 'better' categories.

This study confirms that changes in Vaizey score after PFR reflect patients' subjective perception of relief.⁶ The small numbers of patients in the 'worse' and 'much worse' groups (3 and 1, respectively) may explain why a larger subjective improvement did not correspond with an exactly proportional increase in effect size.

The estimates for MIC and SDC yielded moderately consistent results. Various methods were used to estimate the MIC, to compensate for the disadvantages of each method. The change method and SDC suggested an MIC value of -5, while the 95% limit cut-off point resulted in an MIC of -7 and the ROC method in an MIC of -2 or -5. When applying the ROC method, the MIC depends on whether one chooses the cut-off point with the lowest percentage of overall misclassification (MIC=-2) or takes into account the lowest absolute number of misclassified patients and an MIC beyond the measure-

ment error (SDC) (MIC=-5). In this case, we preferred an MIC of -5, as this was also supported by the results of the change method. Future research is needed to determine which value represents the MIC best.

The interpretation of the MIC is different for the individual and group level. The average change in Vaizey score of -3.01 reflects the improvement in the total group, whereas 62 individuals achieved an improvement of at least five points on the Vaizey score (32%). This result can be considered moderate, but it should be interpreted in view of the inclusion of only severely faecally incontinent patients.

This study confirms that the Vaizey score is a reproducible score,^{3,4} although its internal consistency is poor at baseline. The FU and Vaizey change scores have adequate internal consistency except for the 'medication use' item. Since the individual Vaizey items do not all seem to measure the same construct, we suggest that the Vaizey scale structure should be revised in case of a low internal consistency or in case of non-responsive items. Future research is needed to determine whether the use and summing up of Vaizey subscales results in different conclusions.

Avery et al. indicate in their review on questionnaires to assess urinary and anal incontinence a grade C recommendation for the Vaizey score. This means that the score is in the early stage of development and further study is required and encouraged.¹³ No questionnaires were identified that meet the grade A criteria (highly recommended: validity, reliability and responsiveness established with rigor) and only three attained a grade B status, including the Fecal Incontinence Quality of Life Scale (FIQL),¹⁴ the Manchester Health Questionnaire¹⁵ and the Birmingham Bowel and Urinary Symptoms Questionnaire.¹⁶ These questionnaires are considerably longer than the 7-item Vaizey score.

The choice for selecting an outcome measure is not only dependent on its psychometric properties, but above all on the primary interest: this might be the assessment of symptoms, condition-specific quality of life, or a combination of both. Furthermore, available time to complete the questionnaire and the setting (clinic/home) should be taken into account.

Limitations

First, this study was conducted as part of a large diagnostic cohort study, which was not originally set up as a psychometric study. Second, GPE data was available for only 53 patients. In contrast to the entire sample of 194 patients, this probably produced less stable estimates for the effect sizes, correlations, test-retest reliability, and MIC and SDC estimates. Third, the GPE was chosen as the anchor in assessing the criterion validity of the Vaizey score. Although this is no 'gold standard', it was believed to best represent patients' opinions about improvement. Besides, the results presented here might have been different if a 7-, 9- or 15-point GPE scale had been used instead of a 5-point scale, as such scales differentiate more fully between the improved and deteriorated categories and probably produce less error. However, the correlation between the GPE score and the Vaizey change scores in this study ($r=-0.55$) was higher

than the correlation found by Maeda et al. between an 11-point scale measuring change in patients' subjective perception after biofeedback treatment and the Vaizey score ($r=0.45$).⁶

In view of these limitations, our results should be interpreted with caution.

The Vaizey score is currently being used as the primary outcome in an ongoing physiotherapy trial.¹⁷ The trial results will be used to verify the conclusions of the present study.

CONCLUSIONS

We conclude from our results that the psychometric properties of the total Vaizey score are not convincing. More research with larger samples is required to define its psychometric properties and to justify its use in research and routine clinical practice.

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CHAPTER 5

A RANDOMISED PHYSIOTHERAPY TRIAL IN PATIENTS WITH FAECAL INCONTINENCE: DESIGN OF THE PHYSIOFIT-STUDY

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Published in: BMC Public Health 2007;7:355

ABSTRACT

Background

Faecal incontinence (FI) is defined as the recurrent involuntary excretion of faeces in inappropriate places or at inappropriate times. It is a major and highly embarrassing health care problem which affects about 2 to 24% of the adult population. The prevalence increases with age in both men and women.

Physiotherapy interventions are often considered a first-line approach due to its safe and non-invasive nature when dietary and pharmaceutical treatment fails or in addition to this treatment regime. Two physiotherapy interventions, rectal balloon training (RBT) and pelvic floor muscle training (PFMT) are widely used in the management of FI. However, their effectiveness remains uncertain since well-designed trials on the effectiveness of RBT and PFMT versus PFMT alone in FI have never been published.

Methods/Design

A two-armed randomised controlled clinical trial will be conducted. One hundred and six patients are randomised to receive either PFMT combined with RBT or PFMT alone. Physicians in the University Hospital Maastricht include eligible participants. Inclusion criteria are (1) adults (aged ≥ 18 years), (2) with faecal incontinence complaints due to different aetiologies persisting for at least six months, (3) having a Vaizey incontinence score of at least 12, (4) and failure of conservative treatment (including dietary adaptations and pharmacological agents). Baseline measurements consist of the Vaizey incontinence score, medical history, physical examination, medication use, anorectal manometry, rectal capacity measurement, anorectal sensation, anal endosonography, defecography, symptom diary, Fecal Incontinence Quality of Life scale (FIQL) and the PREFAB-score. Follow-up measurements are scheduled at three, six and 12 months after inclusion. Skilled and registered physiotherapists experienced in women's health perform physiotherapy treatment. Twelve sessions are administered during three months according to a standardised protocol.

Discussion

This section discusses the decision to publish a trial protocol, the actions taken to minimise bias and confounding in the design, explains the choice for two treatment groups, discusses the secondary goals of this study and indicates the impact of this trial on clinical practice.

Trial registration

The Netherlands Trial Register ISRCTN78640169.

BACKGROUND

Background of faecal incontinence

Faecal incontinence (FI) is a major health care problem, that is particularly embarrassing and affects about 2 to 24% of the adult population, increasing to 47% in the institutionalised elderly.¹⁻⁷ The actual prevalence is likely to be higher due to the common underreporting of FI as a result of patients' embarrassment to visit a physician or unawareness of possible treatment options.⁸ FI can be defined as the recurrent involuntary excretion of faeces in inappropriate places or at inappropriate times,⁷ and covers a wide spectrum from involuntary but recognised passage of gas, liquid, or solid stool (urge incontinence) to unrecognised anal leakage of mucus, fluid, or stool (passive incontinence).⁹ Faecal continence is based on a combined interplay of sensory, motoric and reservoir functions. Incontinence occurs if one or more of these components fail and when compensatory mechanisms fall short. Obstetric trauma is one of the major causes of FI in women. Several colorectal, urological or gynaecological interventions can cause FI as well. Specific neurological diseases associated with FI include diabetes, multiple sclerosis, Parkinson's disease, stroke, and spinal cord injury.

FI is often thought to be associated with older people, as a natural aspect of ageing. Recently, it has been estimated that 6% of the persons aged 60 years and older suffer from involuntary loss of faeces and 3% from involuntary loss of both faeces and urine.⁸ In addition, the prevalence of FI will increase in the next fifteen years due to double ageing. However, younger people are often affected as well.¹ FI interacts with multiple aspects of daily life resulting in a number of difficulties; having to stay at home near a toilet, having to avoid social contacts including relationships or sexual contact, having feelings of depression and low self-esteem.² Moreover, it is often difficult to explain experiencing FI to others, like family or a partner. A lot of these implications are due to the unpredictable character of FI and the fear of odour or soiling.¹⁰

Apart from a social burden to the patient, FI also has a significant impact on the health care budget. Limited data are available on healthcare costs for FI.^{9,11} Recently, total costs in the Netherlands were estimated at €2169 per faecally incontinent patient a year.¹²

Estimates of health care costs and prevalence numbers for FI vary greatly and depend on the clinical setting and on the definition of FI based on severity and frequency.^{1,13,14}

Physiotherapy in faecal incontinence

Treatment of faecally incontinent patients consists of conservative as well as surgical interventions. Surgical interventions aim to anatomically and functionally correct the rectum, anus, pelvic floor or anal sphincters. Conservative interventions consist of dietary adaptations, medication, absorbent materials, and physiotherapy. Physiotherapy interventions are non-invasive and inexpensive, require no sophisticated equipment and have hardly any adverse effects. Besides, physiotherapy does not exclude

any other form of treatment.¹⁵ Therefore, physiotherapy is often attempted before surgical treatment.

Physiotherapy FI management includes pelvic floor muscle training (PFMT), biofeedback (BF), rectal balloon training (RBT) and electrical stimulation (ES). PFMT aims to restore muscular strength, coordination and timing of contractions. With BF the patient gets information about the activity of the pelvic floor muscles by way of a visual display. RBT, as a modality of BF, is used to decrease the sensory threshold in case of passive FI and to increase the sensory threshold in case of urge FI. In addition, RBT aims to synchronise contraction of the external anal sphincter with relaxation of the internal anal sphincter. ES is used to increase awareness and isolated contraction of the anal sphincters. Often, one or more physiotherapy interventions are combined, depending on the underlying cause of FI.

Over 60 uncontrolled trials exist on the use of BF for the management of FI.¹⁶ Some authors conclude that BF is the treatment of choice for FI on the basis of these observational studies.¹⁷ An overall cure and improvement rate of 72% has been reported.¹⁸ However, the results of a Cochrane review on the effects of BF and/or PFMT for the treatment of FI in adults were based on eleven randomised controlled but heterogeneous trials and showed that some elements of BF therapy and sphincter exercises might have a therapeutic effect, but this is not certain.¹⁶ Moreover, it was suggested that RBT improved continence more than sham training. This is in agreement with other authors who consider lowering the threshold volume for discrimination of rectal distension an important factor in the success of BF.^{15,19-21}

In an attempt to assess the effect of the different BF components (balloon/pressure BF, electromyographic BF) in the management of FI, two complicated cross-over trials have been performed.^{15,20} Group comparison was impossible to assess due to the small sample in one trial¹⁵ and the single case experiments in the other.²⁰

A second Cochrane review evaluated ES in adult patients with FI.^{16,22} Insufficient data was available to allow reliable conclusions on the effect of ES in the management of FI.²² A recently published randomised controlled trial examined whether anal ES, in the absence of any adjunct exercises or advice, would improve symptoms more when compared with 'sham' ES. It is concluded that 1 Hz (sham) was as effective as 35 Hz. Possibly, the main effect is not sphincter contraction but sensitisation of the patient to the anal area, or simply the effect of intervening per se (Hawthorne effect).²³

Finally, the results of a recent large cohort study on ES and PFMT with BF in patients with severe FI indicated that pelvic floor muscle rehabilitation leads to a modest improvement in severity of FI, squeeze pressure and maximal tolerated volume.²⁴

Although uncertainty exists on the exact extent of the effectiveness of physiotherapy treatment in FI, results are promising.¹⁸

Relevance

Evaluating effectiveness of physiotherapy treatment has been impeded by lack of enough quality studies, lack of standardisation, a large variety in outcome measures and methodological flaws.²⁵ Ultimately, there is a need to evaluate treatment options in FI, especially since FI considerably reduces quality of life. Besides, FI has a significant impact on the health care budgets, and the prevalence of FI is increasing due to double ageing.²⁶

PFMT has been proven effective in the treatment of stress urinary incontinence.²⁷ Since the anal sphincter and puborectalis muscle form a part of the same pelvic floor as the closing urethral system, expectations are raised for the same positive results of pelvic floor muscle re-educative techniques in FI. Next to PFMT, some authors consider lowering the threshold volume for discrimination of rectal distension as an important factor in the success of physiotherapy.^{15,19-21} No study has reported adverse events from either PFMT or RBT, and it seems unlikely that these treatments itself could cause symptoms to worsen.

Apparently, PFMT and RBT seem to have a role in the treatment of faecally incontinent patients. Since well-designed trials on the effectiveness of RBT and PFMT versus PFMT alone in patients with FI have never been published, a randomised controlled trial on this topic will be carried out (Physiotherapy in Faecal Incontinence Trial – PhysioFIT-study).

METHODS/DESIGN

Study design, research question and hypothesis

A two-armed randomised controlled clinical trial will be conducted in patients diagnosed with FI (Figure 5.1), serving the following research question: what is the effectiveness of rectal balloon training and pelvic floor muscle training compared to pelvic floor muscle training alone in patients with FI?

It is hypothesised that patients receiving the combined training will have a larger reduction on the primary outcome measure.

Figure 5.1 Flowchart of the PhysioFIT-study.
t, point in time; VS, Vaizey score; MH, medical history; M, medication use; PE, physical examination; DI, diary; AM, anorectal manometry; RC, rectal capacity measurement; AS, anorectal sensation; AE, anal endo-sonography; D, defecography; FIQL, Fecal Incontinence Quality of Life Scale; PREFAB, modified PRAFAB-score; GPE, Global Perceived Effect-score; PFMT, pelvic floor muscle training; RBT, rectal balloon training; VS (-), reduction on VS <4 points; VS (+), reduction on VS ≥4 points; GPE (+), score 1-7 on GPE scale; GPE (-), score 8 or 9 on GPE scale.

Study population

The study population consists of patients living in the Netherlands. Participants are eligible when they are diagnosed with FI after general diagnostics. The randomised controlled trial is carried out at the University Hospital Maastricht, where two colorectal surgeons (CB, WG) include eligible participants. Inclusion criteria are (1) adults

(aged ≥ 18 years), (2) FI complaints due to different aetiologies persisting for at least six months, (3) Vaizey incontinence score of at least 12,²⁸ (4) and failure of conservative treatment (including dietary adaptations and pharmacological agents). Inclusion criteria 2, 3 and 4 indicate that patients with moderate to severe FI complaints are of interest. Patients diagnosed with an anorectal tumour within the past two years, absent squeeze pressure of the anal sphincter, chronic diarrhoea (always fluid stool three or more times a day), overflow incontinence, proctitis, ulcerative colitis, Crohn's disease, soiling (defined as leakage of a minimal amount of faeces out of the anal canal), previous ileo-anal or colo-anal anastomosis and/or rectal prolapse in situ are excluded. Participants who received physiotherapy during the previous six months or who are intellectually and/or linguistically incapable to understand therapy are excluded as well.

Recruitment, informed consent and randomisation

The medical ethics committee of the University Hospital Maastricht/Maastricht University approved the PhysioFIT-study. The physician points out during the first visit that the patient is eligible to participate in the trial. The patient is provided with patient information sheets and information on the diagnostic tests. Participating patients sign informed consent at the beginning of the second visit, allowing eligible participants two months to decide to participate.

An independent research assistant performed the blocked randomisation (size of four). A computer-generated randomisation list is prepared in advance. The independent research assistant contacts the physiotherapist by telephone to inform about the referral and the allocated treatment.

Blinding

The research doctor, performing the general diagnostics, and the researcher, dealing with the data-entry, are blinded during baseline measurements but not during follow-up measurements. However, the physician involved in measuring the primary outcome measure is blinded and blinded double data-entry is performed to assure correct data-entry. It is impossible to blind patients and participating physiotherapists for treatment allocation. However, physiotherapists are not involved in the diagnostic work-up and follow-up measurements. The outcome assessors are blinded.

Baseline and follow-up measurements

Given the multiple aetiologies in patients with FI, no test should be expected to yield consistent results in all patients with FI. Therefore, no standard protocol is available for the assessment and diagnosis of FI. The chosen tests at baseline (except for the questionnaires) are recommended by the American Gastroenterological Association, being procedures of value (symptom diary, physical examination, anal endosonography, anorectal manometry, rectal and anal sensory testing) and being procedures of possible value (defecography).²⁹ As these diagnostic tests are part of the standard diagnos-

tic assessment in the University Hospital Maastricht, participation in the trial will not burden the patient additionally. The following baseline measurements are done during the first and second patient visit:

- **Vaizey incontinence score:** this score reflects the severity of FI and ranges from 0 (complete continence) to 24 (complete incontinence) (Table 5.1).²⁸ The Vaizey score is a widely used severity score and it is reported to be a reproducible score, which correlates highly with physicians’ clinical impression.²⁸ Recently, it has also been demonstrated that higher Vaizey scores are associated with more reported problems in general health domains³⁰ and that changes in Vaizey score reflect patients’ subjective perception of relief.³¹

Table 5.1 Faecal incontinence score according to Vaizey et al.²⁸

Incontinent	Never	Rarely	Sometimes	Weekly	Daily
Solid stool	0	1	2	3	4
Liquid stool	0	1	2	3	4
Gas	0	1	2	3	4
Alteration in lifestyle	0	1	2	3	4

	No	Yes
Need to wear a pad or plug	0	2
Taking constipating medication	0	2
Lack of ability to defer defecation for 15 minutes	0	4

Minimum score=0=perfect continence; maximum score=24=totally incontinent.
Never: no episodes in the past 4 weeks; rarely: one episode in the past 4 weeks; sometimes: > one episode in the past 4 weeks, but < once a week; weekly: one or more episodes a week but < one a day; daily: one or more episodes a day.

- **Medical history:** the medical history assesses gender, age, duration of complaints, risk factors (surgical, obstetric, neurological, metabolic) and medication use.
- **Physical examination:** digital rectal examination is used to assess anal pressure at rest and pressure during contraction. Sphincter defects can be located and palpated, especially during contraction. In women, the presence of a rectocele can be established.
- **Anorectal manometry:** anorectal manometry is performed to measure anal sphincter pressures. The test is performed in the left lateral position and flexed hips to 90 degrees. Anal manometry takes place according to the water-perfused technique. The single use manometric catheter (Medtronic Inc., Skovlunde, Denmark) is introduced, which is connected to a computer-assisted polygraph (Polygraf™ ID, Medtronic Inc., Skovlunde, Denmark) and stabilised in the anal sphincter complex. After positioning the catheter in the maximal pressure area, the basal sphincter pressure, maximum squeeze pressure and rectal anal inhibitory reflex are measured.
- **Rectal capacity measurement:** the measurement is performed by introducing a single use manometric catheter (Medtronic Inc., Skovlunde, Denmark) in the rectum, which is connected to a computer-assisted polygraph (Polygraf™ ID, Medtronic Inc., Skovlunde, Denmark). The catheter is inflated with air with slow

increments. The volumes are recorded at which first sensation is noticed (sensory threshold), an urge to evacuate is produced (urge sensation), and volume at which pain becomes intolerable (maximum tolerated volume).

- Anorectal sensation: a St. Mark's glove pudendus electrode is placed on the index finger and introduced into the anal canal. An increasing electrical current (mAmp) is gradually applied until the threshold of sensation is reached. Anorectal sensation is measured at two places: three o'clock and nine o'clock in the transversal plane.
- Anal endosonography: anal endosonography provides an anatomical image of the anal sphincters and pelvic floor. This technique gives an instant excellent image of the condition of the sphincters and establishes the size of a possible internal or external anal sphincter defect. Anal endosonography is performed with an ultrasound scanner with radial endoscopic probe and a 7.5-MHz transducer. The probe is covered with a lubricant and a condom. After application of a final lubricant the probe is introduced in the anal canal with the patient in the left lateral or prone position. The probe is slightly withdrawn, so all the different levels of the anal sphincter complex can be visualised.
- Defecography: defecography is a dynamic radiological study of attempted defecation and shows both the functional aspects of defecation and defects in anorectal anatomy. Patients are instructed to drink contrast medium diluted water before examination. The test starts with the patient in left decubital position. Through an injection pistol barium paste is manually injected in the rectum. In female patients gel is also injected via a syringe in the vagina. The perineum is located with a gel solution as well. Subsequently, the entire X-ray table is tilted upright 90^0 and the patient is seated on a specially developed radiolucent defecography chair. After the test the patient is instructed to drink sufficiently to eliminate the contrast.
- Diary: the diary gives insight into the defecation pattern, involuntary incontinence episodes, pad use, presence of urge feelings, presence of urinary incontinence, alteration in daily activities, ability to infer defecation and medication use of a patient. The diary monitors three weeks. Since FI often goes together with urinary incontinence, additional questions will be included concerning the severity of urinary incontinence. Since limited information is available about the quantity of absorbent material use and the associated costs, the diary will enclose additional questions to get more insights into this domain and estimate accompanying health care costs.
- Fecal Incontinence Quality of Life Scale (FIQL): the FIQL is a disease-specific quality of life scale, comprising four scales: Lifestyle (10 items), Coping/Behaviour (9 items), Depression/Self Perception (7 items), and Embarrassment (3 items). The FIQL has shown to be reliable and valid.³² A separate quality of life scale is included as FI is considered to highly influence patients' daily life since uncertainty exist on when incontinence episodes may occur.
- PREFAB-score: the PREFAB-score is a modified version of the widely used and accepted PRAFAB-score in the urinary incontinence field. The PRAFAB-score is a sim-

ple and quickly obtained score which summarises several objective and subjective components of the severity of urinary incontinence complaints (ranging from 5–20).³³ The score has proven to be reliable, valid and responsive.³⁴ Since most of the PRAFAB-score, except for the ‘amount’ item, is applicable to faecally incontinent patients as well, it is suggested to adapt the PRAFAB-score to faecally incontinent patients. From the patients’ point of view, two aspects about excretion of faeces are reported to be highly embarrassing and affecting social life and body image: whether the excretion can be held up by a pad or not and whether excretion can be smelled by the environment or not. These two aspects, reflecting social limitations, are therefore integrated in the modified PRAFAB-score (min–max=6–20; range=15).

- Global perceived effect (GPE) (Table 5.2): this subjective score reflects the change in complaints after physiotherapy treatment compared to the period before treatment and ranges from 1–9 points. A score of 8 or 9 indicates much worsening of complaints and is an indication to consider co-interventions.

Table 5.2 Global perceived effect (GPE score)

To which degree have your complaints changed compared to the period before physiotherapy treatment?	
At the moment, the complaints are	
1.	Very much improved
2.	Much improved
3.	Moderately improved
4.	Slightly improved
5.	Unchanged
6.	Slightly worse
7.	Moderately worse
8.	Much worse
9.	Very much worse
..... compared to the period before physiotherapy treatment	

Follow-up measurements are scheduled at 3, 6 and 12 months after inclusion. Primary outcome measure is the Vaizey score.²⁸ Success after physiotherapy is defined as a reduction in Vaizey score of more than four points (16.7%) compared to baseline measurement. Recently, it is reported that patients who rated their situation to be better, improved four points on the Vaizey score.³¹ Secondary outcome measures are anorectal resting and squeeze pressure, rectal capacity, anorectal sensation, diary results, FIQL, PRAFAB-score and GPE score.

Sample size

The sample size is calculated based on data from a recently published cohort study on pelvic floor rehabilitation in faecally incontinent patients.³¹ Patients who reported their situation to be better after pelvic floor rehabilitation showed a mean improvement of 4.33 points on the Vaizey score and patients who rated their situation to be equal or (much) worse showed a mean improvement score of one point (baseline average Vaizey score is 18). As subjective improvement can not be projected as decimals on the Vaizey score, we calculated two sample sizes: (1) with exact scores and (2) with abso-

lute scores to correspond with the Vaizey score and estimate of the minimally important change (MIC). The absolute difference in improvement of four points between the patients who rated themselves to be better and the patients who rated themselves to be equal or (much) worse is considered the MIC on the Vaizey score between both trial arms.³¹

The first sample size is calculated using an average Vaizey score of 13.67 and a standard deviation of 8.45 for sample 1 and an average Vaizey score of 17 and a standard deviation of 3.90 for sample 2, which resulted in a sample size of 106 patients. The second sample size is calculated using an average Vaizey score of 13 and a standard deviation of 8.45 for sample 1 and an average Vaizey score of 17 and a standard deviation of 3.90 for sample 2, which resulted in a sample size of 73 patients. The sample size is in a range of 73 to 106 patients and we decided to include at least 106 patients to feel confident about the required sample size.

All calculations assume a one-sided effect (first trial arm has a better outcome than second trial arm), an alpha of 0.05, a power of 80% and an expected drop-out of 10%. It is expected that the drop-out will be minimal since patients will be greatly motivated to participate and adhere to the protocol, by experiencing such an embarrassing and socially restricting disorder.

Referral

After diagnosis and inclusion, patients are referred to an extramural private practice, preferably nearby their home address. Private practices are selected with physiotherapists experienced in women's health, who are registered at the Dutch Association for Physical Therapy for Pelvic Floor Disorders and Pre- and Postnatal Healthcare (NVFB). They are all educated and trained in the performance of invasive techniques, as is used during digital testing and RBT. In total, 90 physiotherapists participate throughout the Netherlands. The initial physiotherapy session should be within three weeks after referral. The patient hands over an acknowledged doctor's referral letter and fifteen rectal balloons (group 1) to the physiotherapist before the initial session. FI is not recognised as a chronic disorder and insurance coverage is patient dependent. However, physiotherapy in this trial is considered as being part of usual care, as included patients would also be excellent candidates for physiotherapy outside the scope of this trial. Since January 2006 a referral letter is not obligatory anymore.

Physiotherapy intervention

Treatment is administered according to a standardised protocol, which has been developed by clinicians and physiotherapists specialised in the field of pelvic floor disorders. Physiotherapy consists of pelvic floor muscle training (PFMT) and rectal balloon training (RBT; only trial arm one). Patients receive 12 sessions within three months after the initial session, starting with two sessions a week during three weeks, and thereafter reducing to one session a week. The first session takes 45 minutes and all

subsequent sessions 35 minutes. A case report form physiotherapy (CRF) is filled out for each physiotherapy session.

Physiotherapists are aware of details about the medical history, physical examination and diary, but blinded for findings of anal manometry, rectal capacity measurement, anorectal sensation, anal endosonography, defecography, FIQL and PREFAB-score. During physiotherapy sessions, information is obtained by performing peri-anal inspection, digital rectal evaluation and rectal balloon training (trial arm 1).

Pelvic floor muscle training

PFMT is offered to all included patients and consists of selective voluntary contractions and relaxations of the pelvic floor muscles and the anal sphincter. Awareness of these muscles is necessary, as is the avoidance of the use of surrounding muscles like the adductors or abdominals. PFMT aims at maximising strength, improving duration of strength, and improving timing and coordination of contractions. One treatment cycle starts with a series of 8–12 single maximum contractions, sustained for 1–3 seconds. During treatment the series are extended from 1–3, and the sustained maximum contraction from 1–3 seconds to 6–8 seconds. A one-minute rest is allowed between the cycles. One cycle is repeated three times. Furthermore, patients perform three sustained submaximal contractions of 30 seconds, with one-minute rest in between. Patients are also instructed to perform home exercises daily. At home the cycles and sustained contractions are done in the same way and three times daily. The exercises are practiced in different starting positions and circumstances, simulating as much as possible daily life. Patients' compliance is encouraged because the home exercises determine to a great extent the success of physiotherapy. Digital rectal examination is used to measure the ability of pelvic floor muscles to contract and to quantify the strength. A contraction is considered to be sufficient if the pelvic floor and anal sphincter lift inward.

Rectal balloon training

A rectal balloon attached to a syringe is introduced in the rectum and slowly inflated with air to imitate rectal contents. Sensory threshold, urge sensation, and maximal tolerated volume are assessed. Patients with an insensitive rectum are trained to discriminate and respond to smaller rectal volumes of distension until a normal level of sensory threshold is reached or further improvement cannot be expected. In this way, patients receive an earlier warning from stool entering the rectum and impending defecation. In addition, patients can counteract reflex inhibition of the internal sphincter due to awareness of stool in the rectum.¹⁵ Patients with a hypersensitive rectum are trained to tolerate larger volumes by the use of progressive distension and urge resistance until a normal level of urge sensation is reached or the urge does not diminish with time or is uncomfortable. Patients with weakness of the pelvic floor muscles learn to contract these muscles immediately and strongly in response to rectal filling.

Details of each session

Details of each physiotherapy session are presented in the Appendix.

Co-interventions

After three months follow-up the physician evaluates the Vaizey score and the GPE score. In case of an improvement of less than four points on the Vaizey score and/or a GPE score of eight or nine, the clinician decides upon further therapeutic steps. If necessary, co-interventions are allowed after three months follow-up, although this should be avoided as much as possible. Examples of co-interventions are sacral nerve stimulation, anal repair or lavage. During physiotherapy treatment, patients are not allowed to use anal tampons.

The most common used drugs in patients with moderate to severe FI are Loperamid and sometimes Norit. Medical interventions and/or diet changes are first tried in the conservative management of patients with FI. If patients do not benefit from medical interventions, usual care implies referral for physiotherapy interventions. So, the majority of patients do not use drugs before physiotherapy management since they appeared not to be helpful. Nevertheless, patients who use drugs before starting with physiotherapy interventions are instructed to follow an unchanged stable medical regimen. Therefore, ongoing and change in medication use are monitored during the trial. In the analysis we will account for co-interventions, such as medication use.

Data analysis

In case of longitudinal missing data, imputation is performed with the last observation carried forward method. Main outcome is tested according to the intention to treat principle. Both treatment groups are checked for comparability regarding baseline data. An independent t-test is done for continuous variables and a Chi-square test for categorical variables. Analyses are performed to compare differences within groups between baseline and short-term follow-up (3 months) and long term follow-up (6 and 12 months) regarding primary and secondary outcome measures. A subgroup analysis is done for the patients with and without co-interventions. Analysis of variance is used to determine the difference in mean change of outcome measure between clinical subgroups. A two-tailed P-value of 0.05 is considered to indicate statistical significance. Data analysis is done using SPSS version 13.0 (SPSS, Inc., Chicago, Illinois). Confounders that are taken into account are presence of urinary incontinence, use of anal tampons, compliance and medication use.

Compliance

The ability of the patient to perform exercises at home is documented at the beginning of all physiotherapy sessions and if not, the reason is asked.

DISCUSSION

Why publish a study protocol

It is decided to publish the study protocol before obtaining results as we think it is important to share our thoughts and decisions about designing this study with other researchers in the field. Furthermore, a research article offers only a limited space to explain and reflect on the methods of a trial. Finally, researchers in the field are aware of which study is ongoing, which prevents wasteful duplication of research effort.

Bias and confounding

To minimise information bias, the performance of physiotherapists experienced in women's health is standardised. First, only physiotherapists registered at the NVFB are able to participate in this study. Registration approval indicates satisfying the outlined quality criteria by the NVFB. Second, participating physiotherapists receive a general and physiotherapy protocol of the PhysioFIT-study composed by experts in the field. Third, participating physiotherapists attend a general meeting. An individual visit is organised when a patient is referred to a non-attending physiotherapist. During the meeting, the physiotherapists were instructed on the general and physiotherapy protocol, a DVD-film illustrated the intended rectal balloon training and questions and discussions were raised. Some small adaptations were made in the protocol based on remarks from the physiotherapists.

To further minimise information bias, adaptations were made to the rectal balloons of the supplier. The current balloon reached the sigmoid colon, as soon as the balloon was filled with approximately 180 mL, possibly resulting in colic pains. This prevented the measurement of a higher maximal tolerated volume, although 180 mL is rarely reached. As this balloon was inadequate for this trial, adapted rectal therapy balloons were developed in cooperation with the supplier.

In this trial, it is impossible to blind on all levels. Most importantly, the measurement of the primary outcome is blinded as well as the outcome assessors. Blinded double data-entry is performed to assure correct data-entry.

Confounders that are taken into account are presence of urinary incontinence, use of anal tampons, patients' compliance and medication use. The presence of urinary incontinence can influence the perception of patients' quality of life and estimation of severity of FI, thereby distorting the scores on the FIQL, PREFAB and GPE scores. Furthermore, patients' compliance to the home exercise program determines therapy intensity and indirectly therapy success. Therefore, the physiotherapist asks each session patients' compliance to the home exercise program as formulated. The patient gives an explanation in case of insufficient compliance. Finally, the use of anal tampons and medication use can influence the number of involuntary events and stool consistency, as measured during the visits to the physician and in the diary.

Trial arms and patient selection

The effectiveness of rectal balloon training is assessed in a randomised controlled trial using two trial arms. A third trial arm with RBT as a single treatment option is not included, for the following reasons: RBT, as a solitary therapy option, has another suggested goal than PFMT. PFMT is hypothesised to improve strength, coordination and endurance. RBT is hypothesised to teach patients with an insensitive rectum to discriminate and respond to smaller rectal volumes of distension until a normal level of sensory threshold is reached. Patients with a hypersensitive rectum are taught to tolerate larger volumes by the use of progressive distension and urge resistance until a normal level of urge sensation is reached. In our first trial arm RBT is performed to strengthen the effect of PFMT. To compare this trial arm with only PFMT, the additional effect of RBT can be assessed.

In addition, a review by Mørkved, based on seven randomised controlled trials published between 1999 and 2005 on conservative treatment (PFMT with or without BF and ES) concluded that PFMT with and without BF seems to be effective in reducing FI.³⁵ This shows us that many physiotherapy intervention trials are designed with PFMT being at least part of the intervention. Adding a trial arm enables us to assess the effect of a more optimised physiotherapy intervention. Based on this and consensus in patients with moderate to severe FI, it is decided that PFMT alone should be at least the minimum standard of care in this disabled patient group.

Finally, as a result of strict in- and exclusion criteria we expect to include only one patient a week, resulting in an unacceptable long inclusion and follow-up period or insufficient power when extending to three trial arms.

Secondary goals

Secondary goals of the PhysioFIT-study are to assess the psychometric properties of the PREFAB-score in clinical practice. In addition, baseline measurements are checked for their ability to predict outcome of physiotherapy.

Furthermore, the responsiveness of the Vaizey score is reviewed. The Vaizey score is a more common used severity score in surgical trials. But less is known about the responsiveness of the Vaizey score in physiotherapy trials. One study with comparable in- and exclusion criteria and study population made a first attempt to correlate the Vaizey score with a global perceived effect score.³¹ Patients with a decrease of 9 points rated their situation 'much better' than before physiotherapy treatment, patients with a decrease of 4 points rated their situation 'better' and the group that rated their situation '(much) worse or equal' had a decrease in 1 point (patients were asked in a standardised interview to score their current situation on a scale of 1 (much worse), 2 (worse), 3 (no change), 4 (better), or 5 (much better)). We decided that rating the patients' situation as 'better' is a minimally important change, expressed in an absolute number and not in a certain percentage.

Next to the Vaizey score we included the subjective GPE score (range 1–9) in our trial as an additional outcome measure, as we believe it is important to further investigate the responsiveness of the Vaizey score in patients with FI in physiotherapy trials. Finally, it is expected that the outcome after physiotherapy interventions is dependent on the baseline Vaizey score; therefore we will perform a stratified analysis on baseline severity scores and change scores.

Transfer of knowledge and implementation

The results of this trial will be used for recommendations in the guideline of physiotherapy in faecally incontinent patients. This guideline will be developed according to the method of clinical practice guideline development of the Royal Dutch Society for Physical Therapy (KNGF).³⁶

Spin off

Often, a trial is designed with the objective to make a difference in clinical daily practice. This section elaborates on what we believe might influence clinical practice.

Based on the results of this trial, baseline measurements are checked for their ability to be a prognostic factor for a good or poor outcome after physiotherapy. In this way, patients can be selected who benefit most of the proposed physiotherapy intervention. Furthermore, this trial can give information on the number of physiotherapy sessions after which more sessions are of no further benefit. Continuation or ending of therapy will have the same outcome at that point. Finally, if the effectiveness of a certain physiotherapy intervention is established, physicians can make a well-founded decision for referral to physiotherapy treatment, instead of proceeding with surgical treatment. In conclusion, this trial might add to clearer referral patterns and better cooperation between physicians and physiotherapists.

In case of insufficient result of physiotherapy and proceeding to surgical treatment, the received physiotherapy intervention can still be of value. Patients already learned to be aware of their pelvic floor muscles and know how to contract them in an environment without post-surgical stress. Besides, their pelvic floor is probably in a better condition. Future research in this area can focus on the effect of different pre-operative physiotherapy interventions on the success of surgery in FI.

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APPENDIX

Appendix Details of each physiotherapy session

Session 1 (45 minutes)

- Explanation and information

In the first physiotherapy session, expectations of both the patient and therapist are discussed and the anatomy of the pelvic floor and anal sphincters is explained, as well as the functioning of the pelvic floor muscles. Furthermore, the patient receives details about the treatment program. Informing the patient will stimulate the compliance of the patient and improve the patient-therapist relationship.³⁷ For long-term success, the provision of training and information is important, especially with regard to pelvic floor use and toilet behaviour, which should be incorporated into patients' daily life.

Exercise therapy given in combination with the provision of information and training is an example of a so-called behavioural technique, in which certain steps are important: the exchange of information and explanation ('thinking'), the awareness and feeling of the pelvic floor, posture and movement ('feeling'), exercising and training the pelvic floor ('doing') and encouraging compliance to therapy over both the short and long term ('keep on doing').³⁸

- Explanation of pelvic floor muscle training (group 1 and 2) and rectal balloon training (group 1)

Session 2 (35 minutes)

- Inspection and digital rectal examination

Additional information for the physiotherapist is obtained by inspection and digital rectal examination. Inspection provides information about the perineal movement during contraction and straining of the pelvic floor. Digital rectal examination reveals pain complaints. Besides, pelvic floor muscle and external anal sphincter muscle strength is assessed according to the Oxford score, ranging from 0 (no muscle contraction) to 5 (strong contraction).³⁹ Endurance of submaximal strength (maximum 30 sec) of these muscles and exhaustion of a maximum contraction (maximum 5 times 1 sec) is also determined. Finally, the sensory threshold, urge sensation, and maximal tolerated volume are assessed.

- Treatment according to treatment arm

Session 3 to 11 (35 minutes)

- Evaluation

-Discuss problems with the patient

-Evaluation by way of digital rectal examination to assess pelvic floor muscle and external anal sphincter muscle strength according to the Oxford score (session 9).

- Treatment according to treatment arm

Session 12 (35 minutes)

- Evaluation
- Discuss problems with the patient
- Evaluation by way of digital rectal examination to assess pelvic floor muscle and external anal sphincter muscle strength according to the Oxford score. In addition, endurance of submaximal strength (maximum 30 sec) of these muscles and exhaustion of a maximum contraction (maximum 5 times 1 sec) is determined.
- Treatment according to treatment arm
- Evaluation of treatment program and compliance to treatment program

CHAPTER 6

RECTAL BALLOON TRAINING AS ADD-ON THERAPY TO PELVIC FLOOR MUSCLE TRAINING IN ADULTS WITH FAECAL INCONTINENCE: A RANDOMISED CONTROLLED TRIAL

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ABSTRACT

Background and aims

Faecal incontinence (FI) is embarrassing and socially restricting, resulting in poor quality of life. Rectal sensation may be more important than sphincter strength to relieve symptoms. A single-blind, randomised controlled trial among adults with FI compared the effectiveness of rectal balloon training (RBT) and pelvic floor muscle training (PFMT) versus PFMT alone.

Methods

We randomised eighty patients, recruited from the Maastricht University Medical Centre. Primary outcome was based on the Vaizey score. Secondary outcomes were the Fecal Incontinence Quality of Life Scale (FIQL), nine-point global perceived effect score, anorectal manometry, rectal distension volumes, and thresholds of anorectal sensation. Analyses were by intention to treat.

Results

Forty patients were assigned to combined RBT with PFMT and 40 to PFMT alone. Adding RBT did not result in a significant improvement in the Vaizey score (mean difference, -1.19; 95% confidence interval (CI), -3.79–1.42; $P=0.37$). Secondary outcomes favouring RBT were: Lifestyle subscale of the FIQL (0.37; 95% CI, 0.02–0.73; $P=0.039$), global perceived effect (-1.01; 95% CI, -1.75– -0.27; $P=0.008$), maximum tolerable volume (49.35; 95% CI, 13.26–85.44; $P=0.009$), and fatigue of the external anal sphincter (0.65; 95% CI, 0.26–1.04; $P=0.001$). Overall, 50% of patients were considered improved according to the estimated minimally important change (Vaizey change ≥ 5).

Conclusions

RBT with PFMT was equally effective as PFMT alone. Secondary outcomes show beneficial effects of RBT on urgency control, global perceived effect and lifestyle adaptations. Fifty percent of patients had a clinically relevant improvement, so referral for physiotherapy intervention should be considered before surgery.

INTRODUCTION

Faecal incontinence (FI) is defined as 'the complaint of involuntary loss of faeces' and affects 2–24% of community-dwelling adults.^{1,2} It is astonishing that patients regularly accept having FI, with only 5–27% seeking medical help.^{3,4} This may result from embarrassment, the erroneous belief that FI is a normal part of aging, or the perception that no treatment is available. However, established interventions for FI are available, such as dietary changes, medication, and physiotherapy.⁵ If medication or dietary adaptations offer no relief, physiotherapy intervention is an attractive alternative to surgery, because it is inexpensive, non-invasive, and without any adverse effects.⁶⁻⁸

Biofeedback, including rectal balloon training (RBT), and pelvic floor muscle training (PFMT) are elements of physiotherapy treatment. PFMT is recommended as an early intervention in FI treatment and is accepted as an effective intervention for urinary incontinence.⁹ The multifactorial nature of FI symptoms means that not every patient will benefit from PFMT to the same extent. Moreover, changes in sphincter strength are not necessarily linked to changes in symptoms. It is reported that rectal sensation may be more important than sphincter strength in attempts to relieve symptoms.¹⁰⁻¹²

RBT is used to improve rectal sensitivity by stepwise reductions in rectal balloon distension, in order to distinguish smaller rectal volumes, or resist urge by using progressive distension, or use a voluntary anal squeeze to counteract the recto-anal inhibitory reflex in response to rectal filling. Even though precise mechanisms responsible for improvement after PFMT or biofeedback interventions remain unclear, some have argued that rectal sensitivity training is the most important element of biofeedback.¹⁰⁻¹²

Lack of reliable, well-controlled research in the field of physiotherapy has hampered recommendations for pelvic physiotherapy interventions.⁷ Knowledge of effective physiotherapy elements enables guidance and improves referral patterns to pelvic physiotherapists, thereby preventing surgical treatment or wasting of other healthcare resources, and improving cooperation between physicians and physiotherapists. We hypothesised that a combined physiotherapy program with RBT and PFMT, aimed at treating the multifactorial origin of FI, would show more symptom relief. The purpose of this study was therefore to assess the effectiveness of RBT as an add-on therapy to PFMT in adults with FI.

METHODS

Study design and patients

In a two-armed parallel randomised, single-blind controlled trial patients were included from August 2006 to May 2009 at the Maastricht University Medical Centre, the Netherlands. An experienced colorectal surgeon consecutively included adult patients who reported having had FI for more than six months, with a Vaizey incontinence score ≥ 12 (range 0–24), and failure of dietary measures and medication. Patients

diagnosed with an anorectal tumour within the past two years, absent squeeze pressure of the anal sphincter, chronic diarrhoea (always fluid stool three or more times a day), overflow incontinence, proctitis, ulcerative colitis, Crohn's disease, soiling (defined as leakage of a minimal amount of watery faeces from the anal canal), previous ileo-anal or colo-anal anastomosis, and/or rectal prolapse in situ were excluded. Participants who had received physiotherapy during the previous six months or who were considered unable to comply with therapy were also excluded. Many patients appeared to have minor day- or night-time soiling. Patients with minor soiling were included as long as it was not due to overflow incontinence.

After signing informed consent, participants received either PFMT combined with RBT, or PFMT alone. The Medical Ethics Committee of the Maastricht University Medical Centre approved the study. A more detailed overview of this trial, registered as number ISRCTN78640169 (The Netherlands Trial Registry), was published elsewhere.¹³

Randomisation and masking

Patients were assigned in a 1:1 ratio with a random permuted block size of four. To conceal treatment allocation, an independent research assistant prepared a computer-generated randomisation list and treatment allocation. An independent physician performed enrolment. It was impossible to mask patients and therapists. To minimise bias, we ensured no involvement of therapists in the diagnostic and follow-up measurements, blinded double-data entry of baseline data and blinded primary outcome assessment.

Outcome measures

The primary outcome measure was the Vaizey grading system for FI severity, ranging from 0 (complete continence) to 24 (complete incontinence).¹⁴ Secondary outcome measures were the Fecal Incontinence Quality of Life Scale (FIQL),¹⁵ a nine-point global perceived effect score (GPE) (1=very much improved, 9=very much worse), anorectal manometry (resting pressure and squeeze pressure of the anal canal), rectal capacity measurement (sensory threshold, urge sensation, maximum tolerable volume), and thresholds of anorectal sensation.

All outcomes were measured at baseline and as soon as possible after completion of physiotherapy treatment. Baseline diagnostic work-up also included medical history taking, physical examination, endoanal ultrasound, and defecography.

Physiotherapy program

Nationwide, 90 physiotherapists working in private practices were trained to provide supervised physiotherapy close to the patients' home. Individual treatment was administered according to a standardised protocol, developed by clinicians and physiotherapists specialised in the field of pelvic floor disorders. Patients attended a maximum of 12 sessions within nine weeks. Sessions lasted 35 minutes and about 45 minutes in the group with rectal sensitivity training. Pelvic floor muscle assessment

following the PERFECT scheme was evaluated at baseline and at the final session.¹⁶ The assessment involved 'Power', evaluated by means of the modified Oxford grading scheme (0=no discernible muscle contraction, 5=strong muscle contraction), 'Endurance', defined as seconds (maximum 30) sustaining submaximum contraction, and 'Repetitions', as number (maximum five) of maximum contractions. Exercise regimens were patient-tailored and planned according to baseline assessments. To increase the chance of successful outcome, patients were instructed to practice the training program three times daily at home. Both groups received education and advice on anatomy, defecation mechanisms, and toilet behaviour.

Pelvic floor muscle training

PFMT consisted of selective voluntary contractions and relaxations of the pelvic floor muscles and the anal sphincter, practiced in different starting positions.¹³ Training aimed to maximise strength, improve duration of strength, and improve timing and coordination of contractions. Digital rectal examination was used to measure the ability of pelvic floor muscles to achieve maximum contraction and to quantify the strength. A contraction was considered sufficient if the pelvic floor and anal sphincter lifted inward and upward.

Change in neuromuscular function was promoted by basing PFMT on principles of overload and specificity, maintenance, and reversibility, similar to previously established recommendations for PFMT training dosage.¹⁷

Rectal balloon training

A rectal balloon attached to a syringe was introduced into the rectum and slowly inflated with air. Sensory threshold, urge sensation, and maximum tolerated volume were assessed. Patients with an insensitive rectum were trained to distinguish and respond to smaller rectal volumes of distension until a normal level of sensory threshold was achieved. In addition, patients with a hypersensitive rectum were trained to tolerate larger volumes by means of progressive distension and urge resistance, until a normal level of urge sensation was achieved. Coordination training tried to enhance the voluntary anal contraction in response to rectal filling, thereby counteracting the recto-anal inhibitory reflex, combined with reinforcement of rectal sensitivity and sustained external anal sphincter contractions to improve sphincter strength.

Statistical analyses

Missing values were checked prior to the analyses. A pre-calculated sample size of 106 participants was deemed sufficient to detect a 4.33 point difference in our primary outcome variable between groups ($\alpha=0.05$, $\beta=0.20$, one-sided).¹³

Statistical analyses were performed using SPSS software, version 17.0 (IBM Corporation, Somers, NY, USA). First, observed group differences were analysed using the independent samples t-test. Adjusted group differences were analysed using ANCOVA with post-intervention measurements as dependent variable and baseline measures as

covariates. Baseline to post-intervention comparisons were based on intention-to-treat analysis. In case of missing values, data were completed using the multiple imputation procedure, in which each missing value was replaced by a set of multiple values, estimated from regression models and available data. We identified 15 core factors, which were used as predictors for the missing values in each imputation model. We generated five multiple imputed datasets for each intervention group.^{18,19} The pooled results are presented, with a P-value of 0.05 indicating statistical significance.

RESULTS

Patients

The flow chart (Figure 6.1) shows the study flow after 101 patients had been assessed for eligibility. Eighty patients (79%) gave informed consent and were randomly assigned to either group 1 (RBT+PFMT, n=40) or group 2 (PFMT, n=40). Table 6.1 shows baseline demographic and clinical characteristics of the responders (n=80) vs. non-responders (n=20), showing a difference between the groups in time since onset of FI symptoms (P=0.043). After randomisation, groups were comparable in terms of baseline demographic and clinical characteristics (Table 6.2). No adverse events were reported for the subjects in each group. Ten patients (12.5%) dropped out during or after physiotherapy treatment. Completion rates did not differ between groups (P=0.31). After the intervention, patients were evaluated at a mean of 6.8 weeks (SD 5.3) after completing physiotherapy.

Table 6.1 Baseline demographic and clinical characteristics of responders and non-responders: % (n) unless otherwise stated

	Responders (n=80)		Non-responders (n=20)	
	%	n	%	n
Age (years) (mean; SD)	59.3 (11.9)	80	60.6 (10.2)	20
Gender (female)	90.0	72	95.0	19
Time since onset of FI symptoms (months) (mean; SD)	78.4 (99.5)	80	114.2 (90.5)	18 ^a
Vaizey score (mean; SD)	17.8 (2.8)	80	17.4 (3.1)	19 ^b
Nature of incontinence		80		19 ^b
Passive FI	10.0	8	5.3	1
Urge FI	33.8	27	47.4	9
Mixed FI	56.3	45	47.4	9

SD, standard deviation; FI, faecal incontinence.

^aTwo missing values.

^bOne missing value.

Figure 6.1 Flow chart describing enrolment, allocation, number of drop-outs and lost to follow-up.

Table 6.2 Baseline demographic and clinical characteristics: % (n) unless otherwise stated

	RBT + PFMT group (n=40)		PFMT group (n=40)	
	%	n	%	n
Age (years) (mean; SD)	58.3 (10.8)		60.2 (12.9)	
<45	10.0	4	10.0	4
45-54	17.5	7	17.5	7
>54	72.5	29	72.5	29
Gender (female)	90.0	36	90.0	36
BMI (mean; SD)	25.3 (4.6)		24.8 (3.4)	
Time since onset of FI symptoms (months) (mean; SD)	78.5 (104.9)		78.3 (95.1)	
Parity (mean number; SD)	2.1 (1.1)		2.3 (1.1)	
0	13.9	5	8.3	3
1	5.6	2	5.6	2
2	47.2	17	47.2	17
≥3	33.3	12	38.9	14
Vaizey score (mean; SD)	17.4 (3.0)		18.2 (2.6)	
Nature of incontinence				
Passive FI	10.0	4	10.0	4
Urge FI	32.5	13	35.0	14
Mixed FI	57.5	23	55.0	22
Origin				
Surgery	12.5	5	17.5	7
Delivery	10.0	4	7.5	3
Spontaneous	17.5	7	12.5	5
Gradually	7.5	3	7.5	3
Other	15.0	6	10.0	4
Unclear	37.5	15	45.0	18
Previous gynaecological surgery	58.3	21	41.7	15
Hysterectomy	47.2	17	41.7	15
Current constipating medication (yes)	20.0	8	15.0	6
IAS/EAS deficiency (yes)	52.5	21	42.5	17
IAS deficiency (yes)	27.5	11	25.0	10
EAS deficiency (yes)	42.5	17	30.0	12
Urinary incontinence (yes)	37.5	15	55.0	22
Co-morbid conditions (yes)				
Cardiovascular problems	22.5	9	25.0	10
Diabetes mellitus	7.5	3	10.0	4
Lung problems	12.5	5	5.0	2

RBT, rectal balloon training; PFMT, pelvic floor muscle training; SD, standard deviation; BMI, Body Mass Index; FI, faecal incontinence; IAS, internal anal sphincter; EAS, external anal sphincter.

Post-intervention follow-up

Table 6.3 shows the outcomes of post-intervention group comparisons. The adjusted mean difference on the Vaizey score was 1.19 point larger in group 1 than in group 2 (-5.58 vs. -4.39); this difference was not significant ($P=0.37$). Secondary outcome measures that improved in favour of group 1 were the mean GPE score (mean difference, -1.01; 95% confidence interval (CI), -1.75– -0.27; $P=0.008$), the Lifestyle subscale of the FIQL (mean difference, 0.37; 95% CI, 0.02–0.73; $P=0.039$), maximum tolerable volume (mean difference, 49.35 mL; 95% CI, 13.26–85.44; $P=0.009$), and fatigue of the external anal sphincter (mean difference, 0.65; 95% CI, 0.26–1.04; $P=0.001$). No significant differences were found in the total FIQL score and its Coping/Behaviour, Depression/Self perception or Embarrassment subscales, resting pressure, squeeze pressure, thresholds of anorectal sensation, the rectal distension volumes of sensory threshold and urge sensation, Oxford score of the external anal sphincter and pelvic floor, endurance of the external anal sphincter, or endurance and fatigue of the pelvic floor.

Figure 6.2 illustrates the change in Vaizey score in both groups after the intervention. Improvement rates (reduction in Vaizey change score ≥ 1) were 82.5% and 76% for groups 1 and 2, respectively ($P < 0.78$, Chi-square); 7.5% and 8.5% remained stable (Vaizey change score = 0) ($P < 1.00$, Chi-square), and 10% and 15.5% deteriorated (increase in Vaizey change score ≥ 1) ($P < 0.74$, Chi-square). 51% and 48% of patients were considered importantly improved based on the minimally important change, as determined before²⁰ (reduction in Vaizey change score ≥ 5) ($P < 1.00$, Chi-square). Six patients in group 1 and two in group 2 reported complete continence for gas, liquid, and solid stools, whereas eleven patients in group 1 and five in group 2 reported complete continence for liquid and solid stools.

The average time between first and final physiotherapy session was 12.4 weeks (SD 6.7); 33 patients (82.5%) completed 12 sessions in group 1, compared to 30 patients (75%) in group 2 ($P = 0.59$).

Figure 6.2 Change in Vaizey score in both groups after the intervention. MIC, minimally important change.

Table 6.3 Outcomes of group comparisons after intervention (intention-to-treat)^a

	Mean (SD) change from baseline		Mean difference (95% CI) in change from baseline as observed	Adjusted mean difference (95% CI) in change from baseline ^b	P-value
	RBT + PFMT group (n=40)	PFMT group (n=40)			
Vaizey score (range 0-24)	-5.50 (5.8)	-4.47 (5.0)	-1.03 (-3.59, 1.54)	-1.19 (-3.79, 1.42)	0.37
FIQL total (range 4-16)	1.67 (2.3)	0.79 (2.3)	0.88 (-0.28, 2.03)	1.01 (-0.07, 2.08)	0.066
Lifestyle subscale (range 1-4)	0.45 (0.8)	0.21 (0.8)	0.24 (-0.16, 0.63)	0.37 (0.02, 0.73)	0.039
Coping/Behaviour subscale (range 1-4)	0.52 (0.7)	0.20 (0.6)	0.32 (-0.03, 0.66)	0.32 (-0.02, 0.66)	0.063
Depression/Self perception subscale (range 1-4)	0.23 (0.6)	0.04 (0.7)	0.19 (-0.14, 0.52)	0.24 (-0.04, 0.53)	0.092
Embarrassment subscale (range 1-4)	0.47 (1.0)	0.34 (0.8)	0.13 (-0.35, 0.62)	0.14 (-0.32, 0.59)	0.55
Post-intervention GPE (range 1-9)	2.91 (1.4)	3.92 (1.7)	-1.01 (-1.75, -0.27)	-	0.008
Resting pressure (mmHg)	7.86 (22.1)	6.05 (23.3)	1.82 (-9.31, 12.95)	5.46 (-4.73, 15.64)	0.29
Squeeze pressure (mmHg)	6.26 (46.6)	10.20 (33.7)	-3.95 (-24.77, 16.88)	2.85 (-18.96, 24.65)	0.79
Threshold rectal sensation (mAmp)	1.94 (9.5)	1.23 (4.9)	0.70 (-5.03, 6.44)	1.65 (-3.12, 6.42)	0.48
Threshold anal sensation (mAmp)	-0.78 (6.8)	0.87 (7.4)	-1.65 (-5.90, 2.60)	-0.72 (-4.28, 2.84)	0.68
Sensory threshold (mL)	6.68 (39.6)	-0.13 (42.9)	6.80 (-17.12, 30.72)	8.57 (-16.26, 33.39)	0.48
Urge sensation (mL)	29.08 (60.3)	-0.93 (48.0)	30.00 (-6.77, 66.77)	32.37 (-8.85, 73.59)	0.11
Maximum tolerable volume (mL)	39.80 (77.8)	-3.28 (54.0)	43.08 (6.43, 79.72)	49.35 (13.26, 85.44)	0.009
Oxford score EAS (range 0-5)	0.52 (1.2)	0.93 (1.3)	-0.42 (-0.99, 0.16)	-0.25 (-0.78, 0.28)	0.36
Endurance EAS (range 0-30 sec)	10.47 (12.7)	7.34 (12.0)	3.13 (-3.71, 9.97)	1.40 (-5.14, 7.94)	0.66
Fatigue EAS (range 0-5)	1.39 (1.8)	0.24 (1.3)	1.15 (0.42, 1.88)	0.65 (0.26, 1.04)	0.001
Oxford score PF (range 0-5)	0.48 (1.2)	1.16 (1.4)	-0.68 (-1.31, -0.04)	-0.52 (-1.13, 0.09)	0.09
Endurance PF (range 0-30 sec)	12.14 (12.0)	11.48 (11.2)	0.67 (-5.39, 6.72)	-3.24 (-7.33, 0.86)	0.12
Fatigue PF (range 0-5)	1.44 (1.9)	1.10 (2.2)	0.34 (-0.74, 1.41)	-0.28 (-1.12, 0.57)	0.51

SD, standard deviation; RBT, rectal balloon training; PFMT, pelvic floor muscle training; CI, confidence interval; FIQL, Fecal Incontinence Quality of Life Scale; GPE, global perceived effect; EAS, external anal sphincter; PF, pelvic floor.

^aPooled results are presented.

^bAdjusted for the baseline value of the parameter.

DISCUSSION

This study assessed the effect of adding RBT to PFMT in patients with FI of mixed aetiology. We hypothesised that both groups would show symptom reduction, with group 1 having additional benefits from supplementary attention to rectal sensitivity training. Although this study provided no evidence for an add-on effect of RBT to PFMT, some of the secondary outcomes show beneficial effects of RBT on the control of urgency, external anal sphincter function, subjective rating of improvement, and lifestyle adaptations. Half of the patients demonstrated clinically important improvement on the Vaizey score, based on the minimally important change.²⁰

Several aspects should be taken into account when interpreting our results. Firstly, various outcome measures have been used to evaluate pelvic physiotherapy studies,^{14,21} and no outcome measures have a high level of scientific rigor, due to the lack of a criterion standard.²² We decided to use the Vaizey score as the primary outcome since it incorporates several items regarding severity of incontinence. Although the score correlates with patients' subjective perception of relief^{20,23} and has a significant relation with frequency of reported problems in general health domains,²⁴ its psychometric properties need further study.^{20,22} Furthermore, it is commonly reported in the literature that while outcomes measuring anatomical and functional characteristics give us an idea of physiological changes in response to treatment, doubts have been raised about their usefulness to evaluate treatment outcome.^{21,25} Therefore, patient's response in terms of subjective relief of FI symptoms is an attractive alternative as a primary outcome.^{21,26} Taking subjective improvement into account, our study found that RBT resulted in a significantly better outcome.

Secondly, the number of included patients was lower than anticipated, resulting in a power of 73.2%. Twenty patients were deemed eligible, but refused to participate for various reasons. Given that only a few colorectal surgeons in the Netherlands have specialised and experienced in this group of patients, patients sometimes had to travel far to visit our specialised centre (Maastricht University Medical Centre). In addition, only specialised physiotherapists with ample experience were included in this trial. Although 90 physiotherapists were trained nationwide to facilitate attendance of physiotherapy care sessions, this implied additional travel time in certain areas of the Netherlands. Although we had expected that enough patients would satisfy our inclusion criteria, practical and financial issues thus resulted in non-responders.

Furthermore, it was impossible to blind therapists or patients, which may have introduced bias. However, the questionnaires that asked participants for subjective judgments were completed at home, minimising interaction between patients and health care professionals.

Additionally, the percentage of missing post-intervention data was 12.5. We aimed to ensure valid imputation by generating five imputation datasets based on a comprehensive set of predictors. It is unlikely that this approach greatly influenced our results.

RBT is assumed to be able to restore anorectal sensory perception of rectal filling, resulting in earlier warning of pending defecation, urge resistance in patients with a hypersensitive rectum, improved rectal compliance, and improved coordination between rectal distension and pelvic floor muscle function. From this perspective, differences between the groups in rectal sensation threshold and sensory threshold were not as expected. By contrast, urge sensation and maximum tolerable volume increased in group 1, indicating better control of urge resistance.

Since we included a diverse patient group with passive, urge and mixed FI, we aimed to normalise the rectal capacity thresholds. Patients in group 1 received all elements of RBT, although emphasis was placed on those elements appropriate for each type of FI. The interpretation of changes in rectal distension volumes is complicated, since no consensus exists on standards of normal ranges, so conclusions about improvement at the individual level should be interpreted with caution. This was why we analysed at group level, which may have distorted effects since we included different types of FI. Rectal distension volumes may also have been influenced by the potential presence of rectal contents.

Fatigue reduction of the external anal sphincter was significantly greater in group 1. This may be explained by the additional coordination training in group 1, consisting of a forceful contraction of the external anal sphincter following rectal distension. The remaining outcomes of pelvic floor muscle assessment did not differ between the groups, confirming our expectations.

Some of the presumed mechanisms by which RBT reduces symptoms are inconsistent with our results and hypothesis. This may indicate loosely related proxy measures of symptom relief or lack of precise knowledge on putative mechanisms of RBT. The only mechanisms explaining the effect of RBT for which our results show evidence is that of control of urge resistance and forceful external anal sphincter contraction to counteract the recto-anal inhibitory reflex. It remains questionable whether these mechanisms are associated with symptom relief, as our primary outcome did not differ significantly between the two groups, although our results demonstrate symptom relief in terms of less lifestyle adaptations and better subjective improvement rating.

We can only speculate whether the number of sessions and total treatment period were sufficient to show a dose-response relation. This relation has already been established in patients with stress or mixed urinary incontinence, in that at least three months of supervised PFMT should be offered as a first-line approach.²⁷ Future research should elucidate whether increasing the number of sessions results in further improvement. This would provide stronger evidence for a dose-response relation and biological plausibility of physiotherapy treatment.

Our results are difficult to compare with other studies, due to differences in methodology and outcomes. A meta-analysis of 11 trials of biofeedback therapy in FI found no differences comparing biofeedback with non-biofeedback therapy (OR, 1.19; 95% CI, 0.69–2.05; $P=0.54$) or comparing various modes of biofeedback (OR, 1.28; 95% CI, 0.74–2.22; $P=0.38$).²⁸ Similar conclusions were reported by Norton et al., who found

that hospital-based computer-assisted sphincter pressure biofeedback yielded no greater benefit than advice or advice combined with instructions on sphincter exercises, for all outcomes.²⁹ Contrary to our findings, Heymen found that a program combining PFMT and biofeedback was more effective than PFMT alone.³⁰

Since the primary outcome did not differ between the groups, we also conducted combined group analyses. The results show that PFMT with or without RBT reduced the Vaizey score by a mean of 5.0 points (95% CI, -6.25– -3.72; $P=0.00$), equal to the estimated minimally important change.²⁰ Secondary outcomes also changed significantly from baseline ($P=0.00$), except for the physiological variables of squeeze pressure, anorectal sensation, and rectal distension volumes. Furthermore, a substantial proportion of the patients improved according to several definitions: 79.3% with a Vaizey score reduction of ≥ 1 , 71% with a GPE score of 1 to 4, and 20% being completely continent for liquid and solid stools. Our physiotherapy program resulted in 50.5% of the patients being importantly improved. It is not certain whether the observed improvement was solely due to the physiotherapy program, as we did not include a true control group reflecting patient expectations or the natural course of FI.²⁹ However, given the nature of our patient group, with a long history of moderate to severe FI symptoms, it seems unlikely that untreated patients would improve to the same extent. Based on this observation, we recommend physiotherapy first before proceeding to surgery.

In conclusion, our study provides no evidence for an add-on effect of RBT to PFMT, although some of the secondary outcomes show beneficial effects of RBT on the control of urgency, external anal sphincter function, subjective rating of improvement, and lifestyle adaptations. Selection of patients benefitting most from RBT remains to be confirmed in studies allowing subgroup analyses.

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CHAPTER 7

PREDICTORS OF A FAVOURABLE OUTCOME OF PHYSIOTHERAPY IN FAECAL INCONTINENCE: SECONDARY ANALYSIS OF A RANDOMISED TRIAL

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Submitted

ABSTRACT

Background

It is unclear which patients with faecal incontinence (FI) are likely to benefit from physiotherapy treatment, which is relevant for medical decision-making and patient counselling. This study aimed to identify patient characteristics predicting a favourable outcome of physiotherapy.

Methods

Eighty patients with FI, with a mean age of 59.3 (SD±11.9), were recruited at the Maastricht University Medical Centre and enrolled in a randomised controlled trial, which assessed the effect of adding rectal balloon training to pelvic floor muscle training. Treatment groups were combined for prediction modelling. Candidate predictors were obtained from patient demographics, clinical and physical examination, and questionnaires. Favourable outcome was defined as a combination of: (1) Vaizey score reduction ≥5 points, and (2) 'slightly' to 'very much improved' on the nine-point global perceived effect score. Predictors were identified by univariable and multivariable logistic regression analyses.

Results

Thirty-seven patients (46.3%) had a favourable outcome. Multivariable analysis showed that longer time since FI onset appeared to be associated with an unfavourable outcome (odds ratio [OR], 0.68; 95% confidence interval [CI], 0.47–1.00; $P=0.05$), whereas the use of constipating medication (OR, 3.79; CI, 0.99–14.51; $P=0.05$), any obstetric factor (OR, 2.15; CI, 0.94–4.89; $P=0.07$), and experiencing minor embarrassment (OR, 2.12; CI, 1.10–4.09; $P=0.03$) predicted a favourable outcome.

Conclusions

The small subset of identified predictors for a favourable outcome may be relevant in patient counselling and targeting physiotherapy treatment more efficiently, especially as they are available early in the diagnostic process.

INTRODUCTION

Faecal incontinence (FI) is the complaint of involuntary loss of faeces¹ and is associated with embarrassment, depression, and societal effects. A complex interplay of various physiological variables is involved in maintaining continence: recto-anal sensitivity, rectal capacity, stool consistency, cognition, and pudendal nerve and pelvic floor muscle function. FI is not restricted to elderly persons or nursing home residents, but also affects postpartum women, particularly as a result of third- or fourth-degree anal sphincter rupture.² Cross-sectional studies showed prevalence rates ranging from 2.2–25%, while underreporting is a common feature of this patient group due to embarrassment about seeking help or lack of awareness of available and effective interventions.³ The majority of patients who seek help respond favourably to physiotherapy treatment, with reported success rates of 49–72%.⁴ However, a Cochrane review of controlled studies of biofeedback and exercises for FI showed insufficient evidence to determine which patients are most likely to have a favourable outcome.⁵ Identifying patient characteristics that are amenable to change by physiotherapy treatment may prevent referral to surgical interventions, which are considered equally effective, but costly and burdensome because of their invasive nature and serious adverse events.⁶ Identifying predictors based on pre-treatment assessments is essential to support early clinical decision-making and may lead to more realistic expectations of the outcome, as well as a more efficient use of resources and referral. Only a few studies have examined which patient characteristics are likely to predict the greatest benefit from physiotherapy treatment. Conflicting data has been reported on the predictive value of age, sex, symptom severity, underlying cause, manometric data, sphincter lesions, and pudendal neuropathy, due to heterogeneity of study populations, outcome measures, treatment methods, and definition of favourable outcome.^{7,8}

If individual patient characteristics can be related to treatment response, physiotherapy treatment can be made more effective by using targeted interventions. We recently completed a two-armed randomised controlled trial examining the effect of rectal balloon training as an add-on therapy to pelvic floor muscle training.⁹ We performed secondary analyses aimed at identifying baseline characteristics that predict a favourable outcome of physiotherapy treatment for patients with FI.

PATIENTS AND METHODS

We report on secondary analyses of data from a randomised controlled trial conducted between August 2006 and May 2009. An extensive description of the design is provided elsewhere and summarised below.⁹

Patients

Eighty consecutive adults with FI, recruited at the Maastricht University Medical Centre in the Netherlands, were randomly assigned to receive rectal balloon training as an add-on therapy to pelvic floor muscle training (n=40) or pelvic floor muscle training alone (n=40).⁹ Inclusion criteria were: FI complaints lasting at least six months, Vaizey incontinence score of at least 12, and failure of medication and dietary adaptations. Exclusion criteria were: having been diagnosed with an anorectal tumour within the past two years, absent squeeze pressure of the anal sphincter, chronic diarrhoea (always fluid stools, three or more times a day), overflow incontinence, proctitis, colitis ulcerosa, Crohn's disease, previous ileo-anal or colo-anal anastomosis, and rectal prolapse in situ. Participants who had received physiotherapy during the previous six months or who were considered unable to understand instructions were also excluded. The Medical Ethics Committee of the Maastricht University Medical Centre approved the study.

Selecting distinct predictive factors based on literature reports in advance was not feasible due to conflicting conclusions of the articles. We therefore included a wide range of candidate predictors obtained from patient demographics, clinical and physical examination, and questionnaires at baseline (Appendix).

Selecting predictive factors

Medical history and physical examination

The medical history and physical examination were obtained by one physician and included demographics, symptoms of incontinence (onset, type and degree of FI, associated urinary incontinence, and underlying causes), bowel habits (stool consistency and constipation), previous history (systemic diseases, any urological, neurological, anorectal, or metabolic factor), and any obstetric factor (including high birth weight ($\geq 4,000$ gr), episiotomy, instrumental delivery, prolonged second stage of labour (>2 h), and breech delivery). Rupture after vaginal delivery was separately evaluated as independent predictor. The primary outcome measure was the Vaizey grading system, ranging from 0 (complete continence) to 24 (complete incontinence).¹⁰ This severity score assesses the frequency of loss of gas, liquid stool, and solid stool, the ability to defer defecation for 15 minutes, alterations in lifestyle, and the use of pads and constipating medication. To minimise bias, double data entry was used for the medical history and physical examination.

Anorectal functional tests

Anorectal function was evaluated with the following series of tests: anorectal manometry, rectal capacity measurement, anorectal sensation, anal endosonography, and defecography.

Questionnaires

Condition-specific quality of life was measured with the Fecal Incontinence Quality of Life (FIQL) Scale, ranging from 4 to 16 and comprising four subscales: Lifestyle (10 items), Coping/Behaviour (9 items), Depression/Self Perception (7 items), and Embarrassment (3 items).¹¹ A higher score indicates a better quality of life. We also asked patients to score their perceived change after the treatment, using a nine-point global perceived effect score (GPE), ranging from: 1=very much improved to 9=very much worse. These questionnaires were completed at home, ensuring unbiased results as the physician and physiotherapist were unaware of the responses.

Physiotherapy treatment

After signing informed consent and randomisation, patients received a standardised course of a total of 12 treatment sessions within nine weeks. Pelvic floor muscle training included improving awareness of the pelvic floor and anal sphincter, strength and endurance training, and home exercises. Exercises also comprised simulating daily life activities. Rectal balloon training was done according to the sensory and coordination training protocol.^{4,5} Participating physiotherapists were instructed by means of a booklet and a written protocol, and an educational meeting in which we showed them an instruction film on rectal balloon training. Baseline pelvic floor muscle assessment included the Oxford grading scheme,¹² endurance (submaximum contraction up to 30 seconds), and fatigue (maximum contraction up to five repetitions) of the external anal sphincter and pelvic floor, in addition to rectal capacity measurements.

Outcome

No single test or score exists that can benchmark favourable treatment of FI, due to the lack of a criterion standard.¹³ Patient improvement after treatment was selected as favourable outcome, representing the dependent variable in the predictive model. In agreement with other studies,¹⁴ favourable outcome was defined as the following combination: (1) a reduction of the Vaizey score by at least five points, and (2) 'slightly' to 'very much improved' on the GPE score. The first criterion is derived from a study by the same authors who reported an estimate for the minimally important change in the Vaizey score.¹⁵

Statistical analyses

Missing predictive and outcome variables were completed using the multiple imputation procedure, in which each missing value was replaced by a set of multiple values, estimated from regression models and available data. We generated five multiple imputed datasets, separately for each intervention group.^{16,17} The pooled results are presented.

We explored the assumption of linearity between the continuous predictors and the change in Vaizey score and GPE using visual inspection. In case of non-linearity, the continuous variables were transformed to better approach linearity. Multicollinearity

was examined using the variance inflation factor (VIF) and the reciprocal tolerance statistic ($1/\text{VIF}$). VIF values indicate whether a predictor has strong linear associations with the other predictors. The average VIF value of all predictors should not be substantially larger than 1 and the reciprocal tolerance statistic should not be lower than 0.2.¹⁸

The associations between the outcome immediately after treatment and each of the candidate predictors were analysed using univariable logistic regression analyses. We used the Akaike Information Criterion, which corresponds to selection of predictors at a 0.157 significance level.¹⁹ Univariable odds ratios (OR) and their corresponding 95% confidence interval were calculated. Selected predictors derived from univariable logistic regression were included in the multivariable logistic regression model.

For the multivariable analyses, a three-stage model was used. The initial prediction model included demographic and medical history variables available at the time the patients first visited the physician. The second model examined the added effect of anorectal functional tests in predicting the outcome of physiotherapy. Finally, the third model included variables available at the time the patient was referred to a physiotherapist. Hence, models 1 and 2 provide physicians with information before referral to physiotherapy, whereas model 3 examines the added effect of variables from physiotherapy diagnostics. To evaluate the influence of treatment allocation, we added this variable to the final set of predictors (model 4).

A forced entry approach with a significance level of 0.10 was used to build each multivariable model. A liberal P-value increases the chances of obtaining true predictors. Regression coefficients and related standard errors and P-values were then estimated across all multiply imputed datasets using Rubin's Rules.²⁰ Analyses were performed using SPSS software, version 17.0 (IBM Corporation, Somers, NY, USA). Reported P-values are two-tailed.

RESULTS

Patients

Eighty patients, predominantly female (90%), were included, with a mean age of 59.3 (SD±11.9). Ten patients (12.5%) dropped out before or during physiotherapy treatment. Patients were evaluated at a mean of 6.8 weeks (SD 5.3) after completing physiotherapy. Patients reported median of four years having elapsed since the onset of FI symptoms (interquartile range 2 to 6). The mean baseline Vaizey score changed from 17.8 (SD±2.8) to 12.8 (SD±5.9) after treatment, an average reduction of 5.0 points (95% CI, -6.3 to -3.7). A reduction in Vaizey score of at least five points was achieved by 50.5% of treated patients, while 71% had a 'slightly' to 'very much improved' GPE score. Favourable outcome, defined as the combination of these two outcomes, was achieved by 37 patients (46.3%).

Univariable analyses of response to physiotherapy treatment

Candidate predictors were first analysed using univariable logistic regression (Appendix). The results for predictors with a P-value <0.157 are summarised in Table 7.1. We identified ten factors predicting outcome of physiotherapy treatment. The following predictors, obtained from demographics and medical history, were associated with a favourable outcome (positive regression coefficient (B) and OR >1.0): a higher total score on the FIQL and its Embarrassment subscale, use of constipating medication, and any obstetric factor. Factors associated with an unfavourable outcome (negative B and OR <1.0) were longer time since onset of FI, liquid stool incontinence, and no distinction between flatus and faeces. Predictors obtained from physical examination were not associated with a favourable or unfavourable outcome. Only one variable from the anorectal functional tests predicted the outcome of physiotherapy: a higher urge sensation threshold was associated with an unfavourable outcome. Finally, two predictive factors from physiotherapy diagnostics were identified: better results for endurance and fatigue of the pelvic floor were associated with an unfavourable outcome.

Table 7.1 Association between demographics and medical history, anorectal functional tests, physiotherapy diagnostics and favourable outcome after treatment: n (%), unless otherwise stated

	n (%)	B (SE)	P	OR	95% CI
Demographics and medical history					
FIQL total score (mean; SD) (range 4-16)	9.4 (2.6)	0.14 (0.09)	0.13	1.15	0.96–1.38
Embarrassment subscale (mean; SD) (range 1-4)	2.1 (0.8)	0.62 (0.30)	0.04	1.86	1.04–3.33
Current constipating medication (yes)	14 (17.5)	0.89 (0.62)	0.15	2.44	0.73–8.19
Any obstetric factor	40 (55.6)	0.77 (0.49)	0.12	2.15	0.82–5.63
Longer time since onset of FI symptoms (mean; SD) (m)	78.4 (99.4)	−0.28 (0.17)	0.09	0.75	0.54–1.04
Liquid stool incontinence (≥ 1/m)	79 (98.8)	−0.41 (0.28)	0.14	0.66	0.38–1.15
No distinction flatus/faeces (≥ 1x/m)	42 (52.5)	−0.25 (0.14)	0.07	0.78	0.59–1.02
Anorectal functional tests					
Urge sensation (mean; SD) (ml)	103.2 (47.1)	−0.01 (0.01)	0.12	0.99	0.98–1.00
Physiotherapy diagnostics					
Endurance pelvic floor (mean; SD) (range 0-30 sec)	14.3 (10.9)	−0.04 (0.02)	0.08	0.96	0.92–1.01
Fatigue pelvic floor (mean; SD) (range 0-5)	3.6 (1.7)	−0.29 (0.15)	0.06	0.75	0.56–1.01

B (SE), estimate with corresponding standard error; OR, odds ratio; CI, confidence interval; FIQL, Fecal Incontinence Quality of Life Scale; FI, faecal incontinence; m, months; sec, seconds; predictors of favourable outcome shown in bold.

Multivariable analyses of response to physiotherapy treatment

Variables with a P-value <0.157 based on univariable analyses were included in the multivariable logistic regression model. The initial multivariable model (model 1 from the three-stage modelling) included all predictors identified from demographics and medical history (Table 7.2). Longer time since onset of FI, a higher score on the Embar-

rassment subscale of the FIQL, use of constipating medication, and any obstetric factor independently appeared to predict response and therefore remained in the model. In the second model, we added the significant predictor from the anorectal functional tests, i.e. urge sensation threshold, to the variables established in the first model, which did not add to the association with treatment response. Next, significant predictors from physiotherapy diagnostics (fatigue and endurance of the pelvic floor) were added to the variables determined in the first model, but yielded no additional predictors. After adding group allocation in the final multivariable model (model 4), the variables established in the first model remained significant. Ultimately, longer time since onset of FI proved to predict an unfavourable outcome (B, -0.38; OR, 0.68; P=0.05), whereas the use of constipating medication (B, 1.33; OR, 3.79; P=0.05), any obstetric factor (B, 0.77; OR, 2.15; P=0.07), and a higher score on the Embarrassment subscale (B, 0.75; OR, 2.12; P=0.03) proved to predict a favourable outcome. The Embarrassment subscale of the FIQL contains items with regard to feeling ashamed, worry about others smelling stool, and the concern about leaking stool without even knowing it.

Table 7.2 Multivariable logistic regression analyses

	Model 1			Model 2			Model 3			Model 4		
	B (SE)	P		B (SE)	P		B (SE)	P		B (SE)	P	
Longer time since onset of FI symptoms (m)	-0.49 (0.22)	0.03		-0.40 (0.19)	0.04		-0.56 (0.23)	0.02		-0.38 (0.19)	0.05	
Embarrassment subscale	1.13 (0.50)	0.03		0.67 (0.34)	0.05		0.97 (0.40)	0.02		0.75 (0.34)	0.03	
Current constipating medication	1.35 (0.70)	0.05		1.36 (0.69)	0.05		1.33 (0.77)	0.08		1.33 (0.69)	0.05	
Any obstetric factor	0.95 (0.45)	0.04		0.93 (0.44)	0.04		0.95 (0.47)	0.04		0.77 (0.42)	0.07	
Liquid stool incontinence	-0.46 (0.36)	0.19										
No distinction flatus/faeces	-0.22 (0.16)	0.16										
FIQL total score	-0.21 (0.17)	0.22										
Urge sensation (ml)				-0.01 (0.01)	0.12							
Endurance pelvic floor							-0.05 (0.04)	0.17				
Fatigue pelvic floor							-0.27 (0.23)	0.23				
Group allocation										-0.24 (0.53)	0.65	

B (SE), estimate with corresponding standard error; FI, faecal incontinence; m, months; FIQL, Fecal Incontinence Quality of Life Scale.

DISCUSSION

The aim of this study was to assess which independent factors can predict a favourable outcome of physiotherapy treatment for patients with FI. The total FIQL score was only associated with favourable outcome in univariable analysis. By contrast, longer time since onset of FI symptoms appeared to be associated with an unfavourable outcome in multivariable analysis, whereas liquid stool incontinence, no distinction between flatus and faeces, urge sensation threshold and fatigue and endurance of the pelvic floor proved to predict an unfavourable outcome in univariable analysis. The multivariable analyses showed that three predictive factors were associated with a favourable

outcome: the Embarrassment subscale of the FIQL score, the use of constipating medication, and any obstetric factor.

Our findings indicate that only a small subset of predictive factors is important in predicting favourable or unfavourable physiotherapy outcome. Only predictive factors obtained from the medical history were associated with response to treatment. Results of physical examination, diagnostic tests, and physiotherapy assessment did not provide additional predictive factors, indicating that patients who are suitable candidates for physiotherapy treatment can be identified in an early stage of the diagnostic process. Early knowledge of factors predicting a favourable or unfavourable outcome of physiotherapy is useful for medical decision making and patient counselling.

Potential study limitations should be taken into account. Predictors of favourable outcome of physiotherapy treatment are inconsistently reported in the literature, which complicated pre-specification of the most important predictors to be included in the prediction model. To overcome this problem, we initially tested the association between each potential predictor and the outcome. Selection of predictors was based on the Akaike Information Criterion, which uses a liberal P-value. This limits the possibility that a true predictor would be missed.

Symptom severity and subjective perception of improvement do not seem to correlate.²¹ Therefore, we defined favourable outcome as a combination of subjectively reported improvement and a reduction in the Vaizey score, which means we were stricter than most other studies, which used only one criterion.^{22,23} This reduced the reported number of patients with a favourable outcome.

As a rule of thumb, multivariable logistic regression models should be used with a minimum of ten outcome events (i.e. favourable outcome) per predictor variable.²⁴ In our study, events per variable ranged between 5.3 (model 1) to 7.4 (model 2 and 4). However, others consider this rule too conservative and conclude that this rule can be relaxed, based on the finding that model performance problems were uncommon with 5–9 events per variable.²⁵

The generalisability of the predictive factors is limited by our study's stringent inclusion and exclusion criteria. The population under study included patients with moderate to severe FI complaints, who were unresponsive to medical or dietary measures. The predictive factors need further exploration for use in other FI patients not represented in this study.

To our knowledge, embarrassment as a predictive factor has never been studied before. Less embarrassment was associated with a favourable outcome of physiotherapy treatment. It is possible that compliance, motivation, cooperation, and expectations from patients who are less embarrassed are different, and this may have influenced treatment outcome. Since the degree of embarrassment was not related to the time since the onset of symptoms (Pearson $r=-0.046$, $P=0.69$), the association was not explained by patients seeking help at an earlier stage and the prevention of symptom worsening. Degree of embarrassment was also not related to the severity of FI symptoms (Vaizey score) prior to physiotherapy treatment (Pearson $r=0.093$, $P=0.42$), so a

favourable outcome of physiotherapy treatment for less embarrassed patients is unlikely to be explained by mild symptoms.

The use of constipating medication was associated with favourable outcome of physiotherapy treatment. We recently determined that the use of constipating medication barely changed following physiotherapy treatment.¹⁵ This may indicate that the use of constipating medication is associated with a patient's self-confidence and a more favourable coping strategy, rather than with symptom severity.

The variable 'any obstetric factor' appeared to predict a favourable outcome. Obstetric factors included high birth weight ($\geq 4,000$ gr), episiotomy, instrumental delivery, prolonged second stage of labour (>2 h), and breech delivery. One or more obstetric factors were reported by 55.6% of the included women. These factors may result in perineal, anal sphincter, or nerve damage. Patients with one or more obstetric factors seem to have the potential to benefit from physiotherapy treatment, which is a promising finding in view of the large group of patients having obstetric factors. Rupture after vaginal delivery was not included in the 'any obstetric factor' predictive factor, as there was a high prevalence in our patient group (66.7%). This factor was found to have a favourable but non-significant association with treatment outcome in univariable analysis. This is in agreement with previous studies,^{7,22,23,26,27} although two studies found that biofeedback was most beneficial in patients with structurally intact sphincter muscles.^{28,29} Unfortunately, we were unable to differentiate between degrees of rupture, which hampered our interpretation for patients with severe ruptures. As third- and fourth-degree ruptures are an important aetiological factor for postpartum FI,² knowing whether these patients will benefit from physiotherapy treatment is of great interest.

In contrast to other studies,^{7,23,27} longer time since onset of symptoms was associated with an unfavourable outcome of physiotherapy treatment. This may be explained by the prior ineffective attempts to treat the disorder, like failure of medication and/or dietary adaptations as defined by our inclusion criteria, resulting in decreased motivation and possibly poor outcome. Symptoms may also worsen if patients seek help only after years.

We did not find an association between demographics (age or gender) and outcome, whereas one study found that treatment success was more likely in women,¹⁴ and a few studies have reported a positive association between favourable treatment outcome and older age.^{14,22,23} In line with most studies, we found that the physiological variables obtained from anorectal manometry, rectal capacity measurements, endo-anal ultrasound, defecography, and anorectal sensory thresholds had no predictive value.^{23,27,30,31} Nevertheless, a few studies reported better outcomes for patients with a higher squeeze pressure^{7,27,32} and lower rectal sensory thresholds.³⁰ Hence, our results add evidence to the belief that test results may be proxy measures of symptoms, but at the same time attempt to explain the underlying pathophysiology of FI.⁷ Interpretation of the association between physiological variables and favourable treatment outcome is further impeded by the substantial variation in test protocols.

We did not find an association between the severity of symptoms and the outcome of physiotherapy treatment, as was reported by other authors,^{22,23,26,30,31} although some authors reported an association between good clinical response and mild symptomatology^{7,27} or severe symptomatology.¹⁴

Identifying patients who are likely to benefit most from physiotherapy is crucial for clinical practice. Our study provides a small subset of factors that independently predict favourable outcome of physiotherapy treatment in patients with moderate to severe FI. Moreover, we considered various predictive factors that have never been considered before. These predictors may be relevant in patient counselling and targeting physiotherapy treatment more efficiently, especially as they are available early in the diagnostic process. Future research is warranted to find out if selecting patients for physiotherapy treatment based on the predictors found in this study may result in better outcomes. In addition, more studies are needed to confirm these predictors in other settings and in patients not represented in this study, such as mild symptomatology.

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APPENDIX

Appendix Association between demographics, medical history, physical examination, baseline tests, physiotherapy diagnostics and favourable outcome following physiotherapy: n (%), unless otherwise stated

Demographics	n (%)	B (SE)	P	OR	95% CI
Age (y) (mean; SD)	59.3 (11.9)	0.02 (0.02)	0.23	1.02	0.98–1.07
Gender (female)	72 (90)	−0.17 (0.82)	0.83	0.83	0.24–5.97
BMI (mean; SD)	25.1 (4.0)	0.00 (0.06)	0.94	1.00	0.89–1.11
Vaizey baseline (mean; SD) (range 12–24)	17.8 (2.8)	0.00 (0.08)	0.97	1.00	0.85–1.18
FIQL total score (mean; SD) (range 4–16)	9.4 (2.6)	0.14 (0.09)	0.13	1.15	0.96–1.38
Subscale 1 Lifestyle (mean; SD) (range 1–4)	2.7 (0.9)	0.09 (0.27)	0.74	1.09	0.64–1.87
Subscale 2 Coping/behaviour (mean; SD) (range 1–4)	1.9 (0.7)	0.43 (0.33)	0.20	1.53	0.80–2.95
Subscale 3 Depression/Self Perception (mean; SD) (range 1–4)	2.7 (0.8)	0.42 (0.31)	0.19	1.51	0.82–2.80
Subscale 4 Embarrassment (mean; SD) (range 1–4)	2.1 (0.8)	0.62 (0.30)	0.04	1.86	1.04–3.33
Medical history					
Onset of FI symptoms (mean; SD) (m)	78.4 (99.4)	−0.28 (0.17)	0.09	0.75	0.54–1.04
Presence of urge FI	27 (33.8)	0.57 (0.52)	0.28	1.76	0.64–4.89
Presence of passive FI	8 (610)	−1.04 (0.85)	0.22	0.35	0.07–1.88
Presence of mixed FI	45 (56.3)	−0.17 (0.49)	0.73	0.85	0.33–2.20
Frequency defecation (<7 times/week)	10 (12.5)	−0.79 (0.73)	0.28	0.45	0.11–1.91
Thin stool consistency	4 (5)	−0.85 (1.29)	0.51	0.43	0.03–5.35
Soft mushy stool consistency	38 (47.5)	0.24 (0.47)	0.62	1.27	0.50–3.20
Solid stool consistency	24 (30)	0.02 (0.50)	0.96	1.03	0.38–2.75
Varying stool consistency	13 (16.3)	0.00 (0.61)	0.99	1.00	0.30–3.34
Mucus loss during defecation (≥1/m)	19 (23.8)	−0.07 (0.19)	0.72	0.94	0.65–1.35
Pad use (≥1/m)	70 (87.5)	−0.12 (0.17)	0.48	0.89	0.63–1.24
Flatus incontinence (≥1/m)	71 (88.8)	−0.18 (0.18)	0.32	0.84	0.60–1.18
Liquid stool incontinence (≥1/m)	79 (98.8)	−0.41 (0.28)	0.14	0.66	0.38–1.15
Solid stool incontinence (≥1/m)	62 (77.5)	0.02 (0.18)	0.91	1.02	0.72–1.45
Social restriction (≥1/m)	78 (97.5)	0.09 (0.28)	0.75	1.09	0.63–1.89
Current constipating medication	14 (17.5)	0.89 (0.62)	0.15	2.44	0.73–8.19
Sensation of incomplete evacuation (≥1/m)	55 (68.8)	−0.07 (0.14)	0.64	0.94	0.72–1.22
Sensation of anal prolapse (≥1/m)	11 (13.8)	0.04 (0.22)	0.87	1.04	0.68–1.59
Loss of large amount (≥1/m)	34 (42.5)	−0.08 (0.17)	0.66	0.93	0.67–1.29
No distinction flatus/faeces (≥1/m)	42 (52.5)	−0.25 (0.14)	0.07	0.78	0.59–1.02
Urinary incontinence (≥1/m)	37 (46.3)	0.10 (0.13)	0.45	1.10	0.86–1.42
Constipation	21 (26.3)	−0.19 (0.53)	0.72	0.83	0.29–2.34
Parity (0,1,2,≥3)	2.2 (1.1)	−0.03 (0.26)	0.92	0.97	0.59–1.62
Rupture after vaginal delivery	48 (66.7)	0.30 (0.52)	0.56	1.35	0.49–3.72
Any obstetric risk factor	40 (55.6)	0.77 (0.49)	0.12	2.15	0.82–5.63
Any ano- and colorectal risk factor (e.g. surgery for haemorrhoids, fistulas)	23 (28.8)	0.58 (0.51)	0.26	1.79	0.65–4.90
Any gynaecological risk factor (e.g. hysterectomy)	36 (50)	0.18 (0.48)	0.71	1.20	0.47–3.05
Any urological risk factor (e.g. Burch operation)	12 (15)	0.74 (0.69)	0.28	2.10	0.55–8.03
Any neurological risk factor (e.g. hernia, TIA)	11 (13.8)	−0.95 (0.72)	0.19	0.39	0.09–1.59
Any metabolic risk factor (e.g. diabetes mellitus, thyroid disorders)	9 (11.3)	0.84 (0.76)	0.27	2.32	0.52–10.39
Co-morbidity (cardiovascular or lung disease)	26 (32.5)	0.32 (0.50)	0.53	1.37	0.52–3.64

	n (%)	B (SE)	P	OR	95% CI
Physical examination					
Perineal and/or perianal scar tissue	19 (23.8)	0.22 (0.54)	0.68	1.25	0.43–3.58
Impaired sensibility	8 (10)	0.17 (0.84)	0.84	1.18	0.23–6.17
Paradoxal squeezing	17 (21.3)	−0.34 (0.62)	0.58	0.71	0.21–2.40
Defect anal sphincter complex	9 (11.3)	−0.08 (0.72)	0.91	0.92	0.23–3.75
Rectocele	9 (11.3)	−0.61 (0.75)	0.42	0.54	0.13–2.37
Baseline tests					
Resting pressure (mean; SD) (mmHg)	38.1 (22.4)	0.00 (0.01)	0.92	1.00	0.98–1.02
Squeeze pressure (mean; SD) (mmHg)	83.7 (42.4)	0.00 (0.01)	1.00	1.00	0.99–1.01
Threshold rectal sensation (mean; SD) (mAmp)	7.4 (5.1)	−0.04 (0.06)	0.44	0.96	0.86–1.07
Threshold anal sensation (mean; SD) (mAmp)	8.5 (4.8)	−0.02 (0.06)	0.69	0.98	0.87–1.01
Sensory threshold (mean; SD) (ml)	47.7 (26.6)	−0.01 (0.01)	0.20	0.99	0.97–1.01
Urge sensation (mean; SD) (ml)	103.2 (47.1)	−0.01 (0.01)	0.12	0.99	0.98–1.00
Maximal tolerable volume (mean; SD) (ml)	156.1 (60.3)	−0.01 (0.00)	0.23	1.00	0.99–1.00
Presence anterior rectocele	40 (55.6)	0.14 (0.54)	0.80	1.15	0.40–3.31
Presence of intussusception/rectumprolapse	30 (37.5)	0.58 (0.54)	0.29	1.79	0.61–5.20
Presence of entero- or sigmocele	6 (7.5)	−0.57 (1.04)	0.58	0.56	0.07–4.33
Presence of EAS deficiency	29 (36.3)	−0.22 (0.49)	0.65	0.80	0.31–2.09
Presence of IAS deficiency	21 (26.3)	−0.45 (0.55)	0.41	0.64	0.22–1.86
Presence EAS or IAS deficiency	38 (47.5)	−0.30 (0.46)	0.51	0.74	0.30–1.84
Presence of anismus	23 (28.8)	0.32 (0.58)	0.59	1.37	0.44–4.30
Physiotherapy diagnostics					
Paradoxal lifting	10 (12.5)	0.21 (0.85)	0.81	1.23	0.23–6.47
Paradoxal straining	3 (3.8)	−0.27 (1.25)	0.83	0.76	0.07–8.85
Pain during palpation	19 (23.8)	−0.32 (0.57)	0.58	0.73	0.24–2.22
Oxford score EAS (median)	2.1 (1.0)	0.09 (0.23)	0.69	1.10	0.69–1.73
Endurance EAS (range 0–30 sec)	12.7 (10.9)	−0.02 (0.02)	0.49	0.99	0.95–1.03
Fatigue EAS (range 0–5)	3.8 (1.7)	−0.19 (0.14)	0.17	0.82	0.62–1.09
Oxford score PF (median)	2.4 (1.1)	0.03 (0.25)	0.92	1.03	0.63–1.66
Endurance PF (range 0–30 sec)	14.3 (10.9)	−0.04 (0.02)	0.08	0.96	0.92–1.01
Fatigue PF (range 0–5)	3.6 (1.7)	−0.29 (0.15)	0.06	0.75	0.56–1.01
Sensory threshold (mean; SD) (ml)	31.4 (21.4)	0.00 (0.02)	0.72	1.00	0.97–1.04
Urge sensation (mean; SD) (ml)	89.1 (44.5)	0.00 (0.01)	0.56	1.00	0.98–1.01
Maximal tolerable volume (mean; SD) (ml)	178.3 (69.2)	0.00 (0.01)	0.59	1.00	0.99–1.01

B (SE), regression coefficient with corresponding standard error; OR, odds ratio; 95% CI, 95% confidence interval; y, years; SD, standard deviation; BMI, body mass index; FIQL, Fecal Incontinence Quality of Life Scale; FI, faecal incontinence; m, months; EAS, external anal sphincter; IAS, internal anal sphincter; PF, pelvic floor.

CHAPTER 8

A CRITICAL EVALUATION OF THE VAIZEY SCORE, WEXNER SCORE AND THE FECAL INCONTINENCE QUALITY OF LIFE SCALE FOR CLINICAL USE IN PATIENTS WITH FAECAL INCONTINENCE

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ABSTRACT

Purpose

This study aims to assess the psychometric properties and minimally important change (MIC) of the Vaizey score, Wexner score and the Fecal Incontinence Quality of Life Scale (FIQL) for use in the evaluation of patients with faecal incontinence (FI).

Methods

Eighty patients with FI with a mean age of 59.3 ($SD \pm 11.9$) were enrolled in a randomised controlled trial. The patient-reported outcomes were tested for internal and external responsiveness, longitudinal construct validity and interpretability.

Results

All total scores proved to have both adequate to excellent responsiveness and longitudinal construct validity, and changes were in agreement with subjective improvement. Due to variability in MIC estimates, they should be used as indicators. All patient-reported outcomes showed psychometric or practical limitations.

Conclusions

The instruments available to date to evaluate severity and quality of life in FI do not yet attain the highest levels of psychometric soundness. As the focus of patients may differ from that of physicians, it is recommended to include several measures for evaluation. So far, there are suggestions that the Wexner score is most suitable for severity assessment and the FIQL for evaluating quality of life.

INTRODUCTION

Faecal incontinence (FI) is defined as the complaint of involuntary loss of faeces.¹ Affected patients often suffer in silence and only one-third seeks medical advice.² Reporting severity of FI and its consequences on quality of life (QoL) are of great relevance, although no consensus exists on the optimal patient-reported outcome (PRO).³ In evaluating a patient, one should assess the patients' subjective perception of improvement for example by using the global perceived effect score.⁴⁻⁶ Furthermore, grading scales assessing type of stool loss are in general use, however lack information on frequency of bowel accidents and are therefore unable to discriminate between patients with small differences.^{7,8} Other multi-item scoring systems have emerged,⁹⁻¹³ of which the Vaizey score¹⁰ and Wexner score⁹ are most often used as PRO in physiotherapy trials.¹⁴⁻²⁰ Both scales combine items regarding frequency and type of stool loss, pad use and alteration in lifestyle. Additionally, the Vaizey score assesses the ability to defer defecation and the use of constipating medication, and reduces the emphasis on the need to wear a pad.¹⁰

Since FI is a major stressor in life, QoL outcomes are frequently used in evaluating FI. The Fecal Incontinence Quality of Life Scale (FIQL) is a condition-specific QoL questionnaire²¹ and more commonly used as secondary outcome in evaluating patients with FI.^{17,18,21,22} Besides evaluating treatment, PROs may be of use with regard to clinical decision making as well as communication among health providers and involved insurance companies.

However, the application of the Vaizey and Wexner scores and the FIQL in standard practice is hampered since the psychometric properties, in terms of validity, reliability and responsiveness have not been rigorously evaluated before. Avery et al. concluded that the Vaizey and Wexner score were in the early stage of development, and the FIQL was recommended as some validity, reliability and responsiveness was indicated.²⁰ In the meantime, efforts from our research group to validate the Vaizey score in patients undergoing physiotherapy treatment resulted in the conclusion that the psychometric properties of the Vaizey score are inconclusive.²³

Additionally, it is important that the numerical value of a PRO is interpretable. Thus far, it is unclear which change scores are minimally important, which is of significance in calculating the sample size of a trial and interpreting results of physiotherapy effectiveness studies. We performed secondary analyses on a recently completed randomised controlled trial²⁴ which evaluated the add-on effect of rectal balloon training to pelvic floor muscle training, with the goal to critically evaluate the psychometric properties and minimally important change (MIC) of the Vaizey score, Wexner score and the FIQL for use in the evaluation of patients with FI.

MATERIALS AND METHODS

Study design and subjects

The study population consisted of participants included in a randomised controlled trial.²⁴ Adult patients with FI were included at Maastricht University Medical Centre between August 2006 and May 2009 when they reported having had FI for more than six months, with a Vaizey incontinence score ≥ 12 (range 0–24), and failure of dietary measures and medication. Patients were excluded if diagnosed with an anorectal tumour within the past two years, absent squeeze pressure of the anal sphincter, chronic diarrhoea (always fluid stool three or more times a day), overflow incontinence, proctitis, ulcerative colitis, Crohn's disease, previous ileo-anal or colo-anal anastomosis and/or rectal prolapse in situ. Participants who had received physiotherapy during the previous six months or who were considered unable to comply with therapy were excluded as well. The Medical Ethics Committee of the Maastricht University Medical Centre approved the study.

Outcome measures

Baseline measurements consisted of medical history taking, physical examination, anal manometry (measurement of sphincter pressure during rest and squeezing), rectal capacity measurement (evaluation of threshold of first detectable sensation, urge sensation and maximum tolerable volume), anorectal sensitivity, endoanal ultrasound (assessment of internal and external abnormalities) and defecography (imaging of rectum during rectal evacuation). After the diagnostic work-up, patients were referred for a standardised physiotherapy program, comprising 12 sessions within a maximum of nine weeks and were followed up at a mean of 6.8 weeks (SD=5.3).

Patient-reported outcomes

Four questionnaires were completed at baseline and follow-up. The primary outcome was based on the Vaizey score, which was completed by an independent physician, and ranges from 0 (complete continence) to 24 (complete incontinence). The Vaizey score assesses the severity of FI (Table 8.1). The Wexner score was derived from the Vaizey score (without the items 'urgency' and 'medication use' and a higher weighting for 'pad use') and ranges from 0 (complete continence) to 20 (complete incontinence) (Table 8.1).

A nine-point global perceived effect (GPE) score asked patients to score their perceived change after physiotherapy treatment. Categories ranged from 'very much improved' to 'very much worse'.

The FIQL is a 29-item condition-specific QoL questionnaire, ranging from 4–16 and composing of four multi-item subscales: Lifestyle (10 items), Coping/Behaviour (9 items), Depression/Self Perception (7 items), and Embarrassment (3 items).²¹ A higher score indicates better QoL. Next to one question on general health and one on self perception, items are partly scored on a 4-point scale ranging from 'most of the time' to

‘none of the time’, and partly from ‘strongly agree’ to ‘strongly disagree’. The FIQL and GPE score were completed at home, thereby avoiding social desirable answers.

Table 8.1 Faecal incontinence score according to Vaizey and Wexner

Incontinent	Never	Rarely	Sometimes	Weekly/ usually	Daily/always
Solid stool ^a	0	1	2	3	4
Liquid stool ^a	0	1	2	3	4
Gas ^a	0	1	2	3	4
Alteration in lifestyle ^a	0	1	2	3	4
Wears pad ^b	0	1	2	3	4
				No	Yes
Need to wear a pad or plug ^c				0	2
Taking constipating medication ^c				0	2
Lack of ability to defer defecation for 15 min ^c				0	4

Never: no episodes in the past four weeks; rarely: 1 episode in the past four weeks; sometimes: >1 episode in the past four weeks, but <1 a week; weekly/usually: 1 or more episodes a week but <1 a day; daily/always: 1 or more episodes a day.

^aVaizey and Wexner items.

^bOnly Wexner item.

^cOnly Vaizey item.

Psychometric properties and statistical analyses

Distributional characteristics and responsiveness

The total PRO scores and individual items of the Vaizey and Wexner score were calculated at baseline and follow-up. Based on reports of effectiveness of physiotherapy treatment, patients were expected to improve after physiotherapy, although patients may respond differently.²⁵ The paired t-test was used to test the significance of the mean change of the total scores. Responsiveness refers to the ability of a PRO to detect change over time in the construct to be measured.²⁶ The standardised response mean (SRM) statistic was used to determine the magnitude of the instrument’s responsiveness between baseline and follow-up (group level internal responsiveness).²⁷ The SRM is defined as the mean change divided by the standard deviation (SD) of this change. A large SRM indicates that the change is large relative to the mean change variability in the measurements.²⁸ SRM values of 0.20, 0.50 and 0.80 correspond to small, moderate, and large change.²⁷

The external responsiveness was assessed by calculating the correlation coefficients between the changes in the individual items and total scores and the 9-point GPE score (Pearson or Spearman *r*).²⁷ The GPE score was used to represent an indication of change in the condition of the patient (anchor). In addition, we determined the area under the receiver operating characteristics (ROC) curve (AUC) for the PRO total scores, which is the probability that a questionnaire correctly classifies patients as improved or not improved, according to an external criterion (GPE).^{27,29} The categories ‘slightly better’ to ‘much more better’ indicated ‘important improvement’. The AUC was considered excellent with values ≥ 0.90 , adequate with values between 0.70–0.89 and poor with values < 0.70 .

Furthermore, responsiveness is considered to be a measure of longitudinal construct validity,³⁰ which is the degree to which change scores are related to scores of other instruments that measure the same construct.²⁶ In the absence of a gold standard, we expected the Vaizey and Wexner change scores to be highly convergent as they have overlap with regard to some sub items. Besides, we hypothesised that the Vaizey and Wexner change scores were related to the changes in the FIQL total score and its subscales, as the subscale items 'alteration in lifestyle' and 'pad use' specify aspects of QoL. Pearson correlation coefficients were calculated to evaluate the pre-specified hypotheses and were considered excellent with absolute values ≥ 0.60 , adequate with absolute values between 0.31–0.59 and poor with absolute values ≤ 0.30 .

Variability (the presence of floor or ceiling effects) may influence the responsiveness and will therefore be presented. Patients with high initial total scores have a greater change potential whereas the amount of change is limited in patients with low initial scores. Ceiling and floor effects were considered to be present if $\geq 15\%$ of responders had the worst (highest) and most desirable (lowest) possible outcome, respectively, in terms of individual items and the total scores.³⁰

Interpretability

Interpretability is the degree to which one can assign a qualitative meaning to an instrument's change in scores.²⁶ For each PRO, we calculated the mean scores at baseline and follow-up with the corresponding SRM, separately for each GPE category. The Wilcoxon signed rank test was used to test the hypothesis that there was no mean change in the measures over two time points in each GPE category.

Several anchor-based and distribution-based methods are proposed to determine the MIC. However, the MIC methodology is still in the developmental stage.^{28,31} It is argued that distribution-based methods, which use statistical measures as a value for MIC, should not be used because they assess minimal *detectable* change, but they lack information on whether this change is minimally important.³¹ Anchor-based methods use an external criterion to operationalise clinically important change. Two anchor-based methods were calculated:

1. The mean change method proposed by Jaeschke.^{6,32} The GPE was used as anchor and calculates the mean change in scores of patients who rated themselves as 'slightly better' minus the mean score of the patients who were 'unchanged'.
2. Receiver Operator Curve (ROC) analysis. We dichotomised the GPE for this analysis: persons who indicated that they were 'slightly better' to 'much more better' were labeled as 'importantly improved' while 'unchanged' to 'much more worse' was labeled as 'not importantly improved'. The ROC method aims to identify the optimal cut-off point for the change scores, that is, the value for which the sum of the percentages of false-positive and false-negative classifications ($[1 - \text{sensitivity}] + [1 - \text{specificity}]$) is smallest.^{27,32} We calculated the sensitivity and specificity of the MIC estimate assessed by the mean change method, by using the cut-off points provided by the ROC method.

To account for baseline differences, we performed additional analyses for calculating the MIC. Relative change scores were calculated as a percentage of change from baseline, by dividing the change score by the baseline score.³³ For the mean change method, we used the difference between the mean relative change scores of the 'slightly better' and 'unchanged' response categories.

Data were completed using the multiple imputation procedure in which each missing value was replaced by a set of multiple values, estimated by regression models and available data. We generated five multiple imputed datasets.^{34,35} Statistical analyses were performed using SPSS software, version 17.0 (IBM Corporation, Somers, NY, USA). Pooled results are presented, with a two-tailed P-value of 0.05 indicating statistical significance.

RESULTS

Distributional characteristics

101 patients were assessed for eligibility and 80 patients were randomised. Ten patients (12.5%) discontinued physiotherapy treatment or dropped out and 80 patients were analysed according to the intention to treat principle. The predominantly female population (90%) had a mean age of 59.3 (SD=11.9) and time since onset of FI symptoms of 78.4 months (SD=99.5). Patients experienced mainly mixed FI (56.3%), followed by urge FI (33.8%) and passive FI (10%).

Details for the distributional characteristics of the PROs are given in Table 8.2. The mean baseline total scores for the Vaizey, Wexner and FIQL were respectively 17.8 (SD=2.8; range=12–24), 15.4 (SD=2.9; range=7–20), and 9.4 (SD=2.6; range=4–16). The mean change in Vaizey score after physiotherapy treatment was 5.0 points (SD=5.4, $P<0.000$), whereas the Wexner score reduced with 4.8 points (SD=5.2, $P<0.000$), and the FIQL improved 1.2 points (SD=2.4, $P<0.000$). Table 8.3 shows the patient's perception of change in terms of GPE. The mean GPE score was 3.4 ($n=80$, SD=1.6). 57 patients rated subjective improvement (GPE=1 to 4), 18 patients rated themselves as unchanged, and only five patients showed subjective deterioration (GPE=6 to 9), which precludes interpretation of change in deteriorated patients.

Table 8.2 Distributional characteristics and responsiveness of Vaizey score, Wexner score and FIQL Scale

	Baseline Mean (SD) (median)	% Ceiling	% Floor	FU Mean (SD) (median)	Mean Change (SD) (median)	SRM ^a	Spear- man r^b
Vaizey score							
Urge	3.6 (1.2) (4)	-	-	3.1 (1.7) (4)	-0.5 (2.0) (0)	-0.3	0.41*
Pads	1.8 (0.7) (2)	-	-	1.5 (0.9) (2)	-0.3 (0.8) (0)	-0.4	0.22
Flatus	2.9 (1.3) (3)	47.5	11.3	2.4 (1.5) (3)	-0.6 (1.4) (0)	-0.4	0.15
Liquid	3.3 (0.9) (3)	45.0	1.3	2.0 (1.5) (2)	-1.3 (1.3) (-1)	-1.0	0.42*
Solid	2.1 (1.4) (2.5)	17.5	22.5	1.2 (1.4) (0.5)	-0.9 (1.6) (-1)	-0.6	0.32*
Adjustment	3.7 (0.8) (4)	87.5	2.5	2.4 (1.7) (3)	-1.4 (1.6) (-1)	-0.9	0.53*
Medication	0.4 (0.8) ^c (0)	-	-	0.3 (0.7) ^c (0)	-0.1 (0.9) (0)	0.1	0.17
Total Vaizey	17.8 (2.8)	1.3	6.3	12.8 (5.9)	-5.0 (5.4)	-0.9	0.60 ^{d*}
Wexner score							
Pads	3.3 (1.4)	77.5	12.5	2.7 (1.7)	-0.6 (2.0)	-0.3	0.08
Total Wexner	15.4 (2.9)	5.0	0.0	10.6 (5.3)	-4.8 (5.2)	-0.9	0.44 ^{d*}
FIQL Scale							
Lifestyle ^e	2.7 (0.9)	6.3	1.3	3.0 (0.8)	0.3 (0.8)	0.4	-0.33 ^{d*}
Coping ^e	1.9 (0.7)	0.0	4.3	2.3 (0.9)	0.4 (0.7)	0.5	-0.45 ^{d*}
Depression ^e	2.7 (0.8)	2.5	0.0	2.9 (0.7)	0.1 (0.7)	0.2	-0.38 ^{d*}
Embarrass- ment ^e	2.1 (0.8)	5.3	18.5	2.5 (1.0)	0.4 (0.9)	0.5	-0.32 ^{d*}
Total FIQL	9.4 (2.6)	0.0	0.0	10.6 (2.8)	1.2 (2.4)	0.5	-0.48 ^{d*}

FIQL, Fecal Incontinence Quality of Life Scale; SD, standard deviation; FU, follow-up.

*Significant at the 0.05 level.

^aSRM, standardised response mean; mean change score per category divided by the standard deviation of the change scores.

^bSpearman correlation coefficients between the changes in the total and individual item scores and patient's perceived benefit assessed by the GPE.

^cCoded as follows: (0)=no, (2)=yes.

^dPearson r .

^eFIQL subscales.

Responsiveness

Internal and external responsiveness

Total Vaizey score showed excellent internal and external responsiveness based on the SRM (-0.9), AUC (0.886), and Pearson correlation ($r=0.60$) (Table 8.2). The total Wexner score showed excellent internal responsiveness (SRM=-0.9), and adequate external responsiveness ($r=0.44$, AUC=0.842). The total FIQL showed adequate internal and external responsiveness (SRM=0.5, $r=-0.48$, and AUC=0.765). The individual items of the Vaizey score showed inconsistent results for the responsiveness. The items 'liquid stool incontinence' and 'adjustment' had excellent internal responsiveness (SRM=-1.0 and -0.9, respectively) and adequate external responsiveness ($r=0.42$ and 0.53, respectively), whereas 'medication use', 'pads' and 'flatus incontinence' had poor internal and external responsiveness. The item 'pad use' of the Wexner score had poor internal (SRM=-0.3) and external responsiveness ($r=0.08$). The FIQL Coping and Embarrassment subscales had adequate internal (SRM=0.5) and external responsiveness ($r=-0.45$ and -0.32, respectively). The Lifestyle and Depression subscales had poor internal responsiveness (SRM=0.4 and 0.2, respectively) and adequate external responsiveness ($r=-0.33$ and -0.38, respectively).

Table 8.3 Patient's perception of change in terms of global perceived effect (GPE), and mean changes in the Vaizey score, Wexner score and FIQL Scale at FU

GPE score	n (%)	Baseline Mean (SD)	FU Mean (SD)	Change scores ^a	SRM ^b	P ^c
Vaizey						
1. Much more better	9 (11.3)	17.1 (3.1)	5.7 (6.1)	-11.4 (5.8)	-2.0	<0.01
2. Much Better	17 (21.3)	17.0 (2.8)	9.5 (4.9)	-7.5 (4.7)	-1.6	<0.00
3. Moderately better	18 (22.5)	18.3 (3.2)	12.9 (4.8)	-5.4 (3.3)	-1.6	<0.00
4. Slightly better	13 (16.3)	19.0 (2.0)	14.40 (5.5)	-4.6 (4.7)	-1.0	<0.01
5. Unchanged	18 (22.5)	17.8 (2.7)	17.4 (3.3)	-0.4 (4.1)	0.1	<1.00
6. Slightly worse	2 (2.5)	18.5 (1.3)	16.3 (1.8)	-2.20 (0.7)	-3.1	<0.50
7. Moderately worse	1 (1.3)	12.5	13.5	1.00	0.3	-
8. Much worse	2 (2.5)	15.3 (0.8)	15.7 (0.7)	0.4 (1.0)	0.4	<1.00
9. Much more worse	0	-	-	-	-	-
Improved	57 (71.3)	17.9 (2.9)	11.1 (5.9)	-6.8 (5.0)	-1.4	0.00 ^d
Not improved	23 (28.8)	17.5 (2.7)	16.9 (3.2)	-0.5 (3.7)	-0.1	<0.88
Wexner						
1. Much more better	9 (11.3)	13.5 (3.8)	5.1 (5.0)	-8.5 (3.9)	-2.2	<0.01
2. Much Better	17 (21.3)	14.6 (3.0)	7.4 (4.1)	-7.3 (4.2)	-1.7	<0.00
3. Moderately better	18 (22.5)	15.7 (2.8)	10.5 (4.3)	-5.2 (3.4)	-1.5	<0.00
4. Slightly better	13 (16.3)	16.4 (2.3)	12.3 (4.7)	-4.1 (3.5)	-1.2	<0.01
5. Unchanged	18 (22.5)	16.3 (2.1)	14.9 (3.5)	-1.4 (3.8)	-0.4	<0.45
6. Slightly worse	2 (2.5)	14.4 (2.9)	14.1 (5.0)	-0.3 (3.1)	-0.1	<0.66
7. Moderately worse	1 (1.3)	13.2	11.3	-1.9	-	-
8. Much worse	2 (2.5)	12.3 (3.6)	12.1 (2.5)	-0.3 (3.9)	-0.1	<0.66
9. Much more worse	0	-	-	-	-	-
Improved	57 (71.3)	15.2 (3.0)	9.1 (5.0)	-6.1 (4.0)	-1.5	<0.00 ^d
Not improved	23 (28.8)	15.8 (2.4)	14.4 (3.6)	-1.4 (3.7)	-0.4	<0.39
FIQL						
1. Much more better	9 (11.3)	8.7 (2.1)	12.6 (3.3)	3.8 (3.0)	1.3	<0.02
2. Much Better	17 (21.3)	10 (2.9)	11.9 (2.6)	1.9 (2.8)	0.7	<0.02
3. Moderately better	18 (22.5)	10.0 (2.2)	11.1 (2.4)	1.1 (1.4)	0.8	<0.04
4. Slightly better	13 (16.3)	8.3 (2.7)	9.3 (2.0)	1.0 (1.5)	0.7	<0.05
5. Unchanged	18 (22.5)	9.5 (2.7)	9.4 (2.8)	-0.1 (1.3)	-0.1	<0.98
6. Slightly worse	2 (2.5)	7.9 (2.3)	9.7 (3.6)	1.8 (2.9)	0.6	<0.72
7. Moderately worse	1 (1.3)	9.4	7.0	-2.4	-	-
8. Much worse	2 (2.5)	10.4 (2.4)	9.0 (1.2)	-1.4 (1.8)	0.8	<0.66
9. Much more worse	0	-	-	-	-	-
Improved	57 (71.3)	9.4 (2.6)	11.2 (2.7)	1.8 (1.7)	1.1	0.00 ^d
Not improved	23 (28.8)	9.4 (2.6)	9.3 (2.7)	-0.1 (2.4)	0.0	<0.78

Pooled SD's based on categories with n>1; FIQL, Fecal Incontinence Quality of Life Scale; FU, follow-up.

^aVaizey: min, max=-24, 12; Wexner: min, max=-20, 13; FIQL: min, max=-12, 12.

^bSRM, standardised response mean; mean change score per category divided by the standard deviation of the change scores.

^cWilcoxon signed rank test.

^dPaired samples t-test.

Longitudinal construct validity

Table 8.4 shows the correlation coefficients between the changes in PROs. The Vaizey score was excellently associated with the Wexner score ($r=0.91$). Both the Vaizey and Wexner score were adequately associated with the FIQL total score ($r=-0.53$ and -0.45 , respectively), adequately associated with the Lifestyle, Coping and Embarrassment FIQL subscales, and poorly associated with the Depression subscale.

Table 8.4 Correlation coefficients between changes in PROs from baseline to follow-up

	Vaizey score	Wexner score
Wexner score	0.91 ^b	-
FIQL total	-0.53 ^b	-0.45 ^b
Lifestyle subscale	-0.38 ^a	-0.35 (P=0.058)
Coping/Behavior subscale	-0.53 ^b	-0.46 ^b
Depression/ Self Perception subscale	-0.26 ^a	-0.21 (P=0.086)
Embarrassment subscale	-0.43 ^b	-0.37 ^b

^aCorrelations significant at the 0.05 level (2-tailed).

^bCorrelations significant at the 0.01 level (2-tailed).

Ceiling and floor effects

Ceiling effects are observed when related to the individual items of the Vaizey score and the item ‘pad use’ of the Wexner score, which might be explained by the smaller range of response categories (Table 8.2). From an improvement perspective it is not worrying to observe ceiling effects. Floor effects are observed for the individual item ‘solid stool incontinence’ (Vaizey) and the Embarrassment subscale of the FIQL.

Interpretability

The change in Vaizey score per GPE category is according to what was expected. Patients who rated themselves to be improved had higher changes compared to unchanged patients (SRM=-1.4 vs. -0.1) (Table 8.3). This result was further confirmed by the results of the Wilcoxon signed rank test (P=0.00 vs. P<0.88). The same pattern and conclusions applied to the Wexner score and FIQL Scale. In addition, the SRMs per GPE category showed a rather consistent pattern for all three PROs, although the SRM estimates for the small number of patients who deteriorated (n=5) provided no reliable conclusion.

Variability existed in the MIC estimates of the Vaizey and Wexner, according to the different methods, whereas the estimates for the FIQL were rather consistent (Table 8.5). The MIC estimates for the Vaizey score based on the absolute change scores ranged from -3 to -5, for the Wexner score from -2 to -3 and for the FIQL from 1.1 to 1.2. The ROC method provided the MIC estimates with the lowest misclassification rates, which were 20%, 17.5% and 33.8% for the Vaizey, Wexner and FIQL, respectively. The MIC for the Vaizey score based on the relative change scores was -25.1% (mean change method) and -14.3% (ROC). An MIC of -25.1% means 25.1% improvement from baseline (e.g., change from 24 to 18 (most severe patients) or change from 12 to 9 (least severe patient)). The MIC estimates calculated with the mean change method were higher for both the absolute and relative change scores. For the purpose of easy interpretation, Table 8.6 gives a simplified overview of the results originating from this study.

Table 8.5 Minimally important change according to two anchor-based methods

	Vaizey score (range 0–24) [abs]	Wexner score (range 0–20) [abs]	FIQL (range 4–16)
Absolute change scores			
1. Mean change method	-4.2 [-5] (Se=0.65; Sp=0.89)	-2.7 [-3] (Se=0.78; Sp=0.78)	1.1 (Se=0.61; Sp=0.86)
2. ROC method	-3.0 [-3] (Se=0.80; Sp=0.82)	-2.0 [-2] (Se=0.87; Sp=0.70)	1.2 (Se=0.59; Sp=0.90)
Range MIC (min, max)	2.0 (-3, -5)	1.0 (-2, -3)	0.1 (1.1, 1.2)
Relative change scores			
1. Mean change method	-25.1%	-19.5%	17.8%
2. ROC method	-14.3%	-12.6%	12.5%

Abs, absolute values; FIQL, Fecal Incontinence Quality of Life Scale; MIC, minimally important change; Se, sensitivity; Sp, specificity.

Table 8.6 Simplified overview of study results

PRO properties	Vaizey	Wexner	FIQL	Lifestyle subscale	Coping subscale	Depression subscale	Embar-rassment subscale
Internal responsiveness	++	++	+	-	+	-	+
External responsiveness	++	+	+	+	+	+	+
Longitudinal construct validity	+/++	+/++	+	+	+	-	+
Interpretability	+	+	+	NA	NA	NA	NA
Range MIC	-3 to -5	-2 to -3	1.1 to 1.2	NA	NA	NA	NA
Easy to sum up	++	++	-	-	-	-	-

PRO, patient-reported outcome; FIQL, Fecal Incontinence Quality of Life Scale; MIC, minimally important change; ++=excellent; +=adequate; -=poor; NA, not applicable.

DISCUSSION

To be useful in clinical trials PROs should have acceptable reliability, validity, responsiveness and interpretability. The aim of this study was to determine the psychometric properties of the Vaizey and Wexner score, and the FIQL for use in the evaluation of patients with FI and to provide a clinically meaningful interpretation of changes in the PROs.

It should be considered that when interpreting our results, the most appropriate methods to determine the responsiveness statistic and the MIC are a matter of debate.²⁷ The SRM was used to assess the internal responsiveness as it is not dependent on sample size and based on variability of change.^{27,28} It may however vary as a function of treatment effectiveness, such as physiotherapy compared to surgery or mild compared to severe FI.^{27,28} External responsiveness, assessed by the AUC and the correlation coefficient, may be influenced by the choice of the anchor.^{27,28,36}

It is also arbitrary which method should be used to determine the MIC, given the shortcomings of each method.³¹ Some recommend using several anchor-based methods and to examine various distribution-based estimates as supportive information.³⁷ Others conclude that using multiple methods does not work.³¹ To estimate the MIC we relied on anchor-based approaches, since they consider minimally important change rather than minimally detectable change. The choice of the anchor and definition of

important change is crucial in anchor-based approaches.²⁷ The GPE score does not represent a 'gold standard', but is an alternative offering a reasonable estimate of true change. Moreover, the GPE is easy to obtain and the best measure from the individual's perspective.²⁸

We considered 'slightly better' as MIC. Although it is unknown whether this change is considered important by patients, higher change scores for all PROs were observed when comparing the 'slightly better' and 'unchanged' group.³⁷ The ROC method is preferred when false positive and false negative classifications are equally weighted. Besides, the ROC method uses all data and is thus more powerful.³¹ In general, the ROC method results in lower estimates.³⁸

This study is one of the first that performed an elaborative psychometric evaluation of the PROs under study based on FI patients that received physiotherapy treatment.^{3,10,39}

The Vaizey and Wexner score both aim to evaluate the FI severity. Strengths include that they are easy to use, rapidly completed and both show adequate to excellent responsiveness following physiotherapy treatment.^{10,40} Furthermore, they changed in agreement with subjective improvement, which is in line with other studies.^{3,23,39} Data from other studies suggest adequate test-retest reliability for the Vaizey and Wexner score,^{10,23} although one study found unsatisfactory results in patients after colorectal surgery.⁴¹ The construct validity seemed adequate for both when related to clinical impression,¹⁰ or when the Vaizey score was related to either patients' perception of bowel control³⁹ or the Hallböök or COREFO questionnaires.⁴¹

However, the Vaizey score has several limitations.³⁹ The urgency item is not applicable in patients with passive FI alone or urgency without incontinence. Hence, the score may not adequately reflect symptom severity in these patients. The Vaizey score had also poor internal consistency,²³ which may result from the suggestion that it is composed of two multi-item scales, i.e. incontinence and social impact.⁴¹

Moreover, the amount of leakage is not represented, and the scoring of pad use may be biased in patients with concomitant urinary leakage. Pad and medication use showed also poor responsiveness following physiotherapy,²³ indicating that a large improvement is necessary to be self-confident enough to abstain from incontinence material and constipating medication.

Given the limitation associated with the additional items that the Vaizey score assesses compared to the Wexner score, together with only minor differences in psychometric properties and the absence of another highly recommendable questionnaire, the Wexner score seems preferable to assess severity of FI.

The impact of FI on QoL is evident and its assessment is therefore deemed an essential aspect in the evaluation and treatment of FI. Only a few condition-specific QoL instruments are available.^{21,42-44} The Gastrointestinal Quality of Life Index,⁴⁴ the Manchester Health Questionnaire⁴³ or its modified version⁴⁵ have items that remain too general and therefore sacrifice specificity and responsiveness.⁴⁶ The FIQL is designed for use in clinical trials and outcome research in adults with FI. Furthermore, it is developed to be

responsive to the condition in question and is based on a clearly described conceptual framework.^{21,46,47} Rockwood²¹ concluded that the FIQL subscales demonstrated acceptable internal consistency, test-retest reliability and adequate discriminate and convergent validity. Our findings showed that the Depression subscale had inadequate internal responsiveness and longitudinal construct validity. The FIQL total score had acceptable responsiveness, test-retest reliability and longitudinal construct validity. Based on our results and in the absence of other recommendable QoL scales, we agree with Avery to recommend the FIQL for evaluating QoL of patients with FI.²⁰

Practical issues related to the 29-item FIQL include the complicated addition of items and time-consuming completion, which holds consequences for clinical use. Finally, to be useful in the Dutch situation, further validation of the translated version of the FIQL is required.⁴⁷

Despite the variability in MIC estimates in our study, recommendations may be valuable in planning new physiotherapy trials, as sample size and statistical power are dependent on the magnitude of the MIC.

A plausible MIC estimate for the Vaizey score would be five points, as this was found in the mean change method and supported by earlier results.²³ However, this would be penalised by increased patient misclassification (29 vs. 20%) compared to the ROC method (MIC = -3). Another study also found a MIC of three points,³ although results are difficult to compare because it included deteriorated patients in the 'unchanged' group, used a five-point GPE, and the difference between the groups was rounded off downwards.³ The same study provided a MIC of three points for the Wexner score (mean change method) which corresponds with our findings. With regard to the FIQL, a MIC estimate of 1.2 seems reasonable, based on our rather consistent results and the fact that the ROC method is the preferred anchor-based method. To our knowledge, this is the first study that reports on interpretability of the FIQL.

When interpreting MIC estimates, it is known that the MIC is a variable concept, depending on the method, definition of important improvement, type of anchor, differences between perceived effectiveness between patients, variation across different patient populations, and baseline values.³⁷ MIC estimates based on relative change scores were calculated to account for baseline differences. It is shown that patients with higher baseline values require higher change scores to be considered importantly improved.

In clinical practice, MIC interpretation shifts from proportions of misclassifications in groups to probabilities in individual patients.⁴⁸ Furthermore, the presented MICs should not be considered as fixed values but be used as an indication. Choosing a specific MIC may depend on the baseline value, consequences in patient care, such as type of intervention or the consequence of being misclassified. Our study should be considered as a first step in improving our knowledge on the interpretation of the PROs and establishing their usefulness. Future clinical trials should explore MIC estimates for important clinical subgroups, and support and perhaps further narrow the range of MIC values.

In conclusion, the current instruments available to evaluate severity and QoL in FI do not yet attain the highest levels of psychometric soundness. As the focus of patients (embarrassment, fear, preventive measures) may differ from that of physicians (frequency and amount of lost stool, type of FI), it is recommended to include several evaluative measures, such as the combination of a severity scale, subjective rating of improvement, and a QoL instrument. Integration of objective and subjective items into one PRO in order to capture both the focus of the patient and physician may affect the psychometric soundness. Given the lack of a psychometrically robust PRO, developments in this research area are of great interest.⁴⁹ So far, the Wexner score seems most suitable for severity assessment and the FIQL for evaluating QoL.

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CHAPTER 9

GENERAL DISCUSSION

This chapter presents an overview of the main findings of the performed studies, and discusses important strengths and weaknesses, which were not fully highlighted in the previous chapters. It also positions the results of the studies in the context of other scientific evidence in the field of faecal incontinence (FI) management, followed by a discussion on the implications for current health care practice. Finally, it offers some general recommendations for future research.

MAIN FINDINGS

This thesis started with a survey of the state of the art regarding different aspects of the management of patients with FI. Subsequently, it presented a systematic review assessing the aetiological factors for postpartum FI. A third- or fourth-degree sphincter rupture appeared to be the only factor which is strongly (anal incontinence) or moderately (flatus incontinence) associated with postpartum FI. Furthermore, two studies reported on assessing the psychometric properties and minimally important change for the Vaizey score, Wexner score and Fecal Incontinence Quality of Life Scale (FIQL) for use in physiotherapy research. These outcome measures do not as yet meet the highest levels of psychometric quality. There are indications that the Wexner score is most suitable for severity assessment and the FIQL for evaluating quality of life. Finally, we carried out a randomised controlled trial to examine the effectiveness of rectal balloon training (RBT) as an add-on therapy to pelvic floor muscle training (PFMT). In terms of the primary outcome (Vaizey score), RBT appeared equally effective as PFMT alone, whereas secondary outcomes showed beneficial effects of rectal balloon add-on training on the control of urgency, subjective improvement, fatigue of the external anal sphincter and lifestyle adaptations. Secondary analyses of the trial data provided predictors for a favourable outcome of physiotherapy treatment, viz. less embarrassment, any obstetric factor and the use of constipating medication. A longer duration since the onset of FI symptoms was associated with an unfavourable outcome.

METHODOLOGICAL CONSIDERATIONS

Patients and providers

When generalising the findings of the studies based on our randomised controlled trial, we have to take into account that women were overrepresented (90%) in the study population (chapter 6). This imbalance is not attributable to selection bias, but is inherent to the disorder, and is comparable to findings in other studies.¹⁻⁵ Furthermore, we have to interpret our results in view of the patient group we included. Patients had to have experienced moderate to severe symptoms for at least six months after failure of medication and/or dietary advice. Our inclusion criteria selected those patients who had not benefited from simpler conservative measures, such as dietary adjustments, whose FI was not due to another underlying disorder such as ulcerative

colitis, and who had no impediments as regards receiving and completing physiotherapy treatment, such as language restrictions or absence of squeeze pressure.

Severity of symptoms was defined as a Vaizey score of at least 12 points (range 0–24). This score could be obtained and interpreted in several ways, but in our opinion indicates moderate to severe symptoms, as the patient experiences at least moderate health problems in terms of impairments in functions and structures, limitations in activities and/or restrictions in participation, as classified by the 'International Classification of Functioning, Disability and Health' (ICF).⁶ In view of the large proportion of included patients who experienced double incontinence (46.3%), an additional impact on their quality of life was to be expected.^{7,8}

At the start of our trial, we intended to include 106 patients within 14 months (chapters 5 and 6), a figure which was based on the FI prevalence rates in cross-sectional surveys, the availability of the few physicians specialising in FI, and the inclusion rate from a previous study at the same medical centre with similar inclusion criteria.¹ Unfortunately, we were only able to include 100 and randomise 80 patients, which may be attributable to several factors. First of all, it has frequently been reported that there is a discrepancy between reported prevalence figures and number of patients seeking help. Despite more liberal values in today's society, it is still very difficult to admit FI to others, including your closest relatives. Some 67% of patients avoid seeking help due to this embarrassment, due to the misconception that it is a normal part of ageing or due to unawareness of treatment options. Although these reasons are not legitimate, such ideas still prevail in this patient group. Second, when patients ultimately do seek help, not all general practitioners seem to be aware of the wide range of non-surgical and surgical treatment options, and even today they sometimes only prescribe incontinence absorption materials.⁹ It has been reported that only 17.3% of the patients who consulted a general practitioner with FI were referred for further diagnostic work-up (primary and secondary care, based on 60 practices, n=965.070 consultations, International Classification of Primary Care (ICPC) code D17), with 30% being referred for physiotherapy treatment.¹⁰ Third, having all the required standardised forms completed by only one physician (who was responsible for the inclusions) was a time-consuming process which competed with the regular daily workload. We could have included more centres to increase the inclusion rate, but probably at the cost of reduced internal validity. In a multicentre trial, the quality of data gathering is influenced by different physicians being involved, the availability and variability of equipment, more problematic communication and difficulties checking the data. Another option might have been to involve general practitioners in the inclusion process. However, statistics of the Netherlands Information Network of General Practice (LINH) showed that reports of FI to general practitioners were very rare: only 0.02% of all consultations in 60 practices comprised FI (ICPC code D17).¹⁰ Data from the National Database on Paramedical Care (Landelijke informatie-voorziening Paramedische Zorg (LiPZ)) showed similar results.¹¹ Furthermore, a 2004 report from the Netherlands Institute for Health Services Research (NIVEL) showed that the prevalence rates of presenting to

the general practitioner with FI symptoms among men and women aged 45–64 years were 0.1/1000/year and 0.5/1000/year, respectively.¹² It therefore did not seem useful to us to pursue this option. Finally, our patient population consisted mostly of women with a mean age of 60 years. Given these characteristics, obstetric risk factors, hormonal and age-related physiological changes, or other co-morbidity, like cardiac disease or respiratory problems, were likely to be frequently present. Even more important is the co-occurrence of other pelvic floor dysfunctions, such as urinary incontinence or rectal prolapse. A notable aspect is the high co-occurrence of urinary incontinence in our patient populations (46.3%). FI treatment will probably only be given priority if FI is the prevailing disorder.

In order to determine the specific long-term effect of an intervention under study, included patients need to abstain from any co-intervention. However, this might be unethical, especially for those patients who do not improve to a satisfactory level.¹³ In our trial, two thirds of all patients attending the post-intervention follow-up were not receiving any additional intervention, whereas one third were receiving other treatments (protocol deviation), such as colonic irrigation or percutaneous nerve evaluation. The intention-to-treat principle indicates that all randomised patients have to be included in the analysis, regardless of whether they have received co-interventions. This hampers the interpretation of specific long-term effects.

Outcome measures

The literature review showed that no outcome measure was available which satisfied the criteria for excellent validity, reliability, responsiveness or interpretability. At the start of the randomised controlled trial project, described in chapters 5 and 6, we decided to use the Vaizey score as the primary outcome measure as this score not only includes items relating to severity and frequency, but also to urgency, pad use and social restrictions. This also enabled us to calculate the sample size required for our study from Vaizey score data from a previous Dutch study with a comparable study population.¹⁴ Because a validated Dutch version of the questionnaire was not available, we decided to use their translated version of the Vaizey questionnaire. This was also the case for the FIQL, which is currently the only condition-specific quality of life measure for FI. As it has been recommended to include quality of life as an outcome measure especially for disorders characterised by embarrassment and social life restrictions,¹⁵ we decided to translate the FIQL for use in the Dutch population, checking the translation by means of a backtranslation procedure. However, it is always questionable whether or not a construct retains its validity when an instrument is translated from one language or culture to another.¹⁶

In our trial, we used the 9-point global perceived effect (GPE) score to rate subjective improvement. This represents a modification of the method used by Jaeschke et al., who originally proposed a 15-point GPE for use as an anchor.¹⁷ We are aware that validation of this subjective outcome is problematic, and patient's response may be biased by the state of mind, a 'placebo' effect (patients want to believe that they have

improved) and the retrospective character of the GPE (recall bias). Nevertheless, this outcome is used more and more, probably because certain restrictions, such as psychometric properties or weak correlation with severity of FI symptoms, also apply to more objective outcome measures, such as questionnaires or diagnostic tests. Moreover, 'objective' measures are also influenced by various circumstances, for instance in the subjective evaluation of endosonography.

The number of categories to be included in the GPE is arbitrary, and the number of response categories varies between studies.^{14,18-21} We chose to use a 9-point scale because we felt that nine response categories were necessary to describe all relevant categories without losing the ability to discriminate. We did not include 'completely recovered' at the extreme of the GPE, because we wanted a questionnaire with mirrored response categories. Moreover, 'complete recovery' would then be based on the post-intervention follow-up, which only captures a short period during which symptoms can evolve. It is generally reported that symptoms may relapse over time, especially in the absence of follow-up interventions or lack of adherence to the required home exercise programme.

Methodological quality

In the past decades, physiotherapy has focused strongly on scientific research and evidence-based practice. The volume and quality of the evidence used to guide physiotherapy interventions have grown, and the number of randomised controlled trials and systematic reviews of physiotherapy interventions has doubled every 3.5 years.²²⁻²⁵ Evidence from randomised trials and systematic reviews has been used for the development of national and international physiotherapy guidelines, reflecting the transition from clinically based practice to evidence-based practice.^{13,26,27} Furthermore, several initiatives, such as the CONSORT statement (Consolidated Standards of Reporting Trials), checklists to assess the methodological quality of studies, and grading the quality of evidence and strength of recommendations for guideline purposes, have contributed to higher methodological quality standards.^{28,29} We were able to perform our trial, described in chapters 5 and 6, while closely following the revised CONSORT statement published in 2010.^{28,29} Although lack of blinding of patients and therapists may have introduced bias in our trial, this is a generally recognised limitation of physiotherapy trials, which may preclude maximum scores being obtained when using lists of methodological quality criteria that include items on blinding of patients or therapists.^{30,31}

POSTPARTUM FAECAL INCONTINENCE AND PREVENTION

After birth, most of the attention is devoted to the newborn baby, even though the mother may experience a variety of delivery-related complications, such as severe blood loss or sphincter rupture. In addition, the postpartum period is a highly demanding period for the mother, involving emotional aspects, care for the other children, lack of sleep and household or work activities.^{7,32} Sphincter rupture during delivery occurs in approximately 27% of women.³³ We found that a third- or fourth-degree sphincter rupture is the most important factor to be strongly (anal incontinence) or moderately (flatus incontinence) associated with postpartum FI (chapter 3). Mechanisms involved in postpartum incontinence may relate to a denervation injury, or to supportive tissue or anatomical injuries.³⁴ Up to 10% of young mothers experience FI after childbirth, while 33% experience urinary incontinence.³⁵ This is worrying, as (temporary) FI symptoms after vaginal delivery or caesarean section may predispose to (chronic) FI later in life.^{32,35,36}

Assessment of the severity of anal sphincter injury is relevant for estimating the risk of postpartum pelvic floor dysfunctions and for deciding on further policy. This assessment can be done by a well-trained gynaecologist or with the aid of imaging techniques to visualise the anal sphincters. Both endoanal sonography and endoanal MRI have been reported to be accurate tools for mapping internal and external anal sphincter defects.^{37,38}

Role of physiotherapy in primary and secondary prevention

Recognition of nerve, supportive tissue or anatomical system injuries following vaginal delivery is particularly relevant for primary and/or secondary prevention of FI immediately after delivery or later. Primary prevention aims to remove the causes of a disease,³⁵ while secondary prevention focuses on the detection of asymptomatic dysfunction and providing treatment to end its progress.³⁵

When a person sustains a sports injury, counselling and treatment mostly starts the next day, whereas women experiencing a third- or fourth-degree sphincter rupture often receive treatment only after six weeks.^{39,32} Despite the common occurrence of FI and other pelvic floor dysfunctions during pregnancy and after (vaginal) delivery,³² usual preventive care for postpartum women thus still seems to be insufficient,⁴⁰ and inefficient⁴¹ as well as poorly organised⁴¹ in terms of information provision (including risk and prognosis), education, assessment, identification of women at risk and early intervention.^{32,42}

In terms of the three main stages of injury healing, pelvic physiotherapy may have a role to play in each stage to prevent symptoms (primary prevention) or reduce symptoms (secondary prevention). During the first stage, i.e. the inflammatory or acute stage (0 to 10 days), attention should focus on improved wound healing, for instance by giving information on toilet behaviour (preventing straining and stretching), abdominal pressure reduction and having softer stools.³⁹ During the second stage, the

proliferative phase (4 to 21 days), physiotherapy should be directed at sensory (awareness of pelvic floor and urge to defecate) and motor training (coordination/proprioception) with the aim of stimulating the organisation and orientation of new collagen connective tissue.³⁹ Physiotherapy in the third stage, the remodelling stage (21 to 265 days), should involve selective stimuli and active therapy to prepare for daily activities and sports.³⁹

It has been hypothesised that pelvic floor muscle training before or during pregnancy may be beneficial to improve external anal sphincter function and hence prevent incontinence after birth.³⁵ A trained muscle may be less prone to injury, may have a greater strength reserve, or may be easier to retrain following damage.³⁵

Furthermore, research has shown that offering advice on pelvic floor muscle training to all pregnant or postpartum women, whether they have incontinence symptoms or not, might be effective in late pregnancy and during the postpartum period.³⁵ This will only be effective, however, if the protocol is intensive enough (sufficient exercise dosage and sufficient contact with a health care professional to teach, supervise and encourage training).³⁵ In addition, there is support for the widespread recommendation that pelvic floor muscle training is an effective treatment for older women with persistent postpartum stress urinary incontinence to reduce urinary leakage.³⁵ Based on the arguments outlined above, physiotherapy treatment is expected to have a beneficial effect in the prevention of FI.

Multidisciplinary screening

Multidisciplinary screening of the pelvic and urogenital regions in postpartum women at risk for incontinence should be organised during follow-up visits after delivery. This may include a visit to a midwife, gynaecologist, general practitioner or pelvic physiotherapist. Current knowledge suggests that sphincter injury is the most consistent and important factor associated with the development of postpartum FI (chapter 3).³⁵ From this perspective, it seems reasonable that this high-risk group should be monitored carefully and should consult an expert on pelvic floor dysfunctions following delivery. The added value of pelvic physiotherapy may be the additional attention the women receive as a result of longer consultation times, the teaching of pelvic floor muscle awareness and pelvic floor muscle exercises that can address their self-management in the primary or secondary prevention, and providing instructions on heavy lifting. Although the (small) evidence base supports a beneficial effect for the primary and secondary prevention of FI in the postpartum period, the feasibility and effectiveness of postpartum prevention and treatment programmes by pelvic physiotherapists need further investigation. One might also consider a one-time physiotherapy consultation, prior to possible referral for pelvic physiotherapy (Figure 1; C).⁴³ This might involve a written request by a referring physician to a pelvic physiotherapist to examine and evaluate a patient to generate information regarding (functional) diagnosis and prognosis, in order to facilitate their management plan, specifically regarding the options for physiotherapy intervention. It could provide a valuable tool to enhance

the appropriateness of referral and improve communication among professionals. Research on other disorders has shown that primary care physicians used the opportunity of a one-time physical therapy consultation and were satisfied with the outcome and process of consultation.⁴³ A pilot study might elucidate the feasibility and efficiency of such a one-time consultation for patients with pelvic floor dysfunctions.

Further research should focus on optimising screening programmes to identify women at risk for incontinence (persistent symptoms, sphincter rupture),⁴⁴ as well as on the short- and long-term effectiveness of prevention programmes targeting at postpartum women at risk, and on ways of ensuring the organisation and implementation of such programmes (in terms of embedding in usual care, costs and availability of skilled professionals). Barriers to be overcome include health insurance companies refusing to cover the costs of assessment and treatment, and the problems of promoting participation in primary and secondary prevention programmes even when women do not (or not yet) perceive symptoms. Finally, it would be interesting to investigate the role of genetic predisposition in relation to the development of pelvic floor dysfunctions. A familial inheritance pattern was recently demonstrated in a cross-sectional study which showed that a maternal history of pelvic organ prolapse was an independent risk factor for such prolapse.⁴⁵

PELVIC PHYSIOTHERAPY: EFFECTIVENESS, PREDICTIVE FACTORS AND PATIENT SELECTION

Selecting patients suitable for pelvic physiotherapy is associated with the question: whether and to what extent are patients with FI, based on type and severity of FI, amenable to symptom relief when treated with pelvic physiotherapy? And what kind and intensity of pelvic physiotherapy is required to be effective?

Effectiveness

Before considering physiotherapy, specific conditions should be excluded as they require other (surgical) interventions. These include acute trauma of the anal sphincters which requires surgery, treatable causes of diarrhoea (inflammatory bowel disease or infection), changes in defecation pattern due to colorectal cancer, rectal prolapse in situ or grade three or four internal haemorrhoids, leakage of faeces due to constipation, or neurologic disease.¹³ After failure to normalise the consistency of the faeces and optimise the defecation pattern (by means of medication, use of gastro-colon reflex, posture or diet), physiotherapy might be indicated.

To date, there is not enough evidence from trials to decide whether electrical stimulation is helpful, so it is not possible to recommend this treatment for FI.^{44,46} Although biofeedback (including RBT) to augment PFMT may be effective compared to education, lifestyle and dietary interventions, this remains unproven.^{13,44} It is also questionable whether biofeedback has an additional effect compared to PFMT alone.^{13,44} Based

on the existing evidence, no treatment modality can be recommended as the mainstay of treatment.⁴⁷

However, consensus exists on the following recommendations.⁴⁸

- Offering patients advice and information is regarded as an essential element of physiotherapy treatment. It will help patients understand their disorder and create coping strategies.
- In view of the low costs and morbidity and the (weak) evidence suggesting effectiveness, PFMT is recommended as an early intervention as part of other conservative management (education, lifestyle and dietary interventions).
- In case of insufficient symptom relief, the use of various elements of biofeedback is recommended. This is based on the numerous (mainly uncontrolled) trials and low associated morbidity.

Rectal balloon training

RBT is not a widely used treatment modality in the Netherlands, probably because its effectiveness is largely unknown. However, the suggested working mechanism seems promising for (selected) patients with FI.

The inclusion criteria for our trial (chapters 5 and 6) were intended to generate a homogeneous group with maximum sensitivity to the interventions under study, viz. PFMT and RBT. Performing PFMT requires at least contraction of the pelvic floor muscles and anal sphincter. We therefore excluded patients having no squeeze pressure, since electrical stimulation is more suitable for these patients (chapter 1).⁴⁶

As RBT aims to improve rectal sensitivity, we considered including only patients with poor sensitivity, but we were unable to define poor sensitivity in advance. Rectal sensitivity can be assessed in several ways (rectal capacity measurement, threshold of rectal sensation), and defining poor sensitivity is hampered by the lack of an accepted method with clear cut-off values (chapters 1 and 6). Moreover, methods and protocols differ across medical centres.

RBT with PFMT was equally effective as PFMT alone (chapter 6). Based on secondary outcomes, however, our findings suggest that RBT may be beneficial for patients with urge FI and patients with rapid fatigue of the external anal sphincter. In addition to the physiological effects, it is promising that adding RBT to PFMT resulted in better perceived outcomes and beneficial changes in lifestyle adjustments. The ideal method to identify patients who would benefit most from the addition of RBT would be subgroup analyses for every type of FI and category of severity. We were unable to perform such subgroup analyses, however, because of lack of power. Adding RBT to PFMT could enhance results in the short term, especially for patients with impaired urgency control, but whether RBT is the overriding factor in symptom improvement needs further exploration. The limited evidence from previous studies,^{4,49} combined with the results of our trial, suggests promising opportunities for RBT, but will not change the existing recommendations as yet.

Factors predictive of physiotherapy outcome

In chapter 8 we identified a small set of factors that predict a favourable outcome of physiotherapy treatment, based on the total sample of included patients. Longer time since the onset of FI appeared to be associated with unfavourable outcome, whereas the use of constipating medication, any obstetric factor and less embarrassment predicted a favourable outcome. Since these factors are identified in an early stage of the diagnostic process, it is useful to keep them in mind when targeting interventions and counselling patients. In addition to these identified predictive factors, agreement exists on several factors that seem to influence the prognosis and outcome of pelvic physiotherapy, such as patient-therapist interaction² and patient motivation.⁵⁰ It is also recognised that pelvic physiotherapy may not be appropriate for patients with neurological deficits involving severely impaired rectal sensitivity and motor control.¹³ Furthermore, institutionalised patients with substantial co-morbidity or cognitive impairments are unlikely to benefit from physiotherapy (chapter 1).

Adherence to the treatment programme is a prerequisite for evaluating the real effect of physiotherapy treatment.⁴⁷ Adherence can be defined as ‘the extent to which patients follow the instructions they are given for prescribed treatments’. Short-term supervised adherence should be distinguished from long-term non-supervised adherence, as they are characterised by different barriers and skills.⁵¹ Adherence can be promoted by influencing self-efficacy through health education (written personal exercise advice, setting realistic goals, information about possible relapse, feedback). Long-term adherence, sometimes lifelong, is particularly relevant as inactivity will make the muscles revert to their pre-trained status and make symptoms reoccur.⁵² Therefore, it may be worthwhile to optimise adherence and treatment outcome and to implement a ‘reminder therapy’: a regular check-up to prevent recurrence (Figure 1; H). One might consider a brief history-taking and diagnostic check-up to elucidate whether the diagnosis and circumstances (e.g. comorbidity) are unchanged, followed by a short treatment period, or in the case of a changed situation, referral to the general practitioner for further management.^{51,53}

Additionally, an intensive protocolised physiotherapy programme, in terms of number of treatment sessions and total duration in weeks, based on strength training principles, supports the idea of a dose-response relation.⁵⁴ Effective training dosage for strength training targets specific muscles with 8–12 slow-velocity, close to maximum contractions, performed three times a day at least 2–3 times a week for more than five months.⁵⁴ In addition to increasing strength, a proper PFMT programme should include endurance and coordination training while giving attention to applications in daily life situations.⁵²

It is not possible to predict the likely outcome of physiotherapy treatment on the basis of diagnostic tests assessing FI.⁵⁵ This suggests that most currently available additional tests have no value in terms of predicting the results before patients are referred for physiotherapy, although they might be more useful in selecting patients for surgical treatment options.⁵⁵ It is therefore recommended that patients receive physiotherapy

treatment as a first-line approach, because it has no adverse effects and entails relatively low costs, as has also been suggested by others.⁵⁶⁻⁵⁸

Another strategy, not fully evaluated yet, is to establish a 'pilot treatment' (Figure 1; E).⁵⁹ Such a pilot treatment acknowledges the lack of certainty about the most optimal programme, including the number of sessions and the exercise protocol, and the fact that factors predicting successful outcome need to be confirmed. Thus, a pilot treatment can be regarded as a diagnostic tool that can guide treatment, even if physiotherapy fails.⁶⁰ A pilot may prove cost-effective and save resources, provided that it is clear how many sessions are at least required to observe improvement.

SELECTING AN OUTCOME MEASURE FOR EVALUATING PHYSIOTHERAPY TREATMENT

If we want outcomes to be accepted as primary outcomes in clinical studies, we need to know their psychometric properties, including the extent to which changes in scores on the instrument reflect changes in health status that the patient would consider important. So far, no consensus exists about the ideal outcome measure to evaluate physiotherapy treatment for FI. Since changes in diagnostic measures do not consistently correlate with symptom improvement, this supports the suggestion that diagnostic measures should not be used as a primary outcome.¹⁵ Nevertheless, they are often used as secondary outcomes, to determine the underlying cause of FI and to guide clinical decision making. Furthermore, summary scores acknowledge that the severity of FI is more complex than is assumed by grading scales. We evaluated the Vaizey and Wexner summary scores, which were originally developed by clinicians. It has been argued that clinical scores focus too heavily on quantitative measurements, whereas subjective improvement and impact on quality of life should be the basis of any evaluation of FI.¹⁵ The results of our study suggest that the Wexner score is most suitable for severity assessment and the FIQL for evaluating quality of life. It is however useful to include a combination of measures for evaluation, such as a severity scale, a subjective rating of improvement and a condition-specific quality of life instrument. Obviously, developing an outcome measure that meets the high quality standards for psychometric soundness is a research area that deserves more attention, especially as we currently base our conclusions about the effectiveness on outcomes that do not yet meet the highest standards of psychometric soundness.

IMPLICATIONS FOR CLINICAL PRACTICE

Implications for clinical practice and recommendations for further research are summarised in a simplified diagram in Figure 9.1.

National practice guidelines and quality indicators

Current pelvic physiotherapy practice is going through major new developments. In 1995, the Royal Dutch Society for Physical Therapy (KNGF) implemented a quality policy, which includes the development and implementation of evidence-based national practice guidelines to improve the quality and efficiency of care.⁶¹ Currently, an Evidence-Based Statement for Faecal Incontinence is being developed. This Evidence-Based Statement is a shortened version of a clinical guideline, which is a review of current knowledge about the diagnostic and therapeutic options in FI. The development of this statement is relevant as it provides evidence about the effectiveness of physiotherapy treatment, supports physiotherapists in their decision-making process and reduces differences in practice routines. Moreover, it demonstrates the value of physiotherapy for patients with FI to insurance companies, health policy makers, other medical disciplines and patients. The additional evidence presented in this thesis will be integrated in this Evidence-Based Statement (Figure 1; E). After completion, translation of the statement will make it accessible internationally and stimulate further development in the process of updating the guidelines.

Particular attention should be devoted to the implementation of the Evidence-Based Statement to overcome the gap between actual practice and practice as proposed by the statement. The use of quality indicators has been suggested to evaluate guideline adherence.⁶² A distinction can be made between structure, process and outcome indicators.⁶³ Process indicators are measurable elements of health care processes for which there is evidence or consensus that they are indicative of the quality of health care.^{62,63} Process indicators should preferably be derived systematically from guidelines. Insurance companies, patients and health care providers may be interested in either outcome indicators (results of care provided, such as the patient's health status) or process indicators (focusing on the diagnostic and therapeutic process). Several studies illustrate the process of developing quality indicators.^{62,64,65} Their results may serve as an example for developing specific quality indicators for physiotherapy in pelvic floor dysfunctions. In addition to evaluating guideline adherence, quality indicators are used by insurance companies to develop a pay-per-performance system. Quality indicators can also contribute to transparency of care because of their ability to measure quality of care, and they can help professionals to review and improve their clinical practice routines. The use of quality indicators can be promoted provided that the data gathering is organised, for example, by means of web-based electronic patient dossiers (EPDs). Such EPDs are being developed based on guidelines to be used by physiotherapists to standardise the diagnostic and therapeutic process, which will stimulate evidence-based practice. After completion of the Evidence-Based Statement on FI, this statement can provide the starting point for optimising EPDs for FI (Figure 1; B). The problem with FI is that it is mostly associated with other pelvic floor dysfunctions. The challenge today is to build an EPD for FI that also includes the option of recording associated pelvic floor dysfunctions without burdening the therapist with an unnecessarily extensive EPD taking too much time to complete. If EPDs are to offer

added value and be successfully implemented, there is a need for facilitating strategies such as informing and instructing therapists, synchronising software from various providers, and arranging technical and financial support.

Multidisciplinary incontinence centre

As FI is a multifactorial problem with significant co-occurrence of other pelvic floor dysfunctions, a multidisciplinary approach has been recommended,¹³ for instance organised in a professional facility like a multidisciplinary incontinence centre (Figure 1; D).⁶⁶ Expenditures on incontinence materials have risen rapidly in recent years, up to 160 million euros,⁶⁷ and might rise further due to the ageing population. Optimising FI management might decrease expenditures on these materials. Some measures to face this challenge have been discussed in this thesis, such as organising FI management in multidisciplinary incontinence centres, proper patient selection for physiotherapy, identifying patients with sphincter rupture who are at risk for postpartum FI in the short or long term, recommendations for treatment, contributing to guideline development, and optimising outcome measures for research. If physiotherapy treatment can indeed reduce the expenditures on incontinence materials and/or treatment, health insurance companies might be willing to cover its costs.

RECOMMENDATIONS FOR FURTHER RESEARCH

It is generally known that only one third of patients with FI seek help. Although the diagnostic and therapeutic pathway can be optimised, as long as patients do not seek help they will not benefit from it (Figure 1; F). Although not easy, a first important step would therefore be to trace those patients that rely on incontinence absorption materials or do not consult their doctor. This could be achieved by displaying information leaflets in doctors' waiting rooms, referring people to patient associations, such as the Dutch 'Stichting Bekkenbodem Patiënten', or to informative websites, such as www.bekkenbodemonline.nl. Above all, all health care professionals should recognise signs when they see patients and actively ask them about their problems.⁸ Furthermore, emphasis should be placed on 'cure' instead of 'care', so providers of incontinence materials, who probably have other interests, should be contacted to screen their customer system for patients who may benefit from further diagnostic work-up.

General practitioners are the gate-keepers of the Dutch health care system. When patients ultimately decide to visit a general practitioner, it is important that the problem is discussed and appropriate care is provided. As both general practitioners and physiotherapists play important roles in the management of patients with FI, it is suggested to develop national agreements on collaboration in primary care (Landelijke Eerstelijns Samenwerkings Afspraken or LESA) through joint efforts by the Dutch General Practitioners Society (NHG) and the Royal Dutch Society for Physical Therapy (KNGF), as has already been done for urinary incontinence (Figure 1; A).⁵³ These

agreements should synchronise policies between the two disciplines as well as account for differences between the disciplines regarding tasks and responsibilities.

Since optimal management may be hampered by the time constraints that all physicians face, the value of employing continence advisors should be further explored (Figure 1; G).^{9,68}

A recent consensus report defined FI as the complaint of involuntary loss of faeces (including liquid and solid stools).⁶⁹ There is a need for consistent use of this definition to allow comparisons between prevalence figures and trial outcomes, and to facilitate communication among health care providers or insurance companies. Another aspect, the amount of leakage, is difficult to define, but this factor does contribute to the severity of FI, as it matters whether a patient loses the entire contents of their bowel or just a small amount. Therefore, efforts should be undertaken to define and classify the amount of leakage.

Finally, the effectiveness of rectal balloon training and predictive factors for its success should be further explored among populations not represented in our trial, such as male populations.

CONCLUDING COMMENTS

Pelvic physiotherapy has seen rapid developments in recent years, and this thesis has provided new suggestions to optimise the management of FI patients.

I started this thesis by explaining that faecal incontinence is a disorder which can definitely be considered as a ‘What if it happened to you’ kind of affliction. The thesis has tried to show that FI is a common, but also treatable disorder. Treatment should not only aim at symptom relief, but undoubtedly also involve attempts to improve the patients’ quality of life. Although the field of FI is showing progress, further research is needed to achieve an optimal diagnostic and treatment pathway for FI. I hope that this thesis will inspire many researchers in the near future to invest effort in developing and optimising further research in the interesting field of FI.

Figure 9.1 Simplified flow chart of multidisciplinary relations and suggestions for further improvement of clinical practice.

- A. Development and implementation of national agreements on collaboration in primary care (Landelijke Eerstelijns Samenwerkings Afspraak (LESA))
- B. Electronic Patient Dossier (EPD)
- C. Selection of patients
 - One-time physiotherapy consultation
- D. Multidisciplinary care organised in specialised centres
- E. Development and implementation of Evidence-Based Statement
 - Pilot treatment
- F. Increase percentage of patients who seek help
- G. Continence advisor
- H. Reminder therapy

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SUMMARY

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The main aim of the work summarised in this thesis was to present new evidence concerning different aspects of the management of patients with faecal incontinence (FI), in order to enable physicians and physiotherapists to provide competent individual counselling and treatment to patients with FI who suffer from it. To reach this goal several topics were addressed. First, the state of the art for various aspects of the management of patients with FI was examined. Subsequently, we evaluated which delivery-related factors are important in the development of FI in postpartum women. Furthermore, the psychometric properties and minimally important change of the Vaizey score, Wexner score and Fecal Incontinence Quality of Life Scale for clinical use in patients with FI were assessed. Finally, we studied the effect of adding rectal balloon training to pelvic floor muscle training in patients with FI and investigated the predictive value of patient characteristics on a favourable outcome of physiotherapy treatment.

Chapter 2 presents a review of the state of the art for various aspects of the management of patients with FI. FI is the complaint of involuntary loss of faeces. Anal incontinence is mostly used to include faeces or flatus. Figures on help-seeking behaviour confirm the enormous embarrassment felt by patients affected by it; only one third of patients have the courage to consult a physician. Cross-sectional studies have shown that about 2–24% of people suffer from FI (incontinence for liquid and solid stools), although these rates are likely to be underestimated. The aging of the population will make FI an increasingly important socioeconomic problem in the coming decades, especially since it leads to reduced quality of life and premature institutionalisation, which has important cost implications. Obstetric trauma is one of the major causes of FI in women.

Several diagnostic measures are available to assess the nature and severity of FI or patient-reported quality of life, although guidance for conservative or surgical therapy is not always obvious. If conservative methods, such as pharmacological treatment and/or dietary changes fail, pelvic physiotherapy is usually the next treatment option. It is regarded as safe, inexpensive and well-tolerated. Failure of pelvic physiotherapy is often a criterion for proceeding to surgical management, such as encirclement procedures and sacral nerve stimulation. There is a lack of knowledge about effective modalities of pelvic physiotherapy treatment in FI. Additionally, there are no consistently reported patient characteristics that are likely to predict the greatest benefit from physiotherapy treatment, which is relevant for medical decision making and patient counselling. A wide variety of outcome measures are available to evaluate treatment, such as severity scores, quality of life scales, subjective perception of relief and diagnostic measures, although no consensus exists on the optimal outcome measure. The review outlined in this chapter led to the formulation of the different research topics and aims as presented in this thesis.

Conflicting results are reported about the contribution of maternal, obstetric and foetal characteristics to postpartum FI. **Chapter 3** describes the results of a systematic review to identify delivery-related aetiological factors for postpartum FI. We performed a qualitative analysis of 31 prospective cohort studies. This systematic review, including only longitudinal studies and recognising the importance of separating results for different outcomes, identified that a third- or fourth-degree sphincter rupture was the only factor, which is strongly (anal incontinence) or moderately (flatus incontinence) associated with postpartum FI. No association with other postulated risk factors was found, e.g. high birth weight or instrumental delivery. We have to keep in mind that the potential co-existence of different risk factors impedes the interpretation of the influence of a single delivery-related risk factor.

The design of studies evaluating effectiveness of FI treatment is problematic as no well-validated outcome measures are available. A frequently used outcome measure to evaluate the severity of FI is the 7-item Vaizey score. However, its psychometric properties are unclear and there is a lack of information on the clinically meaningful interpretation of change. The study described in **chapter 4** was conducted as part of a prospective cohort study assessing the outcome of pelvic floor rehabilitation in patients with FI. One hundred ninety-four patients were analysed, 53 of whom provided data on the global perceived effect score. Pelvic floor rehabilitation resulted in a significant reduction of the total Vaizey score and most individual items. The total score changed in agreement with the global perceived effect scores and appeared responsive. The test-retest reliability was adequate or excellent for six individual items and the total score. The internal consistency was low for the total Vaizey score at baseline. The estimates for the minimally important change and smallest detectable change yielded moderately consistent results. A minimally important change of -5 points seemed preferable and yielded the lowest misclassification rate. We conclude from our results that the psychometric properties of the total Vaizey score and its individual items are not convincing. The Vaizey score is in the early stage of development and future research is required to confirm conclusions on the psychometric properties to justify its use in research and routine clinical practice.

Two physiotherapy interventions, rectal balloon training and pelvic floor muscle training, are used in the management of FI. However, evaluating their effectiveness has been impeded by lack of high quality studies, lack of standardisation and a large variety in outcome measures. **Chapter 5** describes the design of a two-armed single-blind randomised controlled trial, which aims to assess the effectiveness of rectal balloon training combined with pelvic floor muscle training, compared to pelvic floor muscle training alone. The trial was conducted at the outpatient department of the Maastricht University Medical Centre. Eligible participants were (1) adults (aged ≥ 18 years), (2) with FI complaints due to different aetiologies persisting for at least six months, (3)

having a Vaizey score of at least 12, and (4) failure of conservative treatment (including dietary adaptations and pharmacological agents). Primary outcome was based on the Vaizey score. Follow-up measurements were scheduled at three, six and 12 months after inclusion. Pelvic physiotherapists administered twelve sessions during three months according to a standardised protocol. Furthermore, this chapter discusses the actions taken to minimise bias and confounding in the design, explains the choice for two treatment groups, discusses the secondary goals of this study and indicates the impact of this trial on clinical practice.

Chapter 6 presents the short-term effectiveness of rectal balloon training as an add-on therapy to pelvic floor muscle training. We randomised eighty patients and analyses were by intention to treat. Forty patients were assigned to combined rectal balloon training with pelvic floor muscle training and 40 to pelvic floor muscle training alone. Rectal balloon training with pelvic floor muscle training was equally effective as pelvic floor muscle training alone. Secondary outcomes favouring rectal balloon training were: Lifestyle subscale of the Fecal Incontinence Quality of Life Scale, global perceived effect, maximum tolerable volume and fatigue of the external anal sphincter. Overall, 50% of patients were considered improved according to the minimally important change (Vaizey score reduction ≥ 5 points), so referral to physiotherapy interventions should be considered before surgery.

Patient selection on the basis of predictive factors will help to improve the management of patients with FI. So far, it is unclear which patients are likely to benefit from physiotherapy treatment, which is relevant for medical decision making and patient counselling. **Chapter 7** reports on secondary analyses of data from the randomised controlled trial and aims to identify patient characteristics predicting a favourable outcome of pelvic physiotherapy treatment. Candidate predictors were obtained from patient demographics, clinical and physical examination, and questionnaires. Favourable outcome was defined as a combination of: (1) Vaizey score reduction ≥ 5 points, and (2) 'slightly' to 'very much improved' on the nine-point global perceived effect score. Predictors were identified by univariable and multivariable logistic regression analyses. Multivariable analysis showed that longer time since FI onset appeared to be associated with an unfavourable outcome, whereas the use of constipating medication, any obstetric factor, and experiencing minor embarrassment predicted a favourable outcome. This small subset of identified predictors for a favourable outcome may be relevant in targeting physiotherapy treatment more efficiently, especially as they are available early in the diagnostic process.

In **chapter 8** we determined, based on data from the randomised controlled trial, the psychometric properties and minimally important change of the Vaizey score, Wexner score and the Fecal Incontinence Quality of Life Scale for use in the evaluation of patients with FI. The patient-reported outcomes were tested for internal and external

responsiveness, longitudinal construct validity and interpretability. All total scores proved to have both adequate to excellent responsiveness and longitudinal construct validity, and changes were in agreement with subjective improvement. Due to variability in the estimates of the minimally important change, they should be used as indicators. All patient-reported outcomes showed psychometric or practical limitations. The instruments available to date to evaluate severity and quality of life in FI do not yet attain the highest levels of psychometric soundness. As the focus of patients (embarrassment, fear, preventive measures) may differ from that of physicians (frequency and amount of lost stool, type of FI), it is recommended to include several measures for evaluation. So far, there are suggestions that the Wexner score is most suitable for severity assessment and the Fecal Incontinence Quality of Life Scale for evaluating quality of life.

Chapter 9 presents an overview of the main findings of the performed studies, discusses important strengths and weaknesses, and positions the results of the studies in the context of other scientific evidence in the field of FI.

SAMENVATTING

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Het doel van de verschillende studies in dit proefschrift was een bijdrage te leveren aan de evidentie met betrekking tot de diagnostiek en behandeling van patiënten met fecale incontinentie en zodoende medici en fysiotherapeuten relevante instrumenten in handen te geven bij advisering en behandeling van deze patiënten.

Hoofdstuk 2 geeft een overzicht van de huidige stand van zaken betreffende verschillende aspecten van de diagnostiek en behandeling van patiënten met fecale incontinentie. Fecale incontinentie is gedefinieerd als het klagen over onvrijwillig verlies van ontlasting (incontinentie voor vloeibare en vaste ontlasting). Flatus incontinentie is gedefinieerd als het klagen over onvrijwillig verlies van flatus. De term anale incontinentie wordt gebruikt als we spreken over fecale en flatus incontinentie. Slechts één derde van de patiënten zoekt hulp voor hun klachten. Dit onderstreept de forse schaamte die patiënten met fecale incontinentie ervaren. Transversaal onderzoek toont aan dat ongeveer 0,4–18% van de populatie last heeft van fecale incontinentie, wat een onderschatting lijkt te zijn. In de komende decennia zorgt de vergrijzing ervoor dat fecale incontinentie een belangrijk sociaal-economisch probleem gaat vormen, vooral ook door sociale isolatie, verminderde kwaliteit van leven en vroegtijdige opname in verpleeg- of verzorgingshuis, resulterend in stijgende kosten. Niet alleen op hogere leeftijd komt fecale incontinentie voor, maar het treft vooral ook jonge vrouwen, met name na een bevalling met complicaties, zoals een inscheuring.

Verschillende diagnostische meetinstrumenten zijn voorhanden om de aard en ernst van fecale incontinentie vast te stellen, hoewel de resultaten niet altijd een leidraad vormen voor verder conservatief behandelen of chirurgie. Als conservatieve behandeling (waaronder het gebruik van constiperende medicatie, dieetaanpassingen, advies over leefstijl of toiletgedrag) onvoldoende leidt tot vermindering van symptomen, wordt bekkenfysiotherapie beschouwd als een eerste optie, omdat het een relatief goedkope behandeling is zonder bijwerkingen. Falen van bekkenfysiotherapie is vaak een criterium om over te gaan tot chirurgische behandeling, zoals de dynamische gracilis plastiek en sacrale zenuwstimulatie. Hedendaags is er onvoldoende bewijs voor de effectiviteit van de verschillende bekkenfysiotherapeutische behandelmodaliteiten bij fecale incontinentie, i.c. electrostimulatie, biofeedback (waaronder rectale ballontraining) en bekkenbodemspiertraining. Daarnaast is het onduidelijk welke patiënten het meest gebaat zijn bij bekkenfysiotherapie. Meer duidelijkheid hierover faciliteert de medische besluitvorming en advisering aan patiënten. Er is een groot aantal meetinstrumenten voorhanden voor evaluatie van behandeling, zoals ernstscores, kwaliteit van leven, subjectief ervaren effect en diagnostische testen, maar er bestaat nog onvoldoende consensus over welk meetinstrument het meest geschikt is. De review beschreven in dit hoofdstuk heeft de basis gevormd voor het formuleren van de onderzoeksvraagstellingen van de studies in dit proefschrift.

In de huidige literatuur worden conflicterende resultaten gerapporteerd met betrekking tot de onderlinge bijdrage van bevallingsgerelateerde factoren (obstetrische, moeder- en kindfactoren) die kunnen leiden tot de ontwikkeling van postpartum fecale incontinentie. **Hoofdstuk 3** beschrijft de resultaten van een systematische review waarin is gekeken naar bevallingsgerelateerde etiologische factoren voor postpartum fecale incontinentie. Er is een kwalitatieve analyse van 31 prospectieve cohort studies uitgevoerd. Alleen longitudinale studies zijn geïnccludeerd en er is rekening gehouden met het uitsplitsen van de resultaten voor verschillende uitkomsten. Een derde- of vierdegraads sfincter ruptuur bleek de enige etiologische factor te zijn, die sterk (anale incontinentie) en matig (flatus incontinentie) geassocieerd is met postpartum fecale incontinentie. We vonden geen bewijs voor een verband met andere veronderstelde risicofactoren, zoals een hoog geboortegewicht of een kunstverlossing. Bij de interpretatie van de resultaten moet worden rekening gehouden met het feit dat verschillende risicofactoren tegelijkertijd kunnen optreden, waardoor het vaststellen van de invloed van een individuele bevallingsgerelateerde risicofactor wordt bemoeilijkt.

Er is gebrek aan valide uitkomstmaten voor het evalueren van effectstudies bij fecale incontinentie. De 7-item Vaizey score wordt vaak gebruikt om de ernst van fecale incontinentie in kaart te brengen, hoewel de psychometrische eigenschappen en de interpretatie van een verandering in de score onduidelijk zijn. De studie beschreven in **hoofdstuk 4** is uitgevoerd als onderdeel van een grootschalige prospectieve cohort studie, die als doel had het effect van bekkenfysiotherapie bij patiënten met fecale incontinentie te onderzoeken. Van de 194 onderzochte patiënten waren er van 53 patiënten data over hun subjectieve oordeel aangaande het resultaat van bekkenfysiotherapie (globaal ervaren effect). Bekkenfysiotherapie resulteerde in een significante verlaging van de totale Vaizey score en de meeste individuele items van de Vaizey score. De totale score bleek te veranderen in overeenstemming met het globaal ervaren effect. De totale score was responsief, hoewel de individuele items inconsistente bevindingen lieten zien. De test-retest betrouwbaarheid was voldoende tot uitstekend voor zes individuele items en de totale score. De interne consistentie was laag voor de totale Vaizey score op baseline. De schattingen voor de minimaal *belangrijke* verandering en de minimaal *detecteerbare* verandering leverden matig consistente resultaten op. Een minimaal belangrijke verandering van 5 punten verbetering lijkt het meest aannemelijk en levert het laagste percentage misclassificatie op. De conclusie is dat de Vaizey score nog in een ontwikkelfase verkeert en verder onderzoek nodig is om onze conclusies ten aanzien van de psychometrische eigenschappen te bevestigen en zodoende het routine gebruik van de Vaizey score voor onderzoek en de klinische praktijk te kunnen rechtvaardigen.

Twee bekkenfysiotherapeutische behandelmodaliteiten, namelijk rectale ballontraining en bekkenbodemspiertraining, worden gebruikt voor de behandeling van patiënten met fecale incontinentie. Onderzoek naar de effectiviteit van beide modali-

teiten wordt bemoeilijkt door gebrek aan studies met voldoende methodologische kwaliteit, gebrek aan standaardisatie tussen de studies en grote variatie in uitkomstmaten.

Hoofdstuk 5 beschrijft het design van een gerandomiseerde interventiestudie, met als doel de effectiviteit van rectale ballontraining gecombineerd met bekkenbodemspiertraining te vergelijken met alleen bekkenbodemspiertraining. Inclusiecriteria waren: (1) ≥ 18 jaar, (2) ≥ 6 maanden fecale incontinentie klachten door verschillende oorzaken, (3) Vaizey score ≥ 12 (range 0–24), en (4) falen van dieetaanpassingen en medicatie. Baseline metingen bestonden uit de Vaizey score (primaire uitkomstmaat), medische voorgeschiedenis, lichamelijk onderzoek, medicatiegebruik, anorectale manometrie, rectale capaciteitsmetingen, anorectale sensibiliteitsmeting, anale endoechografie, defecografie, symptoom dagboek, Fecal Incontinence Quality of Life Scale en de PREFAB-score. Follow-up metingen werden ingepland na 3, 6 en 12 maanden na inclusie van de patiënten. Alle patiënten werden verwezen voor 12 behandelingen bekkenfysiotherapie gedurende drie maanden volgens een gestandaardiseerd protocol.

Hoofdstuk 6 beschrijft de korte termijn resultaten van de interventiestudie. Uiteindelijk zijn 80 deelnemers gerandomiseerd. Daarvan kregen 40 deelnemers 12 behandelingen met rectale ballontraining en bekkenbodemspiertraining en 40 deelnemers kregen alleen bekkenbodemspiertraining. De groep met rectale ballontraining als toevoeging op bekkenbodemspiertraining verbeterde één punt meer op de Vaizey score. Dit verschil was niet significant. De volgende secundaire uitkomstmaten gaven wel een significant verschil ten gunste van rectale ballontraining: de subschaal 'Lifestyle' van de Fecal Incontinence Quality of Life Scale, globaal ervaren effect, maximum tolereerbaar volume en uitputting van de externe anale sfincter. Gezien het feit dat 50% van alle patiënten 5 of meer punten verbeterden op de Vaizey score, verdient het aanbeveling bekkenfysiotherapie te overwegen voordat verwezen wordt voor chirurgie.

Patiëntenselectie, gebaseerd op predictieve factoren, zal de behandeling van patiënten met fecale incontinentie bevorderen. Er bestaat onduidelijkheid over welke patiënten het meest gebaat zijn bij bekkenfysiotherapeutische behandeling, terwijl dat wel relevant is voor medische besluitvorming en advisering aan patiënten. **Hoofdstuk 7** rapporteert over secundaire analyses van data van de interventiestudie en heeft als doel patiëntkenmerken te identificeren die succes van bekkenfysiotherapie kunnen voorspellen. Geselecteerde predictoren komen voort uit de demografische variabelen, klinisch en lichamelijk onderzoek, en vragenlijsten. Succes van bekkenfysiotherapie was gedefinieerd als een combinatie van: (1) verlaging van tenminste 5 punten in de Vaizey score, en (2) 'iets beter' tot 'heel veel beter' op de 9-punts globaal ervaren effect score. Univariabele en multivariabele logistische regressie analyses zijn gebruikt om predictoren te identificeren. De multivariabele analyse liet zien dat een langere duur van klachten was geassocieerd met het ontbreken van succes van bekken-

fysiotherapie, terwijl het gebruik van constiperende medicatie, obstetrische risicofactoren en minder schaamte waren geassocieerd met succes van bekkenfysiotherapie. Deze kleine set van predictoren kan relevant zijn bij het zo efficiënt mogelijk inzetten van bekkenfysiotherapie, vooral omdat de geïdentificeerde predictoren vroegtijdig in het diagnostische proces bekend zijn.

In **hoofdstuk 8** zijn de psychometrische eigenschappen en minimaal belangrijke verandering van de Vaizey score, Wexner score en Fecal Incontinence Quality of Life Scale onderzocht voor gebruik bij de evaluatie van patiënten met fecale incontinentie, gebaseerd op de data van de interventiestudie. De vragenlijsten zijn beoordeeld op interne en externe responsiviteit, longitudinale construct validiteit en interpreteerbaarheid. Alle totaalscores bleken voldoende tot uitstekende responsiviteit en longitudinale construct validiteit te hebben. Veranderingen in de scores waren in overeenstemming met subjectieve verbetering. Wegens variatie in de schattingen voor de minimaal belangrijke verandering moeten deze worden opgevat als een indicatie. Het kiezen van een bepaalde waarde voor de minimaal belangrijke verandering hangt onder meer samen met de consequenties voor de patiëntenzorg. Verder vertonen de huidige vragenlijsten, die de ernst van fecale incontinentie en kwaliteit van leven bij patiënten met fecale incontinentie evalueren, psychometrische of praktische beperkingen. Doordat de focus van de patiënt (schaamte, angst, preventieve maatregelen) kan verschillen van die van een arts (frequentie en hoeveelheid van verloren ontlasting, type fecale incontinentie), wordt aanbevolen bij de evaluatie verschillende meetinstrumenten te betrekken. Het lijkt dat de Wexner score het beste kan worden ingezet om de ernst van fecale incontinentie te bepalen en de Fecal Incontinence Quality of Life Scale bij het evalueren van kwaliteit van leven.

Hoofdstuk 9 geeft een overzicht van de belangrijkste bevindingen van de uitgevoerde studies. Verder worden deze bevindingen bediscussieerd en geplaatst in de context van ander wetenschappelijk bewijs en ontwikkelingen op het gebied van fecale incontinentie.

DANKWOORD

De klassieker Forrest Gump is niet alleen een mooie film maar bracht ook een fantastische zin voort, één van mijn persoonlijke favorieten:

'Life is like a box of chocolates... you never know what you're gonna get.'

Hoe waar is deze zin wel niet als je gaat promoveren. Hoe vaak komt het immers niet voor dat die hele mooie bonbon ineens vieze kersenlikeur blijkt te bevatten. Of het tegenovergestelde is waar: die minder lekker uitziende bonbon heeft een voortreffelijke zoete vulling. Waargemaakte verwachtingen en het onverwachte maken deel uit van een promotietraject. En denkend aan de afgelopen jaren zou ik voor elk soort bonbon wel een toepasselijke situatie kunnen bedenken. Gelukkig kan ik stellen dat mijn box aan herinneringen voornamelijk bestond uit mooie bonbons met mooie vullingen, mede door de verschillende personen die hebben bijgedragen aan de totstandkoming van dit proefschrift.

Ten eerste wil ik mijn begeleiders bedanken.

Beste Rob, bedankt voor alle inzet die je hebt getoond om dit proefschrift tot een goed einde te brengen! Niets was je teveel en je hebt er alles aan gedaan mijn project zo goed en prettig mogelijk te laten verlopen. Onze wekelijkse afspraak op maandag morgen gebruikten we om even de belevenissen van het weekend de revue te laten passeren, koffie te drinken, en natuurlijk verschillende zaken rondom mijn onderzoek te bespreken. Jouw enthousiasme, bron van kennis en relativiseringsvermogen maakten onze afspraak een uitstekend begin van de week. Ik ben heel blij dat we onze samenwerking in de toekomst kunnen voortzetten.

Beste Cor. Als fysiotherapeut en nog onervaren in de wereld die wetenschap heet, kwam ik bij u om kennis te maken. Al snel werd duidelijk hoe klein de wereld is, want ik bleek op de middelbare school bij uw dochter in de klas te hebben gezeten en uw zoon is als fysiotherapeut een collega van mij geweest. Terwijl u een sigaartje rookte en ik uw rol met pepermuntjes op at, heeft u mij ontzettend veel bijgebracht over de beperkingen die patiënten met fecale incontinentie ervaren en hoe gecompliceerd de zorg rondom deze patiënten is. U kon hier vol enthousiasme legio verhalen over vertellen. Daarnaast bood u mij de kans om me te verdiepen in de chirurgische behandeling. Zo heb ik verschillende indrukwekkende operaties mogen bijwonen.

Verder bedankt voor alle inzet die nodig is geweest voor het includeren van alle patiënten voor het onderzoek, met daarna nog het uitvoeren van alle nametingen. Zonder uw tomeloze inzet was de PhysioFIT-studie nooit geweest wat het nu is geworden!

De eindstreep hebben we met het hele begeleidingsteam samen gehaald, en de medaille is zeker ook voor u, al komt dit officieel gezien misschien niet naar voren. Ik denk met veel plezier terug aan onze fijne samenwerking en hopelijk kunt u binnenkort gaan genieten van een welverdiend pensioen samen met uw vrouw en kinderen.

Beste Bary, tijdens mijn werkzaamheden in jouw praktijk, was jij het, die mij voorstelde een promotietraject te gaan beginnen. Dat ik deze weg zou inslaan, had ik me van tevoren nooit voorgesteld, maar al snel was ik overtuigd van de relevantie van het project en heb ik de stap gezet. Bedankt voor deze introductie en al je inspanningen om mijn promotietraject tot een succes te maken. Jouw aanstekelijke karakter, veelzijdigheid aan ideeën en schat aan ervaring werkten voor mij stimulerend en verhelderend. Merci voor diene input, ammesante congres bezoeken en rotsvas vertrouwde in me! Veer zien us.

Beste Erik, ik ben erg blij dat jij uiteindelijk mijn promotieteam kwam versterken! Jouw kennis en ideeën bleken onmisbaar. Ondanks dat jij het verste weg woont, stond je altijd klaar om mij van advies te voorzien, analyses en manuscripten door te nemen en te sparren over van alles en nog wat. Ik denk met veel plezier terug aan onze gezamenlijke besprekingen, uitjes en congresbezoeken. Jouw inzet, gevoel voor humor en opbeurende woorden heb ik erg gewaardeerd en ik hoop in de toekomst nog vaak met jou te mogen samenwerken.

De leden van de beoordelingscommissie, Prof. dr. A.A.M. Masclee, Dr. R.J.F. Felt-Bersma, Dr. G.A. van Koeveringe, Prof. dr. M.H. Prins wil ik danken voor het lezen, beoordelen maar vooral voor het goedkeuren van mijn proefschrift.

Prof. dr. S. Mørkved. I feel honoured for your willingness to participate in the committee for evaluation of this thesis.

Jarno, Bas en Bart, bedankt voor alle moeite die jullie hebben gedaan om de patiënten in te plannen en de nametingen te voltooien. Jullie waren altijd bereikbaar en bereid om mij te helpen waar nodig. Jarno en Bart, hopelijk is jullie promotie in de nabije toekomst ook een feit.

Jos en Harrie, de uitdrukking 'voor een complete chaos heb je een computer nodig' was helaas ook weleens op mij van toepassing. Door jullie snelle hulp en dito oplossing kon ik altijd weer snel aan de slag. Jos, bedankt voor het uitvoeren van de dubbele data-entry, en de ontelbare keren dat je mee hebt geholpen te kopiëren en klappers in te binden. Onze gesprekken over verre reizen maakten dit klusje in elk geval veel minder saai!

Conny, bedankt voor het randomiseren van alle patiënten en de logistieke hulp. Het is fijn om te weten dat iemand goed het overzicht weet te bewaren.

Yvonne, bedankt dat je de lay-out van mijn proefschrift hebt verzorgd. Door jou ziet het er nu piekfijn uit!

Jan Nijhuis, Marije Deutekom, Wim van Gemert en Martijn Heymans. Als co-auteurs wil ik jullie bedanken voor jullie bijdrage en kritische blik. Martijn, bedankt dat ik je te

pas en (soms) onpas mocht storen over vragen met betrekking tot de multiële imputatie procedure.

Mischa, bedankt voor het ontwerpen van de cover van dit proefschrift. Hoe concreet mijn ideeën vooraf ook leken, probeer dat maar eens om te zetten tot een geheel. Jij hebt het tot een geheel weten te maken en ik ben erg blij met het resultaat.

Verder wil ik alle collega's en oud-collega's van de afdeling Epidemiologie bedanken. Alle lunchbreaks, babbels bij het koffieapparaat, wandelingen, etentjes en uitjes maken dat ik elke dag met veel plezier naar 'het Deb' ga.

Alle mede-promovendi, van wie een deel nu al gepromoveerd is, wil ik bedanken voor de leuke lunches en uitjes. Ik heb erg veel gehad aan jullie steun en inhoudelijke discussies. Emmylou, jou wil ik in het bijzonder bedanken als 'oud-kamergenoot'. Bedankt voor je plezierige gezelschap en veel succes met jouw promotieonderzoek!

Medewerkers van de FIT-studie, bedankt voor jullie bijdrage en prettige samenwerking.

Verder wil ik alle bekkenfysiotherapeuten bedanken die hebben meegedaan aan de PhysioFIT-studie. Ik ben jullie veel dank verschuldigd, want zonder jullie inzet, feedback, betrokkenheid en enthousiasme was deze studie nooit mogelijk geweest.

Tevens wil ik alle patiënten bedanken die aan de PhysioFIT-studie hebben deelgenomen.

Verder wil ik Medeco B.V. bedanken. Ik waardeer jullie bijdrage aan het onderzoek, in de vorm van het kosteloos aanleveren van de rectale therapieballonnen, enorm.

Yvonne, Nathalie en Petra, hartelijk dank voor alle hand-en-spandiensten.

En dan mijn paranimfen.

Beste Vivian, samen hebben we de opleiding Bewegingswetenschappen en Fysiotherapie doorlopen en hoe toevallig (of niet?) dat we ook nog collega's zijn geworden! Samen met de andere 'fysiodames' hebben we het altijd erg gezellig en het is een feest als iedereen uit alle hoeken van het land weer bij elkaar is. Ik ben zeer vereerd dat jij me bij deze belangrijke dag wil bijstaan. Veel succes met het afronden van jouw promotietraject!

Beste Audrey, jij bent je 's'-je al kwijtgeraakt! Jij bent vanaf het begin mijn kamergenoot (hoewel ik een kort uitstapje heb gemaakt naar een paar deuren verder). Zonder jou zou het werk een stuk minder gezellig zijn. Naast het feit dat we heel wat aflachen, ben je ook een hele steun als het om het serieuze werk gaat. Ik ben er trots op dat jij mijn paranimf bent en gelukkig blijven we voorlopig nog collega's en kamergenoten!

Ik ben ervan overtuigd dat ik kan stellen dat vrienden onmisbaar zijn en ik ben dan ook heel blij dat ik het geluk heb ontzettend lieve mensen om mij heen te hebben. In het bijzonder 'de dansmariekes': bedankt voor jullie trouwe vriendschap en de broodnodige ontspanning. Ik waardeer het enorm dat we altijd voor elkaar klaar staan in voor- en tegenspoed: 'Amigos para siempre'.

Daarnaast wil ik de 'fysiodames' bedanken: Shireen, Vivian B., Vivian W., Ellen en Inez. Wat bijzonder dat we nog steeds contact houden en kunnen bijkletsen over ieders wel en wee!

Lieve (schoon)familie en vrienden, bedankt voor al jullie interesse in mijn onderzoek de afgelopen jaren.

Lieve Bas, Nicole en Joep. Ondanks alles wat jullie (hebben moeten) meemaken staan jullie altijd voor ons klaar en hebben jullie aandacht voor ons. Bedankt voor jullie steun en vriendschap. We gaan er wat moois van maken in de toekomst!

Lieve Ans en Theo, bedankt voor alles wat jullie voor ons doen. Jullie zijn de allerliefste opa en oma voor Rowan, en we kunnen altijd voor alles bij jullie terecht. Niets is jullie teveel en dat waardeer ik enorm. Wat bof ik met jullie!

Lieve Raymond en Cindy, bedankt voor jullie vriendschap, belangstelling en steun. En Raymond, met een tweelingbroer als jou, ben je nooit eenzaam, maar zoals jij het vroeger ooit noemde 'tweezaam'.

Lieve pap en mam. Bedankt voor de geweldige jeugd die ik heb gehad. Een goede basis is het begin en hoe waar is dat. Jullie zijn mijn steun en toeverlaat en de deur staat altijd wagenwijd open. Het is niet voor niets dat jullie telefoonnummer nog steeds in mijn telefoon staat als 'THUIS'. Naast geweldige ouders zijn jullie ook hele lieve grootouders voor Rowan. Iech haw vaan uuch!

En als laatste mijn twee lieve kerels thuis: Erik en Rowan. Lieve Erik, bedankt voor al je hulp en geduld. Zonder jouw liefde, steun en begrip zou mijn proefschrift nooit zijn geworden wat het nu is. Wat ben ik trots als ik jou samen zie met onze lieve knul. Lieve Rowan, wat ben ik blij dat jij in ons leven bent gekomen en daarom sta jij (toen je nog héél klein was) op de cover van mijn proefschrift. Jij bent het zonnestraaltje in huis en het is een feest om na een werkdag weer thuis te komen. Jij maakt ons heel gelukkig. Ik hou van jullie.

Esther

CURRICULUM VITAE

ABOUT THE AUTHOR

Esther Bols was born on October 13, 1979 in Geleen, the Netherlands. She completed secondary school (Gymnasium) at the Jeanne d'Arc College in Maastricht in 1998. In the same year, she started her study Movement Sciences at the Maastricht University. In addition, she started studying Physiotherapy at the Hogeschool Zuyd in Heerlen in 2001. She fulfilled her master internship at the Department of Orthopaedics at the Maasland Hospital in Sittard, the Netherlands, resulting in the master thesis 'early postoperative improvement in cardiovascular fitness after transfusion of autologous blood in total hip arthroplasty'. After graduating from both studies in 2004, she started working as a physiotherapist. From 2005, she was employed as a PhD student at the Department of Epidemiology, Maastricht University (CAPHRI School for Public Health and Primary Care, Maastricht University) on the research project, entitled 'Pelvic physiotherapy in faecal incontinence', resulting in the present thesis. At the same university, she obtained her master degree Health Sciences Research in 2007. Since April 2011, Esther works as a post-doctoral researcher at the Department of Epidemiology, Maastricht University, where she continues her work in the field of incontinence and arthrosis. Esther is married to Erik van Soerland and mother of Rowan.