The effectiveness and acceptability of a computerised guided self-help programme for vaginismus: a mixed methods design

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signature:

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Overview

This volume is separated into three parts:

- **Part 1: Literature review**. A systematic review and meta-analysis of twelve randomised controlled trials that examined the efficacy of psychological treatment for vaginal pain. The aim was to understand whether efficacy of treatment differed for vaginal pain defined as medical or psychiatric in aetiology. Differences and similarities in efficacy were examined on outcomes of pain and sexual function. Effectiveness of psychological treatment was found to be comparable regardless of aetiology, indicating that this distinction may not be helpful for informing treatment decisions.

- **Part 2**: **Empirical paper**. A mixed methods study evaluated a new computerised programme for six women with vaginismus. Change in pain-related fear, penetration behaviour and pain intensity was quantitatively assessed. Interviews were used to qualitatively explore acceptability and change. Pain-related fear and pain intensity reduced over the course of the programme; successful penetration increased. The programme was experienced as convenient, gradual, and supportive, with moments of frustration at progress. The programme also had a positive influence on self-awareness, confidence, normalisation, and approach behaviours, with exposure attributed as the most difficult but important aspect of change.

- **Part 3: Critical appraisal**. An appraisal of methodology used in the empirical study, discussing the potential biases encountered with research allegiance.

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Part 1: Literature Review

Psychological treatments for vaginal pain: a systematic review and meta-analysis

ABSTRACT

Background: Vaginal pain is experienced in numerous health conditions. Classifying these conditions using aetiological factors is often imprecise, but vaginal pain is typically defined as having a medical or psychiatric cause. The primary distinction between the categories is the presence or absence (respectively) of an assumed physiological cause. Whilst causes of pain vary, commonalities in response to vaginal pain exist across conditions, including sexual behaviour, and emotional and cognitive experiences. Exploring how medically and psychiatrically defined vaginal pain responds to psychological treatment could help to understand further similarities and differences of the conditions. **Aim**: To examine the combined and relative efficacy of psychological treatments for vaginal pain problems defined as medical or psychiatric on outcomes of pain and sexual function. **Method**: A systematic search of EMBASE, Medline, PsycINFO and CINAHL was undertaken. Twelve randomised controlled trials were included of which eleven provided data that were entered into a meta-analysis. Standardised mean differences and odds ratios were used to calculate effects. Effect sizes for individual psychological trial arms were also calculated to compare efficacy between vaginal pain types. **Results**: The meta-analysis revealed no significant differences when comparing psychological treatments to medical and psychological alternatives for all vaginal pain disorders, on outcomes of pain and sexual function. Individual effect sizes for psychological treatment arms were similar for both vaginal pain types. **Conclusions:** Effectiveness of psychological treatment was comparable for vaginal pain conditions regardless of their medical or psychiatric categorisation, indicating that the aetiological distinction may not be helpful. This could have clinical implications for the type of treatments offered for vaginal pain. Further research in this area is needed to support these findings.

INTRODUCTION

There are numerous vaginal pain disorders that interfere with sexual functioning. Some of these are defined as pain disorders with medical aetiology and others as sexual dysfunctions with psychiatric aetiology. The definitions will be explored in an attempt to understand their unique and shared characteristics.

Medically defined vaginal pain

Vulvodynia is a term used to describe chronic pain of the vulvar (external genital region). Vulvodynia can be broken down into subtypes: generalised (pain across entire vulva), localised (pain in specific area of vulva), provoked (pain on contact), and unprovoked (pain without contact). Combinations of the subtypes exist and terminology is variable, e.g. vestibulodynia is a term used to describe pain localised to the vestibule. Vulvodynia is not diagnosed if certain conditions are present that cause pain, such as particular skin disorders or sexually transmitted infections. However, vulvodynia is still medically defined, because it is assumed to have underlying organic causes. Interestingly, these causes are largely unknown and it is widely accepted that the aetiology of vulvodynia remains unidentified (Lonkey, Edwards, Gunter & Haefner, 2011; Lotery, McClure & Galask, 2004). A number of possibilities have been suggested, including genetic vulnerabilities, immune factors, hormonal imbalances, and alternation in the sensitivity of nerves (Haefner et al., 2005); factors that are not readily identifiable. This can also be the case with 'medically unexplained pain' conditions, such as fibromyalgia, which is often diagnosed in the absence of a clear causal explanation. This does not imply that pain is caused by non-medical or psychological factors, but that the clinical tools available are not able to identify a cause. Vulvodynia may follow

infection, such as recurrent thrush, even after it has been successfully been treated (Paavonen, 1995). But often there is no identifiable trigger. A population survey showed that the onset of vulvodynia was attributed to situational factors, such as tampon insertion or intercourse, suggesting that pain pre-existed attempts at penetration. Interestingly higher levels of stress were found in women who developed vulvodynia than those who did not, which could point towards a biopsychological understanding of pain onset (Sutton, Bachmann, Arnold, Rhoads & Rosen, 2008).

Viewed medically, vulvodynia is diagnosed using physical indicators. For example, provoked localised vulvodynia can be assessed using Friedrich's (1987) three criteria: 1) pain on vestibular contact or attempted penetration, 2) tenderness when localised pressure is applied to the vestibule (vaginal opening), and 3) vestibular erythema (redness). Classified as a pain disorder, its impact on functioning is not necessary for a diagnosis; however, the pain can have a considerable impact on aspects of life, including sexual function. Reasons for impaired sexual functioning may appear obvious, particularly in the provoked subtypes. Pain can interfere with intercourse, reduce arousal, diminish desire, and elicit fear and avoidance behaviours (Hallam-Jones, Wylie, Osborne-Cribb, Harrington & Walters, 2001; Masheb, Lozano-Blanco, Kohorn, Minkin & Kerns, 2004). Dyspareunia (pain on intercourse) is a defining characteristic of provoked vestibulodynia (Goldstein, Pukall & Goldstein, 2009). In a medical sense, dyspareunia and vestibulodynia are seen as part of the same condition; however, when a psychiatric classification of dyspareunia is introduced the picture becomes more ambiguous.

Psychiatrically defined vaginal pain

DSM-IV-TR (APA, 2000) criteria are used in this review, as it commenced before the publication of the DSM-5 (APA, 2013). The DSM-5 revised classifications of vaginal pain disorders, implications of which are discussed below.

The DSM-IV-TR defined sexual dysfunction as an interference with sexual responsivity or pleasure, causing marked distress or interpersonal difficulty. The sexual dysfunctions span numerous areas of sexual response (e.g. arousal, desire, orgasm), but this review focuses on the two that are associated with sexual pain: dyspareunia and vaginismus. Dyspareunia is described as 'recurrent or persistent genital pain associated with sexual intercourse'. The criteria specify that dyspareunia is not caused exclusively by effects of substances or a general medical condition. Vaginismus is described as 'recurrent or persistent involuntary spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse'. Similarly, this is not accounted for by medical causes. This definition does not explicitly mention pain; however, it is categorised in the sexual pain section. Therefore, the distinguishing criterion between dyspareunia and vaginismus is a vaginal muscle spasm, the certainty of which has been disputed.

Several reviews have concluded that evidence to support the existence of a vaginal spasm is weak (Binik, 2005, 2010a, 2010b). This was evidenced through experimental research, which found no differences between vaginismus and dyspareunia on a range of measures, including: penetration of a finger, muscle tension or pain during intercourse (De Kruiff, Ter Kuile, Weijenborg & van Lankveld, 2000). Women with vaginismus reported more difficulty with sexual intercourse and women with dyspareunia reported more pain on examination, but these differentiating factors are not reflected in DSM-IV criteria. The sample size in this study was small; therefore, subtle dissimilarities may have been detected with more power. Other empirical

research found that gynaecologists could detect greater muscle tension and more frequent vaginal spasms in vaginismus, compared with dyspareunia (from vestibulodynia) and healthy controls. However, only 28% of women with vaginismus exhibited the spasm on penetration. Interestingly, it was fear and avoidance behaviours that were more frequently reported; 73% in the vaginismus group refused electromyographic sessions (measure of activity in vaginal muscle) compared with 0% in dyspareunia and control group. Women with vaginismus were also rated significantly higher by gynaecologists on defensive behaviours that interfered with the examination (Reissing et al., 2004). This fits with other research that has identified specific cognitive and behavioural (as opposed to physical) elements of vaginismus, such as increased catastrophising of pain, negative self-image, feelings of sexual disgust, fears of intimacy and loss of control (Borg, Peters, Schultz & de Jong, 2012; de Jong, Overveld, Schultz, Peters & Buwalda, 2009). Such cognitions have been attributed to the cause of vaginismus (Reissing, 2012), as they would precede anticipatory anxiety and consequent muscle tension found in vaginismus. But these cognitions are not reflected in the DSM and are more consistent with chronic pain presentations. The inconclusiveness of the literature has led to a change in the diagnostic criteria in the most recent DSM-5 publication (APA, 2013).

Integrating diagnoses

The DSM-5 (APA, 2013) no longer separates vaginismus and dyspareunia; instead it has increased overlap between them by introducing a more inclusive diagnosis. Genito-Pelvic Pain/Penetration Disorder (GPPPD) incorporates both diagnoses into one classification (see Box 1). GPPPD consists of four broad domains: penetration, pain, fear and muscle tension, each of which can be assessed separately. For example, women may experience pain but still

manage penetration, or they could experience minimal pain during penetration, but have a marked fear of penetration. These domains do not rely so heavily on the assessment of aetiological factors (such as a spasm); instead the focus is on symptomatology and impact on functioning.

Box 1

DSM-5 (APA, 2013) criteria for Genito-Pelvic Pain/Penetration Disorder (GPPPD)

- Persistent or recurrent difficulties with one or more of the following:

  1) Vaginal penetration during intercourse

  2) Marked vulvovaginal or pelvic pain during vaginal intercourse or penetration attempts

  3) Marked fear of or anxiety about vulvovaginal or pelvic pain, in anticipation of, during, or as a result of vaginal penetration

  4) Marked tensing or tightening of the pelvic floor muscles during attempted vaginal penetration.

- Symptoms have persisted for a minimum of 6 months.
- Symptoms cause clinically significant distress in the individual.
- The sexual dysfunction is not better explained by a nonsexual mental disorder or as a consequence of severe relationship distress or other significant stressors and is not attributable to the effects of substance/medication or another medical condition.

A minimum of one symptom is needed to be present for a diagnosis, therefore GPPPD should capture a wider range of vaginal pain problems. The previously mentioned vulvodynia subtypes could also overlap with GPPPD. For example, women with provoked vulvodynia experience pain on touch, which is likely to be aggravated during sexual activity. This may interfere with penetration and may lead to a marked fear of sexual activity, all of which is present in the absence of a known medical cause (Paavonen, 1995). It may not have been the intention of the DSM-5 classification to include vulvodynia,

but due to the overlap in symptomatology, GPPPD is a widely inclusive classification that disregards aetiology.

Theory of pain

Pain theory increasingly recognises chronic pain as a disorder with common biological and psychological features regardless of where the pain is felt (Tracey & Bushnell, 2009). Melzack and Wall's (1965) pioneering pain-gate model understands pain to occur through two main processes: 1) a neuronal response, in terms of noxious sensory input received and processed at the dorsal horn, and 2) the top-down influence on pain at the spinal level, through psychological processes, such as attention, mood and memory.

The neuronal response in chronic pain is often explained by the concept of sensitisation. Sensitisation is the amplification of neuronal excitability, which leads to a greater number of signals being processed as pain at the dorsal horn. This process has been identified both centrally (in central nervous system) and peripherally (in the nerve fibres) in vulvodynia (Bohm-Starke, 2010; Tympanidis, Terenghi & Dowd, 2003). Changes in the central nervous system have also been identified in vaginismus (Frasson et al., 2009), but evidence for this is slim compared to vulvodynia.

Psychological components of provoked vestibulodynia have been integrated into a biopsychological model to explain the influence of vulval pain on sexual dysfunction (Basson, 2012). The model suggests that stress in combination with numerous premorbid psychological factors (e.g. anxiety, depression, harm avoidance, hypervigilance and traits of perfectionism) can induce neuronal changes in the form of sensitisation. In turn, the pain is maintained or heightened by acquired risk factors, such as beliefs of sexual inadequacy and diminished sexual motivation. This dynamic reciprocal model highlights how biological and genetic factors interact with behavioural and

cognitive factors. Similar top-down influences have been found in women with vaginismus (Borg et al., 2012). It could be conceived that women, for example, with pre-morbid anxiety or harm avoidance could be more vulnerable to developing vaginismus if they encounter difficult sexual experiences or develop negative self-beliefs.

Anxiety and fear-avoidance models can be applied to pain processing irrespective of pain aetiology. Leeuw, Goossens, Linton, Crombez, Boersma and Vlaeyen (2007) developed a model that described how pain is maintained by avoidance due to a range of unhelpful cognitive strategies. These included: catastrophising about pain (e.g. imagining self as bedbound or paralysed by pain); pain-related anxiety (e.g. hypervigilance to unpleasant sensations); and pain-related fear (threatened by anticipation of pain). Continual perceptions of threat and feelings of anxiety can increase excitability and sensitivity of the pain system (Norton & Asmundson, 2004). Avoidance behaviours diminish opportunities to challenge perceived threat and prevent physical benefits such as muscle strength. In terms of vaginal pain, these concepts also apply (Payne, Binik, Amsel & Khalifé, 2005). Whether diagnosed with vaginismus, dyspareunia or vulvodynia, pain can increase threat perception, anxiety and avoidance, all of which could be modified using psychological techniques.

It is not only anxiety that influences pain; general negative affect can exacerbate pain (Janssen, 2001). Experimental studies have demonstrated that pain is perceived as significantly worse when a sad mood is induced (Boettger, Schwier & Bär, 2011; Tang, Salkovskis, Hodges, Wright, Hanna & Hester, 2008). This is supported by neuroimaging studies that have found increased activation in the anterior cingulate cortex and the amygdala (both involved in the emotional processing of pain), suggesting an exacerbation of pain perception when accompanied by sadness (Berna, Leknes, Holmes,

Edwards, Goodwin & Tracey, 2010; Yoshino et al., 2010). Berna et al., (2010) developed a model that could accurately predict severity of pain experience using pain-based cognitions as an independent variable. This provides neural evidence for pain mechanisms responding to mood and cognition.

So far, the evidence suggests there may be physiological distinctions between medically defined conditions (vulvodynia subtypes) and psychiatrically defined vaginal pain conditions (vaginismus & dyspareunia). However, there is less evidence for psychological distinctions; similar cognitive, behavioural and emotional responses are found across pain conditions.

This raises the clinical question: would medically and psychiatrically defined vaginal pain conditions respond similarly or differently to psychological interventions? For example, if the cause of sexual avoidance is localised pain, it could be hypothesised that psychological intervention may not be effective. However, addressing fear of pain may activate descending inhibitory pathways that in turn act on localised pain. Light can be cast on this question by examining how the varying conditions respond to psychological intervention. While this cannot establish which aspects of the conditions are the same or distinct, it can add to the existing evidence which bears on the classification of the disorders and how they are best treated.

Psychological interventions for vaginal pain

Treatments for vaginal pain disorders have tended to correspond with the medical versus psychiatric distinction. Treatment for vaginismus has primarily been psychological; in a systematic review of vaginismus, the large majority of trials evaluated systematic desensitisation or cognitive-behavioural therapy (Melnik, Hawton & McGuire, 2012). Some medical interventions for

vaginismus have been evaluated, such as botox and bupivacaine injections which aim to minimise pain (Pacik, 2011). Dyspareunia has tended to be treated medically, because it is commonly a secondary diagnosis to vulvodynia (rather than a discrete sexual dysfunction). In turn, treatment typically targets the pain itself rather than the impact the pain causes on penetration (Binik, 2005). In a review of vestibulodynia that examined dyspareunia outcomes, almost all studies evaluated medication, surgery or physiotherapy (Andrews, 2011). The assumption could be that if the pain reduces so too will the dyspareunia. The majority of effectiveness research for vulvodynia has examined medical treatment, but some psychological interventions (exposure, CBT & mindfulness) have been examined (Murina, Berniorio & Palmiotto, 2008; Bergeron et al., 2001; Brotto, Basson, Carlson & Zhu, 2013) and show promising effects.

Aims of review

Vaginal pain conditions generally get treated medically or psychologically in accordance with their definitions, but the evidence had not been evaluated in a systematic way. If psychological treatments are found to be effective for both medically and psychiatrically defined conditions on outcomes of pain and sexual function, this could imply that aetiological distinctions are not so important, and that behavioural, emotional and cognitive response to pain are comparable. This review brings the evidence together by exploring the efficacy of psychological interventions for an inclusively-defined group of vaginal pain problems. Previously, one systematic review has examined the effectiveness of CBT for a more inclusive group of vaginal pain problems, and concluded it was a worthwhile treatment for improving sexual function (LoFrisco, 2011). Taking this further, the current review will combine evidence for all

psychological interventions for medically and psychiatrically defined vaginal pain. This will help to answer the following review questions:

1. How effective are psychological interventions for medically and psychiatrically defined vaginal pain on outcomes of pain and sexual functioning?
2. Is there a difference in effectiveness of psychological treatment between medically and psychiatrically defined vaginal pain on outcomes of pain and sexual functioning?

METHOD

Search strategy

A search of the literature was undertaken in August 2013 using multiple electronic databases (EMBASE 1974- Aug 2013; Medline 1946- Aug 2013; PsycINFO 1967- Aug 2013; CINAHL 1981- Aug 2013). These searches were updated to include studies from 2013-mid-April 2014. References of relevant systematic reviews were also searched. Searches only captured articles published in peer-reviewed journals. Combinations of the following search terms were used in full text searches: *vaginismus, (superficial)* dyspareunia, *sexual dysfunction & pain, (provoked, localised) vulvodynia, vestibulodynia, vestibulitis*, and variations of *randomised controlled trial* (see Appendix 1 for full search strategy). These terms were derived from the inclusion criteria, as well as previous systematic reviews (Andrews, 2012; McGuire & Hawton, 2001; Melnik, Hawton & McGuire, 2012).

Inclusion criteria

Studies were included in the review if the following criteria were met:

- Participants were women over 16 years with vaginismus, dyspareunia or vulvodynia (all subtypes)
- Participants were randomised or quasi-randomised
- Studies had at least one psychological treatment arm compared with a control or other treatment arm (psychological or medical)
- Effectiveness was evaluated using one or more of the following outcomes: pain, sexual functioning (behavioural & cognitive measures) or related psychological distress (e.g. sexual anxiety).

Exclusion criteria

Studies were excluded if the following criteria were met.

- Participants were women with vaginal pain due to known medical conditions (e.g. endometriosis, sexually transmitted infections, cancer, inflammatory problems, dermatoses, menopause) or with deep dyspareunia or chronic pelvic pain (beyond the scope of the current review)
- Studies were published in languages other than English.

Study identification

A total of 1548 studies were retrieved from initial electronic and reference searches after de-duplication (see Figure 1 for study flow). The author and another clinical psychologist independently sifted all studies retrieved from the electronic searches. Any discrepancies in decision about inclusion or exclusion of studies were discussed and agreed.

The 1517 studies that were excluded based on titles and abstracts either failed to meet the population criteria or were non-randomised designs or reviews. Thirty one studies were read in full and 20 were excluded for the

following reasons: 16 had no psychological treatment arm (Bornstein & Abramovici, 1997; Bornstein, Livnat, Stolar & Abramovici, 2000; Bornstein, Tuma, Farajun, Azran & Zarfati, 2010; Bornstein, Zarfati, Goldik & Abramovici, 1995; Donders & Bellen, 2012; Donders, Dreher, Bellen & Fiews, 2013; Farajun, Zarfati, Abramov, Livoff & Bornstein, 2012; Foster, Dworkin & Wood, 2005; Foster et al., 2010; Murina, Bianco, Radici, Felice, Di Martino & Nicolini, 2008; Murina, Graziottin, Felice, Radici & Tognocchi, 2013; Nyirjesy, Sobel, Weitz, Leaman, Small & Gelone 2001; Petersen, Giraldi, Lundvall & Kristensen, 2009) and/or were found to be non-randomised clinical trials (Fowler, 2000, McKay, Kaufman, Doctor, Berkova, Glazer & Redko 2001; Peters, Carrico & Boura, 2011) and two were unavailable in English (Bazin et al., 2011; Zukerman, Roslik & Orvieto, 2005). Two studies included reproduced data from original studies (Bohm-Starke, Brodda-Jansen, Linder & Danielsson, 2007; ter Kuile, van Lankveld, Groot, Melles, Neffs & Zandbergen, 2007) and were used to obtain additional information about the relevant included studies, but data were not double counted. Update searches retrieved 288 studies, of one which met inclusion criteria, giving a total of 12 included studies.

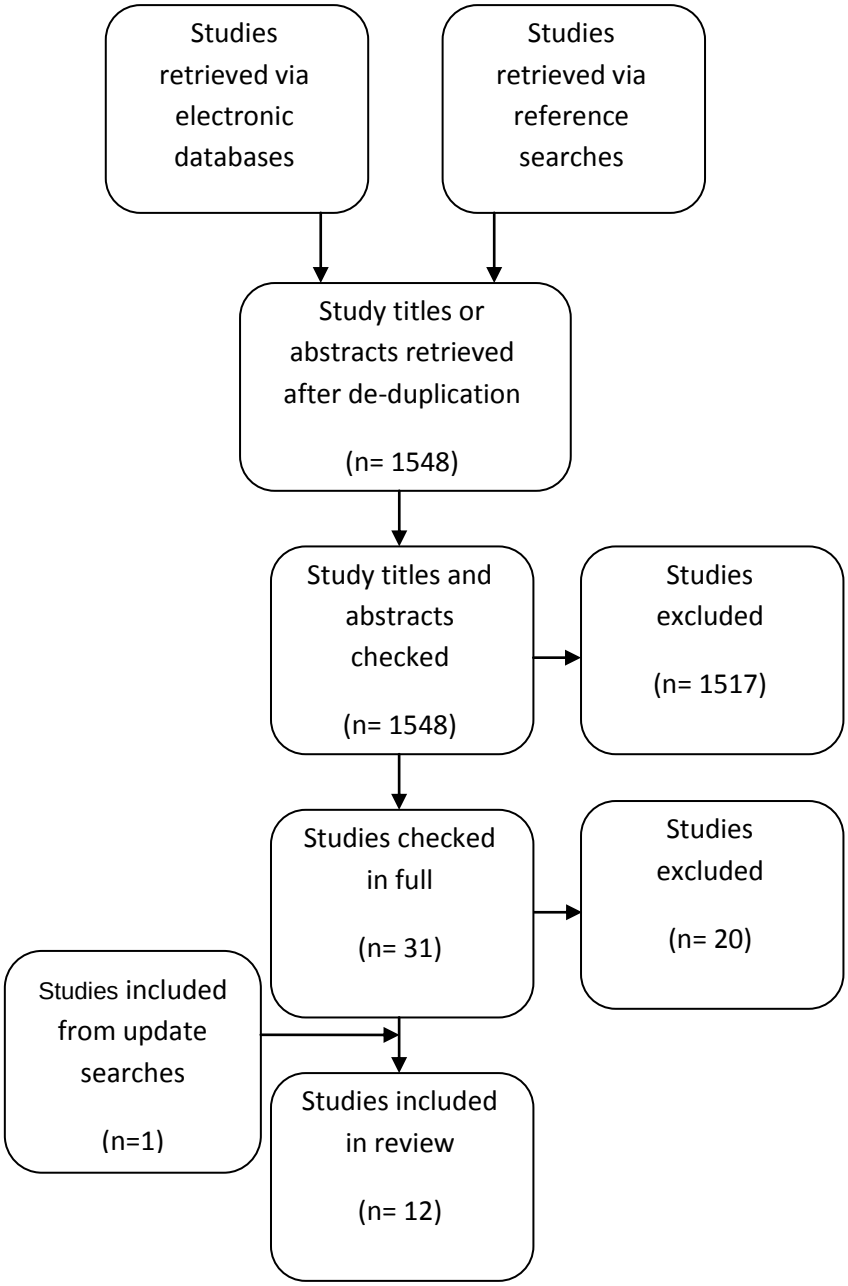

Figure 1: Flow chart of included and excluded studies

Data extraction

The large majority of included studies reported continuous data, from which the means, standard deviations and sample sizes were extracted. If the study used categorical data, events-based outcomes were extracted. If the required data were not included in the published article, authors were contacted. Data from three studies were obtained from the authors. One study provided full

data after partially including RCT data as part of a regression model (Desrochers, Bergeron, Khalifé, Dupuis & Jodoin, 2010). Two studies (van Lankveld, Everaerd & Grotjohann, 2001; van Lankveld, ter Kuile, de Groot, Melles, Nefs & Zandbergen, 2006) provided raw datasets from which the means and standard deviations were calculated. This explains variation in N in the analyses, accounted for by missing data. Calculations from van Lankveld et al., (2006) were based on single questionnaire items rather than composite scores. Data from one study were no longer available (Schnyder, Schnyder-Lüthi, Ballinari & Blaser 1998), although another meta-analysis reported odds ratios for this study (McGuire & Hawton, 2001). Data from Danielsson, Torstensson, Brodda-Jansen & Bohm-Starke (2006) included medians and interquartile ranges; therefore, data could not be converted using standardised methods (Hozo, Djulbegovic & Hozo, 2005; Deeks, Higgins & Altman, 2008). Limited categorical data allowed for the use of odds ratios instead.

Data analysis

Using Review Manager 5 software version 5.2 (The Cochrane Collaboration, 2012), means and standard deviations entered into random effects meta-analyses to calculate mean differences or standardised mean differences. Event-based outcomes were entered into random effects meta-analyses to calculate odds ratios. The analyses examined a range of psychological treatments: CBT (individual, group & self-help), behavioural (biofeedback & exposure), hypnotherapy and supportive therapy. These were compared to medical treatments or control conditions: surgery, medication, topical cream or waiting list. The main outcomes examined were: pain and sexual functioning.

Effect sizes (Cohen, 1992) from baseline to post-treatment were calculated for psychological treatment arms. This enabled comparisons to be made between

trials, in particular, to examine differences between vaginal pain defined as psychiatric (vaginismus) or medical (vulvodynia).

Quality of studies

The NICE (2012) methodology checklist for randomised controlled trials was used to assess the quality of included studies. The checklist assessed four domains: 1) selection bias (randomisation method, allocation concealment & comparability of groups at baseline), 2) performance bias (blinding & equivalent care), 3) attrition bias (drop-out) and 4) detection bias (reliability of outcomes). As the studies examined psychological interventions, blinding was less applicable. However, studies that attempted to address this by using independent assessors were viewed as superior on that domain to studies that did not. The level of risk was rated as 'low, 'high' or 'unclear' on each domain, based on the accumulated risk of the items. Ratings were undertaken by the researcher and an independent assessor. Disagreements were resolved through discussion, ensuring that criteria were applied consistently across studies. Quality of the studies was used to inform the integrity of treatment effects.

Researcher allegiance can impact considerably on effects found in controlled trails (Leykin & DeRubeis, 2009); therefore, this was assessed using a process developed by Gaffan, Tsaousis and Kemp-Wheeler (1995). Studies were assessed on several domains: citing previous research supporting a particular treatment; discussing the superiority of a particular treatment; including a description of treatment that exceeds 10 sentences; authorship of treatment; and having a sole active treatment condition. Research was rated as having 'strong', 'moderate', 'weak' or 'no' allegiance.

All quality criteria can be found in Table 1.

Table 1

Study Characteristics for included studies

Study ID	Population	Intervention/ Comparator	Outcomes	Selection bias	Performance bias	Attrition bias	Detection bias	Research allegiance
Al-sughayir 2005	36 women with vaginismus (DSM-IV) Aged 17-40 (mean age 23) Outpatient psychiatric clinic, Saudi Arabia Mean duration of problem 9.52 months	1. Hypnotherapy (n=18) Once weekly 45-60 minutes, wife only (mean sessions 4.7) 2. Behaviour therapy (n=18) Once weekly 45-60 minutes, both wife and husband (mean sessions=10)	Sex related anxiety (5 point scale) Wife's sexual satisfaction (5 point scale) Husband's sexual satisfaction (5 point scale)	Design: quasi-randomised Randomisation method: alternate allocation Allocation concealment: not reported No significant differences between demographic and outcome measures at baseline Risk of selection bias: high	Blinding: Independent psychology assessment pre and post treatment. No additional treatments during study Treatment administered until symptoms reduced (considerable variation in number of sessions offered) Risk of performance bias: high	Follow-up: none Dropout: Group 1: post-treatment 2/18 Group 2: post-treatment 3/18 Data for 5 dropouts not included No ITT analysis reported Risk of attrition bias: unclear	Follow-up duration inadequate Basic outcome measures not well defined; not validated No measure of sexual functioning or pain Attempt to blind investigators to treatment exposure Risk of detection bias: high	Strong allegiance to hypnotherapy
Bergeron et al. (2001) [Bergeron Khalifé, Glazer & Binik (2008) [follow-up]	78 women with vestibulodynia Mean age 26.8 Canada	1. Vestibulectomy (n=22) 30 minute operation; information given before and after surgery by gynaecologist 2. Biofeedback (n=28) Self-insertion of EMG sensor into vagina; twelve 45-minutes sessions over 8 weeks. 3. Group CBT (n=28)	Vestibular pain index (11-point scale) Pain intensity of vaginal intercourse (11-point scale) McGill Pain Questionnaire (MPQ): Pain Rating Index & Sensory scale	Design: randomised Randomisation method: blocked Allocation concealment: not reported No significant differences between groups on demographic or pre-treatment outcomes.	Blinding: outcomes administered by independent clinical associate No additional treatments requested during study. 5/76 at post-treatment and 17/51 at 2.5 year follow-up reported using other means of pain relief, This did not differ as function of treatment.	Follow-up: 6 months Dropout: Group 1: 7/22 pre-treatment; 3/13 6-month follow-up. Significantly higher dropout pre-treatment than in groups 2 & 3. Group 2:1/28 pre-treatment; 2/27 post-treatment; 8/25 6-month follow-up. Significantly more dropouts at follow-	Follow-up duration appropriate Established and reliable outcome measures used Attempt to blind investigators to treatment exposure Risk of detection bias: low	Moderate allegiance to CBT

		Eight 2-hour sessions over 12 weeks; led by psychologists.	Sexual Information Scale Frequency of sexual intercourse Global Severity Index of Brief Symptom Inventory (BSI-GSI) Credibility	Participants randomly assigned to 1 of 2 gynaecologists (group 1) or 1 of 2 psychologists (groups 2 & 3). Adherence to manual: Group 2: 57% compliance with homework Group 3: 0.87 inter-rater reliability and 65% compliance with homework Risk of selection bias: low	Risk of performance bias: unclear	up. Group 3: 1/28 pre-treatment dropout. No significant differences in demographic or pre-treatment outcomes in completers or dropouts at 6-month and 2.5 year follow-up 2.5 year follow-up Group 1: n=15 Group 2:n=17 Group 3: n=19 ITT analysis undertaken. Risk of attrition bias: low		
Desrochers et al., 2010	97 women with vestibulodynia Mean age: Group 1: 26 Group 2: 27 Mean onset of problem 5.5 years	1. Group CBT(n=52) Ten 90-minute sessions, run by trained and supervised psychotherapists 2. Topical Treatment (n=45) 8 weeks corticosteroid cream (1%) applied to vestibule twice a day for 13 weeks, plus lubricant during penetration and education. Prescribed by 2 gynaecologists. Discontinue after 8 weeks if no improvement.	Gynaecological examination Pain during intercourse (0-10 visual analogue scale) MPQ-PPI Frequency of intercourse Female Sexual Functioning Index (FSFI) Sexual satisfaction	Design: randomised Randomisation method: blocked Allocation concealment: not reported No significant differences between groups on demographic or clinical outcomes, apart from pain duration (longer in group 2). Adherence to manual checked	Blinding: unknown Concurrent treatments: unknown Risk of performance bias: unclear	Follow-up: 6 months Dropout (based on unpublished data) Group 1: 13/52 post-treatment; 4/39 follow-up Group 2: 15/45 post-treatment; 1/30 follow-up No significant differences found between dropouts and completers on demographic or clinical variables apart from fear of pain (higher in dropouts)	Follow-up duration appropriate Established and reliable outcome measures used Investigators not blind to treatment exposure Risk of detection bias: unclear	Moderate allegiance to CBT

			Brief Symptom Inventory/Global Severity Index (BSI-GSI) Pain catastrophising Scale (PCS) Pain Intercourse Self-Efficacy Scale	Risk of selection bias: low		ITT analysis undertaken Risk of attrition bias: low		
Brown Wan, Bachmann & Rosen (2009)	53 women with vulvodynia (generalised and provoked) Non-responders from previous trial of dietary therapy Mean age 47	1. CBT based self-management (n=26) Twelve 2-hour weekly group sessions; delivered by nurse practitioner, psychologist and physiotherapist 2. Amytriptyline (tricyclic antidepressant) (n=13) 10mg a day for 6 weeks, increased to 20mg if well tolerated, for remainder of study 3. Amytriptyline + Triamcinolone cream (corticosteroid) (n=14) 10mg a day for 6 weeks, increased to 20mg if well tolerated for remainder of study. Plus once daily application of triamcinolone 5mg cream on affected area Cream discontinued at 6	McGill Pain Questionnaire (MPQ): Pain Rating Index	Design: randomised prospective Randomisation method: computer generated Allocation concealment: envelopes. Twice as many participants randomised to group 1, than groups 2 & 3 No significant differences between groups on demographic or pre-treatment outcomes for completers. ITT revealed significantly more pain in group 3 than group 1 at baseline Adherence: Group 1 evaluated by attendance (81% attended all) and self-reported	Blinding: neither participants nor staff blinded to treatment Discontinued any other treatments for pain during study Risk of performance bias: unclear	Follow-up: none Dropout: Group 1: 5/26 post-treatment Group 2: 2/13 post-treatment Group 3: 3/14 post-treatment No significant differences in refusal or dropout rates between groups. ITT analysis undertaken for baseline scores only Risk of attrition bias: low	Follow-up duration inadequate Established and reliable outcome measures used Investigators not blind to treatment exposure Risk of detection bias: high	Weak allegiance, no treatment notably favoured

		weeks		compliance. Groups 2 & 3 evaluated by tablet count and cream measurement (100% compliance). No significant difference in adherence between groups. Risk of selection bias: low				
Danielsson et al., (2006)	46 women with vestibulodynia Mean age: Group 1: 25.8 Group 2: 23.3 Outpatient vuvlar clinic, Sweden	1. Lidocaine (local anesthetic) (n=23) 2% gel, 5% ointment; applied 5-6 times a day for 2-4 months 2. EMG biofeedback (n= 23) Vaginal sensor applied 3 times a day for 10 minutes per session at home	Pain pressure thresholds Short form 36 (SF-36) Prime Care Evaluation of Mental Disorders (PRIME MD) Quality of Life (QOL) 0-100 visual analogue scale Sexual functioning 0-100 visual analogue scale Coital pain 0-100 visual analogue scale	Design: randomised Randomisation method: computer generated Allocation concealment: not reported Group differences at baseline not adequately reported Adherence: Group 1: 95% 5 times per day; 50% used ointment only after 2 months Group 2: 0% 3 times per day; 56% 2 times per day. Risk of selection bias: unclear	Blinding: not reported Concurrent treatments: unknown Risk of performance bias: unclear	Follow-up: 6 and 12 months Dropout: Group 1: 4/23 post-treatment; 4/19 12 month follow-up Group 2: 5/23 post-treatment; 1/18 12 month follow-up No ITT analysis reported Risk of attrition bias: unclear	Follow-up duration adequate Established and reliable outcome measures Investigators not blind to treatment exposure Risk of detection bias: unclear	Weak allegiance, neither treatment notably favoured
Masheb, Kerns, Lozano, Minkin &	50 women with vulvodynia (generalised & provoked)	1. CBT (n=25) Ten weekly 60 minute sessions	Physician assessment: speculum, cotton swab and erythema.	Design: randomised Randomisation method: computer generated	Blinding: treatment blinded to gynaecologists undertaking examinations. Participants and	Follow-up: 6 and 12 months Dropout:	Follow-up duration adequate Established and	Strong allegiance to CBT

Richman (2009)	Mean age 43 University students	2. Supportive Psychotherapy (n=25) Ten weekly 60 minute sessions Both treatments delivered by doctoral level research therapists	Multidimensional Pain Inventory (MPI) McGill Pain Questionnaire (MPQ) Female Sexual Functioning Index (FSFI) Beck Depression Inventory (BDI) Pain Anxiety Symptom Scale (PASS) Global improvement rating (scale 0-5) Satisfaction and credibility rating (scale 0-10)	Allocation concealment: not reported. Group assignment revealed post-assessment. Adherence to manuals checked by supervisors. No significant differences between groups on demographic, psychiatric or pain history outcomes. Risk of selection bias: low	researchers not blind to treatment. Concurrent treatments: participants excluded if started psychotherapy, psychopharmalogical or pain treatment in past month Risk of performance bias: unclear	Group1: 2/25 post-treatment; 1/23 at 6 and 12 month follow-up Group 2: no drop-out No ITT analysis reported Risk of attrition bias: unclear	reliable outcome measures Attempt to blind investigators to treatment exposure Risk of detection bias: low	
Schnyder et al., (1998)	44 women with vaginismus (DSM-III; acquired & lifelong) Mean age 28 Mean duration of problem 4.02 years Outpatient	1. In vivo desensitisation (n=21) Dilators introduced manually by physician 2. In vitro desensitisation (n=23) Dilators introduced verbally by physician Both groups 10-15 minutes of desensitisation 5 times a	Successful intercourse (outcome measure not described in detail)	Design: quasi-randomised Randomisation method: alternate allocation. Two participants requested not to be in group1 and were put in group 2 Allocation concealment: not reported No significant	Blinding: not reported Concurrent treatments: unknown Risk of performance bias: unclear	Follow-up: 10 months Dropout: 8/44 at follow-up Dropout differences by group not reported No ITT analysis reported Risk of attrition bias: high	Follow-up duration adequate Unknown whether outcome measure is reliable Investigators not blind to treatment exposure Risk of detection bias: high	Moderate allegiance to behavioural treatment

	obstetrics and gynaecology department, Switzerland	week. Therapy sessions every 2 weeks Number of sessions not reported Therapy sessions Treatment delivered by two experienced sex therapists		differences between groups on demographic variables or problem severity. Significantly more patients in group 2 found it difficult to talk about sex and held negative attitudes about their bodies. Risk of selection bias: high				
Ter Kuile, Melles, de Groot, Tuijnma-Raasvel & van Lankveld (2013)	70 women with vaginismus (lifelong) and their partners Mean age 28.9 Mean duration of problem 10.41 years Outpatient Clinic Psychosomatic Gynecology and Sexology, Netherlands	1. Exposure Therapy (n=35) Maximum of three 2-hr sessions within 1 week, plus two follow-up sessions over 5 weeks. In vivo desensitation, self-controlled; verbally directed by therapist. Treatment delivered by 4 female psychologists and one experienced social worker 2. Waiting list (n=35)	Successful intercourse (as recorded in a diary) Golombok Rust Inventory of Sexual Satisfaction (GRISS) Fear of Sexuality Questionnaire (FSQ) Female Sexual Distress Scale (FSDS)	Design: Randomised Randomisation method: blocked and stratified Allocation concealment: Yes No significant differences between groups on demographic variables or sexual dysfunctions. However control group significantly higher baseline sexual distress scores (FSDS) Risk of selection bias: unclear	Blinding: not reported Concurrent treatments: unknown Risk of performance bias: high	Follow-up: 6 and 12 weeks Dropout: Group 1: 2/35 post-treatment, 2/33 follow up Group 2: 3/35 post-treatment ITT analysis undertaken Risk of attrition bias: low	Follow-up duration brief Established and reliable outcome measures Investigators not blind to treatment exposure Risk of detection bias: unclear	Strong allegiance to CBT
van Lankveld et al., (2001)	55 women (vaginismus n=28; dyspareunia n=25)	1. CBT bibliotherapy (n=125) Given manual to read; 10 weeks duration with telephone support	Golombok Rust Inventory of Sexual Satisfaction (GRISS) Maudsley Marital Questionnaire	Design: randomised (partially) Randomisation method: blocked card draw. 21/125 couples in	Blinding: assessed by psychologist not involved in treatment. Concurrent treatments: unknown	Follow-up: 10 weeks Overall dropout: Group 1: 14/125 post-treatment; 11/111 follow-up	Follow-up duration brief Established and reliable outcome measures	Strong allegiance to CBT

	Subset of couples with numerous sexual dysfunctions Mean age of subsets unknown, overall mean age of females 35 Outpatient gynaecology and sex clinic, Netherlands	Group 1 broken down further into: randomised participants (n=104) (self-initiated contacts if having difficulties); and non-randomised participants (n=21) (scheduled telephone contacts) 2. Waiting list (n=98)	(MMQ) Intimate Contact Body Scales Self-rated evaluation of treatment (4 item scale) Compliance	treatment condition were not randomised Allocation concealment: unclear No significant differences between groups on demographic variables or sexual dysfunctions. Risk of selection bias: high	Risk of performance bias: unclear	Group 2: 10/98 post-treatment-up; 3/88 follow-up Subgroup dropout: None Attrition rates not compared ITT analysis undertaken Risk of attrition bias: low	Attempt to blind investigators to treatment exposure Risk of detection bias: unclear	
van Lankveld et al., (2006)	117 couples (women with vaginismus, DSM-IV-TR, lifelong only) Mean age females 28.6 Mean age partners 31 Mean duration of problem 11 years Outpatient sexology clinic, Netherlands	1. Group CBT (n=43) CBT manual and CD-ROM, plus ten 2-hour group sessions (female partner only) 2. Bibliotherapy CBT (n=38) CBT manual and CD-ROM, plus six biweekly 15-minute telephone calls 3. Waitlist control (n=36) 12 weeks on waiting list 3 months treatment duration Treatment delivered by 10 therapists (7 senior, 3 junior)	Primary Endpoint Questionnaire (levels of penetration achieved) Mini International Neuropsychiatric Interview (MINI) Female Sexual Function Index (FSFI) Maudsley Marital Questionnaire (MMQ) Golombok Rust Inventory of Sexual Satisfaction (GRISS)	Design: randomised Randomisation method: urn allocation, by someone not involved in assessment or treatment. Allocation concealment: not reported No significant differences between groups on demographics, treatment history or sexual functioning Risk of selection bias: low	Blinding: 3/4 assessors blinded to treatment;1/4 assessor involved in treatment delivery and data collection Physical examiners blinded to treatment Concurrent treatments: unknown Risk of performance bias: unclear	Follow-up: 3 and 12 months Dropout: 24/117 couples post-treatment. Significantly fewer dropouts in group 3 (3/36) than group 1 (11/43) & 2 (10/38). At 3 months follow-up, 1/33 couples in group 1 and 3/27 in group 2 dropped out. No significant differences found on demographic or sexual functioning between dropouts and completers. ITT analysis undertaken	Follow-up duration adequate Established and reliable outcome measures Attempt to partially blind investigators to treatment exposure Risk of detection bias: unclear	Strong allegiance to CBT

			Sexual and Physical Abuse Questionnaire (SPAQ)			(last score carried forward) Risk of attrition bias: low		
Weijmar-Schultz et al., (1996)	14 women with vestibulodynia and unable to have intercourse Mean age 24 Dutch gynaecology department	1. Behavioural therapy + placebo surgery (n=7) Hospitalised for one night and given local anaesthetic 2. Surgery + behavioural therapy (n=7) Hospitalised for one night and surgical excision under local anaesthetic performed Participants excluded if initiated other pain treatment in the past 2 months	Problem severity (5-point scale)	Design: randomised Randomisation method: not reported Allocation concealment: not reported Comparability of groups at baseline not reported Risk of selection bias: high	Blinding: participants were blinded Comparability of care during study duration unclear. All participants hospitalised to make groups as equal as possible. Risk of performance bias: low	Follow-up: none, but post-treatment scores collected on average 3 years after treatment Wide-ranging times for post-treatment outcome (8-56 months) Substantial differences in treatment duration Group 1: 17 months Group 2: 11 months No ITT analysis No evaluation of sexual functioning Risk of attrition bias: high	Follow-up duration unclear Basic outcome measure; unknown whether outcome is reliable Risk of detection bias: high	Weak allegiance, neither treatment notably favoured

RESULTS

Included studies

Of the 12 studies included in the review, ten provided post-treatment data for a total number of 417 participants. Four studies provided follow-up data with a maximum number of 180 participants. One was a follow-up study for which data are presented together with the original study (Bergeron et al., 2001). Five studies examined vaginismus; five examined vestibulodynia; and two examined a mix of provoked and generalised vulvodynia. One study had three active treatment arms, one study had two active treatment arms and one control arm, seven studies had two active treatment arms, and two studies had one active treatment arm and one control arm. In terms of psychological treatment, six studies evaluated CBT, six evaluated behavioural therapy, one evaluated hypnotherapy and one supportive therapy. When entering these into the meta-analysis, CBT and behavioural treatments were most frequently selected as the active treatment when compared with other psychological treatments. Of the active comparisons, five studies compared psychological treatment with another psychological treatment, two with surgery, and three with medication. Three compared psychological treatments with a waiting list control. The mean age across studies was 30 years.

Risk of bias

All studies included in the review were evaluated for risk in terms of selection bias, performance bias, attrition and detection bias (see Table 1). Selection bias was rated as high in four studies, unclear in two studies and low in five studies. Performance bias was rated as high in two studies, unclear in eight studies and low in one study. Attrition bias was rated as high in two studies, unclear in three studies and low in six studies. Detection bias was rated as high in four studies and unclear in five studies and low in two studies. Overall, most studies were rated as unclear due to a lack of reported information or an inability to adequately blind psychological

treatment. Research allegiance was also evaluated for all studies. This was rated as weak in three studies, moderate in three studies and strong in five studies.

Treatment effects from the meta-analysis

Combining multiple studies into one analysis will inevitably result in a degree of heterogeneity. Heterogeneity is the level of variation among studies and represents both clinical variation, such as the population or intervention, and methodological variation, such as the study design. Heterogeneity was calculated using the I^2 statistic. In line with similar reviews, thresholds used for heterogeneity levels were: <25% low, 25-50% moderate, and >50% high. All results can be found in [Table 2].

Forest plots (Appendix 2) compare two treatment groups on one outcome at a time. The means, standard deviations and number of participants from each study arm are used to calculate a mean effect. The mean effect is represented by a diamond, which can be seen to favour one treatment over another depending on where it falls along the y-axis. If the diamond touches the x-axis, the difference between groups is not significant (indicated by the p value).

Table 2

Comparisons of psychological treatment with other treatments or controls

Study ID	N	Populations	Treatment	Comparator	Outcome	Z	P	Heterogeneity
Bergeron et al., (2001) Desrochers et al., (2010) Brown et al., (2009)	143	Vestibulodynia Provoked/generalised vulvodynia	CBT	Medical (surgery, topical cream & medication)	General pain (not limited to intercourse) post-treatment	1.40	0.16	Moderate (25%)
Bergeron et al., (2001) Desrochers et al., 2010	111	Vestibulodynia	CBT	Medical (surgery & topical cream)	Pain on intercourse post-treatment	0.77	0.44	High (77%)
					Pain on intercourse follow-up	0.07	0.10	Moderate (45%)
Bergeron et al., (2001) Desrochers et al., (2010)	111	Vestibulodynia	CBT	Medical (surgery & topical cream)	Sexual functioning post-treatment	0.21	0.83	None
					Sexual functioning follow-up	0.79	0.43	None
Danielsson et al., (2006) Weijmar-Schultz et al., (1996)	46	Vestibulodynia	Behaviour Therapy	Medical (medication & surgery)	Symptom elimination (follow-up)	0.41	0.68	None
Bergeron et al., (2001) Masheb et al., (2009) van Lankveld et al., (2006)	148	Vestibulodynia Provoked/generalised vulvodynia Vagnismus	CBT	Other psychological (biofeedback, supportive & bibliotherapy)	Pain on intercourse post-treatment	0.32	0.75	Low (15%)
Bergeron et al., (2001) Masheb et al., (2009)	83	Vestibulodynia Provoked/generalised vulvodynia	CBT	Other psychological (biofeedback & supportive)	Pain on intercourse follow-up	1.08	0.28	High (69%)
Bergeron et al., (2001) Masheb et al., (2009)	83	Vestibulodynia Provoked/generalised vulvodynia	CBT	Other psychological (biofeedback & supportive)	Sexual functioning post-treatment	1.03	0.30	None
Al-sughayir (2005) Masheb et al., (2009) van Lankveld et al., (2006)	134	Vaginismus Provoked/generalised vulvodynia	Cognitive & behaviour therapies	Other psychological (hypnotherapy, supportive & bibliotherapy)	Sexual anxiety post-treatment	1.60	0.11	Low (23%)
van Lankveld et al., (2001) van Lankveld et al., (2006)	88	Vaginismus Dyspareunia	Bibliotherapy	Waitlist control	Frequency of sex	2.00	0.05*	None
van Lankveld et al., (2006) ter Kuile et al., (2013)	66	Vaginismus	Cognitive & behaviour therapies	Waitlist control	Pain on intercourse post-treatment	0.89	0.37	High 91%
van Lankveld et al., (2006) ter Kuile et al., (2013)	66	Vaginismus	Cognitive & behaviour therapies	Waitlist control	Fear of intercourse post-treatment	0.28	0.78	High 88%

Psychological versus medical treatment

Three studies (n=143) of good quality compared post-treatment effects of CBT with medical treatment on outcomes of general pain (not limited to intercourse). No significant difference between groups was found. Two studies compared post-treatment effects of CBT with medical treatment on outcomes of pain on intercourse. No significant difference was found. At 6 months follow-up, no significant effect was detected (Appendix 2, Analyses 1.1-1.3).

Two studies compared post-treatment effects of CBT with medical treatment on outcomes of sexual functioning. No significant difference between groups was found. At 6 months follow-up, no significant effect was detected (Appendix 2, analysis 1.4 & 1.5).

Two studies (n=46) of unclear and low quality compared post-treatment odds ratios of behaviour therapy versus medical treatment on outcomes of symptom elimination (absence of related symptoms). No significant difference was found. See Appendix 2, analysis 2.1 for forest plot.

Overall, when comparing psychological treatment to active medical treatments on a range of outcomes, no significant effects were found. This suggests that psychological and medical treatments were equal in their effectiveness; pre-post change (discussed later) suggested that overall, psychological interventions led to improvements, with effect sizes ranging from small to large on pain and sexual functioning.

CBT versus other psychological interventions

Three studies (n=148) of good quality compared post-treatment effects of CBT with other psychological interventions (biofeedback, supportive & bibliotherapy) on

outcomes of pain on intercourse. No significant difference between groups was found. Two of these studies compared effects at follow-up (1 & 2.5 years) and no significant effect was found (See Appendix 2, analysis 3.1 & 3.2). Observationally, vestibulodynia seemed to benefit more from other psychological treatments than from CBT, whereas for vaginismus, no difference was apparent. However, a non-significant difference remained even if the vaginismus study was removed (p=0.20).

Two studies (n=83) compared post-treatment effects of CBT with other psychological interventions (biofeedback & supportive) on outcomes of sexual functioning. No significant difference between groups was found (Appendix 2, analysis 3.3). Data from Schnyder et al., (1998) could not be obtained from the original publication; however, a previous meta-analysis (McGuire & Hawton, 2001) calculated odds ratios for this study that compared two forms of behavioural treatment, and found no significant difference on outcomes of successful penetration (Z=0.52, p=0.60).

Three studies (n=142) of mixed quality compared post-treatment effects of cognitive or behavioural therapy with other psychological therapies (hypnotherapy, supportive & bibliotherapy) on outcomes of sexual anxiety. No significant difference between groups was found. The study on vestibulodynia (Masheb et al., 2009) did not seem to favour either treatment, whereas, the two studies on vaginismus appeared to favour CBT. When the vestibulodynia study was removed, the effect became significant (Z=1.94, P=0.05, ES=0.47, CI=-0.00-0.95), although one of the vaginismus studies was rated as high risk on three domains of bias (Appendix 2, analysis 3.4).

On the whole, when comparing psychological treatment to other active psychological treatments on a range of outcomes, no significant differences were

found, suggesting that psychological treatments were equally in their effectiveness; pre-post change (discussed later) suggested that most psychological interventions improved pain on intercourse and sexual functioning, with effect sizes ranging from small to large. Biofeedback however had no effect on pre-post change on outcomes of sexual function or activity.

Psychological treatment versus waitlist control

Two vaginismus studies (n=88) of adequate quality compared effects of bibliotherapy with waitlist control on outcomes of frequency of sex. A significant effect was found (Z=2.00, P=0.05, ES=0.43, CI=0.86-0.01), suggesting that psychological treatment is better than no treatment (Appendix 2, analysis 4.1 for forest plot).

Two vaginismus studies (n=66) compared effects of CBT and exposure treatment with waitlist control on outcomes of pain and fear of intercourse. No significant difference was found on either outcome (see analyses Appendix 2, analyses 4.2 & 4.3).

Effect sizes

Baseline to post-treatment effects were calculated for psychological treatment arms. Thresholds for size of effect were: >0.1 (small); >0.3 (moderate) and >0.5 (large) (Cohen, 1992). Effects were considered in terms of aetiology: medically defined versus psychiatrically defined conditions. Effects could not be calculated for three studies where the required data were not included in the studies (Al-sughayir, 2005; Weijmar-Schultz et al., 1998; Danielsson et al., 2006) or where pre-treatment data were not included for all outcomes. Effect sizes can be found in Table 3.

Effect sizes for medically defined pain

Four studies examined the impact of psychological treatment on general pain. Effects of CBT ranged from small to large. The effect of biofeedback was small ES=0.16 (Bergeron et al., 2001) and supportive therapy large ES=0.64 (Masheb et al., 2009).

Three studies examined the impact of psychological treatment on pain on intercourse. Effects of CBT were large, ES=0.61 (Bergeron et al., 2001), ES=0.69 (Desrochers et al., 2010), ES=0.53 (Masheb et al., 2009). The effect of biofeedback was also large ES=0.71 (Bergeron et al., 2001). Supportive therapy produced a moderate effect ES=0.48 (Masheb et al., 2009). The same studies also examined the impact of psychological treatment on sexual functioning. Effects of CBT ranged from small ES=0.16 (Bergeron et al., 2001) to large ES=0.69 (Masheb et al., 2009). A small effect was found for supportive therapy ES=0.15 (Masheb et al., 2009). No effect was found for biofeedback.

Two studies examined the impact of psychological treatment on the frequency of sexual activity. A very small effect was found in one study ES=0.12 (Bergeron et al., 2001). No effect of CBT or biofeedback was found.

Effect sizes for psychiatrically defined pain

Two studies examined the impact of psychological treatment on pain on intercourse found a large effects for CBT ES=0.69, exposure ES=2.29 and bibliotherapy ES=0.64. Van Lankveld et al., (2006) also evaluated the ability to undertake penetrative behaviours (excluding sex) and found a large effect size for CBT ES=1.19 and bibliotherapy, ES=0.93 (van Lankveld et al., 2006).

One study that examined the impact of bibliotherapy on sexual functioning found a moderate effect for a dyspareunia subset ES=0.31, and large effect for a vaginismus subset ES=1.46 (van Lankveld et al., 2001). This study also examined the impact of bibliotherapy on the frequency of sexual activity and found no effect for the dyspareunia subset, but in the vaginismus subset, a large effect size was found ES=0.49 (van Lankveld et al., 2001).

Table 3

Effect sizes for psychological treatment arms

Study ID	Populations	Outcome	Treatment	Effect size*
Bergeron et al., (2001)	Vestibulodynia	General pain	CBT	0.09
	Provoked/		Biofeedback	0.16
Brown et al., (2009)	generalised		CBT	0.31
Desrochers et al. (2010)	vulvodynia		CBT	0.44
Masheb et al., (2009)			CBT	**0.78**
			Supportive	**0.64**
Bergeron et al., (2001)	Vestibulodynia	Pain on intercourse	CBT	**0.61**
	Provoked/		Biofeedback	**0.71**
Desrochers et al., (2010)	generalised		CBT	**0.69**
Masheb et al., (2009)	vulvodynia		CBT	**0.53**
			Supportive	0.48
Bergeron et al., (2001)	Vestibulodynia	Sexual functioning	CBT	0.16
	Provoked/		Biofeedback	No effect
Desrochers et al., 2010	generalised		CBT	0.44
Masheb et al., (2009)	vulvodynia		CBT	**0.69**
			Supportive	0.15
Bergeron et al., (2001)	Vestibulodynia	Sexual activity	CBT	0.12
		frequency	Biofeedback	<0.1
Desrochers et al., 2010			CBT	<0.1
van Lankveld et al., (2006)	Vagnismus	Pain on intercourse	Bibliotherapy	**0.64**
			CBT	**0.69**
ter Kuile et al., (2013)			Exposure	**2.29**
ter Kuile et al., (2013)	Vagnismus	Fear of intercourse	Exposure	**0.89**
van Lankveld et al. (2006)	Vagnismus	Other penetration	Bibliotherapy	**0.93**
		behaviours	CBT	**1.19**
van Lankveld et al. (2001)	Dyspareunia	Sexual functioning	Bibliotherapy	0.31
	Vaginismus		Bibliotherapy	**1.46**
ter Kuile et al., (2013)			Exposure	<0.1
van Lankveld et al. (2001)	Dyspareunia	Sexual activity	Bibliotherapy	<0.1
	Vaginismus	frequency	Bibliotherapy	0.49

*Bold indicates a large effect size

Summary of effect sizes

Psychological treatments for vulvodynia produced wide-ranging effect sizes for general pain; but, when focusing on sexual pain specifically, effect sizes were larger. Two vaginismus studies that examined pain found large treatment effects. This

could indicate that psychological interventions have an impact on sexual pain regardless of assumed aetiology.

In terms of general sexual function, CBT for vulvodynia produced a range of effect sizes, from small to large; however, this was not true for the frequency of sexual activity. For vaginismus, large effects were found for sexual function and frequency. Interestingly, within one study, a large effect was found for sexual frequency in a vaginismus subset, but no effect was found in the dyspareunia subset (van Lankveld et al., 2001). This could indicate a differential effect of treatment on the two sexual dysfunctions. This study was rated as having a high risk of selection bias and analyses were undertaken on a small subset of a wider population, which could impact on the reliability of findings.

DISCUSSION

A systematic review and meta-analysis on the effectiveness of psychological interventions for medically and psychiatrically defined vaginal pain was undertaken. Twelve studies were included that compared psychological treatments with medical treatments, other psychological treatments and control groups. Outcomes of pain and sexual function were examined.

Summary of primary findings

The primary aim of the review was to examine the effectiveness of psychological treatment for an inclusive group of vaginal pain conditions. Overall, no significant differences were found between psychological and medical treatments on all outcomes, nor were significant differences found between two active psychological treatments. This suggests equality in treatment effectiveness. A significant effect of psychological treatment (bibliotherapy) for vaginismus was found only when

compared with a waitlist control group. The included studies varied in terms on their quality. Many studies were rated as having an unclear risk of bias due to information not being reported. Two studies included in the meta-analysis stood out as being particularly at risk of bias (Alsughayir, 2005 & Weijmar-schultz et al., 1996). These studies were included in only one analysis each.

No studies on psychiatrically defined vaginal pain were found to compare psychological with medical treatment. This could be due to the psychiatric definitions which might have steered research away from medical comparators. No trials on medically defined vaginal pain were found to compare psychological treatment with a control group. This could be due to ethical concerns about withholding treatment for pain. These gaps in the research literature limited evaluation of combined effectiveness of treatment of vaginal pain types.

Explanations of primary findings

The finding that psychological and medical treatment for vulvodynia pain may be of equal effectiveness is interesting, because medical treatment is often a first-line option (Updike & Wiesenfeld, 2005; Mandal et al., 2010). This raises a question; what elements of pain are such disparate treatments acting on? Medical interventions (including medication & surgery) could be influencing physiological, bottom-up aspects of pain by acting on the peripheral pain system, reducing sensitivity or removing nerve endings; whereas psychological treatments could be mediating pain and sensitivity via top-down processes, such as emotional, cognitive or behavioural aspects of pain. It is unlikely that such a polarised explanation is valid. A biopsychological model would propose that both psychological and biological processes are influencing pain in a reciprocal way (Melzack & Wall, 1965). Only one trial in this review combined psychological and medical placebo, so it is difficult to evaluate interactions between psychological and biological processes on functioning. Weijmar-Schultz et al., (1996) created a placebo surgery condition by

fabricating an operation, and compared it with a real vestibulectomy in the active arm. Both groups also received behaviour therapy which could have confounded placebo effects; however, no differences were found on symptom reduction or elimination between the two treatment arms. With a total sample size of 14, generalisation from this study is limited. In addition, this study was rated as high risk on three domains of quality. Nevertheless, the combined treatment design is useful. Other research has explored the utility of multicomponent treatment models for vestibulodynia (Backman, Widenbrant, Bohm-Starke & Dahlöf, 2008), but evidence on a larger scale is needed to provide informative results about the contribution of medical versus psychological input. Vaginismus trials did not include any comparisons with medical treatment. This prevented direct comparisons with vulvodynia, so conclusions about the relative effectiveness of medical treatment for vaginismus could not be drawn.

The finding that psychological treatments were equally effective for both medically and psychiatrically defined vaginal pain makes it difficult to identify an active component of treatment. As discussed, pain is viewed as a multidimensional problem; therefore different psychological approaches could be acting on different aspects of the pain. For example, supportive therapy may help address emotional distress; CBT may help to address unhelpful cognitions; and behavioural therapy may help with avoidance behaviour, all of which are top-down processes that can influence pain. Similar treatment effects were also found on outcomes of sexual function. Such equivalence is reminiscent of the much debated dodo verdict that suggests all credible psychological therapies have equal effectiveness due to common therapeutic factors, rather than to therapeutic methods specific to therapy type (Wampold, Minami, Baskin & Callen Tierney, 2002). However, this has not been demonstrated when comparing psychological treatments for physical health conditions. One might argue that common therapeutic factors found in medical

settings would be quite different to those found in purely psychological settings. Without a control group it is difficult to determine whether treatments are equally effective or ineffective. Two trials that used control conditions demonstrated a significant effect in the meta-analysis (van Lankveld et al, 2001 & 2006). This implied that psychological treatment is better than no treatment for vaginismus, but this cannot be generalised to vulvodynia.

Due to the variability in treatments and outcomes in the included studies, only two analyses included populations of both medically defined and psychiatrically defined disorders. There was an indication of a differential response to treatment when observing these analyses, in which vaginismus appeared to benefit more from cognitive and behavioural therapies than vestibulodynia, when compared with other psychological interventions on outcomes of sexual anxiety. This supports the idea of vaginismus as an anxiety-driven disorder as opposed to pain-driven disorder. This observation is limited to three studies, one of which had a high risk of bias. With a greater number of studies, a differential effect could be verified or disputed more robustly. Without such data, it is difficult to hypothesise an explanation.

Summary of secondary findings

The secondary aim of this review was to examine differences in effectiveness between vaginal pain types. Individual baseline to post-treatment effects were calculated for each psychological treatment arm to allow for direct comparisons. Two vaginismus studies found large effects for psychological treatment on outcomes of sexual pain (van Lankveld et al., 2006; ter Kuile, Melles, de Groot, Tuijnman-Raasveld & van Lankveld, 2013); these were comparable to and greater than effect sizes found for vulvodynia. Similarly, large effect sizes were found for both medically and psychiatrically defined vaginal pain on outcomes of sexual functioning. This might suggest that pain reduction and sexual function can be improved with

psychological treatment regardless of the perceived aetiology. A difference was found between vaginal pain types when examining sexual frequency. Effects in a vaginismus population were large, but small or no effects were found for the medically defined conditions. This was also evidenced within one trial with two population subsets; a large effect on bibliotherapy for sexual frequency was found for vaginismus, but no effect was found for dyspareunia (van Lankveld et al., 2001). This could indicate a possible differential response to treatment.

Explanations of secondary findings

Equal effect sizes were found across vaginal pain types for pain reduction; this could be understood using a biopsychological model. As discussed in the introduction, commonalities exist in terms of the psychological responses to pain across conditions, including anxiety, fear/avoidance, depression and hypervigilance; therefore, psychological treatment could help to address such factors which maintain and exacerbate pain. A biopsychological model would assume that shared features amongst the vaginal pain conditions could respond similarly to treatment. Vaginismus trials with pain outcomes are needed to confirm this argument as most are focused on sexual and psychological functioning. Reasons for excluding pain as an outcome were not given, but as discussed, it could be due to vaginismus being classified a sexual dysfunction, rather than a pain disorder (DSM-IV-TR, 2000). This may change in conjunction with the new GPPPD classification (APA, 2013), which acknowledges pain as one of four components of a penetration problem.

Equal effect sizes in general sexual function were also found across vaginal pain types, but a discrepancy occurred when focusing specifically on frequency of sexual activity. Vaginismus populations improved on this outcome, whereas vulvodynia did not. One hypothesis for this differential effect could be that psychological treatments for medically defined pain focus exclusively on pain management and discount

factors such as arousal and desire, which may influence sexual frequency. Treatment for vaginismus may be grounded in a more holistic psychological approach to sexual experience (Masters & Johnson, 1966, 1970; Winze & Carey, 2001). However, this does not account for the large effects found for general sexual functioning and small or no effects found for sexual frequency. Sexual function and frequency are somewhat wide-ranging and ambiguous outcomes. Better-defined outcome measures and dismantling studies could help inform which aspects of the programme influenced the various aspects of sexual functioning.

Limitations

The conclusions of this review should be considered in light of several limitations, as well as the variability in the quality of studies outlined above. The sample sizes in most of the included studies were small compared to other RCTs that have evaluated treatment for pain conditions. In a review of psychological therapies for chronic pain, studies were excluded if they contained fewer than 20 participants per treatment arm (Williams, Eccleston & Morley, 2012). If this rule were applied to the current review, six of the included studies would have been excluded. The small sample sizes observed across studies could suggest difficulty with recruitment. This cannot be explained by prevalence rates alone, as pain during sex is experienced by approximately 17% of women; however, the rate of help-seeking populations of women with sexual difficulties is as low as 22% (Moreira et al., 2005). In addition to small samples, a maximum of three studies were included in any one meta-analysis due to the heterogeneity of treatments and outcomes. Together, these factors result in an increased risk of type 2 error (false negatives), and compromise the generalisation of findings and make it challenging to form a clear picture of efficacy. Larger studies would help to detect smaller effects and draw firmer conclusions. The majority of studies compared two or more active treatments; only two studies used a control condition. Without controls, only equality in effectiveness can be concluded.

No vulvodynia studies compared treatment to a control; therefore it cannot be concluded that psychological treatments are better than no treatment. Moreover, when comparing size of effects between studies, it is hard to ascertain whether differences are attributable to the treatment itself, rather than a feature of the trial design.

This review did not include other vaginal pain conditions without identifiable organic causes that can also affect sexual function, such as chronic pelvic pain and deep dyspareunia (Verit, Verit & Yeni, 2006). As previously mentioned, the new GPPPD diagnosis (DSM-5; APA, 2013) could embrace such conditions on the basis that they interfere with the ability to engage in penetrative behaviours. Widening the inclusion criteria for vaginal pain populations would have allowed for more a robust analysis and an exploration of analogous or differential treatment effects for all pain problems linked to sexual dysfunction.

Clinical and research implications

This review has highlighted a lack of studies that compared psychological and medical treatments for psychiatrically defined conditions, and a lack of studies that used a control condition for the medically defined conditions. Only one study used an integrated medical and psychological treatment approach.

It has long been debated whether psychiatrically or medically defined vaginal pain problems should remain separate (Binik, 2005), and with the new GPPPD diagnosis, more research into integrative classifications and treatment approaches could help to improve care for pain-induced sexual dysfunction. Multicomponent models of pain have been investigated and applied to vulvodynia, mainly in the form of pain management programmes (Sadownik, Seal & Brotto, 2000; Munday, Buchan,

Ravenhill, Wiggs & Brooks, 2007); but these have not used controlled methodology, nor considered the relative impact of psychological and physiological treatment components on pain. It could be helpful to investigate multimodal treatment approaches, by varying the proportion of psychological and medical input to see whether differential effects are found. Or, vaginal pain could be placed on a spectrum; at one end would be women with no identifiable physiological cause and significant psychological difficulties, and on the other end would be women with clear physiological causes and few psychological difficulties (although physiological causes are not always detectable, Lonkey et al., 2011). Along this spectrum, effects of treatment could be explored to help determine what type of treatment is suitable for varying presentations. Vaginal pain conditions falling in the middle of the spectrum may benefit most from a combination of medical and psychological interventions, and it would be useful for larger controlled trials could examine both singular and multicomponent treatments. Qualitative research could supplement this, to help understand experiences and treatment preferences of women with vaginal pain.

This review was based on a small number of trials, several of which were low in quality. However, psychological and medical treatments were found to be equally effective in improving pain and sexual functioning for medically defined vaginal pain. If this finding it supported by further research, it could be argued that the less invasive method of treatment should be offered first. NICE guidelines for mental health conditions (e.g. depression & generalised anxiety disorder) often sequence treatment options in terms of invasiveness, recommending non-medical options first if treatments are equally effective (NICE, 2009; 2011). Applying a biopsychological understanding of vaginal pain and allowing patients a choice of treatments, could also help to improve outcomes.

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Part 2: Empirical paper

The effectiveness and acceptability of a computerised guided self-help programme for vaginismus: a mixed methods design

ABSTRACT

Aim: To evaluate the effectiveness and acceptability of a computerised guided self-help programme for women with vaginismus. **Method**: Six women with vaginismus followed a behavioural programme delivered online and supported by weekly telephone calls. A multiple case design was used to quantitatively evaluate effectiveness on primary outcomes of penetration, pain-related fear and pain intensity. Data were analysed using: 1) graphical representations, 2) correlations, 3) the reliable change index, and 4) benchmarking. A thematic analysis on post-treatment interviews was undertaken to examine acceptability and change experienced as a consequence of the programme. **Results:** Over the course of the programme, pain-related fear reduced and successful penetration attempts increased; these outcomes were strongly correlated. Pain on penetration reduced, but was low in intensity from initial attempts. At the end of treatment, three women had successful intercourse; two successfully used objects to penetrate; and one did not improve significantly. The programme was experienced as easy to access, use and understand. The progressive nature of the programme and clinical support was found to be helpful, although frustration was experienced when progress was slow. Other themes found were: increased self-awareness and confidence, normalisation, and a willingness to approach the problem rather than avoid it. Exposure to penetration was identified as the most difficult but most important aspect of change. **Conclusions:** A guided computerised programme is an acceptable and effective way to treat vaginismus. Larger scale studies could evaluate whether its clinical effectiveness is comparable with face-to-face interventions, and if so it could be an economical alternative intervention.

INTRODUCTION

Vaginismus is a female sexual dysfunction, which, according to the DSM-IV-TR (APA, 2000) can be diagnosed if a woman experiences a 'recurrent or persistent involuntary contraction' of the outer vaginal muscles in response to attempted penetration. The involuntary contraction is a physiological response that restricts the vaginal passage and makes penetration painful or unachievable. Although the response is physical, the cause is not explained by medical or physiological factors. Some women develop vaginismus when they first become sexually active (classified as lifelong) and others develop it following a specific experience (classified as acquired). The 2000 classification was recently revised in the DSM-5 (APA, 2013), in which dyspareunia ('recurrent or persistent genital pain associated with sexual intercourse'; APA, 2000) and vaginismus were merged into a new diagnostic category termed 'genito-pelvic pain/penetration disorder' (GPPPD), covering a wider range of pain and penetration problems. This change arose from a lack of evidence for exclusivity between the two diagnoses (Binik, 2010a, 2010b), in addition to problems accurately identifying vaginal muscle contractions as a diagnostic factor (Reissing, Binik, Khalife, Cohen & Amsel, 2004). This new GPPPD diagnosis will take time to permeate relevant NHS settings and diagnostic criteria for vaginismus are still being used today. The current research was initiated before the DSM-5 was published, therefore, the population studied focused on vaginismus.

Large population surveys have demonstrated prevalence rates of sexual dysfunctions in females (including problems with pain, lubrication, arousal, desire and orgasm) in the range of 44-51%, with vaginal pain problems comprising 12-17% (Moreira et al., 2005; Shifren et al., 2009; Vahdaninia, Montazeri, & Goshtasebi, 2009). This does not reflect the prevalence of vaginismus per se, as problems could include chronic pain conditions, such as vulvodynia, which can interfere with sexual function. However, Spector and Carey (1990) found a similar rate (16%) of

vaginismus in women attending sexual health clinics. In the general population, a more conservative estimate of 1% has been suggested (Winze & Carey, 2001).

There is a lack of evidence for predictors of vaginismus, but associated cognitions have been found. These include: catastrophising about pain, holding a negative self-image, fearing intimacy, fearing loss of control and feeling disgusted about sex (Borg, Peters, Schultz & de Jong, 2012; de Jong, Overveld & Schultz, 2009; Reissing, 2012). Such beliefs may have existed prior to the problem, creating a susceptibility to developing vaginismus. But most research has explored beliefs in populations in which the problem is established; therefore, cognitions may serve to maintain the fear-pain response. Although cognitive explanations can be useful for understanding idiosyncratic presentations and applying CBT techniques, the evidence for treatment suggests a powerful behavioural component (ter Kuile, Lankveld, de Groota, Melles, Neffs & Zandbergen, 2007).

Wijma and Wijma (1997) put forward a behavioural model (Figure 2), which includes components of classical and operant conditioning. Initially an association is formed between penetration (unconditioned → conditioned stimulus) and pain (unconditioned → conditioned response). Thereafter, the response is maintained by negative reinforcement (avoidance of pain). This model recognises that internal events strengthen and maintain the avoidance response, including mood states (e.g. fear) and unhelpful cognitions. Once avoidance of penetration is established, sexual functioning is impaired.

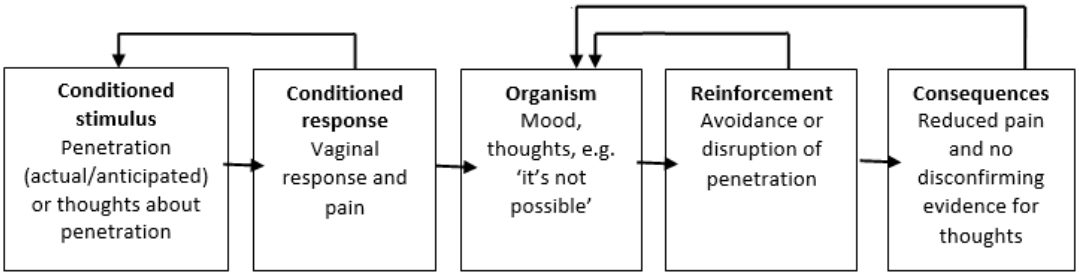

Figure 2: Behavioural model of vaginismus

A behavioural understanding of vaginismus has been used to develop treatment approaches, such as graded exposure (described below). Presently, there are no evidence-based guidelines to specifically inform the treatment of vaginismus; but the British Association for Sexual Health and HIV recommend routinely treating vaginismus with behavioural intervention (Crowley, Richardson & Goldmeier, 2006). Behavioural techniques can be supported by cognitive strategies (e.g. targeting associated cognitions) to help reduce fear and encourage approach behaviours. Cognitive-behavioural approaches have been commonly evaluated in controlled clinical trials on vaginismus (Melnik, Hawton & McGuire, 2012).

Typically treatment involves five main components (Hawton, 1985; Jeng, Wang, Chou, Shen & Tzend, 2006; Masters & Johnson, 1970; Winze & Carey, 2001) as follows:

1) Education. This component helps the person (and her partner) to understand and normalise the problem. Education in itself can help to alleviate anxiety and establish rationale and motivation for treatment.

2) Relaxation. As a fear-driven disorder, vaginismus is characterised by sexual anxiety, but is also frequently co-morbid with general anxiety (Watts & Nettle, 2010). Therefore, learning how to identify tension and relax muscles is an important part of the treatment. As vaginismus is accompanied by avoidance, relaxation can be a gentle way to re-introduce self-focus.

3) Pelvic floor exercises. These exercises are introduced to teach women to differentiate between sensations of contracting and relaxing the muscles around the vagina. Pelvic floor exercises help to increase awareness of vaginal tension and can also help to increase a sense of control, both of which can help when penetration is attempted (Kegel, 1948).

4) Sensate focus. This involves exploration of bodily touch without the pressure of penetrative activities. Removing penetration from sexual activity allows women to build confidence (with themselves and their partners) and enjoy a tactile experience without a fear of pain.

5) Exposure. Women are asked to insert objects of graded sizes (called trainers) into their vaginas. Introducing the physiological sensations of penetration (conditioned stimulus) with reduced anxiety (habituation) helps to eliminate the vaginal spasm (conditioned response) that causes the pain (Masters & Johnson, 1970). When women are comfortable using the trainers, a transition to sexual intercourse can be made. Graded exposure has been argued to be the most important component for driving behaviour change (ter Kuile et al., 2007; 2009).

Due to the multi-factorial treatment for vaginismus, the relative impact of each component remains unclear. Some have argued that outcomes can be predicted by factors external to treatment, such as the quality of the patient's relationship or motivation of the patient's partner (Hawton, 1995). Melnik, Hawton & McGuire (2012) undertook a systematic review examining the efficacy of psychological treatment for vaginismus. From five trials that were included, no significant differences were found between treatment types. Due to the small number of trials, the authors suggest that the findings should be interpreted cautiously. Larger trials could help to identify smaller treatment effects. The largest trial included in the review examined 117 women with lifelong vaginismus (Van Lankveld, ter Kuile, de Groot, Melles, Nefs & Zandbergen, 2006). It compared group CBT, bibliotherapy

and waiting list control. Both treatment conditions used a self-help manual, but the bibliotherapy group only had minimal contact with a professional (brief weekly phone calls, as opposed to weekly CBT groups). The study reported that both treatment arms led to successful vaginal intercourse; interestingly, more so in the bibliotherapy (18%) than the CBT group (9%) and control (0%) post-treatment. This effect was maintained at 12 months, in which the rate of successful vaginal penetration in the CBT group was 21% and in the bibliotherapy 15%. Support for bibliotherapy has also been evidenced in another RCT, in which self-reported complaints reduced following treatment (van Lankveld, Everaerd & Grotjohann, 2001).

There is limited good quality evidence despite vaginismus being recognised as a prevalent sexual dysfunction. Overall, results from these trials evidence do not evidence a high rate of success, but show that self-help in the form of bibliotherapy is a plausible alternative to face-to-face treatment.

A large global study of sexual attitudes and behaviours revealed that 44% of women with a sexual problem did not seek help (Moreria et al., 2004). Of those who did, 16% did so via anonymous routes (such as books, helplines or the internet), which is similar to the rate of people who sought help from a doctor (19%). Shrifen et al., (2009) found a higher rate of help-seeking via professionals (35%) most of whom were gynaecologists or GPs; however, only 6% specifically visited their doctor to address the sexual problems and most women preferred the clinician to initiate the conversation. Several reasons for not seeking help include: embarrassment, concern that the doctor would be uncomfortable, not having enough time, not being asked by a clinician, and not seeing the problem as serious or treatable (Moreria et al., 2004; Vahdaninia et al., 2009). Barriers to help-seeking are wide-ranging, but seem to relate to a shared anxiety of disclosure. Help-seeking behaviours may also be culturally dependent.

In the past decade, a surge of health related information has become available online. Patients often use the internet to investigate possible causes and treatments for their symptoms, instead of or before seeking professional help (Ybarra & Sunman, 2006). This mode of help-seeking may be particularly pertinent for those who are embarrassed to talk about their difficulties, but also for those who have less time or resource to attend appointments; information can be accessed immediately. Numerous computer-based self-help programmes have emerged in the past decade for the treatment of anxiety and depression, and have been found to be a highly acceptable mode of treatment based on adherence rates (80%) and overall satisfaction (86%) (Andrews, Cuijpers, Craske, McEvoy & Titov, 2010). Computerised interventions targeting sexual behaviour have also been reviewed, and show stronger effects than face-to-face interventions at improving knowledge of sexual health, as well as a small effect on promoting safer sex (Bailey et al., 2010).

There is a lack of validated computer-based programmes for sexual dysfunction, and so far no computerised interventions specifically for vaginismus have been formally evaluated. Some women may access information on the internet, and there are websites that offer purchasable self-help guides (www.vaginismus.com) and blogs, which aim to support women (www.livingwithvaginismus.blogspot.co.uk). However, it is unclear how the guides were developed and whether or not they are effective.

From examining the evidence base for vaginismus and the use of computerised interventions, a computer-based self-help programme could be an accessible, effective an acceptable way of treating vaginismus. The current research aimed to answer the following questions:

1. Following the use of a computerised self-help programme for vaginismus, will there be a change in: ability to achieve penetration, intensity of pain, pain-related fear, general anxiety and relationship satisfaction?
2. Will a computerised self-help programme for vaginismus an acceptable and helpful form of treatment for participants who experienced it?

METHODS

Participants

Participants were six women seen in an inner London genitourinary medicine clinic (see Table 1 for demographics). All participants were assessed by a gynaecologist or sexual health specialist and diagnosed with vaginismus. Women were included if they were over 18 years old and had capacity to consent. Relationship status and heterosexuality were not inclusion criteria because the programme was designed so that penetration could be achieved with or without a penis. Women were excluded if they were: pregnant; starting, undergoing or completing menopause (as pain can be related to changes at this stage); insufficiently fluent in English to read and understand the self-help guide; did not have access to a computer at home or were unable to use a computer. Women with vaginal pain on penetration due to other factors for example, tissue or nerve damage, sexually transmitted infections, urinary tract infections, lack of lubrication) would be excluded from a vaginismus diagnosis.

Of eight women who consented and started the programme, two dropped out. One moved abroad three weeks into the programme and contact could not be maintained. The other went abroad for a month after two weeks of starting the programme and did not re-engage on her return.

Table 4

Participant demographics

ID	Age	Ethnicity	Type	Last attempt at sexual intercourse	Pain rating for last penetration attempt	Relationship status	Previous treatment
P1	30s	White European	Lifelong	8 months	10/10 (intercourse)	Heterosexual relationship 2 years	None
P2	20s	White European	Acquired (1 year)	3 days	10/10 (intercourse)	Heterosexual relationship 3.5 years	None
P3	30s	White European	Lifelong	1 month	5/10 (trainer 4)	Not in a relationship	Trainers self-directed
P4	30s	White European	Acquired (3 years)	2 months	5/10 (intercourse)	Heterosexual relationship 2 years	Vibrator self-directed
P5	20s	White British	Lifelong	1 month	5/10 (intercourse)	Heterosexual relationship 6 months	Trainers self-directed
P6	<20	Asian British	Acquired (3 months)	4 days	8/10 (intercourse)	Heterosexual relationship 3 months	None

Process

Participants were offered a choice between standard care (face-to-face psychological therapy) and the computerised self-help programme. It was explained to participants that the difference between treatments was the mode of delivery, not the content. Participants were given an information sheet (Appendix 3) and were allowed one week to consider their choice. If they opted for the computerised programme they were invited to attend an initial meeting with the researcher. This meeting lasted approximately 45 minutes and involved: explaining the treatment process in detail, obtaining consent (Appendix 4), demonstrating how to use the programme, giving participants trainers (supplied by the clinic) and completing baseline questionnaires.

Based on previous research (van Lankveld et al., 2006), the programme was designed to run for 6-8 weeks, with the possibility to extend to 9 weeks depending on the pace of the individual. If by 9 weeks participants had not achieved their goals, they were invited to have a review with the researcher and their care was continued in the GUM clinic if necessary. Weekly telephone contacts with the researcher of 15-20 minutes were scheduled as part of the intervention.

Intervention

The self-help programme was developed using existing literature, consisting of textbooks, articles and empirical research pertaining to specific clinical techniques. A bibliotherapy guide used in a previous RCT (van Lankveld et al; 2006) was also obtained from the authors and translated to help inform the current programme. As a predominantly behavioural intervention, behaviour change techniques were integrated to try to promote change, including: goal-setting, goal-review, self-monitoring, feedback, behavioural practice, graded tasks and self-reward (Abraham & Michie, 2008). A draft of the programme was sent to specialists in the field (sexual health clinicians and researchers) for consultation. Feedback from six specialists was collated and revisions were made accordingly.

The programme was developed and run using internet based software Prezi (www.prezi.com). This software facilitates the development of a mind-map, in which information and key concepts are linked together in a simple sequential way, using text, diagrams and video. The steps in the programme were set out visually on a pathway (see Appendix 5 for a screenshot of the programme).

The following components were included in the programme.

- **Step 1: Educational material.** This step explained the diagnosis of vaginismus, explored possible causal factors and maintenance cycles, used

anatomical diagrams to demonstrate physiological change, and included fictional vignettes about experiences of vaginismus. Participants moved to Step 2 when they had finished all of the reading.

- **Step 2: Relaxation and pelvic floor exercises**. This step asked participants to introduce relaxing activities into their routine, as well as practising more formal relaxation techniques. Progressive muscle relaxation was implemented to help participants notice differences between muscle tension and relaxation, and become more aware of tension held throughout the body. The pelvic floor exercises aimed to increase identification of tension specifically in the vaginal area and to promote a sense of control over these muscles. Participants moved to Step 3 when they had tried progressive muscle relaxation three or four times, had done pelvic floor exercises every day for at least a week and were able to notice differences in tension and relaxation.
- **Step 3: Body awareness**. This step required participants to explore their body using touch and massage, either alone or with their partners. Body awareness specifies that no penetrative activities should take place during this exercise, in order to remove any related fear or pressure. Encouraging sexual exploration whilst feeling relaxed can help to build confidence and allow for an enjoyable sensual experience. This aimed to help women feel more prepared for the final stage of the programme. Participants were given the choice of whether or not to undertake body awareness because some women already feel confident with touching their bodies. Those who chose to do body awareness, moved to Step 4 when they had tried it three or four times and felt ready to move on to the trainers.
- **Step 4: Exposure.** This step involved gradual exposure to penetration to familiarise participants with the sensations of objects contained in their vagina whilst feeling relaxed. Women chose to start using their fingers or trainers. The size of the object contained was increased slowly and women only moved

on to the next sized object when they could contain the previous object with no pain or minimal discomfort. Most participants aimed to attempt sexual intercourse; therefore, the transition between the trainers and their partner's penis was made. Participants without partners could choose to transition to a vibrator, which mimics sexual intercourse more closely than the trainers. Treatment ended when participants were able to have successful penetration either with their partner's penis (or a vibrator if no partner) or if the participant reached the 9 week maximum duration period.

Participants received a telephone call each week for the duration of the programme from the researcher (trainee clinical psychologist). The first call occurred on the day that participants gained access to the programme and focused on setting goals for the ensuing 6-8 weeks. All calls made after the first contact followed a similar informal structure. First, diary activity (self-monitoring) from the week was discussed, in terms of what activities were undertaken and reflecting on the fear and pain scores. Second, any difficulties or points of interest identified were explored using a problem-solving approach. Third, goals were reviewed and feedback was given regarding the progress of the participant. Fourth, a plan was made collaboratively for the upcoming week. Finally, participants had the chance to ask any final questions. The researcher undertaking the telephone contacts had regular supervision from a senior clinical psychologist.

Ethical approval for the evaluation of this intervention was obtained from the Central London NRES Committee (Ref: 13/LO/0487; Appendix 6).

Quantitative outcomes

The programme was evaluated using several outcome measures selected on the basis of functional and psychological factors experienced with vaginismus.

The following two outcomes were collected daily for the duration of the programme. Data were collected using online diaries; each participant had an anonymous account and entered outcomes in code to preserve confidentiality.

- **Pain-related fear.** As fear maintains avoidance in vaginismus, a fear score was collected daily throughout the programme, in addition to a week of baseline scores. Participants were asked to rate how fearful they were of pain associated with penetration on a scale from 0-10, 0 being 'not at all fearful' and 10 being 'extremely fearful'. Psychometric properties of this exact scale have not been previously evaluated; however, a similar scale used in a trial of vaginismus found good internal consistency (α =0.82) and test-retest reliability (r =0.73; ter Kuile et al., 2007).
- **Programme activities**. To monitor the frequency and duration of activities included in the programme, participants recorded what activity they tried, whether or not it was successful, and how long they did it for. Embedded within this information were questions from the Primary Endpoint Questionnaire (PEQ; van Lankveld et al., 2006; Appendix 7). The 7-item measure looks at whether penetration on a range of levels (self or partner one finger or two fingers, other objects inserted by self or partner and sexual intercourse) was attempted and successful in the past 4 weeks. Scores ranged from 0-7; successful or partially successful attempts scored 1 and unsuccessful or no attempt scored 0. Internal consistency of the PEQ has been found to be satisfactory (α = 0.72). The PEQ was adapted to accommodate the use of trainers instead of fingers, and for women who did not have partners as follows: one finger scored equivalently to trainer 1 (smallest), two fingers equivalently to trainer 2 (second smallest), other objects equivalently to trainers three and four, and intercourse equivalently to a vibrator. PEQ scores for P4 were also prorated to account items that

required a partner's finger. P4 had a successful smear test during the programme which was coded as successful object inserted by other.

Pain was rated every time the participant engaged in a penetrative behaviour.

- **Intensity of vaginal pain.** As pain is associated with attempted penetration in vaginismus, a pain rating was collected throughout the programme on days that penetration activity occurred. Pain was measured on a numerical rating scale from 0-10, 0 being 'no pain' and 10 being worst possible pain' (McCaffery & Pasero, 2001). Moderate construct validity (r=0.85; Ritter, González, Laurent & Lorig, 2006) and test-retest reliability (r=0.90) has been found (Lundeberg et al., 2001).

Two further outcome measures were administered at baseline, mid-treatment and post-treatment.

- **Anxiety**. Heightened anxiety is associated with women with vaginismus (Watts & Nettle, 2010). The GAD-7 is a seven-item measure of general anxiety, which examines worry, nervousness, restlessness, irritability, trouble relaxing, and feeling afraid or panicked. Scores range from 0-21, and the cut-offs used were: 0-4 (none), 5-10 (mild), 11-15 (moderate), 15-21 (severe) (Department of Health, 2011). The GAD-7 has been found to have good internal consistency (Cronbach's α=0.89; Lowe et al., 2008), high test-retest reliability (0.83) and optimal levels of sensitivity and specificity (89% & 82% respectively; Spitzer et al., 2006).
- **Relationship satisfaction**. Interference with sexual function can impact considerably on relationships (Byers, 2005). For participants who were in relationships the Couples Satisfaction Index (CSI-16; Funk & Rogge, 2007) was administered. This is a 16-item measure that examines the extent to which the relationship offered happiness, strength, comfort, reward,

satisfaction and other emotional aspects. It gives a single score between 0-81 and has been found to have high levels of internal consistency (α= 0.98) and strong convergent validity with other existing measures (Funk & Rogge, 2007).

Qualitative outcomes

As this programme used a novel mode of treatment delivery, semi-structured interviews were used to gain a more in-depth understand of participant's experiences. All interviews were conducted within a week of finishing the programme and aimed to explore two broad domains: 1) Acceptability of the programme (e.g. likeability, usability, comprehensibility & the process of treatment), and 2) what changes occurred as a consequence of the treatment and why. The interview schedule included acceptability questions derived from feasibility research (Brug, Schaalma, Kok, Meertens & Van der Molen, 2000; Collings et al., 2012). The interview also encompassed questions from the Client Change Interview (Elliott, Slatick & Urman, 2001; Appendix 8), which explored aspects of the programme participants found helpful or difficult, changes they noticed in themselves, what was attributed to change, as well as a question on suggested improvements.

Design

A multiple-case literal replication design was used to measure quantitative change. This design uses the same methodology as a single-case design, but is considered more robust, as a larger sample size allows for the replication of findings (Yin, 1994). Literal replication means that similar outcomes are predicted for each case because they are selected for their likeness. In this research, similarity was defined on the basis of the specified inclusion and exclusion criteria, and adherence to the programme. This design uses a small number of participants; therefore, typical sampling logic does not apply. The aim is not to generalise to the target population,

but to closely explore a theory and practice-derived application over time. Yin (1994) suggests at least 2-3 cases are required to provide compelling data, or enough to reach saturation. To account for the small sample, a larger number of data points are collected.

The qualitative data would be used to understand further the acceptability of the programme and a more in-depth account of change. This follows a 'partially mixed sequential dominant status design' (Leech & Onwuegbuzie, 2009), as the two methods of data collection are undertaken sequentially, and greater emphasis is placed on the quantitative outcomes.

Quantitative analyses

The data were analysed in four ways:

1. **Graphical representation**. Change was observed graphically for outcomes collected for the duration of the programme. This method has been used in single-case analyses to infer change based on observed scores following the introduction of an intervention (Barlow, Nock & Hersen, 2008).
2. **Correlation**. As data were not normally distributed, Spearman rank correlations were used to calculate associations between pain-related fear and penetration for each participant.
3. **Reliable Change Index**. All outcomes were analysed using the reliable change index (RCI; Jacobson & Truax, 1991). The RCI calculates whether individual participants improved, did not change or deteriorated between two time points. Internal consistency coefficients were used to calculate reliable change scores; if unavailable, test-retest reliability was used. Reliable change criterion for all outcomes can be found in Table 5.
4. **Benchmarking**. Effect sizes were calculated and compared to previous RCTs on outcomes of penetration and fear to help determine whether levels of change were meaningful. Effect sizes for pre-post change were used.

Table 5

Reliable Change Criterion for all outcomes

Outcome measure (range)	Mean at assessment	Standard deviation at assessment/ standard error of change	Reliability coefficients	Reliable Change score*
PEQ (0-7)	1.71	0.95 / 0.34	Internal consistency 0.72 (van Lankveld et al., 2006)	**1.39**
GAD-7 (0-21)	9.71	6.65 / 3.12	Internal consistency 0.89 (Spitzer et al., 2006)	**6.11**
CSI (0-81)	63.33	16.87 / 3.37	Internal consistency 0.98 (Funk & Rogge, 2007)	**6.61**
Pain scale (0-10)	7.14	2.27 / 1.02	Test retest reliability 0.90 (Lundeberg et al., 2001)	**1.99**
Fear scale (0-10)	7.71	1.60 / 1.18	Test-retest 0.73 (ter Kuile et al., 2007)	**2.30**

Note: *The level of change necessary on each outcome to detect a reliable change

Qualitative analysis

Interviews were transcribed verbatim. Qualitative data were used to understand the experience of the programme in detail and to explore factors that were not captured by quantitative outcomes. Interviews followed certain lines of enquiry (acceptability and change), so thematic analyses were conducted under these pre-defined headings. Similar methods have been used in previous research, with a specific interest in uncovering themes that relate to acceptability (Finucane & Mercer, 2006) and change (Orford et al., 2006). Under the two broad headings of acceptability and

change, themes were explored using Braun and Clarke's (2006) suggested method for qualitative analysis. The following phases were carried out: 1) familiarisation with data; 2) generating initial codes; 3) searching for themes; 4) reviewing themes; and 5) defining and naming themes. The software NVivo10 was used to help organise data into themes. To enhance the credibility of the analysis, another researcher examined all data and a consensus was reached (Barker & Pistrang, 2005).

It is recommended that researchers state their position regarding the research topic. I am a trainee in my late-twenties who has previously worked in a sexual health setting and implemented similar behavioural techniques with women with vaginismus. I hold the opinion that CBT is the most appropriate treatment for vaginismus. As the author of the computer programme, I was keen for it to be helpful; but, I did not hold the assumption that it would be a success, because no computer-based programmes have been evaluated previously.

RESULTS

Six women completed the programme. Days taken to complete the programme ranged from 38-61 (*M*=54.8 days). Least time was spent on Step 1 (education) *M*=2.3 days; most time was spent on Step 4 (exposure) *M*=42.3 days (see Table 6). Three women chose not to complete Step 3 (body awareness), reporting that comfort with bodily touch was not a problem for them. P1, P2 and P6 ended the programme when they were able to have full sexual intercourse with minimal or no pain. P3 had no partner so ended the programme when she was able to use her vibrator comfortably. P4 and P5 had not attempted intercourse by the end of treatment. P5 reported that she intended to attempt intercourse in the near future. P4 decided to continue using her vibrator until she felt comfortable before attempting intercourse.

Table 6

Programme duration and penetration outcome

ID	Baseline /treatment duration	Days per phase (steps 1-4)	Contact amount (number & total call duration)	Hours of penetration exposure (excluding intercourse)	Penetration at end of treatment (pain rating)
P1	10 days 52 days	3, 14, 0, 45	8 calls (2.5 hours)	10.6 hours	Intercourse (0/10)
P2	8 days 38 days	1, 17, 7, 21	7 calls (1.9 hours)	2.6 hours	Intercourse (0/10)
P3	7 days 60 days	1, 7, 11, 48	10 calls + 1 email (2.7 hours)	10.1 hours	Vibrator (1/10)
P4	7 days 61 days	7, 7, 0, 56	9 calls + 1 email (2.5 hours)	8.9 hours	Vibrator (3/10)
P5	8 days 59 days	1, 12, 0, 46	9 calls (2.6 hours)	6.8 hours	Trainer 4 (2/10)
P6	8 days 59 days	1, 10, 11, 38	10 calls (2.6 hours)	4.3 hours	Intercourse (2/10)

Graphical representation

Outcomes of pain-related fear and penetration (PEQ) were collected every day for the duration of the baseline and programme. Pain ratings were collected at every penetration attempt. These outcomes are represented graphically (Figure 3, Figure 4 & Figure 5) and have been corroborated with diary activity and ethnographic data (notes from telephone sessions) to try to understand the observed patterns.

Figure 3 shows pain-related fear ratings for all participants. A general downward trend can be observed, in which fear reduced over time. All participants' scores reduced by ≥50% subsequent to the exposure phase; the mean number of days penetration activities were attempted (before ≥50% reduction) was 11.7, but the variance was substantial (SD= 9.8).

When observing scores in the baseline, four participants seemed to have stable fear scores (P2, P3, P4 & P5), whereas the other two experienced a dip in fear before stabilising. P1 reported that this dip was due to knowing that treatment would commence soon. P6 reported that talking about the programme made her feel less fearful of intimacy with her partner during the baseline period.

When observing the patterns of change during treatment, some shifts in fear appear quite erratic. In some cases this was accounted for by the introduction of new stages of the programme. For example, P4's fear drops from 9 to 0 on the first day that she attempted the trainers, doing so without any pain. Her scores increase again when attempting to use trainer 4, with which she made minimal improvement with thereafter. P1 shows a peak of pain-related fear near the end of the programme; this represented the first time she had successful intercourse (corresponding pain score was 0). A more steady reduction in pain-related fear was observed for other participants. For P2 & P5 these reductions appeared to mirror successful attempts at using the trainers, but for P3 and P6 the reduction was not clearly linked to specific activities. P3 reported that her fear of at the end of treatment remained at 2/10 because without a partner, she had not been able to attempt sexual intercourse.

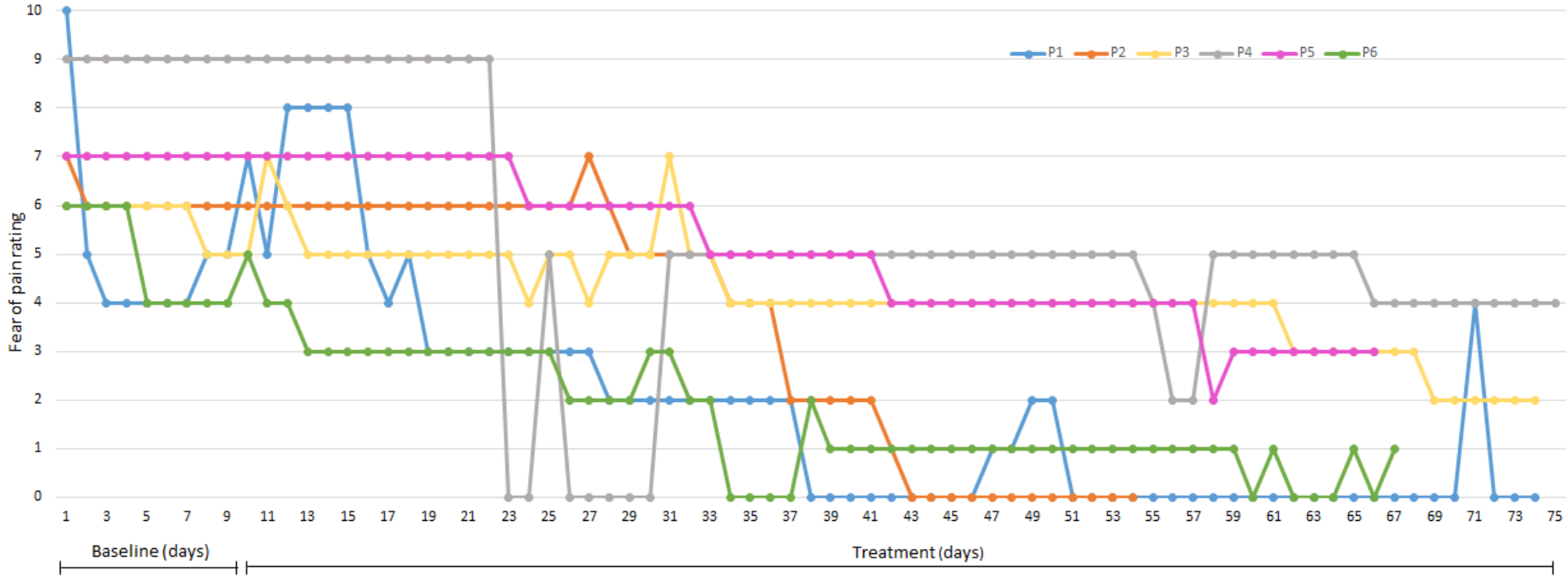

Figure 3: Pain-related fear scores for duration of programme

Figure 6 shows penetration behaviours for all participants. It should be noted that as the PEQ assesses penetration over the last 4 weeks, scores are cumulative and therefore less erratic than the pain-related fear scores. A general upward trend can be observed, in which the number of successful penetration behaviours increases over time. Five participants' scores increased by ≥50% in a mean number of 23 days (SD=9.14).

Around week three (22-27 days) all six participants show an increase in penetration behaviour, subsequent to steady scores. P1 increased sharply by 3 points when she attempted trainers 1-4 all in one day; her score continues to increase over the following weeks, accounted for by penetration activities with her partner and intercourse at the end. P2 demonstrated a steadier increase in penetration behaviours, increasing the sizes of trainers and partner's fingers progressively over time and ending with intercourse. P3 and P6 were already engaging in some penetration behaviours before the programme started, so change was less remarkable. P6 was the only participant who was attempting sexual intercourse during the programme alongside the trainers; however, intercourse went from being unsuccessful at baseline to successful (indicated by 1 point increase at day 26). P5 had a temporary decline in functioning because she used trainer 4 for more than four weeks, so scores for the smaller trainers expired. P4 only increased one point in this scale, but in fact her use of trainers/vibrator led to an increase of 3 points, whilst penetration activities with her partner decreased by 2 points.

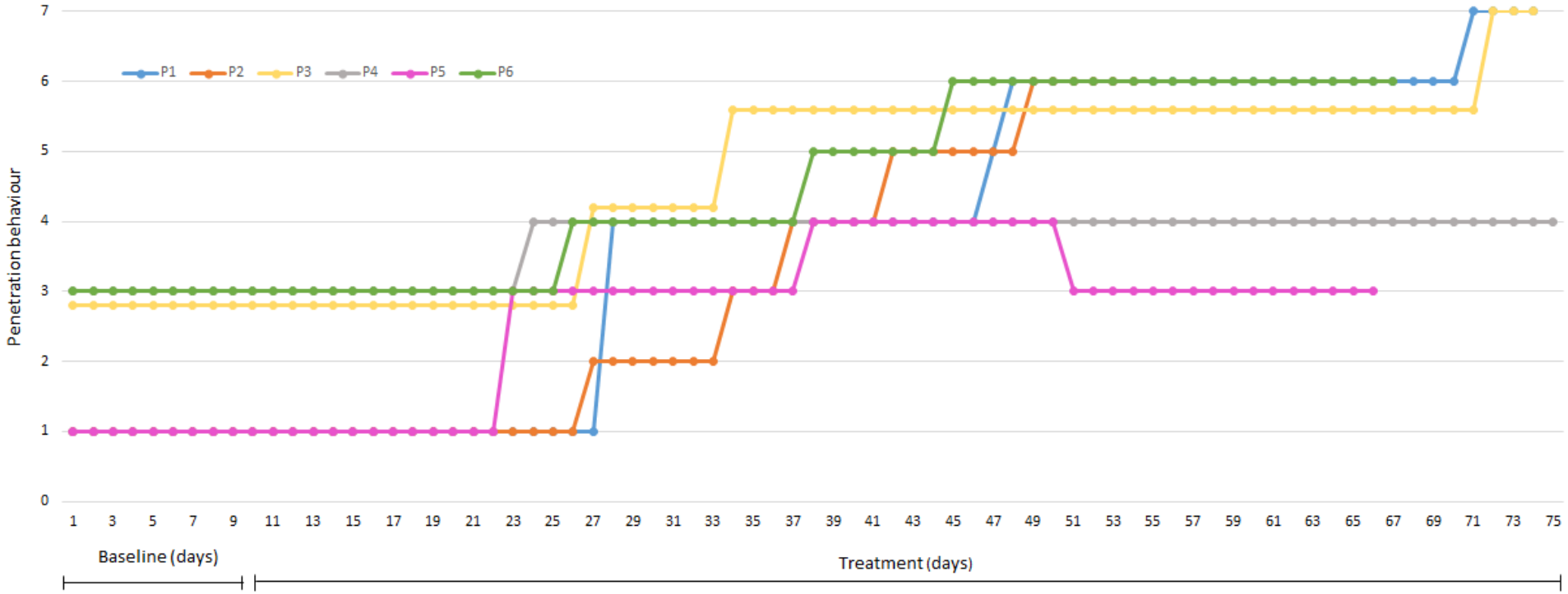

Figure 4: Penetration behaviours for duration of programme

Pain ratings were collected on days when penetration activity was attempted. The programme was designed to prepare participants for comfortable penetration and this resulted in pain scores tending to be low at the initial penetration attempt. Ratings from trainer 3 have been presented (Figure 5) because all participants used this trainer during the exposure phase. None of the women scored more than 2/10, and ratings tended to go down to 0/10 after several attempts. Initial attempts at trainer 4 were rated higher on pain (range 0-6), but after several attempts these also reduced to 2/10 or less in all women except P5 whose pain remained at 3/10.

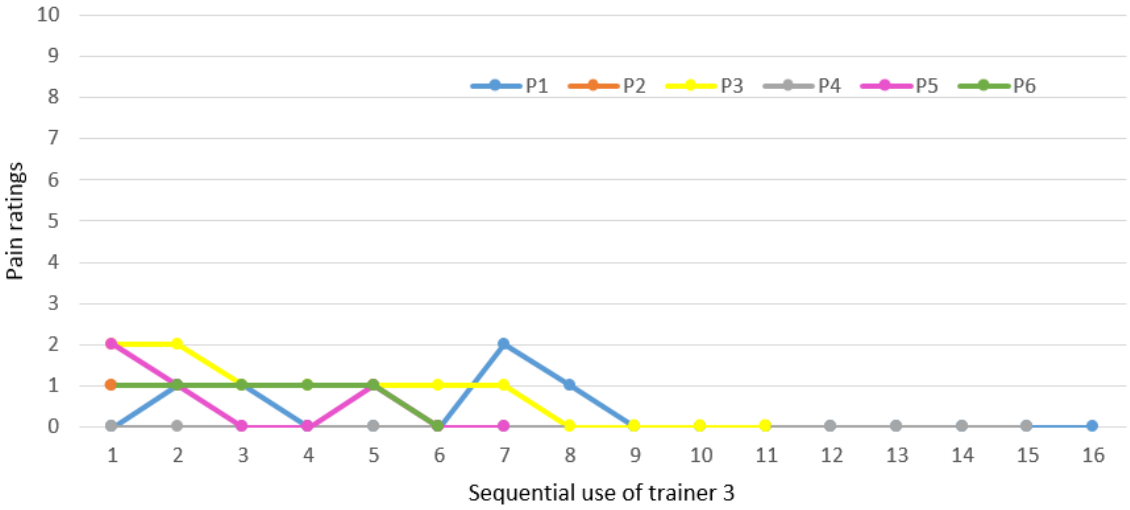

Figure 5: Pain ratings for trainer 3

Correlations

To evaluate associations between pain-related fear and penetration, scores for each participant for the duration of the programme were correlated. All correlations indicated a significant strong negative relationship between fear and penetration ($p<0.0001$), which helps to confirm the observational data above. The strength of these associations could indicate a dependability of the two variables on one another.

Table 7

Correlations between pain-related fear and penetration

Participant	Spearman's r
P1	-0.81
P2	-0.94
P3	-0.88
P4	-0.78
P5	-0.96
P6	-0.84

Reliable Change Index

Change from baseline to end of treatment was evaluated using the Reliable Change Index. When fear scores in the baseline phase were unstable, the score representing the highest level of functioning was selected in order to detect the smallest amount of change. Pre-treatment pain ratings were estimates given by participants of their last penetration attempt (intercourse or trainers). These related to penetration attempts ranging from 3 days to 8 months ago; therefore, the reliability of this outcome is questionable. End of treatment pain ratings were taken from the last penetration attempt of intercourse (P2, P3, P6), vibrator (P3, P4) and trainer 4 (P5). The graphs below represent three levels of change based on the reliability of each outcome. The top left triangle represents a reliable improvement; the section within the dotted lines represents no change; and the bottom right triangle represents a reliable deterioration.

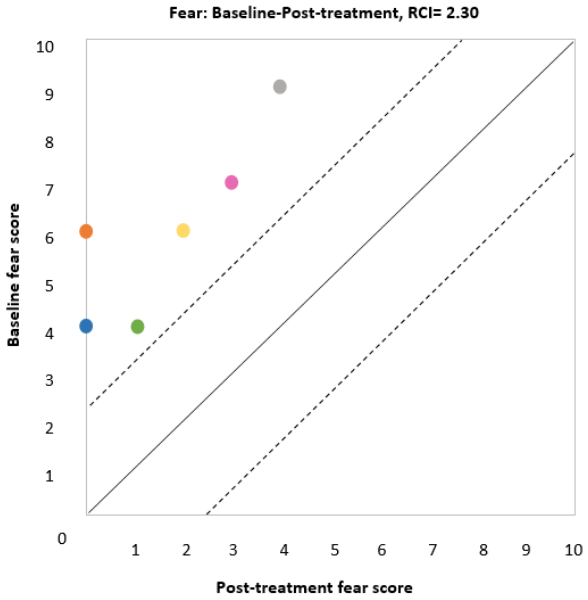

Figure 6: RCI for pain-related fear

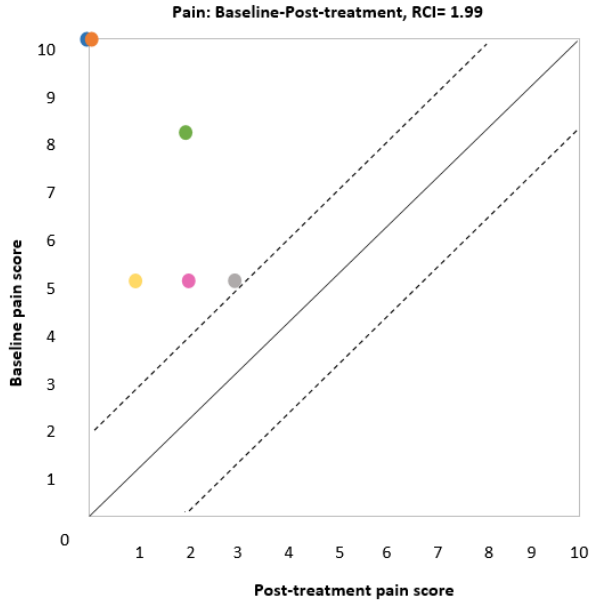

Figure 7: RCI for pain

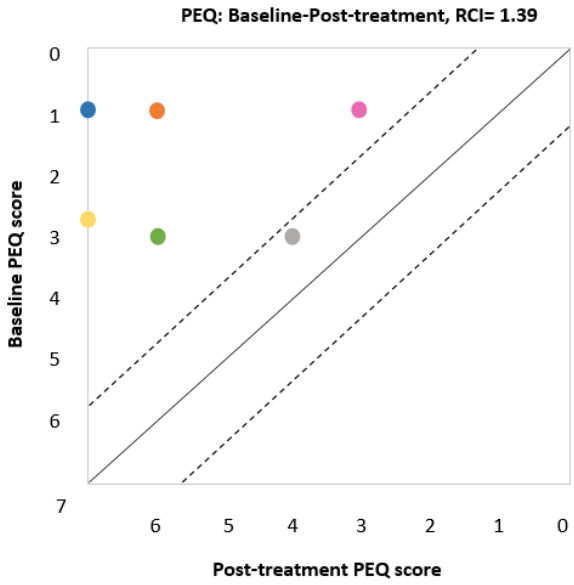

Figure 8: RCI for penetration behaviour

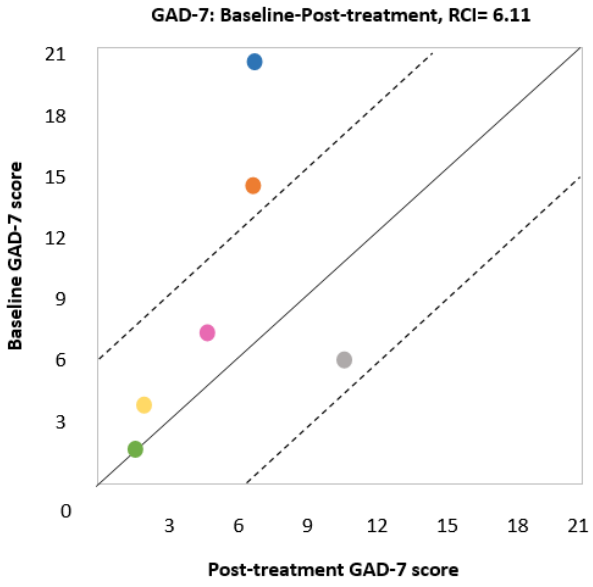

Figure 9: RCI for anxiety

All participants showed a reliable improvement in pain-related fear (Figure 6). P4 who had not attempted intercourse by the end of treatment still experienced a reduction in fear, possibly because she had some success using the smaller trainers and vibrator. However, on intensity of pain (Figure 7) P4 only showed a marginal improvement. All other participants demonstrated a reliable improvement in pain intensity. Two women (P1 & P2) showed pain scores which decreased from 10 to 0. This seems an extreme change and could be explained by the inaccuracy of the pre-treatment rating; however, pre-treatment pain was severe enough to prevent successful intercourse and post-treatment intercourse was successful and pain-free.

Five of the six women improved reliably on penetration behaviours (Figure 8). It could be argued that this outcome is inherent in the exposure phase of treatment as patients are asked to engage in penetration behaviours; but, in order for these to be scored as 'successful' a low/tolerable level of pain is required or penetration cannot occur successfully (supported by pain ratings, Figure 5).

General anxiety outcomes (Figure 9) showed that two women reliably improved and four women showed no change. General anxiety was not a specific target of the intervention, but the programme contained techniques that focused on anxiety based thoughts and physiology, which may have impacted positively on general anxiety.

Couples satisfaction outcomes were only available for four participants. For three participants (P2, P4, P5) no reliable change was found, but their baseline scores were already high (M=70.3, SD=0.6) indicating good relationship satisfaction from the outset. P6 reliably deteriorated on this outcome, but reported that this was not

linked to sexual issues. P3 was not in a relationship and P1 ended her relationship at the end of treatment, but also reported that this was not related to sexual issues.

Benchmarking

Table 8 includes pre-post treatment effect sizes for fear and non-coital penetration calculated from the current study and benchmarked against previous RCTs. These RCTs were selected as benchmarks because they used similar populations and data were available on shared outcomes. Fear was benchmarked against an RCT that evaluated an exposure-based treatment for life-long vaginismus (Ter Kuile, Melles, de Groot, Tuijnman-Raasveld & van Lankveld, 2013). Penetration behaviour was benchmarked against an RCT that evaluated bibliotherapy for life-long vaginismus and reported on non-coital penetration (van Lankveld et al., 2006).

The RCTs demonstrated large effects on pain-related fear and penetration; the current study also found large effect sizes for both outcomes. As a multiple-case design the effects of the current study are not intended to be representative of a wider population; however, it is useful to know that obtaining a large effect size is not unrealistic, when comparing to findings from controlled trials.

Table 8

Benchmarking pre-post effect sizes for penetration and fear

Outcome	Pre-treatment M / SD (n)	Post treatment M / SD (n)	Effect size
Pain-related fear			
RCT (ter Kuile et al., 2013)	3.10 / 1.11 (35)	1.98 / 0.92 (35)	d = 1.10
Current research	6.00 / 1.90 (6)	1.67 / 1.63 (6)	d = 2.44
Non-coital penetration			
RCT (van Lankveld et al., 2006)	0.8 / 1.4 (38)	2.3 / 1.8 (38)	d = 0.93
	1.80 / 1.30 (6)	4.83 / 1.17 (6)	d = 2.45

Qualitative analysis

All six participants were interviewed at the end of the programme (*M*=26 minutes). Data were categorised under two broad headings (acceptability and change) and within these categories a general thematic analysis was undertaken (see Appendix 9 for annotated example). Fourteen themes were found within five domains (see Table 9). The domains were informed by the types of questions asked and are as follows: 1) Practicality of use, which discusses experiences of the interface and usability of the programme; 2) Process of treatment, which examines the journey through the programme; 3) Suggestions for improvement; 4) Cognitive changes identified; and 5) Behavioural attributions to change identified. Themes are supported by extracts from the interviews.

Table 9

Categories, domains, themes and sources

Category	Domain	Theme	Number of sources
Acceptability of programme	1. Practicality of use	1.1 Convenience	6
		1.2 Understanding the content	6
		1.3 Negotiating privacy	3
	2. Process of treatment	2.1 Steps to penetration	5
		2.2 Feeling supported	6
		2.3 Frustration at progress	5
	3. Suggestions for improvement	3.1 Easier way to navigate	3
		3.2 More information	3
		3.3 More clinician input	3
Change experienced through programme	4. Cognitive change	4.1 Awareness of self	6
		4.2 Normalising the problem	5
		4.3 Feeling more confident	6
	5. Behavioural	5.1 Approach from avoidance	6

	attributions to change	5.2 Trainers difficult but necessary	6

1. Practicality of use

Themes in this domain are focused on the practicalities of using the programme and relate primarily to interview questions on likeability, usability and comprehensibility (see Appendix 8).

1.1 Convenience

All six women reported that the programme was convenient to use because it was easy to access and/or it fitted flexibly into their routine. Ease of access often related to using the Prezi and completing the daily diary online. Fitting the programme into their routine was described as flexible because they could undertake programme activities at times that suited them and did not have to attend face-to-face appointments.

> *"I thought it was really easy. It didn't take much time. It kind of gave you no excuses to do it because it was so easy [laughs] to use and do it." (P5)*

> *"I think everything was relatively easy because I could use it on my phone on the go, so filling in the diary, if I'd forgotten to do it the night before I could do it on the train." (P6)*

> *I mean for me it would have been really difficult I mean almost impossible to do the programme if I had to come here [to hospital] every week, and I think that has delayed me to ask for help in the past" (P1)*

1.2 Understanding the content

All of the women commented on how they could understand or relate to the content of the Prezi. This was often in response to the interview question on comprehensibility, but it was also raised elsewhere. The women explained how they connected with the

stories, the style of written language, and more generally with the simplicity of the content.

> *"If someone is reading for the first time, especially a woman who is actually experiencing these kind of issues, it's like, you feel like it's helpful straight away it's not really anything complicated, the language is really simple." (P4)*

> *"I liked the stories, so it wasn't just the information that I had to consume, but it was something more personal, so I could relate to some of the stories as well… it made it much easier to make sense in my head, instead of just reading through information and trying to take it on board." (P2)*

> *"The Prezi, I think there is something a bit more grown-up about it and because it starts with explaining it, it straight away doesn't make you feel stupid or patronised which I think is important." (P5)*

1.3 Negotiating privacy

Only three women commented on issues of privacy, but it appeared to be an important issue for those who raised it. Opinions on privacy were somewhat divergent. P4 said she found it difficult to ensure the privacy of calls during work hours. P5 and P6 found the trainers difficult to conceal at home, but found other aspects of the programme, such as accessing it online and using anonymous codes enhanced a sense of privacy. Therefore, a mixed picture of privacy was found.

> *"Sometimes I end up standing on the street like in a quiet corner talking about inserting things and stuff and I really hope no-one can hear me." (P4)*

> *"I think they're [trainers] probably the hardest element because they're the least subtle, private, you know when you live in a house with other people" (P5)*

> *"Because there were codes it didn't really matter what I was writing down even if it was in public." (P6)*

2. Process of treatment

Themes in this domain concentrated on the experience of following the programme, including the stepped structure, support received along the way and a sense of progress throughout treatment. These themes were not clearly linked to specific questions, but tended to be discussed in the context of acceptability.

2.1 Steps to penetration

Five women reported that they liked the progressive nature of the programme. They liked being able to work through the initial phases (Steps 1-3) before the penetration phase, explaining that it gave them time to prepare and build up skills to help them to feel ready.

> *"I thought it was really well structured, so when you got to the point of starting to use the trainers, you had built very good foundations." (P1)*

> *"I liked the fact that it gave the whole thing, the reading about it, the understanding of how it worked, the relaxation was important, so it wasn't just focused on, you know the practical bit." (P3)*

> *"What I liked was the way it built it up in stages, that I did at first initially think it was going to jump straight into things like trainers and stuff like that so. It was nice that I had time to progress." (P6)*

2.2 Feeling supported

All of the women commented on the weekly telephone calls being supportive or helpful. They reported that it was important to speak to someone with knowledge about the problem and who could help guide them through the programme at the right pace. They also said it felt as if they were not alone. Several women explained that whilst the Prezi offered more general information, the calls provided individualised support.

"You really need someone there and someone who also knows, their expertise, they can tell you try this or that." (P1)

"I think that helped a lot, it did, just because you know someone is there with you. I know it's very personal problem, but you still need someone there." (P2)

"I think that the value of the phone call is that it is tailored around what you are experiencing, so obviously there are always going to be things that don't quite fit…so I think that it was really helpful to be able to talk to someone and say what was going on." (P3)

2.3 Frustration at progress

Five of the six women reported that they had sometimes felt frustrated at the amount of time it was taking to progress and/or the effort that was required to see an improvement (primarily in relation to trainer use). The women progressed at their own pace and were only advised not to move onto larger sizes if they were experiencing pain. The level of frustration appeared to be influenced by the amount of progress made, for example, P4 who showed the least improvement on penetration expressed the most amount frustration. Two women commented that when progress seemed slow, the diary helped them to reflect that overall progress was still being made.

"Because it's frustrating, you know you have to do it but sometimes I am so tired and sleepy, I am too tired for this today and sometimes it's just being tired of being frustrated, it's not very helpful." (P4)

"In the beginning I was a little bit inpatient to kind of, oh get on with this, what's happening? And actually there was a lot of work you needed to do emotionally, mentally, and also you know, like all the different stages of preparing," (P1)

"Sometimes, you know there would be moments where I was not succeeding as much as I thought I would be. I think that [diary] gave me the feeling that I was still making progress."(P3)

2. Suggestions for improvement

Women were asked about ways in which they might improve or change the programme. Common suggestions related to orienting the content of the Prezi, being given more information and having more clinician input.

2.1. Easier way to navigate

The Prezi software was not able to remember the last place the person viewed the programme. This meant that the women had to search through the content to find where they last were. Three women commented on this, and two suggested having software that could remember their last location or have bookmarks to help find the information more easily.

> *"If they were just hyperlinks you could just click and it would jump to like the second step. Because I think if I remember correctly, it went back to the start and you had to keep clicking." (P6)*

> *"If you have it in some kind of book format you can straight away find where you left it, you can go back to things, whereas with this one, or maybe I am just useless technologically, but I didn't know how to do it so I had to kind of going like de de de [clicking] until I found the right bit." (P1)*

2.2. *More information*

Three women wanted more information included in the Prezi. Two women had sought additional information on the Kegel exercises; one mentioned using another resource to help with orgasm.

> *"I think maybe once I looked something up elsewhere. I think it was the Kegel exercises I just wasn't, I think it is just because it is a hard thing to explain, but I was just trying to make sure I was doing it right." (P5)*

"If there's any way of making it more personal, so more stories that you could relate to, plus more information...there was a book I used for, to try and make myself orgasm". (P2)

2.3. More clinician input

Three women said that additional support could have been helpful in terms of the amount of clinical contact and the time necessary to complete the programme.

"I think if someone actually approaches you or seeks help and they are still at this early stage they will need a lot more psychological support to work through the preliminary issues." (P3)

"Maybe you can give some support in this area [feeling frustrated], because I found this the most difficult thing and the thing that, maybe something you could help a little bit more in this particular area." (P4)

3. Cognitive change

The themes in this domain relate to cognitive changes that women identified as a consequence of the programme. They were asked to identify changes that occurred and were prompted to comment on how they related to particular elements of the programme. It was interesting that women talked mostly about cognitive, rather than the behavioural change, the latter of which was captured by the quantitative outcomes.

3.1. Awareness of self

Five of the women commented on how they had become more aware of their reactions, both cognitively and physically. Women learnt to notice when they were tense or relaxed and this related to specific techniques in the programme (Kegels and relaxation). They also reported being more aware of thoughts linked to their fears of penetration. Some of them described how they would try to respond to these moments of awareness differently. P1 and P2 said that programme had helped them to notice difficulties they experienced with anxiety more generally.

"It [Kegels] just helps you realise how much in control you can be of those muscles. In terms of the other relaxation exercises, I didn't use those loads, but what I did learn was that in myself when was good to use them and when wasn't good to use them" (P5)

"It's naturally coming into my head, all of the bad thoughts 'it's going to hurt' or I naturally tense up. It is like a habit now. So the main thing for me is not just to relax but try and make positive thoughts out of negative." (P2)

"When I know we are about to have sex, I keep thinking in my head, kind of mentally freezes, like I don't know if this is going to hurt or not, so I keep having to tell myself in my head 'oh this is going to be fine, I can do this" (P6)

3.2. Normalising the problem

Five women said that the programme had made them view vaginismus as a common or more manageable problem. Expectations about the programme being helpful seemed to be low initially, with indications that vaginismus was seen to be an unsolvable rare problem. After the programme, the problem seemed to be viewed as less overwhelming and manageable.

"At the very beginning maybe I thought it was this like this terrible thing that no-one else in the world had, even if I knew. I think the programme has made me process more the fact that other people, maybe not many, but other people have the same problem." (P3)

"I honestly thought that something is wrong with me or the relationship… Where after reading through the programme, what vaginismus was, it made it sound much easier like it's a small problem, I think 3% of women have it in the world, so I am not the only one, it's treatable, and you don't need any medicine or anything" (P2)

"I didn't feel like I'm part of, like I'm different, I knew that inside. But now I'm like no, I'm just completely normal. So it's a huge thing for me" (P1)

3.3. Feeling more confident

When asked about changes they had noticed in themselves, all of the women said they felt more confident. Confidence often related to interactions in their relationships, such as, approaching sexual behaviours, and talking to partners about what they wanted sexually. Some women were also more optimistic about the future, for example, more confident with the idea of starting new relationships and being able to tackle the problem again if it returns.

> *"It gave me some confidence back that, I can do that…I didn't really have much problem with them so it was kind of like this is something I can do and I will be able to do bigger sizes at some point. Definitely motivated me and gave me confidence that I can do that at all." (P4)*

> *"I think it's also made me more confident in being able to talk with a partner about things, things that are causing me pain, things that I don't like or just anything sexual. Because before I couldn't really and I think this has encouraged me to do that." (P6)*

> *"Now I see people and I think, if I like them, oh it could be a relationship…whereas before I would probably get a bit more nervous if I like someone, because I would think if we get together I would have a problem" (P1)*

4. Behavioural attributions to change

The programme was reported to decrease avoidance and increase approach behaviours; this is a change in itself, but was also attributed as the cause of the cognitive changes outlined above. Trainer use was identified as the most difficult aspect of the programme, but one of the main contributors to change.

4.1. *Approach from avoidance*

All six women commented on how the programme had encouraged them to approach the problem, whereas before they had been more inclined to avoid doing something about it. This is partly inherent in the programme, as it asks women to undertake exercises and face the problem. Approach behaviours were often related to the

clinical support and monitoring (diary and calls), which seemed to motivate the women to engage with the programme. One woman (P4) said although she had used the trainers more, she had become more avoidant of sexual activities with partner, because she wanted to be comfortable using the trainers first.

> *"It helped me to motivate myself, because sometimes when you have a problem you try to avoid to handle it, even though it's very important and you convince yourself that you need to go through this, sometimes you want to just not face it." (P2)*

> *"If I was on my own, I could do that on my own probably, but it would be, I just like, sometimes you give up." (P4)*

> *"Knowing that I had to write something down and that somebody would see it, kind of motivated me to do something during that day, because otherwise I would have probably been lazy." (P6)*

4.2. Trainers difficult but necessary

All of the women reported that the trainers had been a main reason for change in the programme. This may be expected, because this is the stage where they directly faced the feared situation. The women described how they had realised trainers of increasing size could be inserted without pain. They also identified this as the most difficult aspect of the programme, but one that had to be undertaken in order to progress. The women often reported that they were willing to tolerate some discomfort when using the trainers in order to reach their goals.

> *"Maybe for me the biggest change was like trying these dilators, because it like as I said, you can talk about it, you can read about it but at the end of the day that's not what is going to help." (P4)*

> *"The trainers, although I think they took a bit of getting used to, which I had the opportunity to do before, I think there's no other way around that and actually, I think they're probably the hardest element." (P5)*

"It was obviously for me the most ground-breaking one was the trainers, because I thought 'oh my God something can kind of come in there!" (P1)

DISCUSSION

The effectiveness and acceptability of a new computerised guided self-help programme for vaginismus was evaluated on six women. A multiple case design was used to assess quantitative change over time, in addition to pre- and post- treatment outcome measures. Qualitative interviews were used to evaluate the programme's acceptability and change experienced as a result of the programme.

Summary of quantitative results

Pain-related fear was observed to reduce over the course of the programme. A reliable improvement was found from baseline to end of treatment in all women, and a large effect size was comparable to previous research (ter Kuile et al., 2013). Changes in reported fear tended to be associated with exposure activity; a ≥50% reduction in pain-related fear occurred in all women after commencement of the exposure phase and strong negative correlations between fear and penetration suggest that these factors were associated.

Successful penetration attempts were observed to increase over the course of the programme. A reliable improvement from baseline to the end of treatment was found in five women, and a large effect size was found in line with previous evidence (van Lankveld et al., 2006). Approximately three weeks into the programme, all of the women embarked on exposure using trainers, giving an indication of the time needed to prepare for exposure. By the end of treatment, three women had successful intercourse; one without a partner was successfully using her vibrator; and two had not yet attempted intercourse. Pain on penetration was observed to reduce with successive penetration attempts; but, for most women, pain was low in intensity from the initial attempts, suggesting that tension was already minimised.

No clear patterns of change were found on outcomes of generalised anxiety or relationship satisfaction.

Summary of qualitative results

Practically, participants found the programme easy to access, use and understand. There was inconsistency regarding privacy; some found it enhanced a sense of privacy (e.g. having information online), whereas others mentioned difficulties concealing trainers and phone conversations.

The process of treatment was experienced as helpful in terms of its progressive nature (building up skills in preparation for exposure) and the support received along the way. The clinical contact and monitoring of activity was experienced as having a motivational influence. There was some experience of progress being slower than expected, accompanied by feelings of frustration.

In terms of the change identified, women became more aware of tension in their bodies and were able to identify negative thoughts attached to sexual activity. Their view of vaginismus seemed to change from being an untreatable problem to a common and manageable problem. Confidence was reported to improve in relation to sexual functioning and interpersonal behaviours more broadly. Behavioural change inherent in the programme were also identified as helpful; generally, moving from avoidance to approaching the problem, and specifically, the use of the trainers.

To help conceptualise a more inclusive picture of the computerised vaginismus programme, a model has been developed (Figure 10) that includes both quantitative and qualitative findings. This is a hypothetical conceptual model and would need to be developed through further research. It includes the practical aspects of treatment that were found to be acceptable and which could influence initial and continued engagement with the programme. It identifies ongoing processes that might occur throughout treatment and which are likely to require the input of a clinician. Finally, it

includes changes that may be expected as the programme is followed. These changes are not intended to reflect a sequential order, but highlight possible experiences along the journey of treatment.

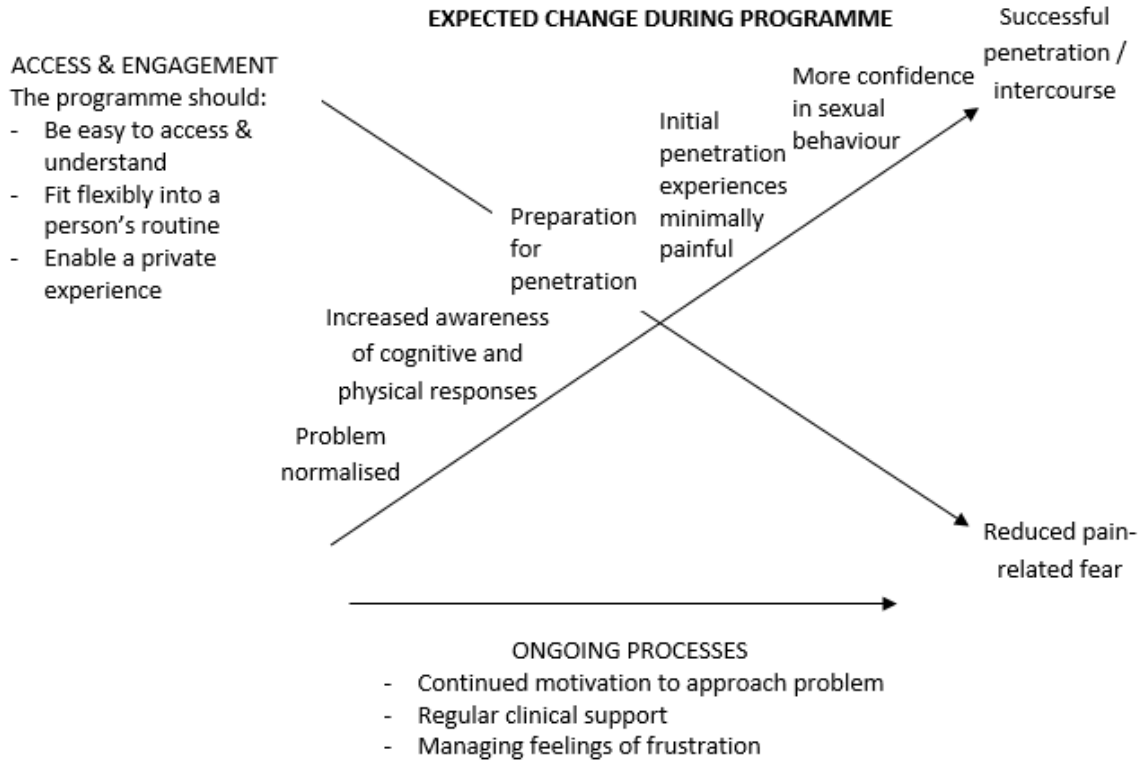

Figure 10: Model of vaginismus programme

Theoretical explanations

Quantitative and qualitative findings suggested that pain-related fear reduced in conjunction with successful penetration experiences. This is consistent with behavioural models of vaginismus such as Wijma and Wijma's (1997) model outlined in the introduction. Negative reinforcement (from avoidance) is removed as gradual exposure provides evidence of non-threatening outcomes. With repetition, the association between the conditioned stimulus (fearful anticipation of penetration) and the conditioned response (muscle tension/pain) is weakened. This model implies that pain-related fear reduces subsequent to exposure, which seemed to be the case in this study. However, for a minority of the women, pain-related fear started to reduce before exposure. This could relate to the pre-exposure phases of treatment and is

supported by the ‘steps to penetration’ theme, in which women liked the fact they have time to learn skills in preparation for exposure. Such preparation is used when treating other anxiety-based disorders using systematic desensitisation, equipping people with relaxation skills first to minimise fear during exposure (Wolpe, 1968).

Penetration activities were inherent in the programme, but that did not guarantee their success. For example, three of the women had previously been recommended to use trainers or a vibrator without regular clinician input (see Table 4), but had only been partially successful in using these (this statement is supported by high pre-treatment pain scores and the fact they were still seeking help). Therefore, the programme seemed to offer something additional that encouraged the women to engage in penetration activities more frequently and with more success.

One explanation that relates to the ‘feeling supported’ theme from the qualitative analysis is that the clinical input received throughout the programme motivated the women to engage with the exercises. Research suggests that having some clinical contact (not necessarily face-to-face) makes a difference when compared with no contact at all. This has not been evidenced for vaginismus, but in an RCT of a computerised intervention for depression, weekly email contact from a therapist led to greater improvement in symptoms (ES= 1.14), compared with no additional guidance (ES=0.66; Berger, Hämmerli, Gubser, Andersson, & Caspar, 2011). Even without clinical input the effect is large, and it would be interesting to see whether the vaginismus programme could be effective without any support.

Another explanation for the success of penetration could be the frequency of activities undertaken. The programme asked participants to engage in regular practice, and the theme ‘approach from avoidance’ demonstrated that women had previously not been attending to the problem. A behavioural approach would acknowledge that regular exposure is vital for the association between fear and pain to be weakened. Frequency of practice could also relate to the support received,

without which women may have been inclined to slip back into cycles of avoidance, particularly as most women reported that trainers were initially uncomfortable. This is analogous to treatment for other anxiety disorders that requires people to endure low levels of distress in order obtain disconfirmatory evidence of the feared outcome.

Whilst some pain was experienced, scores tended to be low even on initial penetration attempts. This could be explained by the graded behavioural approach; women only moved onto larger trainers when they had used previous sizes with no pain or minimal discomfort (Abramowitz, Deacon & Whiteside, 2012). If pain was intolerable, women would be at risk of disengaging with the trainers (positive reinforcement from success needed to outweigh negative reinforcement from pain avoidance). Even when pain scores were low, a change of a little as 2 points on an 11-point pain scale has been found to be clinically meaningful (Farrar, Young, LaMoreaux, Werth & Poole, 2001); so what may appear to be a small change could be important to the patient.

The finding that the programme was convenient (easy to access and fit into routine) highlights the importance of instant and flexible access to resources. This could reflect the wider culture of internet-based help-seeking and fits with findings from a national survey that found the internet was used to access help with health concerns, because it was an easy and fast way to get information (Ybarra & Sunman, 2006). It seems a sensible option to provide information online, leaving clinical time for personalised support the internet cannot offer. Whilst the programme fostered convenience, privacy seemed harder to achieve. Face-to-face contact in hospital settings enables privacy and confidentiality; in this programme, ensuring the privacy of phone calls was the responsibility of the patient. In addition, whilst the programme itself was all online (enhancing privacy), the trainers were necessary material objects that jeopardised privacy. Difficulties concealing trainers would be the same in routine clinical practice; it may be possible to exchange exposure using trainers for fingers

(Mousavinasab & Farnoosh, 2003). Three participants suggested that they would like to be able to navigate the online content more easily and have additional information included in the programme; these suggestions could be integrated into future versions without difficulty. Three women also commented on wanting more clinical input. This is less easily changed, particularly if the intervention is found to be clinically and cost effective with minimal contact (research implications discussed below).

Wider context

It is useful to view these findings within the wider context of computerised programmes for physical and mental health. There has been an expansion of systematic reviews examining the effectiveness of internet-based interventions; many of which have found equal effects between online and face-to-face interventions for chronic pain (Cuipers, van Straten & Andersson, 2008), sexual health knowledge (Bailey et al., 2010), and anxiety and depression (Andrews et al., 2010). So far, internet-based options seem a promising alternative to face-to-face treatment. Others have evaluated individual components of successful internet-based interventions for health; a meta-analysis revealed that the inclusion of more behaviour change techniques increased effectiveness (Webb, Joseph, Yardley & Michie, 2010). Behaviour change techniques were purposefully integrated into the current programme (see interventions section), providing a measurable way to compare it to a wide range of other behavioural interventions (Abraham & Michie, 2008). With a future of internet-based healthcare ahead, it is vital to understand the psychological components of change as well as the size of clinical effects.

Strengths

Use of single case methodology enabled an in-depth evaluation of a novel intervention, supported by qualitative findings. Multiple repeated outcomes allowed for the examination of subtle processes of change through components of treatment, which are not usually detectable using pre-post analyses. This methodology may

seem far-removed from the gold standard randomised designs; but an increasing amount of research is drawing on single case methodology and attempting to integrate randomisation methods to enhance credibility (Kratochwill & Levin, 2010).

Limitations

The multiple case design used could have been improved. Firstly, the baseline phase helps to determine change at the introduction of an intervention. This study had a mean baseline of eight days, which is disproportionate to the intervention duration and may not have been long enough for scores to stabilise. Increasing the baseline duration would allow for stabilisation and strengthen conclusions about causality. Secondly, a follow-up period was not included, which is important for evaluating the longer-term impact of the programme. Ter Kuile et al., (2009) used a similar single-case A-B design and had a follow-up period of 12 weeks, in which successful penetration continued to increase post-treatment. Follow-up data would have been particularly useful to review the progress of P4 & P5 who continued to use the trainers and work towards intercourse. Finally, as the programme was designed to be flexible, half of the women chose not to do Step 3, body awareness. This diminished the opportunity to observe patterns across all phases of the programme.

Participants were asked to rate pain on scale of 0-10 in order to obtain regular data without burdening them with lengthy questionnaires. This was helpful in terms of data gathering, but the reliability of the scores was more problematic. Pre-treatment pain ratings were estimates of penetration pain, which for some women were not recent experiences (ranging from 3 days to 8 months). The test-retest reliability coefficient taken from ter Kuile et al., (2007) had a stability period of three weeks, which means four of the six women in this study fall outside of that window for their baseline pain ratings. Moreover, pain scores were compared across activities; it could be argued that, for example, whilst a vibrator is similar in size with a penis, emotional factors

associated with intercourse makes these activities impossible to compare. The PEQ was also limited in its measurement, assessing penetration over a four week period and potentially missing subtle changes in behaviour. Adapting the PEQ to measure a two week period could have helped to detect more discrete change and allowed for a better observation of penetration in line with the individual's activity throughout the programme.

Outcomes in this study may have been biased by researcher allegiance, which occurs when a researcher holds a preference for a particular treatment (Leykin & DeRubeis, 2009). Researcher allegiance is associated with factors such as having authorship of the intervention and believing a certain therapeutic approach is superior to others (Gaffan, Tsaousis, & Kemp-Wheeler, 1995). Bias occurs when allegiance influences the outcomes of research. It is difficult to know whether this study was biased by allegiance, but as the author and sole delivering clinician of the intervention, it is possible the findings would have been different if the programme was delivered by someone less invested in the research. This posed a particular risk during the interviews, as participants were being asked by the delivering clinician about their experiences. This is likely to have influenced their expression of negative experiences; having an independent researcher would have helped to minimise bias of favourable outcomes in the qualitative data.

Whilst this research did not explicitly exclude women with complex mental health difficulties or abusive histories, it is unknown whether such participants would benefit from the programme. For example, women who have experienced abuse may need more time to reflect on its impact and women in non-consensual relationships would be prioritised for help with domestic violence. Adaptions could be made for more complex presentations, such as face-to-face sessions in addition to programme access, but this would need to be supported by research.

Research and clinical implications

It is not the intention of a multiple case design to generalise to the target population; however, with data from only 6 women, the wider effectiveness of the vaginismus programme is unknown. A larger controlled trial could provide a more substantial effectiveness evaluation. It would be helpful for the trial to include several arms as follows: 1) guided computerised self-help, 2) unguided computerised self-help, 3) guided bibliotherapy, and 4) unguided bibliotherapy. These arms would help to control for the mode of delivery (computerised versus paper information), as well as the level of clinical input (with or without guidance). It might be expected that more clinical input improves efficacy, in which case it would also be interesting to include a face-to-face arm, to assess the quality of the interaction. If more clinical input is found to increase efficacy, then a cost-effectiveness analysis could help to determine whether additional resource is worth the change in treatment outcome. It might also be hypothesised that a computerised mode would be more accessible, but it is difficult to know whether this would impact on the overall efficacy of the intervention.

It would be helpful to use additional quantitative outcomes to evaluate change identified by the current study's qualitative findings. For example, a measure of confidence and other behavioural changes (such as interactions within the relationship) could help to assess the wider impact of the intervention. With a larger dataset it would be interesting to consider whether relationship satisfaction mediates the effectiveness of the intervention. The programme is very much focused on the woman holding responsibility for change, but sexual dysfunction is rarely detached from interpersonal issues. Therefore, the development of a programme which emphasises systemic couples work could be another avenue of research. The programme was also focused on penetration; opening the content up to address sexual wellbeing more generally, including aspects such as desire, arousal and orgasm, could provide a more holistic intervention. A measure of sexual function that

considers all of these factors, such as the Female Sexual Function Index (Rosen et al., 2000), could be used to assess effectiveness.

If the computerised programme for vaginismus is clinically comparable (in terms of the size of effect achieved) and more economical (resource required) with face-to-face interventions, women could be offered the choice between routine care and the computer programme, or possibly a mix of the two. The programme could help to save resources, because it is paperless, and time is saved due to outcomes being collected online and the intervention being conducted using brief telephone contact. Waiting-lists could be reduced, as clinicians could make three contacts per hour instead of one. The flexibility of the programme could also help more women to access treatment for vaginismus, in particular for women for whom time is a practical barrier to help-seeking or for those who are embarrassed to speak face-to-face about sexual issues. As we move towards a future of online healthcare, validating and implementing such interventions in real clinical settings should be a priority.

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Part 3: Critical Appraisal

Psychotherapy research today is scrutinised on numerous indicators of quality and bias. These indicators have tended to focus on design methodology, but biases can also arise from interpersonal factors, such as the allegiance of the researcher. Allegiance occurs when the researcher believes that active components and effects of one treatment surpass those of another. This belief can lead to a bias in research if 'investigators' allegiances are responsible for the advantages found for their preferred treatments' (Leykin & DeRubeis, 2009, p56). Links between allegiance and outcome have been demonstrated empirically, though the mechanisms behind these links are less clear. In this critical appraisal, I will reflect on my own allegiance to my research on vaginismus and try to establish potential biases that occurred.

Before highlighting potential problems with allegiance, it should be noted that within the history of psychology, allegiance may have been necessary for the development of theory and intervention. One could argue that paradigm shifts were borne out of allegiance; tension between theories and theorists helped to drive research forward. Dissatisfaction with one psychological approach led to the breakdown and displacement of allegiance. For example, in Miller's reflections on the cognitive revolution, he reports, 'When I became dissatisfied at Harvard between B.F. Skinner's strict behaviorism and S.S. Stevens' psychophysics, I turned to Jerry Bruner's social psychology, and in 1960 that led to the creation at Harvard of the Center for Cognitive Studies.' (Miller, 2003, p142). It seems impossible to have no allegiance in such a diverse field of science; however, it should be recognised as a potential biasing factor in research.

In a review of reviews, 30 meta-analyses comprising 1248 primary studies on psychological treatment were examined for allegiance effects (Munder, Brütsch, Leonhart, Gerger & Barth, 2013). Seven percent of the variance in outcome was explained by research allegiance (r=0.26). This association was maintained when controlling for treatment type, patient population and setting. Interestingly, the

review also examined allegiance of the authors of the included meta-analyses. Strong allegiance to the concept of research allegiance was found to moderate the outcomes of primary studies (r=0.39). Therefore, even when trying to evaluate allegiance impartially, researchers are still at risk of holding influential biases.

Quality measures have been developed to try to capture levels of allegiance. I came across these when undertaking my literature review, in which I chose to implement a measure developed by Gaffan, Tsaousis, & Kemp-Wheeler (1995). It attempts to assess several domains of allegiance, including: citing previous research that supports a particular treatment; discussing the superiority of a particular treatment; including a lengthy description of a particular treatment; having authorship of a treatment; and researching only one active treatment condition. When applying this quality measure to my own research I discovered that I met criteria for bias on all domains! This was somewhat of a surprise as it suggested that my research was high risk in terms of allegiance bias. Recognition of this encouraged me to reflect on the domains of allegiance in relation to my research.

Authorship

As there were no available computerised self-help programmes for vaginismus I set about writing the programme content, drawing on the available literature. The theoretical concepts were grounded in evidence, but the content and visual presentation were influenced by my own writing style, creative vision and understanding of theory. For example, the programme had fictional vignettes and hand-drawn anatomical diagrams, the inclusion of which enhanced the sense of ownership and personalisation. Other individualised components of the programme, such as the use of online resources (links to websites, audio files), also contributed to a feeling that the programme was in some way unique. After the programme content was drafted it was sent to specialists for comment and revised accordingly. This process helped me to feel more confident about the quality of the product and

the desire to claim ownership of it. Taking these reflections into account, there is no doubt that I held a strong allegiance to the programme, but in what ways could authorship have biased my research?

It has been suggested that the delivery of treatment may differ depending on whether the delivering clinician is also the author (McLeod, 2009). Being familiar with the content might improve the quality of delivery compared to clinicians who may be less knowledgeable about the specific intervention. Could authoring the content have improved my delivery of the programme or could a more experienced psychologist have delivered the programme effectively without knowing the content in its entirety? Knowing the content is surely not enough to deliver it in an optimal therapeutic way; this would undermine a fundamental premise of clinical psychology, which is the therapeutic relationship between clinician and patient. But being familiar with the content could indeed help the patient access the right information and increase competence in the delivering clinician. It has been argued that to know whether or not a treatment is effective, it should be implemented by 'people who know what they are doing', together with independent monitoring of the treatment (Hollon, 1999, p110). This argument fits with what is expected from real clinical practice; trained clinicians delivering specialist interventions with supervision. Without formal evaluation, it is difficult to know how well I delivered the vaginismus programme and whether this was influenced by allegiance.

Hollon (1999) commented that authorship of treatment in itself does not necessarily map on to one's clinical work. For example, a doctor may be interested in developing CBT-based treatment for pain, but clinically works in a medical framework. Vaginismus is commonly treated in genitourinary medicine clinic; a multidisciplinary setting with primary diagnostic input received from medical professionals. My external supervisor was a gynaecologist and there were times in

the research when I asked him whether the source of a participant's pain could be better explained by an underlying chronic pain condition, such as provoked vestibulodynia. For other participants I also wondered whether longer term psychological help in the form of couple's therapy (as opposed to CBT model) could be more helpful. Researching in an MDT setting and keeping an open mind to the biological factors and alternative therapeutic models was a useful way to inhibit strong allegiance effects.

Being the author of the vaginismus programme certainly influenced my desire for the intervention to work effectively and be acceptable to patients. I was able to identify clues to my own authorship bias when listening to the clinical interviews at the end of treatment. Participants did not know that I had written the programme, although some of my responses to their experiences were noticeably confirming. For example, I observed that in several of my responses I said 'that's good', or 'I'm glad'. Even though I was consciously trying to be impartial, I had built up therapeutic rapport with the participants over the course of the programme and it felt invalidating not to comment on their progress. Therefore, being both the author of the programme and the interviewer could have certainly created a bias.

Superiority

Prior to this research, I worked in a sexual health setting and implemented CBT-informed treatment for sexual dysfunction; so it was my opinion that this was the most appropriate form of help for vaginismus. Undertaking my literature review confirmed this belief, as it was the most commonly evaluated treatment (Melnik, Hawton & McGuire, 2012). Moreover, it was the only model in which I was trained to manage sexual dysfunction; other less commonly used approaches such as hypnotherapy or psychodynamic psychotherapy were outside my therapeutic repertoire.

The belief that one treatment is superior to another has been called the 'Mecca effect' (Shaw, 1999). This is not to say that one treatment is always seen to be the best, but one modality may be seen as superior for a certain client group, such as dialectical behaviour therapy for borderline personality disorder or CBT for worry. Often, clinicians will have a preferred modality of treatment and it is reasonable to assume that the Mecca effect and allegiance are strongly connected. What is less clear is whether this effect always leads to biased outcomes.

In my study, it could be hypothesised that the outcomes were more successful than if I had attempted to deliver a psychodynamic-informed approach, but this would be attributable to my own limitations as a psychologist rather than the influence of allegiance. Research has shown that behavioural activation delivered by generic mental health staff with minimal training can result in good clinical outcomes in depression (Ekers, Richards, McMillan, Bland & Gilbody, 2011). It would be interesting to know whether similar outcomes could be obtained if the vaginismus programme was delivered by staff who did not view CBT as superior. This could help to alleviate allegiance effects and minimise bias. Bearing in mind that the telephone calls drew on fundamental therapeutic techniques (not just problem-solving), the capacity to do 'therapy' could also influence the effectiveness of the intervention. In any clinical setting, patients and staff would expect treatment to be delivered by a professional who is skilled in that area, so why not in research too? Shaw (1999) argued that if outcomes are improved by allegiance, then it should be fostered to achieve better results for all patients. The practicalities of fostering allegiance, however, are difficult to imagine, and the notion of brainwashing may come to mind! But if a measure of individual allegiance (including beliefs about superiority) was developed, it could be a useful starting point to examine allegiance effects in practice. So far, allegiance indicators have been applied to entire studies, rather than to individual researchers or clinicians.

Investment in outcome

Investment in outcome was not defined as a separate item in Gaffan et al.'s (1995) measure, but I think it is closely linked to authorship and superiority beliefs. We know from the evidence that allegiance is positively associated with outcome, but the mechanism behind this is less well understood. It has been suggested that if someone is invested in a particular outcome, s/he will draw on the knowledge s/he has to fit with the desired result (Markman & Hirt, 2002). Applying this concept to my research, when a participant was finding it difficult to insert one of the trainers, I would draw on all the behavioural techniques I had used previously to help them achieve that outcome. But would I have been so invested in helping them achieve penetration behaviour if it was not one of my main outcome measures? If my primary outcome was relationship satisfaction, maybe I would have focused on the dynamics between the couple and drawn more on systemic theory instead. The primary outcomes were selected in accordance to the CBT model in use, suggesting influences of treatment superiority from the outset.

The process of setting up and seeing research through from beginning to end is often costly; whether it is time, effort, financial or emotional cost. For me, I think the emotional cost influenced my sense of allegiance most. Feelings of fear and frustration often accompanied the research process, and the idea that the programme would fail evoked such emotions. Without this emotional investment, would I have been so attentive to the success of the treatment? This is likely to reflect the feelings of other researchers, who have a strong desire certain for outcomes. This is represented more generally in publication bias; neutral and negative outcomes are far less likely to be published, indicating that findings do not represent the desires of researchers. Interestingly, experimental research has examined desirability in relation to games, and a quote from one article summed up

my experience well: ‘knowing that one has a lot of money riding on the outcome of a game might cause greater attention to evidence supporting the desirable outcome, but might also cause restraint of one’s stated optimism as a way of protecting oneself from disappointment’ (p113, Krizan & Windschitl, 2007). Although I did not have real money riding on the outcome, I felt that I paid more attention to the evidence that supported a successful outcome, rather than contemplating that the treatment might be unsuccessful for some people. I also found that I was on occasion telling myself ‘it doesn’t matter’ about the findings as long as I had my data, which could have been my attempt to protect myself from disappointment. Of all the aspects of allegiance, I think for me, emotional investment could be the most powerful driver of bias.

Sole active condition

Another indicator of research allegiance identified by Gaffan et al., (1995) was the inclusion of a sole active treatment condition. This criterion may be more applicable to larger RCTs where it is common to have at least one active treatment group. In my research project it would have been difficult to arrange a second treatment condition. However, the fact I had not thought to compare the programme with another condition or even a control made me wonder about the influence of allegiance. On reflection, I could have collected pre- and post- treatment outcomes from women who opted for of routine care (face-to face treatment) or from those who were waiting to be assessed.

Citing supporting research

Undertaking a systematic review on psychological treatments for vaginismus helped me to identify treatment modalities other than CBT (in RCTs only), such as hypnotherapy. But I realised the good quality evidence for alternative treatments was thin. I realised that I had not considered non-RCT research into alternative

treatments which led me to run a quick search of non-CBT, non-RCT research for vaginismus. I discovered empirical investigations and reviews from a range of theoretical backgrounds, including psychodynamic, humanistic, and behavioural/biofeedback (Barnes, Bowman & Cullen, 1984; Hiller, 1996; Kleinplatz, 1998). However, this literature tended to be small-scale, outdated, and had not been replicated or supported by more robust evidence. Without the CBT literature, few good studies remained. Therefore, on this domain of allegiance bias, I would argue that citing evidence to support a CBT approach was a true reflection of the evidence-base, rather than a biased inclusion of specific research. Re-visiting my superiority beliefs, it seems in the case of vaginismus, CBT may be viewed as 'Mecca' for a reason.

Implications and recommendations

Although allegiance and attempts to moderate its effects are starting to be evaluated as indicators of quality research, as it stands, most researchers still consciously or unconsciously bias their findings by holding an allegiance to certain treatments. Without guidance or recommendations to inform researchers about how bias can be minimised, they will continue to be at risk of influencing outcomes. Reflecting on my own research allegiance has impacted on the way that I would approach research in the future. I have developed several recommendations:

1. It would be advantageous to develop a clinician's manual for the vaginismus programme. This would diminish effects of authorship and help to standardise treatment. It would also allow for adherence checks, which would be useful for monitoring differences in the quality of delivery.
2. Use of a manual would not be enough to eliminate allegiance effects; for example, a clinician with a pre-existing preference for CBT might be more prone to allegiance bias than a clinician who has a pre-existing preference for systemic therapy. It could be helpful if researchers (as with qualitative indicators of credibility) state their

position on the research in hand, for example, what psychological models they practice in and whether they expect certain results. A measurement of individual clinician preferences could be useful to understand effectiveness research in relation to allegiance; but to my knowledge, there is no such validated questionnaire in psychological research.

3. When developing and evaluating a treatment programme, it would be desirable for the author not to deliver it directly to patients. This is already an established methodological criterion in RCTs, in which studies that use independent researchers and attempt to mask investigators are seen to be more robust. Training clinicians to deliver the programme could still pose an allegiance bias, but less so than the author or researchers themselves. The aforementioned manual would allow the programme to be delivered by independent professionals (e.g. nurses, gynaecologists, psychologists), who would be less invested in the programme's success. This also applies to outcome collection (questionnaires and interviews), which should be obtained independently where possible.
4. It would be useful to compare the programme to another treatment condition to see whether CBT is more effective than other approaches. This could help challenge allegiance to CBT. As the evidence for vaginismus is limited largely to behavioural and cognitive approaches it would be difficult to create distinct treatment conditions, particularly because treatments such as hypnotherapy and psychodynamic approaches may not be amenable to a computerised format. But different components could be evaluated, for example, one condition could be purely behavioural (exposure), one could be cognitive (thought-focused), and another could be CBT (integration of techniques). This could help to demonstrate differences in treatment effectiveness and help to identify mechanisms of change.
5. It would be helpful for a reliable and valid measure to be developed, that could quantify and help to predict levels of bias in response to allegiance variables. For example, a quasi-randomised trial may be classified as high risk on selection bias

and the results of the research would be treated with caution. So what if a study was at high risk of allegiance bias? Would we treat it with the same level of caution and interpret the results accordingly? Allegiance information could be corroborated with other biasing factors to get a more complete assessment of risk. Further research in this area could help to clarify these questions.

Final thoughts

This reflection has led me to identify several improvements that could help to reduce allegiance bias. But to reduce is not to eliminate, and allegiance will always be inescapable to some extent. Allegiance in psychological treatment is entwined with the therapeutic relationship; so removing allegiance bias is more complex and could be argued to be unethical. I believe allegiance can be advantageous in guiding the progression of psychological research, which is often driven by positive experiences of implementing treatments with patients. I would not have been driven to develop and evaluate a CBT-based programme for vaginismus if I had not experienced the successful implementation of this approach beforehand. If we want to encourage psychologists to undertake research alongside their clinical work, the likelihood is they will evaluate treatments that they feel enthusiastic about.

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Appendix 1: Literature review search strings

OVID (Psychinfo, EMBASE & Medline)

1. vaginismus or dyspareunia or superficial dyspareunia or vestibulodynia or vulv* vestibulitis or vulvodynia
2. provoked adj3 (vulvodynia or vestibulodynia)
3. localised adj3 (vulvodynia or vestibulodynia)
4. sexual dysfunction and vagin* and pain*
5. sexual dysfunction and female and pain*
6. 1-5
7. random or randomi*ation or randomi*ed controlled trial or control* trial or controlled clinical trial or random* adj 2 (trial or allocate* or assign* or sampl*)
8. clinical adj2 trial
9. double blind or single blind
10. 7-10
11. 6 and 10

CINHAL

1. CINAHL; (vaginismus OR dyspareunia OR superficial AND dyspareunia OR vestibulodynia OR vulv* AND vestibulitis OR vulvodynia)
2. CINAHL; (provoked AND vulvodynia OR provoked AND vestibulodynia OR localised AND vulvodynia OR localised AND vestibulodynia)
3. CINAHL; (sexual AND dysfunction AND vagin* AND pain)
4. CINAHL; (sexual AND dysfunction AND female AND pain*)
5. CINAHL; 11 OR 12 OR 13 OR 14
6. CINAHL; (random OR randomi*ation OR randomi*ed AND controlled AND trial OR controlled AND clinical AND trial)
7. CINAHL; (random* AND trial OR random* AND allocate* OR random* AND assign* OR random* AND sampl*)
8. CINAHL; (clinical AND trial OR double AND blind OR single AND blind)
9. CINAHL; 16 OR 17 OR 18
10. 20. CINAHL; 15 AND 19

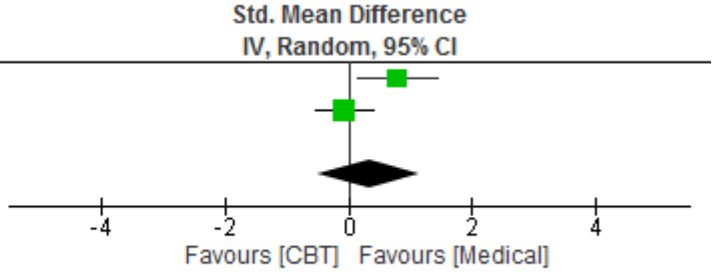
Appendix 2: Literature review forest plots

Analysis 1.1
Comparison: CBT versus medical treatment
Outcome: General pain post-treatment

Analysis 1.2
Comparison: CBT versus medical treatment
Outcome: pain on intercourse post-treatment

	CBT			Medical Treatment				Std. Mean Difference
Study or Subgroup	Mean	SD	Total	Mean	SD	Total	Weight	IV, Random, 95% CI
BERGERON2001	6	2.13	27	3.93	3.25	15	46.4%	0.79 [0.13, 1.44]
DESROCHERS2010	5.46	2.75	39	5.67	3.32	30	53.6%	-0.07 [-0.55, 0.41]
Total (95% CI)			**66**			**45**	**100.0%**	**0.33 [-0.51, 1.17]**

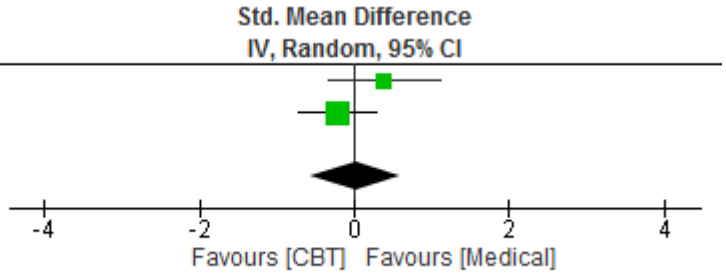
Heterogeneity: Tau² = 0.28; Chi² = 4.30, df = 1 (P = 0.04); I² = 77%
Test for overall effect: Z = 0.77 (P = 0.44)



Analysis 1.3
Comparison: CBT versus medical treatment
Outcome: pain on intercourse 6 months follow-up

Study or Subgroup	CBT Mean	CBT SD	CBT Total	Medical Treatment Mean	Medical Treatment SD	Medical Treatment Total	Weight	Std. Mean Difference IV, Random, 95% CI
BERGERON2001	4.46	2.47	27	3.41	3.17	10	39.7%	0.39 [-0.35, 1.12]
DESROCHERS2010	5.21	2.87	35	5.87	3.07	29	60.3%	-0.22 [-0.71, 0.27]
Total (95% CI)			**62**			**39**	**100.0%**	**0.02 [-0.56, 0.60]**

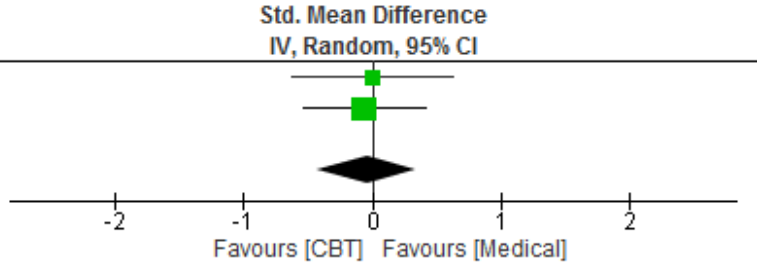
Heterogeneity: Tau² = 0.08; Chi² = 1.81, df = 1 (P = 0.18); I² = 45%
Test for overall effect: Z = 0.07 (P = 0.95)



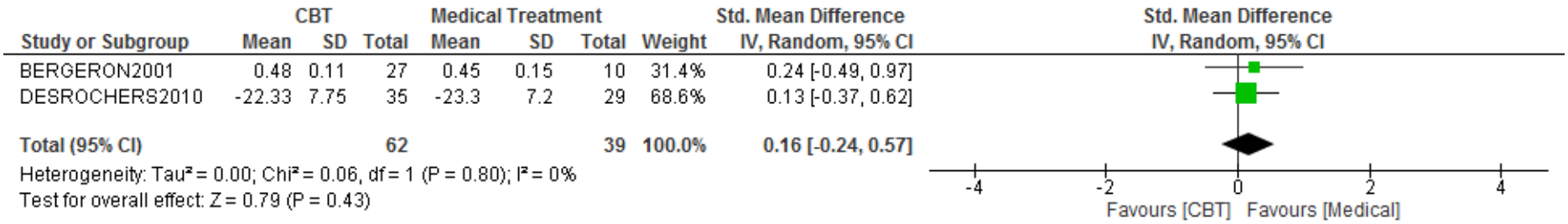
Analysis 1.4
Comparison: CBT versus medical treatment
Outcome: sexual functioning post-treatment

Study or Subgroup	CBT Mean	CBT SD	CBT Total	Medical Treatment Mean	Medical Treatment SD	Medical Treatment Total	Weight	Std. Mean Difference IV, Random, 95% CI
BERGERON2001	0.49	0.12	27	0.49	0.14	15	36.3%	0.00 [-0.63, 0.63]
DESROCHERS2010	-23.03	7.59	39	-22.53	7.63	30	63.7%	-0.06 [-0.54, 0.41]
Total (95% CI)			**66**			**45**	**100.0%**	**-0.04 [-0.42, 0.34]**

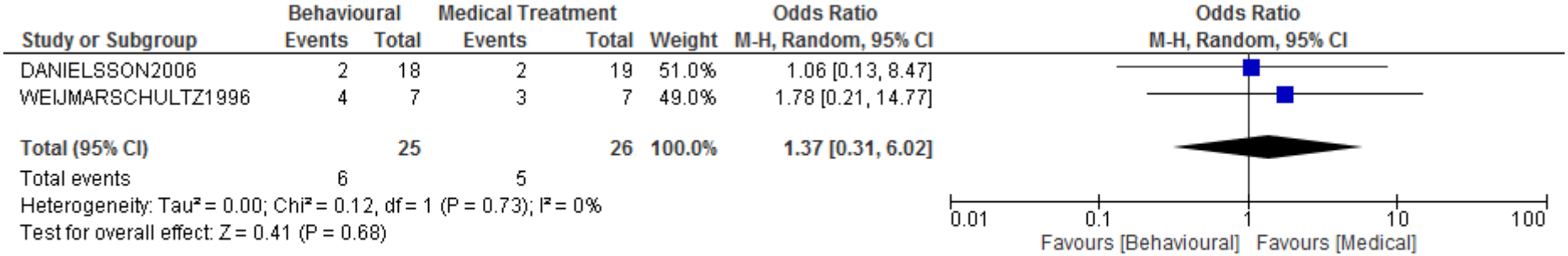
Heterogeneity: Tau² = 0.00; Chi² = 0.03, df = 1 (P = 0.87); I² = 0%
Test for overall effect: Z = 0.21 (P = 0.83)



Analysis 1.5
Comparison: CBT versus medical treatment
Outcome: sexual functioning 6 month follow-up



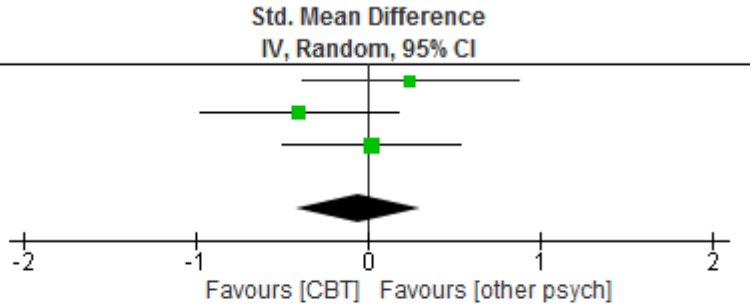
Analysis 2.1
Comparison: Behavioural versus medical treatment
Outcome: symptom elimination follow-up



Analysis 3.1
Comparison: CBT versus other psychological treatment
Outcome: pain on intercourse post-treatment

Study or Subgroup	CBT Mean	CBT SD	CBT Total	Other psych Mean	Other psych SD	Other psych Total	Weight	Std. Mean Difference IV, Random, 95% CI
BERGERON2001	27.75	15.09	27	23.79	17.23	15	27.7%	0.24 [-0.39, 0.88]
MASHEB2009	-8.1	6	23	-5.6	6.2	25	33.0%	-0.40 [-0.98, 0.17]
VANLANKVELD2006	-2.19	1.58	31	-2.22	1.76	27	39.3%	0.02 [-0.50, 0.53]
Total (95% CI)			**81**			**67**	**100.0%**	**-0.06 [-0.41, 0.30]**

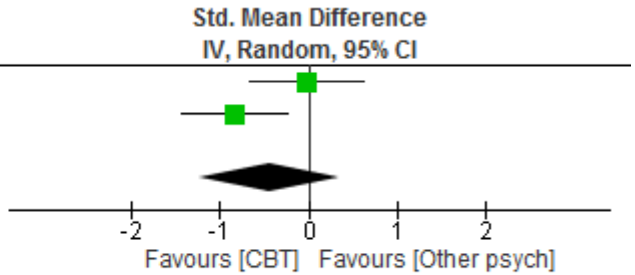
Heterogeneity: Tau² = 0.02; Chi² = 2.35, df = 2 (P = 0.31); I² = 15%
Test for overall effect: Z = 0.32 (P = 0.75)



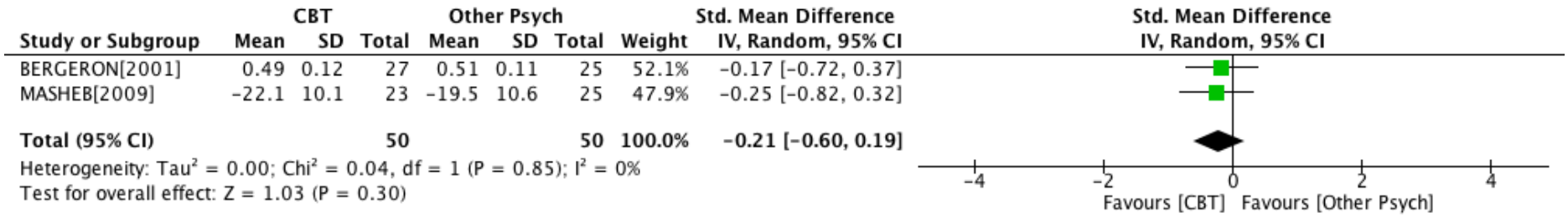
Analysis 3.2
Comparison: CBT versus other psychological treatment
Outcome: pain on intercourse follow-up

Study or Subgroup	CBT Mean	CBT SD	CBT Total	Other psych Mean	Other psych SD	Other psych Total	Weight	Std. Mean Difference IV, Random, 95% CI
BERGERON2001	19.96	15.6	19	20.32	18.31	17	48.6%	-0.02 [-0.68, 0.63]
MASHEB2009	-8.1	1.3	22	-7	1.3	25	51.4%	-0.83 [-1.43, -0.23]
Total (95% CI)			**41**			**42**	**100.0%**	**-0.44 [-1.23, 0.36]**

Heterogeneity: Tau² = 0.23; Chi² = 3.21, df = 1 (P = 0.07); I² = 69%
Test for overall effect: Z = 1.08 (P = 0.28)



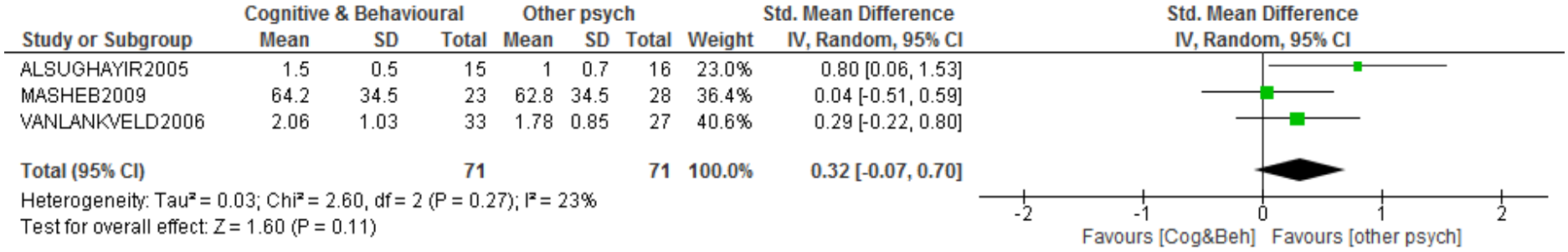
Analysis 3.3
Comparison: CBT versus other psychological treatment
Outcome: sexual functioning post-treatment



Analysis 3.4

Comparison: Cognitive & Behavioural versus other psychological treatment

Outcome: sexual anxiety post-treatment

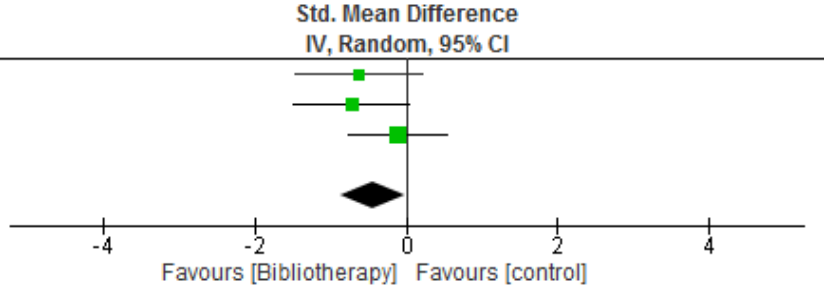


Analysis 4.1

Comparison: Bibliotherapy versus control
Outcome: sexual frequency post-treatment

Study or Subgroup	Bibliotherapy Mean	SD	Total	Waitlist Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
VANLANKVELD2001	7	1.84	9	7.96	1.24	16	25.8%	-0.63 [-1.47, 0.21]
VANLANKVELD2001	6.62	1.76	13	7.73	1.22	15	30.6%	-0.72 [-1.49, 0.05]
VANLANKVELD2006	5.33	3.36	27	5.71	2.84	14	43.5%	-0.12 [-0.76, 0.53]
Total (95% CI)			**49**			**45**	**100.0%**	**-0.43 [-0.86, -0.01]**

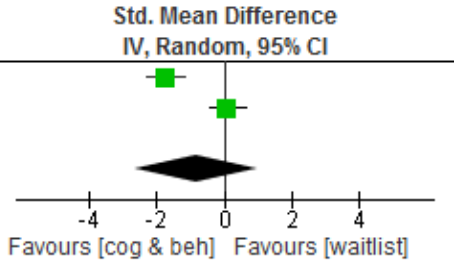
Heterogeneity: Tau² = 0.00; Chi² = 1.67, df = 2 (P = 0.43); I² = 0%
Test for overall effect: Z = 2.00 (P = 0.05)



Analysis 4.2
Comparison: CBT and behavioural treatment versus waitlist
Outcome: Pain on intercourse

Study or Subgroup	Cognitive & Behavioural treatment Mean	SD	Total	Waitlist Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
TERKUILE2013	-3.34	1.8	35	-0.67	1.18	35	49.9%	-1.73 [-2.29, -1.18]
VANLANKVELD2006	-2.19	1.58	31	-2.35	1.64	23	50.1%	0.10 [-0.44, 0.64]
Total (95% CI)			**66**			**58**	**100.0%**	**-0.82 [-2.61, 0.98]**

Heterogeneity: Tau² = 1.60; Chi² = 21.57, df = 1 (P < 0.00001); I² = 95%
Test for overall effect: Z = 0.89 (P = 0.37)



Analysis 4.3

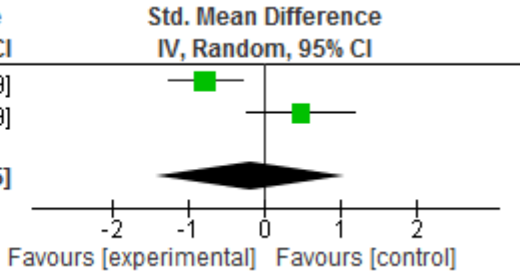
Comparison: CBT and behavioural treatment versus waitlist

Outcome: Fear of intercourse

	Cog & Beh treatment			Waitlist Control				Std. Mean Difference
Study or Subgroup	Mean	SD	Total	Mean	SD	Total	Weight	IV, Random, 95% CI
TERKUILE2013	2.2	0.92	35	3.01	1.12	35	52.2%	-0.78 [-1.27, -0.29]
VANLANKVELD2006	2.06	1.03	33	1.6	0.52	10	47.8%	0.48 [-0.24, 1.19]
Total (95% CI)			**68**			**45**	**100.0%**	**-0.18 [-1.41, 1.05]**

Heterogeneity: Tau² = 0.70; Chi² = 8.15, df = 1 (P = 0.004); I² = 88%

Test for overall effect: Z = 0.28 (P = 0.78)



Appendix 3: Empirical study Patient Information Sheet

Imperial College Healthcare NHS Trust

[redacted]

Participant information and consent sheet

A computerised self-help guide for women with vaginismus

(Students research project)

You are being invited to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information. Ask us if anything is unclear or if you would like more information. Take time to decide whether or not you want to take part.

What is the purpose of the study?
The purpose of this study is to see whether a computerised self-help programme can help women with vaginismus.

Vaginismus is associated with vaginal pain and difficulties with penetration, which can be a very distressing problem. Estimates suggest that approximately 1% of women in the general population have this condition. Treatment for vaginismus is usually delivered face-to-face by a sexual health specialist. At the moment, there is no computer-based version of treatment, but there is an increasing demand for help to be available online. Therefore, this study is going to examine whether a computerised self-help programme can be a helpful way to treat vaginismus.

Why have I been chosen?
Women who have been given a diagnosis of vaginismus will be asked whether or not they want to take part in the study. This is because the programme has been designed specifically to help women experiencing this problem. We are aiming to get about 10 women to take part in this research.

Do I have to take part?
It is up to you whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard care you receive.

What will happen to me if I take part?
If you decide to take part you will meet with the researcher who will explain how the computer programme works and what you need to do. A week after this meeting, you will be given access to the programme and you work through it at home. You will have weekly telephone contacts (10-15 minutes) with the researcher to help guide you through the programme which is estimated to last 6-8 weeks.

The computerised programme will involve reading information and following instructions. There will be a number of things you will need to do including:

reading about vaginal pain; learning relaxation techniques and exercises for muscle control; exploring your body; working towards penetration by containing trainers (smooth plastic objects of different sizes) in your vagina whilst feeling relaxed. The exercises will take different amounts of time to complete; some will take 5 minutes and others 30 minutes. However, it is up to you how much you do each day.

You will be asked to complete a daily diary to monitor what activities you are doing each day and your experiences of fear and pain (if any). This will also be completed online and should not take more than a minute per day. There will also be some short questionnaires that you will be asked to complete before and after the programme.

At the end it would be helpful to hear about your experiences of using the programme and so the researcher will ask you some questions about what you found helpful or not so helpful over the telephone. This should take about 20-30 minutes. This will be recorded and transcribed after which the recording will be deleted.

Travel expenses will not be reimbursed for journeys made to the clinic.

What are the alternative treatments?
You have the option to choose whether or not you want to take part in this study. If you do not want to then you can have routine treatment which involves the same exercises, but instead you will be meeting weekly face-to-face with a sexual health specialist.

What are the potential benefits?
Previous research has shown that similar programmes for women with vaginismus have helped them to achieve sexual intercourse. This study is hoping to produce similar outcomes, aiming to help women have vaginal penetration, reduce fear of vaginal penetration and reduce pain on penetration.

What if something goes wrong?
We do not anticipate any risk will come to the women who take part in this study.
As vaginismus is associated with pain on attempted penetration, it is likely that there will be some physical discomfort during the training phase of the programme. This is a normal part of treatment although the idea is to minimise your pain or discomfort as much as possible. This will be done by introducing the training element slowly and only when you have completed the previous steps and feel ready.

If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated by members of staff you may have experienced due to your participation in the research, National Health Service or UCL complaints mechanisms are available to you. Please ask your research doctor if you would like more information on this.

In the unlikely event that you are harmed by taking part in this study, compensation may be available.

If you suspect that the harm is the result of the Sponsor's (University College London) or the hospital's negligence then you may be able to claim compensation. After discussing with the researcher, please make the claim in writing to Amanda Williams who is the Chief Investigator for the research and is based at University College London. The Chief Investigator will then pass the claim to the Sponsor's Insurers, via the Sponsor's office. You may have to bear the costs of the legal action initially, and you should consult a lawyer about this.

What happens when the research study stops?
After the programme has finished, you will be given the choice of coming for a follow-up appointment with the researcher if you think the intervention has not been successful or if you need some additional support, to discuss your needs. A decision will then be made about how the genitourinary clinic can provide further support or whether a referral will be made to a more appropriate service. You will continue to have access to the computerised programme.

Will my taking part in this study be confidential?
In compliance with the Data Protection Act 1998, all information which is collected about you during the course of the research will be kept strictly confidential. Any information about you which leaves the hospital will have your name and address removed so that you cannot be recognised from it. Your GP will be notified of your participation in this study with your permission if you consent.

What will happen to the results of the research?
The results of the study will be retained and written up as part of Doctoral research conducted at University College London (UCL). The UCL Records Office maintains archived records in a safe and secure off site location. Access to stored records is strictly controlled. The results could also be published in a journal. Any publication will uphold confidentiality and anonymity.

Who is organising the research?
This study is being conducted as part of Doctoral research at University College London, with approval from Imperial Healthcare NHS Trust.

[redacted]
Main contacts for research
If you are interested in taking part in this research, you can:

- Contact the researcher (Esther Flanagan) directly via telephone or email (details below)
- Inform the healthcare worker you are currently seeing that you would like to take part in the study, and then the researcher will contact you to arrange a meeting.
- Or, contact Esther Flanagan or David Goldmeier to find out more about this research before you decide whether or not you want to take part.

Ms Esther Flanagan
Trainee Clinical Psychologist
[redacted]

Dr David Goldmeier
Consultant Gynaecologist

For independent advice and support you can contact Imperial College patient advice and liaison service (PALS) offers help, support, information and advice to patients and their relatives, friends and carers.
PALS
[redacted]

Appendix 4: Empirical paper consent form

Imperial College Healthcare NHS Trust

[redacted]

INFORMED CONSENT FORM
Title of Project: A computerised self-help guide for women with vaginismus
Principal Investigator: Amanda Williams

Please initial each box once you have read it.

I confirm that I have read and understood the subject information sheet dated for the above study and have had the opportunity to ask questions which have been answered fully.	
I understand that my participation is voluntary and I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected.	
I understand that sections of any of my medical notes may be looked at by the researcher or regulatory authorities where it is relevant to my taking part in this research. I give permission for these individuals to access my records that are relevant to this research.	
I understand that the interview at the end of the programme will be recorded	
I agree to the referrer/GP being informed of my participation in the study	
I agree to take part in the above study.	

Name of Participant	Signature	Date
_____	_____	_____
Name of Person taking Consent	Signature	Date
_____	_____	_____
Name of Principal Investigator	Signature	Date

Appendix 5: Empirical paper screenshot of programme

Appendix 6: Empirical paper ethics approval letter

NHS
Health Research Authority

NRES Committee London - Central
Skipton House
80 London Road
London
SE1 6LH

Telephone: 020 797 22560

03 May 2013

Dr Amanda Williams
Reader in Clinical Health Psychology
UCL
1-19 Torrington Place London
WC1E 6BT

Dear Dr Williams

Study title:	**The effectiveness and acceptability of a computerised self-help guide for women with vaginismus**
REC reference:	**13/LO/0487**
IRAS project ID:	**112067**

The Research Ethics Committee reviewed the above application at the meeting held on 24 April 2013. Thank you for attending to discuss the application.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the Co-ordinator Julie Kidd, NRESCommittee.London-Central@nhs.net.

Ethical opinion

The Committee reviewed the above study. The Chief Investigator Dr Amanda Williams and Student Esther Flanagan attended to discuss the application.

In discussion, the Committee noted the following ethical issues.

- Current treatment is face to face or via new home computerised system.
- Members were content that the participants would be given the information sheet and a week to consider the study prior to giving consent.
- Members found the study to be generally well written except no risks had been included in the PIS, members felt that this would be especially important for new patients. That

said, the researcher had added risks in the IRAS form A22.

- The researcher will be advised to ensure that the clinic telephone number is included in the PIS and not her personal one.
- Members noticed that there was no information as to whether the product was a drug or a device.
- The PIS mentions similar programmes, and the Committee would be interested in seeing if the results of the similar programmes are the same.

Dr Amanda Williams and Ms Esther Flanagan joined the meeting. Discussion took place as follows:-

a. The Chair asked if the research would change standard of care if participants opt for the computerised system. Miss Flanagan assured the Committee that all participants would get support and the reason for the computerised system is so that participants do not have to take time off work; the level of contact is the same but the new version would be via the telephone.
b. The Chair asked if participants opt for face to face would they also get the demonstration of the computerised programme and any medical devices to be used. Miss Flanagan said all information is available for all participants, currently all devices are used without specific demonstration at home anyway.
c. The Chair pointed out to Miss Flanagan that no risks had been included in the PIS, and asked her to add them if necessary.
d. The Chair asked Miss Flanagan to explain the programme. Miss Flanagan explained that the system used a programme called PREZI, which is an interactive programme that includes diagrams whereby participants follow a path and can move both back and forth and if necessary seek help on the phone.
e. The Chair asked Miss Flanagan to ensure that the clinic number was on the PIS rather than her own. Miss Flanagan agreed to remove her personal number.
f. The Chair asked why an online gmail account had been set up for participants and asked if it was secure. Miss Flanagan said that it was a good way to collect data and the format would be the same for all. Miss Flanagan said no names would be entered only codes for names and data.
g. The Chair asked what other programmes similar to hers where available. Miss Flanagan said there was a programme similar in the Netherlands but no computerised guide was available only protocols of face to face, with no self-help forum.
h. Miss Flanagan confirmed that only researchers had access to PREZI.
i. The Chair asked Miss Flanagan to ensure that participants get at least a week before giving consent.

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non NHS sites

The Committee has not yet been notified of the outcome of any site-specific assessment (SSA) for the non-NHS research site(s) taking part in this study. The favourable opinion does not therefore apply to any non-NHS site at present. I will write to you again as soon as one Research Ethics Committee has notified the outcome of a SSA. In the meantime no study procedures should be initiated at non-NHS sites.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

It is responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

Document	*Version*	*Date*
Advertisement	1	08 March 2013
Evidence of insurance or indemnity	B1262F10103012	30 July 2012
Interview Schedules/Topic Guides	1	08 March 2013
Investigator CV: Amanda Williams		10 January 2013
Letter from Statistician	1	31 January 2013
Other: Student CV: Esther Flanagan		11 January 2013
Other: Data Protection		10 January 2013
Participant Consent Form	1	08 March 2013
Participant Information Sheet	1	08 March 2013
Protocol	1	08 March 2013
Questionnaire: Validated Questionnaire	1	08 March 2013
REC application	112067/424769/1/994	11 March 2013
Summary/Synopsis	1	08 March 2013

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Frances Goodhart declared interest.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

13/LO/0487	Please quote this number on all correspondence

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr John Keen
Chair

Email: NRESCommittee.London-Central@nhs.net

Enclosures: *List of names and professions of members who were present at the meeting and those who submitted written comments*
"After ethical review – guidance for researchers"

Copy to: *Dr Clara Kalu*
Lucy Parker, Imperial College Healthcare

Appendix 7: Empirical paper Primary Endpoint Questionnaire

1	In the last 4 weeks, have you had sexual intercourse (full vaginal penetration by a penis) • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
2	In the last 4 weeks, have you inserted a finger into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
3	In the last 4 weeks, have you inserted two fingers into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
4	In the last 4 weeks, has your partner inserted their finger into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
5	In the last 4 weeks, has your partner inserted two fingers into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
6	In the last 4 weeks, have you inserted any other object into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
7	In the last 4 weeks, has your partner inserted any object into your vagina? • Not attempted • Attempted, but not successful • Attempted, and sometimes successful • Attempted, and always successful
	End of the questionnaire. Thank you for completing the questionnaire.

Appendix 8: Empirical paper interview schedule

Questions 2-4 aim to elicit information on acceptability. Questions 1, 5-9 aimed to elicit information on change and are derived from Elliott, Slatick & Urman's (2001) Client Change Interview.

1. **What was your overall experience of using the computer programme?**

2. **Likeability**
   What did you like or dislike about the programme? Please give examples. For example, what kinds of things were appealing and helpful or what kind of things were hindering, unhelpful, negative or disappointing?

3. **Usability**
   How easy or difficult was it to use the programme? For example, accessing and filling out information online, finding way around the content.

4. **Comprehensibility**
   How easy or difficult was it to understand the programme? For example, understanding the language used in the programme, following the instructions.

5. **Were there things in the therapy which were difficult or painful but still OK or perhaps helpful? What were they?**

6. **What changes, if any, have you noticed in yourself since the programme started?** (For example, are you doing, feeling, or thinking differently from the way you did before? What specific ideas, if any, have you gotten from therapy so far, including ideas about yourself or other people? Have any changes been brought to your attention by other people?) Prompt for both positive and negative changes.

7. **Is there anything that you wanted to change that hasn't since therapy started?**

8. **Attributions**
   In general, what do you think has caused these various changes? In other words, what do you think might have brought them about? (Including things both outside of therapy and in therapy) Prompt for specific parts of the programme.

9. **Suggestions**
   Do you have any suggestions for us, regarding the research or the therapy? Do you have anything else that you want to tell me?

Appendix 9: Empirical paper qualitative annotation