

REVIEW PAPER

Psychological Treatment for Vaginal Pain: Does Etiology Matter? A Systematic Review and Meta-Analysis

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ABSTRACT

Introduction. Classification of vaginal pain within medical or psychiatric diagnostic systems draws mainly on the presumed presence or absence (respectively) of underlying medical etiology. A focus on the experience of pain, rather than etiology, emphasizes common ground in the aims of treatment to improve pain and sexual, emotional, and cognitive experience. Thus, exploring how vaginal pain conditions with varying etiology respond to psychological treatment could cast light on the extent to which they are the same or distinct.

Aim. To examine the combined and relative efficacy of psychological treatments for vaginal pain conditions.

Methods. A systematic search of EMBASE, MEDLINE, PsycINFO, and CINAHL was undertaken. Eleven randomized controlled trials were entered into a meta-analysis, and standardized mean differences and odds ratios were calculated. Effect sizes for individual psychological trial arms were also calculated.

Main Outcome Measures. Main outcome measures were pain and sexual function.

Results. Equivalent effects were found for psychological and medical treatments. Effect sizes for psychological treatment arms were comparable across vaginal pain conditions.

Conclusions. Effectiveness was equivalent regardless of presumed medical or psychiatric etiology, indicating that presumed etiology may not be helpful in selecting treatment. Research recommendations and clinical implications are discussed. **Flanagan E, Herron KA, O'Driscoll C, and Williams AC de C. Psychological treatment for vaginal pain: Does etiology matter? A systematic review and meta-analysis. J Sex Med 2015;12:3–16.**

Key Words. Vaginismus; Dyspareunia; Vestibulodynia; Vulvodynia; Sexual Dysfunction; Psychotherapy

Introduction

This review examines three disorders under the collective heading of vaginal pain: vulvodynia, vaginismus, and dyspareunia. While the basis for distinction has been challenged because of shared psychological and physiological symptomatology [1,2], etiological factors are still used to differentiate diagnoses. This review attempts to combine data from treatment trials for vaginal pain and assess whether outcomes of psychological treatment differ according to etiological distinctions.

Vulvodynia—chronic pain in the vulval region—can be generalized or localized, provoked by contact or unprovoked. Combinations of these subtypes exist, and multiple terminologies are current. For instance, vestibulodynia is a term used to describe vulvodynia localized to the vestibule. It is unclear exactly what processes underlie vulvodynia, although physiological etiology is to a degree assumed [3,4]. Several theories have been proposed, including changes in sensitivity of the peripheral nervous system [5,6]. Diagnosis is made on the basis of pain on contact, tenderness to local pressure, and vestibular erythema. For the purpose

of this review, the term “medically defined” refers to disorders that are presumed to be medical in their etiology. While no particular psychological characteristics are required for a diagnosis, unsurprisingly, vaginal pain impacts various aspects of sexual desire and performance [7,8].

Dyspareunia (painful intercourse) is often diagnosed in conjunction with provoked vulvodynia and sometimes these terms are used interchangeably [2]. However, dyspareunia is also defined in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM). In the Fourth Edition, Text Revision (DSM-IV-TR [9]; replaced in 2013 by DSM-5, but current for this review), sexual dysfunction, defined as interference with sexual responsiveness or pleasure that causes marked distress or interpersonal difficulty, included two painful conditions: (i) dyspareunia, defined after exclusion of other medical causes as “recurrent or persistent genital pain associated with sexual intercourse”; and (ii) vaginismus, defined as “recurrent or persistent involuntary spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse”. The distinguishing criterion between dyspareunia and vaginismus was vaginal muscle spasm, the certainty of which has been disputed [1,10,11]. For the purpose of this review, the term “psychiatrically defined” refers to vaginismus and dyspareunia where they are defined as primarily psychiatric in their etiology.

In the recently introduced DSM-5 classification, vaginismus and dyspareunia are classified together under the broader label of “genito-pelvic pain/penetration disorder” (GPPPD) [12]. Four criteria are assessed separately: (i) persistent or recurrent difficulties in vaginal penetration during intercourse; (ii) marked vulvovaginal or pelvic pain during intercourse or penetration attempts; (iii) marked fear of or anxiety about vulvovaginal or pelvic pain in anticipation of, during, or as a result of penetration; and (iv) marked tensing or tightening of the pelvic floor muscles during attempted penetration. Any one criterion is sufficient for diagnosis: for example, some women experience pain but still manage penetration, whereas others cannot manage penetration because of actual or anticipated pain. These four domains do not rely so heavily on the assessment of etiological factors (such as a spasm); instead the focus is on symptomatology and impact on functioning. Thus, provoked vulvodynia effectively falls under GPPPD by virtue of involving pain on touch, likely to be aggravated during sexual penetration, in the absence of a

known medical cause, with the risk of leading to a marked fear of sexual activity [13].

Studies that have examined differences between vaginismus and dyspareunia have shown inconclusive results. A review of electromyogram (EMG) studies concluded that muscular responses in vaginismus could not be accurately differentiated from those in dyspareunia and vulvodynia [1]. A small, possibly underpowered study found no difference in ease of penetration (by a finger), muscle tension, redness, or pain during intercourse [14]. Another study reported greater muscle tension and more frequent vaginal spasms on gynecological examination in vaginismus than in dyspareunia (from vestibulodynia), but still in less than one-third of the women with vaginismus [15]. Interestingly, fear and avoidance behaviors were frequently reported, which are characteristic of chronic pain [16]. Seventy-three percent of the vaginismus group refused EMG sessions (none in dyspareunia and control groups) and were found more difficult to examine by gynecologists. This corresponds with psychological correlates of vaginismus, such as increased catastrophic thinking about pain and feelings of disgust [17,18]. Catastrophic thinking, not reflected in the DSM definition, is consistent with chronic pain presentations.

Chronic pain is increasingly characterized as a disorder with common biological and psychological features regardless of pain location [6]. Melzack and Wall’s [19] pioneering pain-gate model, now universally accepted, was the first to integrate neuronal response with noxious sensory input and “descending influences” representing cognitive and emotional processes (e.g., attention, mood, and memory). Changes in the nervous system, both centrally and peripherally, include the amplification or suppression of neuronal response and a failure to activate descending inhibition. Known as sensitization, these changes have been identified in vulvodynia, and rather less reliably in vaginismus [20–22]. Basson’s [23] model of provoked vestibulodynia proposed that pain generates sexual dysfunction even when a physiological cause such as central sensitization can be identified and that premonitory psychological factors including anxiety, depression, harm-avoidance, and vigilance to somatic experience exacerbate pain and adversely affect sexual function. The problem is then maintained by acquired risk factors, including beliefs of sexual inadequacy and diminished sexual motivation. Similar top-down influences, such as heightened harm-avoidance, have been found in women with vaginismus, and it could be conceived that

such tendencies may have preceded the development of vaginismus [17]. Conceptualizing pain using a stress-vulnerability model emphasizes analogous characteristics of the vaginal pain types in terms of response and maintenance, rather than etiology.

An important psychological model of pain is that of fear and avoidance, whereby fearful cognitive and emotional responses, including vigilance to pain and other somatic sensations, generate avoidance of activities [16,24]. Behavioral avoidance diminishes opportunities to test expectations of anticipated pain, exacerbating fears, lowering mood, and in turn increasing emotional content of pain processing [25–28]. Cycles of fear and avoidance apply to vaginal pain as well, with no reason to expect differences between vaginismus, dyspareunia, and vestibulodynia [29]. This model of pain can be used to conceptualize the four criteria of GPPPD, where attempted penetration causes distress, avoidance, and muscle tension (as part of a protective reaction to pain), which produce further avoidance and fear. Could standard psychologically based treatment for pain [30] therefore work as well for each of the disorders subsumed under GPPPD?

Treatment for vaginal pain has largely corresponded with identification of the pain as being of medical and psychiatric etiology. Treatment for vaginismus has primarily been psychological and aimed at reestablishing comfortable penetration. A systematic review showed a predominance of systematic desensitization and cognitive-behavioral therapy (CBT), but some local medical interventions for pain in vaginismus, such as botox and bupivacaine injections, have been trialed [31,32]. Treatment for dyspareunia has primarily been medical, possibly due to its overlap with provoked vulvodynia, with a focus on pain reduction to restore normal sexual function [1]. Treatment for vulvodynia has also been primarily medical, usually in the form of pharmacotherapy, surgery, or physiotherapy [33]. Promising effects have been shown for psychological treatments for vulvodynia, including exposure, CBT, and mindfulness, which have also been shown to be effective in other types of chronic pain [30,34–36]. Few randomized trials have evaluated a multidisciplinary treatment approach to vaginal pain.

Aims

The combined efficacy of psychological treatment for vaginal pain conditions (vulvodynia, vaginismus, and dyspareunia) has not been systematically examined, other than by one review that found CBT to be effective in improving sexual function [37]. We aim to broaden the evaluation of combined efficacy by including all psychological treatments with statistical analyses of the data. This review also aims to compare efficacy of psychological treatment according to presumed etiology to add to the evidence with regard to whether the disorders should be distinguished or combined.

Methods

A search of peer-reviewed journals was undertaken using EMBASE, MEDLINE, PsycINFO, and CINAHL, from the earliest date to April 2014. The reference lists of relevant systematic reviews were also searched. The following terms, derived from diagnostic criteria and previous systematic reviews, were entered in combination into full text searches: *vaginismus*, *(superficial) dyspareunia*, *sexual dysfunction and pain*, *(provoked, localised) vulvodynia*, *vestibulodynia*, *vestibulitis*, and variations of *randomised controlled trial* (see Supporting Information Appendix S1 for full search strategy). Two authors independently sorted all studies from the searches using titles and abstracts; discrepancies in decisions were discussed and agreed upon.

Methods

Inclusion Criteria

To be included in the review, studies had to involve women over 16 years with vaginismus, dyspareunia, or vulvodynia; involve evaluation of at least one psychological treatment (defined as a psychological rationale for cognitive, emotional, or behavioral change); be randomized or quasi-randomized; and use the outcome measures of sexual functioning (behavioral and cognitive measures) and pain.

Inclusion Criteria

Exclusion Criteria

Studies were excluded from the review if they involved women with known primary diagnoses, such as endometriosis, sexually transmitted infections, cancer, inflammatory problems, dermatoses, or menopause; involved women with deep dyspareunia or chronic pelvic pain; or were published in languages other than English.

Exclusion Criteria

Main Outcome Measures

The main outcome measures used in this review were measures of pain and sexual function. These were selected on the basis of their shared use

across studies and relevance to the aims of the review. Pain is a shared feature of vaginismus, dyspareunia, and vulvodynia, despite etiological assumptions, and different disorders might show similar or differential responses to psychological treatment. Sexual dysfunction defines vaginismus and dyspareunia and is a common secondary diagnosis in vulvodynia; therefore, it could also be used to determine whether these disorders showed comparable or differential responses to psychological treatment.

Data Extraction and Analysis

For continuous data, means, standard deviations, and sample sizes were extracted post-treatment and at follow-up. For categorical data, event-based outcomes were used. Authors were contacted where data were not available from the published article. All data were analyzed using Review Manager version 5.2 [38]. Standardized mean differences were calculated for continuous data and odds ratios for event-based outcomes, both using random-effects meta-analyses. Change from baseline to post-treatment was also calculated for psychological treatment arms. Thresholds for size of effect were >0.1 (small), >0.3 (moderate), and >0.5 (large) [39].

Possible bias was estimated using a methodology that samples selection bias (randomization method, allocation concealment, and comparability of groups at baseline), performance bias (blinding and equivalent care), attrition bias (dropout), and detection bias (reliability of outcomes) [40]. In psychological treatments, blinding of patients is rare and that of therapists even rarer, so the use of blind assessment of patients was used as a quality marker. Ratings were made by two authors independently and compared. Quality ratings can be found in Table 1 (full quality assessment supplied in Supporting Information Appendix S2).

Results

Included Studies

A total of 1,548 studies were retrieved from initial electronic and reference searches after de-duplication (see Figure 1 for PRISMA diagram). The 1,517 studies that were excluded from the initial screen either failed to meet the population criteria (e.g., chronic pelvic pain, endometriosis, cancer) or were nonrandomized trials or reviews. Twenty studies were read in full and excluded for the following reasons: 13 had no psy-

chological treatment arm [52–64] and/or were found not to be randomized clinical trials [65–67]; two were unavailable in English [68,69]; two [70,71] included reproduced data from other included studies [45,50] and were used to supplement information but not duplicated. The update searches retrieved 288 studies, of which one met inclusion criteria, giving a total of 12 studies.

Meta-Analyses

Of the 12 studies that met inclusion criteria, one did not include sufficient data in the original publication, and the author no longer had the data [47]. Data from 11 studies were included in the meta-analysis: six presented sufficient data in original publications [35,41,44,46,48], one of which was a follow-up study [42]; two included limited categorical data, which allowed for the use of odds ratios [45,51]; and three were supplemented by descriptive statistics [43] and raw datasets [49,50] from authors.

Eleven studies, of which 10 provided posttreatment data for 417 women and four provided follow-up data for a maximum of 180 women, were entered into the meta-analysis. Participants had a mean age of 30. Four studies examined vaginismus; one study examined vaginismus and dyspareunia, five vestibulodynia, and two a mix of provoked and generalized vulvodynia. One study had three active treatment arms, one had two active treatment arms and a control arm, seven studies had two active treatment arms, and two studies had one active treatment arm and a control arm. Six studies evaluated CBT, six behavioral therapy, one hypnotherapy, and one supportive therapy. CBT and behavioral treatments were most often selected as the active treatment in analyses when compared with other psychological treatments. Pharmacotherapy was used for three control arms, and surgery for two. Three studies used a waiting list control.

Risk of Bias

Selection bias was rated as high in four studies, unclear in two, and low in five. Performance bias was rated high in two studies, unclear in eight, and low in one. Attrition bias was rated high in two studies, unclear in three, and low in six; detection bias was rated high in four studies, unclear in five, and low in two. The main reason for unclear ratings was lack of reported information or difficulty blinding treatment personnel or patients. See Table 1 for ratings of bias.

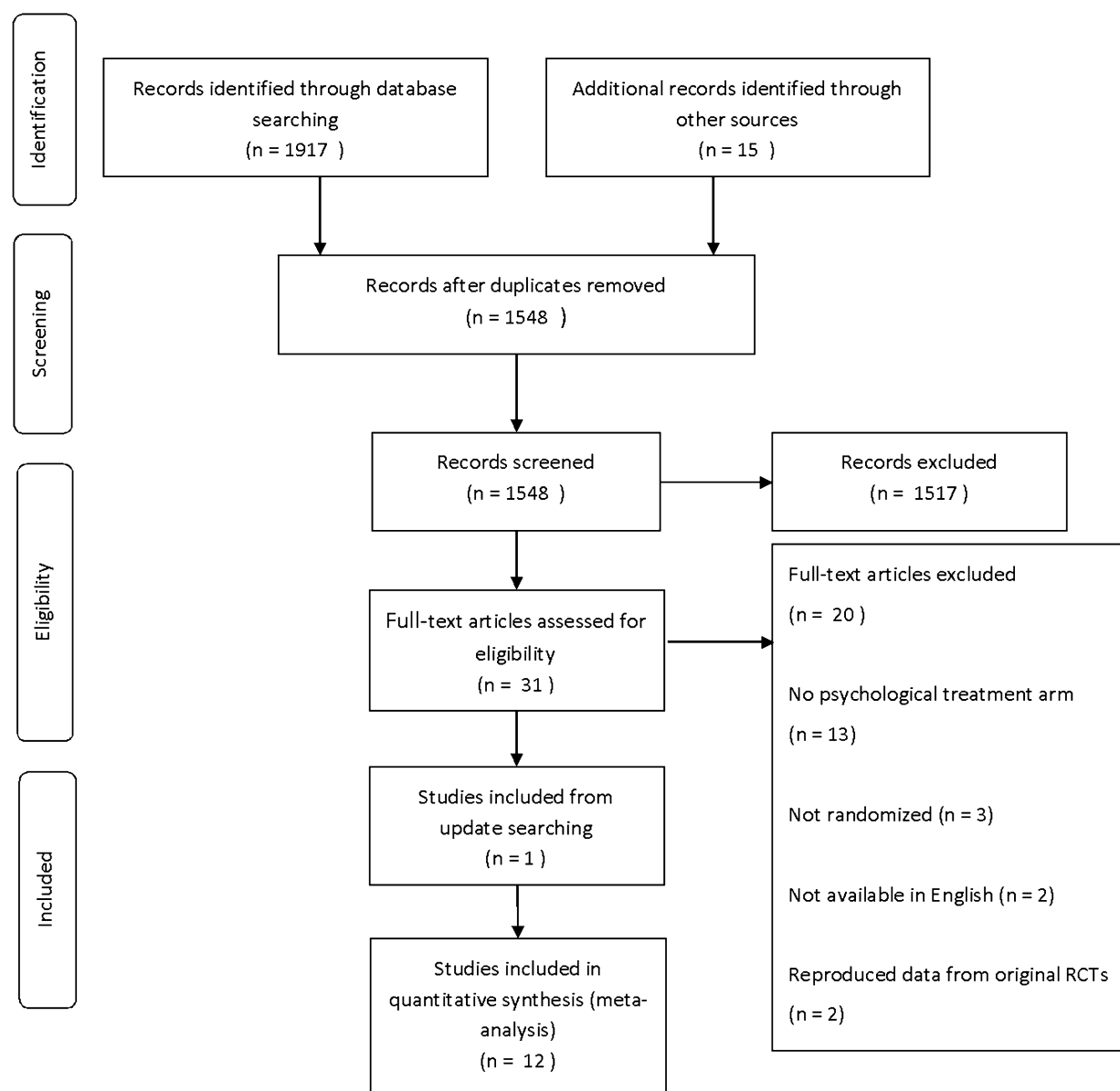


Figure 1 PRISMA flow diagram of study selection.

Treatment Effects from the Meta-Analysis

Heterogeneity was calculated using the I^2 statistic, using heterogeneity thresholds of <25% (low), 25–50% (moderate), and >50% (high). All results can be found in Table 2 (forest plots supplied in Supporting Information Appendix S3).

Psychological Versus Medical Treatment

Three studies of good quality ($n = 143$) compared CBT with medical treatment with regard to post-treatment outcomes of general pain (not limited to intercourse); no significant effect was found. Two studies compared CBT with medical treatment on

posttreatment and 6-month follow-up outcomes of pain with regard to intercourse; no significant effect was found for either. Two studies compared CBT with medical treatment with regard to post-treatment and 6-month follow-up outcomes of sexual functioning; no significant effect was found. Two studies of unclear and poor quality ($n = 46$) compared behavior therapy with medical treatment; posttreatment odds ratios of symptom elimination showed no significant effect. Overall, the lack of significant effects could suggest that psychological and medical treatments were equally effective or ineffective.

Table 1 Study characteristics for included studies

Study ID	Population	Treatment/comparator	Outcomes	Selection, performance, attrition, and detection bias
Al-Sughayir 2005 [41]	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. Behavior therapy (n = 18) Once weekly, 45–60 minutes, both wife and husband (mean sessions = 10)	<ul style="list-style-type: none"> Sex-related anxiety (5-point scale) Wife's sexual satisfaction (5-point scale) Husband's sexual satisfaction (5-point scale) 	High High Unclear High
Bergeron et al. 2001 [35] (Bergeron et al. 2008 [42], 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 gynecologist 2. Biofeedback (n = 28) Self-insertion of electromyographic sensor into vagina; 12 45-minute sessions over 8 weeks 3. Group CBT (n = 28) Eight 2-hour sessions over 12 weeks; led by psychologists	<ul style="list-style-type: none"> Vestibular pain index (11-point scale) Pain intensity of vaginal intercourse (11-point scale) McGill Pain Questionnaire: Pain Rating Index & Sensory scale Sexual Information Scale Frequency of sexual intercourse Global Severity Index of Brief Symptom Inventory Credibility Gynecological examination 	Low Unclear Low Low
Desrochers et al. 2010 [43]	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, as well as education; prescribed by 2 gynecologists; discontinued after 8 weeks if no improvement	<ul style="list-style-type: none"> McGill Pain Questionnaire—Present Pain Intensity Frequency of intercourse Female Sexual Function Index Sexual satisfaction Global Severity Index of Brief Symptom Inventory Pain Catastrophizing Scale Pain Intercourse Self-Efficacy Scale McGill Pain Questionnaire—Pain Rating Index 	Low Unclear Low Unclear
Brown et al. 2009 [44]	53 women with vulvodynia (generalized and provoked) Nonresponders 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. Amitriptyline (tricyclic antidepressant) (n = 13) 10 mg a day for 6 weeks; if well tolerated, increased to 20 mg for remainder of study 3. Amitriptyline + triamcinolone cream (corticosteroid) (n = 14) 10 mg amitriptyline a day for 6 weeks, increased to 20 mg for remainder of study if well tolerated, plus once daily application of 5 mg triamcinolone cream on affected area; cream discontinued at 6 weeks	<ul style="list-style-type: none"> McGill Pain Questionnaire—Pain Rating Index 	Low Unclear Low High
Danielsson et al. 2006 [45]	46 women with vestibulodynia Mean age: Group 1: 25.8 Group 2: 23.3 Outpatient vulvar clinic, Sweden	1. Lidocaine (local anesthetic) (n = 23) 2% gel, 5% ointment; applied 5–6 times a day for 2–4 months 2. Electromyographic biofeedback (n = 23) Vaginal sensor applied three times a day for 10 minutes per session at home	<ul style="list-style-type: none"> Pain pressure thresholds Short Form 36 Prime Care Evaluation of Mental Disorders Quality of Life (0–100, visual analog scale) Sexual functioning (0–100, visual analog scale) Coital pain (0–100, visual analog scale) 	Unclear Unclear Unclear Unclear
Masheb et al. 2009 [46]	50 women with vulvodynia (generalized & provoked) Mean age 43 University students	1. CBT (n = 25) Ten weekly 60-minute sessions 2. Supportive psychotherapy (n = 25) Ten weekly 60-minute sessions; both treatments delivered by doctoral-level research therapists	<ul style="list-style-type: none"> Physician assessment: speculum, cotton swab, and erythema Multidimensional Pain Inventory McGill Pain Questionnaire Female Sexual Function Index Beck Depression Inventory Pain Anxiety Symptom Scale Global improvement rating (scale 0–5) Satisfaction and credibility rating (scale 0–10) 	Low Unclear Unclear Low

Table 1 Continued

Study ID	Population	Treatment/comparator	Outcomes	Selection, performance, attrition, and detection bias
Schnyder et al. 1998 [47]	44 women with vaginismus (DSM-III; acquired & lifelong) Mean age 28 Mean duration of problem 4.02 years Outpatient obstetrics and gynecology department, Switzerland	1. In vivo desensitization (n = 21) Dilators introduced manually by physician; 10–15 minutes of desensitization 5 times a week 2. In vitro desensitization (n = 23) Dilators introduced verbally by physician; 10–15 minutes of desensitization 5 times a week 3. Therapy sessions (n = 44) Treatment delivered every 2 weeks by two experienced sex therapists; number of sessions not reported	• Successful intercourse (outcome measure not described in detail)	High Unclear High High
Ter Kuile et al. 2013 [48]	70 women with vaginismus (lifelong) and their partners Mean age 28.9 Mean duration of problem 10.41 years Outpatient clinic in psychosomatic gynecology and sexology, Netherlands	1. Exposure therapy (n = 35) Maximum of three 2-hour sessions within 1 week, plus two follow-up sessions over 5 weeks; in vivo desensitizations, 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 • Fear of Sexuality Questionnaire • Female Sexual Distress Scale	Unclear High Low Unclear
Van Lankveld et al. 2001 [49]	55 women (vaginismus n = 28; dyspareunia n = 25) Subset of couples with numerous sexual dysfunctions Mean age of subsets unknown, overall mean age of females 35 Outpatient gynecology and sex clinic, Netherlands	1. CBT bibliotherapy (n = 125) Given manual to read; 10 weeks' duration with telephone support; broken down further into randomized participants (n = 104) (self-initiated contacts if having difficulties) and nonrandomized participants (n = 21) (scheduled telephone contacts) 2. Waiting list (n = 98)	• Golombok–Rust Inventory of Sexual Satisfaction • Maudsley Marital Questionnaire • Intimate Contact Body Scales • Self-rated evaluation of treatment (4-item scale) • Compliance	High Unclear Low Unclear
Van Lankveld et al. 2006 [50]	117 couples (women with vaginismus, DSM-IV-TR, lifelong only) Mean age of females 28.6 Mean age of 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 (level of penetration achieved) • Mini International Neuropsychiatric Interview • Female Sexual Function Index • Maudsley Marital Questionnaire • Golombok–Rust Inventory of Sexual Satisfaction • Sexual and Physical Abuse Questionnaire	Low Unclear Low Unclear
Weijmar Schultz et al. 1996 [51]	14 women with vestibulodynia and unable to have intercourse Mean age 24 Dutch gynecology department	1. Behavioral therapy + placebo surgery (n = 7) Hospitalized for one night and given local anesthetic 2. Surgery + behavioral therapy (n = 7) Hospitalized for one night and surgical excision under local anesthetic performed Participants excluded if they initiated other pain treatment in the past 2 months	• Problem severity (5-point scale)	High Low High High

CBT = cognitive-behavioural therapy

CBT Versus Other Psychological Treatments

Three studies of good quality (n = 148) compared CBT with other psychological treatments (biofeedback, supportive therapy, and bibliotherapy) with regard to posttreatment outcomes of pain on intercourse; no significant effect was found, nor was one found at later follow-up (1 and 2.5 years) for the two of the three that performed such follow-up. Two studies (n = 83) compared CBT with other psychological treatments (biofeedback

and supportive therapy) with regard to sexual functioning; no significant effect was found. Although data from one study could not be obtained [47], a previous meta-analysis [72] calculated odds ratios for this study comparing two forms of behavioral treatment and found no significant effect on successful penetration ($Z = 0.52$, $P = 0.60$).

Three studies of mixed quality (n = 142) compared cognitive or behavioral therapy with other

Table 2 Effects of psychological treatment compared with other treatments or controls

Study ID	N	Populations	Treatment	Comparator	Outcome	Z	P	Heterogeneity
Bergeron et al. 2001 [35] Desrochers et al. 2010 [43] Brown et al. 2009 [44]	143	Vestibulodynia Provoked/generalized vulvodynia	CBT	Medical (surgery, topical cream, and medication)	General pain post-treatment	1.40	0.16	Moderate (25%)
Bergeron et al. 2001 [35] Desrochers et al. 2010 [43]	111	Vestibulodynia	CBT	Medical (surgery and topical cream)	Pain on intercourse post-treatment Pain on intercourse follow-up	0.77 0.07	0.44 0.10	High (77%) Moderate (45%)
Bergeron et al. 2001 [35] Desrochers et al. 2010 [43]	111	Vestibulodynia	CBT	Medical (surgery and topical cream)	Sexual functioning post-treatment Sexual functioning follow-up	0.21 0.79	0.83 0.43	None None
Danielsson et al. 2006 [45] Weijmar Schultz et al. 1996 [51]	46	Vestibulodynia	Behavior therapy	Medical (medication and surgery)	Symptom elimination (follow-up)	0.41	0.68	None
Bergeron et al. 2001 [35] Masheb et al. 2009 [46] Van Lankveld et al. 2006 [50]	148	Vestibulodynia Provoked/generalized vulvodynia Vaginismus	CBT	Other psychological (biofeedback, supportive, and bibliotherapy)	Pain on intercourse post-treatment	0.32	0.75	Low (15%)
Bergeron et al. 2001 [35] Masheb et al. 2009 [46]	83	Vestibulodynia Provoked/generalized vulvodynia	CBT	Other psychological (biofeedback and supportive)	Pain on intercourse follow-up	1.08	0.28	High (69%)
Bergeron et al. 2001 [35] Masheb et al. 2009 [46]	83	Vestibulodynia Provoked/generalized vulvodynia	CBT	Other psychological (biofeedback and supportive)	Sexual functioning post-treatment	1.03	0.30	None
Al-Sughayir 2005 [41] Masheb et al. 2009 [46] Van Lankveld et al. 2006 [50]	134	Vaginismus Provoked/generalized vulvodynia	Cognitive and behavior therapies	Other psychological (hypnotherapy, supportive, and bibliotherapy)	Sexual anxiety post-treatment	1.60	0.11	Low (23%)
Van Lankveld et al. 2001 [49] Van Lankveld et al. 2006 [50] Van Lankveld et al. 2006 [50]	88	Vaginismus Dyspareunia	Bibliotherapy	Waitlist control	Frequency of sex	2.00	0.05*	None
Van Lankveld et al. 2006 [50] Ter Kuile et al. 2013 [48]	66	Vaginismus	Cognitive and behavior therapies	Waitlist control	Pain on intercourse post-treatment	0.89	0.37	High (91%)
Van Lankveld et al. 2006 [50] Ter Kuile et al. 2013 [48]	66	Vaginismus	Cognitive and behavior therapies	Waitlist control	Sexual anxiety post-treatment	0.28	0.78	High (88%)

*Statistically significant, $P \leq 0.05$
CBT = cognitive-behavioural therapy

Table 3 Effect sizes for psychological treatment arms

Study ID	Population	Outcome	Treatment	Effect sizes, small to large
Bergeron et al. 2001 [35]	Vestibulodynia	General pain	CBT	0.09
Brown et al. 2009 [44]	Provoked/generalized		Biofeedback	0.16
Desrochers et al. 2010 [43]	vulvodynia		CBT	0.31
Masheb et al. 2009 [46]			CBT	0.44
			CBT	0.78
			Supportive	0.64
Bergeron et al. 2001 [35]	Vestibulodynia	Pain on intercourse	CBT	0.61
Desrochers et al. 2010 [43]	Provoked/generalized		Biofeedback	0.71
Masheb et al. 2009 [46]	vulvodynia		CBT	0.69
			CBT	0.53
			Supportive	0.48
Bergeron et al. 2001 [35]	Vestibulodynia	Sexual functioning	CBT	0.16
Desrochers et al. 2010 [43]	Provoked/generalized		Biofeedback	No effect
Masheb et al. 2009 [46]	vulvodynia		CBT	0.44
			CBT	0.69
			Supportive	0.15
Bergeron et al. 2001 [35]	Vestibulodynia	Sexual activity frequency	CBT	0.12
Desrochers et al. 2010 [43]			Biofeedback	<0.1
			CBT	<0.1
Van Lankveld et al. 2006 [50]	Vaginismus	Pain on intercourse	Bibliotherapy	0.64
Ter Kuile et al. 2013 [48]			CBT	0.69
			Exposure	2.29
Ter Kuile et al. 2013 [48]	Vaginismus	Fear of intercourse	Exposure	0.89
Van Lankveld et al. 2006 [50]	Vaginismus	Other penetration behaviors	Bibliotherapy	0.93
			CBT	1.19
Van Lankveld et al. 2001 [49]	Dyspareunia	Sexual functioning	Bibliotherapy	0.31
Ter Kuile et al. 2013 [48]	Vaginismus		Bibliotherapy	1.46
			Exposure	<0.1
Van Lankveld et al. 2001 [49]	Dyspareunia	Sexual activity frequency	Bibliotherapy	<0.1
	Vaginismus		Bibliotherapy	0.49

psychological therapies (hypnotherapy, supportive therapy, and bibliotherapy) with regard to sexual anxiety; no significant effect was found. Removing the study on vestibulodynia to leave two studies on vaginismus showed a significant reduction in anxiety ($Z = 1.94$, $P = 0.05$, effect size [ES] = 0.47, confidence interval [CI] = -0.00–0.95), although one of the remaining studies was rated as high-risk on three domains of bias [41]. As before, psychological treatments appeared to be as effective or ineffective as one another across outcomes.

Psychological Treatment Versus Waitlist Control

Two studies of adequate quality ($n = 88$) compared bibliotherapy with waitlist control with regard to frequency of sexual activity and found a significant effect ($Z = 2.00$, $P = 0.05$, ES = 0.43, CI = 0.86 to 0.01). Two studies, also of adequate quality ($n = 66$), compared CBT and exposure treatment with waitlist control with regard to posttreatment outcomes of pain and fear of intercourse; no significant effects were found.

Effect Sizes for Psychological Treatment

Posttreatment effects in comparison with baseline can be found in Table 3 and are reported first for studies that examined medically defined disorders and second for psychiatrically defined disorders.

Medically Defined Disorders

Four studies examined the impact of psychological treatment on pain not limited to intercourse. Effects of CBT ranged from small to large. The effect of biofeedback was small and that of supportive therapy large. Three studies examined pain on intercourse; effects of CBT and biofeedback were large, while supportive therapy produced a moderate effect.

Three studies examined the impact of psychological treatment on sexual functioning. Effects of CBT ranged from small to large. A small effect was found for supportive therapy, and no effect was found for biofeedback. Two studies examined the frequency of sexual activity; no or very small

effects of CBT were found, and no effect was found for biofeedback.

Psychiatrically Defined Disorders

Two studies that examined the impact of psychological treatment on pain on intercourse found large effects for CBT, exposure, and bibliotherapy. One study also examined ability to undertake penetration other than in sexual activity and found a large ES for both CBT and bibliotherapy [50]. One study examined the impact of bibliotherapy on sexual functioning and found a moderate effect for a dyspareunia subset and a large effect for a vaginismus subset [49]. This study also examined frequency of sexual activity, finding no effect for the dyspareunia subset and a moderate effect in the vaginismus subset.

Summary of Effect Sizes

Psychological treatments for vulvodynia produced wide-ranging effects for general pain but more consistently large effects for sexual pain, comparable to those for vaginismus. This suggests that such treatments have an impact on sexual pain regardless of assumed etiology. In terms of sexual function, CBT for vulvodynia produced a range of effect sizes, from small to large. Large effect sizes were found for vaginismus.

For frequency of sexual activity in vulvodynia, no or minimal effects were found. Interestingly, within one study, a moderate to large effect was found for sexual frequency in a vaginismus subset, but no effect was found in the dyspareunia subset [49]. This finding might suggest differential treatment effects between vulvodynia/dyspareunia and vaginismus, although the study was rated as high-risk with regard to selection bias, and the subset was small.

Discussion

A systematic review and meta-analysis on the effectiveness of psychological interventions for vaginal pain defined as primarily medical or psychiatric in etiology found 12 studies that compared psychological treatments with medical treatments, other psychological treatments, or control groups. Overall, no significant differences were found between psychological and medical treatments for vulvodynia on any outcome. Unfortunately, we found no vaginismus trials that compared psychological with medical treatment, preventing direct comparisons and conclusions about relative effectiveness. No significant differences were found for

vulvodynia and vaginismus when comparing two psychological treatments, also supporting the proposal that treatments are equally effective or equally ineffective.

Without a control group, it is difficult to determine whether treatments are equally effective or equally ineffective. Three trials that used control conditions demonstrated significant effects of treatment, all for vaginismus [48–50]. Whether or not it denotes treatment gains, the finding of equivalence from medical and psychological treatments for vulvodynia is of interest, partly because medical treatment is generally the first-line option [73,74], with psychological treatment often unavailable [75]. It is also of interest because it raises the question of whether the treatments have distinct actions with similar effects on pain or act through common pathways. A simple view would posit that medical interventions, such as medication or surgery, affect pain via changes to the peripheral pain system, whereas psychological treatments mediate pain experience via top-down changes in emotional and cognitive aspects of pain. A biopsychological perspective would rather envisage both psychological and biological processes influencing pain in recursive loops [19]. Even this is an oversimplification given the diversity of treatment methods and their different targets, highlighting the difficulty in casting light on etiology by examining treatment response. While it is frequently the case that different psychological therapies have equal effectiveness, which is hypothesized to be due to common therapeutic factors rather than specific therapeutic methods [76], it is less usual to find equivalence of psychological and medical treatments. One small, low-quality trial in the review combined psychological and medical treatments for vestibulodynia [51], comparing placebo and real surgery, both in combination with behavior therapy, and found similar symptom reduction in the two treatment arms. This cannot be further dismantled to identify effects. While multidisciplinary approaches are recommended and used in clinical practice, this recommendation was not reflected in the randomized controlled trial evidence. This limited the possibility of comparisons of single and multicomponent treatments.

Only two analyses included populations with both medically defined and psychiatrically defined disorders. There was an indication of a differential response to treatment of sexual anxiety from these analyses: vaginismus patients appeared to benefit more from cognitive and behavioral therapies than vestibulodynia patients, when compared with other

psychological interventions. Given the lack of direct comparisons, we calculated study-wise treatment effect sizes for each arm of psychological intervention. Two vaginismus studies found large effects on sexual pain comparable to and larger than effect sizes found for psychological treatment in vulvodynia [48,50]. Similarly, large effect sizes on outcomes of sexual functioning were found for both medically and psychiatrically defined vaginal pain.

So does this suggest that pain reduction and sexual function can be improved with psychological treatment, regardless of the perceived etiology? A more psychologically oriented explanation would point to commonalities across conditions in response to pain in terms of anxiety, fear and avoidance, vigilance to pain, and depression, all of which can be addressed using psychological treatment, with consequent improvement in pain experience. Because vaginismus trials did not in general evaluate effects on pain (instead evaluating effects on sexual functioning, consistent with the classification of vaginismus as a sexual disorder), we found no data with which to address this question [9]. It is possible that the inclusiveness of the definition of GPPPD in DSM-5 may encourage inclusion of pain as a primary outcome of treatment [12].

Limitations

Our interpretation of findings is limited by the variable quality of the studies, with much risk uncertain where information was not reported. The total number of studies included makes our conclusions tentative, but attempting to systematically evaluate the evidence provides a platform for future review work and helps to identify areas lacking adequate empirical work. In addition, sample sizes in particular were generally very small, raising issues of type II error, and presumably associated with low rates of help-seeking rather than of prevalence [77,78]. Diversity of interventions and of outcomes meant that no more than three studies were eligible to combine in any analysis, making it hard to draw conclusions across the field. Inactive controls were used in only three studies, with the remainder comparing active treatments, from which equivalence of outcome is hard to interpret. Only one study [49] included dyspareunia as a distinct condition using a psychiatric classification, which may signify the interchangeable nature of the vaginal pain conditions.

This review included a limited number of medically defined conditions (vulvodynia subtypes) associated with sexual dysfunction. Adding excluded conditions, such as chronic pelvic pain

and deep dyspareunia, might have increased the volume of data and strength of conclusions [79].

Conclusions

This review highlighted the shortage of studies comparing psychological and medical treatments for psychiatrically defined vaginal pain, along with that of studies that use a controlled design for treatment trials for medically defined vaginal pain. Therefore, questions remain about the relative and overall efficacy of psychological treatments for vaginal pain. If the preliminary finding that psychological treatments are equally effective for all vaginal pain types is supported by further research, it could be argued that the less invasive method of treatment should be offered first. UK guidelines for mental health conditions (e.g., depression and generalized anxiety) often recommend nonmedical options first if treatments are equally effective [80,81]. Applying a biopsychological understanding of pain and allowing patients a choice of treatments could also help to improve outcomes. While a multidisciplinary approach may be recognized as the ideal in clinical practice, this was not reflected in the evidence; only one trial attempted to evaluate a combined medical and psychological approach [51].

Starting from the new definition of GPPPD in DSM-5, more research into integrative classifications and treatment approaches could help to improve care for pain-induced sexual dysfunction. Multicomponent models of pain have been applied to medically defined vaginal pain in the form of pain management, but these have not used controlled trial methodology, nor have they considered the relative impact of psychological and physiological treatment on pain [82,83]. Better-defined outcome measures in single-component and multicomponent controlled treatment studies could help determine which aspects of treatment influence the various aspects of pain and sexual functioning. Supplementary qualitative research could help to explain the experiences and treatment preferences of women with vaginal pain.

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Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher's website:

Appendix S1 Search strings.

Appendix S2 Full quality assessment for included studies.

Appendix S3 Forest plots from meta-analyses.