Pain Medicine, 22(12), 2021, 2863–2875 doi: 10.1093/pm/pnab265

Advance Access Publication Date: 28 August 2021

Original Research Article

A Network Analysis of Selected Psychosocial Factors in Vulvodynia and Its Subtypes

Claudia Chisari, MSc,* Ioannis Begleris, PhD, Mani B. Monajemi, MD,* Fiona Lewis, MD,[†] Rona Moss-Morris, PhD,* Whitney Scott, PhD,*,[‡] and Lance M. McCracken , PhD,*

*Health Psychology Section, Institute of Psychiatry, Psychology, and Neuroscience (IoPPN), King's College London, London, UK; [†]St. John's Institute of Dermatology, Guy's & St. Thomas's NHS Foundation Trust, Guy's Hospital, London, UK; [‡]INPUT Pain Management Unit, Guy's and St. Thomas' NHS Foundation Trust, London, UK; and [§]Department of Psychology, Uppsala University, Uppsala, Sweden

Correspondence to: Lance M. McCracken, PhD, Department of Psychology, Uppsala University, Box 1225, 751 42 Uppsala, Sweden. Tel: +46 73-899 97 47; Fax: +46 18-471 21 23; E-mail: Lance.McCracken@psyk.uu.se.

Funding sources: This study represents independent research funded by the National Institute for Health Research (NIHR) Biomedical Research Centre at South London and Maudsley National Health Service (NHS) Foundation Trust and King's College London. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR, or the Department of Health and Social Care.

Conflicts of interest: The authors have no conflicts of interest to declare.

Abstract

Objective. Psychosocial factors are related to pain and sex-related outcomes in provoked vulvodynia and possibly in mixed and spontaneous vulvodynia. However, a broader behavioral framework, such as the psychological flexibility model, has received limited attention in this context. Recently, additional psychosocial variables have also emerged that appear relevant to vulvodynia, including perceived injustice, body-exposure anxiety during intercourse, and unmitigated sexual communion. The present study applied network analysis to explore relations between psychological flexibility, newly emerging psychosocial variables relevant to vulvodynia, and their associations with vulvodynia outcomes. The study also explored potential differences across vulvodynia subtypes. Design. An online crosssectional study of 349 participants with vulvodynia (112 provoked, 237 spontaneous/mixed) was carried out. Methods. Participants completed self-report questionnaires, including questions on pain and sexual outcomes, depression, facets of psychological flexibility, body-exposure anxiety during intercourse, unmitigated sexual communion, and perceived injustice. Networks were computed for the total sample and for provoked and mixed/spontaneous vulvodynia subsamples. Results. Perceived injustice, pain acceptance, and depression were "central" factors among the included variables, in all models. Psychological flexibility processes were relevant for all networks. Depression was more central in the network for mixed/spontaneous vulvodynia; body-exposure anxiety during intercourse was most central for the provoked subtype. Conclusions. Among the included variables, perceived injustice, pain acceptance, depression, and psychological flexibility appear to be important in vulvodynia. As different factors are significant across subtypes, tailored treatment approaches are suggested.

Key Words: Vulvodynia; Genito-Pelvic Pain/Penetration Disorder; Chronic Pain; Psychological Flexibility; Network Analysis; Psychosocial Factors

Introduction

Vulvodynia is characterized by persistent vulval pain [1, 2]. Vulvodynia can be spontaneous, provoked by pressure, or mixed [2]. Provoked vestibulodynia (PVD), defined by pain triggered by pressure on the vulval vestibule, is the most studied type of vulvodynia [3]. The lifetime prevalence of vulvodynia is 10–28% [4], and

pain can occur during daily activities [5], resulting in significant impacts on functioning [1]. The etiology of vulvodynia remains unknown [2, 6, 7], and biomedical treatments achieve only modest improvements in pain and sexual functioning [8], suggesting that a broader conceptualization is needed.

Several studies [9–13] have highlighted the importance of pain-specific [14], sex-related [10], and social factors [15] in PVD. Psychological variables such as pain catastrophizing, self-efficacy, and general anxiety have been researched in vulvodynia [16–18] and appear relevant in this population. In addition to these variables, there may be wider models worth investigating in vulvodynia, including the psychological flexibility model [19]. Psychological flexibility is defined as "the capacity to engage in personally important activities whilst remaining in conscious and open contact with thoughts, feelings, and sensations" [20, 21]. Psychological flexibility is a broad behavioral model that focuses on the function of experiences in relation to behavior rather than the specific content of thoughts and feelings, and it can accommodate content-specific psychosocial variables [19]. Psychological flexibility can be summarized as behavior that is "open (acceptance and cognitive defusion), aware (present-moment awareness and self-as-context), and engaged (values-based and committed action)" [20, 21]. The acceptance component of psychological flexibility was related to lower pain and greater sexual functioning in one study on PVD [22]. Given the relevance of psychological flexibility in relation to pain more broadly [21, 22], as well as growing evidence for Acceptance and Commitment Therapy [23], a treatment approach that fosters psychological flexibility, this model appears worth exploring in vulvodynia.

In addition to the psychological flexibility model, there are other emerging variables, consisting of specific feelings, thoughts, or perceptions potentially relevant to vulvodynia. In contrast to the contextual variables included in the psychological flexibility model, these variables can be regarded as reflecting specific psychological content. For instance, women with vulvodynia experience pain invalidation, which could potentially lead to perceived injustice, and this could adversely influence outcomes [15, 24]. Furthermore, women with PVD report changes to their body image, perceiving themselves as "unattractive" or "not women anymore." [25] Related body-exposure anxiety and avoidance during sexual activities may lead to greater pain and sexual dysfunction in PVD [25]. Lastly, as women with vulvodynia engage in intercourse despite their pain [26], sexual motivation may be an important avenue to investigate. Unmitigated sexual communion—"meeting a partner's sexual needs to the exclusion of one's own"-may lead to worse pain and sexual dysfunction in PVD [27]. These variables appear promising, relatively specific to the experience of vulvodynia, and worth further study.

Psychological flexibility, perceived injustice, bodyexposure anxiety, and unmitigated sexual communion appear promising for an improved understanding of vulvodynia outcomes. Simultaneous investigation of thoughts, feelings, and perceptions alongside broader behavioral processes allows for an understanding of the complex interrelations among these variables, which represent levels of context and content. There is also a need to understand potential differences across subtypes, as the situations in which the pain occurs differ. Such differences could have implications for the development of individualized treatments [28].

Network analysis represents a novel data-driven approach that examines complex interrelationships among variables in a way that traditional methods (e.g., multiple regression) cannot accommodate [29]. A key advantage is that network analysis can identify key variables within a network of selected variables, accounting for all possible linkages simultaneously and allowing unexpected links to emerge [29, 30]. Therefore, using network analysis, the present study aimed to 1) identify the complex relations among a) psychological flexibility variables, b) selected, newer, content-specific psychosocial factors, and c) vulvodynia outcomes when simultaneously considered, and 2) explore possible subtype differences in the networks derived.

Methods

This was an online cross-sectional study of women living with vulvodynia. The present study includes analyses and findings from baseline data, part of a larger online longitudinal study. The larger longitudinal study explored changes in psychosocial factors and outcomes in vulvodynia at baseline and 3 months' follow-up through the use of self-report questionnaires between November 2019 and June 2020. Baseline data reported in this article were collected via the online survey platform Qualtrics between November 2019 and February 2020, and all participants were given an anonymous ID link. The King's College London Ethics committed approved this study (HR-19/20-14507).

Recruitment for the present study was pursued through the Vulval Pain Society and on social media. Participants were eligible if they self-reported having a confirmed diagnosis of vulvodynia from a health professional (defined as "idiopathic vulval pain of three months or more" [2]), were 18 years of age or older, were not pregnant, and were experiencing vulvodynia at study entry. A sample of 349 women provided informed consent and completed the questionnaires. The questionnaire included demographics, questions about vulvodynia, and measures assessing psychosocial factors, pain severity, pain interference, sexual functioning, and sexual satisfaction variables.

Questionnaires

Vulvodynia-Related Questions

Questions about vulvodynia subtypes were included. These included whether the pain was spontaneous (i.e., pain occurring without a trigger such as pressure), provoked by pressure/touch, or mixed (a combination of provoked and spontaneous). The onset of vulvodynia was determined by asking participants whether they

experienced vulvodynia from the first time they attempted intercourse (primary vulvodynia) or whether dyspareunia occurred after pain-free intercourse (secondary vulvodynia). Questions on the duration of pain, its location, and whether participants were taking pain medications were also asked; the latter were entered by participants into free text boxes.

Chronic Pain Acceptance (Openness). Chronic pain acceptance is defined as "a process of engaging in activities with pain and refraining from unsuccessful efforts to control pain so that important life activities are pursued" [20]. To measure pain acceptance, the eight-item version of the Chronic Pain Acceptance Questionnaire (CPAQ-8) was used [31]. The CPAQ-8 consists of eight items on a seven-point scale, where 0 is "never true" and 6 represents "always true" and a higher score reflects greater levels of pain acceptance. A systematic review examined a range of questionnaires measuring chronic pain acceptance and showed that the CPAQ performed best in terms of its psychometric properties compared with other questionnaires [32]. We computed the internal consistency of this measure in the present study, and it displayed high internal consistency ($\alpha = 0.80$).

Present-Moment Awareness (Awareness).

The Tacting of Function Scale (TOF) is a questionnaire that measures the psychological process of tacting functional relations [33]. This is also defined by "one's ability to notice and reflect the purposes or guiding influences on one's behavior

"[33]. The measure consists of 10 items on an eightpoint scale, where 0 is "never true" and 7 is "always true." A higher score on this measure reflects greater tacting of functional relations. The TOF demonstrated good psychometric properties in a sample of college students [33]. We computed the internal consistency of the TOF, and it was adequate ($\alpha = 0.80$).

Committed Action (Engagement).

Committed action is defined as "flexible persistence in values-based or goal-directed behavior" [19]. To measure this construct, the eight-item version of the Committed Action Questionnaire (CAQ-8) was used [34]. The CAQ-8 consists of eight items on a seven-point scale, where 0 is "never true" and 6 represents "always true" and a higher score reflects greater levels of committed action. Past research has indicated that the CAQ-8 has good psychometric properties [34], and it had adequate internal validity in the present study ($\alpha = 0.82$).

Depression. To measure depression, we used the Patient Health Questionnaire-9 (PHQ-9) [35]. The PHQ-9 consists of nine items that reflect the frequency of symptoms of depression on a four-point scale, where 0 is "not at all" and 3 is "nearly every day." A higher score on this measure reflects greater levels of depressive symptoms. Past research has indicated that the PHQ-9 has good psychometric properties and can discriminate

between people with and without a diagnosis of depression in people with pain [36]. This measure showed adequate internal validity in the present study ($\alpha = 0.84$).

Body-Exposure Anxiety and Avoidance During Sexual Activities. To measure body image in the context of sexual relations, the Body Exposure Anxiety and Avoidance During Sexual Activities questionnaire (BESAQ) was used [37]. On the BESAQ, respondents rate 28 items on a five-point scale ranging from 0 ("never") to 4 ("always or almost always"). Higher scores reflect "greater self-conscious or anxious attentional focus on their body's appearance and stronger desires and attempts to avoid the exposure of certain aspects of their body to sexual partners" [37]. Past research in women with PVD [25] has indicated that this measure has good psychometric properties. This measure had high internal validity in the present study ($\alpha = 0.95$).

Perceived Injustice. Perceived injustice is defined as "a multidimensional construct, comprising elements pertaining to the severity and irreparability of loss, blame, and a sense of unfairness" [38]. This was originally explored in people with musculoskeletal injury [38]. To measure pain-related perceived injustice, we used the Injustice Experiences Questionnaire (IEQ) [38]. On the IEQ, participants rate the 12 items, such as "it all seems so unfair" and "most people don't understand how severe my condition is," on a five-point numerical scale, where 0 is "not at all" and 4 is "all the time." Higher scores on this scale reflect greater perceived injustice. The IEQ has been used in people with several chronic pain conditions [38] and PVD [15], demonstrating good psychometric properties. For the present study, we adapted the IEQ by replacing the word "injury" used in the questionnaire description instructions "vulvodynia." The following description shows how the IEQ was adapted for the present study: "When vulvodynia happens, it can have profound effects on our lives. This scale was designed to assess how vulvodynia has affected your life." This measure had high internal validity in this study ($\alpha = 0.89$).

Unmitigated Sexual Communion. The Unmitigated Sexual Communion Scale (USC) is a three-item scale assessing unmitigated sexual communion, defined as "consistently placing others' needs before one's own, worrying excessively about others' problems, and focusing on others to one's detriment" [27]. The three items were rated on a five-point scale ranging from 0 (strongly disagree) to 5 (strongly agree). Higher scores on the USC indicate greater unmitigated sexual communion. This measure was used in women with dyspareunia, displaying good psychometric properties [27]. This measure showed high internal validity in the present study ($\alpha = 0.86$).

Outcome Variables

Pain Severity and Interference. To measure pain severity and pain interference, the Brief Pain Inventory Short

Form (BPI) was used [39]. For pain severity, people rate their worst, least, present, and average pain in the prior week on a scale from 0 ("no pain") to 10 ("pain as bad as you can imagine"). A pain severity score index is computed by averaging the four ratings. For pain interference, participants rate the extent to which pain interference, participants rate the extent to which pain interference on the following domains: "general activity, mood, mobility, work, relationships, sleep, and enjoyment of life." These are rated on a scale from 0 ("no pain") to 10 ("pain as bad as you can imagine"). A pain interference score is computed by averaging the seven ratings. In the present study, the BPI had high internal consistency for pain severity ($\alpha = 0.88$) and pain interference ($\alpha = 0.90$).

Sexual Functioning. To measure sexual functioning, the Female Sexual Function Index (FSFI) [40] was used. This measure includes 19 items on a six-point scale assessing different domains of sexual functioning, such as desire, lubrication, etc. Higher scores reflect greater sexual functioning. Past research has indicated that the FSFI has good psychometric properties in women with PVD [41], and it had adequate internal consistency in the present study ($\alpha = 0.95$).

Sexual Satisfaction. To measure sexual satisfaction, the Global Measure of Sexual Satisfaction scale (GMSEX) [42] was used. The GMSEX consists of five items on a seven-point scale, with different anchors for each item. Higher scores on this measure reflect greater sexual satisfaction. Past research has indicated that the FSFI has good psychometric properties, including in women with PVD [43, 44], and it had high internal consistency in the present study ($\alpha = 0.95$).

Statistical Analyses

IBM SPSS Statistics 26 and Python (using NetworkX) [45] were used to perform statistical analyses. Missing data occurred in 10.8% of the sample. Mean imputation was first conducted at the questionnaire level if, for each participant, at least half of the items within each questionnaire were completed. For participants for whom more than half of the items were missing on a given questionnaire, multiple imputations were conducted, resulting in 20 multiply imputed datasets. Before and after imputations, normality tests were carried out. Data were also explored to assess potential differences across the vulvodynia subtypes in demographic variables and pain levels. Correlation analyses in SPSS with the pooled correlation coefficients were conducted. Three correlation analyses were conducted with the pooled data: total sample (provoked, mixed, spontaneous), provoked sample, and mixed/spontaneous sample.

Network Development

The present study aimed to explore and create networks in the total sample and by vulvodynia subtype (provoked versus mixed/spontaneous). Three network models were constructed in Python, and their properties were obtained with the NetworkX package [45]. Network models explore relations between variables, or "nodes," through partial correlations and present them in a graphical format, with each link referred to as an "edge." The visual representation of a network provides researchers with the opportunity to examine the structure of a group of variables and their patterns of relations in a way that traditional approaches cannot accommodate [29, 30].

The networks are, again, based on partial correlations between the nodes and correlations between pairs with all other correlations taken into account, enabling researchers to examine the unique relationships between nodes. To detect correlations as a function of sample size for this study, a sample of at least 85 participants was required according to the Pearson correlation coefficient power curve [46]. The Fruchterman-Reingold force-directed algorithm [47], implemented in NetworkX, was used to plot the networks. This algorithm forces nodes with many and strong edges to be placed centrally and strongly connected node pairs to be placed closely while also minimizing overlap of edges and nodes. The weight of the edge reflects the strength of the relation conditional on all other possible relations. The seismic color bar within the Matplotlib library [48] was used to represent the direction and strength of the edges for positive relationships (colored in blue) and negative relationships (colored in red), where darker colors correspond to stronger associations between nodes.

Nodes have different sizes in the networks. Larger nodes represent stronger associations with the rest of the nodes within the graph, whereas smaller nodes represent weaker associations with the rest of the nodes within the graph. To interpret a network, multiple features are examined. These include the distance between nodes, how strongly connected nodes are with each other, and the proximity of a node to the center of the network. In addition to visual inspection, we used NetworkX to calculate the following centrality indices: closeness, degree, and strength [49]. Closeness refers to the inverse of the sum of distances to all other nodes. Degree refers to the number of edges each node has within the network. Strength includes how strongly connected a node is with other nodes and the sum of the weights of the relations with which a node is involved. As strength centrality encapsulates both degree and closeness indices, it is considered the most informative centrality index. Predictability is the degree of variance in each node that can be explained by variance in nodes to which it is connected, an estimate akin to R^2 ; this was estimated for the three networks.

Results

Tables 1 and 2 display the disease and demographic characteristics of the sample (n = 349). The mean age of participants was 32.6 years (standard deviation [SD] 11.4), and they were mostly white. Among the participants, 32% reported provoked vulvodynia, 58.5% reported

Table 1. Participants' demographic characteristics (n = 349)

Demographic Factor	Mean (SD) or n (%)	
Age, y, mean (SD)	32.6 (11.4)	
Ethnic group, n (%)		
White	322 (92.1)	
Asian	6 (1.8)	
Mixed	6 (1.8)	
Black	4 (1.2)	
Other	11 (3.1)	
Relationship status, n (%)		
Alone	48 (13.8)	
With partner and/or children	200 (57.3)	
With other family members	69 (19.8)	
With friends/flatmates	32 (9.1)	
Work status, n (%)		
Employed	183 (52.4)	
Employed part-time because of pain	31 (8.9)	
Unemployed because of pain	28 (8)	
Unemployed for other reason	10 (2.9)	
Other (retired, homemaker, student, etc.)	97 (27.8)	
Relationship status, n (%)		
In a relationship	265 (75.9)	
Not in a relationship because of pain	38 (10.9)	
Not in a relationship for other reason	46 (13.2)	
Sexually active, n (%)		
Yes	195 (55.9)	
No because of pain	132 (37.9)	
No for other reason	22 (6.3)	

Table 2. Participants' pain and health-related characteristics (n = 349)

Pain and Health-Related Factor	Mean (SD) or n (%)	
Pain duration, y, mean (SD)	7.7 (7.5)	
Pain site, n (%)		
All vulva	179 (51.3)	
Clitoris	5 (1.4)	
Vestibule	162 (46.4)	
Vagina	3 (0.9)	
Pain trigger, n (%)		
Spontaneous	33 (9.5)	
Provoked	112 (32.1)	
Mixed	204 (58.5)	
Pain onset, n (%)		
Primary	120 (34.4)	
Secondary	229 (65.6)	
Reported comorbidities, n (%)	233 (67)	
Fibromyalgia	17 (4.8)	
Bowel conditions	42 (12.03)	
Depression and/or anxiety	128 (36.7)	
Bladder and kidney conditions	29 (8.3)	
Sexual/reproductive health conditions	57 (16.3)	
Skin conditions	13 (3.7)	

mixed vulvodynia, and 9.5% reported spontaneous vulvodynia. More than half of participants (66%) classified themselves as having secondary vulvodynia, and the remainder reported primary vulvodynia. Participants reported an average pain duration of 7.7 years (SD 7.5), and more than half of them (67%) reported living with another physical or psychiatric condition. When pain duration and demographic variables were explored across

subtypes, no significant differences were found. Reflecting this, the smallest P value, when vulvodynia subtypes were compared on pain or demographic data, was 0.81.

Table 3 shows the mean scores of the psychosocial variables and pain and sexual outcomes. Participants had an average pain severity score of 3.77 (SD 2.09) and an average interference score of 4.23 (SD 2.46). Similarly, participants had a mean depression score of 11.44 (SD 5.96) and a sexual functioning score of 40.01 (SD 22.28), corresponding to moderate depressive symptoms [35] and the presence of sexual dysfunction [40].

Networks

Network 1: Total Sample

The first network (total sample N = 349) is presented in Figure 1. For this network, three network "communities," groups of closely aligned nodes, emerged. Community 1 included unmitigated sexual communion, present-moment awareness, body-exposure anxiety, and committed action. Community 2 included sexual functioning and satisfaction. Community 3 included depression, perceived injustice, pain acceptance, pain interference, and pain severity. Centrality estimates for each node are presented in Figure 2. On the basis of strength centrality indices, the node with the highest centrality was pain interference, followed by depression, sexual satisfaction, and perceived injustice. In terms of predictability, the node with the highest predictability was pain interference (64%), followed by pain severity (53%), sexual satisfaction (50%), sexual functioning (48%), and pain acceptance (42%). Table 4 shows the predictability of the nodes for each network.

Network 2: Provoked Vulvodynia

The second network of women with provoked vulvodynia (n = 112) is presented in Figure 3. For this network, three network communities emerged. Community 1 included pain interference, perceived injustice, pain acceptance, and pain severity. Community 2 included sexual functioning, sexual satisfaction, and unmitigated sexual communion. Community 3 included body-exposure anxiety during intercourse, committed action, present-moment awareness, and depression. For this network, centrality estimates for each node are presented in Figure 4. On the basis of strength centrality indices, the node with the highest centrality was body-exposure anxiety/avoidance during intercourse, followed by pain interference, sexual satisfaction, and pain acceptance. In terms of predictability, the node with the highest predictability was sexual satisfaction (66%), followed by sexual functioning (61%), pain interference (57%), pain acceptance (45%), and body-exposure anxiety during intercourse (43%). Table 4 shows the predictability of the nodes for each network.

Table 3. Means and standard deviations of the total scores for the study variables

Variable	Mean (SD)
Pain interference	4.23 (2.46)
Pain severity	3.77 (2.09)
Depression	11.44 (5.96)
Committed action	28.18 (6.83)
Present-moment awareness	51.01 (9.02)
Pain acceptance	22.51 (7.93)
Perceived injustice	33.15 (9.89)
Unmitigated sexual communion	7.81 (3.84)
Body-exposure anxiety and avoidance during intercourse	39.04 (26.20)
Sexual functioning	40.01 (22.28)
Sexual satisfaction	19.32 (11.40)

Network 3: Mixed/Spontaneous Vulvodynia

The third network of women with mixed/spontaneous vulvodynia (n=237) is presented in Figure 5. For this network, three communities emerged. Community 1 included unmitigated sexual communion, present-moment awareness, body-exposure anxiety, and committed action. Community 2 included sexual functioning and sexual satisfaction. Community 3 included depression, perceived injustice, pain acceptance, pain interference, and pain severity. Centrality estimates for each node are presented in Figure 6. On the basis of strength centrality

indices, the node with the highest centrality was pain interference, followed by perceived injustice, sexual satisfaction, and depression. In terms of predictability, the node with the highest predictability was pain interference (61%), followed by pain severity (53%), sexual satisfaction (50%), sexual functioning (46%), and depression (37%). Table 4 shows the predictability of the nodes for this network.

Discussion

This study applied network analysis to explore the complex interrelationships between psychological flexibility processes, selected newer psychosocial factors, and vulvodynia outcomes. The investigated psychosocial factors were significantly related to pain and sex-related outcomes across three networks. Perceived injustice, depression, pain acceptance, and body-exposure anxiety were highly interconnected within networks. Differences emerged among vulvodynia subtypes; this could have implications for theory and the design of tailored treatments.

Comparison of Networks

The mixed/spontaneous and total sample networks were very similar, likely because there were more women with

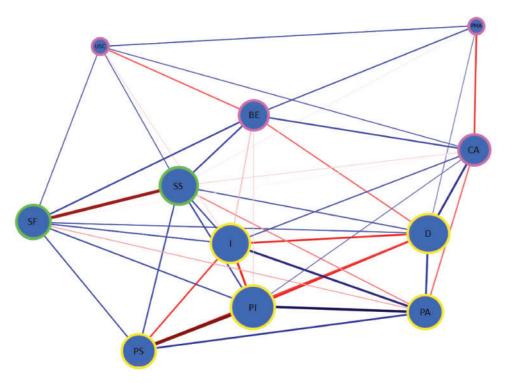


Figure 1. Network model of the total sample (mixed, spontaneous, and provoked vulvodynia). Red edges indicate positive relationships; blue edges indicate negative relationships. BE= body-exposure anxiety/avoidance during intercourse; CA= committed action; D= depression; I= perceived injustice; PA= pain acceptance; PI= pain interference; PMA= present-moment awareness; PS= pain severity; SF= sexual functioning; SS= sexual satisfaction; USC= unmitigated sexual communion. Nodes of community 1 (unmitigated sexual communion, present-moment awareness, body-exposure anxiety, and committed action) are outlined in pink. Nodes of community 2 (sexual functioning and satisfaction) are outlined in green. Nodes of community 3 (depression, perceived injustice, pain acceptance, pain interference, and pain severity) are outlined in yellow.

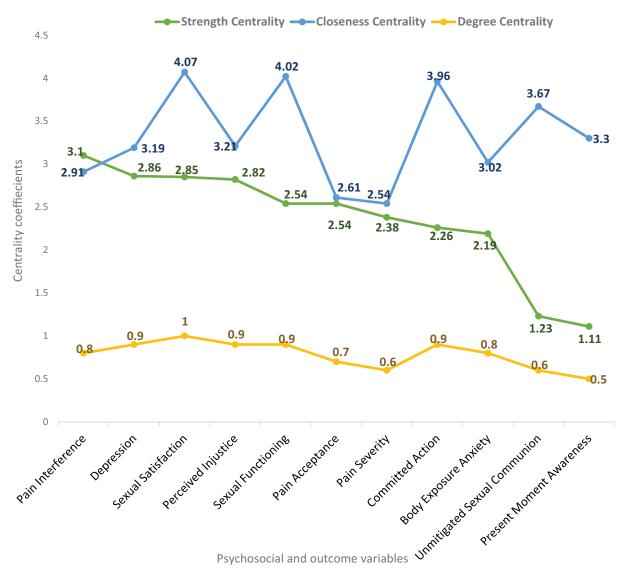


Figure 2. Centrality measures of nodes for the total sample network (n = 349).

mixed than provoked or spontaneous pain overall. However, in an analysis focusing on the networks for mixed/spontaneous and provoked vulvodynia, important similarities and differences were present. Perceived injustice, depression, and pain acceptance were important psychosocial factors in both models. Although injustice in vulvodynia has been researched only once in relation to PVD [15], the findings are consistent with studies highlighting the adverse impact of injustice in people with chronic pain [15, 50–53]. Greater perceptions of the condition's being unfair may contribute to a view of living with this condition as uncontrollable and to a sense of helplessness [51, 54].

The role of injustice within the network may reflect the nature of pain experienced by women with vulvodynia. Notably, in the present study, mean scores of injustice were higher than earlier work in a mixed sample of people with pain after whiplash injury [50]. Past research in vulvodynia has shown a significant diagnostic delay [4, 6], and patients reported experiences of pain invalidation and being told that their pain was all "in their head" [15, 24]. Consistently, previous studies have also highlighted that women, in general, are more likely to experience pain invalidation and stereotyping [55–57]. These experiences may contribute to a greater sense of unfairness and a feeling that others do not take their condition seriously, which are components of injustice [38]. This is consistent with conceptualizations of pain as a social experience that threatens our social needs: the need for fairness and justice [58].

Aside from injustice, the role of depression within the network is consistent with studies in vulvodynia and the wider pain literature [12, 18, 59]. Prospective data have shown that depression is a risk factor for vulvodynia and is related to symptom exacerbation [12, 13, 59]. Notably, depression was a relatively more central node in the mixed/spontaneous model than in the provoked model, and it had strong connections to pain outcomes

Table 4. Predictability (R^2) of nodes for each network

Nodes	Network 1 Total Sample (Ranking)	Network 2 Provoked (Ranking)	Network 3 Mixed/Spontaneous (Ranking)
Pain interference	64% (1)	57% (3)	61% (1)
Pain severity	53% (2)	38% (6)	53% (2)
Sexual satisfaction	50% (3)	66% (1)	50% (3)
Sexual functioning	48% (4)	61% (2)	46% (4)
Pain acceptance	42% (5)	45% (4)	36% (6)
Depression	39% (6)	31% (8)	37% (5)
Committed action	36% (7)	30% (9)	36% (6)
Perceived injustice	35% (8)	35% (7)	33% (7)
Body-exposure anxiety	30% (9)	43% (5)	25% (8)
Present-moment awareness	18% (10)	13% (11)	19% (9)
Unmitigated sexual communion	15% (11)	14% (10)	17% (10)

only in the mixed/spontaneous vulvodynia subtype. Such findings suggest that feelings of loss and distress may be more salient for women who also experience spontaneous forms of vulval pain. This may be because experiences of spontaneous pain are likely to interfere with more aspects of life, resulting in considerably more impact.

In addition to depression, pain acceptance played an important role in both networks. These results are consistent with one previous study on PVD [22] and numerous studies in other chronic pain conditions, where lower pain acceptance was related to higher pain interference and intensity and lower mood [22, 60, 61]. In

vulvodynia, lower pain acceptance may decrease women's motivation to engage in intercourse and may be associated with an unhelpful focus on pain and its unpleasant characteristics; this could result in more pain and worse sexual outcomes. Because more than 80% of women with PVD engage in sex despite the pain [26], acceptance is a potentially important target.

The most notable difference between the networks was the role of body-exposure anxiety. Body-exposure anxiety was the most important node of the network for the provoked subtype, whereas it was relatively less important in the network for the mixed/spontaneous

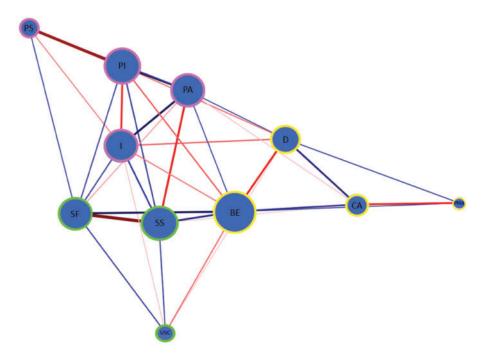


Figure 3. Network model of provoked vulvodynia. Red edges indicate positive relationships; blue edges indicate negative relationships. BE= body-exposure anxiety/avoidance during intercourse; CA= committed action; D= depression; l= perceived injustice; PA= pain acceptance; PI= pain interference; PMA= present-moment awareness; PS= pain severity; SF= sexual functioning; SS= sexual satisfaction; USC= unmitigated sexual communion. Nodes of community 1 (unmitigated sexual communion, present-moment awareness, body-exposure anxiety, and committed action) are outlined in pink. Nodes of community 2 (sexual functioning and satisfaction) are outlined in green. Nodes of community 3 (depression, perceived injustice, pain acceptance, pain interference, and pain severity) are outlined in yellow.

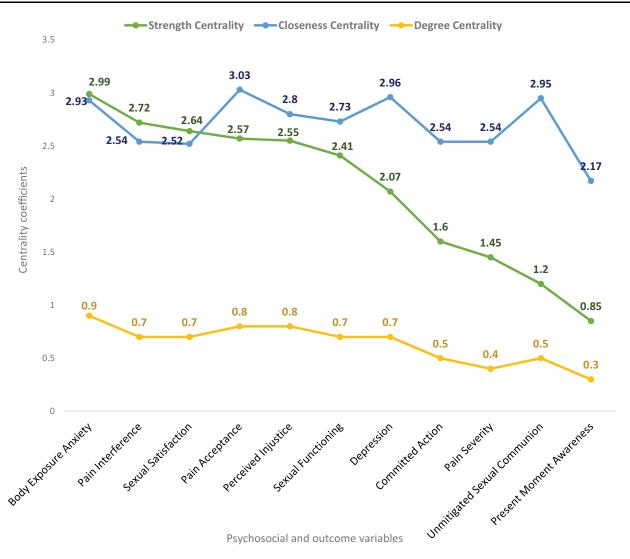


Figure 4. Centrality measures of nodes in the network of provoked vulvodynia (n = 112).

subtype. These findings suggest that avoidance-promoting processes relating specifically to intercourse and body image are relevant to consider in PVD. This difference between networks makes sense, given the intercourse-specific aspect of PVD compared with women who also have spontaneous vulvodynia. Body-exposure anxiety has been explored in PVD in only one previous study [25]. This is consistent with body image research in other pain conditions [62, 63] and with qualitative findings showing that women with PVD perceive themselves to be "unattractive" [64], which highlights how vulvodynia profoundly affects a woman's body perception.

Theoretical and Clinical Implications

The present findings suggest several clinical implications. Because pain severity, pain interference, sexual functioning, and sexual satisfaction appear as highly central nodes, these appear as important outcomes in this population. Furthermore, perceived injustice, pain acceptance, and depression may be important treatment targets in

vulvodynia, regardless of subtype. This is in addition to other well-established psychological targets, such as pain catastrophizing, self-efficacy, and general anxiety, as demonstrated in other studies [14, 16-18]. Given the high scores of perceived injustice, future research should explore processes that are contributing to this, such as invalidating communication within relationships or with health care professionals. All three facets of the psychological flexibility model were significantly related to outcomes across all networks, suggesting that psychological flexibility represents a potentially relevant framework in vulvodynia. However, the networks indicate a much more important role for pain acceptance and committed action than for present-moment awareness, suggesting that these may be key areas to focus on in future studies for people with vulvodynia. Of course, this finding requires replication.

In addition to treatment development that focuses on fostering psychological flexibility, the present data suggest that a specific focus on injustice and on bodyexposure anxiety in PVD may be needed. The fact that

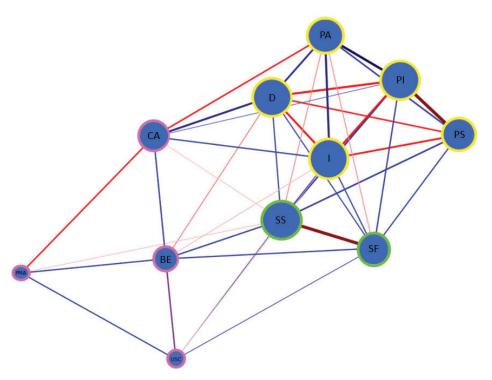


Figure 5. Network model of mixed/spontaneous vulvodynia. Red edges indicate positive relationships; blue edges indicate negative relationships. BE= body-exposure anxiety/avoidance during intercourse; CA= committed action; D= depression; l= perceived injustice; PA= pain acceptance; Pl= pain interference; PMA= present-moment awareness; PS= pain severity; SF= sexual functioning; SS= sexual satisfaction; USC= unmitigated sexual communion. Nodes of community 1 (unmitigated sexual communion, present-moment awareness, body-exposure anxiety, and committed action) are outlined in pink. Nodes of community 2 (sexual functioning and satisfaction) are outlined in green. Nodes of community 3 (depression, perceived injustice, pain acceptance, pain interference, and pain severity) are outlined in yellow.

content-specific variables like injustice and bodyexposure anxiety were in "communities" with psychological flexibility variables in the networks points to the potential utility of tailoring within process-based treatments, such as Acceptance and Commitment Therapy [23]. This might involve orienting treatment participants to contexts of particular relevance, such as injustice experiences, noticing the responses coordinated by these experiences, and then practicing more flexible response options in these situations. For instance, if injustice experiences are responded to with behaviors that interfere with valued activities, treatment might first help a person to notice this pattern, take a perspective that is less influenced by injustice, and actively pursue what matters to them.

A meta-analysis of randomized controlled trials of psychological interventions for vaginal pain, including vulvodynia, found that effect sizes were comparable across vaginal pain conditions, indicating that presumed etiology may not help select treatment [65]. However, that meta-analysis did not specifically compare women with PVD and those with spontaneous/mixed pain. Although randomized controlled trials are considered the gold standard for evaluating treatments [66], group averages have the limitation of overlooking individual differences [28, 66, 67], and there are limitations in applying

findings and models that are based on nomothetic methods to the individual case. In contrast, the present findings suggest that as differences across subtypes are present, treatment tailoring may lead to greater improvements. A within-person, idiographic approach, such as single-case experimental designs, may help to further characterize the complexity of psychosocial processes and challenges occurring across the vulvodynia spectrum.

Limitations

Several limitations warrant consideration. This study was cross-sectional, and causal inferences cannot be made. In the future, longitudinal networks may disentangle the direction of effects while maintaining the advantages of network analysis. Secondly, the analyses of network differences between subtypes include judgments of differences in centrality and predictability and not tests of statistical significance. Such statistical tests in networks remain in the early stages of development. Third, the measures explored were self-reported. Limitations are inherent in the subjective nature of self-reported measures. However, given the inherently subjective nature of the pain, sexual, and psychosocial experiences, use of self-report measures is probably necessary and appropriate in this context. Fourth, clinical information was self-reported and therefore could not be confirmed with medical records.

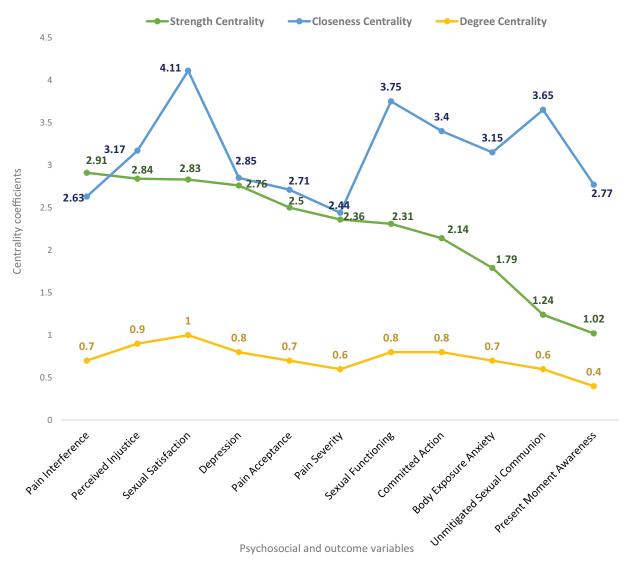


Figure 6. Centrality measures of nodes in the network of mixed/spontaneous vulvodynia (n = 237).

Furthermore, there were more women with mixed vulvodynia than with other subtypes in the total sample. Thus, the lack of differences between the mixed/spontaneous and total sample networks should be interpreted carefully given this uneven split. Fifth, although certain variables appeared to be "central" in the networks, this does not mean they are the most important of all the possible psychosocial variables studied in this population, and there are other more widely researched variables in PVD [16–18] that were not included. However, our focus was on the psychological flexibility model and more recent psychosocial factors to expand the existing understanding of vulvodynia. Finally, the sample was particularly narrow in representation in that it consisted of predominantly white participants.

Conclusion

This study applied network analyses to vulvodynia and its subtypes. Among the psychosocial factors investigated, perceived injustice, depression, and pain acceptance appeared important. Depression appeared to play a more prominent role in women with mixed/spontaneous vulvo-dynia, whereas body-exposure anxiety was relatively more influential within the provoked model. Facets of psychological flexibility played a role across all networks, suggesting the potential utility of the psychological flexibility model. Tailoring may yield added benefits during the development of psychosocial treatments for vulvodynia and its subtypes, as different psychosocial challenges may influence key outcomes for different people.

Authors' Contributions

All authors have significantly contributed to this research study.

Acknowledgments

We thank the Vulval Pain Society and all the women who took part in this research.

References

- Bornstein J, Preti M, Radici G, Stockdale CK, Vieira-Baptista P. Vulvodynia: A neglected chronic pain diagnosis. Pain 2019;160 (7):1680-1.
- Bornstein J, Goldstein AT, Stockdale CK, et al.; International Pelvic Pain Society (IPPS). 2015 ISSVD, ISSWSH, and IPPS consensus terminology and classification of persistent vulvar pain and vulvodynia. J Sex Med 2016;13(4):607–12.
- 3. Wesselmann U, Bonham A, Foster D. Vulvodynia: Current state of the biological science. Pain 2014:155(9):1696–701.
- Harlow BL, Stewart EG. A population-based assessment of chronic unexplained vulvar pain: Have we underestimated the prevalence of vulvodynia? J Am Med Women Assoc 2003;58 (2):82.
- Shallcross R, Dickson JM, Nunns D, Mackenzie C, Kiemle G. Women's subjective experiences of living with vulvodynia: A systematic review and meta-ethnography. Arch Sex Behav 2018;47 (3):577–95.
- Xie Y, Shi L, Xiong X, et al. Economic burden and quality of life of vulvodynia in the United States. Curr Med Res Opin 2012;28 (4):601–8.
- Chalmers KJ, Madden VJ, Hutchinson MR, Moseley GL. Local and systemic inflammation in localized, provoked vestibulodynia. Obstet Gynecol 2016;128(2):337–47.
- Klann AM, Rosenberg J, Wang T, Parker SE, Harlow BL. A systematic review of drug treatment of vulvodynia: Evidence of a strong placebo effect. BJOG 2018;125(10):1216–24.
- Chisari C, Chilcot J. The experience of pain severity and pain interference in vulvodynia patients: The role of cognitive-behavioural factors, psychological distress and fatigue. J Psychosom Res 2017;93:83–9.
- Reed BD, Haefner HK, Punch MR, et al. Psychosocial and sexual functioning in women with vulvodynia and chronic pelvic pain. A comparative evaluation. J Reprod Med 2000;45(8):624–32.
- Santerre-Baillargeon M, Rosen NO, Steben M, et al. Does self-compassion benefit couples coping with vulvodynia?
 Associations with psychological, sexual, and relationship adjustment. Clin J Pain 2018;34(7):629–37.
- Khandker M, Brady SS, Vitonis AF, et al. The influence of depression and anxiety on risk of adult onset vulvodynia. J Womens Health 2011;20(10):1445–51.
- 13. Chisari C, Monajemi MB, Scott W, Moss-Morris R, McCracken LM. Psychosocial factors associated with pain and sexual function in women with vulvodynia: A systematic review. Eur J Pain 2021;25(1):39–50.
- 14. Benoit-Piau J, Bergeron S, Brassard A, et al. Fear-avoidance and pelvic floor muscle function are associated with pain intensity in women with vulvodynia. Clinical J Pain 2018;34(9):804–10.
- 15. Pâquet M, Bois K, Rosen NO, et al. Why us? Perceived injustice is associated with more sexual and psychological distress in couples coping with genito-pelvic pain. J Sex Med 2016;13 (1):79–87.
- 16. Rosen NO, Bergeron S, Pukall CF. Recommendations for the study of vulvar pain in women, part 1: Review of assessment tools. J Sex Med 2020;17(2):180–94.
- 17. Ekdahl J, Flink I, Engman L, Linton SJ. Vulvovaginal pain from a fear-avoidance perspective: A prospective study among female university students in Sweden. Int J Sex Health 2018;30 (1):49–59.
- Govind V, Krapf JM, Mitchell L, et al. Exploring pain-related anxiety and depression in female patients with provoked vulvodynia with associated overactive pelvic floor muscle dysfunction. Sex Med 2020;8(3):517–24.

19. Hayes SC, Luoma JB, Bond FW, Masuda A, Lillis J. Acceptance and commitment therapy: Model, processes and outcomes. Behav Res Ther 2006;44(1):1–25.

- McCracken LM. Learning to live with the pain: Acceptance of pain predicts adjustment in persons with chronic pain. Pain 1998;74(1):21–7.
- McCracken LM, Vowles KE, Eccleston C. Acceptance of chronic pain: Component analysis and a revised assessment method. Pain 2004;107(1-2):159–66.
- Boerner KE, Rosen NO. Acceptance of vulvovaginal pain in women with provoked vestibulodynia and their partners: Associations with pain, psychological, and sexual adjustment. J Sex Med 2015;12(6):1450–62.
- Hann KE, McCracken LM. A systematic review of randomised controlled trials of Acceptance and Commitment Therapy for adults with chronic pain: Outcome domains, design quality, and efficacy. J Context Behav Sci 2014;3(4):217–27.
- Nguyen RH, Ecklund AM, MacLehose RF, Veasley C, Harlow BL. Comorbid pain conditions and feelings of invalidation and isolation among women with vulvodynia. Psychol Health Med 2012;17(5):589–98.
- Maillé DL, Bergeron S, Lambert B. Body image in women with primary and secondary provoked vestibulodynia: A controlled study. J Sex Med 2015;12(2):505–15.
- Reed BD, Harlow SD, Sen A, et al. Prevalence and demographic characteristics of vulvodynia in a population-based sample. Am J Obstet Gynecol 2012;206(2):170-e1–170.e9.
- 27. Muise A, Bergeron S, Impett EA, Rosen NO. The costs and benefits of sexual communal motivation for couples coping with vulvodynia. Health Psychol 2017;36(8):819–27.
- 28. Hayes SC, Hofmann SG, Stanton CE, et al. The role of the individual in the coming era of process-based therapy. Behav Res Ther 2019;117:40–53.
- Contreras A, Nieto I, Valiente C, Espinosa R, Vazquez C. The study of psychopathology from the network analysis perspective:

 A systematic review. Psychotherap Psychosom 2019;88
 (2):71–83.
- Cramer AO, Waldorp LJ, Van Der Maas HL, Borsboom D. Complex realities require complex theories: Refining and extending the network approach to mental disorders. Behav Brain Sci 2010;33(2-3):178–93.
- 31. Fish RA, McGuire B, Hogan M, Morrison TG, Stewart I. Validation of the Chronic Pain Acceptance Questionnaire (CPAQ) in an Internet sample and development and preliminary validation of the CPAQ-8. Pain 2010;149(3):435–43.
- 32. Reneman MF, Dijkstra A, Geertzen JH, Dijkstra PU. Psychometric properties of chronic pain acceptance questionnaires: A systematic review. Eur J Pain 2010;14(5):457–65.
- Pierce BG, Levin ME. Preliminary validation and reliability assessment of a 10-item Tacting of Function Scale. J Context Behav 2019;12:322–8.
- McCracken LM, Chilcot J, Norton S. Further development in the assessment of psychological flexibility: A shortened Committed Action Questionnaire (CAQ-8.). Eur J Pain 2015;19 (5):677–85.
- 35. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: Validity of a brief depression severity measure. J Gen Intern Med 2001;16 (9):606-13.
- 36. Choi Y, Mayer TG, Williams MJ, Gatchel RJ. What is the best screening test for depression in chronic spinal pain patients? Spine J 2014;14(7):1175–82.
- 37. Cash TF, Maikkula CL, Yamamiya Y. "Baring the body in the bedroom": Body image, sexual self-schemas, and sexual functioning among college women and men. Electron J Hum Sex 2004 2004;29(7):1–9.

- Sullivan MJ, Adams H, Horan S, et al. The role of perceived injustice in the experience of chronic pain and disability: Scale development and validation. J Occup Rehabil 2008;18 (3):249–61.
- 39. Cleeland CS, Ryan KM. Pain assessment: Global use of the Brief Pain Inventory. Ann Acad Med Singap 1994;23(2):129–38.
- 40. Rosen C, Brown JH, Leiblum S, et al. The Female Sexual Function Index (FSFI): A multidimensional self-report instrument for the assessment of female sexual function. J Sex Martial Ther 2000;26(2):191–208.
- 41. Masheb RM, Lozano-Blanco C, Kohorn EI, Minkin MJ, Kerns RD. Assessing sexual function and dyspareunia with the Female Sexual Function Index (FSFI) in women with vulvodynia. J Sex Martial Ther 2004;30(5):315–24.
- 42. Lawrance KA, Byers ES, Cohen JN. Interpersonal exchange model of sexual satisfaction questionnaire. In: Fisher TD, Davis CM, Yarber WL, Davis S, eds. Handbook of sexuality-related measures: A compendium. New York, NY: Routledge; 1998:525–30.
- 43. Jodoin M, Bergeron S, Khalifé S, et al. Male partners of women with provoked vestibulodynia: Attributions for pain and their implications for dyadic adjustment, sexual satisfaction, and psychological distress. J Sex Med 2008;5(12):2862–70.
- 44. Rancourt KM, Rosen NO, Bergeron S, Nealis LJ. Talking about sex when sex is painful: Dyadic sexual communication is associated with women's pain, and couples' sexual and psychological outcomes in provoked vestibulodynia. Arch Sex Behav 2016;45(8):1933–44.
- Hagberg A, Swart P, Chult, SD. Exploring Network Structure, Dynamics, and Function Using NetworkX. Los Alamos, NM: Los Alamos National Lab; 2008.
- Cohen J. Statistical Power Analysis for the Behavioral Sciences. Hillsdale, NJ: Lawrence Erlbaum Associates; 1988.
- 47. Fruchterman TM, Reingold EM. Graph drawing by force-directed placement. Softw Pract Exp 1991;21(11):1129–64.
- 48. Hunter JD. Matplotlib: A 2D graphics environment. Comput Sci Eng 2007;9(3):90–5.
- Koschützki D, Lehmann KA, Peeters L, et al. Centrality indices.
 In: Brandes U, Erlebach T, eds. Network Analysis. Berlin: Springer; 2005:16–61. doi:10.1007/978-3-540-31955-9_3.
- Sullivan MJ, Adams H, Martel MO, Scott W, Wideman T. Catastrophizing and perceived injustice: Risk factors for the transition to chronicity after whiplash injury. Spine 2011;36(Suppl 25):S244–9.
- 51. Margiotta F, Hannigan A, Imran A, Harmon DC. Pain, perceived injustice, and pain catastrophising in chronic pain patients in Ireland. Pain Pract 2017;17(5):663–8.
- Miller MM, Scott EL, Trost Z, Hirsh AT. Perceived injustice is associated with pain and functional outcomes in children and adolescents with chronic pain: A preliminary examination. J Pain 2016;17(11):1217–26.

- 53. Sullivan MJ, Davidson N, Garfinkel B, Siriapaipant N, Scott W. Perceived injustice is associated with heightened pain behavior and disability in individuals with whiplash injuries. Psychol Inj Law 2009;2(3-4):238–47.
- 54. Martel ME, Dionne F, Scott W. The mediating role of pain acceptance in the relation between perceived injustice and chronic pain outcomes in a community sample. Clin J Pain 2017;33 (6):509–16.
- 55. Samulowitz A, Gremyr I, Eriksson E, Hensing G. "Brave men" and "emotional women": A theory-guided literature review on gender bias in health care and gendered norms towards patients with chronic pain. Pain Res Manag 2018;2018:6358624.
- Nguyen RH, Turner RM, Rydell SA, MacLehose RF, Harlow BL. Perceived stereotyping and seeking care for chronic vulvar pain. Pain Med 2013;14(10):1461–7.
- Braksmajer A. Struggles for medical legitimacy among women experiencing sexual pain: A qualitative study. Women Health 2018;58(4):419–33.
- Karos K, Williams AC, Meulders A, Vlaeyen JW. Pain as a threat to the social self: A motivational account. Pain 2018;159 (9):1690-5.
- Bair MJ, Robinson RL, Katon W, Kroenke K. Depression and pain comorbidity: A literature review. Arch Intern Med 2003; 163(20):2433–45.
- Kratz AL, Davis MC, Zautra AJ. Pain acceptance moderates the relation between pain and negative affect in female osteoarthritis and fibromyalgia patients. Ann Behav Med 2007;33 (3):291–301.
- 61. Vowles KE, McCracken LM, Eccleston C. Processes of change in treatment for chronic pain: The contributions of pain, acceptance, and catastrophising. Eur J Pain 2007;11(7):779–87.
- Moseley GL. I can't find it! Distorted body image and tactile dysfunction in patients with chronic back pain. Pain 2008;140 (1):239–43.
- 63. Lotze M, Moseley GL. Role of distorted body image in pain. Curr Reumat Rep 2007;9(6):488–96.
- 64. Ayling K, Ussher JM. "If sex hurts, am I still a woman?" The subjective experience of vulvodynia in hetero-sexual women. Arch Sex Behav 2008;37(2):294–304.
- 65. Flanagan E, Herron KA, O'Driscoll C, de C Williams A. Psychological treatment for vaginal pain: Does etiology matter? A systematic review and meta-analysis. J Sex Med 2015;12 (1):3–16. doi:10.1111/jsm.12717.
- 66. Fisher AJ, Medaglia JD, Jeronimus BF. Lack of group-to-individual generalizability is a threat to human subjects research. Proc Natl Acad Sci U S A 2018;115(27):E6106–15. https://doi.org/10.1073/pnas.1711978115.
- 67. Fisher AJ, Reeves JW, Lawyer G, Medaglia JD, Rubel JA. Exploring the idiographic dynamics of mood and anxiety via network analysis. J Abnorm Psychol 2017;126(8):1044–56.