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Persistent Genitopelvic Pain: Classification, Comorbidities, Chronicity, and Interpersonal Factors

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Abstract

Purpose of Review To review recently published literature on vulvodynia classification, comorbidities, chronicity, and interpersonal factors, and to outline critical gaps between research findings and clinical practice.

Recent Findings The classification of vulvodynia varies in terms of its specificity and degree of focus on pain and psychosocial correlates, with recent conceptualizations supporting a biopsychosocial framework. The presence of comorbidities in those with vulvodynia is associated with poorer symptom outcomes and psychosocial wellbeing, as well as longer-term symptom presentation. Longitudinal research has indicated that the course of vulvodynia can vary, with some women experiencing complete pain remission, pain remission with relapse, or pain persistence. There has been a significant increase in psychosocial research in vulvodynia, with much attention on the role of interpersonal factors in the experience of pain and in sexual and relationship satisfaction. Partner dynamics and sexual motivation have figured prominently in this area of research.

Summary The field of vulvodynia research has grown exponentially over the past few years, with an increase in attention to factors and issues outside pathophysiological processes in the vulva. Despite empirically-based recommendations for treatment options, treatment algorithms remain inconsistent and are based on poorly supported treatment options.

Keywords Genitopelvic pain · Vulvodynia · Classification · Comorbidities · Psychosocial factors · Interpersonal relationships

Introduction

The field of persistent genitopelvic pain, specifically vulvodynia, has evolved greatly over the past few years. Vulvodynia, defined as idiopathic chronic vulvar pain, has a prevalence of about 7% [1, 2]. Potential risk factors for the development of vulvodynia include yeast [3] and urinary tract [1] infections, a history of depression and anxiety [4], comorbid chronic pain conditions [5, 6], and the experience of early chronic stress [7]. Much recent attention in the field of vulvodynia has been paid to its classification, comorbidities, and chronicity, as well as its interactive effects within relationships. This review will focus on recently published articles in these domains.

Classification of Vulvodynia

Decades of research into the centrality of the pain component of vulvodynia have radically changed its original conceptualization

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as a psychosexual condition to that of a chronic pain condition that affects the individual (and their romantic partner) in many ways, including sexually, psychosocially, and physiologically. Consequent changes in clinical practice include a full assessment of the pain of vulvodynia and its correlates, resulting in a more efficient diagnostic process and increased streamlined and empirically-based treatment planning [8••]. In addition, multiple classifications of vulvodynia exist, allowing flexibility in its diagnosis and reflecting the biopsychosocial nature of the pain. The most commonly used classification systems for genital pain, the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) [9] and the International Classification of Diseases (ICD-11) [10], will be reviewed in this section, and a recently published multi-societal classification of chronic vulvar pain and vulvodynia will be introduced.

The DSM-5 introduced new terminology, Genitopelvic Pain/Penetration Disorder (GPPPD), to replace the DSM-IV-TR [11] category of Sexual Pain Disorders, which consisted of dyspareunia and vaginismus. Part of the rationale for this change was to help address the overlapping symptoms/characteristics (e.g., heightened muscle tension) of dyspareunia and vaginismus that sometimes made their differential diagnosis difficult. Although the category of GPPPD has been criticized for being so broad as to preclude specific vulvodynia-related diagnoses [12], its focus on pain, difficulties with sexual activities involving vaginal penetration, fear and anxiety in response to the (anticipated) pain, and muscle tension, closely reflect the empirically-based correlates of vulvodynia and related conditions.

It is important to note that GPPPD is included in the Sexual Dysfunctions chapter of the DSM-5 and not in the chapter specific to pain conditions. This decision likely reflects the breadth of specialists (e.g., those in sexual medicine, psychology, pelvic floor physical therapy)—not just pain specialists—involved in its assessment and treatment, as well as the varied recommended treatment options that target the pain as well as the psychosocial components of vulvodynia [8••]. Indeed, the definition of GPPPD represents a strong attempt to align with the results of numerous research studies supporting the conceptualization and treatment of vulvodynia as a biopsychosocial condition [13–16].

The most recent edition of the ICD-11 Classification of Chronic Pain [17] includes vulvodynia within the category of Chronic Primary Visceral Pain [18]. In addition, the ICD-11 includes vulvodynia and related conditions in two separate sections, likely reflecting the input of pain *and* sexual medicine specialists in the development of the ICD-11: (1) Pain Related to the Vulva, Vagina, or Pelvic Floor and (2) Conditions Related to Sexual Health, in which vulvodynia is included as a sexual pain disorder. The classification of vulvodynia as a vulvovaginal/pelvic pain condition and/or as a sexual pain disorder in the ICD-11 allows providers in different specialties to choose which diagnosis best fits their clients/patients and likely has implications for treatment options; for example, classifying it as pain related to the vulva may reflect a localized symptom presentation that may benefit from localized treatment (e.g., vestibulectomy), whereas classifying vulvar pain as a sexual pain disorder may emphasize the pain and its sexual/interpersonal consequences, which may be best addressed via psychotherapy. The presence of both of these diagnoses within a single classification system supports the importance of the pain *and* psychosocial (e.g., sexual and interpersonal) aspects of vulvar pain and vulvodynia.

Recently, an empirically based classification of chronic vulvar pain and vulvodynia was published and approved by two sexual health and one pain organization [19••, 20•]. This classification reflects the clinical reality of how one reaches a diagnosis of vulvodynia: diagnosing vulvodynia in the absence of known factors that can lead to chronic vulvar pain, and basing vulvodynia subtypes (e.g., provoked and localized) on pain characteristics (e.g., location). The potential associated factors—comorbidities and other pain syndromes, genetics, hormonal factors, inflammation, musculoskeletal, neurologic mechanisms, structural defects, and psychosocial factors—are empirically-based given that they reflect factors that are known to contribute to the development and/or maintenance of vulvodynia. This classification supports the centrality of the pain in vulvodynia and chronic vulvar pain conditions and is also aligned with research findings and clinical practice, like the DSM-5 and ICD-11 systems.

Related to the classification of persistent genitopelvic pain is recent attention to, and attempts to classify, the experience of persistent, distressing genitopelvic sensations other than pain. An excellent example of a persistent, distressing genitopelvic condition involving a primary sensory complaint other than pain is Persistent Genital Arousal Disorder (PGAD) [21•]. PGAD is characterized by persistent, unwanted sensations of genital arousal (i.e., genital sensitivity and/or swelling) that occur in the absence of sexual desire [22]. It is estimated to affect approximately 1% of individuals [23]. Interestingly, there appears to be some overlap between PGAD and vulvodynia on some characteristics; for example, those with PGAD often also experience other comorbid genitopelvic pain conditions (e.g., interstitial cystitis and irritable bowel syndrome, see below for more information on comorbidities). In addition, despite the primary complaint of unwanted arousal in PGAD, almost half (44.3%) of individuals with PGAD describe their arousal symptoms as painful [24]. This finding is fascinating because there is much overlap between many terms used to describe pain and arousal (e.g., throbbing, aching); however, the relationship between arousal and pain is not yet well understood. Pukall and colleagues [21•] have proposed an overarching framework of "genitopelvic dysesthesias" which encompasses genitopelvic pain (e.g., vulvodynia, chronic vulvar pain), arousal (e.g., PGAD), pain and arousal (e.g., PGAD with pain), as well as other unpleasant

sensations (e.g., itching, numbness). Although this framework has yet to be formally recognized and such a process will take many years and much agreement among different societies and organizations, it may prove useful in streamlining the existing diverse classifications of conditions involving unpleasant genitopelvic sensations. As such, it may also help to increase clinical and research attention to other kinds of unpleasant genitopelvic sensations—promoting more efficient and effective identification and treatment of these distressing conditions. More consideration is needed to understand where and how PGAD should be classified within existing systems; however, its conceptualization and treatment options would benefit from a biopsychosocial perspective—similar to that applied to persistent genital pain.

Comorbidities With and Chronicity of Vulvodynia

Women with vulvodynia often report comorbid pain conditions (e.g., fibromyalgia, interstitial cystitis/painful bladder syndrome, irritable bowel syndrome) [5, 6]. Nguyen and colleagues [5] found that 45% of women with vulvodynia reported one or more comorbid pain conditions, and Reed and colleagues [25] found that women with vulvodynia are 2.3 to 3.4 times more likely to experience comorbid chronic pain. This relationship is bidirectional; women with fibromyalgia, interstitial cystitis, and irritable bowel syndrome are also 2.3 to 3.3 times more likely to experience vulvodynia than women without these conditions [25, 26]. The presence of additional comorbidities is also associated with poorer symptom outcomes and psychosocial wellbeing.

Women with vulvodynia and comorbid pain conditions (as compared to those with few/no comorbidities) report significantly greater feelings of invalidation and isolation, more severe vulvar pain symptoms (e.g., longer duration of symptoms, greater pain ratings), greater interference with daily activities, more visits to healthcare providers, and greater depressive and anxiety symptoms [27–29]. Using a cluster analysis to characterize subgroups of vulvodynia, Reed, Plegue, Williams, and Sen [30] found that the presence of other comorbid pain conditions was associated with poorer general health, the presence of psychiatric disorders, and greater vulvar pain. The presence of comorbid pain conditions, in addition to other factors such as pain severity and the presence of anxiety, has also been associated with the chronicity of vulvodynia.

Recent longitudinal research has indicated that the course of vulvodynia is not necessarily predictably chronic for all sufferers. Reed et al. [31] found that half of their sample experienced symptom remission within 6–30 months; however, those who experienced a relapse or whose pain persisted past 30 months reported higher pain severity, a longer duration of symptoms, and were more likely to experience comorbid fibromyalgia than those whose pain remitted. Furthermore, a 7-year study reported that those with persistent pain were more likely than those with a decreasing pain pattern to be of older age at pain onset, have pain at a location other than the vaginal entrance, and experience more anxiety [32]. In addition, another study indicated that those with primary vulvodynia (vulvar pain that started at first vaginal penetration) were less likely to experience remission than those with secondary vulvodynia (vulvar pain that started after a period of pain-free activities involving vaginal penetration) [33]. Taken together, the results of these studies emphasize the heterogeneity of vulvodynia in terms of the pattern of chronicity and the presence of various subgroups (e.g., primary versus secondary, with or without comorbid pain conditions or anxiety). Acknowledging this heterogeneity, future research may seek to develop and evaluate treatment protocols specific to different presentations of vulvodynia. For example, women who experience widespread pain and/or multiple comorbidities in addition to their vulvar pain may benefit from a different approach from those who experience localized vulvar pain.

Psychosocial Factors

In the past few years, there has been a large amount of high-quality research on pathophysiological contributors to vulvodynia, including microbial [34, 35], inflammatory [36–41], genetic [42, 43], hormonal [44], pelvic floor [45–50], central/neural [51–53], and environmental [54] factors. There has also been an impressive, perhaps unprecedented, amount of recent research investigating the role of psychosocial factors in the expression of vulvodynia. This section will focus on cutting-edge research on psychosocial factors, specifically, relationship factors—including partner dynamics and sexual motivation.

Partner Dynamics

The Interpersonal Emotion Regulation Model of women's sexual dysfunction posits that partner dynamics play a key role in the onset and maintenance of genitopelvic pain [55••]. This conceptual framework suggests that distal and proximal interpersonal factors modulate couples' emotion regulation, and consequently, women's pain and both partners' sexual and psychological adjustment. Emotion regulation is proposed as a mediating pathway given the threat value of this type of pain and the negative affect it generates.

Intimacy and romantic attachment are two distal factors that have received recent empirical attention. Intimacy, defined as a recursive process involving empathic responses and self-disclosure [56], was studied using dyadic cross-sectional, self-report and laboratory observation designs. Findings indicated that both partners' observed and self-

reported empathic responses were associated with their better sexuality outcomes [57] and greater relationship satisfaction [58], whereas both partners' greater perceived self-disclosure was associated with their greater sexual satisfaction [57]. In a laboratory-based study of 42 heterosexual couples where experimentally-induced vaginal pressure served as a simulation of vaginal sensations during penetration, men were asked to estimate their partner's level of pressure pain. Results showed that the level of congruence between actual and estimated ratings of pain was higher when partners reported higher relationship satisfaction and women perceived their partner to be more responsive—a component of intimacy [59, 60]. Feeling understood, accepted, and cared for in a romantic relationship may contribute to creating trust and safety between partners and facilitate verbal and nonverbal sexual communication about the pain.

Two cross-sectional studies conducted among couples coping with vulvodynia showed that when they reported lower levels of sexual communication, women reported greater pain, and both partners reported lower sexual and relationship satisfaction [61, 62]. Further, results from a dyadic daily diary study indicated that more non-sexual physical affection among partners was linked to higher sexual and relationship satisfaction, as well as to greater sexual function (e.g., desire, arousal) [63]. Overall, recent findings suggest that couples who engage in physical and emotional intimacy may be better able to mitigate the negative impact of vulvodynia. Nevertheless, some partners struggle with being close and responsive to one another. Romantic attachment—a second distal factor—may underlie their ability to be comfortable with intimacy in their relationship.

Hazan and Shaver [64] identified two types of adult insecure attachment, along a continuum: anxious attachment, characterized by a fear of abandonment, and avoidant attachment, by a fear of intimacy. These ways of relating to a romantic partner are thought to stem from an individual's childhood interactions with a significant caregiver and would be relatively stable across time. Prior cross-sectional work has shown that insecure attachment was associated with sexuality outcomes in women with vulvodynia (lower sexual satisfaction and sexual function) and their partners (lower sexual satisfaction) [65]. A recent dyadic prospective study conducted among couples coping with a common subtype of vulvodynia, provoked vestibulodynia, indicated that women's higher levels of attachment anxiety and avoidance were associated with their greater genital pain intensity 2 years later. This association was mediated by women's lower levels of pain self-efficacy. Findings suggest that when women are more afraid of losing their partner, in the case of anxious attachment, or have more difficulty asking for support, in the case of avoidant attachment, this may make it more challenging for them to cope with their pain, and ultimately lead to greater pain. Interestingly, a study found that when women with vulvodynia and their partners reported greater anxious attachment, they perceived each other as being less responsive, i.e., less empathic [66]. This finding suggests potential bidirectional associations between attachment and intimacy. Both may also be associated with negative childhood experiences.

Indeed, women with vulvodynia report higher levels of child maltreatment, i.e., sexual, physical, or psychological abuse, and psychological and physical neglect [4]. A dyadic cross-sectional study further indicated that higher levels of child maltreatment in this population were associated with women's lower sexuality outcomes and greater pain, whereas partners' higher levels of maltreatment were associated with their own lower sexuality and relationship outcomes, as well as women's lower relationship satisfaction [67]. Negative childhood experiences may contribute to some couples' less adaptive responses to the pain [68].

Partner responses to pain are the most studied proximal factor. Solicitous (e.g., expressions of attention and sympathy) and negative (e.g., expressions of hostility, frustration) responses are thought to inhibit adaptive emotional processes, whereas facilitative (e.g., affection and encouragement of adaptive coping) responses are thought to promote them. Findings from dyadic cross-sectional and daily experience studies indicate that greater woman-perceived and partner-reported solicitous and negative responses are associated with women's greater pain and both partners' lower sexual function and satisfaction, in addition to lower relationship satisfaction. Such responses may reinforce avoidance of pain and sex and thus prevent the development of alternative, pain-free sexual activities. Facilitative responses, on the other hand, are associated with positive sexuality and relationship outcomes for both partners, as well as women's lower pain, as they can promote better co-regulation of emotions [55••], in addition to women's greater sexual assertiveness [69].

Other cognitive-affective factors associated with both partners' sexuality and relationship outcomes include self-compassion [70], contingent self-worth [71, 72], perceived injustice [73], and couples' daily anxiety and depressive symptoms [74]. Taken together, the body of evidence on partner dynamics suggests a significant involvement of romantic relationship factors in the experience of vulvodynia and associated sexual difficulties.

Sexual Motivation

Sexual motivation—that is, what drives people to engage in sexual activity—has been integrated into biopsychosocial models of genitopelvic pain given growing evidence of its important influences [55••, 75••]. Interest in sexual motivation has been driven by an apparent paradox in affected women's sexual behavior; although women with vulvodynia are generally avoidant of sexual activity to reduce the pain, over 85% persist with regular penetrative sex [76]. It has been posited theoretically [75••] and

supported by qualitative reports [77] that this paradox may be explained by taking into account other motivational factors that frequently relate to the relationship, such as a desire to please their partner, avoid conflict, and feelings of guilt, or fear of losing the relationship. Such interpersonal goals are especially relevant in the context of vulvodynia, as couple interactions and dyadic coping are highly influential to their distress and pain management [55••].

In a cross-sectional study, women with vulvodynia were less likely to report engaging in sexual activity in pursuit of positive relationship outcomes such as intimacy (i.e., approach goals) and more likely to report sexual activity in order to avoid negative relationship outcomes such as partner disappointment (i.e., avoidance goals), compared with control women [78]. Affected women also reported lower approach and higher avoidance sexual goals compared with their own partners, who did not differ in their goals relative to controls, pointing to the heavier burden that the women carry. Nonetheless, there is the potential for both costs (increased nociceptor sensitization, increased nerve proliferation) [79••] and benefits (e.g., enhanced relational intimacy, pain acceptance) [57, 80] of persisting with painful intercourse, underscoring the importance of establishing the specific impacts of different motivations.

Three aspects of sexual motivation have received recent attention in couples affected by vulvodynia: sexual goals (i.e., the reasons for engaging in sexual activity), sexual communal strength (i.e., the extent to which people are motivated to meet their partner's sexual needs), and unmitigated sexual communion (i.e., being motivated to meet a partner's sexual needs to the exclusion of one's own needs). With regard to sexual goals, in a daily experience study, on days when women with vulvodynia reported higher approach goals, they reported less pain [81]. Moreover, they and their partners attended to more positive thoughts and feelings during sex (e.g., pleasurable sensations, connectedness) and, in turn, both women and partners reported greater sexual functioning and relationship satisfaction. Being more approach-oriented may promote more emotional expression and validation and a more positive interpersonal context for the sexual interaction overall [55••].

In contrast, on days when women reported higher avoidance goals, both they and their partners attended more to negative sexual cues (e.g., signs of disinterest, distress, or pain), and, in turn, women reported greater pain and both partners reported poorer sexual function [81]. Prior studies have linked avoidance goals to more negative pain-related emotions (e.g., pain-related fear) [82]. Thus, holding stronger avoidance goals could promote less adaptive emotion regulation strategies (e.g., avoidance) by sensitizing the couple to focus on the negative interference of pain to their (sexual) lives.

One of the primary reasons that women with vulvodynia report for continuing painful sexual activities is a desire to meet their partners' sexual needs [83–86]. For this reason, Muise and colleagues [87, 88] conducted another daily experience study to examine sexual communal strength and unmitigated sexual communion in this population. They found that on days when participants reported being more motivated to meet a partner's sexual needs (i.e., higher sexual communal strength), both affected women and their partners reported greater sexual functioning, sexual satisfaction and relationship satisfaction, and less anxiety, and women reported lower pain during intercourse. However, on days when women were overly focused on a partner's sexual needs to the detriment of their own needs (i.e., higher unmitigated sexual communion), couples reported poorer sexual, relationship, and psychological well-being, and women experienced more pain. Heightened sexual distress was found to account for the daily associations between women's unmitigated sexual communion and their own greater pain, depression, and anxiety.

In summary, and consistent with Rosen and Bergeron's [55••]) recent Interpersonal Model of Emotion Regulation applied to genitopelvic pain, avoidance sexual goals and unmitigated sexual communion might be risk factors for poor emotion regulation related to sex, whereas approach goals and sexual communal strength may promote more adaptive emotional processing for both members of the couple. In this way, distinct sexual motivations have differential implications for women's vulvar pain and couples' adjustment to the pain.

Conclusions

The field of vulvodynia research has been growing exponentially over the past few years, with a recent emphasis on topics that extend beyond a unitary focus on pathophysiological processes of the vulva. Indeed, there has been a greater focus on classification, comorbidities, chronicity, and the influence of relationship factors on pain and sexual and relationship satisfaction. However, the research findings related to partner dynamics and sexual motivation described in this paper have not been translating to clinical practice as well as one might expect given their evident clinical implications. On a larger scale, despite the publication of explicit, empirically-based recommendations for the treatment of vulvodynia (i.e., psychotherapy focusing on pain reduction and sexuality/relationship factors, pelvic floor physical therapy, vestibulectomy for provoked vestibulodynia) [8••], research has shown that first-line interventions remain predominantly medical (e.g., pharmacological agents), with no consistent treatment approach; only a minority of women received psychotherapy in one study [89•].

More efforts are needed to ensure that new findings reach their intended audiences, including women with vulvodynia and their partners so that they can better advocate for their health and well-being within a multidisciplinary treatment approach, as well as clinicians delivering treatment services and other key decision-makers (e.g., hospital administrators, funding bodies).

Targeted training workshops and book chapters for clinicians, in addition to enhanced accessibility of treatment manuals (e.g., freely available on websites), are two ways to promote the uptake of psychological interventions. Importantly, increasing cross-discipline communication is crucial to the improvement of best practices in the clinic. Empirically based knowledge sharing initiatives aimed at the general public such as the social media campaign #itsnotinyourhead (http://whri.org/itsnotinyourhead/) are another important way to bridge the knowledge-to-action gap, direct attention to up-to-date treatment resources, and enhance communication among all those affected by and involved in caring for women with vulvodynia and their partners.

Compliance with Ethical Standards

Conflict of Interest Sophie Bergeron, Natalie O. Rosen, and Robyn Jackowich each declare no potential conflicts of interest.

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- •• Of major importance

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