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Female Sexual Pain Disorders: a Review of the Literature on Etiology and Treatment

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Abstract Female sexual pain disorders, although highly prevalent and increasingly studied, remain a distressing complaint for women and their partners. Empirical evidence points to a multifactorial conceptualization of the etiology, course, and associated difficulties of sexual pain; thus, treatment options span the medical, pelvic floor rehabilitation psychological and multimodal. Given the interpersonal context in which sexual pain occurs, recent work has underscored the importance of considering the dyadic framework in research and treatment. This review presents understanding from across disciplines focusing on the impact of sexual pain on the woman and the couple, proposed etiologic pathways and risk factors related to its development and course, and current treatment options. Recommendations for research point to an urgency for multidisciplinary exchanges in the development of conceptual models and refinement of targeted interventions.

Keywords Female sexual pain · Genito-pelvic pain/penetration disorder · Dyspareunia · Genital pain · Vulvodynia · Vaginismus

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Introduction

Prevalence and Significance

Chronic pain problems involving the female reproductive system are major health concerns in women of all ages. Despite significant advances in the field, these conditions are still poorly understood, with only 60 % of afflicted women seeking treatment and 52 % of those never receiving a formal diagnosis [1•]. The sexual pain disorders, dyspareunia and vaginismus—now classified in the DSM-5 as a single entity termed genito-pelvic pain/penetration disorder [2]—are thought to affect 14 to 34 % of younger women and 6.5 to 45 % of older women [3]. Current DSM-5 diagnostic criteria for genito-pelvic pain/penetration disorder include persistent or recurrent difficulties with one or more of the following for at least 6 months and resulting in clinically significant distress: (1) vaginal penetration during intercourse; (2) marked vulvovaginal or pelvic pain during vaginal intercourse or penetration attempts; (3) marked fear or anxiety about vulvovaginal or pelvic pain in anticipation of, during, or as a result of vaginal penetration; and (4) marked tensing or tightening of the pelvic floor muscles during attempted vaginal penetration [2]. An overlapping condition, not based on DSM classification, is vulvodynia, or chronic unexplained vulvar pain, which is also associated with pelvic floor muscle

dysfunction and superficial dyspareunia [4•]. A recent population-based study suggests that the prevalence of vulvodynia is 8 % [1•]. Provoked vestibulodynia (PVD)—an acute recurrent pain localized within the vulvar vestibule and experienced primarily during sexual intercourse—is suspected to be the most frequent cause of vulvodynia and dyspareunia in premenopausal women. These common gynecological pain conditions all result in significant sexual, psychological, and relationship impairments, which are as much a source of distress for the patient and her partner as the pain itself.

Conceptual models of sexual pain espouse a multifactorial view, with empirical evidence suggesting the existence of multiple etiologic pathways leading to the development and persistence of the pain and associated psychosexual and relationship difficulties. Initial onset may be triggered by biomedical/mechanical trauma to the genitalia, with ensuing inflammation, pelvic floor muscle dysfunction, and other local changes leading to nociceptor sensitization and further peripheral and central alterations in pain processing [5]. The experience of pain, combined with a lack of proper recognition by health professionals [1•] may interact with individual predispositions to generate varying degrees of distress in patients and their partners. In turn, cognitive, behavioral, affective, and interpersonal factors may modulate the pain experience and associated negative sequelae, as not all women who have an initial experience of pain are at risk of suffering from a persistent condition [6], or of developing sexual dysfunction [5].

This review will outline the negative consequences of sexual pain and its proposed etiologic mechanisms in women, including biomedical, psychological, and relationship factors. Current treatment approaches from medical, physical therapy, psychosocial, and integrated perspectives will be reviewed, and recommendations for future research will be formulated.

Consequences and Associated Difficulties

The experience of sexual pain has wide-reaching consequences for affected women and their partners' psychological, sexual, and relationship well-being. Controlled cross-sectional studies show that women with sexual pain report heightened psychological distress, including anxiety, depressive symptoms, lower self-esteem, and body image concerns [7–12]. A community-based study highlights that disorders of depression and anxiety were significantly more prevalent as consequences of vulvar pain when compared to healthy controls [13•]. Further, up to 45 % of women with vulvodynia report a comorbid pain condition, such as interstitial cystitis or fibromyalgia, and this is associated with increased feelings of isolation and invalidation [14].

Women with a sexual pain disorder report disruptions to every aspect of their sexuality compared to women without these conditions, including lower desire, arousal, sexual satisfaction, and frequency of orgasm and intercourse [15–19]. Indeed, affected women typically score in the clinical range of sexual dysfunction for low desire and arousal [20]. They also report more sexual anxiety, a greater tendency to perceive sexual cues as negative (i.e., erotophobia), more negative and less positive cognitions about penetration, poorer sexual communication, and lower sexual self-esteem compared to pain-free controls [11, 18, 19, 21–23].

Male partners of women with sexual pain also suffer consequences. In one of the first studies to examine the impact of women's sexual pain on male partners, the partners reported more depressive symptoms when compared to a control group of healthy men [24]. Other studies have not replicated this finding [25, 26]. However, male partners have reported decreased sexual satisfaction and an increased prevalence of sexual difficulties (e.g., erectile dysfunction) compared to male partners of women without sexual pain [25, 23].

Although most affected couples remain quite satisfied with their overall romantic relationships [27], some studies have found significantly lower self-reported relationship satisfaction in women with sexual pain compared to those without [17, 28, 29]. The sexual relationship is an integral part of the overall quality of an intimate relationship [30]; thus, it is not surprising that couples, including those who are in mixed-sex or same-sex relationships, report that sexual pain has a negative toll on their intimacy and sexuality [31]. In one study, 73 % of male partners reported that their partner's pain had a negative impact on their relationship [25]. Women have also reported that their pain negatively affects their ability to feel close to and show affection toward their partners [32]. In qualitative studies, women report feelings of guilt, shame, and inadequacy as a sexual partner, as well as fears of losing or disappointing their partner because of the pain [33, 34]. These findings illustrate the significant strain that sexual pain can put on a relationship and suggest that relationship stressors may further perpetuate the pain and associated consequences. Understanding associated consequences and maintaining factors will help direct clinical attention. Equally important is an understanding of the potential pathways that can lead to sexual pain.

Etiology

Biomedical Factors

Most sexual pain problems of the genital skin and mucus membranes are transient and are caused by inflammation from acute genital infections, such as candidiasis, trichomoniasis, and genital herpes, or an infection of the greater vestibular glands.

The causes of vulvodynia, or chronic, unexplained vulvar pain, have not yet been elucidated. Recent studies have shown

at least four possible pathways that may modify risk of developing this disorder: (1) hormonal changes, (2) neurological changes, (3) inflammation, and (4) hypertonic pelvic floor muscles. First, evidence on the role of combined hormonal contraception (CHC) in the development of vulvodynia is inconclusive, as several cross-sectional studies have shown that CHC use significantly increases the risk of developing vulvodynia [35, 36], while a recent large population-based study showed that CHC use did not increase the risk of vulvodynia [37]. Nevertheless, it has been demonstrated that CHCs induce morphologic changes in the vestibular mucosa [38] and increase its sensitivity [39]. Second, various studies have examined the role of vulvar nerve fiber density in vulvodynia. However, the results are contradictory in terms of the presence or absence of intraepithelial and dermal nerve fibers [40–44]. Additionally, using quantitative sensory testing methodologies, vulvodynia patients report lower pain thresholds than pain-free women, as well as increased pain intensity at supra-threshold levels [45]. Third, it has also been postulated that an inability to clear vulvovaginal infections and the resulting inflammation may lead to the development of vulvodynia. Although the literature contains inconsistent histopathological findings [43, 46, 44, 47], the former point is supported by an experimental mouse model of vulvodynia that confirmed that repeated vulvovaginal infections with *Candida albicans* led to chronic vulvar pain behavior and increased vulvar innervation [48]. Finally, vulvodynia may be associated with some degree of pelvic floor muscle dysfunction. Using electromyography recordings, several studies have demonstrated increased resting muscle tone, impaired voluntary relaxation and decreased voluntary muscle contractile ability in women with vulvodynia compared to asymptomatic women [49]. Working with physical therapists, Reissing et al. [50] found that women with vaginismus demonstrated significantly higher vaginal/pelvic muscle tone and lower muscle strength than women with PVD or controls. Single "candidate" gene research has been performed in the field of vulvodynia, and genetic polymorphisms have been linked to the development of vulvar pain [51–56]. Although to date, there is evidence for heritability in other chronic pain syndromes [57], no heritability studies have been performed in the context of vulvodynia. Some conditions that may contribute to deep dyspareunia include pelvic inflammatory disease, endometriosis, adnexae pathologies (e.g., ovarian cysts), uterine pathologies (e.g., leiomyoma, adenomyosis), and the pelvic congestion syndrome.

Psychological Factors

Similar to biomedical factors, the psychological factors involved in the etiology of female sexual pain disorders are multifactorial and varied. In a large-scale cross-sectional study, female adolescents experiencing pain during sexual intercourse were more likely to report a history of sexual abuse, fear of physical abuse, trait anxiety, and potentially harmful vulvar hygiene behaviors compared to adolescent females reporting no pain [58]. In this same study, adolescents who reported a lifetime occurrence of sexual abuse were 1.9 times more likely to report sexual pain compared to those who did not experience sexual abuse [58]. Using a case-control study, researchers attempted to examine the role of psychosocial stressors in the etiology of vulvodynia. Compared to unaffected women, women with vulvodynia were almost three times more likely to have experienced severe childhood physical or sexual abuse or to have lived in severe fear of any abuse as a child [59]. A community-based study showed that the vulvovaginal pain was four times more likely among women with antecedent depression or anxiety compared to women without and that these disorders were also significantly more prevalent as consequences of the vulvar pain when compared to healthy controls [13]. In contrast, some studies have found no association between sexual pain and depressive symptoms [18, 60, 61].

In keeping with the biopsychosocial model, there is empirical evidence to suggest that cognitive interpretations such as attributions or beliefs about the pain contribute to its increased intensity [62] and thus play a role in pain modulation and management. Women with sexual pain report more catastrophizing about their pain (i.e., an exaggerated and pessimistic perspective) compared to healthy control women [60, 45] and also demonstrate higher levels of hypervigilance toward the pain when compared to a neutral stimulus [63]. Higher levels of catastrophizing, fear of pain, and hypervigilance and lower levels of self-efficacy correlate with increased pain in women with PVD, while higher levels of anxiety and avoidance and lower levels of self-efficacy were associated with their increased sexual dysfunction [64]. Moreover, there is evidence of a prospective relationship between cognitive variables and PVD, where higher levels of pain catastrophizing and lower levels of self-efficacy were shown to predict worse treatment outcomes in a randomized trial evaluating cognitive-behavioral therapy (CBT) [65], and increases in pain self-efficacy over a 2-year period were associated with better pain, sexual function, and sexual satisfaction outcomes [66].

Continued efforts to understand psychological risk factors for sexual pain are integral to mitigating affected women's distress. Current understanding is limited by cross-sectional designs and a dearth of prospective research, yet there are strong implications that early psychological stressors such as abuse, fear of abuse, and anxiety play a role in the development of sexual pain disorders.

Relationship Factors

Given the sexual context in which female sexual pain is most often triggered, research has increasingly focused on the role

of relationship factors in sexual pain. Partner responses, the most studied of the relationship factors, can be negative (e.g., hostility), solicitous (e.g., sympathy), and facilitative (e.g., affection and encouragement of adaptive coping). In cross-sectional and daily diary studies, greater facilitative partner responses were associated with women's lower intercourse pain [67] and better sexual functioning [68], as well as couples' greater relationship and sexual satisfaction [69, 67]. Conversely, greater negative and solicitous partner responses are associated with greater pain [26, 67, 70, 71] and more depressive symptoms in women [72], as well as lower sexual functioning [68] and relationship and sexual satisfaction in couples [69]. Whereas facilitative responses may promote couples' use of adaptive coping strategies and shared emotion regulation in the face of pain, solicitous and negative partner responses may reinforce avoidance of pain and sex and disrupt couples' pain-related coping and emotion regulation.

Couples' greater ambivalence over emotional expression (a marker of poor emotion regulation) has been associated with reductions in their sexual satisfaction and function, psychological adjustment, and relationship adjustment [73]. Furthermore, studies examining male partners' pain-related cognitions showed that their lower pain catastrophizing was associated with women's lower pain [74] and partners' greater pain acceptance was associated with their own lower incidence of depressive symptoms [75]. Couples' beliefs regarding women's pain can also affect outcomes, as male partners' higher negative pain attributions have been shown to predict their own greater psychological distress, and poorer relationship and sexual satisfaction in the presence of women's greater pain intensity [76]. Finally, while partners accurately tracked women's pain during intercourse over a period of 2 months, they generally underestimated their female partner's pain, and those whose relationship satisfaction varied more day to day demonstrated poorer tracking accuracy for the woman's pain [77]. Together, these studies highlight the many ways that partners' own experiences of the pain may directly or indirectly influence women's pain and couples' psychological, sexual, and relational outcomes.

Couples affected by sexual pain may be more likely to experience relational obstacles than couples in the general population. For example, women with sexual pain are more likely than women without sexual pain to have insecure romantic attachments [78], and couples affected by sexual pain report lower sexual communication than pain-free couples [25, 23]. In turn, lower sexual communication and insecure romantic attachments are associated with women's greater sexual distress [79] and couples' lower sexual function [79, 80], sexual satisfaction [80], and relationship satisfaction [79]. Moreover, couples affected by sexual pain have greater self-reported and observed intimacy, which is associated with their greater sexual satisfaction, sexual function, pain self-efficacy, or the belief that they can manage the pain, and lower sexual distress [81, 82]. Finally, examination of interpersonal sexual goals of women with sexual pain found that women with more significant approach goals (i.e., having sex to pursue intimate connection) in comparison with those with lower avoidance goals (e.g., having sex to avoid relational conflict) were associated with greater sexual and relationship satisfaction and a lower incidence of depression [83]. Together, these studies underscore the importance of targeting the dyadic context of female sexual pain.

Treatment

Medical Treatments

Vulvodynia is difficult to treat. Multiple treatments have been used, including vulvar care measures; topical, oral, and injectable medications; and surgery. However, most evidence for treating vulvodynia is based on clinical experience, descriptive or observational studies, or the reports of expert committees. Few randomized clinical trials (RCTs) have been performed to verify the efficacy of medical treatments [84–87]. Furthermore, a confounding factor is the improvement of pain symptoms in up to 40 % of patients with vulvodynia with no treatment [88]. The most commonly prescribed topical medication is lidocaine 5 %. This can be applied as needed for symptomatic relief and 30 min before sexual intercourse. Although nightly vulvar application of lidocaine 5 % reduced pain during sexual intercourse in a prospective study [89], in a randomized placebo-controlled trial, lidocaine cream was found to be less effective than topical placebo [87]. Topical application of cromolyn 4 % at the vulva in a placebo-controlled randomized, double-blind study demonstrated no statistically significant difference in symptom reduction compared to placebo use [84]. Likewise, the effectiveness of topical nifedipine did not exceed that of placebo in a double-blind study of 30 women with vulvodynia [85]. Capsaicin is available to treat neuropathic pain and was effective in the treatment of vulvodynia in a small retrospective study [90]. However, its extreme irritant effects limit its usefulness in treatment of sexual pain. Estrogen has been used topically with varying results [91]. Nevertheless, application of estradiol 0.03 % and testosterone 0.1 % is effective in women who have developed vestibulodynia while taking oral contraceptives [92]. A common oral treatment is the use of antidepressants, such as tricyclic antidepressants (TCAs), selective serotonin reuptake inhibitors (SSRIs), and selective norepinephrine reuptake inhibitors (SNRIs) [93]. Descriptive studies regarding the use of antidepressants to treat vulvodynia report success rates of 27 to 100 % [93]. However, a double-blind randomized placebo-controlled trial that included 133 women with localized vulvodynia showed that desipramine as monotherapy or in combination with topical lidocaine failed to reduce vulvodynia pain more than placebo [87]. In addition, oral

anticonvulsants (e.g., gabapentin and carbamazepine) have been used to treat vulvodynia [91]. Although observational studies and case reports suggest that anticonvulsants reduce pain for some women, no RCTs studying the use of anticonvulsants to treat vulvodynia have been published. In terms of injectable medications for vulvodynia, intralesional steroids and bupivacaine injections have been successful for some patients with localized vulvodynia [94]. Additionally, interferon α and injection of botulinum toxin A have been successfully used for the treatment of vulvodynia. However, the only randomized, placebo-controlled, double-blind study comparing botulinum toxin A with placebo resulted in significant reduction of equal value in both groups after 6-month follow-up [86].

Finally, surgical management for vestibulodynia has demonstrated success rates of 60 to 90 % [95]. One RCT showed that improvements resulting from vestibulectomy were maintained at a 2.5-year follow-up and that this treatment was superior to psychological and biofeedback interventions in terms of pain reduction [96]. The extent to which vestibulectomy can improve sexual function and satisfaction is not known. Although vestibulectomy seems to be effective, insufficient data on complication rates precludes its recommendation as the initial treatment for vulvodynia [91].

Specific treatments for deep dyspareunia are based on the underlying condition. Treatment of the causative factor, such as a myomectomy for uterine leiomyomatoma or the removal of ovarian cysts, usually results in improvement of the dyspareunia.

Physical Therapy Treatments

Pelvic floor physical therapy is a widely used and well-accepted treatment option for sexual pain [97]. Many studies now support the effectiveness of different modalities targeting pelvic floor muscle dysfunctions [98], although no RCT focusing on a comprehensive physical therapy program has been published. Education, electromyographic (EMG) biofeedback, manual and guided insertion techniques, and electro-therapeutic methods are all important components of physical therapy interventions. Stretching and myofascial release techniques in particular are thought to facilitate muscle relaxation, normalize tone, improve blood circulation and mobility in the pelvi-perineal region, and adjust postural imbalances, as well as increase the size of the vaginal opening and desensitize the area [99, 100].

The use of EMG biofeedback to treat sexual pain was first popularized by Howard Glazer [101], who retrospectively evaluated its effectiveness in a sample of 33 women with PVD. At 6-month follow-up, 52 % of participants were pain-free, and 79 % who had been abstaining from intercourse were able to resume this activity. In another retrospective study involving a sample of 43 women with vulvodynia, Glazer [102] showed that treatment gains were maintained 3 to 5 years post-treatment.

Bergeron et al. [96] published a 2.5-year follow-up study of women with PVD who had been randomized to vestibulectomy, biofeedback, or cognitive-behavioral group therapy. Results showed that for all three arms of the study, treatment gains were maintained and pain intensity was further reduced at follow-up, suggesting that biofeedback has stable, long-lasting positive effects. In another randomized trial, Danielsson et al. [103] compared biofeedback to lidocaine for the treatment of PVD. A significant improvement was found in 66 % of the participants at 12-month follow-up.

Studies have also investigated the effects of various electrotherapeutic modalities in the treatment of sexual pain. In a prospective study involving 29 participants, Nappi et al. [104] showed improvements in pain, pelvic floor contractility, and muscle relaxation using electrical stimulation. Similarly, Fitzwater et al. [105] conducted a retrospective study involving 66 women with chronic pelvic pain and spasm of the levator ani—a pelvic floor muscle. Electrical stimulation resulted in a reduction in muscle tension and pain. Finally, in a randomized controlled trial in 40 women with PVD, Murina et al. [106] found a reduction in pain and an improvement in sexual function following transcutaneous electrical nerve stimulation (TENS) treatment.

Two studies have focused on a comprehensive physical therapy program rather than a single component such as biofeedback. A first retrospective study investigating such a program in a sample of 35 women diagnosed with PVD yielded a 52 % success rate with regard to pain reduction and a significant improvement in participants' sexual functioning [107]. Using a prospective methodology, another study in a sample of 11 women diagnosed with PVD, Gentilcore-Saulnier et al. [108] demonstrated that a physical therapy program led to the normalization of the pelvic floor. Women also reported a significant reduction in pain during vaginal palpation and significant decreases in pain during intercourse and during a gynecological examination, as well as improved sexual function [109].

Psychosocial Treatments

Given the multifaceted nature of the etiology, modulation and impact of sexual pain, a treatment model that can target pain as well as its associated psychological, sexual, and relational consequences, could have a presumed advantage over interventions targeting only the pain. In one of the first randomized clinical studies, group CBT for women with PVD that targeted pain management and sexual functioning resulted in significant improvements in sexual function and psychological adjustment alongside the other two interventions, vestibulectomy and biofeedback [110]. Among other empiric interventions, this eight-session CBT included

psychoeducation, exposure exercises, pain journaling, and cognitive restructuring. While CBT did not outperform the other interventions on pain improvements, participants in this arm demonstrated a lower attrition rate than the vestibulectomy group and were also more satisfied with their treatment than the biofeedback group [110]. These improvements in pain and sexual functioning for women receiving CBT were maintained at 6-month and 2.5-year follow-ups [96]. In a retrospective, uncontrolled follow-up study of women treated with CBT for superficial coital pain and vaginismus, 56 % of women reported a reduction in coital pain problems, but also reported having a greater sense of self as a sex partner and as a woman following treatment [111], thus highlighting the potential to see improvements in areas beyond the pain itself. To distinguish active psychotherapeutic properties from supportive effects of therapeutic interventions, a randomized trial examining the efficacy of individual CBT for vulvodynia compared to a supportive psychotherapy demonstrated that CBT resulted in significantly greater improvement in pain severity and sexual function between pre- and post-treatment, with gains being maintained at 1-year follow-up [112]. The first randomized trial examining cognitive-behavioral sex therapy for vaginismus yielded a 15 % success rate, suggesting that simply talking about the problem in the therapist's office may not be sufficient [113]. Another randomized trial of therapist-aided exposure therapy for vaginismus showed that 89 % of study participants were able to achieve vaginal intercourse post-treatment, indicating that exposure may be a crucial ingredient of therapeutic success [114]. Results from these studies demonstrate the effectiveness of psychosocial interventions for sexual pain while also indicating the potential benefit of addressing the multifactorial nature of sexual pain.

Given the potential effectiveness of mindfulness in the management of other persistent pain conditions [115, 116], researchers have also begun to develop and test mindfulness-based CBT interventions for sexual pain. One example is a mindfulness and CBT program delivered in a group setting over four 2-h sessions and consisting of education about PVD and pain, CBT skills to address problematic thoughts, progressive muscle relaxation, and mindfulness exercises, as well as sex therapy, including a discussion of non-penetrative pleasuring [117]. An assessment of the efficacy of this four-session integrated mindfulness-based group therapy for PVD compared to a wait-list control showed significant improvements between pre- and post-treatment and from post-treatment to 6-month follow-up in pain catastrophizing, pain hypervigilance, cotton swab-provoked allodynia, and sexual distress, but not for pain experienced during sexual intercourse [118]. Mindfulness may be an important component to the management of pain-related distress in the context of sexual pain.

There have been repeated recommendations to include the partner in therapy given the interpersonal factors involved in women's sexual pain [81, 73]. Cognitive-behavioral couple therapy (CBCT) for couples with vulvodynia is described as an integrative pain management and sex therapy intervention delivered to couples over 12 weekly 60-min sessions and includes psychoeducation, introduction of pain management techniques, communication skill building, sexual re-approach, discussion and development of sexual narratives, and mindfulness- and acceptance-based exercises [119]. Pilot testing of this novel treatment yielded significant pre- to post-treatment improvements in pain during sexual intercourse and sexual functioning for women and in sexual satisfaction for women and partners. Exploratory analyses indicated large improvements in pain catastrophizing for both members of the couple and both partners' perception of women's pain self-efficacy, as well as improvements in other psychological and interpersonal outcomes, highlighting the potential benefit of a couple-based approach [119]. While limited by a small sample size, these promising results underscore the importance of continued efforts in understanding the benefits and mechanisms of psychosocial and psychotherapeutic interventions for women's sexual pain disorders, for the woman and the couple.

Integrated, Multimodal Treatments

Despite some successes with 1D medical, physical, and psychological therapies, the multifactorial etiology of female sexual pain points to the potential value of integrated, multimodal approaches to sexual pain treatment [5]. Indeed, recent standard operating procedures call for biopsychosocial and multidisciplinary approaches to the assessment and treatment of female sexual pain conditions [120]. To date, a small number of quantitative [121–124] and qualitative studies [125, 126] have examined the impact of treatment programs for women's sexual pain that integrate medical, physical, and psychological therapies. For example, in an uncontrolled, prospective study, Backman and colleagues found that 6 months after completing a program of concurrent sex therapy and pelvic floor physical therapy, women reported significant reductions in dyspareunia, greater frequency of intercourse, and improvements in sexual functioning [123]. In the most comprehensive series of studies to date, Brotto and colleagues reported on the outcomes, including patient-perceived benefits, of a 10- to 12-week multidisciplinary vulvodynia program consisting of educational seminars, medical management, group psychological skills training, and pelvic floor physiotherapy [121, 122, 125]. Women participating in the program experienced significant pre- to post-treatment reductions in dyspareunia and sexual distress and significant improvements in sexual functioning and sexual satisfaction; these gains were maintained 3 months later [121]. Additionally, women perceived many benefits of the program including feeling supported and

empowered; acquiring knowledge, skills, and tools; and experiencing improvements in emotional well-being [125].

Despite these encouraging results, this small body of literature contends with methodological concerns such as small sample sizes [124, 123], retrospective designs [124], and a lack of randomization or control groups [121–124], which confounds treatment effects with potential placebo responses.

Conclusions and Future Research Directions

Future research and treatment progress in the field of sexual pain will require an expansion of our research methodologies and greater use of rigorous designs. Novel methodologies such as 4D ultrasound to measure pelvic floor muscle activity without introducing a measurement bias should be prioritized. Heritability research is still lacking, yet findings to date suggest that genetic factors warrant further study. Longitudinal designs following cohorts of young women from prepuberty to adulthood are ambitious, but probably the only way to better understand the roles of risk factors such as childhood maltreatment, hormonal changes and contraception, and inflammatory processes, as well as the mechanisms by which they contribute to the development of sexual pain.

Despite the fact that most sexual activity involves two partners, research in the area of sexual pain has tended to include only the afflicted woman. Future research should include both members of the couple in order to examine the role of dynamic interpersonal processes in the experience of pain and associated psychological distress and sexual impairment. Observing couples in the laboratory could limit recall biases associated with self-report measures and correlational designs and capture interactions as they are unfolding in real time. Dyadic diary methodologies also offer the opportunity to measure a phenomenon closer to its occurrence, with the added benefit of examining it in the natural environment of participants. Experimental research conducted in the laboratory with nonclinical samples could also facilitate the identification of basic mechanisms involved in relating couple variables, pain and sexuality outcomes. Dyadic data analytic strategies that take into account the interdependency of couple data, such as the actor partner interdependence model and the common fate model [127], should be privileged for all of these designs.

As for treatment research, there is a great need for more RCTs in order to provide evidence-based treatment options to patients and their partners. Too many studies in the field still rely on small clinical samples, which are probably biased in the direction of increased symptomatology. Efforts need to be made to recruit research participants from the community and to access those who do not seek help and are not involved with the healthcare system. New treatments should be theory-driven rather than purely developed on a trial and error basis and then tested within an experimental framework. For psychosocial and physical therapy interventions, multiple measures administered during treatment could capture potential mechanisms of change. Although most now espouse integrated approaches (e.g., [3]), which address the multifactorial nature of sexual pain conditions, few studies have examined whether they are actually superior to single modality approaches, and no RCT has focused on testing any kind of multimodal framework.

Finally, the fragmentation of disciplines impedes progress in our field. There is an urgent need for (1) increased dialogue among sub-disciplines and across disciplines and (2) research endeavors that incorporate both biomedical and psychosocial variables and questions focusing on the interactions therein. Such studies will contribute to the development of more refined, biopsychosocial etiological models of sexual pain and will inform the development of targeted interventions.

Compliance with Ethics Guidelines

Conflict of Interest Sophie Bergeron, Serena Corsini-Munt, Leen Aerts, Kate Rancourt, and Natalie O. Rosen declare no conflicts of interest.

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