2015 ISSVD, ISSWSH, and IPPS Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia

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On behalf of the consensus vulvar pain terminology committee of the International Society for the Study of Vulvovaginal Disease (ISSVD), the International Society for the Study of Women's Sexual Health (ISSWSH), and the International Pelvic Pain Society (IPPS)

Introduction: In 2014, the executive council of the International Society for the Study of Vulvovaginal Disease, the boards of directors of the International Society for the Study of Women's Sexual Health, and the International Pelvic Pain Society acknowledged the need to revise the current terminology of vulvar pain, on the basis of the significant increase in high-quality etiologic studies published in the last decade.

Materials and Methods: The new terminology was achieved in the following 4 steps. The first involved a terminology consensus conference with representatives of the 3 societies, held in April 2015. Then, an analysis of the relevant published studies was used to establish a level of evidence for each factor associated with vulvodynia. The terminology was amended on the basis of feedback from members of the societies. Finally, each society's board accepted the new terminology.

Results and Conclusions: In 2015, the International Society for the Study of Vulvovaginal Disease, International Society for the Study of Women's Sexual Health, and International Pelvic Pain Society adopted a new vulvar pain and vulvodynia terminology that acknowledges the complexity of the clinical presentation and pathophysiology involved in vulvar pain and vulvodynia, and incorporates new information derived from evidence-based studies conducted since the last terminology published in 2003.

Key Words: vulvar pain, vulvodynia, terminology, vulvar vestibulitis, vestibulodynia

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For more than a decade, gynecologists, dermatologists, vulvar pain specialists, and researchers have used the 2003 International Society for the Study of Vulvovaginal Disease (ISSVD) terminology as a guide to diagnosing vulvar pain (see Table 1).[1] This terminology evolved over years of discussion on the nature of idiopathic vulvar pain, especially that occurring during sexual activity involving vaginal penetration, which was commonly referred to as “vestibulitis” (see Table 2).[1,2] The 2003 terminology[1] divides vulvar pain into the following 2 overarching categories: vulvar pain related to a specific disorder, and vulvodynia, defined as “vulvar discomfort, most often described as burning pain, occurring in the absence of relevant visible findings or a specific, clinically identifiable, neurologic disorder.” Clinically, the use of the term vulvodynia, as directed by the 2003 terminology, came to mean pain “of unknown cause”; in other words, the term “vulvodynia” implies that the pain is idiopathic.

Since 2003, the category of “identifiable causes” for vulvar pain has evolved substantially, as have factors “potentially associated” with vulvodynia. These developments are based on the results of numerous studies examining a wide range of possible etiological factors (e.g., inflammatory, genetic, musculoskeletal, neurosensory, neuropathic) and treatment avenues (e.g., pelvic floor physical therapy, surgical intervention, psychological intervention, oral medication). These studies have led to the conclusion that vulvodynia is likely not one disease but a constellation of symptoms of several (sometimes overlapping) disease processes, which will benefit best from a range of treatments based on individual presentation (e.g., Refs. 3,4). These studies have widened the scope of etiological considerations and have resulted in the need to update the description and nomenclature of persistent vulvar pain in a new terminology. In addition, several important pain characteristics of vulvodynia have been introduced (e.g., intermittent and constant pain pattern), consistent with how pain conditions in general are classified.[5]

Vulvar pain has been a focus of several professional organizations in addition to the ISSVD, the 2 most prominent are the International Society for the Study of Women's Sexual Health (ISSWSH) and the International Pelvic Pain Society (IPPS). To take advantage of the experience and knowledge of these 3 societies and to obtain a broad-based consensus, a meeting with representatives from all

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TABLE 1. 2003 ISSVD Terminology and Classification of Vulvar Pain

A. Vulvar pain related to a specific disorder
   1. Infectious (e.g., candidiasis, herpes, etc)
   2. Inflammatory (e.g., lichen planus, immunobullous disorders, etc)
   3. Neoplastic (e.g., Paget disease, squamous cell carcinoma, etc)
   4. Neurologic (e.g., herpes neuralgia, spinal nerve compression, etc)
B. Vulvodynia
   1. Generalized
      a. Provoked (sexual, nonsexual, or both)
      b. Unprovoked
      c. Mixed (provoked and unprovoked)
   2. Localized (vestibulodynia, clitorodynia, hemivulvodynia, etc)
      a. Provoked (sexual, nonsexual, or both)
      b. Unprovoked
      c. Mixed (provoked and unprovoked)

3 societies was arranged. In addition, members from 3 other organizations were also invited: the American College of Obstetricians and Gynecologists, which publishes vulvodynia guidelines, the American Society for Colposcopy and Cervical Pathology, which strives to improve clinician competence, performance, and patient outcomes through educational activities focused around the study, prevention, diagnosis, and management of lower genital tract disorders, and the National Vulvodynia Association, which is a major nonprofit organization that promotes vulvodynia research and education to members of the scientific and clinical communities and lay public. It was agreed that the proposed terminology resulting from the consensus meeting would be brought to a discussion and vote by each society. This article discusses the development of the 2015 consensus terminology and classification of persistent vulvar pain based on this consensus process.

MATERIALS AND METHODS

Objective

The objective of the consensus process was to develop an evidence-based terminology on the basis of the input from experts experienced in the diagnosis, evaluation, treatment, and/or study of persistent vulvar pain in women. The terminology and resulting definitions had to be applicable across disciplines and useful for both clinical and research settings.

The Process

The ISSVD, ISSWSH, and IPPS organized an international meeting to reach a consensus on the terminology of persistent vulvar pain, on April 8–9, 2015, in Annapolis, Maryland.

The leadership of the 3 societies identified individual members with expertise in the care and/or research of persistent vulvar pain and invited them to participate. In addition to representatives of these 3 societies, representatives from the American College of Obstetricians and Gynecologists, American Society for Colposcopy and Cervical Pathology, and National Vulvodynia Association were also present.

The meeting began with a description of the development of the 2003 terminology, followed by presentations on relevant anatomy, histopathology, and neuroanatomy, given by experts in the field. Subsequent topics included physical examination and psychometrics as well as discussion of the possible contributing factors to vulvar pain (i.e., systemic, psychosocial, neuroproliferative, musculoskeletal, inflammatory, genetic, and central nervous system-related factors).

The members were then divided into small groups to develop proposals for the new terminology. Six different proposals were presented to the entire consensus group. The 2 terminologies that received most support were further discussed and united to form the final terminology proposal, which was unanimously accepted at the meeting. At that point, several working groups were charged with the postmeeting mission of reviewing the published studies on factors potentially associated with the genesis and expression of the pain of vulvodynia. The studies were rated using the Oxford Centre for Evidence Based Medicine (2011)[6] and levels of evidence assigned to each associated factor. The assigned levels of evidence were discussed among the members until consensus was reached.

The terminology proposal was presented and discussed by e-mail with the ISSVD membership and then discussed at the 2015 ISSVD World Congress. At the world congress, a slightly modified version of the consensus terminology was approved, and subsequently, the Boards of Directors ISSWSH and IPPS discussed and approved the same terminology in July and August of 2015.

RESULTS

The title of the new terminology is the "2015 Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia" (see Table 3), referred to as the "2015 terminology" in this article. Although the 2003 terminology[1] had "ISSVD" in its

TABLE 2. Historic Descriptions and Terminologies of Idiopathic Vulvar Pain

Period, author	Term or terminology
First century AD, Soranus	Satyriasis in females
1880, Thomas	Excessive hypersensibility of the nerves
1889, Kellogg	Sensitive points
1889, Skene	Super sensitiveness of the vulva
1928, Kelly	Exquisitely sensitive in hymeneal ring
1976, Weisfogel	The burning vulva
1976, ISSVD	The burning vulva syndrome
1978, Dodson and Friedrich	Psychosomatic vulvovaginitis
1978, Tovell and Young	Vulvodynia or pudendagra
1983, ISSVD task force	Vulvodynia or burning vulva syndrome
1983, Friedrich	Vestibular adenitis
1983, Woodruff and Parmley	Infection of the minor vestibular glands
1986, Peckham	Focal vulvitis
1987, Friedrich	Vulvar vestibulitis syndrome
1988, McKay	Classification: vestibulitis and dysesthetic vulvodynia
1997, Bornstein	Vestibulodynia
1999, ISSVD	Terminology: generalized and localized vulvar dysesthesia
2001, ISSVD	Terminology: provoked and spontaneous vulvar dysesthesia, each has subsets of generalized and localized
2003, ISSVD	2003 ISSVD terminology

Based on Moyal-Barracco et al.[1] and McElhiney et al.[2]

ISSVD indicates International Society for the Study of Vulvovaginal Disease.

TABLE 3. 2015 Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia

A. Vulvar pain caused by a specific disorder[a]
- Infectious (e.g., recurrent candidiasis, herpes)
- Inflammatory (e.g., lichen sclerosus, lichen planus, immunobullous disorders)
- Neoplastic (e.g., Paget disease, squamous cell carcinoma)
- Neurologic (e.g., postherpetic neuralgia, nerve compression, or injury, neuroma)
- Trauma (e.g., female genital cutting, obstetrical)
- Iatrogenic (e.g., postoperative, chemotherapy, radiation)
- Hormonal deficiencies (e.g., genitourinary syndrome of menopause [vulvovaginal atrophy], lactational amenorrhea)

B. Vulvodynia—vulvar pain of at least 3 months' duration, without clear identifiable cause, which may have potential associated factors.

The following are the descriptors:
- Localized (e.g., vestibulodynia, clitorodynia) or generalized or mixed (localized and generalized)
- Provoked (e.g,. insertional, contact) or spontaneous or mixed (provoked and spontaneous)
- Onset (primary or secondary)
- Temporal pattern (intermittent, persistent, constant, immediate, delayed)

[a]Women may have both a specific disorder (e.g., lichen sclerosus) and vulvodynia.

title, the current terminology title reflects its acceptance not only by the ISSVD but also by all 3 societies that are stakeholders in vulvar pain.

The table is divided into 2 sections. The first section is called "Vulvar pain caused by a specific disorder." This section contains vulvar pain conditions for which a cause can be clearly identified (e.g., pain caused by herpes genitalis or genital cutting). Note that one of the causes of vulvar pain is "genitourinary syndrome of menopause." This term has recently been introduced to replace "vulvovaginal atrophy."[7] To avoid confusion, the former term is shown in parentheses. In addition, some women have both vulvar dermatoses and vulvodynia, and this comorbid presentation is the reason that the following footnote was added: "Women may have both vulvar pain caused by a specific disorder (e.g., lichen sclerosus) and vulvodynia."

Other factors, however, may not be as clearly associated with the pain; hence, the second section of the definition is "Vulvodynia: vulvar pain of at least 3 months' duration, without clear identifiable cause, which may have potential associated factors." The minimum duration of the pain is specified, on the basis of a review of the literature showing 3 months of being the most frequently used.[4,8,9]

A special section of the terminology defines the descriptors of vulvodynia, such as "generalized" or "localized," among others. This section reflects the findings that pain characteristics typically used to define that persistent pain conditions[5] may be more useful for classifying vulvodynia subtypes than specifiers based on hypothesized etiology. The possibility of a "mix" (i.e., a combination) of 2 descriptors is also stated for clarification.

The consensus development process identified several important factors potentially associated with vulvodynia. They may be clinically prominent and may help in choosing further evaluation methods or a treatment path. These potential associated factors are mentioned in an appendix to the terminology (see Table 4) and described individually later. Note that because it is not mandatory to determine an associated factor, we added the phrase, "which may have potential associated factors." Factors can co-occur and overlap, such that for a given patient, 2 groups of factors (or more) may be hypothesized to play a role in the pain experience and hence may be identified as treatment targets. This change represents a shift from one in which a single treatment modality would be considered key to one in which an interdisciplinary approach is seen as ideal, given the wide range of potential factors associated with vulvodynia, as well as the possible interactions among them.

Potential Factors Associated With Vulvodynia

Although many studies were reviewed to determine the levels of evidence, we are listing some representative references for each associated factor. For a full listing of references, please refer to the ISSVD website (www.issvd.org).

Comorbidities and Other Pain Syndromes

Associations between vulvodynia and other common pain syndromes are demonstrated in population-based, cross-sectional, and case-control studies. Orofacial pain is an especially strong association.[8] Studies also show that patients with vulvodynia often have more than 1 comorbid condition. In addition, when patients with other pain syndromes are evaluated for vulvodynia, comorbidity is common; in 1 study, almost all patients with interstitial cystitis/painful bladder syndrome had comorbid vulvodynia.[10]

Genetics

Some women with provoked vestibulodynia (PVD) have a genetic predisposition to developing this condition[11] via at least 3 potentially overlapping mechanisms: genetic polymorphisms that increase the risk of candidiasis or other infections, genetic changes that allow prolonged or exaggerated inflammatory responses, and increased susceptibility to hormonal changes associated with oral contraceptive pills.[12]

Hormonal Factors

The use of combined hormonal contraceptives has been associated with an increased risk of developing PVD[13] (but see Ref. 14 for an exception).

TABLE 4. 2015 Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia

Appendix: potential factors associated with vulvodynia[a]
- Comorbidities and other pain syndromes (e.g., painful bladder syndrome, fibromyalgia, irritable bowel syndrome, temporomandibular disorder; level of evidence 2)
- Genetics (level of evidence 2)
- Hormonal factors (e.g., pharmacologically induced; level of evidence 2)
- Inflammation (level of evidence 2)
- Musculoskeletal (e.g., pelvic muscle overactivity, myofascial, biomechanical; level of evidence 2)
- Neurologic mechanisms
  - Central (spine, brain; level of evidence 2)
  - Peripheral: neuroproliferation (level of evidence 2)
- Psychosocial factors (e.g., mood, interpersonal, coping, role, sexual function; level of evidence 2)
- Structural defects (e.g., perineal descent; level of evidence 3)

[a]The factors are ranked by alphabetical order.

Inflammation

Various methods (e.g., histology, immune challenges, animal, in vitro models) have demonstrated increases in inflammatory cells within painful regions of the vulvar vestibule.[15] An increase in the number of mast cells and degranulated mast cells, as well as increased subepithelial heparanase activity,[16] has been associated with vestibular hyperinnervation in women with PVD. An inability to downregulate proinflammatory cytokine activity was demonstrated in whole blood cultures from women with PVD than in controls.[17]

Musculoskeletal

Several causes of pelvic floor hypertonic dysfunction/overactivity have been identified in vulvodynia, including those caused by singular events (e.g., acute vaginal/urinary infection) and insidious factors (e.g., prolonged sitting). These factors may lead to imbalances and functional modifications in the pelvic floor myofascia and neural tissue.[18] Several controlled studies have demonstrated pelvic floor muscle hypertonicity and other dysregulations in women with PVD.[19]

Neurologic Mechanisms

Central. Several controlled studies have demonstrated that women with PVD are more sensitive to various forms of stimulation in nongenital areas of their body (e.g., forearm[20]) than pain-free women. Brain imaging studies have indicated changes in structure, function, and resting state in women with PVD versus those without PVD.[21]

Peripheral.

NEUROPROLIFERATION. An increase in the density of nerve endings in the vestibular endoderm has been demonstrated.[22] These nerve endings have been identified as nociceptors[23] that exhibit an increased density of the vanilloid receptor VR1.[24] Increased innervation has implications for increased sensitivity, as documented in women with PVD.[25]

Psychosocial Factors

Population-based studies have shown that anxiety, depression, childhood victimization, and posttraumatic stress are risk factors for the development of vulvodynia. The odds of vulvodynia were 4 times higher among women with antecedent mood or anxiety disorders compared with women without.[9] Psychological factors associated with greater pain intensity or sexual dysfunction in women with vulvodynia include pain catastrophizing, fear of pain, hypervigilance to pain, lower pain self-efficacy, negative attributions about the pain, avoidance, anxiety, and depression.[26]

Structural Defects

Some women with pelvic organ prolapse complain of vulvar and/or pelvic pain. Two small case series reported that surgical correction of pelvic organ prolapse led to resolution of vulvodynia and pelvic pain in most women undergoing surgery.[27,28]

DISCUSSION

The 2015 terminology of vulvar pain reflects key developments in understanding vulvar disorders and chronic pain over the recent years. The main difference between the 2015 terminology and the 2003 terminology is the addition of "potential associated factors." This addition represents a paradigm shift in the approach to vulvodynia, resulting from research showing that several factors may be associated with the development and maintenance of the condition, rendering vulvodynia being likely the result of a multifactorial process. The inclusion of the associated factors emphasizes that treatment should be chosen according to the characteristics of the individual case and the possible associated factors, rather than as a "one-size-fits-all" approach. For instance, physical therapy could be recommended if musculoskeletal factors were suspected, whereas surgery could be recommended if neuroproliferation were thought to be the main contributing factor.

Over the years, factors that were once claimed to have a causative role in vulvodynia have been later found to be only coincidental. This discovery may also happen with the associated factors listed in the current terminology. Therefore, a level of evidence has been assigned to each factor, based on a review of the literature. The table of associated factors appears in an appendix and not in the main terminology document, because the factors are not actually part of the terminology; the appendix also allows future amendment, when research yields further knowledge, without revising the entire terminology. On the other hand, once a potential associated factor, based on new research, is considered to have a definite causative role in vulvar pain, it may be moved to the first part of the table ("A. Vulvar pain caused by a specific disorder").

Besides the addition of the "potential associated factors," the following revisions took place: "unprovoked" used in 2003 has been replaced with "spontaneous," a term that is commonly used in the pain literature.[29] The title of the terminology has been changed to the "2015 Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia" from the 2003 "Terminology and Classification of Vulvodynia," because the terminology does not pertain only to acute vulvar pain or to vulvodynia. The 2003 definition of vulvodynia, "Chronic vulvar discomfort, mainly described as burning, occurring in the absence of visible relevant findings," has been changed in the 2015 terminology to "Vulvar pain of at least 3 months' duration, without clear identifiable cause, which may have potential associated factors."

No change was made to the 2003 terminology in the division to sections, with the first being "Vulvar pain caused by a specific disorder." This section contains vulvar pain conditions for which a cause has been clearly identified. In addition, the descriptors of vulvodynia regarding location (generalized or localized) and provocation remained as in 2003.[1] However, more descriptors were added, such as onset (primary or secondary) and temporal pattern, to reflect the complexity of vulvar pain presentations seen clinically and in research settings.[30,31]

The strengths of the consensus process include the range of specialties and nationalities and the level of expertise of the committee members. The ability of experts to come to unanimous consensus despite the complexity of the disorder strengthens the validity of the consensus terminology. The limitations of the process involve the necessity of reaching a compromise for each statement, despite the varying opinions that arose, to finalize the terminology.

CONCLUSIONS

The 2015 terminology was reached by a consensus among expert societies. It is expected to replace the previous terminology that was presented in 2003, given that it reflects a more current conceptualization of persistent vulvar pain and vulvodynia based on research, clinical expertise, and tenets important for the classification of chronic pain conditions. We expect that the 2015 terminology will improve clinical care and public health, by stimulating further research into the pathogenesis of vulvodynia, shortening the time to diagnosis in affected women, and streamlining effective management options for women with vulvar pain.

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