THE JOURNAL OF
SEXUAL MEDICINE

ORIGINAL RESEARCH

PAIN

Pelvic Floor Muscle Morphometry and Function in Women With Primary and Secondary Provoked Vestibulodynia

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ABSTRACT

Introduction: Provoked vestibulodynia (PVD) can be categorized as primary PVD affecting women from their first sexual intercourse or secondary PVD, which appears after a period of pain-free intercourse. There is growing evidence that these subgroups may be distinct entities presenting different pathophysiological mechanisms. Although there are documented pelvic floor muscle alterations in provoked vestibulodynia, no study has yet evaluated whether the pelvic floor muscle morphometry or function differed between women with primary and secondary provoked vestibulodynia.

Aim: To assess and compare pelvic floor muscle morphometry and function in women with primary and secondary provoked vestibulodynia.

Methods: A total of 212 women with provoked vestibulodynia (primary = 75 and secondary = 137) participated in the study after completing a gynecologic exam to confirm their diagnosis.

Main Outcome Measure: Pelvic floor muscle morphometry was evaluated at rest and during maximal contraction using 3D/4D transperineal ultrasound and pelvic floor muscle function (tone, strength, speed of contraction, endurance) was assessed with a dynamometric speculum.

Results: Pelvic floor muscle morphometry at rest and during contraction was not statistically different between women with primary and secondary provoked vestibulodynia ($P > .327$ adjusted for the duration of symptoms; $P > .137$ unadjusted t-tests). Regarding pelvic floor muscle function assessed with the dynamometric speculum, no differences were found in tone, strength, speed of contraction, endurance between the 2 groups ($P > .144$ adjusted for duration of symptoms; $P > .118$ unadjusted t-tests).

Clinical Implications: Women with primary and secondary PVD do not differ on pelvic floor muscle morphometric or dynamometric characteristics, suggesting that physical therapy modalities should be offered to both subgroups of PVD.

Strengths & Limitations: The current study used a large and mixed clinical and community sample providing more representative findings. Moreover, the analyses were adjusted for relevant variables such as duration of symptoms. Although the inclusion of nulliparous women below 45 years of age ensured the homogeneity of the sample, it may limit the external validity.

Conclusion: These findings suggest that primary and secondary subgroups of provoked vestibulodynia cannot be differentiated by morphometric or dynamometric characteristics. Pelvic floor muscles alterations in provoked vestibulodynia are therefore not influenced by the onset of the symptoms. **Fontaine F, Dumoulin C,**

Received February 26, 2018. Accepted June 5, 2018.

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https://doi.org/10.1016/j.jsxm.2018.06.001

Bergeron S, et al. Pelvic floor muscle morphometry and function in women with primary and secondary provoked vestibulodynia. J Sex Med 2018;15:1149−1157.

Key Words: Vulvodynia; Provoked Vestibulodynia; Pathophysiology; Pelvic Floor Muscles; Pain Onset; Ultrasound; Dynamometer; Primary Vestibulodynia; Secondary Vestibulodynia

INTRODUCTION

Vulvodynia, a highly prevalent condition affecting 7%−8% of women, is defined as a vulvar pain of at least 3 months' duration, without clear identifiable cause, which may have potential associated factors.[1,2] Vulvodynia can be categorized as provoked (eg, insertional, contact), spontaneous, or mixed (provoked and spontaneous). The leading cause of vulvodynia is provoked vestibulodynia (PVD) where women experience pain to the vulvar vestibule when pressure is applied. Recent studies suggest that PVD can be further characterized according to the onset of the symptoms.[3−8] The resulting subgroups are primary (PVD1) and secondary (PVD2) provoked vestibulodynia, whereby women either experience pain since their first sexual intercourse or tampon insertion or have acquired it after a period of pain-free sexual intercourse, respectively. PVD1 (lifelong) is reported to be less frequent than PVD2 (acquired), with a proportion ranging from 20% to 35% of women with PVD.[3,7,9]

There is growing evidence that these PVD subgroups may be distinct entities with different pathogeneses.[10] Several studies have compared women with PVD1 and PVD2 on specific biomedical and psychosexual variables to achieve a better understanding of the mechanistic differences. Women with PVD1 are found to have higher pain sensitivity during quantitative sensory testing and lower pain thresholds than women with PVD2 for vulvar and non-vulvar areas.[10,11] Regarding self-reported pain intensity during sexual intercourse, there are conflicting data, with some studies finding higher pain in women with PVD1 while others report nonsignificant difference between the 2 subgroups.[8,12−14] Additionally, the painful area of the vulvar vestibule is reported to be mainly posterior in PVD1, whereas the whole vestibule area (ie, posterior, anterior, and clitoral hood areas) is involved more frequently in PVD2.[5,14] In terms of medical history, women with PVD1 are more likely to report a family history of dyspareunia, childhood enuresis, or dysmenorrhea than women with PVD2.[6,14−16] Moreover, women with PVD1 have shown significantly more neural hypertrophy and hyperplasia than those with PVD2.[4,17] Several studies have shown many more significant differences where women with PVD1 are more severely affected than women with PVD2 in relation to pathophysiological variables such as: vulvar pain natural remission, specific immune cell recruitment (ie, CD4-positive T-cell), innate immune system protein genotype (ie, mannose-binding lectin polymorphism), and body image.[7,8,17−20] Although the available evidence has primarily suggested that women with PVD1 fare worse than women with PVD2, some studies reported non-significant differences between the 2 groups on variables such as psychosexual characteristics.[9,14] As underlined in a recent scoping review,[10] small sample sizes of published studies and other methodologic pitfalls have prevented further delineation of distinct pathophysiologic pathways in PVD subgroups. Furthermore, most studies to date have focused on psychosexual and biomedical factors but neglected the examination of important mechanisms such as pelvic floor muscle (PFM) alterations.

The involvement of PFM alterations in the cause of PVD is widely acknowledged.[21−26] Indeed, several studies have shown that women with PVD present an increase in PFM tone, as well as a reduction of strength, control, and endurance when compared with healthy control subjects.[21,24−26] PFM alterations were also significantly found to be associated with pain intensity, with more alterations being related to higher pain intensity.[27] Although PFM alterations are found to be involved in PVD, no study has yet evaluated whether the PFM morphometry or function differs between women with PVD1 and PVD2. An investigation of the differential involvement of PFM alterations in PVD subgroups is relevant to better understand the underlying pathophysiological processes in each PVD subgroups and thus will be of help in tailoring more appropriate treatment modalities.

AIM

The aim of this study was to investigate and compare PFM morphometry and function in women with PVD1 and PVD2. In line with previous studies reporting worse symptoms in women with PVD1,[4,6,7,10] we hypothesized that women with PVD1 would have more pronounced PFM alterations suggestive of increased tone, reduced strength, as well as overall reduced PFM control compared with women with PVD2.

METHODS

Participants

A total of 212 nulliparous premenopausal women were included in this study. Women were recruited through posters in universities, colleges, medical clinics, and stores (32%); newspaper ads (11%); Facebook and web initiatives (30%); word of mouth (8%); public conferences (2%); and through referrals by health care professionals (17%). To be included in the study, women had to meet all of the following inclusion criteria: (1) be between ages 18 and 45 years old; (2) have pain during sexual intercourse that has been present for at least the last 6 months in at least 90% of attempts; (3) have a minimum average pain score of 5 on an 11-point numerical rating scale (NRS) in at least 1

vestibular site during the cotton swab test; and (4) have their PVD diagnosis confirmed by a gynecologist with our team. Exclusion criteria were: (1) urogynecologic and dermatologic conditions; (2) pain not limited to intercourse or other activities that exert pressure on the vulvar vestibule; (3) deep dyspareunia; (4) vaginismus (ie, never having been able to experience vaginal penetration or gynecologic exam[24]); (5) parous women and ongoing pregnancy; (6) ongoing treatment for PVD; or (7) any coexisting significant medical condition likely to interfere with assessment (eg, cardiovascular, hematologic, central nervous system, pulmonary, renal conditions). PVD was categorized as PVD1 when women reported pain since their first sexual intercourse or tampon insertion and as PVD2 when pain appeared after a period of pain-free sexual intercourse.[1]

Procedures

Women interested in participating in the study took part in a telephone screening interview to verify their eligibility. They were then given an appointment with one of our team gynecologists to confirm their PVD diagnosis according to a standardized protocol.[28] The eligible participants had to sign an informed consent form. The study was approved by the Institutional Review Boards of the participating institutions. An experienced pelvic floor physical therapist conducted the PFM assessment. Women were asked to empty their bladder to ensure their comfort during the assessment and avoid any potential influence of bladder fullness on PFM morphometry and function.[29] Thereafter, they were evaluated in a supine position on an examination table with their feet in the stirrups. The physical therapist instructed the women on how to perform a correct PFM contraction and relaxation using digital palpation. The physical therapist then proceeded to the evaluation of the PFM morphometry and function using transperineal ultrasound and an intravaginal speculum dynamometer, respectively.

Main Outcome Measures

PFM Morphometry

PFM morphometry was evaluated with a transperineal ultrasound imaging device (Voluson E8 Expert BT10 [GE Healthcare, Little Chalfont, UK] or Acuson Antares TM [Siemens, Munich, Germany]) connected to a 3D/4D probe (E8 RM6C Transducer [GE Healthcare] and Acuson Antares C7F2 Transducer [Siemens]). To allow an optimal visualization of the PFM structures, the frame rate was set at 2.0–2.8 Hz because the volume acquisition angles were set at maximal in the sagittal plane and the coronal plane. The measurements were taken at rest and during maximal PFM contraction and were repeated twice. The trial showing both most optimal relaxation and contraction according to the hiatus dimensions and anorectal angle displacement was selected for analysis. The PFM morphometry assessment consisted in measuring the following parameters according to a validated method[30–33] in the midsagittal plane: bladder neck position in the *x*-axis and *y*-axis, levator plate angle, anorectal angle; and axial plane: levator hiatus area, levator hiatal anteroposterior, and left-right transverse diameters. The test-retest and interobserver reliability of these parameters were shown to be good to excellent.[34–36] Ultrasound analysis was conducted offline by an analyst blinded to the PVD status using the 4D View v.9.1 (GE Healthcare) and Syngo.fourSight ViewTool v.3.1.0.016 (Siemens Healthcare) software.[21]

Table 1. Baseline characteristics

Characteristic	PVD1 Mean (SD) or %, (n = 75)	PVD2 Mean (SD) or %, (n = 137)	*P* Value
Age (y)	23.2 (3.8)	23.6 (4.2)	.501
BMI (kg/m^2)	22.7 (3.8)	22.5 (4.5)	.755
Place of birth			.368
North America	86.7	90.5	
Europe	5.3	2.2	
Latin/South America	1.3	3.6	
Africa	2.7	2.2	
Asia	2.7	0	
Other	1.3	1.5	
Income in Canadian $			.570
0–19,999$	70.7	62.7	
20,000–39,999$	10.6	21.9	
40,000–59,999$	16	10.2	
≥60,000$	2.7	5.2	
Education			.753
High school	10.7	16.8	
College	46.7	46.7	
Graduate	33.4	25.6	
Postgraduate	9.3	10.9	
Self-reported pain intensity during sexual intercourse (NRS 0–10)	7.1 (1.5)	7.41 (1.5)	.149
Duration of symptoms (y)	5.9 (3.7)	3.1 (2.7)	<.001
Previous treatments received			
Topical lidocaine prior intercourse	8.0	16.1	.098
Psychotherapy	8.0	3.6	.172
Topical estrogen	4.0	8.0	.259
Antidepressant	1.3	0.7	.664
Natural products	2.7	3.6	.702
Age at the first vaginal intercourse (y)	17.1 (2.7)	16.5 (2.4)	.092
Frequency of vaginal intercourse, monthly	4.3 (3.9)	5.6 (6.1)	.057
Using oral contraceptive	73.0	84.0	.085

BMI = body mass index; NRS = Numerical Rating Scale; PVD1 = primary provoked vestibulodynia; PVD2 = secondary provoked vestibulodynia.

PFM Function

The PFM function was evaluated with a pediatric-size intravaginal dynamometric speculum.[26,37,38] To ease the insertion of

Table 2. Pelvic floor muscles morphometry at rest in women with primary and secondary provoked vestibulodynia

Parameter	PVD1 Mean (±SD) N = 75	PVD2 Mean (±SD) N = 137	*P* Value (Unadjusted)*	*P* Value (Adjusted)†
Bladder neck y (cm)	2.65 (0.39)	2.70 (0.45)	.435	.646
Bladder neck x (cm)	−0.34 (0.39)	−0.44 (0.48)	.137	.379
Levator plate angle (°)	28.24 (8.82)	30.12 (10.60)	.194	.327
Anorectal angle (°)	119.41 (12.48)	117.87 (11.25)	.359	.504
Levator hiatus area (cm^2)	10.46 (2.06)	10.72 (2.33)	.430	.811
Levator hiatus AP diameter (cm)	4.54 (0.55)	4.50 (0.64)	.663	.529
Levator hiatus LR diameter (cm)	3.43 (0.41)	3.51 (0.40)	.152	.531

AP = Anteroposterior; LR = Left-right transverse; PVD1 = Primary provoked vestibulodynia; PVD2 = Secondary provoked vestibulodynia.
*Student *t*-tests were used to compare the 2 groups.
†Linear regression analyses adjusting for relevant baseline characteristics were used to compare the 2 groups.

the speculum, a hypo-allergen lubricant was applied on the condom covering the branches. Prior to the evaluation, women were asked to contract their PFM 3 times to become acquainted with and more comfortable using a speculum dynamometer. The following parameters were then evaluated using the method proposed by Morin et al:[39,40] (1) passive forces (N) at minimal vaginal aperture; (2) passive forces (N) at maximal aperture; (3) forces (N) and passive elastic stiffness (PES) were measured at minimal, maximal, and 20 mm aperture during 5 stretch-relax cycles at a constant speed of 5 mm/s, as well as the vaginal aperture (mm) at a common force of 2 N. Parameters were averaged for the last 3 cycles;[39] (4) maximal strength (N) was measured during a 15-second contraction. The strength was computed by subtracting the baseline force from the maximal force obtained during the test; (5) speed of contraction/relaxation and control were measured by the mean rate of force (N/s) of the first contraction (ie, the slope of the ascending curve being related to the speed of contraction and the slope of the descending curve indicating the speed of relaxation), as well as the number of full contractions achieved in 15 seconds; and (6) endurance was measured by asking women to maintain a maximal contraction for 90 seconds. The normalized area under the force curve taken between 10 seconds and 60 seconds after the beginning of the contraction was calculated (% · s). The reliability and validity of these parameters are excellent and have been previously published.[37–43] An observer blinded to the PVD status of the women then proceeded to analyze the data.

Statistical Analyses

A priori sample size calculation was based on minimal differences extracted from the reliability studies for hiatus dimensions, passive forces at minimal aperture, strength, and speed of contraction. A sample of 212 women (with a proportion of 35% of PVD1)[7,9] was required (1) to detect the minimal difference based on the standard error of measurement found in the reliability studies[35,40] and standard deviations available for this population[21,35] at an alpha level of 0.05; and (2) to reach 80% power (eg, passive forces at minimal aperture: SEM = 0.34N; SD = 0.84; total sample 212). PASW Statistics version 20.0 (SPSS Inc, Chicago, IL, USA) was used to perform the statistical analyses. First, multivariate analyses of variance (MANOVA)

Table 3. Pelvic floor muscles morphometry during maximal pelvic floor contraction in women with primary and secondary provoked vestibulodynia

Parameter	PVD1 Mean (±SD) N = 75	PVD2 Mean (±SD) N = 137	*P* Value (Unadjusted)*	*P* Value (Adjusted)†
Changes from baseline during contraction				
Cranioventral displacement of the bladder neck (cm)	0.52 (± 0.32)	0.51 (0.31)	.829	.673
Levator plate angle excursion (°)	8.72 (7.25)	9.28 (7.55)	.598	.597
Anorectal angle excursion (°)	4.07 (13.30)	2.79 (15.72)	.551	.498
Levator hiatus area narrowing (%)	18.41 (14.99)	17.50 (13.22)	.650	.642
Levator hiatus AP reduction (%)	13.91 (7.91)	13.92 (9.17)	.992	.885
Levator hiatus LR reduction (%)	6.81 (10.34)	6.54 (8.82)	.839	.958

AP = Anteroposterior; LR = Left-right transverse; PVD1 = Primary provoked vestibulodynia; PVD2 = Secondary provoked vestibulodynia.
*Student *t*-tests were used to compare the 2 groups.
†Linear regression analyses adjusting for relevant baseline characteristics were used to compare the 2 groups.

Table 4. General pelvic floor muscles tone in women with primary and secondary provoked vestibulodynia

Condition	Parameter	PVD1 Mean (SD) N = 75	PVD2 Mean (SD) N = 137	*P* Value (Unadjusted)*	*P* Value (Adjusted)†
Initial passive resistance	Force (N)	1.33 (0.84)	1.42 (0.84)	.445	.239
Passive resistance at maximal aperture	Force (N)	8.39 (3.55)	8.91 (4.39)	.357	.869
	Aperture (mm)	20.43 (7.65)	21.87 (8.62)	.227	.724
Dynamic stretching (lengthening and shortening cycles)	Force at minimal aperture (N)	0.34 (0.56)	0.41 (0.74)	.456	.455
	PES at minimal aperture (N/mm)	0.63 (0.29)	0.64 (0.36)	.947	.780
	Maximal aperture (mm)	29.79 (7.30)	31.01 (8.38)	.292	.885
	Force at maximal aperture (N)	10.57 (4.19)	11.01 (5.21)	.506	.920
	PES at maximal aperture (N/mm)	0.71 (0.40)	0.71 (0.41)	.995	.515
	Aperture to have a passive force of 2 N (mm)	15.98 (3.03)	15.88 (2.91)	.816	.481
	Force at an aperture of 15 mm (N)	2.11 (1.23)	2.13 (1.41)	.921	.794
	PES at an at an aperture of 15 mm (N/mm)	0.44 (0.25)	0.44 (0.21)	.808	.923
	Hysteresis (N · mm)	45.60 (33.95)	54.96 (47.92)	.103	.497

PES = Passive elastic stiffness; PVD1 = Primary provoked vestibulodynia; PVD2 = Secondary provoked vestibulodynia.
*Student *t*-tests were used to compare the 2 groups.
†Linear regression analyses adjusting for relevant baseline characteristics were used to compare the 2 groups.

were used to compare PFM morphometry and function between women with PVD1 and PVD2 to take into account their potential interrelationships. Second, Student *t*-tests were used to compare the 2 groups followed by linear regression analyses adjusting for relevant baseline characteristics (ie, duration of symptoms). For categorical data, chi-square tests were used. The significance level was set at 0.05.

RESULTS

Sociodemographic Characteristics

A total of 75 women with PVD1 and 137 with PVD2 participated in the study. As presented in Table 1, there were no significant differences in sociodemographic characteristics between women with PVD1 and PVD2. The 2 subgroups had similar age, religion, place of birth, BMI, income, and education. As expected, the symptom duration was significantly different between the subgroups: women with PVD1 had a longer duration of symptoms.

PFM Morphometry at Rest and During Maximal Contraction

Results from a MANOVA, with the PVD subgroups as the independent variable and the PFM morphometry parameters as the dependent variables, showed no significant differences between women with PVD1 and PVD2 for these parameters ($F(13, 197) = 0.929$, $P = .524$; Wilk's $\Lambda = 0.942$, partial $\eta^2 = 0.058$).

Table 5. Pelvic floor muscles contractility in women with primary and secondary provoked vestibulodynia

Condition	Parameter	PVD1 Mean (SD) N = 75	PVD2 Mean (SD) N = 137	*P* Value (Unadjusted)*	*P* Value (Adjusted)†
Maximal strength	Maximal force (N)	3.15 (1.93)	3.33 (2.12)	.533	.343
Speed of contraction/relaxation and control	Number of contractions	7.77 (2.78)	8.49 (3.34)	.118	.144
	Slope of the ascending curve (N/s)	5.83 (4.46)	5.81 (4.82)	.971	.536
	Slope of the descending curve (N/s)	−2.59 (5.86)	−2.02 (6.20)	.515	.894
Endurance	Normalized area under the force curve (% · s)	1879.06 (895.96)	1891.48 (1283.12)	.941	.985

PVD1 = primary provoked vestibulodynia; PVD2 = secondary provoked vestibulodynia.
*Student *t*-tests were used to compare the 2 groups.
†Linear regression analyses adjusting for relevant baseline characteristics were used to compare the 2 groups.

As shown in Table 2, the PFM parameters at rest including the bladder neck position, the levator plate angle, the anorectal angle, and the levator hiatus area were not significantly different between the PVD1 and PVD2 subgroups ($P \geq .137$ unadjusted *t*-tests; $P \geq .327$ adjusted for duration of symptoms).

PFM morphometry during a maximal PFM contraction was based on the changes from baseline and are presented in Table 3. No significant differences were found between the 2 groups ($P \geq .551$ unadjusted *t*-tests; $P \geq .498$ adjusted for duration of symptoms).

PFM Function

Regarding PFM function, no significant difference was found between women with PVD1 and PVD2 using a MANOVA ($F(17,190) = 1.109$, $P = .348$; Wilk's $\Lambda = 0.910$, partial $\eta^2 = 0.090$). As for parameters evaluating PFM tone, *t*-tests and linear regression adjusting for duration of symptoms revealed no significant differences between the 2 groups (Table 4) ($P \geq .103$ unadjusted *t*-tests; $P \geq .239$ adjusted for duration of symptoms).

In terms of muscle contractility, as shown in (Table 5), women with PVD1 and PVD2 showed no significant difference in maximal strength, speed of contraction, control, and endurance ($P \geq .118$ unadjusted *t*-tests; $P \geq .144$ adjusted for duration of symptoms).

DISCUSSION

Given the growing evidence suggesting that PVD subgroups may be distinct entities, most of this evidence is based on studies with small sample sizes, there was a need to investigate the differential involvement of PFM alterations in women with PVD1 and PVD2 in a large sample to better understand their respective pathophysiological pathways. Therefore, the aim of this study was to compare PFM morphometry and function in women with PVD1 and PVD2. Although we hypothesized worse PFM alterations in women with PVD1, the findings revealed no significant differences in PFM morphometry and function between the 2 groups, as assessed with transperineal ultrasound and dynamometry, respectively. The results remained nonsignificant while controlling for duration of symptoms.

In the present sample, the proportion of women with PVD1 was 35%, which is in line with previous published data.[5,9] Also consistent with other studies,[6,13] the 2 groups had similar sociodemographic characteristics (age, BMI, education, income, and culture). The duration of symptoms was found to be longer in women with PVD1 compared with women with PVD2, which corroborates previous findings[4,14] and could be expected, given that women with PVD1 report pain starting from their first penetration attempt.

The results showed no significant difference between women with PVD1 and PVD2 for PFM morphometry at rest and during maximal contraction. Likewise, the PFM function including tone, strength, speed of contraction, and endurance was not found to be significantly different between the PVD subgroups. The results remained nonsignificant while controlling for duration of symptoms. The available studies have thus far focused on investigating PFM alterations in women with PVD in comparison to asymptomatic control subjects rather than comparing PVD subgroups.[21–26,44] The dynamometric and ultrasound parameters used in this study were shown to differ in women with PVD compared to the symptom-free control subjects in previous studies.[21,26] Several conceptual models have explained that PFM alterations may be a causal factor precipitating the onset of pain and could also act as a perpetuator of pain or a consequence of pain in the area.[44,45] Although the exact sequence of events remained unknown and a vicious cycle seemed most likely to happen, a recent study showed that PFM alterations were significantly associated with pain intensity in women with PVD.[27] It is therefore plausible that the similarity in pain intensity observed between the 2 subgroups may have contributed to the absence of significant differences regarding PFM alterations. Supporting the confounding influence of pain intensity, Maillé et al[8] reported that women with PVD1 had significantly more body image alterations than PVD2 but also found higher pain intensity in the PVD1 subgroup. Further analyses also revealed that body image alterations were significantly associated with pain intensity. Likewise, Brotto et al[14] found significant differences in sexual functioning between the 2 subgroups and observed a significant association between pain intensity and sexual function. As discussed in Pukall's review,[10] differences between PVD subgroups in biomedical and psychosexual variables can be influenced by the severity and the duration of vulvodynia, which was controlled in the present study. The absence of difference in PFM alterations in women with PVD1 and PVD2 observed in the current study contrasts with the results in other studies, suggesting a worse symptomatology in women with PVD1 in terms of biomedical and psychosexual outcomes.[3–8] However, none of these studies controlled for pain intensity or duration in their subgroup analyses. It is therefore difficult to discuss the potential influence of PVD severity on their findings. Another explanation could be that most of these studies included small sample sizes and clinical samples. The findings of the present study concur with a large cohort study of 441 women by Reed et al,[46] who conducted cluster analyses to determine data-driven subgroups based on the pain characteristics in women with vulvodynia. They observed that the presence of spontaneous pain and comorbid pain conditions were significant variables for subgroup separation while PVD1 and PVD2 were not found significant to delineate subgroups. Also in line with the current results, Aerts et al[9] showed no significant differences in the pain, sexual, or psychosocial profiles of 269 women with PVD1 and PVD2. Overall, these large studies do not support distinct pathophysiologic pathways in PVD1 and PVD2 in terms of PFM alterations, pain characteristics, or psychosexual variables. It is possible that other relevant factors may discriminate between PVD subgroups but further

rigorous studies are needed using a large sample and a sound methodology.

The current study allowed the investigation of PFM alterations in PVD subgroups using a large and mixed clinical and community sample providing more findings that are more representative of the general population of women with PVD than studies restricted to relatively small clinical samples. Following Pukall's[10] recommendations, analyses were adjusted for relevant variables such as duration of symptoms. Despite the strengths of the study, some limitations should be acknowledged. To ensure the homogeneity of the sample, we only included women that were nulliparous and below 45 years of age, which may have limited external validity. However, this prevented the introduction of bias related to obstetric injury to the PFMs and other types of pain such as vulvar atrophy in postmenopausal women. Having a confirmed diagnosis conducted by a gynecologist specialized in vulvodynia ruled out the possibility of including women affected by other genital pain that share similar symptoms (eg, vaginismus, deep dyspareunia, infections).

CONCLUSIONS

The present results advance our understanding of the involvement of PFMs in PVD1 and PVD2. No significant differences in PFM alterations were found between PVD1 and PVD2. In contrast with other studies reporting pathophysiologic differences,[4,6,7,10] we found that PFM alterations are similar across PVD subgroups. In light of these findings, it does not appear relevant to differentiate PVD1 and PVD2 subgroups from a PFM perspective. This is particularly important, given that pelvic floor physical therapy is suggested as a first-line treatment for PVD.[47] These results support the conclusion that the implication of PFM alterations in PVD are not affected by the timing of the onset of the symptoms, suggesting that similar physical therapy modalities can be offered to both subgroups.

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Conflict of Interest: The authors report no conflicts of interest.

Funding: None.

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REFERENCES

1. Bornstein J, Goldstein AT, Stockdale CK, et al. 2015 ISSVD, ISSWSH, and IPPS consensus terminology and classification of persistent vulvar pain and vulvodynia. **J Sex Med 2016;13:607-612.**
2. Harlow BL, Kunitz CG, Nguyen RH, et al. Prevalence of symptoms consistent with a diagnosis of vulvodynia: population-based estimates from 2 geographic regions. **Am J Obstet Gynecol 2014;210:40.e41-40.e48.**
3. Witkin SS, Gerber S, Ledger WJ. Differential characterization of women with vulvar vestibulitis syndrome. **Am J Obstet Gynecol 2002;187:589-594.**
4. Leclair CM, Goetsch MF, Korcheva VB, et al. Differences in primary compared with secondary vestibulodynia by immunohistochemistry. **Obstet Gynecol 2011;117:1307-1313.**
5. Bornstein J, Maman M, Abramovici H. "Primary" versus "secondary" vulvar vestibulitis: one disease, two variants. **Am J Obstet Gynecol 2001;184:28-31.**
6. Granot M, Friedman M, Yarnitsky D, et al. Primary and secondary vulvar vestibulitis syndrome: systemic pain perception and psychophysical characteristics. **Am J Obstet Gynecol 2004;191:138-142.**
7. Nguyen RH, Mathur C, Wynings EM, et al. Remission of vulvar pain among women with primary vulvodynia. **J Low Genit Tract Dis 2015;19:62-67.**
8. Maillé DL, Bergeron S, Lambert B. Body image in women with primary and secondary provoked vestibulodynia: a controlled study. **J Sex Med 2015;12:505-515.**
9. Aerts L, Bergeron S, Corsini-Munt S, et al. Are primary and secondary provoked vestibulodynia two different entities? A comparison of pain, psychosocial, and sexual characteristics. **J Sex Med 2015;12:1463-1473.**
10. Pukall CF. Primary and secondary provoked vestibulodynia: A review of overlapping and distinct factors. **Sex Med Rev 2016;4:36-44.**
11. Burrows LJ, Klingman D, Pukall CF, et al. Umbilical hypersensitivity in women with primary vestibulodynia. **J Reprod Med 2008;53:413-416.**
12. Heddini U, Bohm-Starke N, Nilsson KW, et al. Provoked vestibulodynia—Medical factors and comorbidity associated with treatment outcome. **J Sex Med 2012;9:1400-1406.**
13. Sutton KS, Pukall CF, Chamberlain S. Pain, psychosocial, sexual, and psychophysical characteristics of women with

primary vs. secondary provoked vestibulodynia. **J Sex Med 2009;6:205-214.**

14. Brotto LA, Sadownik LA, Thomson S, et al. A comparison of demographic and psychosexual characteristics of women with primary versus secondary provoked vestibulodynia. **Clin J Pain 2014;30:428-435.**
15. Goetsch MF. Vulvar vestibulitis: prevalence and historic features in a general gynecologic practice population. **Am J Obstet Gynecol 1991;164:1609-1614.**
16. Greenstein A, Sarig J, Chen J, et al. Childhood nocturnal enuresis in vulvar vestibulitis syndrome. **J Reprod Med 2005; 50:49-52.**
17. Leclair CM, Leeborg NJ, Jacobson-Dunlop E, et al. CD4-positive T-cell recruitment in primary-provoked localized vulvodynia: potential insights into disease triggers. **J Low Genit Tract Dis 2014;18:195-201.**
18. Babula O, Linhares IM, Bongiovanni AM, et al. Association between primary vulvar vestibulitis syndrome, defective induction of tumor necrosis factor-alpha, and carriage of the mannose-binding lectin codon 54 gene polymorphism. **Am J Obstet Gynecol 2008;198:101.e101-101.e104.**
19. Babula O, Bongiovanni AM, Ledger WJ, et al. Immunoglobulin E antibodies to seminal fluid in women with vulvar vestibulitis syndrome: relation to onset and timing of symptoms. **Am J Obstet Gynecol 2004;190:663-667.**
20. Lambert B, Bergeron S, Desrosiers M, et al. Introital primary and secondary dyspareunia: Multimodal clinical and surgical control. **Sexologies 2012;21:9-12.**
21. Morin M, Bergeron S, Khalife S, et al. Morphometry of the pelvic floor muscles in women with and without provoked vestibulodynia using 4D ultrasound. **J Sex Med 2014;11:776-785.**
22. ter Kuile MM, Both S, van Lankveld JJ. Cognitive behavioral therapy for sexual dysfunctions in women. **Psychiatr Clin North Am 2010;33:595-610.**
23. Bergeron S, Rosen NO, Morin M. Genital pain in women: Beyond interference with intercourse. **Pain 2011;152:1223-1225.**
24. Reissing ED, Binik YM, Khalife S, et al. Vaginal spasm, pain, and behavior: an empirical investigation of the diagnosis of vaginismus. **Arch Sex Behav 2004;33:5-17.**
25. Thibault-Gagnon S, Morin M. Active and passive components of pelvic floor muscle tone in women with provoked vestibulodynia: A perspective based on a review of the literature. **J Sex Med 2015;12:2178-2189.**
26. Morin M, Binik YM, Bourbonnais D, et al. Heightened pelvic floor muscle tone and altered contractility in women with provoked vestibulodynia. **J Sex Med 2017;14:592-600.**
27. 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. **Clin J Pain 2018. Mar 9. [Epub ahead of print].**
28. Bergeron S, Binik YM, Khalifé S, et al. Vulvar vestibulitis syndrome: reliability of diagnosis and evaluation of current diagnostic criteria. **Obstet Gynecol 2001;98:45-51.**
29. McLean L, Normandeau C, Hodder J. The impact of state of bladder fullness on tonic and phasic activation of the pelvic floor muscles in women. **J Electromyogr Kinesiol 2016; 27:60-65.**
30. Dietz HP, Shek C, Clarke B. Biometry of the pubovisceral muscle and levator hiatus by three-dimensional pelvic floor ultrasound. **Ultrasound Obstet Gynecol 2005; 25:580-585.**
31. Dietz HP, Wilson PD, Clarke B. The use of perineal ultrasound to quantify levator activity and teach pelvic floor muscle exercises. **Int Urogynecol J Pelvic Floor Dysfunct 2001;12:166-169.**
32. Kruger JA, Heap SW, Murphy BA, et al. Pelvic floor function in nulliparous women using three-dimensional ultrasound and magnetic resonance imaging. **Obstet Gynecol 2008;111:631-638.**
33. Thyer I, Shek C, Dietz HP. New imaging method for assessing pelvic floor biomechanics. **Ultrasound Obstet Gynecol 2008; 31:201-205.**
34. Majida M, Braekken IH, Umek W, et al. Interobserver repeatability of three- and four-dimensional transperineal ultrasound assessment of pelvic floor muscle anatomy and function. **Ultrasound Obstet Gynecol 2009;33:567-573.**
35. Braekken IH, Majida M, Engh ME, et al. Test-retest reliability of pelvic floor muscle contraction measured by 4D ultrasound. **Neurourol Urodyn 2009;28:68-73.**
36. Weinstein MM, Jung SA, Pretorius DH, et al. The reliability of puborectalis muscle measurements with 3-dimensional ultrasound imaging. **Am J Obstet Gynecol 2007;197; 68.e1-6.**
37. Dumoulin C, Bourbonnais D, Lemieux MC. Development of a dynamometer for measuring the isometric force of the pelvic floor musculature. **Neurourol Urodyn 2003;22:648-653.**
38. Morin M, Gravel D, Bourbonnais D, et al. Application of a new method in the study of pelvic floor muscle passive properties in continent women. **J Electromyogr Kinesiol 2010;20:795-803.**
39. Morin M, Gravel D, Bourbonnais D, et al. Reliability of dynamometric passive properties of the pelvic floor muscles in postmenopausal women with stress urinary incontinence. **Neurourol Urodyn 2008;27:819-825.**
40. Morin M, Dumoulin C, Gravel D, et al. Reliability of speed of contraction and endurance dynamometric measurements of the pelvic floor musculature in stress incontinent parous women. **Neurourol Urodyn 2007;26:397-404.**
41. Morin M, Bourbonnais D, Gravel D, et al. Pelvic floor muscle function in continent and stress urinary incontinent women using dynamometric measurements. **Neurourol Urodyn 2004;23:668-674.**
42. Morin M, Dumoulin C, Bourbonnais D, et al. Pelvic floor maximal strength using vaginal digital assessment compared

to dynamometric measurements. **Neurourol Urodyn 2004; 23:336-341.**

43. Dumoulin C, Gravel D, Bourbonnais D, et al. Reliability of dynamometric measurements of the pelvic floor musculature. **Neurourol Urodyn 2004;23:134-142.**
44. Zolnoun D, Hartmann K, Lamvu G, et al. A conceptual model for the pathophysiology of vulvar vestibulitis syndrome. **Obstet Gynecol Surv 2006;61:395-401.**
45. Thomten J, Linton SJ. A psychological view of sexual pain among women: applying the fear-avoidance model. **Womens Health (Lond) 2013;9:251-263.**
46. Reed BD, Plegue MA, Williams DA, Sen A. Presence of spontaneous pain and comorbid pain conditions identifies vulvodynia subgroups. **J Low Genit Tract Dis 2016;20:57-63.**
47. Goldstein AT, Pukall CF, Brown C, et al. Vulvodynia: Assessment and treatment. **J Sex Med 2016;13:572-590.**