Umbilical Hypersensitivity in Women With Primary Vestibulodynia

Lara J. Burrows, M.D., M.Sc., Daisy Klingman, M.D., Caroline F. Pukall, Ph.D., and Andrew T. Goldstein, M.D.

OBJECTIVE: To provide evidence that primary vestibulodynia (PV) is a congenital defect in tissue derived from the primitive urogenital sinus.

STUDY DESIGN: Twenty-two women with PV, 16 with secondary vestibulodynia (SV) and 8 controls were included in this study. Subjects underwent a complete history and physical examination, including assessment with a vulv algosimeter to measure the sensory and pain detection thresholds in the vulvar vestibule, deltoid and umbilicus.

RESULTS: The median vestibular sensitivity was 5 g in the PV group and 10 g in the SV group (p = 0.77). The median umbilical pain thresholds for the PV, SV and control groups were 115, 675 and 500 g, respectively. Women with PV displayed a significantly higher level of umbilical sensitivity (a substantially lower pain threshold) compared with women with SV and the control group (p = 0.0001 and 0.002, respectively). There was no difference in umbilical sensitivity between the SV and control groups.

CONCLUSION: Because both the umbilicus and vulvar vestibule are derived from primitive urogenital sinus, this suggests that women with PV may have a congenital abnormality in urogenital sinus–derived epithelium.

Keywords: pain measurement, pain threshold, umbilicus.

Vestibulodynia (formerly known as vulvar vestibulitis syndrome) is a common cause of sexual pain in women with a reported prevalence of up to 16%. Patients with this disorder typically experience severe introital dyspareunia that is frequently described as burning, cutting and/or searing on vaginal penetration.

This pain is localized specifically to the vulvar vestibule. In patients with primary vestibulodynia (PV), the pain has been present since the first tampon use or intercourse. Those
with secondary vestibulodynia (SV) have had painless tampon insertion or intercourse with subsequent development of vestibular pain; thus, this condition is acquired. It is important to be able to distinguish between these 2 disorders as their management and prognosis may differ considerably.4

The underlying pathophysiology of vestibulodynia (primary or secondary) has not been completely elucidated; however, several studies have identified a proliferation of C-afferent nociceptors in the vestibular mucosa.5-9 These studies have shown that there may be up to a 10-fold increase in the density of nerve endings in the vestibular mucosa of women with vestibulodynia compared with controls.6 This neuronal hyperplasia may at least partially explain the extreme allodynia experienced by women with this disorder. Other studies have suggested that altered central sensory processing may play a role in the development and/or maintenance of vestibulodynia, implying that this disorder is at least partially acquired.10 However, these studies did not differentiate between PV and SV. It is very possible that PV and SV may have entirely different etiologies, as some studies have indicated differences in medical history and pain characteristics in these 2 groups of vestibulodynia patients.11,12

The vulvar vestibule and the umbilicus are derived from the primitive urogenital sinus.13 When specifically questioned, many patients with PV report umbilical hypersensitivity. Given that the vulva and umbilicus are of the same embryologic origin, demonstrating both umbilical and vulvar hypersensitivity in patients with PV would lend support to the concept that PV may represent a congenital defect.

The aim of this study was to describe umbilical sensitivity in women with PV. The authors hypothesized that women with PV would exhibit significantly increased umbilical sensitivity compared with unaffected women and women with SV.

Materials and Methods

All patients were recruited from the office of the senior author (A.G.). This practice is devoted almost exclusively to the treatment of vulvovaginal disorders. This study received approval from the local institutional review board, and all patients signed an informed consent form before participation.

Patients who met criteria for PV and SV based on their history were offered participation in this study. The control group was composed of women who were seen for routine gynecologic care. The median ages of the PV, SV and control groups were 26, 27 and 25 years, respectively. All subjects were Caucasian with the exception of 1 African American woman (in the SV group). Five of the 8 (63%) controls, 11 of 22 (50%) of women in PV group and 9 of 16 (56%) of women in the SV group were using a form of hormonal contraception.

A vulvalgesiometer was used to determine tactile detection and pain thresholds in the vulvar vestibule and umbilicus. This mechanical device exerts standardized pressures through the use of springs with different compression rates. A disposable cotton swab attached to one end of the device is the only part of the instrument that comes into contact with the area being tested. This instrument exerts 3 g–1 kg of pressure and has been shown to replicate the burning pain that women with vestibulodynia often describe experiencing during sexual intercourse.14 The vulvalgesiometer has been used in previous studies examining pain sensation in women with vestibulodynia.15

The tactile detection threshold was defined as the pressure at which the participant first perceived that something was touching her but was not painful. The pain threshold was defined as the point at which she first detected the sensation of pain. To determine these thresholds, the lowest pressure of the vulvalgesiometer was first applied (i.e., 3 g) and consecutively higher ones were applied with an interstimulus interval of 15 seconds. Testing ended when the participant’s pain threshold was reached.

In this study, the vulvalgesiometer was used to determine tactile detection and pain thresholds of women with PV, SV and the control group in 3 areas of the body: over the right deltoid muscle, the center of the umbilicus and the posterior vulvar vestibule. These areas were tested in the above order for all participants in order to flow well with the rest of the general physical examination. Previous research has indicated that the order of testing at vulvar and nonvulvar sites does not affect threshold results.16 Results of vulvalgesiometer testing
and demographic data were recorded by a medical assistant on a separate form created for this study.

Data were analyzed using SPSS 12.0.1 (SPSS Inc., Chicago, Illinois). Descriptive analyses were conducted where appropriate. The Mann-Whitney U test was used to assess for significant differences in tactile detection and pain thresholds among the 3 groups.

Results
A total of 46 women participated in this study. There was no significant difference in use of oral contraceptive pills among the 3 groups. Twenty-two women had PV, 16 had SV and 8 were controls. The median threshold values for the 3 study groups are presented in Table I. The median umbilical pain thresholds for the PV, SV and control groups were 115, 675 and 500 g, respectively. Women with PV displayed a significantly higher level of umbilical sensitivity (a substantially lower pain threshold) compared with women with SV and the control group (p = 0.0001 and 0.002, respectively). There was no difference in umbilical sensitivity between the SV and control groups. There was also no difference in deltoid sensitivity among the three groups.

As expected, both the PV and SV groups had significantly lower vestibular pain thresholds compared with the control group (p = 0.0001 and 0.002, respectively). There was no difference in umbilical sensitivity between the SV and control groups. There was also no difference in deltoid sensitivity among the three groups.

Conclusion
Other authors have suggested that the coexistence of vestibulodynia and interstitial cystitis is evidence that these conditions may represent a generalized disorder of urogenital sinus–derived epithelium.17 As this association has been found in patients as young as 4 years old, they theorized that these conditions may represent a congenital defect, specifically in urogenital sinus–derived epithelium. The most important finding of the current study was that women with PV had a significant increase in umbilical sensitivity (a substantially lower pain threshold) compared with women with SV and a control group. These data give further merit to the theory of a congenital defect in tissue derived from the primitive urogenital sinus in this subset of women with vestibulodynia.

Other explanations or contributing factors may at least partially explain our findings. Pukall et al10 found that patients with vestibulodynia may have a systemic (i.e., nonlocalized) hypersensitivity to tactile and pain stimuli. Thus, a generalized altered perception of pain may contribute to or explain the increased umbilical sensitivity in women with vestibulodynia. However, this would not explain why there was no difference in pain perception in the deltoid region between the groups. Alternatively, Harlow and Stewart19 found that vulvodynia is more likely to develop in women with a history of physical or sexual victimization. It is possible that a woman with vestibulodynia and with a history of victimization may perceive umbilical palpation as invasive and have a lower umbilical pain threshold. In addition, physical therapists who treat vulvar and pelvic pain frequently encounter myofascial trigger points and pain in the anterior abdominal wall.20 These explanations could account for umbilical hypersensitivity in women with vestibulodynia, but they do not fully account for the differences between women with PV and SV found in the present study. Future research should investigate these 2 subsets of vestibulodynia separately to more fully understand the differences in these 2 presentations of vestibulodynia.

As expected, women with vestibulodynia were significantly more sensitive to vestibular pain compared with control women. This finding has been reported in other studies (e.g., Pukall et al10) and is consistent with the clinical presentation of patients

<table>
<thead>
<tr>
<th>Group</th>
<th>Tactile detection</th>
<th>Pain</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Deltoid</td>
<td>Umbilical</td>
</tr>
<tr>
<td>PV</td>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>SV</td>
<td>10</td>
<td>12.5</td>
</tr>
<tr>
<td>Control</td>
<td>5</td>
<td>7.5</td>
</tr>
</tbody>
</table>

Table I  Median Vestibular Tactile Detection and Pain Thresholds in Women With PV, SV and Control Women Over 3 Body Areas
with vestibulodynia. It is likely that vestibular hyperinnervation can at least partially explain this result.\textsuperscript{6-9} Interestingly, women with PV exhibited significantly lower vestibular pain thresholds than women with SV. A potential explanation for this unique finding could be altered hormonal status. Bohm-Starke et al\textsuperscript{21} found increased vestibular mucosa sensitivity in women who used oral contraceptives compared with women not taking such contraceptives. However, there was no difference in our sample between the PV and SV groups with respect to oral contraceptive use. It is likely that women with PV may have a more severe form of vestibulodynia than women with SV; however, further research is needed to explore this finding more fully.

Limitations of this study include a relatively small sample size and control group. The greatest limitation was in making the diagnosis of primary versus secondary vestibulodynia, as this is strictly a clinical diagnosis and therefore we cannot be completely certain that patients were categorized appropriately.

In conclusion, our data show that women with PV display umbilical hypersensitivity, implying that this may be a congenital disorder. The notion that PV and SV have different etiologies is a relatively new one and to date, most studies on vestibulodynia have not differentiated between these 2 categories. Our findings also suggest that testing umbilical hypersensitivity in patients with vestibulodynia may aid in diagnosis and have a significant impact on the subsequent management of these patients.

References