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A longitudinal case-control analysis of pain symptoms, fear of childbirth, and psychological well-being during pregnancy and postpartum among individuals with vulvodynia

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ABSTRACT

Objective: Little research has examined changes in chronic vulvar pain (vulvodynia) symptoms with pregnancy and childbirth, nor fear as it relates to pregnancy/delivery amongst individuals with vulvodynia. The purpose of this study was to examine change in pain symptoms from pregnancy to postpartum amongst women with vulvodynia, as well as pain anxiety, fear of childbirth, and anxiety and depressive symptoms.
Design: Prospective Case-Control Study.
Setting: Online survey.
Participants: Fifty-Seven pregnant individuals with a diagnosis of vulvodynia, and 41 pregnant control participants who reported being free of vulvar pain. Participants were recruited from the community and from hospital-based clinics for this study.
Measurements and Findings: Online surveys were administered to women diagnosed with vulvodynia and pain-free control participants during pregnancy and at three and six months postpartum. The survey contained both investigator-developed items and validated questionnaires, including the Pain Anxiety Symptoms Scale (PASS-20), the Wijma Delivery Expectancy/Experience Questionnaire (W-DEQ) to assess fear of childbirth, the Generalized Anxiety Disorder-7 (GAD-7) measure to assess symptoms of anxiety, and the Patient Health Questionnaire (PHQ-9) to assess symptoms of depression. Linear mixed models with random intercepts for longitudinal analyses indicated statistical improvements for most of the vulvar pain outcomes in the postpartum period amongst women with vulvodynia, including reduced pain intensity at three ($p = 0.005$) and six months ($p = 0.013$) postpartum for those women who delivered vaginally. The mean change in pain intensity corresponded though to only a minimal clinical change. Compared to controls, women with vulvodynia reported higher levels of fear of childbirth on the W-DEQ ($p = 0.024$). In both groups, increases in general anxiety on the GAD-7 were found from pregnancy to three ($p = 0.005$) and six months ($p = 0.033$) postpartum. Mode of birth moderated the findings for pain-related anxiety as measured by the PASS-20: only individuals who delivered via caesarean section reported increases in pain anxiety between pregnancy and six months postpartum ($p < 0.001$).
Key Conclusions: Pregnant women with vulvodynia experienced postpartum improvements in vulvar pain symptoms. Mode of birth may play a role in symptom trajectory.
Implications for Practice: Individuals with vulvodynia often have concerns about how pregnancy and childbirth will impact their symptoms. The current findings can be used to help such individuals make reproductive decisions knowing there may be improvements in vulvar pain and increases in anxiety that can occur postpartum. The statistical versus clinical significance of the pain intensity results also highlight the importance of asking each individual what changes in pain symptoms they experience and the meaning of such changes for that person.

Introduction

Vulvodynia is distressing vulvar pain, lasting three months or longer, without a clearly identifiable cause (Bornstein et al., 2016).

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It is estimated to occur in seven to eight percent of females of reproductive age (Harlow et al., 2014). Individuals with vulvodynia often describe a burning, stabbing, and/or rawness in their vulvar area, and many report pain with sexual activity. Provoked Vestibulodynia (PVD) is the most common type of vulvodynia and is characterized by severe, localized pain upon contact to the vaginal opening (Sadownik, 2014).

Vulvodynia is commonly associated with substantial reduction in quality of life. High levels of anxiety about pain are often present (e.g., Govind et al., 2020), as well as elevated rates of anxiety, mood, and post-traumatic stress disorders (Iglesias-Rios et al., 2015; Khandker et al., 2011). Sexual difficulties, such as reduced desire, are also common amongst persons with vulvodynia (Desrochers et al., 2008), and affected individuals report feeling shame, guilt, and inadequacy as women and sexual partners (Ayling and Ussher, 2008; Shallcross et al., 2018). Individuals with vulvodynia often avoid, or endure with distress, activities like gynaecological examinations and penetrative sex due to pain. Almost all (87.8%) patients receiving treatment for PVD in a multidisciplinary program reported that sexual penetration was not possible at baseline at least occasionally because of vulvar pain (Brotto et al., 2015b).

Given the impact of vulvodynia on sexual and psychological health, it is not surprising that fertility and pregnancy experiences may also be affected. For example, some research suggests that people with vulvodynia may be more likely to be nulliparous compared to persons without such pain (Edgardh and Abdelnoor, 2007; Möller et al., 2015). Many affected individuals still have a desire to conceive, however (George et al., 2019), and vulvodynia likely does not have a biological effect on fertility (Nguyen et al., 2012). Instead, fertility may be indirectly impacted by factors such as reduced frequency of sexual intercourse or increased fear of childbirth. When people with vulvodynia do become pregnant, they may experience anxiety regarding pregnancy and delivery. Johnson et al. (2015) interviewed 18 women with vulvodynia who were either currently pregnant or who had given birth within the past six to 12 months, with most reporting anxiety about how pregnancy may negatively impact their vulvar pain symptoms.

Overall, however, little research has examined anxiety as it relates to pregnancy or birth amongst persons with vulvodynia or regarding how vulvodynia symptoms may change with pregnancy and childbirth. Furthermore, patients may be told that their vulvodynia symptoms will improve or be cured (Johnson et al., 2015), but available data stems from studies containing small numbers of pregnant women with vulvodynia. Reed et al. (2003) reported that 17 of the 104 women in their sample became pregnant when experiencing vulvar pain; of these women, seven reported that pregnancy did not affect their pain, five reported improved pain symptoms associated with pregnancy, and five reported worsened pain symptoms. Another study reported that five of 230 women with PVD became pregnant during the study period, with all five reporting improved pain symptoms during pregnancy (Pagano, 1999). More research is needed to understand the trajectory of vulvodynia symptoms from pregnancy to postpartum and to understand how vulvodynia may affect psychological experiences related to pregnancy and birth.

The main objective of this prospective case-control study was to: 1) assess changes in vulvar pain symptoms from pregnancy to postpartum amongst individuals who had been diagnosed with vulvodynia. The additional objectives were to compare individuals with vulvodynia and pain-free control cases with regard to: 2) levels of pain anxiety; 3) fear of childbirth; 4) symptoms of depression; and 5) symptoms of anxiety in the pregnancy and postpartum periods. These latter symptoms were examined given the impact of vulvodynia on quality of life. As reviewed above, prior studies based on small numbers of pregnant women with vulvodynia reported improvement in pain symptoms with pregnancy for approximately one-third (Reed et al., 2003) to all (e.g., Pagano, 1999) participants. However, given that birth is often associated with acute genital and pelvic pain, and is a risk factor for developing persistent pain of this type in non-vulvodynia samples (Cappell and Pukall, 2017; Rosen and Pukall, 2016), we hypothesized that vulvar pain symptoms, including pain intensity, would increase in the postpartum period for persons with vulvodynia. We also hypothesized that participants with vulvodynia would report higher levels of pain anxiety; fear of childbirth; and anxiety and mood symptoms in the pregnancy and postpartum periods compared to controls.

Methods

Participants

Pregnant individuals were recruited for this study starting in April 2013, with data collection continuing until February 2016. This study was based at a Canadian university and affiliated hospital; given the online nature of the study, however, recruitment was not restricted to participants from a certain location. Participants for both groups were recruited using various strategies, including posters provided to physician and midwifery clinics and placed in local hospitals, website advertisements (e.g., senior author's research website), and word of mouth. Recruitment strategies for the vulvodynia group also included vulvar pain-related website advertisements (e.g., National Vulvodynia Association; Vulval Pain Society), and recruitment announcements from the National Vulvodynia Association to their list of patients. In addition, patients who were diagnosed with vulvodynia at investigator-affiliated clinics were identified using existing research databases and notified about the study. Although we did not require a specific gender identity, our recruitment strategies stated we were seeking pregnant women for this study; as such, the samples will be referred to as women in the remainder of this paper.

Pregnant women were included if: 1) they had ever been diagnosed with vulvodynia, or 2) they reported being free from chronic or recurrent vulvar pain. In this study, we operationalized vulvar pain as "pain that is in or on the vulva, at the vaginal opening, and/or pain with sexual intercourse or other activities involving vaginal penetration". Specifically, women with vulvar pain were included if they reported receiving a diagnosis of PVD (or its former term, vulvar vestibulitis syndrome), generalized vulvodynia, or both by a healthcare provider; women with current or past vulvar pain symptoms could be included in this group.

Women in the control group reported that they did not currently experience chronic or recurrent vulvar pain, did not experience such pain at the time they became pregnant, and had never experienced three months or more of such pain (as pain is often considered "chronic" when lasting three months or longer, e.g., Treede et al., 2019). In addition, these women reported that they had never been diagnosed with a chronic or recurrent vulvar pain condition, nor with Vaginismus. We operationalized Vaginismus as "an inability to have vaginal intercourse and/or other vaginal penetration". We also informed women at the time of recruitment that people with Vaginismus are sometimes told they have an "involuntary spasm of the vagina that interferes with intercourse and/or penetration".

In addition to being pregnant at recruitment, all participants were required to be 19 years of age or older and fluent in English. Women were excluded if they: 1) reported experiencing vulvar pain without a diagnosis; 2) reported receiving a diagnosis of another vulvar condition causing pain (e.g., lichen sclerosus) but not vulvodynia; 3) did not reliably report having vulvar pain for three months or more; 4) were not fluent in English; or 5) were

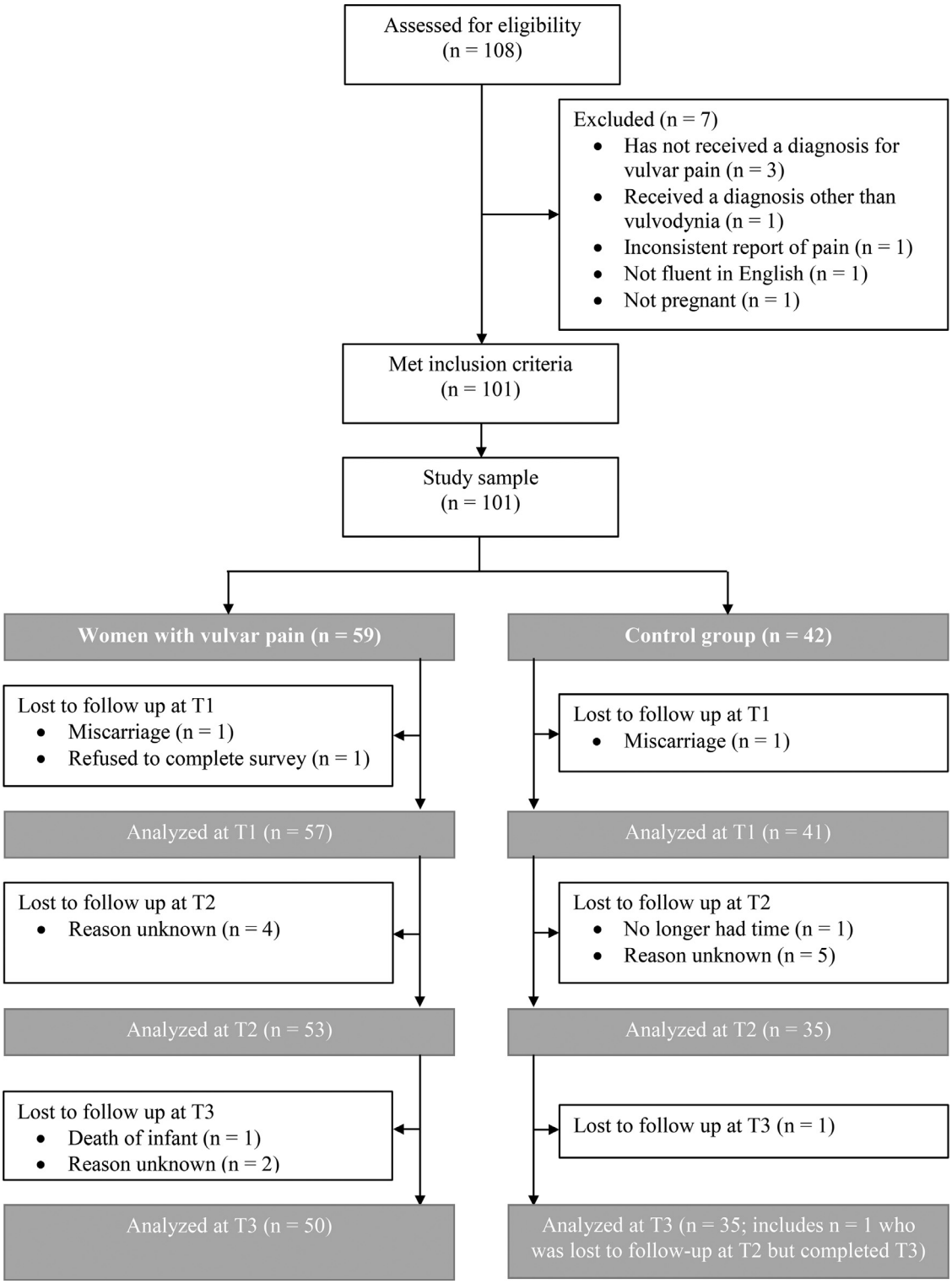

Fig. 1. Flow of participation.

not pregnant at the time of study enrolment. Fig. 1 shows the flow of participation in this study.

This study utilized a convenience sampling method, with sample size dependant on the number of people who responded to our recruitment strategies. The power analysis/sample calculation was conducted for linear mixed model longitudinal analysis using GLIMMPSE software (Kreidler et al., 2013). The results indicated that in order to have 0.90 power to find a significant at $p < 0.05$ level medium size time by treatment interaction effect while assuming monotonic decrease with time in correlation between repeated measures a sample of 96 participants is needed.

Procedures

This was an online, prospective case-control study. Women who were interested in participating were asked to contact the study

coordinator for more information regarding the study. They then completed a telephone screening interview to determine study eligibility. Those persons who met criteria were subsequently sent a link to an online survey (using SurveyMonkey™) at three different time points corresponding with the second or third trimester (T1) before term delivery (i.e., 13–40 weeks), and at three months (T2) and six months (T3) postpartum. Three months was chosen as a follow-up point because pain is often considered chronic when lasting three months or longer (Treede et al., 2019). A six month follow-up was also chosen to assess whether vulvar pain changes occur over a longer period of time following pregnancy and delivery. Other prospective studies of people with vulvodynia have also utilized a six month follow-up (e.g., Brotto et al., 2015a).

Each participant was emailed a unique link to the survey in the few weeks preceding a timepoint; reminders (up to three at each timepoint) were provided if needed to try and increase response rate and minimize non-response bias. We considered a survey lost to follow-up following three reminders. We also emailed participants following their expected due date and sent a congratulatory e-card if a participant informed us their baby had been delivered. Each survey took approximately 20 min or less to complete at each timepoint. An electronically administered consent form was provided in the first survey, and participants were only able to access the survey after informed consent was obtained.

All participants were provided with the option to decline responding to any of the survey questions, and were informed of this option during the consent process. Participants were provided with an electronic $10 gift card for each timepoint in which they participated, for a maximum total of $30 per participant. This study was approved by research ethics board at the University of British Columbia and the Vancouver Coastal Health Research Institute.

Measures

Demographics Questionnaire. This investigator-derived questionnaire asked participants to report various demographic information at T1, including age, relationship status and length, ethnicity, sexual orientation, level of education, and income. This questionnaire also queried number of children, pregnancies, and births, including number of previous caesarean sections.

Labour and Birth Experiences. Participants were asked at T2 whether they had a vaginal or caesarean birth for their recent birth, and about the use of pain medications during their recent labour and birth.

Vulvar Pain Intensity and Distress. At each timepoint, participants were asked if they had regularly experienced vulvar pain since either becoming pregnant with their current pregnancy (T1) or since completing the previous survey in this study (T2 and T3). Participants who answered yes were then asked to report the overall intensity of the vulvar pain they experienced on a Numerical Rating Scale (NRS) of zero (no pain at all) to 10 (worst pain possible), as well as the overall level of distress they experienced about their vulvar pain on a zero (no distress at all) to 10 (most distress possible) NRS.

Change in Vulvar Pain Symptoms. Participants at all timepoints who reported regularly experiencing vulvar pain since becoming pregnant (T1) or since the previous survey (T2 and T3) were asked whether they felt their vulvar pain symptoms had since decreased, increased, or stayed about the same.

Pain or Problems During Intercourse/Penetration: One item from the Patient Health Questionnaire-SADS (Kroenke et al., 2010) was included to assess how much participants had been bothered by pain or problems during sexual intercourse during the past 4 weeks. We adapted this item slightly by changing the term "sexual intercourse" to "sexual intercourse/penetration". This item was assigned a score of zero (not bothered), 1 (bothered a little), or 2 (bothered a lot), and was administered at each timepoint.

Pain Anxiety Symptoms Scale (PASS-20). This 20-item measure was administered at each time point (McCracken and Dhingra, 2002). The PASS-20 contains four subscales to measure cognitive anxiety, escape and avoidance, fearful thinking, and physiological anxiety; however, only the total score was used in this study. Each item is rated on a scale from zero (never) to five (always), with higher scores indicating higher levels of anxiety. The PASS-20 has good internal consistency, reliability, and convergent and divergent validity (McCracken and Dhingra, 2002). It has been used in previous research with pregnant women to examine predictors of postpartum genital/pelvic pain (Glowacka et al., 2014). Cronbach's alpha at baseline in the present study was 0.94.

Wijma Delivery Expectancy/Experience Questionnaire (W-DEQ). The W-DEQ is a widely used measure of fear of childbirth (Wijma et al., 1998). There are two versions of the W-DEQ: version A for use in pregnancy and version B for use in the postpartum. In the current study, version A of the W-DEQ was administered at T1 and version B was administered at T2. Version A asks women to answer how they imagine they will feel during labour and birth; version B asks women to retrospectively answer how they thought their labour and birth actually went. Each version of the W-DEQ contains 33 items which were rated on a scale of one to six. Cronbach's alpha at baseline in the present study was 0.93.

Generalized Anxiety Disorder (GAD)−7. This seven-item measure is widely used to screen for generalized anxiety disorder (GAD; Spitzer et al., 2006). It asks respondents to indicate how much they have been bothered by specific problems (e.g., trouble relaxing) over the past two weeks, with each item scored on a scale from zero (not at all) to three (nearly every day). The GAD-7 has shown good reliability, and is considered a valid instrument for screening for and assessing severity of generalized anxiety symptoms (Spitzer et al., 2006). The GAD-7 has shown clinical utility for screening for GAD during pregnancy and postpartum (Simpson et al., 2014). It was administered at all timepoints in the current study, and Cronbach's alpha at baseline was 0.89.

Patient Health Questionnaire (PHQ)−9. The PHQ-9 is a common tool used to assess for severity of depressive symptoms (Kroenke et al., 2001). It asks respondents to indicate how much they have been bothered by specific problems (e.g., poor appetite or overeating) over the past two weeks, with each item scored on a scale from zero (not at all) to three (nearly every day). The PHQ-9 demonstrates reliability and validity (Kroenke et al., 2001), and is effective at identifying pregnant women with depression (Sidebottom et al., 2012). It has also shown high specificity for identifying depression in the postpartum period (Gjerdingen et al., 2009). It was administered at all timepoints in the current study. Cronbach's alpha at baseline in the present study was 0.85.

Data analysis

The descriptive statistics were calculated for all collected variables representing demographic characteristics, reproduction history, and the circumstances of the birth covered by this study. The two study groups were compared on all of those variables using independent sample *t*-test for continuous variables and chi-square test for categorical variables. Significant group differences are reported below together with the test statistics. Changes in outcomes from pre-birth (T1) to three months postpartum (T2) and six month follow-up (T3) were examined using linear mixed model (LMM) method which analysed the means of continuous outcomes and generalized linear mixed model (GLMM) using logit link function for binomial (vulvar pain, change in vulvar pain) outcomes. All

models were tested with random intercepts only (random effects of time were not included because time was treated as a categorical variable with baseline as a reference category). Therefore, all the final models reported here were conducted with only the random intercept effect in addition to the fixed effects of time, group, and moderators as categorical variables. Vulvodynia status (women with vulvodynia versus control group), mode of birth, and use of pain medication during labour/birth were examined as between-subject moderators of within-subject changes between time points. Each outcome was examined by two models: one including time, vulvodynia vs. control group, and mode of birth (caesarean vs. vaginal) and one including time, vulvodynia vs. control group, and use of pain medication (yes/no). Each model included all three main effects, all three two-way interactions, and the three-way interaction. When none of the effects involving mode of birth or pain medication were significant the models were re-run with just the time and group main effects and interaction.

In addition, since the number of pregnancies was higher in the control group (see Sample Characteristics in the Findings section) and might be related to the longitudinal outcomes, all models were also analysed with pregnancy number as a covariate; if the pattern of significant results was changed (it happened for only one outcome), that change was reported in the Findings section.

For the linear mixed models, the assumptions of normality and homoscedasticity of residuals were evaluated through visual examination of q-q plots and residuals plotted against the fitted values and no violations were observed. For the GLMM models, the most important diagnostics relate to the random effect structure which should be normally distributed and have constant variance across participants. Our evaluation of the q-q plots of the random intercepts from the two GLMM models did not indicate deviation from normality and the overdispersion tests examining consistency of the variance were not significant ($p = 0.999$ for vulval pain and $p = 0.402$ for change in vulval pain) indicating no overdispersion of variance. All analyses were conducted using IBM SPSS v.26 software and R software, and p values equal to or smaller than 0.05 were considered to indicate statistically significant findings.

Findings

Sample characteristics

The flow of recruitment and number of participants included at each stage of the study is shown in Fig. 1. The descriptives for both groups, women with vulvodynia ($n = 57$) and pain-free controls ($n = 41$) are included in Table 1. Participants' age ranged from 24 to 44 years with an average age of 33 years. Women in the control group were more likely to be in a common-law (versus married) relationship than women with vulvodynia (X^2 (1, $N = 94$) = 8.17, $p = 0.007$), and the mean relationship length in the sample was over seven years. The majority of women identified as White and almost all identified as heterosexual. Almost all women also reported at least some college education, with more women with vulvodynia reporting post-graduate education (56%) compared to women in the control group (24%; X^2 (4, $N = 98$) = 12.00, $p = 0.017$). About 80% of women reported an annual income of $60,000 or higher. Over half of the women reported having no children at T1 of the study (56%) while the rest of the participants reported one to three children. Those in the control group reported a higher number of pregnancies than women in the vulvodynia group (t(95) = 2.33, $p = 0.024$). The majority of participants used pain medication during their labour/birth in this study (80.7%) and almost one-third (31.5%) gave birth via caesarean section. Amongst the women with vulvodynia, 84.2% ($N = 48$) reported that they were regularly experiencing vulvar pain before their current pregnancy.

A total of 10 participants dropped out of the study at T2 (four with vulvodynia and six in the control group) and it increased to 13 at T3 due to an additional three dropouts in the vulvodynia group. Comparison of dropouts to those who remained in the study indicated no differences on any characteristics listed in Table 1 and no differences on baseline outcome measures. We assumed the missing data in our study to be MAR (missing at random) and therefore believe that the full information maximum likelihood estimation method adequately deals with missingness since in that method all available information is utilized for parameter estimation (Schafer and Graham, 2002).

Time between baseline and T2 ranged from 56 to 332 days with 92% of participants taking three to nine months between these two data collection points. The potential impact of this wide range of time on the outcome variables was evaluated by correlating the number of days between baseline and T2 with all outcome measures at T2 and T3. Length of time between study points correlated significantly with vulval pain presence indicating that participants with more days between baseline and T2 were more likely to report vulvar pain at T2 ($r = 0.28$, $p = 0.009$); and it correlated with change in vulvar pain symptoms such that participants with longer time between baseline and T2 were more likely to report that vulvar pain stayed the same or decreased at T3 ($r = 0.38$, $p < 0.001$). For those two outcomes, the longitudinal analyses were re-run with days between baseline and T2 as a covariate. The pattern of results was not affected indicating that length time between study points did not impact on pre- to postpartum changes in the study outcomes.

Pain-Related outcomes

Table 2 shows the estimated marginal means and the overall F test values and associated p values for linear mixed model analyses examining the effect of time on all outcomes in the vulvodynia versus control groups moderated by birth mode (vaginal vs. caesarean birth). The use of pain medication during labour and birth did not moderate any effects and is not shown in the table. The intra-class correlations were high, ranging from 0.41 to 0.79, (as would be expected for longitudinal models where repeated measurements are coming from the same participant) and underscore the importance of using mixed models including random intercept effects.

Vulvar Pain: The analysis of proportions of women who indicated having regularly experienced vulvar pain revealed significant time by group interaction and no effects related to birth mode or pain medication use. The simple slope analysis indicated that the proportion of participants reporting regular pain decreased in the vulvodynia group from 76% to 58% between T1 and T3 ($p = 0.050$) whereas it increased in the control group from two% to 20% between T1 and T2 ($p = 0.020$) and stayed somewhat higher than T1 at T3 (14%, $p = 0.073$).

The next three variables, vulvar pain intensity, distress, and change, were measured only for those who indicated regular vulvar pain. Since only one woman in the control group reported regular vulvar pain at pre-birth, the longitudinal analyses for these three variables were conducted only for the vulvodynia group.

Vulvar Pain Intensity & Distress: The time by birth mode interaction was found to be significant for vulvar pain intensity. Simple slope examination indicated that the intensity of participants' vulvar pain decreased between T1 and T2 by one point on a zero to 10 point NRS and between T1 and T3 by one point (Cohen's $d = 0.47$ – medium effect size) for women with vulvodynia who gave birth vaginally ($p = 0.005$ and 0.013, respectively), whereas there was no decrease in intensity for participants who gave birth via caesarean section. Changes in vulvar pain intensity across time were not moderated by the use of pain medication during labour/birth.

Table 1
Characteristics of participants.

Measure	Controls	Women with Vulvodynia	Total
Number of participants	41	57	98
Age (years), mean ± SD (missing $n = 1$)	32.78 ± 4.58	32.93 ± 4.00	32.87 ± 4.23
Relationship status, N (%)			
Single	0	1 (1.8)	1 (1.0)
Dating	0	2 (3.5)	2 (2.0)
Married	30 (73.2)	50 (87.7)	80 (81.6)
Common-Law	11 (26.8)	3 (5.3)	14 (14.3)
Separated	0	0	0
Divorced	0	0	0
Widowed	0	0	0
Other	0	1 (1.8)	1 (1.0)
Length of relationship (years), mean ± SD (missing $n = 4$)	6.90 ± 4.18	7.85 ± 3.37	7.45 ± 3.75
Ethnicity, N (%)			
White	28 (68.3)	48 (84.2)	76 (77.6)
Chinese	4 (9.8)	1 (1.8)	5 (5.1)
South Asian	1 (2.4)	3 (5.3)	4 (4.1)
Black	0	1 (1.8)	1 (1.0)
Latin American	1 (2.4)	0	1 (1.0)
Southeast Asian	1 (2.4)	0	1 (1.0)
Japanese	0	1 (1.8)	1 (1.0)
Korean	2 (4.9)	0	2 (2.0)
Multiple ethnicities	4 (9.8)	3 (5.3)	7 (7.1)
Sexual orientation, N (%)			
Heterosexual	40 (97.6)	55 (96.5)	95 (96.9)
Bisexual	1 (2.4)	1 (1.8)	2 (2.0)
Lesbian	0	1 (1.8)	1 (1.0)
Other	0	0	0
Education, N (%)			
Attended some high school	0	0	0
Graduated high school or earned GED[a]	2 (4.9)	0	2 (2.0)
Attended some college	5 (12.2)	5 (8.8)	10 (10.2)
Graduated 2 year college	4 (9.8)	2 (3.5)	6 (6.1)
Graduated 4 year college and/or university undergraduate degree completed	20 (48.8)	18 (31.6)	38 (38.8)
Post-Graduate degree	10 (24.4)	32 (56.1)	42 (42.9)
Annual household income N (%) (missing $n = 8$)			
Less than $20,000	0	0	0
$20,000-$39,999	2 (4.9)	3 (5.3)	5 (5.1)
$40,000-$59,999	4 (9.8)	2 (3.5)	6 (6.1)
$60,000-$79,999	11 (26.8)	12 (21.1)	23 (23.5)
$80,000-$99,999	4 (9.8)	7 (12.3)	11 (11.2)
$100,000 or more	16 (39.0)	29 (50.9)	45 (45.9)
Prefer not to answer	4 (9.8)	4 (7.0)	8 (8.2)
Number of children, mean ± SD	0.68 ± 0.93	0.54 ± 0.71	0.60 ± 0.81
Number of pregnancies, mean ± SD (missing $n = 1$)	1.58 ± 2.12	0.74 ± 0.97	1.08 ± 1.60
Number of deliveries, mean ± SD	0.68 ± 0.93	0.44 ± 0.60	0.54 ± 0.76
Number of deliveries by caesarean, mean ± SD	0.20 ± 0.60	0.14 ± 0.35	0.16 ± 0.47
Use of pain medication during delivery, N (%)[b]			
Yes	30 (85.7)	41 (77.4)	71 (80.7)
No	5 (14.3)	12 (22.6)	17 (19.3)
Mode of birth, N (%)[b]			
Vaginal	25 (71.4)	34 (66.7)	59 (68.6)
Caesarean (Total)	10 (28.6)	17 (33.3)	27 (31.4)
Caesarean (Planned)	4 (11.4)	9 (17.6)	13 (15.1)

Note: [a]GED = General Education Development. [b] Missing data due to T2 drop-out $n = 10$ plus $n = 2$ no response for mode of birth and $n = 3$ no response for planned caesarean.

Distress associated with vulvar pain showed no significant effects of time and no interaction of time and either of the two moderators.

Change in Vulvar Pain Symptoms. There was a significant T1 to T3 increase (30%, $p = 0.005$) in the proportion of women in the vulvodynia group who reported that their vulvar pain decreased or stayed the same as compared to the beginning of pregnancy or previous data collection time. This main effect of time was not moderated by either birth mode or use of pain medication during labour/birth.

Pain or Problems During Intercourse/Penetration: The analysis of pain or problems during sexual intercourse/penetration on the PHQ revealed two significant two-way interactions of time by group and group by birth mode. The simple effect examination of the first interaction indicated that the pain/problems decreased significantly between T1 and T2 and between T1 and T3 for women with vulvodynia (1.16 vs 0.88, $p = 0.008$, Cohen's $d = 0.34$, and 1.16 vs 0.85 $p = 0.005$, Cohen's $d = 0.38$, respectively), whereas they did not change for women in the control group (0.17, 0.36, and 0.31, for T1, T2, and T3, $ps > 0.05$). Simple effects follow-up of the significant group by birth mode interaction indicated that, averaging across all three time points, women with vulvodynia reported a higher level of pain/problems compared to participants in the control group (1.04 vs 0.16, $p < 0.001$, Cohen's $d = 1.06$,) only amongst women who birthed vaginally. There was no difference between the two groups regarding sexual pain or problems amongst women who gave birth via caesarean section (0.81 versus 0.48, $p = 0.113$). The use of pain medication during labour/birth did not moderate any of the time or group effects.

Table 2
Estimated marginal means for linear mixed model analyses examining effect of time (pregnancy, three months postpartum, six months postpartum) on study outcomes in vulvodynia versus control group moderated by birth mode (vaginal versus caesarean section).

	Time 1		Time 2		Time 3			Main effects		2-way interaction			3-way interaction
Measure	VV	Cntrl	VV	Cntrl	VV	Cntrl	Time	Grp	BM	Time x Grp	Time x BM	Grp x BM	Time x Grp x BM
Vulvar pain[a] (0=no, 1=yes)								*F*	*F*	*F*	*F*	*F*	*F*
								p	*p*	*p*	*p*	*p*	*p*
Vaginal	.77	0.00	.74	.16	.66	.12	.20	22.99	.00	**5.59**	.36	.00	.01
C-section	.71	.10	.71	.29	.47	.11	.264	<0.001	.978	**.019**	.700	.974	.989
Missing n	0	1	6	6	7	6							
Vulvar pain intensity (0–10)													
Vaginal	5.53	–	4.47	–	4.57	–	1.28	–	1.79	–	**5.13**	–	–
C-section	5.32	–	6.34	–	5.25	–	.286	–	.187	–	**.009**	–	–
Missing n	0		6		7								
Vulvar pain distress (0–10)													
Vaginal	5.50	–	4.57	–	4.75	–	.89	–	.20	–	1.33	–	–
C-section	5.31	–	5.63	–	4.87	–	.416	–	.658	–	.271	–	–
Missing n	0		6		7								
Vulvar pain change (0=higher, 1=same/ lower)[b]													
Vaginal	0.58	–	0.75	–	0.81	–	**3.12**	–	.27	–	.74	–	–
C-section	0.50	–	0.50	–	0.88	–	**.048**	–	.605	–	.480	–	–
Missing n	0		7		7								
Pain during penetration (0–2)													
Vaginal	1.27	0.08	0.86	0.23	0.96	0.16	.19	22.72	.17	**3.75**	1.45	**4.95**	.52
C-section	0.85	0.30	0.88	0.60	0.68	0.56	.827	<0.001	.682	**.026**	.237	**.029**	.593
Missing n	2	0	7	7	12	7							
PASS-20 (0–100)													
Vaginal	36.26	35.00	39.85	33.61	36.08	35.92	**5.95**	0.00	0.10	1.65	**5.00**	0.42	1.28
C-section	30.88	29.90	33.71	36.20	36.53	42.28	**0.003**	0.986	0.755	0.196	**0.008**	0.520	0.282
Missing n	0	0	4	6	7	6							
W-DEQ Total (33–198)													
Vaginal	97.47	87.35	95.76	81.56	–	–	0.00	**5.25**	0.03	0.06	2.03	0.28	0.29
C-section	92.07	83.67	95.04	88.22	–	–	0.998	**0.024**	0.856	0.813	0.158	0.599	0.592
Missing n	0	0	6	6									
Generalized anxiety (0–21)													
Vaginal	4.88	4.00	5.74	5.56	5.51	4.96	**3.92**	1.45	0.08	0.74	0.42	0.41	1.38
C-section	4.88	3.20	6.59	3.50	5.71	5.19	**0.022**	0.233	0.784	0.479	0.656	0.523	0.256
Missing n	0	0	5	6	7	6							
Depression (0–27)													
Vaginal	4.65	4.20	4.88	5.48	4.48	5.16	0.28	0.09	0.02	1.23	2.83	0.00	0.93
C-section	5.47	5.60	4.59	3.80	4.35	5.90	0.758	0.763	0.880	0.295	0.062	0.992	0.395
Missing n	0	0	5	6	7	6							

Notes.
[a] Proportion of participants who said they experienced regular vulvar pain since the beginning of pregnancy (T1) or since the last data collection (T2 and T3).
[b] Proportion of participants who said their pain decreased/stayed the same (1) versus increased (0) since the beginning of pregnancy (T1) or since the last data collection (T2 and T3).VV=women with vulvodynia; Cntrl=control group (women without vulvodynia); Grp=women with vulvodynia versus control; BM=birth method (vaginal versus C-section); C-section=caesarean section; PASS-20=Pain Anxiety Symptoms Scale; W-DEQ= Wijma Delivery Expectancy/Experience Questionnaire.

For this outcome, adding number of pregnancies as a covariate changed the group by birth mode interaction effect from significant at $p < 0.05$ to non-significant ($p = 0.067$).

Anxiety and fear related to pain and birth

Pain Anxiety: The PASS-20 scores increased over time but the main effect of time was qualified by a significant interaction of time and mode of birth. The simple effects follow-up indicated that, in both groups, women who gave birth via caesarean sections experienced a significant increase in pain anxiety between T1 and T3 by 8.02 points, $p < 0.001$ (Cohen's $d = 0.45$ – medium size effect), whereas there was no change in pain anxiety symptoms for women who gave birth vaginally. The use of pain medication during labour/birth did not moderate the effect of time.

Fear of Childbirth: Indicating a greater fear of childbirth, the women with vulvodynia reported higher scores on the W-DEQ compared to women in the control group across both T1 and T2 points (the W-DEQ was collected only at T1 to assess participants' feelings about their upcoming birth, and at T2 to assess their feelings experienced during the birth) by 11.67 points (Cohen's $d = 0.57$ – medium size effect). This main effect of group was not moderated by either birth mode or use of pain medication during labour/birth.

Anxiety and mood outcomes

Anxiety Symptoms: Anxiety symptoms, as measured by the GAD-7, increased in women in both groups between T1 and T2 (by 1.14 points, $p = 0.005$, Cohen's $d = 0.27$ – small effect size) and between T1 and T3 (by 0.97 points, $p = 0.033$, Cohen's $d = 0.21$ – small effect size). This main effect of time was not moderated by either birth mode or use of pain medication during labour/birth.

Depressive Symptoms: Scores on the PHQ-9 did not change with time and there were no other significant main or interaction effects on this outcome.

Discussion

This prospective case-control study adds to the small existing literature regarding pregnancy and birth amongst individuals with vulvodynia. To our knowledge, it is the first study to follow women with a vulvodynia diagnosis in pregnancy and postpartum. It is also the first to our knowledge to assess fear of birth in women with vulvodynia using a validated measure. In clinical settings, individuals with vulvodynia often express concern regarding how their pain symptoms will affect and be affected by pregnancy and parturition. The main objective of this study was to assess changes in vulvar pain symptoms from pregnancy to postpartum amongst individuals who had received a vulvodynia diagnosis. We hypothesized that pain symptoms like pain intensity would increase in the postpartum period for persons with vulvodynia. However, our hypothesis was not supported. The results indicated that most of the vulvar pain outcomes actually improved for the individuals with vulvodynia. The findings also suggest that mode of birth, but not use of pain medication during labour and birth, seemed to play a role in the trajectory of some outcomes such as vulvar pain intensity. Specifically, the intensity of vulvar pain decreased between pregnancy and the postpartum timepoints for women with vulvodynia who gave birth vaginally, but not for those women who had a caesarean birth.

We also hypothesized that, compared to controls, participants with vulvodynia would report higher levels of pain anxiety, fear of childbirth, and anxiety and mood symptoms in the pregnancy and postpartum periods. Only our hypothesis regarding fear of childbirth was confirmed: women with vulvodynia reported higher scores on the W-DEQ during both pregnancy and at three months postpartum ~~post-partum~~, which indicated a greater fear of childbirth. During the study period, there were no significant differences between women with vulvodynia and control participants regarding pain anxiety or symptoms of generalized anxiety or depression. Women's symptoms of anxiety did increase following pregnancy and birth, but those changes occurred in both study groups and were not specific to the individuals with vulvodynia. As well, regardless of vulvodynia versus control status, women who had a caesarean section reported that their pain-related anxiety increased from pregnancy to six months postpartum.

With regard to vulvar pain symptoms, the proportion of individuals with vulvodynia who reported regularly experiencing vulvar pain decreased from pregnancy to six months postpartum (76% to 58%). There was also a significant increase in the proportion of individuals (30%) with vulvodynia at six months postpartum who reported that their symptoms had either improved or had not worsened since their last study assessment (compared to when they completed the survey during pregnancy). Women with vulvodynia also reported significant decreases from pregnancy to postpartum with regard to being bothered by pain or problems during sexual penetration. Furthermore, the overall intensity of vulvar pain significantly decreased at both three and six months postpartum compared to pregnancy, but only amongst women who had a vaginal delivery. Previous research has found that women who developed persistent (i.e., three months or more) genito/pelvic pain following childbirth were more likely to have had a caesarean birth (Cappell and Pukall, 2017). As well, individuals who have a caesarean birth may be at risk for developing chronic pain in general (e.g., chronic post-surgical pain; Jin et al., 2016). Although we did not find a between-group difference in rates of vaginal birth, it has been suggested that individuals with vulvodynia may be more likely to have a caesarean section compared to controls (e.g., Nguyen et al., 2012). Various reasons likely exist for why individuals with vulvodynia may give birth via caesarean section (e.g., obstetrical indications; fear of impact of vaginal birth on pain symptoms; Smith et al., 2016). However, the current study suggests that a vaginal birth, if possible, may be associated with less intense vulvar pain over time.

Nevertheless, women with vulvodynia who have vaginal births may still experience distress about their pain, and the current findings need to be examined in terms of their clinical relevance. A change in pain intensity of one point on the NRS, as found in the current study from pregnancy to postpartum, may reflect only a minimal clinical change. The Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT) proposes that pain intensity reductions of 10 to 20 percent represent "minimally important changes" (Dworkin et al., 2008, p.111), whereas a 30 percent or greater reduction is considered to be of at least moderate clinical relevance (Dworkin et al., 2008, 2009). Amongst women with vulvodynia in our study, there were no changes in their level of distress about their vulvar pain, regardless of mode of birth, which may also suggest that overall changes in vulvar pain intensity were not necessarily clinically relevant.

These findings highlight the multidimensional aspects of chronic vulvar pain, and the importance of inquiring about the personal significance of any changes in pain symptoms that a patient may experience. They may also help individuals with vulvodynia develop realistic expectations regarding their vulvar pain symptoms following pregnancy/birth (e.g., that there may be improvement in levels of pain but that pain and/or distress may persist). In their qualitative study of 18 women with a vulvodynia diag-

nosis and/or symptoms, Johnson et al. (2015) found that participants expressed hope that "pregnancy could improve or *cure* their vulvodynia symptoms" (p. 9) based on sources such as physicians and friends. The same study also found that some participants had strong emotional reactions when experiencing increased pain during pregnancy (e.g., fear; devastation). Johnson et al. (2015) highlighted that an absence of information, such as what to expect following birth, contributed to these reactions. The current study helps to fill that informational gap. By providing information regarding the trajectory of vulvar pain symptoms during pregnancy and postpartum, our study may also increase maternity providers' level of comfort when caring for people with vulvodynia. Previous research with 140 maternity care providers (49 midwives, 91 physicians) found that almost 43% of the midwives and 25% of the physicians reported they were not comfortable managing maternity care for women with vulvodynia (Smith et al., 2016).

In addition to pain symptomatology, the current study also examined fear of childbirth, pain-related anxiety, and symptoms of anxiety and depression. Women with vulvodynia reported higher levels of fear of childbirth on the W-DEQ compared to control cases both prior to and after delivery. There are factors that can modify such fear in pregnancy, such as provision of information regarding birth (e.g., Çankaya and Şimşek, 2021), counselling by midwives (Larsson et al., 2019), and cognitive behavioural therapy (e.g., Nieminen et al., 2016). In addition, it has been recommended that a valid assessment measure of fear of childbirth be integrated into routine maternity/midwifery care (Striebich et al., 2018). Such assessment seems particularly relevant when providing care for an individual with vulvodynia who may have elevated and unique concerns related to childbirth.

Given that women with vulvodynia compared to women without are 10 times more likely to have a pre-existing anxiety disorder (Khandker et al., 2011), maternity care providers should also enquire about general mental health. Maternity care providers can then refer to and develop collaborations with experts such as psychologists who can help patients address psychological concerns during pregnancy and beyond. In the current study, general levels of anxiety increased in both groups from pregnancy to postpartum, and pain-related anxiety increased in women who had given birth via caesarean section. The increase in general anxiety symptoms may not represent clinically significant symptoms (a minimal clinically important difference has been estimated to be four points on the GAD-7 for patients undergoing treatment for depression. However, it is acknowledged that the GAD-7 is more sensitive to improvements versus worsening of symptoms; Toussaint et al., 2020). Still, having maternity care providers help patients establish supports during pregnancy may help individuals prepare for and cope with heightened postpartum anxiety symptoms that could be significant for some people.

Moreover, with regard to symptoms of depression, our study did not find fluctuations in depressive symptoms that can occur between pregnancy and postpartum (e.g., Evans et al., 2001). Symptoms of depression during pregnancy are a strong predictor of depression in the postpartum (e.g., Becker et al., 2016), and the generally low PHQ-9 scores in our study may help explain why there was not significant variability in depressive symptoms across time. There were also some participants in the vulvodynia group who were not regularly experiencing vulvar pain symptoms before becoming pregnant; it is possible that mental health symptoms may not have been as pronounced in our sample compared to if we had recruited a group that consisted only of individuals with current vulvar pain.

The strengths of this study involve the use of prospective data collection, use of some validated tools, and inclusion of a control group. This study also has limitations though. For example, the diagnosis of vulvodynia was self-reported and, given the nature of the research design, was not confirmed in this study. We also collected data from different participants at different points in pregnancy (for example, second versus third trimester). Outcomes such as anxiety and related symptoms can show variation between trimesters (e.g., Viswasam et al., 2020; Viswasam et al., 2021), and our study did not account for such variations. Moreover, the sample size was small, and was further decreased as the study continued into the postpartum, which may have introduced some bias. There were no differences found on sample characteristics and baseline outcome measures between participants who continued in the study versus participants who were lost to follow-up; however, it is possible that participants with worsened postpartum mental health or pain symptoms, for example, may have chosen not to complete follow-up. The dropouts also caused decrease in power to find statistically significant results, potentially leaving some findings in the 'marginal significance' zone like the interaction effects for pain with penetration and for depression.

In addition, the sample was quite homogeneous with regard to demographic characteristics, and, as such, cannot be widely generalized. There was less ethnic diversity in our sample than is typically present in our geographical region; the majority of participants also reported higher income compared to the average income in our area and a higher percentage of participants had completed post-secondary education compared to census data (Statistics, 2017). At the same time, participants were not restricted to our geographical region given the online nature of the study. A participant located in a country with universal healthcare versus a participant living in a location with perhaps more limited access to maternity care may experience differences in the outcomes examined (e.g., anxiety) due to systemic factors (such as not knowing if one will have access to a specific type of practitioner). Future research should aim to improve on the limitations of the current study by studying pain symptoms longitudinally in a larger sample of pregnant individuals with vulvodynia. Future research should also obtain more specific information on factors that may contribute to psychological outcomes like fear of childbirth amongst persons with vulvodynia. Another avenue for future research could be the examination of interventions, such as cognitive behavioural therapy, for reducing fear of childbirth amongst women with vulvodynia and coping with increased anxiety symptoms that may occur postpartum.

In conclusion, this prospective case-control study indicated that pregnant women with vulvodynia experienced some postpartum improvements in vulvar pain symptoms. As well, some symptoms like pain intensity may be moderated by mode of birth amongst women with vulvodynia. Compared to during pregnancy, a significant proportion of the individuals with vulvodynia reported that their vulvar pain symptoms either improved or did not worsen in the postpartum period, and the proportion reporting regular vulvar pain significantly decreased. Mental health symptoms, such as increased symptoms of anxiety, may also change from pregnancy to postpartum. Clinicians can use these findings to help counsel pregnant individuals with vulvodynia. The findings may help individuals with vulvodynia decide about birthing options and prepare for physical and psychological changes that can occur during postpartum.

Ethical approval

This research was approved by the Behavioural Research Ethics Board at the University of British Columbia and by the Vancouver Coastal Health Research Institute.

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Conflict of interest

None declared.

CRediT authorship contribution statement

Kelly B. Smith: Conceptualization, Methodology, Investigation, Writing – original draft, Writing – review & editing, Project administration, Funding acquisition. **Bozena Zdaniuk:** Formal analysis, Writing – original draft, Writing – review & editing. **Smruthi O. Ramachandran:** Writing – original draft, Writing – review & editing, Visualization. **Lori A. Brotto:** Conceptualization, Methodology, Resources, Writing – review & editing, Supervision, Funding acquisition.

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