



Predictors of Psychosocial and Functional Outcomes in Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia: Application of the Fear-Avoidance Model

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Abstract: Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia (PGAD/GPD), which affects up to 4.3% of individuals, is a distressing and poorly understood condition characterized by persistent, unwanted, and often painful sensations of genito-pelvic arousal (eg, throbbing) in the absence of sexual desire. PGAD/GPD is associated with significant negative impacts on psychosocial well-being and daily functioning. Recent research has indicated that PGAD/GPD shares many similarities with other forms of chronic genito-pelvic pain. This study applied the fear-avoidance model of chronic pain to PGAD/GPD to identify cognitive and behavioral factors associated with psychosocial and functional outcomes. A total of 263 individuals with PGAD/GPD symptoms completed a cross-sectional online survey of symptom intensity, cognitive and behavioral predictors (symptom catastrophizing, hypervigilance to symptoms, symptom fear and avoidance, self-efficacy), depression symptoms, and role functioning. Symptom catastrophizing, fear of symptoms, avoidance of symptoms, and hypervigilance to PGAD/GPD symptoms were significantly correlated with poorer psychosocial and functional outcomes, whereas higher self-efficacy was significantly associated with lower depression and better role functioning. Two serial parallel mediation models examined the fear-avoidance pathway from PGAD/GPD symptom intensity to depression symptoms and role functioning. In both models, the pathway through symptom catastrophizing, fear of symptoms, and symptom avoidance was significant, but the pathway through symptom catastrophizing, fear of symptoms, and symptom hypervigilance was not. The results of this study provide support for the applicability of the fear-avoidance model to PGAD/GPD. Interventions targeting fear-avoidance factors may help to reduce PGAD/GPD symptom intensity, distress, and increase psychological well-being and daily functioning.

Perspective: This article provides support for the applicability of the fear-avoidance model of chronic pain to Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia (PGAD/GPD). These results suggest that interventions targeting fear-avoidance cognitions and behaviors (catastrophizing, fear, avoidance, hypervigilance) may help to reduce PGAD/GPD symptom intensity and improve psychological well-being and daily functioning.

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Key Words: Persistent genital arousal disorder/genito-pelvic dysesthesia, Psychosocial, Catastrophizing, Hypervigilance, Mediation

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Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia (PGAD/GPD) is a distressing, poorly understood condition characterized by unwanted and often painful genital sensations (eg, tingling, throbbing) that occur without corresponding sexual desire.¹ Symptoms are primarily located in the genital region, inconsistently associated with genital swelling,¹ and typically last for hours/days or are continuously present.² PGAD/GPD symptoms have been described as dysesthesia (ie, an unwanted, unpleasant sensation).³ Prevalence estimates from 4 countries range from 0.6% to 4.3%.⁴⁻⁶ Despite the significant negative psychosocial impact of PGAD/GPD,⁷ little is known about its etiology and effective treatment.^{1,8}

Researchers have observed that PGAD/GPD shares many similarities with other conditions characterized by genito-pelvic discomfort/pain, such as vulvodynia.⁹ Although the hallmark characteristic of PGAD/GPD is unwanted sensations of arousal,^{2,10,11} 35.4% to 44.3% of PGAD/GPD samples report pain symptoms,^{2,10} including genital and pelvic pain, as well as pain during sexual activity.³ In addition, many arousal and pain adjectives overlap (eg, 'throbbing', 'tingling', 'aching').³ Given the high frequency of pain symptoms and PGAD/GPD's dysesthesia quality, the use of chronic pain models in understanding cognitive and behavioral symptom predictors may assist in better understanding PGAD/GPD and guide treatment efforts.

The fear-avoidance (FA) model proposes a biopsychosocial development of chronic pain.^{12,13} According to the FA model, if pain is interpreted as non-threatening and individuals maintain engagement in valued activities, symptom improvement and psychosocial well-being will be promoted. Alternatively, if pain is interpreted as threatening (leading to catastrophizing thoughts about pain, hypervigilance to pain, and fear and avoidance of pain), factors that are adaptive in the acute stage of pain (eg, avoiding re-injury) may paradoxically contribute to the development of chronic pain and poor psychosocial outcomes.^{12,13} The FA model proposes that fear and avoidance driven physical inactivity and deconditioning in turn lead to disability and difficulty engaging in valued activities (eg, less mobility to work, travel, socialize). The FA model adapted for PGAD/GPD is presented in Fig 1. While the model proposes a sequential series of events, it is important to note that recent research also supports interrelationships among the variables.¹⁴ The FA model has been found to be applicable to genital pain conditions (eg, vulvodynia).¹⁵ Outside of the FA model, there is also growing support for the role of self-efficacy in reducing pain intensity and disability,^{16,17} and self-efficacy is associated with lower FA beliefs in chronic pain samples.¹⁸

Some evidence supports the use of the FA model in PGAD/GPD.⁸ For example, individuals with PGAD/GPD report high levels of symptom catastrophizing, which is

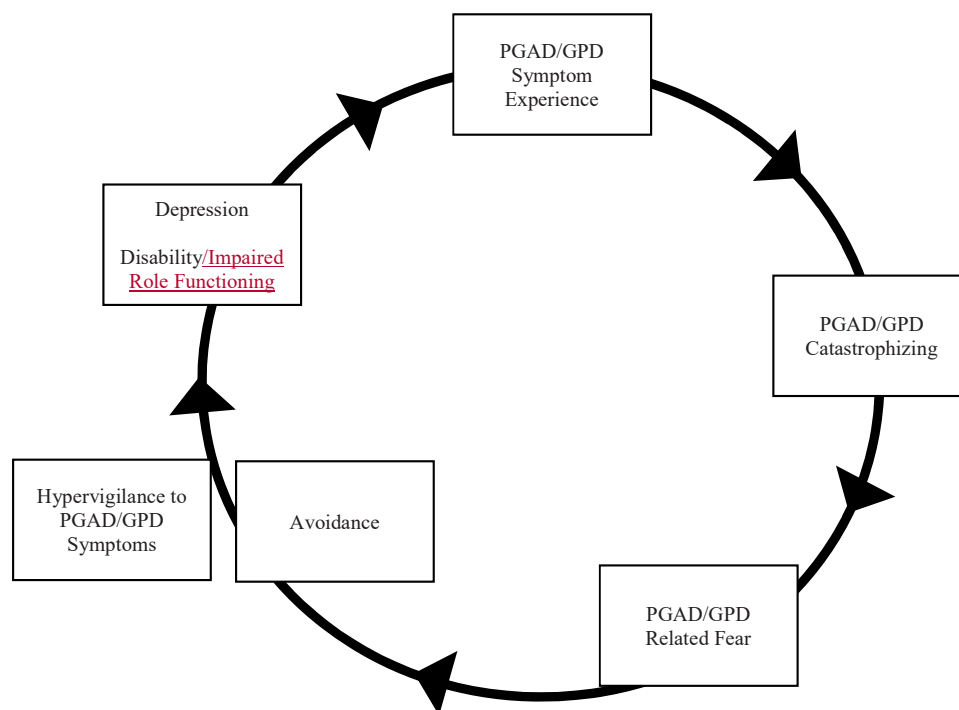


Figure 1. Proposed fear-avoidance (FA) model modified for PGAD/GPD. **Note.** The FA model^{12,13} modified for PGAD/GPD symptoms proposes that if distressing sensations of arousal are interpreted as threatening (eg, catastrophizing thoughts and fear about PGAD/GPD),⁷ one may start avoiding situations or activities that increase PGAD/GPD symptoms and develop increased hypervigilance to PGAD/GPD sensations.¹¹ In turn, avoidance and hypervigilance may lead to negative psychosocial and functional outcomes (eg, interference with completing daily activities such as work and socialization [disability/impaired role functioning], depression symptoms^{7,19}), which may contribute further to the experience and awareness of PGAD/GPD sensations, thus maintaining the cycle. Alternatively, individuals with low levels of catastrophizing, fear, avoidance, and hypervigilance, would theoretically be able to re-engage in daily activities with lower levels of symptom severity, disability, and psychosocial well-being.

associated with greater symptom severity and distress, as well as greater depression and anxiety symptoms.^{7,19} In addition, high rates of hypervigilance to PGAD/GPD symptoms and specific activities/environmental cues (eg, sitting, stress, sexual content in media) may provoke symptoms.^{2,7,20} No study to date, however, has examined the FA model collectively (ie, in its entirety) in a sample with PGAD/GPD. This information could highlight potential targets for intervention—specifically, symptom-related thoughts and behaviors most associated with negative affect, functional disability, and maintenance of symptoms.

The present study aimed to examine the applicability of the FA model to PGAD/GPD in an online, cross-sectional study. The study sought to identify associations among fear-avoidance variables, self-efficacy, PGAD/GPD symptom outcomes, and psychosocial well-being, as well as to develop mediation models to examine the FA model pathway, hypothesizing that significant associations would be found among the FA predictors sequentially as presented in Fig 1.

Methods

Participants

Participants were primarily recruited through online support groups for PGAD/GPD with worldwide membership, as well as through postings on social media, emails to international healthcare providers and researchers who treat and study PGAD/GPD, and previous participants who had consented to be contacted about future research studies on PGAD/GPD. Recruitment occurred between May 2019 and August 2020. Eligible participants were 18 years of age or older, fluent in English, and experiencing symptoms of PGAD/GPD; these symptoms were defined as feelings of persistent genital arousal (genital sensitivity and/or swelling) that occur without subjective feelings of arousal (ie, without sexual desire) and are distressing (a PGAD/GPD symptom distress score greater than 0, where 0 indicates no distress and 10 indicates extreme distress). The PGAD/GPD eligibility criteria were based on recent diagnostic criteria for the condition,¹ and similar criteria have been used in previous online studies.^{6,7,21,22}

Using G*Power (version 3.1.9.3),²³ an a priori power analysis was undertaken to determine the sample size needed to have adequate statistical power to conduct Pearson correlations and serial-parallel mediation analyses with four mediators. Of the proposed analyses, Pearson correlations required the largest sample size; therefore, they were used for the sample size determination. Previous research has found correlations ranging $r = .25$ to $r = .47$ between catastrophizing, PGAD/GPD symptom severity, and psychosocial outcomes (eg, depression and anxiety symptoms).⁷ Based on a conservative estimate of small-to-medium correlations ($r = .20$), an alpha of .05, and a power of .80, a minimum of 191 participants was determined to be needed. In the current study, 263 eligible individuals completed the relevant questionnaires.

Materials

Sociodemographic Information

Participants provided basic sociodemographic information, including age, gender, location, and income (Table 1).

PGAD/GPD Symptom Characteristics

Participants responded to questions regarding the criteria of PGAD/GPD to determine their eligibility, as well as the duration of their symptoms, and whether their symptoms were constant or intermittent. Participants rated the current intensity of their PGAD/GPD symptoms (1: very mild symptoms to 10: extremely severe symptoms) and the distress associated with their symptoms (0: no distress to 10: extreme distress). Participants were also asked to rate the pain associated with their symptoms (0: no pain to 10: extremely painful), as well as the pain associated with specific scenarios (pain with urination, pain with sexual activity, painful orgasm, and pain post-ejaculation).

Predictors of PGAD/GPD Symptoms

Cognitive, emotional, and behavioral predictors were selected based on the FA model.^{12,13} All questionnaires were modified to ask about PGAD/GPD symptoms. Internal consistency (Cronbach's α) for each questionnaire in the present sample is presented in Table 2.

PGAD/GPD Symptom Catastrophizing. The pain catastrophizing scale (PCS)²⁴ is a 13-item self-report measure that asks respondents to think about times when they have experienced pain and to indicate if they engaged in catastrophizing thoughts or feelings. The PCS has been previously used in those with vulvar pain^{25,26} and PGAD/GPD.⁷ In the present study, participants were asked to respond to each item with respect to their experience of persistent genital arousal sensations (example item: 'It's terrible and I think it's never going to get any better') to be inclusive of their overall PGAD/GPD symptoms which may include pain, and throughout the questionnaire, the term 'pain' was replaced with 'genital arousal sensations'. Each item is rated on a 5-point Likert scale from 0 (not at all) to 4 (all the time), with higher scores indicating greater catastrophizing. Total scores of >30 represent clinically significant levels of catastrophizing.²⁴

Fear and Avoidance of PGAD/GPD Symptoms. The pain anxiety symptom scale, short form (PASS20)²⁷ is a 20-item measure of fear and anxiety responses specific to pain. Participants were asked to rate each item based on how often they engage in a list of different thoughts or activities from 0 (never) to 5 (always), with higher scores indicating greater fear and avoidance. In the present study, participants were asked to respond with respect to their experience of persistent genital arousal sensations (example item: 'I will stop any activity as soon as I sense genital arousal sensations coming on'). The PASS20 has four subscales, each comprised of 5 items: cognitive, fear, escape/avoidance, and physiological arousal subscales.

Table 1. Sociodemographic Information (N = 263)

| | % (N) |
|---|------------|
| Gender | |
| Man | 6.5 (17) |
| Non-binary/Genderqueer | 5.3 (14) |
| Other | .4 (1) |
| Woman | 87.8 (231) |
| Sex | |
| Female | 90.5 (238) |
| Intersex | 1.1 (3) |
| Male | 7.6 (20) |
| Other | .8 (2) |
| Currently Reside | |
| Asia | .4 (1) |
| Australia | 5.3 (14) |
| Canada | 14.8 (39) |
| Caribbean | .8 (2) |
| Eastern Europe | 1.9 (5) |
| Latin/South America | 1.1 (3) |
| Middle East | .4 (1) |
| Other | 4.6 (12) |
| United States | 53.6 (141) |
| Western Europe | 17.1 (45) |
| Ethno-Racial Background | |
| African (eg, Ghana, Kenya, Somalia) | .8 (2) |
| African Canadian or African American | 2.3 (6) |
| East Asian (eg, China, Japan, Korea, Taiwan) | 1.5 (4) |
| French Canadian | 1.9 (5) |
| Indigenous (eg, First Nations, Métis, Inuit) | 1.9 (5) |
| Indo-Caribbean (eg, Guyanese with origins in India) | 0 (0) |
| Latin American (eg, Argentina, Mexico, Nicaragua) | 3.8 (10) |
| Other | 10.6 (28) |
| South Asian (eg, India, Sri Lanka, Pakistan) | 1.5 (4) |
| South East Asian (eg, Vietnam, Malaysia, Philippines) | 1.5 (4) |
| White North American | 49.4 (130) |
| White European (eg, England, Greece, Sweden, Russia) | 34.6 (91) |
| Decline Response | 1.1 (3) |
| Education | |
| College/undergraduate degree | 39.2 (103) |
| Graduate/professional school | 28.5 (75) |
| High school graduate | 14.1 (37) |
| Other | 5.7 (15) |
| Some high school | 4.6 (12) |
| Trade school graduate | 7.2 (19) |
| Decline response | .8 (2) |
| Occupational Status | |
| Employed full-time | 32.4 (85) |
| Employed part-time | 14.8 (39) |
| On disability | 14.8 (39) |
| Other | 8.0 (21) |
| Retired | 8.4 (22) |
| Student | 9.9 (26) |
| Unemployed | 11.4 (30) |
| Decline response | .4 (1) |
| Annual Household Income (US Dollar) | |
| \$0–\$19,999 | 18.6 (49) |
| \$20,000–\$39,999 | 12.9 (34) |
| \$40,000–\$59,999 | 13.7 (36) |
| \$60,000–\$79,999 | 13.7 (36) |
| \$80,000–\$99,999 | 7.2 (19) |

Table 1 (Continued)

| | % (N) |
|---|------------|
| > \$100,000 | 22.1 (58) |
| Decline response | 11.8 (31) |
| Where did you hear about the study? | |
| Social media/social media-based support group | 75.3 (198) |
| Healthcare provider | 2.3 (6) |
| Consented to be contacted by email about future studies | 5.3 (14) |
| Own online search | 3.8 (10) |
| Other | 2.7 (7) |
| Decline response/no response | 10.6 (28) |

NOTE. Participants were permitted to select all ethno-racial backgrounds applicable to them, therefore, the total percentage is greater than 100% for that item.

The fear (PASS20-F) and escape/avoidance (PASS20-E) subscales were used in the present study. During the development of the PASS20, it was found to have good internal consistency, test-retest reliability, and construct and predictive validity.²⁷

Hypervigilance to Genital Arousal Sensations. The pain vigilance and awareness questionnaire (PVAQ)²⁸ is a 16-item self-report measure of attention to pain over the past 2 weeks. Each item is rated from 0 (never) to 5 (always), with higher scores indicating greater hypervigilance to pain. In the present study, the term ‘pain’ was replaced with ‘genital arousal sensations’ throughout the questionnaire (example item: ‘I focus on genital arousal sensations’). Previous studies have found strong test-retest reliability as well as strong construct validity for PVAQ through associations with other pain and coping measures in a range of chronic pain samples.^{28,29}

Self-Efficacy. The pain self-efficacy questionnaire (PSEQ)³⁰ asks participants to report how confident they are that they can do a list of 10 activities at present in the context of chronic pain (from 0: not at all confident to 6: completely confident). For the present study, participants were asked to respond with respect to their persistent genital arousal sensations (example item: ‘I can still accomplish most of my goals in life, despite the persistent genital arousal sensations’). The PSEQ has demonstrated convergent validity with other measures of pain-related disability and coping strategies as well as strong psychometric properties (internal consistency and stability over time).³⁰

Psychosocial Outcomes

Depression Symptoms. The hospital anxiety and depression scale (HADS)^{31,32} is a self-report scale with 14 items, divided into a 7-item anxiety symptoms subscale (HADS-A; eg, ‘I feel tense or “wound up”’) and a 7-item depression symptoms subscale (HADS-D;

Table 2. Correlation Matrix

| | <i>M (SD)</i> | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 |
|--|---------------|------------|------------------|------------------|------------------|------------------|------------------|-------------------|-------------------|
| 1. PGAD/GPD symptom intensity (range 1–10) | 6.23 (2.47) | <i>n/a</i> | <i>r</i> = .25** | <i>r</i> = .16* | <i>r</i> = -.01 | <i>r</i> = .24** | <i>r</i> = .21** | <i>r</i> = -.12 | <i>r</i> = -.23** |
| 2. PGAD/GPD symptom catastrophizing (PCS; range 0–52) | 31.03 (12.71) | | α = .94 | <i>r</i> = .72** | <i>r</i> = .46** | <i>r</i> = .45** | <i>r</i> = .56** | <i>r</i> = -.29** | <i>r</i> = -.60** |
| 3. Fear of symptoms (PASS20-F; range 0–25) | 17.35 (5.99) | | | α = .86 | <i>r</i> = .64** | <i>r</i> = .50** | <i>r</i> = .46** | <i>r</i> = -.30** | <i>r</i> = -.55** |
| 4. Escape/Avoidance of symptoms (PASS20-E; range 0–25) | 11.14 (5.63) | | | | α = .71 | <i>r</i> = .37** | <i>r</i> = .40** | <i>r</i> = -.30** | <i>r</i> = -.51** |
| 5. Hypervigilance to symptoms (PVAQ; range 0–80) | 56.21 (12.39) | | | | | α = .83 | <i>r</i> = .26** | <i>r</i> = -.22** | <i>r</i> = -.34** |
| 6. Depression symptoms (HADS-D; range 0–21) | 8.23 (4.79) | | | | | | α = .86 | <i>r</i> = -.42** | <i>r</i> = -.67** |
| 7. Role functioning (SF-36-RF; range 0–100) | 38.40 (41.08) | | | | | | | α = .87 | <i>r</i> = .44** |
| 8. Self-efficacy (PSEQ; range 0–60) | 31.71 (15.40) | | | | | | | | α = .95 |

Abbreviations: PGAD/GPD = Persistent Genital Arousal Disorder/Genito-pelvic dysesthesia; PCS = pain catastrophizing scale; PVAQ = pain vigilance and awareness questionnaire; PASS20 = pain anxiety symptom scale, short form; PSEQ = pain self-efficacy questionnaire; HADS = hospital anxiety and depression scale; SF-36 = 36-item short form health survey. All pain scales modified for PGAD/GPD (see methods).

NOTE. Correlation matrix between fear-avoidance model variables, PGAD/GPD symptoms, and psychosocial outcomes. Means and standard deviations presented in the first column. Cronbach's alpha presented on the horizontal.

NOTE. The potential range for each scale/item is presented in brackets.

* $P < .05$.

** $P < .01$.

eg, 'I still enjoy the things I used to enjoy'). The depression symptoms subscale was used in the present study. Each item is rated from 0 to 3 based on how the respondent has been feeling over the past week, with higher scores indicating greater depression symptoms. Previous studies have found that the HADS demonstrates convergent validity with other measures of depression and anxiety symptoms, as well as acceptable internal consistency with other relevant measures (eg, Beck depression questionnaire, general health questionnaire, state-trait anxiety inventory).^{31,32} Zigmond and Snaith³² recommended that a score above 8 on either the HADS-A or HADS-D should be regarded as a possible case (of either an anxiety or depression diagnosis), and a score above 10 indicates a probable case. These clinical cut-off scores have received support from subsequent reviews and meta-analyses.³¹

Role Functioning. The 36-item short-form health survey (SF-36)³³ is a self-report questionnaire of broad health outcomes. Participants completed the 'Role limitations due to physical health' subscale (SF-36-RF) as a measure of the impact of physical health on daily functioning to capture the disability component of the FA model. The subscale consists of 4 items about whether physical health has interfered with work or other regular daily activities in the previous 4 weeks (yes = 0; no = 100; example item: 'cut down on the amount of time you spent on work or other activities'). Higher scores indicate better role functioning. Previously published studies have found the subscales of the SF-36 reliably demonstrate acceptable internal consistency and high test-retest reliability.³⁴

Procedure

Ethical approval for this study was granted by the Queen's University General Research Ethics Board. Interested participants accessed the online study using a hyperlink provided on study advertisements/posts. Participants first reviewed the Letter of Information and Consent Form which provided information about the nature of the study. If participants chose to participate after reviewing this form, they responded to a question asking them to provide their informed consent and then they proceeded to the remainder of the questionnaires. The survey took approximately 45 minutes to complete (the survey included other questionnaires, not used in the present study). At the end of the study, participants reviewed the debriefing form and had the option to enter their email into a prize draw to win 1 of 4 \$50 (CAD) Amazon gift cards.

Data Considerations

Prior to analyses, the data were examined for missing values, appropriate ranges, normality, and outliers where appropriate. On validated questionnaires with 7 or more items (PCS, PVAQ, PSEQ, HADS-D), if < 15% of the questions were missing for a given individual, those missing values were replaced with the individual's mean response on that questionnaire. If > 15% were missing, then those individuals' responses for that questionnaire were not included in the analyses. Missing data were not replaced for sociodemographic or PGAD/GPD symptom questions. Participants were provided with a decline response option for every question; thus, sample sizes differ across questions. Before conducting analyses, the data were also examined to ensure they met assumptions for bivariate

correlation, multiple regression, and mediation analyses. A correlation matrix and 2 serial parallel mediation analyses were undertaken to examine the relationships among fear-avoidance variables, symptom intensity, and psychosocial outcomes, modeled on the FA model. The serial parallel mediation analyses were run using a custom model in PROCESS V3.5.3.³⁵ Analyses were conducted using the IBM statistical package for the social sciences version 27. Alpha values were set at $P < .05$, and 95% bootstrapped confidence intervals were calculated for the mediation models with 10,000 samples.

Results

Sociodemographics

Three hundred and two (302) surveys were completed; among the respondents, 32 participants were excluded as they did not report current symptoms of PGAD/GPD, and an additional 7 participants were excluded as they reported no distress associated with their PGAD/GPD symptoms. The final sample consisted of 263 participants who were on average 40.06 years of age ($SD = 14.14$; $N = 263$ range: 18–75). The majority of participants were women (87.8%) and were located in North America or Western Europe at the time of participation. A full description of the sociodemographic features of the present sample is presented in Table 1.

PGAD/GPD Symptom Characteristics

Participants, on average, reported that their PGAD/GPD symptoms had been present for a substantial period of time ($M = 11.21$ years, $N = 263$, $SD = 12.93$, range: 0–68 years). The sample was evenly divided between those participants whose symptoms were continuously present (46.0%, $N = 121$) and those who stated they experienced symptom-free periods (54.0%, $N = 142$, 'intermittent symptoms'). The average distress rating was high for the present sample ($M = 7.01/10$, $N = 263$, $SD = 2.72$, range: 1–10) as was the average rating of symptom intensity ($M = 6.23/10$, $N = 262$, $SD = 2.47$, range: 1–10). Additionally, 80.6% ($N = 212$) indicated a non-zero pain rating score associated with their PGAD/GPD symptoms, with an average pain intensity of 4.51/10 ($N = 263$, $SD = 3.32$, range: 0–10).

In terms of the activities that were associated with pain, painful orgasms were reported by 42.4% ($N = 110$) of participants, who rated the intensity of this pain as 5.04 on average (where 0: do not experience this, and 10: extreme intensity). Pain with sexual activity was reported by the majority of participants (59.1%, $N = 146$), and on average, this pain was moderately intense ($M = 4.98$; $SD = 2.76$). Urinary discomfort/pain was also common (54.8%, $N = 144$), and of moderate intensity ($M = 4.27$, $SD = 2.77$). Finally, for participants with a penis and scrotum, 71.4% ($N = 15$) reported post-ejaculatory pain ($M = 5.71$, $SD = 2.78$).

Associations Among Cognitive and Behavioral Fear-Avoidance Factors, PGAD/GPD Symptoms, and Psychosocial Well-being

Associations were examined amongst cognitive and behavioral fear-avoidance variables (symptom catastrophizing [PCS], hypervigilance to symptoms [PVAQ], fear of symptoms [PASS20-F], and escape/avoidance of symptoms [PASS20-E]), symptom outcomes (symptom intensity), psychosocial outcomes (depression symptoms [HADS-D] and role functioning [SF-36-RF]). Means and a correlation matrix are presented in Table 2.

Significant, moderate associations were found between the cognitive and behavioral fear-avoidance variables (PCS, PASS20-F, PASS20-E, PVAQ) and psychosocial outcomes (HADS-D, SF-36-RF). Greater symptom catastrophizing, fear of symptoms, escape/avoidance of symptoms, and hypervigilance to symptoms were significantly associated with greater depression symptoms (r 's ranging .26–.56) and lower role functioning (r 's ranging: $-.22$ to $-.30$). Greater symptom catastrophizing demonstrated the largest association with depression symptoms, whereas greater escape/avoidance of PGAD/GPD symptoms exhibited the largest association with impaired role functioning. Moderate to large positive associations were also observed between the fear-avoidance variables, indicating interrelationships between these factors.

Smaller associations were found between symptom intensity and the fear-avoidance variables and psychosocial outcomes (significant r 's range = .16–.25). The largest positive association between symptom intensity was with symptom catastrophizing ($r = .25$). Symptom intensity was not significantly associated with escape/avoidance of symptoms or role functioning. Finally, self-efficacy to manage symptoms was significantly and negatively associated with all cognitive and behavioral factors from the FA model (r 's range = $-.34$ to $-.60$). Self-efficacy was associated with lower depression symptoms ($r = -.67$) and higher role functioning ($r = .44$).

Fear-Avoidance Mediation Models

Two serial parallel mediation models were created to examine the relationship between PGAD/GPD symptom intensity and (1) depression symptoms (HADS-D) and (2) role functioning (SF-36-RF) through the FA model pathway (symptom catastrophizing [PCS], fear of symptoms [PASS20-F], escape/avoidance of symptoms [PASS20-E] and hypervigilance to symptoms [PVAQ]).^{12,13} Parallel pathways were examined through hypervigilance and avoidance, as these variables are presented in the same step in the FA model.^{12,13} Direct and indirect effects are presented in Table 3.

Depression Symptoms

The first model predicted depression symptoms (Fig 2). In the first pathway, greater symptom intensity was

Table 3. Direct and Indirect Effects for Mediation Models

| PATH | B | SE | 95% CI |
|---|-------|------|---------------|
| Mediation Model 1: Depression Symptoms (HADS-D) | | | |
| Direct Effect of X on Y | .19 | .11 | (-.03,.40) |
| Total Indirect Effect | .22 | .08 | (.05,.39) |
| PGAD/GPD Symptom Intensity → PCS → PASS20-F → PVAQ → HADS-D | -.01 | .01 | (-.03,.01) |
| PGAD/GPD Symptom Intensity → PCS → PASS20-F → PASS20-E → HADS-D | .04 | .02 | (.01,.09) |
| Mediation Model 2: Role Functioning (SF-36-RF) | | | |
| Direct Effect of X on Y | -.98 | 1.09 | (-3.12, 1.16) |
| Total Indirect Effect | -1.09 | .55 | (-2.20, -.01) |
| PGAD/GPD Symptom Intensity → PCS → PASS20-F → PVAQ → SF-36-RF | -.06 | .09 | (-.27,.11) |
| PGAD/GPD Symptom Intensity → PCS → PASS20-F → PASS20-E → SF-36-RF | -.36 | .20 | (-.81, -.03) |

Abbreviations: PGAD/GPD = Persistent Genital Arousal Disorder/Genito-pelvic Dysesthesia; PCS = pain catastrophizing scale; PVAQ = pain vigilance and awareness questionnaire; PASS20-F = pain anxiety symptom scale, fear of symptoms subscale; PASS20-E = pain anxiety symptom scale, escape/avoidance of symptoms subscale; HADS-D = hospital anxiety and depression scale, depression subscale; SF-36-RF = 36-item short form health survey, role functioning subscale. NOTE. All pain scales modified for PGAD/GPD (see methods). 95% bootstrapped confidence intervals provided with 10,000 samples.

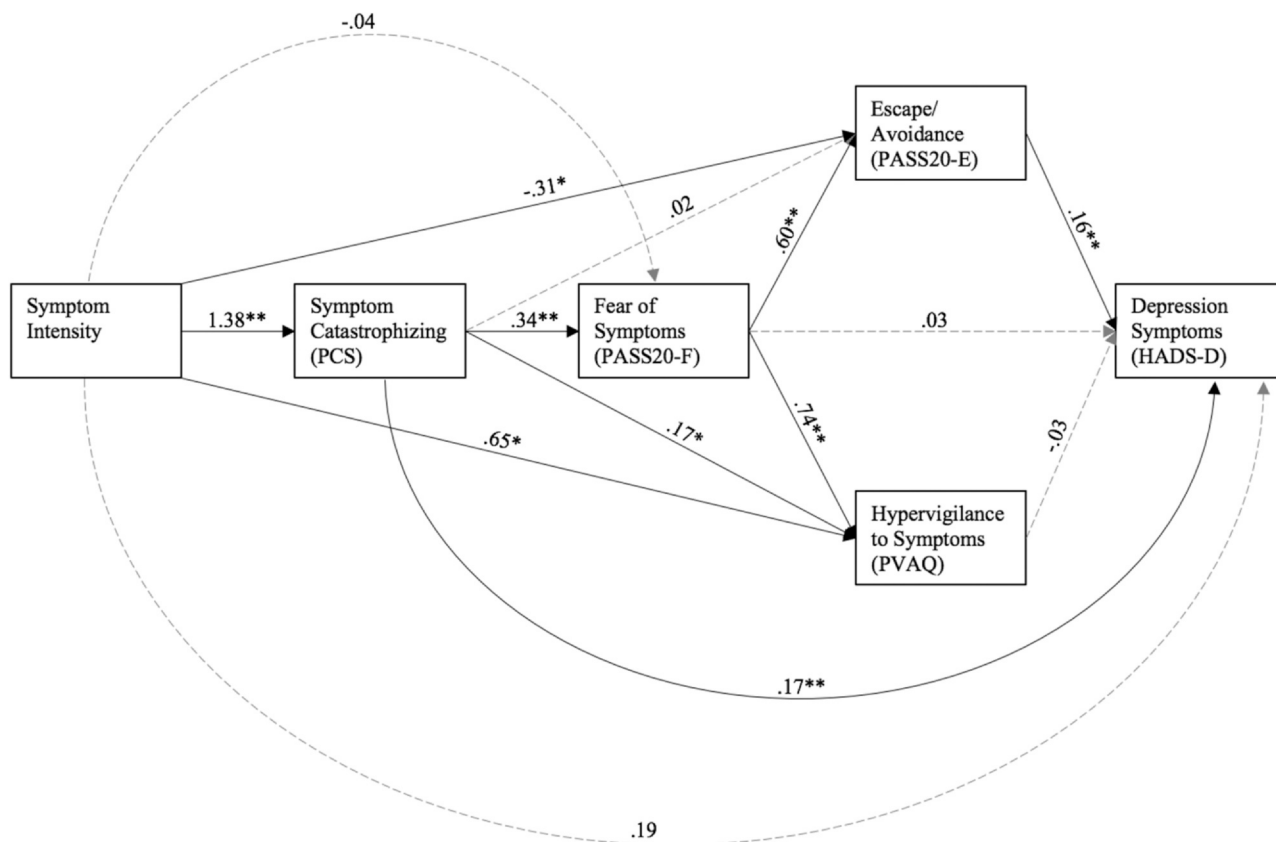


Figure 2. Fear-avoidance mediation model predicting depression symptoms in a sample of individuals with PGAD/GPD (N = 241). Note. Solid lines indicate paths that are significant, and dashed lines indicate paths that are not significant. Unstandardized coefficient presented. PGAD/GPD = Persistent Genital Arousal Disorder/Genito-pelvic Dysesthesia; PCS = pain catastrophizing scale; PVAQ = pain vigilance and awareness questionnaire; PASS20-E = pain anxiety symptom scale, escape/avoidance of symptoms subscale; HADS-D = hospital anxiety and depression scale, depression symptom subscale. All pain scales modified for PGAD/GPD (see Methods). * $P < .05$; ** $P < .01$.

significantly associated with greater symptom catastrophizing ($b = 1.38, P < .001$). Next, greater symptom catastrophizing was significantly associated with greater fear of symptoms ($b = .34, P < .001$), which in turn, was significantly associated with greater escape/avoidance of symptoms ($b = .60, P < .001$). Finally, greater escape/avoidance of symptoms was significantly associated with greater depression symptoms ($b = .16, P = .007$). This overall

pathway from PGAD/GPD symptom intensity to depression symptoms through catastrophizing, fear of symptoms, and escape/avoidance of symptoms was significant ($b = .04, SE = .02, 95\% CI = [.01, .09]$). See Table 3 for indirect pathways.

As stated above, in the second pathway, greater symptom intensity was significantly associated with greater symptom catastrophizing ($b = 1.38, P < .001$)

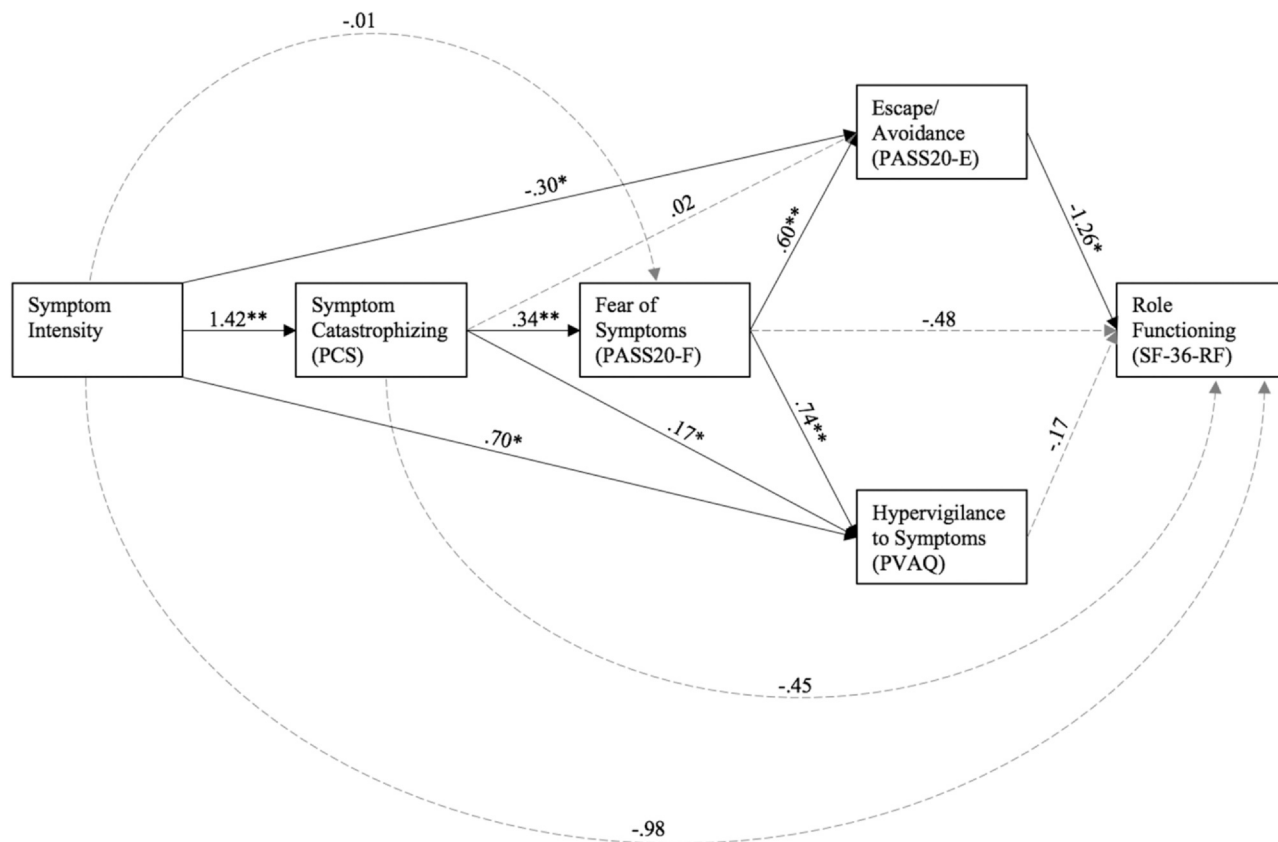


Figure 3. Fear-avoidance mediation model predicting role functioning in a sample of individuals with PGAD/GPD (N = 240). *Note.* Solid lines indicate paths that are significant, and dashed lines indicate paths that are not significant. Unstandardized co-efficient presented. PGAD/GPD = Persistent Genital Arousal Disorder/Genito-pelvic Dysesthesia; PCS = pain catastrophizing scale; PVAQ = pain vigilance and awareness questionnaire; PASS20-E = pain anxiety symptom scale, escape/avoidance of symptoms subscale; SF-36-RF = 36-item short-form health survey, role functioning subscale. All pain scales modified for PGAD/GPD (see Methods). * $P < .05$; ** $P < .01$.

which was significantly associated with greater fear of symptoms ($b = .34$, $P < .001$). Next, greater fear of symptoms was significantly associated with greater hypervigilance to symptoms ($b = .74$, $P < .001$); however, hypervigilance to symptoms was not significantly associated with depression symptoms ($b = -.03$, $P = .27$). This overall pathway from PGAD/GPD symptom intensity to depression symptoms, through catastrophizing, fear of symptoms, and hypervigilance to symptoms, was not significant ($b = -.01$, $SE = .01$, $95\% \text{ CI} = [-.03, .01]$).

Role Functioning

The second model predicted role functioning (Fig 3). In the first pathway, greater symptom intensity was significantly associated with greater symptom catastrophizing ($b = 1.42$, $P < .001$). Next, greater symptom catastrophizing was significantly associated with greater fear of symptoms ($b = .34$, $P < .001$), which in turn was significantly associated with greater escape/avoidance of symptoms ($b = .60$, $P < .001$). Finally, greater escape/avoidance of symptoms was significantly associated with lower role functioning ($b = -1.26$, $P = .03$). This overall pathway from PGAD/GPD symptom intensity to role functioning, through catastrophizing,

fear of symptoms, and escape/avoidance of symptoms was significant ($b = -.36$, $SE = .20$, $95\% \text{ CI} = [-.81, -.03]$).

In the second pathway, again, greater symptom intensity was significantly associated with greater symptom catastrophizing ($b = 1.42$, $P < .001$) and greater symptom catastrophizing was significantly associated with greater fear of symptoms ($b = .34$, $P < .001$). Next, greater fear of symptoms was significantly associated with greater hypervigilance to PGAD/GPD symptoms ($b = .74$, $P < .001$); however, hypervigilance to symptoms was not significantly associated with role functioning ($b = -.17$, $P = .49$). This overall pathway from PGAD/GPD symptom intensity to role functioning, through symptom catastrophizing, fear of symptoms, and hypervigilance to symptoms was not significant ($b = -.06$, $SE = .09$, $95\% \text{ CI} = [-.27, .11]$).

Discussion

This study investigated the associations among cognitive and behavioral fear-avoidance variables, symptom intensity, and psychosocial well-being (depression symptoms, role functioning) in individuals with PGAD/GPD. Although previous studies have examined portions of the FA model within groups of individuals

experiencing PGAD/GPD,⁸ the present study is the first to examine the model collectively.

Consistent with previous studies^{2,10,21,36} PGAD/GPD symptoms were distressing and longstanding, highlighting the need for effective, evidence-based treatments for PGAD/GPD. Although previous studies have found that PGAD/GPD symptoms are often painful^{2,19,37} the prevalence of painful symptoms (80.61%) reported was much higher in the present study. This may be due to how pain was measured (0–10 scale), which could be more sensitive to milder levels of pain than the dichotomous measures used in past studies. For example, 44% of the individuals with PGAD/GPD symptoms responded ‘yes’ (vs no) to a question asking if their symptoms are painful.²

Significant associations were observed between the cognitive and behavioral fear-avoidance variables and psychosocial outcomes. Specifically, greater symptom catastrophizing, fear of symptoms, escape/avoidance of symptoms, and hypervigilance to symptoms were associated with higher depression symptoms and lower role functioning. The direction of these findings is in line with what is theorized by the original FA model.^{12,13}

Symptom catastrophizing was the fear-avoidance variable with the largest association with both depression symptoms and symptom intensity. The average symptom catastrophizing score in the present sample was above the cut-off score indicating clinically significant catastrophizing.²⁴ These findings are consistent with previous research showing that greater symptom catastrophizing is associated with poorer symptom and psychosocial/functioning outcomes for women with PGAD/GPD symptoms.⁷ Indeed, feelings of helplessness (a component of catastrophizing) may be strongly associated with PGAD/GPD given the lack of research on the condition,^{1,8,38} long duration of distressing symptoms,² and frequent barriers to healthcare.²² Previous research found that all the subcomponents of symptom catastrophizing (helplessness, rumination, magnification)²⁴ are associated with greater depression symptoms, anxiety symptoms, sexual distress, symptom severity, and distress in samples of individuals with PGAD/GPD; however, the largest associations are seen between helplessness and the symptom (severity, distress) and psychosocial outcomes.

Overall, the associations between fear-avoidance variables and role functioning were smaller than the associations between fear-avoidance variables and depression symptoms. Although the present sample reported greater role functioning difficulties as compared to normative data from general populations in North America (where most participants resided),^{39,40} other factors that were not captured in the present study may contribute to these difficulties. For example, PGAD/GPD is associated with numerous comorbid health conditions, which could also impact role functioning but are not captured in the present models.^{2,6} Social support may also play an important role in functional outcomes in PGAD/GPD, as it does with other genito-pelvic pain conditions.¹⁵ More information is needed about what influences functional outcomes for individuals with PGAD/GPD, as these may provide additional avenues for intervention and support.

Smaller associations were observed between PGAD/GPD symptom intensity and the fear-avoidance variables. This finding is consistent with previous chronic pain literature where reductions in fear, catastrophizing, and avoidance of pain are more reliably associated with improved functioning and inconsistently associated with changes in pain intensity.^{41,42} In addition, self-efficacy to manage PGAD/GPD symptoms was significantly associated with positive outcomes (lower depression symptoms, higher role functioning). This is consistent with past research, which finds that higher baseline pain self-efficacy is a robust predictor of positive future outcomes in chronic pain samples.⁴³

To examine the pathway through the FA model, 2 serial parallel mediation models were created to predict depression symptoms and role functioning. The pathway from PGAD/GPD symptom intensity through symptom catastrophizing, fear of symptoms, and escape/avoidance of symptoms was significant in both models, while the pathway from PGAD/GPD symptom intensity through catastrophizing of symptoms, fear of symptoms, and hypervigilance to symptoms was not.

Escape/avoidance may play an important role in the impact of PGAD/GPD symptoms on psychosocial/functioning outcomes. Activities that include direct/indirect genital pressure (eg, sitting, sexual activity, walking) frequently provoke or increase PGAD/GPD symptoms.² Avoidance of these activities may lead to significant disruptions in daily functioning. Strategies to improve functioning and psychosocial well-being for individuals with PGAD/GPD may be two-pronged: reducing avoidance behaviors through exposure and/or graded activity scheduling (as used in chronic pain interventions)⁴⁴ and seeking environmental accommodations (eg, cushions, standing desks).

In addition, fear of PGAD/GPD symptoms appears to be an important mechanism in the development and maintenance of symptoms and their psychosocial sequelae. Leiblum and Chivers (2007) proposed that PGAD/GPD may develop from negative attributions and anxiety responses to spontaneous genital arousal experiences.⁴⁵ The corresponding sympathetic nervous system arousal involved in anxiety could in turn increase genital arousal sensations and one’s attention to these sensations. Other theories, such as the excitation transfer hypothesis,⁴⁶ also suggest that physiological arousal experienced during anxiety (eg, increased heart rate, genital sensations) could increase sexual responsivity due to misattributing the arousal to PGAD/GPD rather than to anxiety. Research on whether physiological genital arousal is increased by state anxiety is mixed,⁴⁷ however, no study to date has specifically examined whether this relationship is mediated by attributions made about the physiological anxiety symptoms. These attributions, as supported by the FA model, may be particularly relevant to PGAD/GPD.

In both mediation models, the pathway through symptom hypervigilance was not significant. However, symptom hypervigilance alone was found to have significant correlations with greater PGAD/GPD symptom intensity, greater depression symptoms, and lower role functioning. These findings suggest that symptom hypervigilance may play a more minor mechanistic role in

the association between PGAD/GPD symptom intensity and psychosocial/functional outcomes. One alternative explanation for this finding is that the measure used in the present study (PVAQ) does not fully capture the experience of hypervigilance in PGAD/GPD. The PVAQ primarily focuses on attention to symptoms themselves, however, approximately half of the women with PGAD/GPD report that environmental cues (eg, sexual cues), lead to an increase in their symptoms.² As such, individuals with PGAD/GPD may also experience hypervigilance to environmental stimuli (eg, sexual content in media) which in turn leads them to avoid certain activities or situations and interferes with daily functioning. Future research may seek to further tailor the FA model to the unique experiences of PGAD/GPD. Future research may also examine other sequential models to further investigate the sequence and directions of relationships in the FA model and cumulative interactions.¹⁴

Clinical Implications

The cause of PGAD/GPD is complex and multifactorial; therefore, strictly biomedical approaches to PGAD/GPD treatment do not adequately account for cognitive and behavioral contributors to symptoms and overall psychosocial well-being.⁸ The results of the present study suggest it may be beneficial to target these cognitions and behaviors in treatment. For example, cognitive behavioral therapy could assist individuals experiencing PGAD/GPD symptoms to identify and restructure thoughts associated with fear and catastrophizing of symptoms, and to promote the reduction of anxiety through exposure to feared or avoided situations/symptom cues. Cognitive behavioral therapy interventions have been found to reduce pain severity and improve sexual functioning for individuals experiencing genito-pelvic pain conditions,^{48,49} with catastrophizing, avoidance, and self-efficacy predicting treatment outcomes.⁵⁰

Limitations and Future Directions

Due to the online, self-report design, clinical confirmation of PGAD/GPD diagnosis was not possible. To reduce the impact of this limitation, detailed diagnostic criteria for PGAD/GPD were selected based on recent diagnostic recommendations.¹ PGAD/GPD shares many similarities with other forms of genito-pelvic pain,³ and high agreement between self-reported symptoms and clinical diagnosis has been observed for genito-pelvic pain samples (eg, vulvodynia).^{51,52} The online design also had the potential to reach a broader range of individuals experiencing PGAD/GPD who may not have volunteered to participate in an in-person study given the high rates of associated shame and stigma.²²

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The present study was limited to English-speaking individuals primarily from North America and Western Europe. Future research may seek to extend the present findings to culturally diverse samples. Cultural differences may impact the interpretation of PGAD/GPD symptoms (eg, cross-cultural differences in beliefs/attitudes about sexuality). The scales used in this study were developed to measure psychosocial reactions to pain and were modified for PGAD/GPD. Future research may seek to determine whether the development of new measures specific to PGAD/GPD is needed. Finally, this study was cross-sectional, and therefore, causal conclusions cannot be drawn as the associations observed could be bidirectional. For example, depression symptoms may lead to behavioral inactivity, which in turn leads to greater PGAD/GPD symptoms. Future research may build on these results using longitudinal designs and interventions targeting fear-avoidance cognitions and behaviors (ie, decatastrophizing exercises, paced activities, and exposure exercises to reduce avoidance behaviors).

Conclusions

The results of this study provide support for the applicability of the FA model of chronic pain to PGAD/GPD. Symptom catastrophizing, fear and anxiety about symptoms, hypervigilance to symptoms, and self-efficacy to manage symptoms were significantly associated with depression symptoms and role functioning outcomes. In addition, these results suggest that interventions targeting fear-avoidance cognitions and behaviors may help to reduce PGAD/GPD symptom intensity and distress, as well as the impact of the condition on psychological well-being and daily functioning.

Disclosures

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