

RESILIENCE RESOURCES IN CHRONIC PAIN PATIENTS: THE PATH TO ADAPTATION, 2nd Edition

EDITED BY: Carmen Ramírez-Maestre, Madelon Peters, John Andrew Sturgeon
and Rocio de la Vega
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RESILIENCE RESOURCES IN CHRONIC PAIN PATIENTS: THE PATH TO ADAPTATION, 2nd Edition

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Editorial: Resilience Resources in Chronic Pain Patients: The Path to Adaptation

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Editorial on the Research Topic

Resilience Resources in Chronic Pain Patients: The Path to Adaptation

Individual differences affect the behavior of individuals in response to chronic pain, and such behaviors can lead to either disability or capacity (Ramírez-Maestre and Esteve, 2013). Modern theories of adaptation necessitate a greater emphasis on individual differences in behavioral patterns oriented toward meaningful function despite pain, as well as effective recovery from the negative impacts of pain. Recent empirical studies have acknowledged the positive influence of resilience resources on adjustment to chronic pain (Sturgeon and Zautra, 2016; Hemington et al., 2017; Arewasikporn et al., 2018; Esteve et al., 2018; Ramírez-Maestre et al., 2019). According to these investigations, resilience has emerged as a personal resource that increases the patients' capacity to manage pain effectively. Thus, despite having chronic pain, a resilient individual will be able to experience positive emotions and maintain a higher level of functioning. Patients with these positive characteristics may use more effective coping strategies, have better goal-adjustment, demonstrate greater levels of flexibility and acceptance across situations, and maintain an appropriate functioning level. The studies collected in the present Research Topic are good examples of novel investigation in this context.

There are nine manuscripts in this special issue, including eight original research studies and one narrative review. Three papers analyze the role of positive variables in different stages of the lifespan: Childhood, adolescence, and older adulthood. Hynes et al. review paper identifies family characteristics that are associated with both risk and resilience in children with Juvenile Idiopathic Arthritis (JIA). Their study, which includes seven articles in a narrative review, delineates the contribution of individual and parental resilience mechanisms and resources to resilience outcomes in children with JIA and their families. Hynes et al. highlight children's psychological flexibility, self-efficacy, treatment adherence, pain acceptance, and perceived social support as key contributors to resilience outcomes. The study of Beeckman et al. examines the role of parental protective responses and instructions to engage in activities in adolescents' daily pain-related behavior. The results show an (indirect) adaptive role of parental psychological flexibility on adolescent daily pain-related behavior via its impact on parental protective behavior, highlighting the dynamic, and transactional nature of pain adaptation among adolescents in a family system. Finally, Bartley et al. examined the association of multisystem resiliency with pain and psychological outcomes in a sample of

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older adults with chronic low back pain (cLBP). The aims were to empirically identify domains of resilience based upon psychological, social, and health-related factors and, using cluster analysis, explore whether resiliency phenotypes differ across measures of physical function, pain intensity, disability, and psychological functioning. Their findings indicate that individuals with a more resilient phenotype may present a greater sense of coherence that fosters successful mobilization of resources and navigation of ongoing challenges associated with pain. This study is among a small number to suggest the presence of resiliency profiles based upon psychological, social, and health-related functioning and, further, it suggests that health and psychosocial factors are differentially expressed across older adults with cLBP.

Two papers analyzed the role of resilient variables in the adaptation of women with fibromyalgia. Pastor-Mira et al. aimed (1) to develop a Spanish version of the Goal Pursuit Questionnaire (GPQ); and (2) to explore the relationships between goal preferences and health outcomes, testing the moderating role of affect and the mediating role of chronic pain activity patterns in two studies. In Study 1, they adapted the GPQ to a Spanish-speaking population of women with fibromyalgia. The culturally adapted Spanish version resulted in a shorter version with content changes reflected in the list of proposed activities, as a result of the field testing within the target population. The findings of study 2 revealed that a preference for pain avoidance goals are related to pain, disability, and fibromyalgia impact through activity patterns. Affect ratings showed direct and indirect associations with health outcomes, mainly by increasing task-contingent persistence. The authors conclude that interventions should promote adaptive activity patterns (i.e., task persistence) and reduce activity avoidance.

Also in a sample of women with fibromyalgia, Cejudo et al. examined the effects of a mindfulness-based intervention (MBI) on the improvement of subjective well-being, positive affect, trait emotional intelligence (TEI), mental health, and resilience. A design of repeated measures with a control group (CG) was used: before and after the application of the treatment and a 6 months follow-up. The study demonstrates that MBI may be an effective intervention tool to foster resilience and related constructs.

The remaining four studies examine flexibility, pain-specific resilience, and frustration tolerance as adaptation resources for chronic pain patients, and flourishing as a measure of well-being.

Gentili et al. examined the role of psychological flexibility (measured as avoidance, value obstruction, and value progress) in relation to symptoms (pain intensity and anxiety), and functioning (pain interference and depression) among adults with chronic pain applying for participation in a digital ACT-based self-help treatment. Their results show that psychological flexibility, as a resilience factor, significantly contributed to the prediction of pain interference and depression when adjusting for age, pain, and anxiety.

In a study in people living with HIV and chronic pain, Gonzalez et al. report that greater pain-specific resilience is

significantly associated with lower levels of pain interference and pain catastrophizing, greater use of distraction and coping self-statements, and significantly greater heat pain tolerance. Authors suggest that pain-specific resilience may promote adaptation and positive coping in people living with HIV and chronic pain.

In a longitudinal study, Suso-Ribera et al. assessed a sample of individuals with chronic pain at two time points: 2 weeks before starting medical treatment at a pain clinic, and 6 months after. Authors found a reduction in pain intensity and an improvement in physical functioning. The results of the regression analyses show that a decrease in pain intensity is significantly associated with improvements in physical health and that this association is moderated by frustration tolerance.

Finally, Trompetter et al.'s paper explores, in two samples of individuals with chronic pain, the prevalence and sociodemographic, physical and psychological correlates of flourishing, and complement this exploration with a similar examination of risk for psychopathology. Compared to those without chronic pain, people with chronic pain were as likely to flourish, but more likely to be at risk for specific indices of psychopathology. Both flourishing and risk for depression were related foremost to psychological correlates. While engaged living was the most important correlate of flourishing, pain catastrophizing and psychological inflexibility were the most important correlates of being at risk for depression, suggesting potentially distinct underlying pathways for resilience and vulnerability in chronic pain. The authors suggest the Psychological Flexibility model to explain both poor and optimal functioning in the presence of chronic pain.

CONCLUSIONS

The papers included in this Research Topic demonstrate the positive contribution of a diverse array of psychological characteristics in the adaptation and well-being of individuals with chronic pain across the lifespan. Several clinical implications have been derived from the findings. More research is needed to further illuminate trajectories of effective pain management, which may have significant value for future clinical and empirical models in chronic pain.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

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A Reduction in Pain Intensity Is More Strongly Associated With Improved Physical Functioning in Frustration Tolerant Individuals: A Longitudinal Moderation Study in Chronic Pain Patients

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Objective: The onset and chronification of pain often has devastating consequences on the physical and mental functioning of individuals. Medical interventions are quite efficacious in reducing pain levels. However, changes in physical and mental health status after medical interventions are not proportional. In the past decades, rational/irrational beliefs, especially catastrophizing, have contributed to a better understanding of the pain experience. This study explores whether pain reduction efforts are more beneficial for individuals scoring high in rational thinking (moderation).

Methods: The study design was longitudinal. Patients were assessed twice, 2 weeks prior to the start of medical treatment at the pain clinic and 6 months after. A total of 163 patients with heterogeneous pain (mostly low back and neck pain) participated in the study. Their mean age was 58.74 years ($SD = 14.28$) and 61.3% were female.

Results: Overall, there was a reduction in pain intensity ($t = 4.25$, $p < 0.001$, $d = 0.32$). An improvement in physical functioning ($t = 4.02$, $p < 0.001$, $d = 0.19$), but not mental health ($t = -0.66$, $p = 0.511$, $d = 0.11$) was also observed. In the regression analyses, a decrease in pain intensity was moderately associated with improved physical health ($\beta = 0.87$, $t = 4.96$, $p < 0.001$, R^2 change = 0.177). This association was found to be moderated by frustration tolerance ($\beta = -0.49$, $t = -2.80$, $p = 0.006$, R^2 change = 0.039). Specifically, *post hoc* analyses indicated that changes in pain intensity only correlated with changes in physical health when patients reported high frustration tolerance levels ($r = 0.47$, $p = 0.006$, $M = 7$, $n = 32$), but not when patients were intolerant to frustration ($r = 0.28$, $p = 0.078$, $M = 17$, $n = 41$).

Conclusion: The results suggest that frustration tolerance may render adaptive by facilitating the positive effect that a reduction in pain intensity has on physical health status. The study findings are discussed in the context of personalized therapy with an emphasis on how to maximize the effectiveness of current interventions for pain.

Keywords: chronic pain, physical performance, longitudinal studies, frustration tolerance, moderator variables, personalized medicine, thinking skills

INTRODUCTION

The onset and chronification of pain in previously healthy individuals often has profound and pervasive effects on the people's ability to perform physically, as well as on their overall mental well-being (Mehta et al., 2016; Rayner et al., 2016). Not surprisingly, with estimates of chronic pain ranging from 20 to 30% globally (Fayaz et al., 2016; Chenaf et al., 2018), this disease has become one the leading cause of physical disability and sick leave both in Europe and the United States (Gaskin and Richard, 2012; Breivik et al., 2013).

Medical treatment (i.e., oral drugs and surgery) is the most frequent approach in the management of chronic pain (Turk et al., 2011; Gatchel et al., 2014; Reid et al., 2015), as well as the first-line intervention in pain guidelines (Koes et al., 2010; Sarzi-Puttini et al., 2012; Dowell et al., 2016). While there is evidence to support that medical treatment is effective for pain management (Turk et al., 2011; Finnerup et al., 2015), studies have also indicated that such reductions in pain intensity do not lead to a proportional improvement of physical and mental functioning (Gauthier et al., 2008; Menezes Costa et al., 2011; Bendayan et al., 2017), so it is possible that underlying mechanisms that have shown to contribute to a better understanding of the experience of chronic pain (i.e., psychological factors) might as well influence this relationship.

Consistent with the previous idea, research in the past decades has shown psychological factors clearly contribute to a better understanding of the experience of chronic pain (Edwards et al., 2016; Linton et al., 2018; Serrano-Ibáñez et al., 2018). For instance, catastrophizing, a maladaptive form of thinking characterized by a tendency to exaggerate, worry, and anticipate the worst possible consequences of an event (Leung, 2012; Ramírez-Maestre et al., 2017), has been consistently associated with poorer health status of pain patients across numerous investigations (Vancleef and Peters, 2006; Burns et al., 2015; Fallon et al., 2015; Ramírez-Maestre et al., 2017) and has become a fundamental outcome in pain research (Williams et al., 2013). In fact, important for the present investigation, there is previous evidence suggesting an interaction between pain catastrophizing and pain intensity in their relationship with physical health (Suso-Ribera et al., 2017), thus supporting the idea that an interplay between psychological factors and pain intensity in the prediction of health status exists. The extent to which this interplay also occurs longitudinally (i.e., in the relationship between changes in pain intensity and changes in health status) and whether psychological factors other than pain catastrophizing can also act as moderators of pain-health associations remains unclear.

In relation to the latter, an increasing number of psychological factors are now gaining ground in the pain literature, including pain acceptance, coping, self-efficacy, and injustice (McCracken and Eccleston, 2003; Okifuji and Turk, 2015; Yakobov et al., 2018), among others, which suggests that there might be other potential moderators of the pain-to-health relationship in the pain literature. Also importantly, additional forms of irrational/rational thinking other than catastrophizing (Ellis, 1962), namely, demandingness (i.e., rigid requirements expressed in terms of “musts” and “shoulds”), low frustration tolerance (i.e., evaluating certain circumstances as unbearable), and self-downing (i.e., a tendency to make global negative self-evaluations) are starting to receive attention in pain research. For instance, demandingness, in the form of perfectionism, has been associated with higher pain interference and more negative affect in past research (Hadjistavropoulos et al., 2007). Similarly, low frustration tolerance, which has been argued to bear similarities with low acceptance, and self-downing have been associated with poorer mental health status in past research (Suso-Ribera et al., 2016), while self-downing has also shown to contribute to poorer physical functioning (i.e., activity level) when accounting for the role of other pain-related beliefs, such as the belief in the permanence of pain or the tendency self-blame about pain (Stroud et al., 2000). While the aforementioned findings are encouraging and evidence the important role of psychological factors in the pain experience, the extent to which pain catastrophizing and other forms of thinking can help understand why changes in pain intensity after medical treatment are not necessarily associated with improved functioning remains uncertain.

To shed new light into the aforementioned gap in the literature, this study will investigate the moderating role of the previous forms of thinking, namely demandingness, catastrophizing, frustration intolerance, and self-downing, in the relationship between changes in pain intensity and changes in physical and mental health status in a sample of chronic pain patients following a medical intervention. We hypothesize that improvements in health status after a reduction in pain intensity will be facilitated when individuals score high in rational thinking. Conversely, we expect that irrational thinking will result in an inhibition of the positive effects of pain reduction efforts on adaptation to pain (i.e., moderation). These hypotheses come from studies showing that irrational beliefs, which are defined as a maladaptive appraisal of events in which assumptions about reality are inconsistent with that reality, act as underlying cognitive vulnerability factors for distress in front of negative situations, such

as experiencing chronic pain (Visla et al., 2016; Buschmann et al., 2018). By contrast, rational thinking, which would be characterized by a realistic anticipation and preoccupation about future outcomes (i.e., low catastrophizing), a flexible relationship with the reality in terms of preferences as opposed to demands (i.e., low demandingness), openness to difficult experiences while attempting to reach personal goals (i.e., high frustration tolerance), and a tendency to be self-compassionate and to unconditionally self-accept oneself (i.e., low self-downing), is argued to be in accordance with reality (Visla et al., 2016) and, therefore, it would provide resilient resources for well-being (Cristea et al., 2013; Suso-Ribera et al., 2016). In fact, the promotion of rational thinking is a key treatment goal of Cognitive-Behavior Therapy (CBT), perhaps the most popular and empirically supported psychological approaches to a wide range of health problems, including chronic pain (Cristea et al., 2015). In sum, with the present study we expect to find psychological characteristics in the patient that positively influence the pain reduction to pain adaptation relationship.

MATERIALS AND METHODS

Participants

A total of 163 chronic pain patients with non-cancer, musculoskeletal pain participated in this study. All patients were adults aged eighteen or over. Their mean age was 58.74 years ($SD = 14.28$) and 62.0% of them were female. Almost half of patients had not completed secondary education (49.7%), while a smaller percentage had finished technical or university studies (25.8%). At the time of assessment, 36.2% of patients were working, 11.0% were unemployed, and 52.8% were retired.

Duration of pain prior to intervention ranged from 6 months to 49 years, with a median of 2 years (mean = 5.30, $SD = 7.56$). The main pain locations were the lower back (63.9%) and the neck (11.0%). The remaining pain locations occurred at very low frequencies and are not reported to facilitate the readability of the manuscript. Ethnic characteristics were not explored in this study due to the homogeneity of the sample, which was mostly Caucasian. The large majority of participants (93.9%) were Spanish.

Instruments

Pain Intensity

A numerical rating scale (NRS) was used to measure patients' pain intensity at the time of assessment, with patients being asked to rate their pain intensity from 0 = *no pain* to 10 = *worst possible pain*. Numerical rating scales are the gold standard in the measurement of pain and they are recommended due to their associated compliance rate, responsiveness, and ease of use (Hjermstad et al., 2011).

Health Status

The Spanish form of the Short Form-36 Health Survey (SF-36; Ware and Sherbourne, 1992) was administered to evaluate

the pain patient's physical and mental health status. The 36 items in the SF-36 can be grouped into eight dimensions of health, which are either related to physical health (i.e., physical functioning at daily activities, performance at work, pain intensity, and general health) or mental health (i.e., vitality, social functioning, influence of emotions on functioning, and psychological well-being). Two composite scores can be calculated from these eight factors to obtain a Physical Composite Score (PCS) and a Mental Composite Score (MCS). The use of these two broader constructs is preferred as it eliminates floor and ceiling effects of the eight subscales and reduces the number of statistical comparisons (Ware et al., 1995). However, in the present study the use of the PCS was conceptually problematic because it contains a pain intensity scale (i.e., bodily pain), which would contaminate the relationship between the independent (i.e., numerical rating of pain intensity) and the dependent variable (i.e., physical health). Therefore, the Physical Functioning subscale, which measures the individual's ability to perform in daily activities, was used in the present study as a measure of physical functioning. In accordance with standard practice for the SF-36, all scores were scaled to have a 0–100 range, a mean of 50, and a standard deviation of 10. High scores are interpreted as reflecting better health. Items in the SF-36 use various scale responses and response labels, so the reader is addressed to the validation paper for further information on item content (Ware and Sherbourne, 1992). The internal consistency of the SF-36 was good in the present study ($0.69 < \alpha < 0.93$), consistent with previous reports (Alonso et al., 1998).

Rational/Irrational Beliefs

The short, Spanish version of the General Attitudes and Beliefs Scale (GABS-SV; Gonzalez et al., 1996) was used to evaluate participant's tendency to appraise certain situations in a maladaptive manner (Burgess, 1986; DiGiuseppe et al., 1988). The questionnaire differentiates the four processes or styles of thinking proposed by Ellis (1962): demandingness (e.g., "I must have a pleasant, comfortable life most of the time"), catastrophizing (e.g., "It is a catastrophe to be hassled in life"), low frustration tolerance (LFT; e.g., "I cannot tolerate to fail at important tasks"), and self-downing (e.g., "I would be a worthless person if I achieved poorly at tasks that are important to me"). Each scale is composed of six items with response options ranging from 0 = *strongly disagree* to 4 = *strongly agree*. Thus, the maximum score for each scale is 24. All scales are bipolar, with lower scores reflecting rational thinking. The GABS-SV satisfies the recommendations for the assessment of beliefs: it distinguishes processes from content, evaluates cognition rather than behavior, and it does not include affective wording (Fulton et al., 2010). The internal consistency coefficients we obtained are comparable to those reported in previous research (Suso-Ribera et al., 2016). Specifically, estimates in our sample were 0.66 for demandingness, 0.90 for catastrophizing, 0.82 for LFT, and 0.77 for self-downing. The use of the GABS-SV as opposed to other well-established measures of rational/irrational thinking in the pain literature lies in the fact that only pain catastrophizing is frequently evaluated in

chronic pain settings, while measures for the remaining forms of thinking are missing.

Procedure

Participants in this study were recruited from a previous cross-sectional investigation conducted at the Vall d'Hebron Hospital in Barcelona from early 2013 to late 2015, in which the relationship between irrational beliefs and health status was investigated in a sample of 492 patients (Suso-Ribera et al., 2016). Since the previous study was published, 3 new patients have been recruited, so the current cross-sectional sample is composed of 495 patients. Six months after this cross-sectional evaluation was finished, patients were contacted again to investigate the longitudinal role of irrational beliefs in the recovery of these patients. These longitudinal findings are the ones presented in the current investigation.

Eligibility criteria included experiencing chronic pain (recurrent pain for at least 3 months in duration), being over 18 years of age, and giving written consent to participate. From 2013 to 2015, the clinical history of patients programmed for a first consultation at the pain unit was reviewed to check the eligibility criteria of age and pain duration. Next, potential participants were approached by letter by the lead researcher, CSR, 2 weeks before patients had their first appointment at the pain unit. Patients were asked to return the completed questionnaires on the day of the first visit, so all baseline measures were completed before the onset of medical treatment. On the day of the first medical appointment, either a physician or the lead researcher, CSR, officially enrolled the participants by collecting the written informed consent and the questionnaires. Five months after this first appointment, patients were contacted again by letter, and 1 month later (i.e., 6 months after the first appointment) they returned the new set of completed questionnaires (follow-up assessment). The protocol was the same for both assessment points and included an information sheet, an informed consent document, and the questionnaires. To explore the correlation between changes in pain intensity and changes in health status, both constructs were assessed at baseline and follow-up. By contrast, to test the study hypothesis, irrational beliefs were only measured at baseline.

All patients who completed the baseline assessment ($n = 495$) were contacted again approximately 5 months after the first evaluation. Of these, 163 patients returned the completed questionnaires (32.9%). Reasons for discontinuation could be explored for some patients, but these could not be changed. These reasons mostly included hospital discharge, which resulted in decreased motivation to participate in the study or perceived difficulties in delivering the questionnaires back to us, as well as lack of time and motivation.

All patients received the recommended treatment according to published guidelines (Finnerup et al., 2005; Cruccu et al., 2007; Attal et al., 2010; Brix Finnerup et al., 2010). This included pharmacotherapy (analgesics, non-steroid anti-inflammatory drugs, anticonvulsants, antidepressants, and opioids), interventional treatments (injections, radiofrequency, intrathecal pump implants, and spinal cord stimulation), topic treatments (creams and patches), and non-invasive electrical

stimulation (transcutaneous electrical nerve stimulation and iontophoresis). The goal of the present study is not to discuss the effectiveness of each treatment for pain, but to explore whether a psychological construct, namely rational/irrational thinking, can help understand why changes in pain intensity, if existent, are not unequivocally associated with improved physical functioning. Therefore, a more detailed description of treatments for pain is out of the scope of the present investigation.

The Ethics Review Committee of the Vall d'Hebron Hospital in Barcelona approved the present study and all its procedures.

Statistical Analyses

Because a large subset of patients who responded to the baseline assessment did not respond to the second administration ($n = 332$), we compared their characteristics against those of patients who provided data for both measurements. We used a t -test for independent samples to compare their age, pain duration, pain intensity, health status, and levels of irrational thinking. Cohen's d effect sizes are reported. Additionally, we performed a χ^2 test to explore differences in sex. These results are important to discuss the generalizability of findings. Cronbach's alphas will also be calculated for all the study measures to ensure the internal consistency of scores.

Next, paired-samples t -tests were performed to examine changes in pain-related outcomes and psychological variables after medical treatment. Again, Cohen's d effect sizes are reported. We also investigated sex differences in study variables, which might be informative for the reader and help justify the need to include sex as a covariate in the regression analyses. Additionally, Pearson correlation coefficients were calculated to assess the relationship between changes in pain intensity and changes in health, as well as the bivariate associations between baseline measures. To facilitate the interpretation of results, change scores were computed differently for pain intensity and health outcomes. Because pain intensity was expected to decrease with treatment, the change score was calculated by subtracting from baseline score, the post-treatment rating. By contrast, the physical and mental health status were expected to increase with treatment, so changes in health outcomes were obtained by subtracting from the post-treatment rating, the baseline score. By doing this, positive values in any of the change variables can be interpreted in the same direction, that is, as evidence showing that pain and health status improved.

Finally, a series of hierarchical analyses were performed in order to explore the moderating role of irrational thinking in the relationship between changes in pain intensity and changes in health. In the moderation analyses, variables were centered before creating the interaction term. Age, sex, and pain duration were used as covariates due to their relationship with study variables (Park et al., 2016). In order to interpret the moderation, a probing *post hoc* analysis of single slopes was conducted when a significant moderating effect was found. Significance was set at the alpha level of 0.01 to reduce the risk of Type I errors. To ensure that multicollinearity and influential observations were not a problem in the sample, we calculated the variance inflation factor and the standardized DFBETA, which should be smaller than 2 and 1, respectively (Stevens, 2003). There was no missing data in

the study (the questionnaires were revised with the participants when returned at the pain clinic and any missing information was completed by participants on site).

All analyses were computed using PASW Statistics 22 (IBM Corp., 2013).

RESULTS

Sample Characteristics and Comparison Between Study Completers and Participants Who Dropped Out

As reported in **Table 1**, we compared the baseline demographic and clinical characteristics of patients who completed both assessments ($n_1 = 163$) and patients who only provided data for the baseline evaluation ($n_2 = 332$) by means of a Student's t -test. No differences were revealed in any of the continuous variables, including age, pain duration, pain intensity, health status, or rational thinking (all $p > 0.01$). The χ^2 test did not reveal sex differences between completers and non-completers either (62.0 and 64.2% of females in the sample of completers and non-completers, respectively; $\chi^2 = 0.23$, $p = 0.634$).

Sex Differences in Study Variables

As reported in **Table 2**, we found sex differences in pain intensity ($M_{\text{men}} = 7.32$, $SD_{\text{men}} = 1.80$, $M_{\text{women}} = 8.08$, $SD_{\text{women}} = 1.40$, $t = -3.00$, $p = 0.003$; 95% CI = -1.25 , -0.26 , $d = 0.47$) and physical functioning ($M_{\text{men}} = 38.93$, $SD_{\text{men}} = 23.15$, $M_{\text{women}} = 28.87$, $SD_{\text{women}} = 23.49$, $t = 2.67$, $p = 0.008$; 95% CI = 2.62 , 17.50 , $d = 0.17$). Specifically, women reported having more intense pain and were less able to perform in their daily activities due to their health problems. Sex differences were not observed in age, pain duration, mental health, changes in pain intensity and health outcomes after medical treatment, and rational thinking (all $p > 0.01$).

Changes in Pain and Health Outcomes and Bivariate Associations Between Baseline Scores and Change Scores

Table 3 shows the mean-level differences in study outcomes (pain intensity and health status) after 6 months of medical treatment and the correlations between baseline scores. On average, pre-treatment pain reports fell within the moderate-to-severe range (Jensen et al., 2001a).

Regarding changes at the mean-level, there was a significant reduction in pain intensity ($t = 4.25$, $p < 0.001$; 95% CI = 0.33 , 0.90) and an increase in physical functioning ratings ($t = 4.02$, $p < 0.001$; 95% CI = 2.43 , 7.12) after the intervention. Changes in pain intensity and physical health were between small and medium ($d = 0.32$ and $d = 0.19$, respectively). There were no significant changes in mental health at the group level ($t = -0.66$, $p = 0.511$; 95% CI = -2.22 , 1.11).

The Pearson correlations indicated that pain intensity was significantly associated with poorer physical functioning ($r = -0.56$, $p < 0.001$) and mental health status ($r = -0.32$, $p < 0.001$). Irrational forms of thinking, were generally strongly

intercorrelated and significantly associated with poorer mental health (Pearson correlation coefficients ranged from -0.44 to -0.46 , except for demandingness). Irrational beliefs did not correlate with pain intensity and physical functioning.

Additionally, the bivariate analyses revealed that changes in pain intensity were moderately associated with changes in physical health status ($r = 0.42$, $p < 0.001$) and modestly correlated with changes in mental health ($r = 0.20$, $p = 0.010$).

Moderation of Rational/Irrational Thinking

We explored whether irrational forms of thinking moderated the relationship between changes in pain intensity and changes in health status, with an emphasis on physical functioning as this was the measure of health status that revealed changes after the treatment. As reported in **Table 4**, baseline LFT moderated the relationship between changes in pain intensity and changes in physical functioning ($\beta = -0.19$, $t = -2.67$, $p = 0.008$; 95% CI = -0.52 , -0.08). The negative beta coefficient in the interaction between LFT and changes in pain intensity indicates that LFT reduced the contribution of changes in pain intensity on changes in physical health status. A probing *post hoc* analysis and a graphical representation were performed to help interpret this finding (**Figure 1**). Simple slopes were calculated at ± 1 SD from the mean of LFT and changes in pain intensity. At high levels of LFT ($M = 17$), changes in pain intensity were not related to changes in physical health ($r = 0.28$, $p = 0.078$, $n = 41$). Conversely, at low levels of LFT ($M = 7$) the relationship between changes in pain intensity and changes in physical health was moderate and significant ($r = 0.47$, $p = 0.006$, $n = 32$). Similarly, as reflected in **Figure 1**, the strength of the correlation between changes in pain intensity and changes in physical functioning increased with frustration tolerance. In other words, high frustration tolerance operated in favor of change after treatment (i.e., synergistic additive effect).

The remaining moderation effects were not significant, that is, the moderation of demandingness in the relationship between pain intensity and both physical functioning ($\beta = 0.02$, $t = 0.29$, $p = 0.775$; 95% CI = -0.20 , 0.27) and mental health ($\beta = -0.01$, $t = -0.11$, $p = 0.916$; 95% CI = -0.19 , 0.18), the moderation of catastrophizing in the relationship between pain intensity and both physical functioning ($\beta = -0.10$, $t = -1.43$, $p = 0.154$; 95% CI = -0.33 , 0.05) and mental health ($\beta = 0.05$, $t = 0.58$, $p = 0.564$; 95% CI = -0.11 , 0.20), the moderation of LFT in the relationship between pain intensity and mental health ($\beta = 0.05$, $t = 0.61$, $p = 0.543$; 95% CI = -0.12 , 0.23), and the moderation of self-downing in the relationship between pain intensity and both physical functioning ($\beta = 0.02$, $t = 0.27$, $p = 0.788$; 95% CI = -0.21 , 0.27) and mental health ($\beta = 0.01$, $t = 0.15$, $p = 0.880$; 95% CI = -0.17 , 0.20).

No problems of data fit were detected. Specifically, the variance inflation factor was lower than 2 for all predictors, suggesting no problem of multicollinearity. The standardized DFBETA was smaller than 1 for all cases, so no problems of influential observations were detected either.

TABLE 1 | Means, standard deviations, and statistical differences in baseline scores between completers ($n = 163$) and non-completers ($n = 332$).

	Completers Mean (SD)	Non-completers Mean (SD)	<i>t</i>	<i>p</i>	95% CI	<i>d</i>
Age	58.74 (14.28)	58.47 (14.59)	−0.20	0.842	−3.00, 2.44	0.02
Pain duration	5.30 (7.56)	6.30 (8.71)	1.25	0.213	−0.57, 2.56	0.12
Pain intensity	7.79 (1.60)	7.71 (1.70)	−0.49	0.623	−0.39, 0.23	0.05
Health status						
PF	32.70 (23.80)	32.63 (24.29)	−0.03	0.978	−4.61, 4.48	< 0.01
MCS	40.90 (13.05)	38.72 (13.22)	−1.72	0.087	−4.67, 0.32	0.17
Irrational beliefs						
Demandingness	17.59 (3.64)	17.33 (4.05)	−0.70	0.484	−1.00, 0.47	0.07
Catastrophizing	11.76 (5.92)	12.10 (6.39)	0.56	0.577	−0.84, 1.51	0.06
LFT	12.02 (5.17)	12.59 (5.93)	1.041	0.298	−0.50, 1.64	0.10
Self-downing	6.07 (5.31)	6.95 (5.46)	−0.03	0.978	−4.61, 4.48	0.16

TABLE 2 | Sex differences in study variables.

	Men Mean (SD) <i>n</i> = 62	Women Mean (SD) <i>n</i> = 101	<i>t</i>	<i>p</i>	95% CI	<i>d</i>
Age	55.64 (14.80)	60.64 (13.68)	−2.20	0.030	−9.50, −0.50	0.35
Pain duration	4.80 (6.70)	5.61 (8.06)	−0.67	0.504	−3.23, 1.59	0.11
Pain intensity	7.32 (1.80)	8.08 (1.40)	−3.00	0.003	−1.25, −0.26	0.47
Change in pain intensity	0.71 (2.15)	0.55 (1.63)	0.52	0.603	−0.43, 0.74	0.08
Health status						
PF	32.93 (23.15)	28.87 (23.49)	2.67	0.008	2.62, 17.50	0.17
MCS	43.31 (12.69)	39.41 (13.10)	1.87	0.064	−0.22, 8.03	0.30
Change in PF	5.18 (16.45)	4.53 (14.39)	0.26	0.792	−4.20, 5.49	0.04
Change in the MCS	−1.12 (8.88)	−0.21 (11.77)	−0.52	0.602	−4.34, 2.52	0.09
Irrational beliefs						
Demandingness	17.32 (3.57)	17.75 (3.69)	−0.73	0.466	−1.59, 0.73	0.12
Catastrophizing	11.08 (5.51)	12.18 (6.15)	−1.15	0.253	−2.98, 0.79	0.19
LFT	11.94 (5.35)	12.08 (5.08)	−0.17	0.864	−1.80, 1.51	0.03
Self-downing	5.97 (4.87)	6.14 (5.59)	−0.20	0.843	−1.87, 1.47	0.03

TABLE 3 | Mean-level changes in pain intensity and health after 6 months of medical treatment and Pearson correlations between study variables.

	Mean (SD) baseline	Mean (SD) 6 months	<i>t</i>	95% CI	<i>d</i>	Pearson correlations between baseline scores					
						2	3	4	5	6	7
1. Pain intensity	7.79 (1.60)	7.18 (2.13)	4.25*	0.33, 0.90	0.32	−0.56*	−0.32*	0.06	−0.01	−0.04	−0.01
Health status											
2. PF	32.70 (23.80)	37.48 (26.23)	4.02*	4.43, 7.12	0.19		0.37*	−0.15	0.12	−0.03	−0.06
3. MCS	40.90 (13.05)	40.34 (13.05)	−0.66	−2.22, 1.11	0.04			−0.19	−0.46*	−0.46*	−0.44*
Irrational beliefs											
4. Demandingness	17.59 (3.64)								0.44*	0.41*	0.08
5. Catastrophizing	11.76 (5.92)									0.75*	0.60*
6. LFT	12.02 (5.17)										0.58*
7. Self-downing	6.07 (5.31)										

PF, physical functioning; MCS, mental composite score; LFT, low frustration tolerance. * $p < 0.001$.

DISCUSSION

The aim of this study was to investigate the role of rational and irrational beliefs in the evolution of pain intensity and physical functioning and mental health after 6 months of medical

treatment. Previous research had shown that pain reduction efforts do not necessarily result in improved physical health status (Skljarevski et al., 2010), as correlations between changes in pain intensity and changes in physical disability tend to be modest (Ohrbach and Dworkin, 1998; Sullivan et al., 2008;

TABLE 4 | Moderation of frustration tolerance in the relationship between changes in pain intensity and changes in physical functioning.

DV: change in the PCS	<i>b</i>	β	CI (95%)	<i>t</i>	<i>p</i>	<i>R</i> ² change	<i>F</i> change	<i>p</i>
1 Covariates						0.015	0.78	0.506
Age	−0.09	−0.09	−0.25, 0.06	−1.17	0.053			
Sex	−0.30	−0.01	−4.74, 4.15	−0.13	0.245			
Pain duration	−0.16	−0.08	−0.45, 0.13	−1.07	0.895			
2 Change in pain intensity	3.36	0.41	2.20, 4.52	5.73	< 0.001	0.171	33.14	< 0.001
3 LFT baseline	−0.23	−0.08	−0.65, 0.19	−1.07	0.285	0.002	0.43	0.514
4 LFT × change in pain	−0.30	−0.19	−0.52, −0.08	−2.67	0.008	0.036	7.15	0.008

PCS, Physical Composite Score; LFT, low frustration tolerance. Standardized (β) and unstandardized (*b*) betas refer to the final block of the regression. *R*² change is unadjusted. Change scores were obtained by subtracting from baseline score, the post-treatment rating. Thus, positive change scores reflect a decrease in ratings after treatment (i.e., a reduction in pain intensity and physical health status).

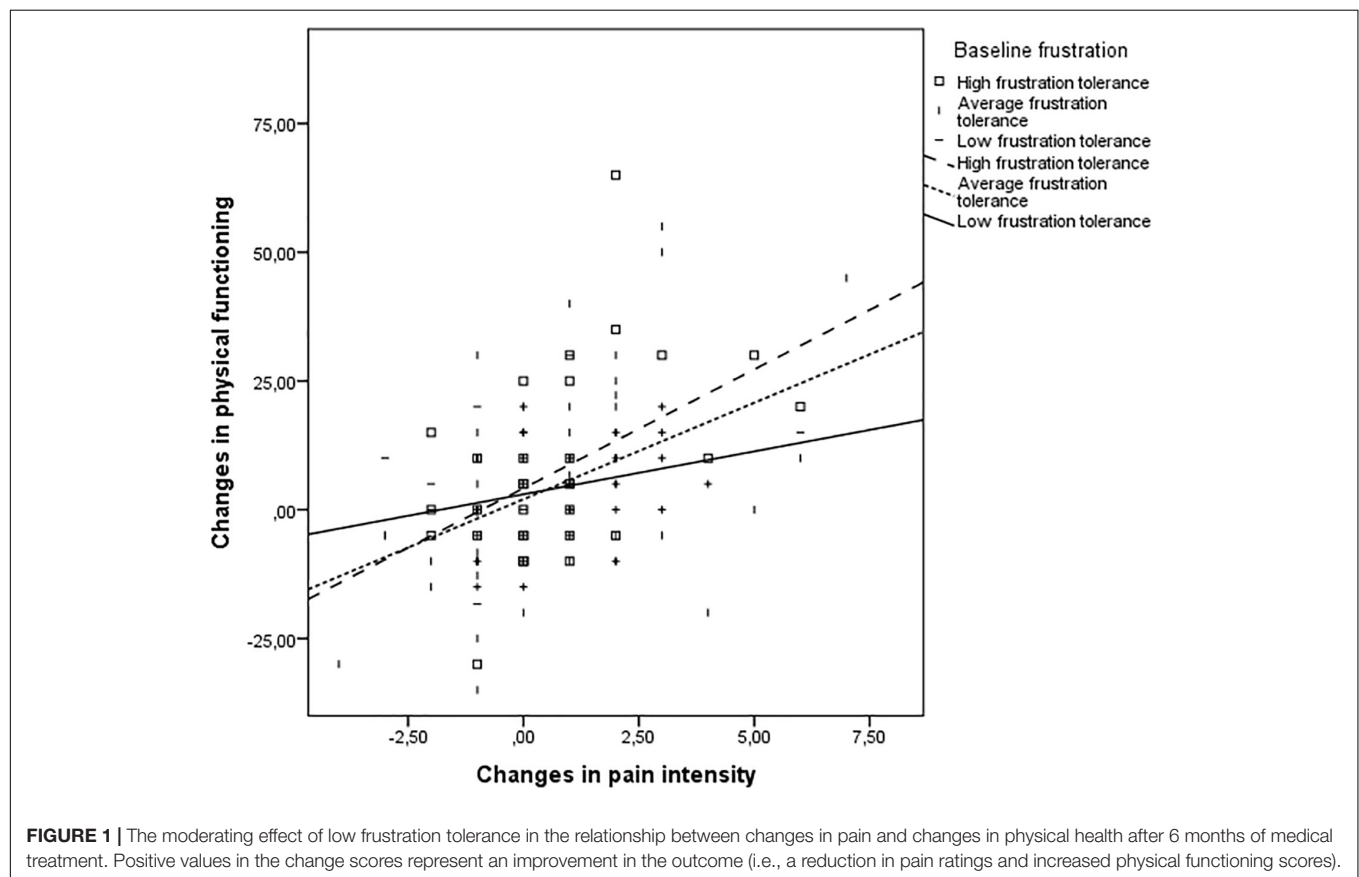


FIGURE 1 | The moderating effect of low frustration tolerance in the relationship between changes in pain and changes in physical health after 6 months of medical treatment. Positive values in the change scores represent an improvement in the outcome (i.e., a reduction in pain ratings and increased physical functioning scores).

Menezes Costa et al., 2011). The current study extends previous findings revealing that psychological factors of the patient, namely LFT, can moderate the relationship between changes in pain intensity and changes in physical health. This might partly explain why reduced pain levels do not unequivocally lead to improved physical functioning.

Contrary to our expectations, only frustration tolerance emerged as a significant moderator in the present investigation. Catastrophizing and, to a lesser extent, demandingness and self-downing or self-criticism have been previously associated with pain outcomes (Stroud et al., 2000; Hadjistavropoulos et al., 2007; Okifuji and Turk, 2015; Ramírez-Maestre et al., 2017). Also importantly, these forms of thinking are moderately

associated with each other (Suso-Ribera et al., 2016), which would justify our study hypotheses (i.e., that all of them would emerge as moderators). While acknowledging the similarities between all the aforementioned forms of thinking, in the next lines we will discuss specific features of frustration tolerance that might have influenced the present study findings. Frustration tolerance is a belief related to the appraisal of situations as being either unbearable (e.g., “I can’t deal with the difficulties life puts me through”) in its irrational pole or tolerable in its rational pole (e.g., “I can tolerate the difficulties life puts me through”) (Suso-Ribera et al., 2016). Thus, different to demandingness, catastrophizing, and self-downing, frustration tolerance taps into aspects of acceptance of reality, regardless

of actual efforts to change that reality (Harrington, 2011). In fact, the similarity between acceptance, a construct that is well established in the chronic pain literature (McCracken et al., 2010), and frustration tolerance has been discussed in previous research (Harrington et al., 2007) and becomes evident when comparing items in the GABS-SV (e.g., “some situations are displeasing and uncomfortable, but I can still function despite them”; David et al., 2010) and items used to assess acceptance of pain (e.g., “I am getting on with the business of living no matter what my level of pain is”; Wicksell et al., 2009). Considering the amount of research showing the importance of acceptance in the context of pain, it is possible that low acceptance of reality, as reflected by high frustration intolerance scores, represents a key distinctive feature of frustration intolerance compared to other forms of thinking, which in turn helps to understand why only frustration tolerance emerged as a significant moderator in the pain-to-health relationship.

In addition to the described differences between frustration tolerance and the remaining forms of thinking, it is also important to note that past pain research has mostly investigated linear associations between thinking styles and outcomes, which are not necessarily generalizable to interaction effects (i.e., moderation). For instance, while pain catastrophizing has been reliably associated with numerous pain-related variables, its moderating role in predicting treatment efficacy (i.e., for whom treatment is more effective) has little support (Wertli et al., 2014), thus indicating that pain interventions will be similarly effective irrespective of baseline levels of pain catastrophizing. By contrast, there is evidence to suggest that acceptance, in the form of psychological flexibility, might explain differential responses to pain interventions (Probst et al., 2018). Consistent with the aforementioned studies, the present investigation revealed that psychological factors that are linearly related to pain outcomes, such as catastrophizing, might not necessarily moderate the effectiveness of interventions and provided further support for the importance of psychological constructs that tap into acceptance of reality (i.e., frustration tolerance) when predicting response to treatment in pain settings. These results should be interpreted as showing that pain treatment effectiveness will be comparable irrespective of baseline catastrophizing, demandingness, or self-downing characteristics of individuals. Additionally, they indicate that the patients' tendency to tolerate discomforting events, such as experiencing pain, will be key in the progression of physical functioning after a medical intervention, maybe because some discomfort (i.e., pain) will still be experienced despite the reduction in pain levels. In other words, it is possible that being open to experience discomfort is more important than being realistic about future outcomes (i.e., low catastrophizing), non-demanding with reality, and self-compassionate (i.e., low self-downing) when it comes to making the most out of medical treatment for pain because some discomfort is likely to be present even if pain intensity is reduced with treatment. While these findings are in line with some previous similar research exploring the moderating role of pain catastrophizing and psychological flexibility in response to treatment (Wertli et al., 2014; Probst et al., 2018), it is important to note that the present is the first investigation

to explore the moderating role of rational thinking in the pain-to-health relationship after medical treatment and one of the first investigations to include all forms of irrational thinking in the same investigation in pain settings, so the reason why moderation only occurred for frustration tolerance and not for the remaining rational beliefs remains speculative at this stage and replication will be needed.

While acknowledging the previous limitation in the conclusions that can be drawn for the present study findings, past research has also shown that the belief that discomforting events cannot be tolerated boosts the negative impact of stressful situations on functioning (Harrington, 2011). By contrast, the belief that difficulties are challenges that can be dealt with is frequent found to be a source of resilience in the face of demanding situations (Esteve et al., 2007; Ramírez-Maestre et al., 2012). This relationship between thinking and outcomes is fundamental to understand how CBT conceptualizes the individuals' functioning. CBT states that people's behavior and emotional states are largely explained by how situations are experienced (Clark and Beck, 2010). Thus, according to this approach, irrational forms of thinking (e.g., catastrophizing about an event) would shape and bias information processing, ultimately leading to maladaptive emotional and behavioral reactions. Indeed, there is research to indicate that a change in irrational thinking is a mechanism explaining the effectiveness of CBT on depression (Cristea et al., 2015). This study evidenced that patients who presented a high frustration tolerance profile were more likely to obtain improvements in physical functioning proportional to the reduction in pain levels, which would support the practice of cognitive flexibility in CBT to increase the tolerance to frustration of these patients. Additionally, several forms of irrational thinking (i.e., catastrophizing, low frustration tolerance, and self-downing) were associated with poor mental health status cross-sectionally, which would provide further support for the important role of thought patterns in understanding emotional states. Only demandingness, which has already been argued to play a modest role when compared with the remaining irrational forms of thinking (Kelly et al., 1998; Suso-Ribera et al., 2016), was not related to mental health.

The moderation of pain-to-health associations after medical treatment is a key finding in the present investigation. The idea that psychological factors can act as moderators of treatment efficacy is not new. In fact, there is an increasing body of research supporting the role of psychological factors as moderators of the effectiveness of psychological interventions (Turner et al., 2007; Miles et al., 2011; Skinner et al., 2012). However, to the best of our knowledge, this is the first study to demonstrate that psychological factors (i.e., frustration tolerance) can also be significant moderators of the effectiveness of medical treatments in pain settings. Specifically, our results indicate that the secondary gains of the intervention (i.e., improved physical functioning as a result of a reduction in pain levels) are higher when individuals present high frustration tolerance. There may be different mechanisms through which frustration tolerance influences the relationship between changes in pain intensity and changes in physical disability. One possibility is that the negativity of frustration intolerance hinders pain reduction

efforts by distorting the perception of physical functionality. Congruent with this idea, one study revealed that depressed patients underestimate their objective levels of physical activity (Huijnen et al., 2010). An ingrained negative belief (e.g., “I can’t deal with physical challenges, such as climbing stairs”) might help create a biased perception that one is physically impaired, which might remain unaltered irrespective of pain reduction efforts. By contrast, a more positive, accepting appraisal of difficulties (e.g., “I can tolerate the pain when doing things that are important to me”) is known to lead to better physical performance (Vowles et al., 2011). Another possibility is that the belief that one cannot manage difficult situations leads to lower mood and, ultimately, to behavioral avoidance, thus contributing to physical disability. Supporting this hypothesis, frustration intolerance has been associated with depressed mood (Buschmann et al., 2018), poor mental health (Suso-Ribera et al., 2016), and low self-esteem (Stephenson et al., 2017). Depressed individuals are, in turn, less active physically (Schuch et al., 2017). Thus, it is possible that frustration-tolerant patients benefit more from the reduction of pain intensity because they present higher mood and remain physically more active. Both hypotheses remain merely speculative at this point.

Sample size was one of the strengths of the present study. Previously reported longitudinal investigations in pain settings have been generally small (i.e., between 40 and 70; for a review, see Jensen et al., 2011), which should make the present work findings relatively robust. However, there are of course a number of limitations in this investigation. Although we explored a set of important psychological factors in the chronic pain literature, especially catastrophizing, the list is far from complete. It is possible, therefore, that other variables frequently considered in pain settings (i.e., acceptance, fear, and perceived injustice) may also moderate the effectiveness of medical interventions. Also in relation to the assessed constructs, it is important to note that all measures were obtained with self-report methods. While this is a frequent practice in pain and health research, it is also true that it is possible that shared method variance might have influenced the results, resulting in stronger associations between variables. At this stage, this remains uncertain for the present study findings. However, the fact that only frustration tolerance and not all rational beliefs were significant moderators in the study makes us think that there is something unique in frustration tolerance which cannot be attributable to shared method variance only. Also importantly, the dropout rate in the study was high (67%) and population was characterized by experiencing heterogeneous pain (mostly low back and neck pain), so the generalizability of findings should be taken with care. While acknowledging this, the sample characteristics in our study (i.e., pain intensity and health status) are comparable (within a 1 SD range) to those of other pain clinics (Keeley et al., 2008; Wetherell et al., 2011), which should make our results useful for a wide number of clinicians and researchers. An additional aspect that should be considered is that the cross-sectional findings with the present study data have been already been reported in previous research (Suso-Ribera et al., 2016). Consequently, we address the readers to the previously reported work for further interpretation of cross-sectional findings. Note, however, that the inclusion of

longitudinal data is clearly new to the present investigation and represents the key aim of the present investigation, for which research questions are largely different from those published previously. Finally, it should be noted that health status can be influenced by many factors other than pain, so we cannot ensure that the physical and mental functioning of patients in our sample was only influenced by pain. To control for this, we used important covariates of health in the regression analyses (i.e., sex and age), but, drawing from existent literature (Cano et al., 2008; Edwards et al., 2016; Cano-García et al., 2017; Kaiser et al., 2017), other candidates surely exist (e.g., medication misuse, treatment modality, anxiety or depressive symptoms, social or family support, and satisfaction with treatment, among others). The fact that the moderation existed while controlling for some important covariates of patient health status should make the present study results robust, but the inclusion of a more comprehensive set of covariates would be desirable to provide further support for the robustness and generalizability of the findings.

While acknowledging the aforementioned shortcomings, we believe that the present study might have important clinical implications. Physical disability due to chronic pain is matter of public concern as the indirect costs of the disease associated with physical limitations (i.e., sick leave, compensations) exceed medical costs for chronic pain patients by a factor of five (Turk, 2002; Gaskin and Richard, 2012). Consequently, it is important to maximize the positive effects that a reduction in pain has on physical functioning so that return to work and daily functioning after an effective pain treatment are enhanced (Hanley et al., 2008; Fedoroff et al., 2013). Thus, the results of the present study may be important in the context of personalized interventions. Personalized therapy has emerged as a result of the heterogeneity of patients’ responses to medical (LeResche et al., 2015) and psychological (Broderick et al., 2016) treatments. The goal of personalized interventions is to detect characteristics of the patient (i.e., genes, personality styles) that explain differences in the effectiveness of interventions (Chapman et al., 2014). The ultimate goal of this approach is to optimize treatment by selecting the most appropriate intervention for each individual. Take, for example, a patient with reports of high pain and poor physical health, arguably due to pain levels. In that situation, one would expect that physical functioning would be improved by decreasing pain intensity. In the light of our findings, this is likely to happen when patients think rationally (i.e., they present high frustration tolerance levels). As opposed to that, a different approach might be needed with patients presenting a low frustration tolerance profile, as they appear to respond similarly to both a decrease and an increase in pain intensity (i.e., no change in physical functioning). Psychological interventions (i.e., CBT) addressing beliefs such as frustration tolerance may therefore be useful in such cases. In fact, the promotion of rational thinking with CBT has already been shown to have positive effects on various health problems, such as hypertensive asthma and breast cancer patients (David et al., 2010). There is also evidence that beliefs can be changed in chronic pain settings (Jensen et al., 2001b; Turner et al., 2007; Morley et al., 2008). In the light of our results, we would expect that, as soon a more rational

form of thinking is adopted, the positive impact that a reduction of pain intensity has on physical functioning will be enhanced. Note, though, that the nature of the present study prevents us from drawing any causal conclusions, so results should be interpreted with caution.

In sum, our results lead us to recommend the assessment of frustration tolerance beliefs before starting pain reduction interventions in pain settings. By doing so, we could personalize treatments by offering psychological treatment (i.e., CBT) to patients scoring low in this form of thinking in conjunction or prior to their usual medical treatment. Further studies are needed to replicate the present study findings, as well as to test whether an early psychological intervention targeting maladaptive beliefs can indeed maximize the secondary gains of pain-reduction efforts (i.e., improved physical functioning).

ETHICS STATEMENT

The Ethics Review Committee of the Vall d'Hebron Hospital in Barcelona approved the present study and all its procedures. The protocol was the same for both assessment points and included an information sheet, an informed consent document, and the questionnaires.

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AUTHOR CONTRIBUTIONS

All authors made substantial contributions to this work—helped in data interpretation, reviewed and discussed the manuscript, and approved the final version of the manuscript after a number of revisions. CS-R, SS-V, and DG-P designed the study. CS-R and LC-G collaborated in data acquisition. CS-R, JO, and DG-P worked in data analysis. CS-R elaborated the first draft of the manuscript.

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Conflict of Interest Statement: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Goal Preferences, Affect, Activity Patterns and Health Outcomes in Women With Fibromyalgia

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Some motivational models understand health behavior as a result of the interaction between goal preferences and mood. However, this perspective has not been explored in fibromyalgia. Furthermore, in chronic pain, it has only been explored with regard to negative affect. Thus, our aims were: (1) to develop a Spanish version of the Goal Pursuit Questionnaire (GPQ); (2) to explore the relationships between goal preferences and health outcomes, testing the moderator role of affect and the mediating role of chronic pain activity patterns. We conducted two cross-sectional studies. In Study 1, after a double translation/back-translation process, we interviewed 94 women attending the Fibromyalgia Unit of the Community of Valencia in order to identify the cultural feasibility and the content validity of the GPQ. Study 2 comprised 260 women. We explored the GPQ structure and performed path analyses to test conditional mediation relationships. Eight activities from the original GPQ were changed while maintaining the conceptual equivalence. Exploratory factor analysis showed two factors: 'Pain-avoidance goal' and 'Mood-management goal' (37 and 13% of explained variance, respectively). These factors refer to patients' preference for hedonic goals (pain avoidance or mood-management) over achievement goals. Robust RMSEA fit index of the final models ranged from 0.039 for pain to 0.000 for disability and fibromyalgia impact. Pain avoidance goals and negative affect influenced pain mediated by task-contingent persistence. They also affected disability mediated by task and excessive persistence. Pain avoidance goals and positive affect influenced fibromyalgia impact mediated by activity avoidance. We also found a direct effect of negative and positive affect on health outcomes. Preference for pain avoidance goals was always related to pain, disability and fibromyalgia impact through activity patterns. Affect did not moderate these relationships and showed direct and indirect paths on health outcomes, mainly by increasing persistence and showing positive affect as an asset and not a risk factor. Intervention targets should include flexible reinforcement of achievement goals relative to pain avoidance goals and positive affect in order to promote task-persistence adaptive activity patterns and decreased activity avoidance.

Keywords: fibromyalgia, goal preferences, activity patterns, affect, health outcomes

INTRODUCTION

Chronic pain and diseases associated with pain are the most important global causes of disability (Rice et al., 2016). Fibromyalgia is a potentially disabling condition characterized by widespread and diffuse musculoskeletal chronic pain not associated with inflammatory or degenerative changes, alongside other physical, affective and cognitive dysfunctions, such as fatigue, non-refreshed sleep, memory problems, decreased attention, and anxiety and depression (Häuser et al., 2015; Arnold et al., 2016). Patients with this chronic pain condition, the cause of which is unknown, usually show high physical and mental comorbidities, such as headaches, irritable bowel syndrome, and rheumatic diseases or stress (Häuser et al., 2015, 2019). Patients with fibromyalgia often report high functional impact, negative consequences in their daily life and negative effects on mood. In addition, reports suggest a high socio-sanitary burden (Häuser et al., 2015). In Europe, estimates indicate a prevalence of 2.5%, and a high proportion of women (Queiroz, 2013). Despite the growing research and scientific literature of recent years, the diagnosis of fibromyalgia is still controversial (Häuser et al., 2019). The 2016 update of diagnosis criteria, based on a self-reported scale, included: generalized pain as defined by pain occurring in at least four of five body regions; a widespread pain index between 4 and 6 and a symptom severity score of ≥ 9 or widespread index ≥ 7 with symptom severity ≥ 5 ; finally, symptoms must be present at a similar level for at least 3 months (Wolfe et al., 2016). Currently, fibromyalgia remains an important clinical challenge and the best treatment approach recommended by experts includes graded physical, pharmacological and psychological strategies depending on the severity of the fibromyalgia condition (Macfarlane et al., 2017; Häuser et al., 2019). The main aim is to increase or maintain the physical, psychological and social functions from a rehabilitation perspective.

Emotions have become a significant topic in chronic pain research, in different theoretical models and at different levels of complexity (Dima et al., 2013). One frequent approach has been to explore the emotion contribution to health outcomes and adaptation in patients with this condition. In this sense, there is broad evidence on the direct and indirect effects of positive and negative affect on physical and psychological health. In general, positive affect appears as an asset and part of the resilience mechanisms whereas negative affect is considered as a vulnerability factor for health in different populations (DeSteno et al., 2013; Chan et al., 2016) including in chronic pain and fibromyalgia (Van Middendorp et al., 2008, 2010; Sturgeon and Zautra, 2013; Estévez-López et al., 2015; Hasset et al., 2016). Studies have also been made of contribution of affect to several fibromyalgia symptoms such as cognitive deficits (Galvez-Sánchez et al., 2018) or fatigue (Estévez-López et al., 2019) with similar relationships for positive and negative affect as those mentioned above. Finally, some authors have shown the relationships of affect to chronic pain patients' activity patterns, exploring its direct effects (Kindermans et al., 2011; Esteve et al., 2017) or its interaction with goal preferences through motivational-affective models

(Vlaeyen and Morley, 2004, 2009), enhancing the affective contextual determinants of activity and adaptation.

In fibromyalgia and chronic pain, disability and also adjustment have been explained from a motivational perspective targeting the role of personal valuable goals on these results (Affleck et al., 2001; Hamilton et al., 2005; Crombez et al., 2012; Vlaeyen and Linton, 2012). In this context, activity limitations due to pain are explained by the simultaneity of several competing goals (Crombez et al., 2012) such as the preference for short-term hedonic goals (i.e., pain avoidance) against long-term achievement goals (i.e., to start or to maintain an activity). Karsdorp and Vlaeyen (2011) performed and validated the Goal Pursuit Questionnaire (GPQ) in people with musculoskeletal complaints to identify the individuals' goal pursuit tendency for hedonic or achievement goals. They explored the relationships of hedonic or achievement goals with pain and disability, and the moderation of negative affect in these relationships. The final version of the GPQ assessed the preference for hedonic goals (pain-avoidance or mood-management goals) in contrast to achievement goals in 16 daily hypothetical situations. They found the endorsement of either pain avoidance or achievement goals were related to pain perception and disability, and that negative affectivity was a significant moderator for pain perception (Karsdorp and Vlaeyen, 2011). As the same authors pointed out, this interaction is similar to the predictions of the mood-as-input model (MAI) which had previously been proposed as a framework to understand the relationships between chronic pain and avoidance or overuse behaviors (Vlaeyen and Morley, 2004; Vlaeyen and Morley, 2009). Indeed Karsdorp and Vlaeyen (2011) designed the GPQ on the basis of this motivational-affective model. The MAI model underlines the informational role of mood in interaction with goals (referred to as stop-rules in the model) in explaining task performance. In people with a preference for hedonic goals, mood informs them whether the activity is pleasurable or not; therefore, positive mood enhances persistence and negative mood encourages disengagement and avoidance. In people with a preference for achievement goals, mood informs them whether goals are reached or not; therefore, positive mood facilitates disengagement and avoidance, and negative mood persistence and overuse. Accordingly, the same mood, in interaction with different goals, encourages either avoidance or persistence behaviors. The model stresses the situational (motivational and affective) determinants of the activity. Affect refers to a predisposition to interpret positively or negatively different stimuli and is more stable than mood. However, we tested the above-mentioned interaction hypotheses, assuming the same effects on avoidance and persistence activity patterns, taking into account that the GPQ was designed to assess people's habitual goal pursuit.

In a meta-analysis of activity patterns and chronic pain, both activity avoidance and excessive persistence (referred to as overuse or overactivity) were associated with poor health outcomes (Andrews et al., 2012). Moreover, there is broad evidence on the important role of activity patterns in chronic pain outcomes (Kindermans et al., 2011; Esteve et al., 2016, 2017). Regarding fibromyalgia patients, avoidance and persistence behaviors have also been linked to more pain and disability

(Van Koulil et al., 2008). To achieve a better understanding of these behaviors and health outcomes in chronic pain, some authors have recommended the investigation of the role of motivational and affective factors from a self-regulation perspective (Vlaeyen and Morley, 2004, 2009; Van Damme and Kindermans, 2015). Research on this issue in fibromyalgia is scarce despite its relevance given the patients' heterogeneity and high prevalence of both avoidance and persistence activity patterns (Van Koulil et al., 2008), the high disabling impact of the problem, the reported low rates of physical activity (McLoughlin et al., 2011; López-Roig et al., 2016), and the perceived difficulties in performing regular physical activity (Pastor et al., 2015; Peñacoba et al., 2017). Our study tested the Karsdorp and Vlaeyen (2011) affect-goals interaction hypothesis, but in a specific chronic pain sample consisting of women with fibromyalgia. Moreover, as a novel contribution, we added the exploration of the effect of these variables on health outcomes through activity patterns, which was recommended by the same authors (Karsdorp and Vlaeyen, 2011). In addition, we studied not only negative affect but also the role of the positive affect in those relationships. Therefore, in the context of a broader research on self-regulation processes and physical activity in fibromyalgia, we conducted two different studies: (1) to develop a culturally adapted version of the GPQ for a Spanish population of women with fibromyalgia, and (2) to explore the relationships of goal preferences to health outcomes by testing the moderator role of affect and the mediator role of the chronic pain activity patterns.

MATERIALS AND METHODS

Design and Procedure

The two studies are the first part of a broader research project which was approved by the Ethics Committees of the Alicante General Hospital and the Miguel Hernandez University. All participants signed the informed consent.

We conducted a descriptive, observational, cross-sectional design in both studies, with the same inclusion criteria: women, aged between 18 and 70 years and with a fibromyalgia diagnosis from the Fibromyalgia Unit (FU) of the Community of Valencia or from other health services in the case of participants from patients' associations.

Regarding Study 1, designed to develop a Spanish version of the GPQ (Karsdorp and Vlaeyen, 2011), the authors of the scale sent us the GPQ Dutch original version and authorized its adaptation. We then conducted a double translation/back-translation process and two consensus meetings. Two independent translators provided two target Spanish versions which were translated back to Dutch by two other independent translators. Translators and back-translators translated into their mother tongue. Discrepancies were solved by consensus and we developed a back-translated Dutch version which was compared for equivalence with the original by a bilingual psychologist (López-Roig and Pastor, 2016). Finally, at the FU setting, we performed a field study with four sub-studies: (1) a group structured interview after group self-administration of the GPQ ($n = 26$); (2) a thinking-aloud study ($n = 16$);

(3) a group self-administration questionnaire comprising only the activities listed in the GPQ to study their frequency in the daily life of fibromyalgia patients ($n = 27$); and (4) a group self-administration of the Spanish version of the WHYMPI-part III (Pastor et al., 1995), which assessed the frequency of several daily life activities ($n = 25$). With these sub-studies we aimed to assess the feasibility of the GPQ, its clarity (instructions, items and answer scale: sub-studies 1–2) and the appropriateness and content validity of the 16 situations listed in the final version of the original questionnaire (sub-studies 1–4) for the Spanish context and fibromyalgia. In this sense, in the adapted GPQ version for these sub-studies, we also asked participants if they considered each situation as usual or “typical” in people like them and, if not, they were asked to describe another activity with similar emotional or painful consequences. With these added questions in each item of the GPQ pilot version (sub-studies 1–3) and the activities listed in WHYMPI-III (sub-study 4) we aimed to identify common activities in the daily life of these patients to adapt any unknown or unfamiliar situation from the original questionnaire, and to check their conceptual and experiential equivalence (López-Roig and Pastor, 2016). We changed the situation in the original questionnaire if more than 50% of participants had not performed the activity and if more than 50% of the participants considered people in their condition did not perform it.

In Study 2, designed to explore the relationships between goal preferences, affect, activity patterns and health outcomes, questionnaires were self-administered in group sessions to other patients attending the same FU setting ($n = 163$) and in an on-line version for participants from patients' associations from the Community of Valencia ($n = 97$). Self-administration lasted over 45 min. The total of 260 participants is over the minimum of 200 suggested as sample size for this kind of studies (Izquierdo et al., 2014; Lloret et al., 2014).

STUDY 1. TRANSLATION AND CULTURAL ADAPTATION OF THE GPQ

Method

Participants

Ninety-four women attending the Fibromyalgia Unit (FU) of the Valencian Community participated in the adaptation process of the GPQ. Most were married (65%) and had primary (37.2%) and secondary studies (32%). At the time of the study 23% of women were working. Mean age was 51.3 ($SD = 10.5$) and the mean of perceived pain intensity was 7.3 ($SD = 1.8$); rank 0 = “no pain at all” and 10 = “the worst pain you can imagine.” See description of this measure in Study 2.

Variables and Instruments

Socio-demographic and clinical variables were measured with an “ad hoc” questionnaire.

Goal Pursuit Questionnaire (GPQ)

This instrument measures the habitual goal pursuit of people with pain, taking into account hedonic or achievement goals

which can be activated at the same time in one situation. We adapted the final version with 16 items answered on a 6 point Likert scale (1 = strongly disagree, 6 = strongly agree) which has shown adequate psychometric properties (Karsdorp and Vlaeyen, 2011). The questionnaire was designed taking into account the above-mentioned MAI model. Items refer to different daily situations or activities related to work, study or leisure, contrasting hedonic and achievement goals. Items belong to three categories: painful situations ($n = 8$), unpleasant non-painful situations ($n = 3$), and pleasant non-painful situations ($n = 5$). People with pain are required to rate their preference for a hedonic goal or an achievement goal, choosing pain avoidance or mood management (avoiding an unpleasant non-pain situation or maintaining a pleasant non-painful situation). People must imagine ‘as vividly as possible’ the situation presented in a vignette and rate their agreement with a specific thought which refers to their preference for hedonic or achievement goal in this specific situation. The final version of the GPQ showed a structure of two factors, both with 8 items, named: ‘Pain-avoidance goal’ (Factor I; $\alpha = 0.88$) and ‘Mood-management goal’ (Factor II; $\alpha = 0.76$). Higher mean scores in each factor indicate stronger preferences for a hedonic goal relative to an achievement goal, that is, to avoid pain (Factor I) or to maintain positive mood (Factor II). Factor I showed low significant and negative relationships with negative affect, sense of responsibility, perfectionism, and fear of negative evaluations. Factor II showed only low significant positive relationships with pain catastrophizing and negative relationships with conscientiousness (Karsdorp and Vlaeyen, 2011). All were coherent with the theoretical predictions.

West Haven Yale Multidimensional Pain Inventory (WHYMPI)-part III

The Spanish version contains 16 items and measures the extent of participation in common daily activities of chronic rheumatic patients (Pastor et al., 1995). Items are answered on a numerical rating scale from 0 (never) to 6 (very often). In this study, our interest was limited to the rate for each individual item.

Data Analysis

Data were analyzed by SPSS-v24. We conducted a descriptive analysis to analyze sample characteristics and items of the GPQ.

Results

Instructions and four activities (items 1, 2, 10, 11) were extended to solve problems of comprehension. In both cases, we took into account the comments and the activities expressed in the group structured interview (sub-study 1) and in the thinking-aloud procedure (sub-study 2). For example, regarding instructions, we emphasized the hypothetical condition of the situations (*Remember they are hypothetical situations. It is possible that you have not experienced them or never will. Please, answer imagining yourself in that situation*). In items 1 and 11 we added by hand not only ‘by computer.’ In item 2 we clarified ‘amazing holidays’ by adding *or some amazing thing which has happened to you*, and in item 10, ‘receive an e-mail,’ we added *or WhatsApp*. Finally, eight situations (items 3, 6, 8-10, 12, 14, 15)

TABLE 1 | Original and alternative activities for the GPQ items.

Original situation (item number) <i>New situation^a</i>	Never done	
	Participants %	Other people with fibromyalgia ^b %
... paint the windows frame (3)	88.9	85.2
... <i>clean the windows</i>		
... load boxes for a move (6)	81.5	70.4
... <i>load the shopping bags or do the shopping</i>		
... study for an exam (8)	63.0	51.9
... <i>read a book</i>		
... finish the assembly line work (9)	92.6	74.1
... <i>organize clothes for the washing machine</i>		
... do a presentation (10)	76.9	53.8
... <i>do a task</i>		
... play an instrument in an orchestra (12)	96.3	70.4
... <i>sewing</i>		
... repair the car (14)	88.9	77.8
... <i>clean the car</i>		
... enjoy writing a report (15)	63.0	51
... <i>enjoy watching TV</i>		

^aIn italics new situations reported by patients; final Spanish version is available upon request. ^bPercentage of participants who consider other people with fibromyalgia do not perform it.

were changed using the alternative situations proposed by women with fibromyalgia (Table 1). Activities reported by patients as alternatives in items 6, 9, and 14 were also reported as frequent or sometimes in WHYMPI (88, 100, and 48% respectively of participants in sub-study 4).

STUDY 2. EXPLORATION OF THE RELATIONSHIPS BETWEEN GOAL PREFERENCES, AFFECT, ACTIVITY PATTERNS AND HEALTH OUTCOMES

Method

Participants

A total of 260 women from the FU ($n = 163$) and from patients’ associations of the Valencian Community ($n = 97$) were recruited. Most were married (71.2%) and had primary (36.9%) and secondary studies (38.5%). Mean age was 51.2 ($SD = 8.7$). At the time of the study 31.5% were working and 21% were on sick leave. The mean time from the first symptoms was 15.9 years ($SD = 11.4$) and from the diagnosis it was 7.9 ($SD = 8.0$). The mean of perceived pain intensity was 6.9 ($SD = 1.4$).

Variables and Instruments

Socio-demographic and clinical variables were measured with the same *ad hoc* scale as in Study 1. In Study 2, we used the culturally adapted version of the GPQ from Study 1. Regarding validity based on the relation to other constructs,

we explored whether high pain catastrophizing would be related to greater endorsement of hedonic goals and whether perfectionism and fear of negative evaluations would be related to greater endorsement of achievement goals (Karsdorp and Vlaeyen, 2011). Our final purpose was to explore whether the preference for hedonic or achievement goals in interaction with positive and negative affectivity would be related to different activity patterns and to health outcomes. Therefore, in the Study 2 we employed:

Pain catastrophizing (PCS)

We used the total score of the Spanish adaptation of the Pain Catastrophizing Scale (García-Campayo et al., 2008). This scale contains 13 items answered on a 5-point Likert scale from 0 (not at all) to 4 (all the time) (rank 0–52). Higher scores represent higher catastrophizing ($\alpha = 0.95$).

Perfectionism

We used the total score of the Spanish version of the Frost Multidimensional Perfectionism Scale (FMPS; Gelabert et al., 2011). This scale contains 35 items answered on a 5-point Likert format from 1 (totally disagree) to 5 (totally agree). Higher total score represents higher perfectionism (range 35–175) ($\alpha = 0.94$).

Fear of negative evaluations

Measured with the total score of the Spanish adaptation of the Brief version of the Fear of Negative Evaluation Scale–Straightforward (BFNE-S; Pitarch, 2010). The scale contains 8 items rated on a 5-point Likert scale (1 = not at all characteristic of me; 5 = extremely characteristic of me; range: 8–40). High total score indicates high fear of negative evaluations ($\alpha = 0.94$).

Positive and negative affect

We used the total score of the corresponding trait version subscales (Positive affect: 10 items; Negative affect: 10 items) of the Spanish adaptation of the Positive and Negative Affect Schedule (PANAS; Estévez-López et al., 2016). Items are rated on a 5-point Likert scale from 1 (not at all or very slightly) to 5 (extremely). Scores range from 10–50 in each case. High total score indicates high positive ($\alpha = 0.90$) or negative affectivity ($\alpha = 0.91$).

Avoidance and persistence activity patterns

We used the Spanish adaptation of the activity patterns scale (Esteve et al., 2016) which contains 24 items rated with a 5-point Likert scale (0 = never, 4 = always) and grouped into eight subscales measuring avoidance (two subscales) persistence (three subscales) and pacing (three subscales). For this study, we only used the subscales related to avoidance and persistence activity patterns: pain avoidance (avoidance behavior related to pain intensity fluctuations; $\alpha = 0.75$), activity avoidance (avoidance behavior related to the own chronic pain condition; $\alpha = 0.55$), task-contingent persistence (persistence in finishing task despite pain; $\alpha = 0.84$), excessive persistence (overuse, persistence without recognition of the own physical limits and with negative rebound effects of this kind of activity; $\alpha = 0.65$), and pain-contingent persistence (activity is variable depending on pain experience; $\alpha = 0.78$). Scores on each scale ranged from 0 to 12.

Pain intensity

Measured with the mean score of the maximum, minimum, and usual pain intensity during the last week and pain intensity at time of the assessment. These items were answered with an 11-point numerical rating scale (0 = “no pain at all” and 10 = “the worst pain you can imagine”). High mean scores indicate high pain intensity ($\alpha = 0.78$).

Disability

We used the Spanish adaptation of the FIQ-R (Salgueiro et al., 2013). Disability was measured with the sum of the first 9 items divided by 3 (rank 0–30). Items are answered on an 11-point numerical rating scale from 0 to 10. Higher scores represent higher disability ($\alpha = 0.89$).

Fibromyalgia impact

The total score of the above-mentioned questionnaire (rank 0–100). Items are answered on an 11-point numerical rating scale from 0 to 10, with different verbal anchors depending on the item. Higher scores represent higher impact perception ($\alpha = 0.93$).

Data Analysis

We conducted a descriptive analysis for sample characteristics and items of the GPQ. With regard to validity analysis based on internal structure, we performed an Exploratory Factor Analysis (EFA) using the maximum likelihood (ML) method and oblique rotation following the recommended standards (Lloret et al., 2017). Previously, we analyzed whether our data fitted the conditions for linear factor analysis (Lloret et al., 2017) and we tested the floor and ceiling effects of each item (percentage of response above 95% in scores 1 and 6). Factors were selected by the scree plot, Kaiser’ rule and baseline theory. We obtained the Kaiser-Meyer-Olkin index and the Bartlett sphericity test to explore the sampling and data adequacy. Items were retained with loading values greater than 0.45. We also calculated the item-corrected scale correlation with the Pearson coefficient. Pearson correlation was also used for assessing the validity of the GPQ based on the relation to other constructs. Statistical significance was set at $p < 0.05$. Cronbach’s alpha and Omega index was calculated for internal consistency of the scales in our sample. Excepting Omega index, these data were analyzed with the SPSS-v24 (Ventura-León and Caycho-Rodríguez, 2017).

Regarding the interaction effect of affect with goal preferences on activity patterns and the mediation of these on health outcomes, we performed a path analysis. Based on raw data, correlations were converted to a covariance matrix. Model fitting was performed by the lavaan package in R (Rosseel, 2012). The results were reported following the recommendations given in the classic study by Raykov et al. (1991).

MVN package in R (Korkmaz et al., 2014) was used to study assumptions of multivariate and univariate normality. Mardia’s multivariate normality test showed no multivariate normality. Shapiro–Wilk univariate normality tests showed non-normality in all the variables. No missing data were found. Twenty-one outliers were detected by the outliers R package (Komsta,

2011), established on the adjusted quantile method based on Mahalanobis distance, and substituted by the median value.

Conditional mediation models were tested, using two (avoidance activity patterns) or three (persistence activity patterns) mediators depending on the model, and one moderator (positive or negative affect in each case). The modeling process started with a complete model (all the predictors, the moderator, the mediators and one dependent variable) and was improved step by step.

Estimation was calculated by maximum likelihood procedure with robust standard errors and a Satorra–Bentler scaled test statistic, due to the non-normality of the data. Models were improved by removing non-significant parameters and by index modification recommendations, until fit criteria were accomplished, and all parameters were significant. A fit-criteria assessment was conducted according to the Hu and Bentler (1999) study. The goodness-of-fit statistical test assesses the magnitude of unexplained variance; a ratio of $\chi^2/df < 2$ suggests an acceptable fit. An RMSEA size below 0.06 suggests a well-fitting model. A CFI above 0.95 indicates a good fit. An SRMR of less than 0.09 also indicates a good fit. The χ^2 statistic provides a conventional measure of model fit. However, because of its sensitivity to sample size, 2 additional fit indices were used to supplement the χ^2 statistic. The choice of these 2 indices was based on Hu and Bentler (1999) recommendation of a 2-index presentation strategy, which was found to provide an optimal balance between type I and type II error rates. All these indicators of model fit will be examined later in order to assess whether the model properly represents the data.

Figures 1, 2 represent the tested structural models, with exogenous and endogenous variables. All variables were observed variables and measured on an interval rating scale. The arrows indicate the directionality of the relationships among the variables. In order to simplify the path

diagram, the hypothesized effects between each variable are represented with one arrow.

Figure 1 represents the first type of model, a moderated (by positive or negative affect) mediational model with the two avoidance activity patterns as mediational variables (pain avoidance and activity avoidance). These models were tested with positive and negative affect and with three different dependent variables (pain, disability, and fibromyalgia impact). Therefore, 6 analyses were performed.

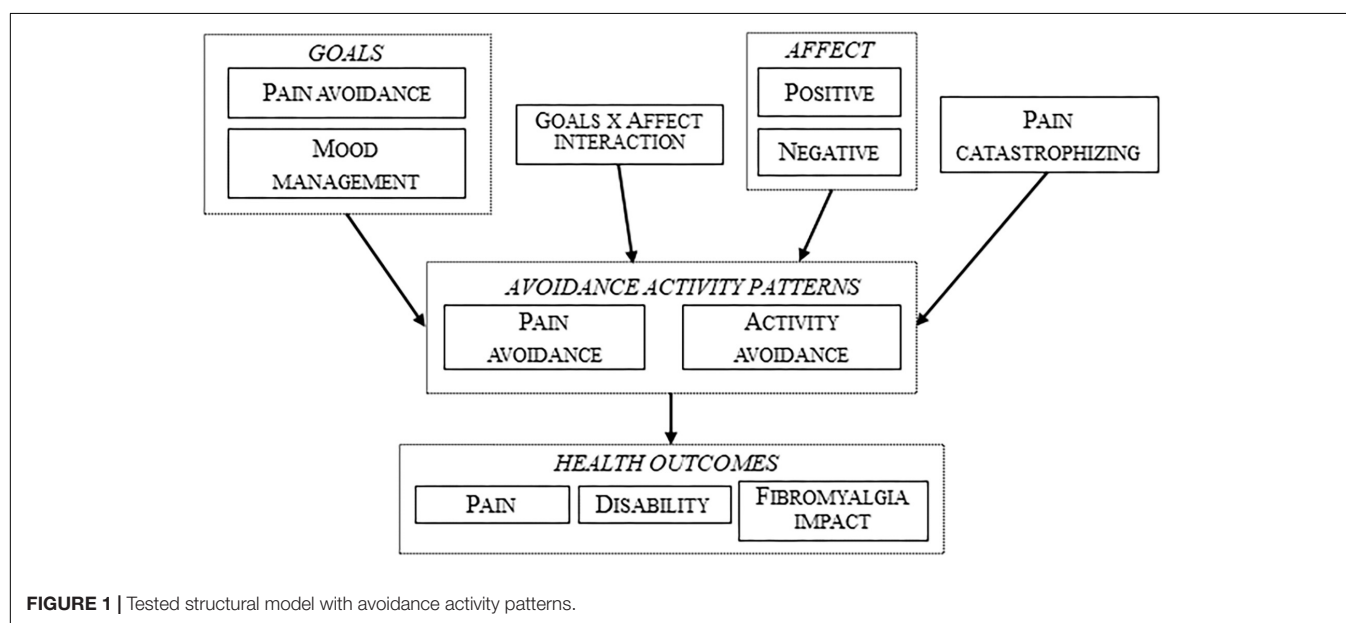
Figure 2 represents the same scenario but with the three persistence activity patterns (task-contingent persistence, excessive persistence, and pain-contingent persistence) as mediational variables.

Following the premises of the fear-avoidance model, pain catastrophizing was only included in the models with avoidance activity patterns. All the models were improved until all the parameters were significant and the global fit indexes were adequate.

Results

GPQ Analysis

All items had answers on all six response options, and they were normally distributed (Kolmogorov–Smirnov test). We found no floor or ceiling effects. Item number 4 showed the highest skewness (1.8) and kurtosis (2.5) (Table 2). The KMO test was 0.90 and the Bartlett test was 1869.9 ($p = 0.000$) indicating the adequacy of the sample and the correlation matrix to perform the EFA. The scree plot showed that two major factors and one minor factor accounted for 49.9% of the variance. Items 5 (doing calculations; factor loading = 0.42) and 8 (reading a book; factor loading = 0.18) did not reach the minimum established factor loading and were removed from the scale. The third factor was not considered as it only accounted for 2.9% of the variance. No items reached the loading criteria (the highest loading was 0.34 for the



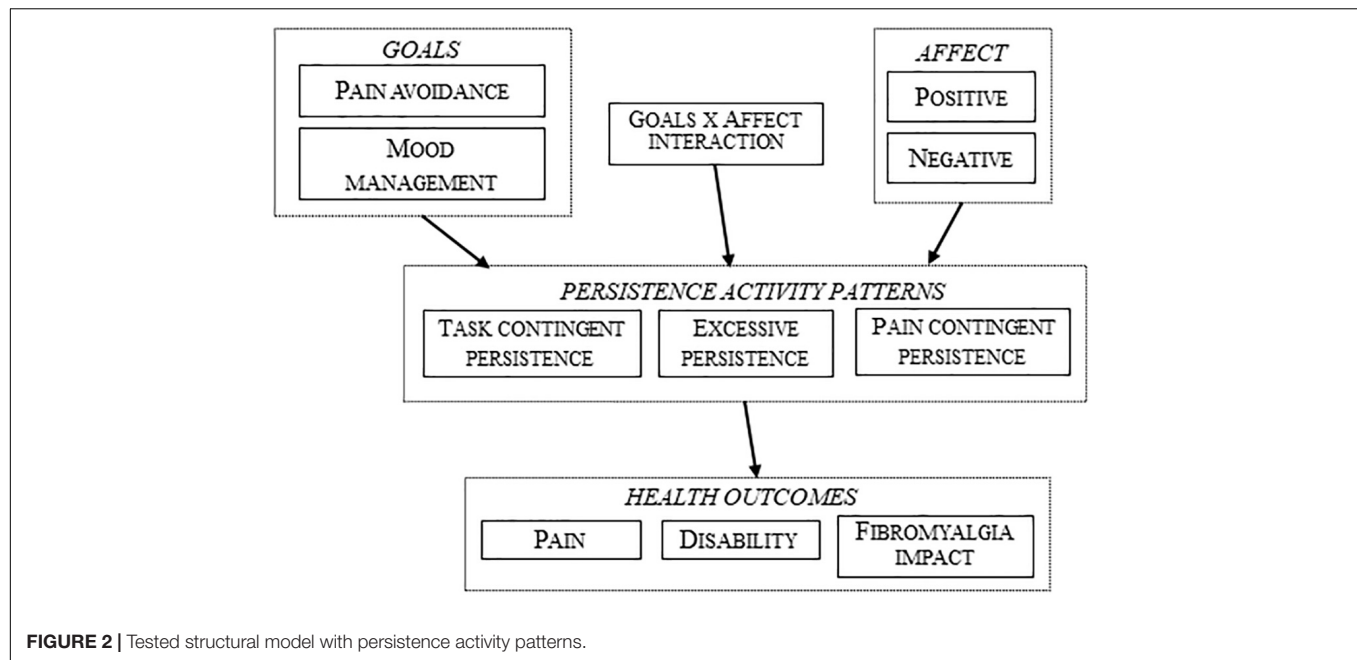


TABLE 2 | Item and factor analysis, descriptive and internal consistency of the GPQ.

Item	I think it is more important...	Loading	<i>M</i> ^a	SD	Sk	K	<i>r</i> _{I-T}	<i>h</i> ²	α/Omega
Factor II. Pain-avoidance goal			3.9	1.3	-0.4	-0.8			0.90/0.93
7	...for the pain in my back to be reduced now, than for the house to be cleaned	0.82	4.2	1.8	-0.6	-1.1	0.75	0.66	0.88
3	...for the pain in my shoulder to be reduced now, than the windows to be cleaned	0.82	4.1	1.9	-0.4	-1.3	0.71	0.62	0.88
14	...for the pain in my forearm to be reduced now, than the car to be cleaned	0.81	4.1	1.7	-0.4	-1.2	0.75	0.66	0.88
6	...for the pain in my upper back to be reduced now, than the shopping to be finished	0.78	3.9	1.8	-0.3	-1.4	0.74	0.63	0.88
11	...for the pain in my wrists to be reduced now, than for the album to be completed	0.74	4.2	1.8	-0.6	-1.0	0.72	0.66	0.88
12	...for the pain in my hands to be reduced now, than for the sewing to be finished	0.72	4.3	1.7	-0.6	-0.8	0.72	0.73	0.88
1	...for the pain in my neck to be reduced now, than for my report to be finished on time	0.49	3.5	1.8	0.02	-1.3	0.50	0.33	0.90
16	...for the pain in my elbow to be reduced now, than for the meeting to be arranged	0.48	3.2	1.7	0.2	-1.2	0.56	0.38	0.90
Factor III. Mood-management goal			2.5	1.1	0.6	0.1			0.81/0.85
10	...to write a nice message (e-mail or WhatsApp), than to finish the task	0.75	2.4	1.5	0.9	-0.2	0.61	0.50	0.76
4	...to read the exciting book now, than to finish the report on time	0.70	1.8	1.3	1.8	2.5	0.61	0.57	0.77
9	...to decrease my boredom, than to organize clothes for laundry	0.68	2.3	1.6	0.9	-0.3	0.62	0.51	0.76
15	...to enjoy watching the TV program, than to finish my chores	0.62	3.0	1.6	0.3	-1.2	0.55	0.44	0.78
13	...to have interesting conversations now, than to make decisions	0.53	3.3	1.6	0.1	-1.0	0.50	0.34	0.79
2	...to tell my holiday stories or something amazing, than to finish my work	0.49	2.3	1.5	0.9	-0.3	0.50	0.42	0.79

GPQ, Goal Pursuit Questionnaire; Sk, Skewness; K, Kurtosis; ^aRank [1–6].

item 12) and they had high loadings in the other factors. A second EFA without these two items (KMO = 0.88; Bartlett test = 1706.6; $p = 0.000$) showed two mayor factors accounted for 50% of the variance. **Table 2** shows the factor pattern matrix with loadings and descriptive data of the items. Factor I ('Pain-avoidance goal': 37% of explained variance; eight items) refers to the choice between pain avoidance goals or achievement goals in different situations. Higher scores reflect stronger preferences for pain-avoidance goals. Factor II ('Mood-management goal': 13% of explained variance; six items) refers to the choice between mood-management goals or achievement goals, with higher scores reflecting stronger preferences for

mood-management goals. Correlation between both factors was moderate ($r = 0.42$, $p \leq 0.01$).

Descriptive data and correlations are in **Table 3**. 'Pain-avoidance goal' was significant and negatively related to perfectionism, and fear to negative evaluation (both $p \leq 0.01$). 'Pain-avoidance goal' and 'Mood-management goal' were significant and negatively related to negative affect ($p \leq 0.05$ for mood management). 'Pain-avoidance goal' was related to more avoidance and less persistence (ranged from $r = 0.52$, $p \leq 0.01$ for pain avoidance activity pattern to $r = -0.12$, $p \leq 0.05$ for pain-contingent persistence). 'Mood-management goal' factor showed significant correlations with only three

TABLE 3 | Pearson correlation coefficients and descriptive statistics for measured variables in the study.

Measure	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
(1) Pain avoidance goal	0.42***														
(2) Mood management goal	0.10	0.03													
(3) Positive affect	-0.24***	-0.14*	-0.16*												
(4) Negative affect	-0.11	-0.10	-0.29***	0.53***											
(5) Pain catastrophizing	0.52***	0.15*	-0.02	-0.17**	0.01										
(6) Pain avoidance	0.23***	0.02	-0.31***	0.04	0.29***	0.35***									
(7) Activity avoidance	-0.46***	-0.14*	0.16*	0.26***	0.05	-0.60***	-0.27***								
(8) Task-contingent persistence	-0.30***	-0.09	0.01	0.49***	0.32***	-0.35***	-0.05	0.55***							
(9) Excessive persistence	-0.12*	-0.15*	0.11	0.31***	0.17**	-0.16*	0.03	0.34***	0.41***						
(10) Pain-contingent persistence	-0.08	0.08	-0.21***	0.21***	0.37***	0.05	0.17**	-0.09	0.16**	0.03					
(11) Pain	-0.02	0.00	-0.27***	0.29***	0.33***	-0.04	0.26***	-0.03	0.24***	0.02	0.56***				
(12) Disability	-0.04	0.00	-0.34***	0.36***	0.44***	0.00	0.37***	-0.05	0.25***	0.05	0.63***	0.88***			
(13) Fibromyalgia impact	-0.26***	-0.07	-0.04	0.54***	0.42***	-0.22	0.11	0.29***	0.42***	0.28***	0.15*	0.29***	0.33***		
(14) Perfectionism	-0.24***	-0.04	-0.16**	0.57***	0.42***	-0.14*	0.03	0.17**	0.39***	0.12	0.00	0.19**	0.21***	0.54***	
(15) Fear of negative evaluation	3.94	2.53	23.16	28.83	28.78	6.18	7.38	6.70	6.62	9.00	6.97	19.96	68.88	101.83	21.26
Mean	1.23	1.06	7.46	9.08	12.18	2.44	2.29	2.63	2.61	2.20	1.41	5.93	16.85	27.55	8.93
SD	-0.41	0.62	0.49	0.20	-0.26	0.13	-0.10	-0.30	-0.16	-0.56	-0.44	-0.79	-0.53	0.36	0.32
Skewness	-0.78	0.03	-0.23	-0.69	-0.77	0.13	-0.39	-0.34	-0.34	-0.12	0.84	0.34	-0.10	-0.51	-1.00
Kurtosis															

*** $p \leq 0.001$, ** $p \leq 0.01$, * $p \leq 0.05$; note: 21 outliers of these data were previously substituted by the median.

activity patterns, ranging from $r = -0.14$, $p \leq 0.05$ for task-contingent persistence to $r = -0.15$, $p \leq 0.05$ for pain-contingent persistence (and with the same value but with positive sign for pain avoidance).

Model Fit

The basic starting models were designed according to **Figures 1, 2**. The fit of the following models was evaluated (**Tables 4, 5**), and figures were generated by the lavaanPlot package in R (Lishinski, 2018), except for simpler multivariate regression models (models without mediation). Non-standardized parameters can be found in tables and standardized parameters are shown in the figures for greater clarity.

Goal models with affect moderation and mediation of the two avoidance patterns

No interaction effects were found between goal preferences ('Pain-avoidance goal' or 'Mood-management goal') and affect (positive or negative) in any tested model for pain, disability or fibromyalgia impact. Moreover, no multivariate models fitted for pain intensity, either testing the model with positive (no model fitted) or with negative affect (only pain catastrophizing predicted pain in a simple univariate model) (**Table 4**).

Regarding disability, the models were exactly the same with positive and negative affect: pain catastrophizing predicted disability directly and indirectly, through activity avoidance; in addition, 'Pain-avoidance goal' showed a significant and indirect path on disability through activity avoidance (**Figure 3**). Affect (positive and negative) did not show any significant contribution.

Fibromyalgia impact was predicted by positive affect directly and indirectly through activity avoidance (negatively in both cases). 'Pain-avoidance goal' also showed a significant and indirect path with this variable, through activity avoidance (**Figure 4**).

Finally, the fitted model, taking into account the negative affect, showed fibromyalgia impact was influenced directly and indirectly, through activity avoidance, by pain catastrophizing. Moreover, the 'Pain-avoidance goal' had an indirect effect on fibromyalgia impact, and negative affect a direct effect (**Figure 5**).

Goal models with affect moderation and mediation of the three persistence patterns

No interaction effects were found between goal preferences ('Pain-avoidance goal' or 'Mood-management goal') and affect (positive or negative) for pain, disability and fibromyalgia impact when we tested models taking into account the mediation of the persistence activity patterns (**Table 5**).

Regarding pain intensity, different models were fitted when we tested goal preference with positive and negative affect. Positive affect showed an indirect path on pain intensity through task-contingent persistence and received the influence of pain intensity with negative sign. In addition, task-contingent and excessive persistence influenced pain with negative and positive sign, respectively. Goal preference did not show any significant contribution in this model. Task-contingent persistence and excessive persistence correlated ($B = 3.56$, $p < 0.001$) (**Figure 6**).

Goal preference played a role in pain intensity when we explored the model with negative affect (**Figure 7**). Both, negative affect and 'Pain-avoidance goal' showed an indirect significant

TABLE 4 | Fitted models with test statistics and path coefficients: goal preferences and affect mediated by avoidance patterns.

Model and fit	Predictor	Dependent Variable	B	SE	z	Effect size
Avoidance patterns						
Pain with Negative affect $\chi^2 = 0.000(0)$; CFI = 1.000; RMSEA = 0.000; SRMR = 0.000	Pain catastrophizing	Pain	0.041	0.007	5.730***	0.126
Disability with Positive affect ^a Disability with Negative affect $\chi^2 = 0.078(1)$, $p \leq 0.780$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.005	Pain catastrophizing	Disability	0.122	0.029	4.185***	0.125
	Pain avoidance goal	Activity avoidance	0.554	0.106	5.255***	0.157
	Pain catastrophizing	Activity avoidance	0.054	0.011	4.812***	
Fibromyalgia impact with Positive affect $\chi^2 = 0.116(1)$, $p \leq 0.734$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.005	Activity avoidance	Disability	0.496	0.157	3.151**	
	Positive affect	Fibromyalgia impact	0.474	0.080	5.888***	0.251
	Pain avoidance goal	Activity avoidance	0.554	0.106	5.255***	0.157
	Positive affect	Activity avoidance	0.054	0.011	4.812***	
Fibromyalgia impact with Negative affect $\chi^2 = 0.509(2)$, $p \leq 0.775$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.009	Activity avoidance	Fibromyalgia impact	2.106	0.408	5.156***	
	Pain catastrophizing	Fibromyalgia impact	0.314	0.095	3.295**	0.290
	Negative affect	Fibromyalgia impact	0.406	0.126	3.233**	
	Pain avoidance goal	Activity avoidance	0.554	0.106	5.245***	0.157
	Pain catastrophizing	Activity avoidance	0.054	0.011	4.811***	
	Activity avoidance	Fibromyalgia impact	2.231	0.404	5.517***	

CFI, Comparative Fit Index; RMSEA, Root Mean Square Error of Approximation; SE, Standard Error; SRMR, Standardized Root Mean Square Residual; ^asame model were obtained with positive and negative affect; ** $p \leq 0.01$, *** $p \leq 0.001$.

TABLE 5 | Fitted models with test statistics and path coefficients: goal preferences and affect mediated by persistence patterns.

Model and fit	Predictor	Dependent variable	B	SE	z	Effect size
Persistence patterns						
Pain with Positive affect $\chi^2 = 1.369(1)$, $p \leq 0.249$ CFI = 0.996; RMSEA = 0.039; SRMR = 0.027	Positive affect	Task-contingent persistence	0.039	0.020	1.991*	0.014
	Task-contingent persistence	Pain	-0.074	0.039	-1.894 ⁺	0.032
	Excessive persistence	Pain	0.099	0.040	2.490*	
	Pain	Positive affect	-0.945	0.369	-2.559*	0.037
Pain with Negative affect $\chi^2 = 0.336(1)$, $p \leq 0.562$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.010	Negative affect	Pain	0.035	0.011	3.182**	0.219
	Negative affect	Task-contingent persistence	0.052	0.018	2.912**	
	Pain avoidance goal	Task-contingent persistence	-0.861	0.121	-7.130***	
	Task-contingent persistence	Pain	-0.063	0.031	-2.016*	0.053
Disability with Positive affect $\chi^2 = 0.000(0)$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.000	Excessive persistence	Disability	0.588	0.139	4.233***	0.122
	Positive affect	Disability	-2.03	0.052	-3.910***	
	Pain avoidance goal	Disability	0.571	0.279	2.046*	
	Negative affect	Disability	0.151	0.046	3.273**	0.139
Disability with Negative affect $\chi^2 = 0.305(1)$, $p \leq 0.581$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.007	Negative affect	Task-contingent persistence	0.052	0.018	2.912**	0.219
	Pain avoidance goal	Task-contingent persistence	-0.861	0.121	-7.130***	
	Negative affect	Excessive persistence	0.121	0.013	7.675***	
	Pain avoidance goal	Excessive persistence	-0.290	0.125	-2.264*	
Fibromyalgia impact with Positive affect $\chi^2 = 1.705(2)$, $p \leq 0.426$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.023	Task-contingent persistence	Disability	-0.591	0.145	-4.086***	
	Excessive persistence	Disability	0.579	0.179	3.229**	0.216
	Positive affect	Fibromyalgia impact	-0.698	0.132	-5.274***	0.202
	Pain avoidance goal	Excessive persistence	-0.451	0.132	-3.402**	0.045
	Pain avoidance goal	Task-contingent persistence	-0.952	0.114	-8.321***	0.215
Fibromyalgia impact with Negative affect $\chi^2 = 0.000(0)$ CFI = 1.000; RMSEA = 0.000; SRMR = 0.000	Positive affect	Task-contingent persistence	0.055	0.019	2.972**	
	Task-contingent persistence	Fibromyalgia impact	-1.332	0.402	-3.314**	
	Excessive persistence	Fibromyalgia impact	2.235	0.407	5.495***	
	Negative affect	Fibromyalgia impact	0.654	0.111	5.866***	0.124

CFI, Comparative Fit Index; RMSEA, Root Mean Square Error of Approximation; SE, Standard Error; SRMR, Standardized Root Mean Square Residual; ⁺ $p = 0.058$, * $p \leq 0.05$, ** $p \leq 0.01$, *** $p \leq 0.001$.

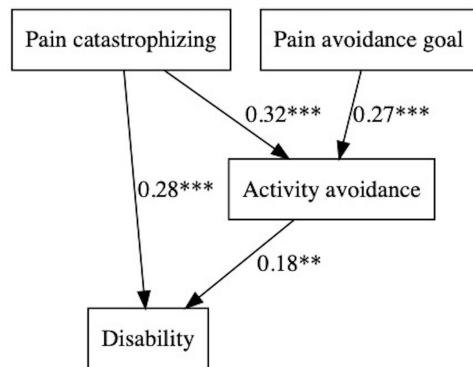


FIGURE 3 | Pain avoidance goal, affect, and activity patterns on disability.

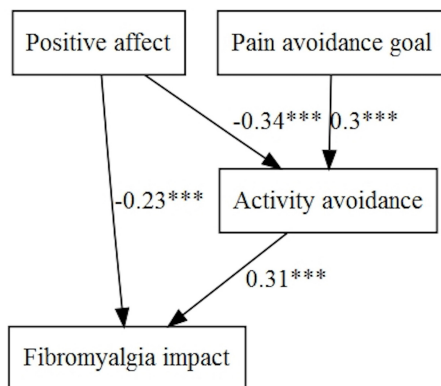


FIGURE 4 | Pain avoidance goal, positive affect, and activity patterns on fibromyalgia impact.

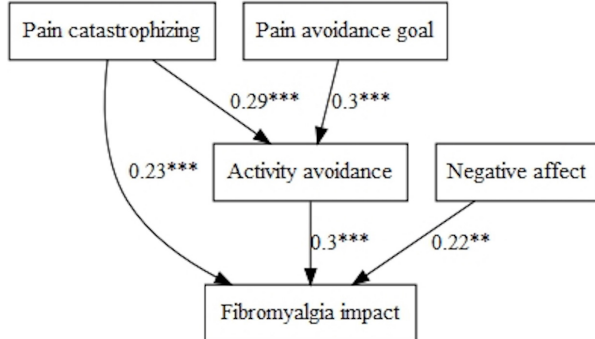


FIGURE 5 | Pain avoidance goal, negative affect, and activity patterns on fibromyalgia impact.

path on pain intensity through task-contingent persistence. Moreover, negative affect showed a positive direct effect on pain.

When we tested the models with disability, we only found direct effects of 'Pain-avoidance goal,' positive affect and excessive persistence in a simple multivariate model

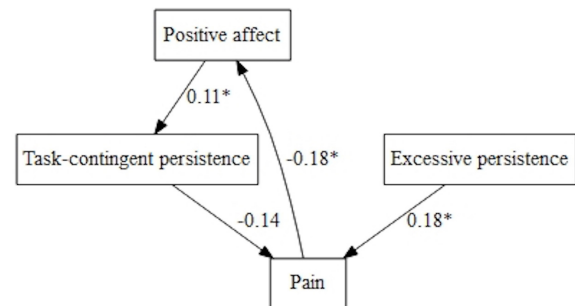


FIGURE 6 | Pain avoidance goal, positive affect, and persistence patterns on pain.

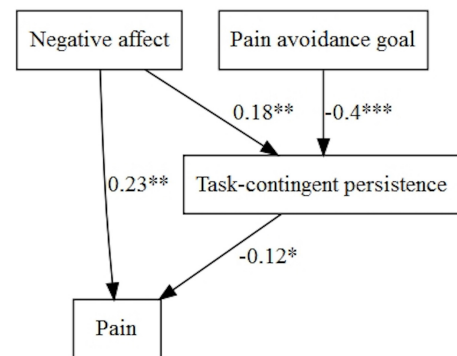


FIGURE 7 | Pain avoidance goal, negative affect, and persistence patterns on pain.

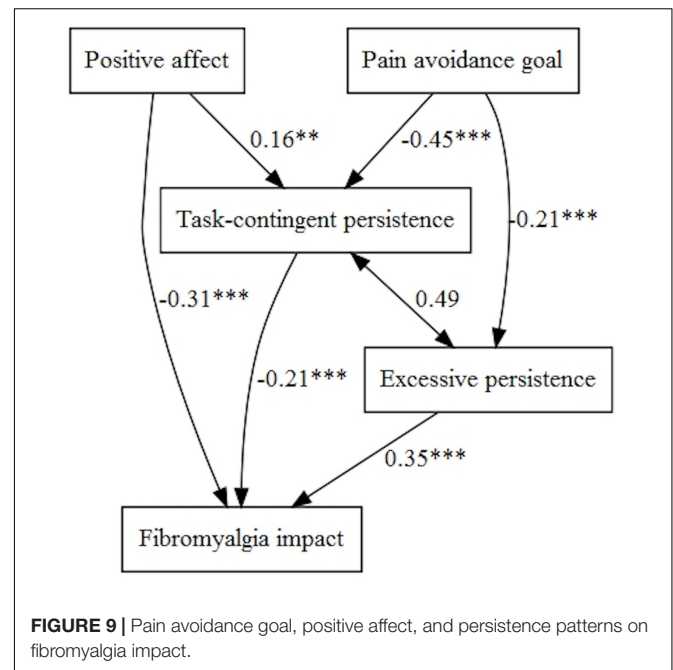
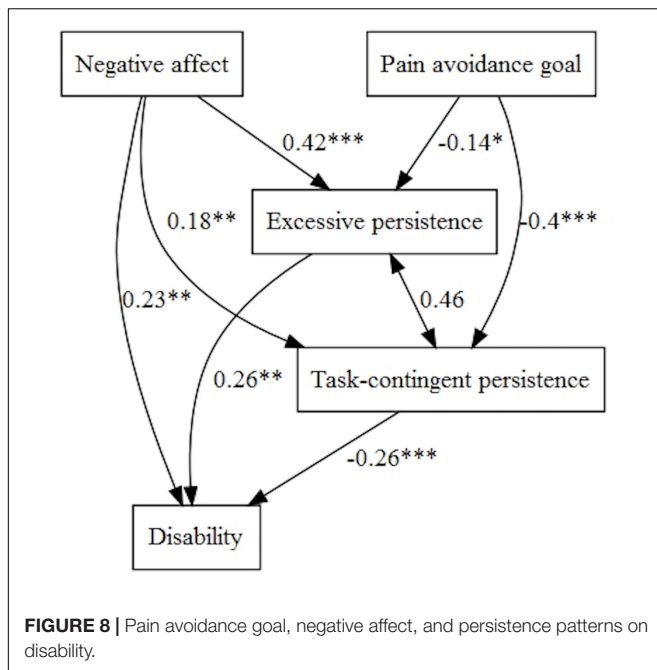
(Table 5). However, when the model was tested with negative affect, this variable (with positive sign) and 'Pain-avoidance goal' (with negative sign) influenced disability directly and indirectly, through task-contingent persistence and excessive persistence (Figure 8). Task-contingent persistence and excessive persistence were correlated ($B = 2.47, p < 0.001$).

Fibromyalgia impact was influenced directly by positive affect (with negative sign) and indirectly through task-contingent persistence. Moreover, 'Pain-avoidance goal' influenced indirectly through task-contingent persistence and excessive persistence (negatively) fibromyalgia impact (Figure 9).

When we tested the model with negative affect, this was the only variable that predicted fibromyalgia impact, which resulted in a simple univariate model (Table 5).

DISCUSSION

This research explored the relationships between goal preferences (preference for hedonic goals in contrast with achievement goals), affect (positive and negative), activity patterns (avoidance and persistence) and health outcomes in fibromyalgia. We took into account the predictions from the MAI model, but were aware of the more stable context represented by the GPQ and the affect measures. As a first step toward this aim, we adapted the



GPQ to a Spanish population of women with fibromyalgia. The culturally adapted Spanish version resulted in a shorter version with changes in several activities made after the field study conducted with the target population. The main consequence of these changes was more representation of situations related to household tasks than in the original GPQ. Although the original and the final back-translated version did not show a high level of coincidence, we underlined the cultural and experiential equivalence to ensure a comprehensible translation and to maintain the concept while also adapting to the cultural target context (López-Roig and Pastor, 2016). The internal structure of the Spanish version reproduced the original GPQ. The two subscales ('Pain-avoidance goal' and 'Mood-management goal') showed high reliability and adequate construct validity in our sample. However, we did not obtain significant relationships with pain catastrophizing. As Karsdorp and Vlaeyen (2011) pointed out, the absence of significant relationships confirm they are different constructs and may be an effect of their different conceptualizations. GPQ compares the relative strength of preference for avoidance goals against achievement goals in a motivational context related to different specific situations. On the contrary, catastrophism is measured in a general context, with no motivational context, and without related situations where goals can compete. In addition, we should bear in mind that the total score of catastrophism includes three different dimensions (magnification, rumination and helplessness) and it is possible they do not have the same relationships with goal preferences, and therefore limit the total correlation score. However, this is not an aim of the present study.

In fibromyalgia, the main effects of goal preferences, affect and activity patterns on disability and adjustment, according to relevant psychological models on chronic pain, have been explored in previous research (Vlaeyen and Morley, 2009).

However, our study explores these constructs in a more complex framework, taking into account the hypotheses of authors who have applied the MAI model to chronic pain (Vlaeyen and Morley, 2004, 2009; Karsdorp et al., 2010; Karsdorp and Vlaeyen, 2011). As a novel contribution, we have studied the mediation of avoidance and persistence activity patterns in the relationships of the goal preferences and affect with health outcomes. Our results showed no interaction effect of affect and goal preferences on activity patterns. Women with fibromyalgia did not use their positive or negative affect as an informational source for task performance, which supports previous results with mood in experimental studies among people with work-related upper extremity pain (Karsdorp et al., 2010) and among people without pain (Ceulemans et al., 2013; Karsdorp et al., 2013). Similarly, we found activity patterns were explained independently by motivational (preference for pain-avoidance goals) and affective (positive and negative affect) factors. In clinical populations with severe and longstanding pain such as fibromyalgia, mood (affect in this study) can be attributed to pain experience and does not have the informational role hypothesized by the MAI model when mood is attributed to the task. This fact, referred to as 'the discounting hypothesis,' suggests that it is possible that there is no interaction between mood and stop-rules (goals) when people attribute their mood to an obvious source (Meeten and Davey, 2011), such as the chronic pain experience in our case.

Results regarding the mediational analyses with the two *avoidance patterns* showed only activity avoidance, in other words avoidance related to the chronic pain condition itself (Kindermans et al., 2011; Esteve et al., 2016), was relevant. In this sense, our findings support the ample evidence available of the fear-avoidance model (Vlaeyen and Linton, 2000, 2012; Leeuw et al., 2007). Catastrophizing thoughts and preference for pain avoidance goals showed a direct and indirect path,

increasing activity avoidance, and disability and fibromyalgia impact perception, in line with previous research with chronic musculoskeletal pain and added evidence to the direct link of activity avoidance with disability and fibromyalgia impact perception (Andrews et al., 2012; Esteve et al., 2016, 2017). It is noteworthy that in these models, positive and negative affect did not show any significant path on disability. However, affect played a different role in the general impact of fibromyalgia. Positive affect was related to less activity avoidance and less fibromyalgia impact, and negative affect showed only a direct effect which increased the patients' perception of fibromyalgia impact.

Finally, no tested model with avoidance patterns was significant for pain intensity. Pain intensity was explained by *persistence patterns*. In the context of a long-term chronic condition (participants had experienced more than 10 years of pain and attended health care tertiary level), the pain is probably integrated in daily experience and persistence would be more relevant as a way of functioning. Affect (positive and negative) and strong achievement goals relative to pain-avoidance goal preferences influenced pain intensity through more endorsement on task-contingent persistence, which was associated with less pain.

Similar to the findings with avoidance patterns, models with persistence were slightly different with positive or negative affect. Only in the model with negative affect, were goal preferences relevant in pain intensity. A strong achievement goal relative to a pain avoidance goal and negative affect increased task-contingent persistence. Negative affect also was directly related to more pain intensity. Regarding disability, the more complex model was obtained with negative affect. This variable and strong endorsement of an achievement goal relative to a pain avoidance goal increased both excessive and task contingent persistence, and these activity patterns were related to more and less disability, respectively. Finally, negative affect also showed a direct path increasing disability. For fibromyalgia impact, the more complex model was obtained with positive affect and, interestingly, showed similar significant paths to negative affect on disability. In these models, task-contingent persistence and excessive persistence predicted better and poorer outcomes respectively in line with previous research (Kindermans et al., 2011; Andrews et al., 2012; Esteve et al., 2016, 2017). These findings provide added evidence of the double adaptive or maladaptive role of persistence on chronic pain outcomes, depending on the kind of persistence and the underlying goals (Van Damme and Kindermans, 2015). In addition, they partially support the avoidance-endurance model of chronic pain (Hasenbring and Verbunt, 2010; Hasenbring and Kindermans, 2018) taking into account the role of negative affect on persistence activity. The role of positive affect as risk factor for overuse, as the model hypothesized, was not supported by our findings. In fact, positive affect appeared as an asset encouraging less avoidance activity and more task-contingent persistence.

Unexpectedly, negative affect increased task-contingent persistence, in contrast with previous research, which found a significant negative relationship between negative affect and this activity pattern (Esteve et al., 2016, 2017). Our result could be explained by the high positive correlation between

task-contingent and persistence activity subscales in our sample. In this sense, we must point out that we employed the original factors of the activity patterns scale, developed with heterogeneous musculoskeletal chronic pain patients (Esteve et al., 2016). The above-mentioned significant correlation alongside the low internal consistency of the excessive persistence subscale, may suggest another internal structure of this scale in a unique sample of women with fibromyalgia. The overuse activity pattern characteristic of some groups of patients with fibromyalgia might make the differentiation of the type of persistence for these patients more difficult. However, a positive significant correlation between negative affect and excessive persistence has been previously reported by the same authors, explained as a way of managing affective discomfort involving in excessive activity (Esteve et al., 2016, 2017). This hypothesis could also be true in fibromyalgia, mainly when these patients usually reported high levels of negative emotions and also of persistence (Vlaeyen and Morley, 2004; Hassett et al., 2008; Van Middendorp et al., 2008, 2010). Finally, the direct paths of positive and negative affect with disability and fibromyalgia impact supported previous research on their beneficial and detrimental role respectively in fibromyalgia adaptation (Van Middendorp et al., 2008; Estévez-López et al., 2015, 2017).

This study has some limitations we should bear in mind. First, we conducted a cross-sectional design with correlational data, which does not allow us to establish causal relationships. Second, all measures were self-reported measures. However, the study represents a first view of the motivational and affective determinants of different activity patterns and health outcomes in fibromyalgia, which should be replicated in prospective studies including also objective measures of activity using accelerometers. Third, as we have mentioned, the activity avoidance and excessive persistence subscales of the Activity Patterns Questionnaire (Esteve et al., 2016) showed low internal consistency in our sample. Future studies should perform a replication of the factor structure of this questionnaire in fibromyalgia. Fourth, the sample in the first study was modest; however, in the context of this phase of cultural adaptation of an instrument, a qualitatively representative sample of the target population is essential (Matsumoto and Van De Vijver, 2011). We can find a large variability in sample sizes, for instance, $n = 5$ (Le Gal et al., 2010; Two et al., 2010) or $n = 14$ (Sánchez-Pérez et al., 2017). In addition, we did not check the final translated version with the original authors in order to contrast the right render of the construct; however, we did take into account the participants' proposals in looking for experiential equivalence in the changed situations of the original questionnaire. Finally, we should point out that as in Karsdorp and Vlaeyen (2011), the effect size of our results was low, possibly due to the complexity of the target.

Despite these limitations, our findings may help to understand motivational and affective issues underlying avoidance and persistence activity in fibromyalgia. In other words, preferences for maintaining a positive mood relative to an achievement goal ('Mood-management goal') did not show any role in activity patterns or fibromyalgia health outcomes, in line with the results of Karsdorp and Vlaeyen (2011) with other pain problems. However, strong endorsement of pain avoidance goals

relative to achievement goals ('Pain-avoidance goal') increased activity avoidance. On the contrary, strong endorsement of achievement goals relative to pain avoidance encouraged both task-contingent persistence and overuse, which showed opposite effects on disability and fibromyalgia impact. Regarding affective issues, positive affect showed significant paths in models with avoidance and persistence patterns. In general terms, positive affect behaved as an asset and a protective factor due to its direct and indirect paths with health outcomes. Women who scored higher on it showed less activity avoidance and more task-contingent persistence and less pain and fibromyalgia impact. Women who scored higher on negative affect showed more task-contingent persistence, which was associated with less pain and disability, but also more excessive persistence or overuse, which was associated with more disability. In addition, negative affect showed direct positive paths to pain and disability, which is also coherent with previous research (Van Middendorp et al., 2008, 2010; Estévez-López et al., 2015, 2017).

Our results did not support the interaction hypothesis of Karsdorp and Vlaeyen (2011). Nevertheless, we believe the 'Pain-avoidance goal' subscale can be useful for a self-regulation perspective in fibromyalgia. This scale could be used as a single scale due to its good psychometric properties and its results with avoidance and persistence activity patterns. As has been mentioned, this scale contrasted preferences for pain avoidance goals relative to achievement goals in eight common daily situations for women with fibromyalgia. Therefore, the scale included in the same context two common goals in pain patients: to reduce pain immediately or to persist in the ongoing task despite pain (Hasenbring and Kindermans, 2018). Both goal preferences were relevant in the avoidance and persistence activities of our participants. Therefore, the pain avoidance-achievement goal "bipolarity" of this scale could be useful in applying the self-regulation perspective in chronic pain.

In summary, this study has shown the relevance of pain avoidance and achievement goal preferences in the same context. These preferences always impacted health outcomes through activity patterns, encouraging activity avoidance (when patients endorsed avoid pain relative to achievement goals) and excessive persistence and task-contingent persistence (when patients endorsed achievement goals relative to pain avoidance goals). Positive and negative affect showed direct and indirect effects on health outcomes. Our results supported the mediational role of activity patterns between goal preferences, affect, and health outcomes, and did not support the moderation of affect in these relationships. These findings allow us to improve the understanding of the complex relationships between goal pursuit, vulnerability (catastrophizing and negative affect), psychological

assets (positive affect), activity patterns and health outcomes in fibromyalgia. In this sense, reinforcing achievement goals relative to pain avoidance (in a flexible way), and positive affect to promote task-persistence adaptive activity and decreased activity avoidance may prove to be suitable targets in interventions to improve chronic pain adaptation.

DATA AVAILABILITY

The raw data supporting the conclusions of this manuscript will be made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethic committee of Miguel Hernández University and Ethic committee of Alicante General Hospital. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

M-AP-M, SL-R, and CP developed the design of the research. All authors participated in the Spanish adaptation process of the GPQ. M-AP-M, SL-R, and FM-Z performed the data analyses. M-AP-M, SL-R, and FM-Z wrote the first draft. All authors contributed and reviewed the final draft. All authors approved the manuscript.

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Multisystem Resiliency as a Predictor of Physical and Psychological Functioning in Older Adults With Chronic Low Back Pain

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Evidence supports the benefits of resilience among older adults with chronic pain. While numerous factors confer resilience, research has largely examined these measures in isolation, despite evidence of their synergistic effects. Conceptualizing resilience from a multisystem perspective may provide a deeper understanding of adaptive functioning in pain. Sixty adults (ages 60+ years) with chronic low back pain completed measures of physical function, pain intensity, disability, and a performance-based task assessing back-related physical functioning and movement-evoked pain (MEP). Depressive symptoms, quality of life, and general resilience were also evaluated. To examine multisystem resiliency, principal components analysis (PCA) was conducted to create composite domains for psychological (positive affect, hope, positive well-being, optimism), health (waist-hip ratio, body mass index, medical comorbidities), and social (emotional, instrumental, informational support) functioning measures, followed by cluster analysis to identify participant subgroups based upon composites. Results yielded four clusters: Cluster 1 (high levels of functioning across psychological, health, and social support domains); Cluster 2 (optimal health and low psychosocial functioning); Cluster 3 (high psychological function, moderate-to-high social support, and poorer health); and Cluster 4 (low levels of functioning across the three domains). Controlling for sociodemographic characteristics, individuals with a more resilient phenotype (Cluster 1) exhibited lower levels of disability, higher quality of life and psychological functioning, and greater functional performance when compared to those with a lower degree of personal resources (Cluster 4). No significant cluster differences emerged in self-reported pain intensity or MEP. These findings signify the presence of resiliency profiles based upon psychological, social, and health-related functioning. Further examination of the additive effects of multiple adaptive behaviors and resources may improve our understanding of resilience in the context of pain, informing novel interventions for older adults.

Keywords: resilience, multisystem, low back pain, aging, psychological, health, social support

INTRODUCTION

Older adults represent the fastest growing population in the United States. As such, increased attention on enhancing the health and well-being of this cohort is imperative. Among health complaints and chronic medical conditions, pain remains a significant area of concern in aging adults, with approximately 18.7 million (53%) adults ages 65 years and older (Patel et al., 2013) reporting they experience bothersome pain (Helme and Gibson, 2001). Further, chronic low back pain (cLBP) impacts 36% of this population and is the leading cause of disability in older adults (Weiner et al., 2003; Molton and Terrill, 2014). In spite of the significant burden of chronic pain in older adults, this group is often subjected to inadequate assessment and suboptimal treatment of pain (Gibson and Lussier, 2012; Molton and Terrill, 2014).

Traditionally, aging has been viewed as a period of frailty, vulnerability, and decline. However, there is considerable variability in the aging process. Indeed, the importance of considering the role of adaptive constructs in promoting successful aging (characterized by decreased disability, greater health-related functioning, and better life engagement) has been highlighted (Rowe and Kahn, 1997, 2015). Understanding factors that could delay or prevent aging-related illnesses and support successful aging would allow for the development of approaches that attenuate disability related to these health conditions. Thus, in the context of functional limitations and decreased quality of life associated with chronic pain, greater emphasis should be placed on identifying factors that ultimately inform targeted interventions for pain in older adults. These investigations should account for the multidimensional nature of pain and the myriad biopsychosocial elements that influence it.

Diminished functioning (e.g., physical disability and work-related interference) and psychosocial interference (e.g., depressed mood, anxiety, pain-related fear, and limited social support) that often accompany chronic pain play a role in disrupted quality of life in individuals with pain. To date, existing research has primarily focused on risk and vulnerability factors related to the maintenance and exacerbation of pain. For example, negative psychological factors (e.g., negative affective states) have consistently been shown to facilitate pain and disability; depression and anxiety are highly comorbid with chronic pain and can significantly impact the pain experience, leading to greater pain severity, impaired functioning, and reduced quality of life (Bair et al., 2003; Lerman et al., 2015). In fact, evidence suggests that in older adults, depression can uniquely contribute to increased risk of developing disabling back pain (Reid et al., 2003). Similarly, reciprocal relationships between symptoms of anxiety and depression and greater pain interference have been demonstrated in the aging population (Arola et al., 2010).

Additionally, negative pain beliefs (e.g., pain catastrophizing and fear-avoidance) are known to adversely influence pain-related outcomes. Consistent evidence suggests that pain catastrophizing (pain-associated rumination, magnification, and helplessness) leads to enhanced pain and greater affective disturbance (Turner and Clancy, 1986; Sullivan et al., 2001).

Likewise, individuals may develop a fear of pain and movement that facilitates avoidance of certain activities following a painful injury, when they view these activities as having the potential to cause re-injury and subsequent pain (Vlaeyen and Crombez, 1999; Vlaeyen and Linton, 2000; Crombez et al., 2012). These fear-avoidance beliefs can increase pain and functional impairment, such as physical deconditioning arising from limited mobility (Rainville et al., 2011; Wertli et al., 2014). Although informative, an emphasis on pathology/vulnerability does not capture the impact of additional contributors on the pain experience, including the potentially protective role of positive, adaptive factors on chronic pain.

While aging has been regarded as a period of loss, this view has been contrasted by mounting evidence that older adults have the capacity for resilience; evidenced by high levels of reported well-being, quality of life, and self-rated successful aging, despite worsening health and substantive physical challenges (i.e., pain) (MacLeod et al., 2016). Although there are competing approaches to the conceptualization and measurement of resilience, it has largely been characterized as a trajectory of positive adaptation in response to significant risk or adversity (Ong et al., 2009). Resilience has also been delineated as a trait-like construct, consisting of personality characteristics and stable psychosocial factors that contribute to adaptive functioning; however, it is argued that this definition lacks precision as it overlooks time-varying and contextually dependent aspects of resilient responding. Further, characterization of resilience as purely dispositional fails to account for the malleability of human functioning or the consideration of how resilience can be promoted through therapeutic intervention. More recent theoretical models have conceptualized resilience as a dynamic process, characterized as an interplay between trait-based resources (e.g., personality factors) and active mechanisms (e.g., cognitive and affective states) that influence adaptive coping responses to pain. This process, in turn, promotes sustainability in meaningful and valued activities, personal growth as a result of one's experience with chronic pain, and the capacity to recover or rebound from disruptions in physiological, emotional, or cognitive functioning (e.g., pain flare-up) (Sturgeon and Zautra, 2010, 2013).

Abundant literature has identified multiple psychological contributors to resilience. For example, optimism (Ferreira and Sherman, 2007; Goodin and Bulls, 2013; Goodin et al., 2013; Cousins et al., 2014), hope (Berg et al., 2008; Howell et al., 2015; Bartley et al., 2019b), positive affect (PA) (Zautra et al., 2005; Finan and Garland, 2015; Hassett and Finan, 2016), self-efficacy (Wright et al., 2008; Wylde et al., 2012; Brembo et al., 2017; Martinez-Calderon et al., 2018; Karasawa et al., 2019), and pain acceptance (McCracken, 1998; Kratz et al., 2007; Jensen et al., 2016) have been associated with adaptive changes across a number of pain and mental health outcomes. Perceived social support also shows benefits in individuals with chronic pain, which may be particularly relevant for older adults as social engagement provides a means of coping with pain (Molton and Terrill, 2014). In fact, perceptions of support are associated with fewer depressive symptoms (Ferreira and Sherman, 2007; Lopez-Martinez et al., 2008; Lee et al., 2016; McKillop et al., 2017),

greater quality of life (Ethgen et al., 2004), lower pain intensity (Lopez-Martinez et al., 2008), and improvements in postsurgical (i.e., lower-limb amputation) functioning (Hanley et al., 2004).

Together with psychological and social functioning, numerous lifestyle and health factors also contribute to resilience. Tobacco usage is associated with a greater incidence and prevalence of pain (Goldberg et al., 2000; Shiri et al., 2010b), while multimorbidity has profound consequences on the occurrence (Schneider et al., 2007) and worsening of pain and physical functioning (Calders and Van Ginckel, 2018). Similarly, sleep has been posited as a key regulator of pain modulation, with effects on somatosensory sensitivity (Campbell et al., 2015; Schrimpf et al., 2015), pain severity (Gerhart et al., 2017), and interference (Kothari et al., 2015). Although sleep and pain are temporally related, sleep quality appears to have a more robust influence on pain symptomatology than vice versa (Finan et al., 2013; Gerhart et al., 2017), and may even serve as a risk factor for pain development and chronification (Gupta et al., 2007; Finan et al., 2013). Likewise, intervening on sleep may have salutary effects on pain, with recent evidence highlighting the influence that treatment-related sleep improvements have on pain intensity (de la Vega et al., 2019). Exercise as a therapeutic modality also confers many health benefits but can be especially potent for pain symptomatology. Increasing evidence suggests that sedentary behavior is inversely associated with functional performance (Lee et al., 2015), with greater physical activity predicting more optimal long-term outcomes in pain and disability (Pinto et al., 2014). Also, acute bouts of exercise yield analgesic effects on pain-evoked laboratory measures (Burrows et al., 2014), yet appear to be differentially influenced by physical activity behavior (e.g., sedentarism and level of physical activity) (Naugle et al., 2017; Ohlman et al., 2018). In turn, sedentary behavior may promote greater adiposity (i.e., body mass index and waist-hip ratio) which can be a risk factor for pain and functional disability (Fanuele et al., 2002; Shiri et al., 2010a; Walsh et al., 2018), presumably through a myriad of pathways such as increased joint loading, biochemical mediators, and mood disturbance (Okifuji and Hare, 2015). The association between pain and obesity is likely reciprocal, however, with chronic pain also potentiating risk for weight gain. Given that obesity is a potentially modifiable factor, some studies have highlighted the efficacy of weight loss interventions in reducing the incidence and severity of pain (Hughes et al., 2018; Dunlevy et al., 2019).

Taken together, there is a wealth of literature supporting the protective effects of psychological, social, and lifestyle factors in the experience of pain. However, much of research has examined these factors in isolation, with limited consideration of their additive contributions. Even more, while existing conceptualizations of resilience have varied widely, it has commonly been defined as a trait-based construct comprised primarily of psychological facets (Windle et al., 2011). Thus, prevailing approaches to the study of resilience may not fully capture the multidimensionality of the construct or how resilient functioning can be promoted through various systems. Extending our current conceptual models may carry important implications in terms of explicating the resources and mechanisms that promote adaptive pain outcomes. Only

a modest literature has addressed the notion of multisystem resiliency. For instance, Agrigoroaei and Lachman (2011) found that a protective composite of psychosocial and behavioral factors (i.e., control beliefs, social support quality, and physical activity) predicted cognitive functioning, above and beyond the effects of sociodemographics, physical health, and cognitive activity engagement. Further, the combination of low-risk lifestyle factors (i.e., smoking, physical activity, adiposity, alcohol use, and diet) was more robustly associated with longer leukocyte telomere length (a marker of cellular aging) in women, as compared to the independent effects of each factor (Sun et al., 2012). Similarly, Johnson et al. (2019) found that a psychosocial and behavioral index of resilience [i.e., optimism, PA, negative affect (NA), active coping, perceived stress, social support, tobacco use, and waist-hip ratio] had a stronger association with telomere length in older adults with knee pain, relative to a composite comprised solely of psychological functioning measures. Overall, these findings provide compelling support for an integrative approach to studying resilience and underscore the importance of exploring these contributions in chronic pain.

The current study sought to address this gap in the literature by examining the association of multisystem resiliency with pain and psychological outcomes in a sample of older adults with cLBP. Given the dimensionality of resilience, several psychosocial resources (i.e., PA, hope, positive well-being, optimism, and social support) and health/lifestyle variables (i.e., waist-hip ratio, body mass index, physical health comorbidities, and smoking status) were considered for inclusion. These measures were selected as they represent modifiable factors with strong, empirical support for their impact on pain and health-related processes. Therefore, the primary aims were to: (1) empirically identify domains of resilience based upon psychological, social, and health-related factors and (2) using cluster analysis, explore whether resiliency phenotypes differ across measures of physical function, pain intensity, disability, and psychological functioning. It was hypothesized that: (1) homogenous subgroups would emerge from patterns of psychological, health, and social resiliency and (2) individuals with more resilient phenotypes (i.e., higher in protective resources) would exhibit higher physical function, lower self-reported pain and disability, and greater psychological functioning.

MATERIALS AND METHODS

Participants and Procedures

This was a cross-sectional study based on a secondary data analysis from the Adaptability and Resilience in Aging Adults (ARIAA) study, a project evaluating the effects of resilience mechanisms on pain modulatory capacity among individuals with cLBP. Sample size estimations were based upon previous pilot data (Bartley et al., 2019b) establishing that 60 participants would provide power of 0.80 at 0.05 (two-tailed) for detecting moderate to large effect sizes between measures of resilience and pain.

Older adults (ages 60+ years) with cLBP ($N = 69$) were recruited from the community via posted fliers, media

announcements, and word-of-mouth referral. All participants provided verbal and written informed consent. Participants were included if they reported at least mild LBP ($\geq 2/10$) occurring on at minimum half of the days during the preceding 3 months. Enrollment in the study was not limited to LBP (due to the presence of medical comorbidities in this population) as long as LBP was an individual's primary pain condition. Exclusion criteria were as follows: recent vertebral fracture; back surgery within the past 6 months; diagnosis of cauda equina syndrome; uncontrolled hypertension ($\geq 150/90$); severe cardiovascular disease (e.g., recent heart attack); neurological disease associated with somatosensory abnormalities (e.g., neuropathy, seizures, and Parkinson's disease); current major medical illness (e.g., metastatic or visceral disease); chronic opioid use; and systemic inflammatory disease (e.g., spondylarthropathies such as ankylosing spondylitis and systemic lupus erythematosus). Participants were provided up to \$100 compensation upon completion of the study.

The University of Florida Institutional Review Board approved all study procedures. Initially, participants were evaluated for study inclusion and exclusion through a brief telephone screen. The following sociodemographic and health data were obtained as part of the screening: self-reported sex, age, and a brief health history including the presence of major medical illnesses, recent back-related injuries or surgeries, and LBP symptoms. If eligible, participants attended two, 2–3.5-h appointments scheduled approximately 1 week apart. During Session 1, eligibility criteria were verified through a self-reported demographic and medical history assessment, and participants completed anthropometric tests, psychosocial questionnaires, and functional performance measures. During the time in between Sessions 1 and 2, participants completed several questionnaires at home. Sensory pain testing was conducted during Session 2 (data not reported), and additional psychosocial questionnaires were also completed at that visit.

Measures

Predictors of Multisystem Resilience

Positive and negative affect schedule

The 20-item Positive and Negative Affect Schedule (PANAS) was used to examine PA and NA (Watson et al., 1988). Respondents were presented with 10 positively valenced and 10 negatively valenced terms that are rated on a five-point scale ranging from 1 (very slightly or not at all) to 5 (extremely) resulting in scale scores for PA and NA, with higher scores indicating increased positive and NA, respectively (only PA scores were included in the current analysis). Reliability tests indicated high internal consistency of items on the PA scale ($\alpha = 0.90$).

Adult dispositional hope scale

The Adult Dispositional Hope Scale (ADHS) is a 12-item questionnaire that includes eight statements measuring two aspects of hope: pathways (e.g., "There are lots of ways around a problem.") and agency (e.g., "I energetically pursue my goals."), as well as four "filler" statements that are not included in scoring (Snyder et al., 1991). Items are rated on a scale ranging from 1 (definitely false) to 8 (definitely true) and respondents

select the number that best describes them for each statement. Higher scores indicate greater trait levels of hope. Reliability analyses from the current investigation revealed Cronbach's α for the ADHS = 0.92, indicating high internal consistency for this measure.

PROMIS positive affect and well-being scale

The Patient-Reported Outcomes Measurement Information System (PROMIS) PA and Well-Being Scale was used to measure PA and overall sense of satisfaction with life (Salsman et al., 2013). This scale consists of 23 items rated on a 1 (never) to 5 (always) scale to indicate how often respondents experienced positive emotion and/or purpose/meaning in life (e.g., "[Lately], I had a sense of balance in my life."). Higher scores reflect greater PA and well-being (Cronbach's $\alpha = 0.97$).

Life-orientation test-revised

Dispositional optimism was evaluated using the Life-Orientation Test-Revised (LOT-R), which consists of 10 items (including four unscored items and three reverse-scored items). Participants were asked to use a 5-point scale ranging from 0 (strongly disagree) to 4 (strongly agree) and rate the degree to which they agreed with the presented statements (e.g., "In uncertain times, I usually expect the best.") (Herzberg et al., 2006). Higher LOT-R scores indicate greater optimism. This measure demonstrated adequate reliability in the sample ($\alpha = 0.73$).

PROMIS support (emotional, instrumental, informational)

To measure social functioning, the short forms of the PROMIS emotional (eight items; e.g., "I have someone who makes me feel appreciated."), instrumental (four items; e.g., "Do you have someone to take you to the doctor if you need it?"), and informational (four items; e.g., "I have someone to turn to for suggestions about how to deal with a problem.") support scales were administered (Hahn et al., 2014). Items are rated on a 1 (Never) to 5 (Always) scale for all three domains, with higher scores indicating greater social support. All three scales were found to have high internal consistency and were also highly reliable with each other: emotional ($\alpha = 0.97$), instrumental ($\alpha = 0.96$), informational ($\alpha = 0.96$), all support measures combined ($\alpha = 0.97$).

Anthropometric tests: body composition

During Session 1, participants' waist (5 cm above the navel) and hip circumferences (widest part of the hips) were calculated (in cm) using a measuring tape, with waist-hip ratio determined by dividing the waist circumference by the hip circumference. Body weight was measured to the nearest 0.1 kg using a digital scale (Healthometer) and height was assessed to the nearest centimeter using a wall stadiometer. Calculation of BMI was determined by weight in kilograms divided by height in meters squared.

Health comorbidities

To determine the presence of physical health comorbidities, participants completed a health status questionnaire whereby they were asked to place an "X" next to any current medical conditions (i.e., high blood pressure, heart disease, diabetes, asthma/breathing problems, kidney/renal disease, thyroid problem, neurological disorder, or other self-reported

health conditions). Medical diagnoses were placed into ICD-10 diagnostic categories for reporting purposes.

Smoking status

Current cigarette smoking status was assessed using the following question: “How would you describe your cigarette smoking?” Possible responses included: “never smoked,” “used to smoke but have now quit,” and “current smoker,” and individuals were categorized as either current smokers (yes) or non-smokers (no).

Study Outcomes

Back performance scale

Functional performance and movement-evoked pain (MEP) were measured using the Back Performance Scale (BPS). The BPS consists of a series of tasks (i.e., Sock Test, Pick-up Test, Roll-up Test, Fingertip-to-Floor Test, and Lift Test) that are designed to measure functional capacity during completion of mobility-oriented activities that have been deemed to be particularly difficult for individuals with back pain (Magnussen et al., 2004; Strand, 2017). An evaluator assesses the degree to which these tasks are completed. Physical functioning scores range from 0 to 3 for each test (total scale score = 0–15), with increasing scores indicating greater difficulty with task performance. MEP was measured by asking participants to rate their current LBP from 0 (no pain) to 100 (most intense pain imaginable) immediately after completion of each of the five tasks on the BPS. MEP was determined from an average of the five pain ratings. Internal consistency was good for this measure ($\alpha = 0.83$).

PROMIS physical function

To evaluate the general physical functioning, the short form of the PROMIS Physical Function measure was administered (Rose et al., 2008, 2014). This scale includes four questions (e.g., “Are you able to do chores such as vacuuming or yard work?”) to examine the difficulty with which an individual is able to complete certain functional tasks. Ratings are made from 5 (without any difficulty) to 1 (unable to do) and lower scores indicate greater difficulty with task performance. This measure demonstrated high reliability among the sample ($\alpha = 0.85$).

PROMIS pain intensity

The three-item PROMIS Pain Intensity short form measure was used to evaluate pain intensity over the past week (Cella et al., 2010). This scale asks respondents to report their average and worst pain during the past 7 days, as well as pain at the time of questionnaire completion by providing a 1 (no pain) to 5 (very severe) pain rating. The PROMIS Pain Intensity scale demonstrated good reliability ($\alpha = 0.81$).

Roland-morris disability questionnaire

The Roland-Morris Disability Questionnaire (RMDQ) is a self-report measure that assesses health status and disability related to LBP (Roland and Morris, 1983). The RMDQ is comprised of 24 statements such as “I stay at home most of the time because of my back” and “I only walk short distances because of my back.” Respondents are instructed to indicate which of the statements describe their current experience. The number of endorsed items is summed to obtain a total score (more items

endorsed = greater disability). Internal consistency was high for this measure ($\alpha = 0.87$).

PROMIS depression scale

The eight-item short form of the PROMIS Depression Scale was used to assess depressive symptoms (e.g., “I felt worthless.”) (Pilkonis et al., 2011). Respondents rate the frequency of their experience of each symptom in the past 7 days from 1 (never) to 5 (always), with higher scores indicating a greater presence of depressive symptoms. The PROMIS Depression Scale demonstrated high reliability ($\alpha = 0.93$).

Brief resilience scale

Trait resilience was examined using the Brief Resilience Scale (BRS), which is a six-item measure examining the ability to bounce back and recover from stressful events and challenges (e.g., “I tend to bounce back quickly after hard times.”) (Smith et al., 2008). Responses are provided using a five-point scale (1 = strongly disagree, 5 = strongly agree), with total scores ranging from 6 to 30. Higher scores on the BRS indicate greater resilience (Cronbach's $\alpha = 0.84$).

World health organization quality of life-brief

The World Health Organization Quality of Life-Brief (WHOQOL-BREF) is a 26-item questionnaire designed to measure quality of life across four domains over the past 2 weeks: physical health, psychological health, social relationships, and environment (Skevington et al., 2004). The first item of the WHOQOL-BREF (i.e., “How would you rate your quality of life?”) was used to examine overall quality of life. This item is rated from 1 (very poor) to 5 (very good).

Statistical Analysis

All analyses were conducted using SPSS 24 and significance level was set at $p \leq 0.05$ (two-tailed). Means, standard deviations, and counts for demographic characteristics were calculated using descriptive statistics. Zero-order correlations were conducted between sociodemographic characteristics and outcome variables (i.e., physical function, MEP, pain intensity, back-related disability, depressive symptoms, general resilience, and quality of life). Demographic variables that were significantly related to outcome variables were controlled for in cluster analyses. The following 11 variables were entered into a PCA to characterize the dimensionality of each resilience measure: PA, dispositional hope, positive well-being, optimism, waist-hip ratio, body mass index, physical health comorbidities, smoking status, emotional support, instrumental support, and informational support. PCA with oblique rotation was used to allow for correlation between factors, with the recommendation that at least three items load on a factor and a difference of ≥ 0.20 was present between cross-loadings (Howard, 2016). Components with eigenvalues > 1 were selected for further analysis and the scree plot was inspected to confirm the number of factors to be retained. Hierarchical cluster analysis employing Ward's clustering method with squared Euclidean distances as the similarity measures was conducted to identify subgroups of individuals that differed across empirically derived resilience domains. Agglomeration coefficients were examined to identify the cluster solution that

best represented the data, with the optimal number being chosen based upon the point at which the percentage change was the largest between the clusters (Milligan and Cooper, 1985). Chi-square analysis for categorical variables or analysis of variance (ANOVA) for continuous variables was employed to examine cluster group differences across demographic composition. Differences across physical function, pain, and psychological outcomes were assessed using multivariate ANOVA's, controlling for the effects of relevant sociodemographic characteristics. Significant findings on multivariate analyses were followed by Sidak-corrected *post hoc* comparisons. To obtain effect size estimates associated with *F*-tests, partial eta-squared (η_p^2) was calculated (small = 0.01, medium = 0.06, and large = 0.14).

RESULTS

Participant Characteristics

Demographic characteristics (means and *SDs*) are reported in Table 1. Participants were mostly female (57%), White/Caucasian

(70%), had a college degree (50%), were married or partnered (52%), and were not employed (85%). Average age was 68 years (range: 60–93 years), duration of back pain was 16.4 years (range: 1–56 years), and participants reported back pain of moderate intensity during the initial session ($M = 5.5$, range = 2–10). Two of the 69 participants discontinued after the first session due to time constraints, and 7 participants who were initially eligible were excluded during their first appointment ($n = 1$ use of exclusion medications, $n = 3$ exclusionary medical condition, $n = 3$ not meeting pain duration criteria), thus leaving 60 participants. Based on ICD-10 classifications (World Health Organization, 2016), the following medical comorbidities/diseases were reported: circulatory and respiratory ($n = 27$, 45.0%), metabolic and endocrine ($n = 14$, 23.3%), genitourinary and renal ($n = 4$, 6.7%), digestive ($n = 3$, 5.0%), skin/subcutaneous tissue ($n = 3$, 5.0%), eye ($n = 3$, 5.0%), musculoskeletal ($n = 1$, 1.6%), nervous system ($n = 1$, 1.6%), infectious disease ($n = 3$, 5.0%), and sleep disorders ($n = 2$, 3.3%). Current smoking was reported among 16.7% ($n = 10$) of the sample.

TABLE 1 | Demographic and clinical characteristics.

Characteristic	Total sample (<i>n</i> = 60)		Cluster 1 High resilience (<i>n</i> = 25)		Cluster 2 High health low PsySoc (<i>n</i> = 13)		Cluster 3 High PsySoc low health (<i>n</i> = 15)		Cluster 4 Low resilience (<i>n</i> = 7)	
	M or N	SD or %	M or N	SD or %	M or N	SD or %	M or N	SD or %	M or N	SD or %
Age (years)	68.1	7.0	69.1	5.8	65.4	5.1	70.0	10.2	65.6	4.7
Sex										
Male	26	43.3	11	44.0	3	23.1	9	60.0	3	42.9
Female	34	56.7	14	56.0	10	76.9	6	40.0	4	57.1
Race										
White/Caucasian	42	70.0	19	76.0	8	61.5	10	66.7	5	71.4
Black/African American	12	20.0	4	16.0	3	23.1	4	26.7	1	14.3
Other	6	10.0	2	8.0	2	15.4	1	6.7	1	14.3
Education										
≤HS diploma	13	21.7	3	12.0	4	30.8	5	33.3	1	14.3
Some college/tech degree	17	28.3	7	28.0	3	23.1	3	20.0	4	57.1
Associates/bachelors	18	30.0	11	44.0	4	30.8	2	13.3	1	14.3
Graduate/professional	12	20.0	4	16.0	2	15.4	5	33.3	1	14.3
Marital status										
Married/partnered	31	51.7	21	84.0	2	15.4	6	40.0	2	28.6
Not married/partnered	29	48.3	4	16.0	11	84.6	9	60.0	5	71.4
Employment										
Employed	9	15.0	4	16.0	2	15.4	2	13.3	1	14.3
Not employed	51	85.0	21	84.0	11	84.6	13	86.7	6	85.7
Income*										
≤\$20,000	21	35.0	5	20.8	5	38.5	7	50.0	4	66.7
\$20,000–39,999	10	16.7	3	12.5	5	38.5	1	7.1	1	16.7
\$40,000–59,999	11	18.3	7	29.2	1	7.7	3	21.4	0	0.0
\$60,000–99,999	8	13.3	6	25.0	2	15.4	0	0.0	0	0.0
≥\$100,000	7	11.7	3	12.5	0	0.0	3	21.4	1	16.7
Back pain duration (years)	16.4	14.2	20.8	16.1	15.8	12.9	9.9	10.8	16.0	12.7

*Some data not reported. HS, high school.

TABLE 2 | Zero-order correlations across sociodemographic characteristics and study outcomes.

	BPS function	BPS pain	PROMIS function	PROMIS pain	RMDQ disability	PROMIS depression	BRS resilience	WHOQOL QOL
Age	0.05	−0.18	0.01	−0.23	−0.11	−0.30*	0.29*	0.27*
Sex	0.18	0.29*	−0.20	0.24	0.23	−0.09	0.09	−0.05
Race	0.16	0.28*	−0.32*	0.42**	0.36**	0.02	−0.16	0.03
Education	−0.12	−0.25*	0.20	−0.35**	−0.38**	−0.03	0.23	0.05
Marital status	−0.13	0.03	−0.25	0.25*	0.21	0.30*	−0.37**	−0.20
Employment	0.14	0.26*	−0.22	0.26*	0.22	0.05	−0.12	−0.01
Income	0.01	−0.36**	0.33*	−0.55**	−0.43**	−0.32*	0.45**	0.45**
Pain duration	−0.00	−0.26*	0.28*	−0.13	−0.35**	−0.14	0.07	0.16

* $p \leq 0.05$, ** $p \leq 0.01$. Sex coded: 0 = male, 1 = female; Race coded: 0 = white, 1 = black/other; Marital Status coded: 0 = married/partnered, 1 = not married/partnered; Employment coded: 0 = employed, 1 = not employed. BPS, Back Performance Scale; PROMIS, Patient-Reported Outcomes Measurement Information System; RMDQ, Roland-Morris Disability Questionnaire; BRS, Brief Resilience Scale; WHOQOL, World Health Organization Quality of Life questionnaire.

Zero-Order Correlations

To identify potential study covariates, zero-order correlations were analyzed across sociodemographic variables and study outcomes (Table 2). In general, age, sex, race, education, marital status, employment, income, and back pain duration were associated with physical function, pain, and psychological outcomes (all p s < 0.04). Hence, analyses assessing cluster group differences across study outcomes included these sociodemographic variables as statistical covariates.

Principal Components Analysis

A PCA was conducted with all 11 items using oblique rotation (direct oblimin), resulting in a four-factor solution. However, on the basis of our item selection criteria (i.e., more than or equal to three items load on a factor), this solution was eliminated as it returned one component containing smoking status. This variable was therefore removed from the model. The resulting analysis revealed the presence of a three-factor solution with eigenvalues over Kaiser's criterion of 1, accounting for 72.4% of the variance in scores. The Kaiser–Meyer–Olkin measure verified the sampling adequacy for the analysis (Kaiser–Meyer–Olkin = 0.78; Bartlett's test of sphericity = <0.001) and all KMO values were above the acceptable limit of 0.50 (Field, 2013). Inspection of the scree plot confirmed inflexions that would justify retaining three factors. Table 3 reports the factor loadings after rotation, with Component 1 representing positive, psychological factors (factor loadings 0.67–0.91), Component 2 denoting health-related functioning (factor loadings 0.60–0.78), and Component 3 reflecting social support (factor loadings 0.77–0.86). The factor loadings from each domain were used in subsequent cluster analysis.

Cluster Analysis Across Resilience Domains

The three composite domains were subjected to Cluster Analysis to identify empirically derived classifications based upon profiles of psychological, health, and social resiliency (Figure 1 and Supplementary Table 1). For ease of interpretation, the health domain was reverse scored, such that lower scores reflected higher waist–hip ratio, body mass index, and health comorbidities. Four clusters were revealed and characterized by

the following: (1) Cluster 1: High Resilience group ($n = 25$, 41.7%): high levels of psychological, health, and social support functioning; (2) Cluster 2: High Health/Low Psychosocial group ($n = 13$, 21.7%): optimal health-related functioning and low levels of psychosocial function; (3) Cluster 3: High Psychosocial/Low Health group ($n = 15$, 25.0%): poor health functioning, high psychological functioning, and moderate-to-high social support; and (4) Cluster 4: Low Resilience group ($n = 7$, 11.7%): low levels of functioning across psychological, social, and health-related factors. There were no sociodemographic differences across cluster groups, with the exception of the High Resilience group (Cluster 1) having the highest proportion of participants who were married or partnered (Table 1); thereby, consistent with previous research (Mun et al., 2019).

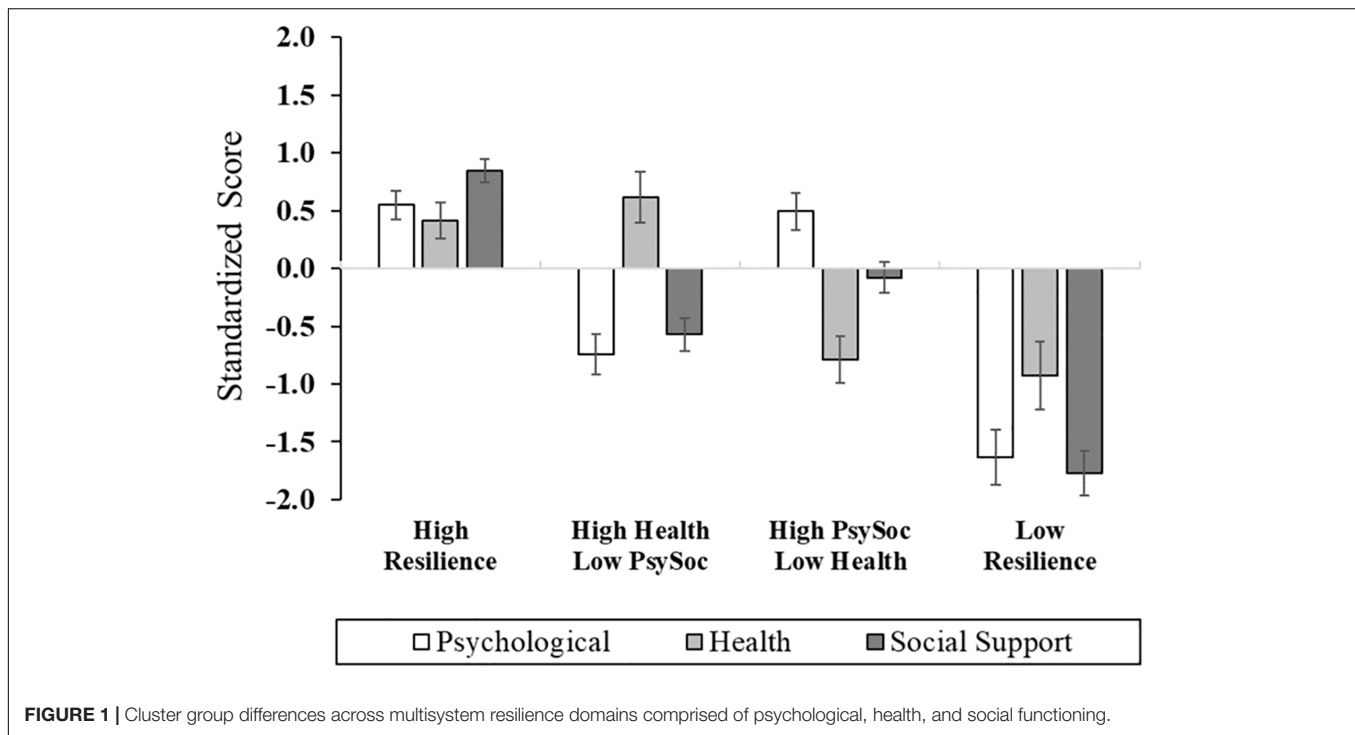
Psychosocial Profiles Across Cluster Group

After adjusting for age, sex, race, education, marital status, employment, income, and back pain duration, significant differences across cluster membership emerged in functional performance, physical function, back-related functional disability, depression, general resilience, and quality of life

TABLE 3 | Principal components analysis loadings across resilience domains.

Measures	Factor 1	Factor 2	Factor 3
Positive affect	0.91	0.01	−0.03
Dispositional hope	0.85	0.03	0.08
Positive well-being	0.69	0.02	0.42
Optimism	0.67	−0.19	0.14
Waist–hip ratio	−0.20	0.78	0.30
Body mass index	−0.04	0.77	−0.04
Health comorbidities	0.34	0.60	−0.36
Emotional support	−0.00	0.09	0.86
Instrumental support	0.26	−0.05	0.80
Informational support	0.33	−0.03	0.77
Eigenvalue	4.36	1.59	1.29
% Variance	43.58	15.91	12.88
% Cumulative variance	—	59.49	72.37

Bolded values were retained in each component.



(Figures 2, 3 and Supplementary Table 2). In particular, functional performance and functional disability due to LBP were poorest among the Low Resilience group, relative to individuals in the High Resilience and High Health/Low Psychosocial cluster groups ($p < 0.05$). *Post hoc* comparisons were non-significant across cluster groups for self-reported physical function, although the difference between the High Health/Low Psychosocial and Low Resilience groups approached significance with a large effect size ($\eta_p^2 = 0.22$). For psychological outcomes, individuals in the Low Resilience group reported the highest levels of depression ($p \leq 0.001$) and lowest quality of life ($p \leq 0.04$), relative to all other groups. Depression was also lower in the High Resilience group ($p = 0.05$), when compared to individuals with low psychosocial resources (Cluster 2). The High Resilience ($p = 0.04$) and High Psychosocial/Low Health ($p = 0.02$) clusters had greater general resilience than the High Health/Low Psychosocial group. In addition, while the Low Resilience group reported statistically lower levels of general resilience relative to the High Psychosocial/Low Health group ($p = 0.04$), these effects only approached significance ($p = 0.07$) when compared with the High Resilience group. No differences in MEP ($p = 0.08$) or self-reported pain intensity ($p = 0.33$) were detected across cluster groups.

DISCUSSION

Although risk factors have been extensively studied in respect to pain, there is a burgeoning literature supporting the role of resilience mechanisms in promoting adaptive pain outcomes. Mounting evidence signifies that a multitude of psychological,

social, and physical/biological factors confer resilience; however, much of the extant literature has focused on psychological resources. Moreover, resilience factors have predominantly been examined in isolation, thus overlooking their potentially synergistic and additive effects. Due to the exponential growth of older adults and global burden of chronic pain in this population, explicating the mechanisms that protect against pain and disability is of critical importance. While a modest literature has considered the cumulative effects of a broad range of personal resources (Agrigoroaei and Lachman, 2011; Sun et al., 2012; Johnson et al., 2019), we have extended previous research by capturing the multifaceted nature of resilience and exploring its influence on pain outcomes among older adults with cLBP.

Aligning with study hypotheses, we found evidence of phenotypic patterns of resilience based upon psychological, social, and health-related functioning. In particular, individuals with a higher array of protective factors exhibited more optimal outcomes in physical function, disability, and psychological processes (despite similar levels of pain), suggesting potentially important benefits of multiple adaptive resources. Overall, these findings signify that individuals with a more resilient phenotype may have a greater sense of coherence that allows them to mobilize resources to successfully navigate the ongoing challenges associated with pain. This would align with Antonovsky's salutogenic model of health (Antonovsky, 1996) which highlights the importance of coping strengths in fostering one's capacity for optimal health and well-being.

While a number of studies have classified patients according to negative psychological and lifestyle variables (Rabey et al.,

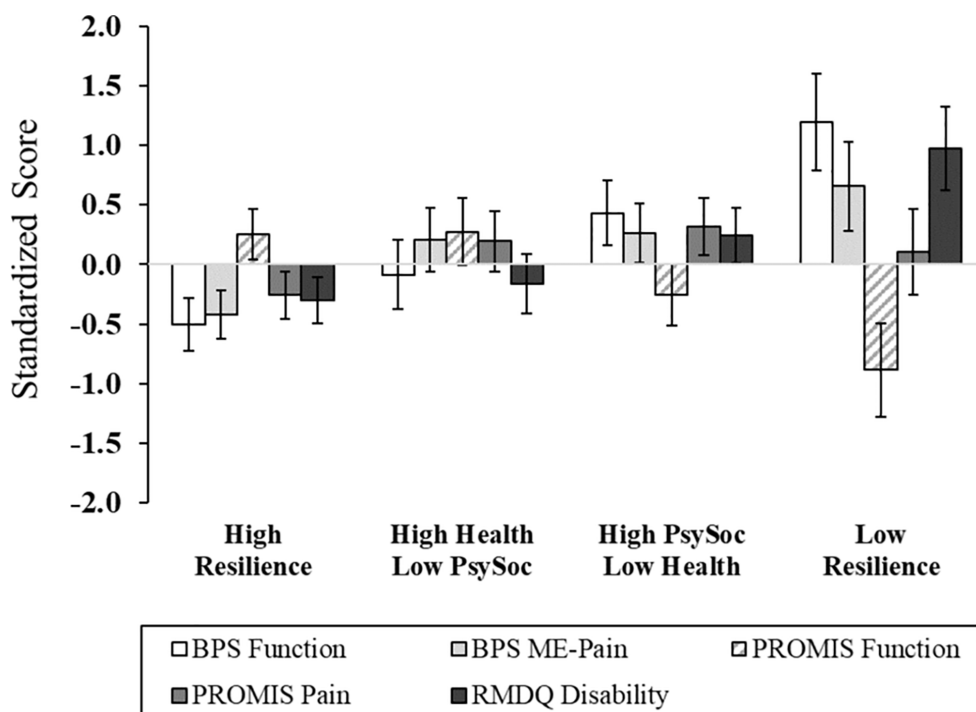


FIGURE 2 | Pain and physical functioning outcomes across multisystem resilience profiles. Relative to Cluster 4 (Low Resilience group), individuals with a greater degree of protective resources had higher functional performance and self-reported physical function, as well as lower disability. There were no group differences in movement-evoked pain or pain intensity. Higher scores on PROMIS function, better physical functioning; BPS, Back Performance Scale; ME, Movement-Evoked; PROMIS, Patient-Reported Outcomes Measurement Information System; RMDQ, Roland-Morris Disability Questionnaire.

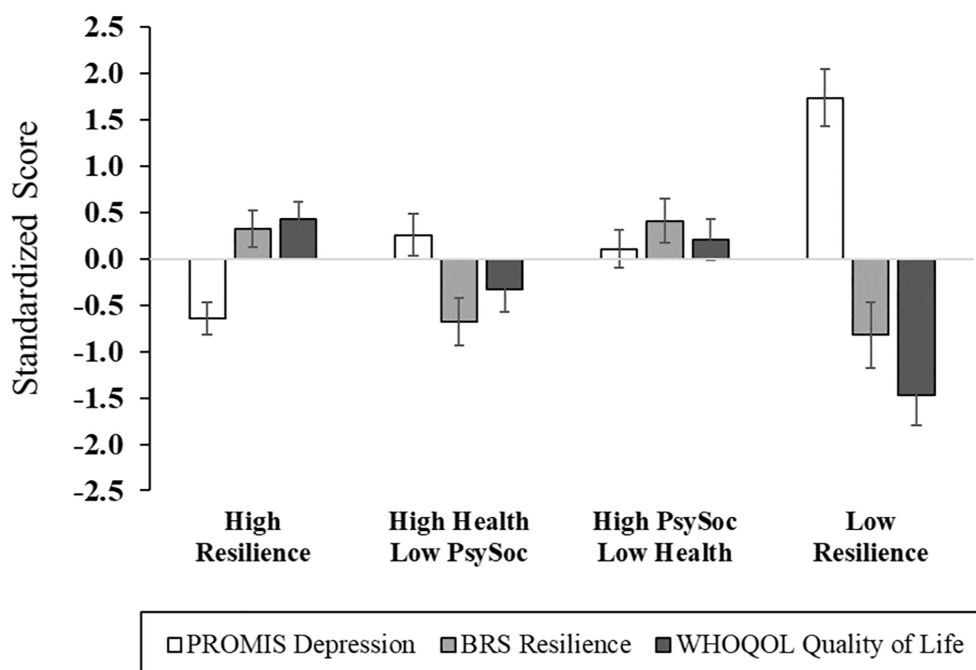


FIGURE 3 | Psychological functioning across multisystem resilience profiles. Compared to Cluster 4 (Low Resilience group), individuals with more resilient phenotypes exhibited lower depressive symptoms, and higher general resilience and quality of life. PROMIS, Patient-Reported Outcomes Measurement Information System; BRS, Brief Resilience Scale; WHOQOL, World Health Organization Quality of Life questionnaire.

2016; Almeida et al., 2018), limited research has stratified subgroups according to sources of resilience. In fibromyalgia, Mun et al. (2019) found that individuals with a higher degree of personal resources (i.e., pain acceptance, resilience, social support, sleep quality) exhibited lower levels of morning pain and depressive symptoms, as well as afternoon pain interference (although effects varied according to level of depression). Similar findings were also observed among patients with chronic neurological/neuromuscular disease, as those with a more resilient profile reported lower interference from pain (Mun et al., 2019). Furthermore, our findings echo a previous study in knee osteoarthritis (Cruz-Almeida et al., 2013), whereby a subgroup characterized by high optimism and low NA exhibited the lowest degree of pain, disability, and somatosensory sensitivity.

Results also suggest that health and psychosocial factors are differentially expressed across older adults with LBP. In particular, when compared to individuals with low resilience (Cluster 4), those with a higher degree of protective resources exhibited lower depression and higher quality of life. However, the findings for general resilience were more robust among individuals with higher social support and positive, psychological function (Cluster 3), thus underscoring the protective nature of psychosocial resources in coping with stress and adversity. Likewise, more favorable outcomes in functional performance and disability were not only observed among individuals with higher overall resilience, but also among those with more adaptive health-related function. This is not entirely surprising as higher disease burden and adiposity may facilitate decrements in functional capacity through mechanisms linked to frailty, psychological comorbidities, physiological dysregulation, increased joint loading, cardiopulmonary reserve, and activity restriction, among others (Fried et al., 2004; Kalyani et al., 2010; Calders and Van Ginckel, 2018). What is more, the influence of these health factors is likely not independent; rather, their effects are interactive and systemic, impacting multiple homeostatic processes to potentiate downstream effects on disability and function (Chapman et al., 2008).

Importantly, our findings have important clinical value as the protective resources we examined are modifiable. For instance, a greater emphasis on enhancing social support and positive, psychological processes may be particularly advantageous for improving adaptive coping, and to some extent, attenuating depressive symptoms in Cluster 2 individuals. Interventions with empirical support for their efficacy (e.g., cognitive-behavioral therapy and spouse-assisted training) (Edwards et al., 2016) are likely to derive some benefit; however, therapies focusing on harnessing resilience through positive, psychological resources [e.g., positive activity interventions (PAIs)] have also shown promise in chronic pain populations (Hausmann et al., 2014, 2017; Muller et al., 2016; Peters et al., 2017). For individuals with poorer health-related functioning (Cluster 3), minimizing the severity of multimorbidity and reducing weight burden through diet and exercise promotion may mitigate functional decline and disability. Ultimately, the development of strategies for the prevention of obesity and medical comorbidities is a

critical directive. Results also suggest that individuals with a low resilient profile would likely benefit from a multimodal approach that optimizes both psychosocial and health-related resources. In particular, combining psychotherapy with lifestyle modification may yield protective benefits in physical and emotional functioning. For individuals in Cluster 1 who appear to be adapting well despite the presence of cLBP, these treatments may be less justified. Surprisingly, while indices of pain intensity (i.e., PROMIS pain intensity, MEP) were lower among individuals with a greater degree of resources (Cluster 1), these effects failed to reach significance across cluster groups. Although it is conceivable that other unmeasured resources may have a more robust influence on pain severity, it is also possible our study was underpowered to detect pain-specific effects. On the basis of the effect sizes observed ($\eta_p^2 = 0.07$ to 0.14), a power analysis revealed that a sample size of 72 to 150 participants would be adequate to detect significant effects in MEP and self-reported pain intensity, respectively. Thus, consideration of these findings in a larger sample is warranted.

Multiple resources shape the expression and development of resilience in chronic pain. While the measurement of resilience is inherently complex (Southwick et al., 2014) due to varying definitions and multiple methods by which to assess this construct, our current models lack precision and fail to account for the multifaceted nature of resilience. Indeed, recent theoretical literature (Liu et al., 2017) posits that resilience should be conceptualized from various levels of analysis that includes intraindividual (e.g., physiological, health behaviors), interpersonal (e.g., personality correlates, coping appraisals), and socio-ecological factors (e.g., socioeconomic status, group membership). While exploring the independent determinants that buffer against negative pain sequelae has clinical utility, recognition of resilience from a multidimensional perspective will likely provide a greater understanding of adaptive capacity. Expanding our current models of resilience and considering new approaches, both theoretically and statistically, in how resilience is conceptualized and assessed in the context of pain will be an important future direction.

Strengths and Limitations

Several strengths of our study merit acknowledgment. To our knowledge, the current investigation is the first to examine resilience from a multidimensional perspective in older adults with cLBP. Participants were phenotyped according to several adaptive resources, offering a novel opportunity to explore how pain, disability, and psychological functioning differ across resiliency profiles. We used an empirical approach to characterize our resilience indices, which provides a statistical, data-driven, and robust method for classification of subgroups. A number of valid and reliable measures were also utilized across the assessment of psychological, social, and health-related functioning. Further, despite the small sample ($N = 60$), large effect sizes (ranging from $\eta_p^2 = 0.17$ to 0.45) were observed for pain and psychological outcomes.

In spite of these strengths, a few limitations are worth noting in the interpretation of results. First, given the nature of

cluster analysis our findings should be considered exploratory. In addition, our relatively modest sample may have influenced our classification of individuals using cluster analysis, with few individuals categorized into particular profiles (e.g., only seven participants comprised the “Low Resilience” cluster). This may have impacted the external validity of the study, thereby compromising generalizability. Future studies with larger sample sizes are warranted to confirm these findings. In light of these limitations, results should be interpreted cautiously. Although we employed a robust, empirical approach to devise our resilience domains, smoking status was eliminated from the analyses due to its retention as an independent factor. Despite smoking being a relevant lifestyle behavior with tremendous health consequences, we have confidence that the omission of this variable did not alter our findings, as it only contributed a small amount of additional variance (4%) to our model. Furthermore, analyses revealed that smoking status did not differ across cluster groups ($p = 0.18$). Nevertheless, there are strengths and challenges to various statistical methods (e.g., factor analysis, z -scores, and median split approach) and future studies should consider the comparison of these approaches, as well as their clinical relevance. Of note, some cross-loadings (>0.30) were observed across psychological, health, and social support factors. Although this may be a limitation, this phenomenon is also anticipated given the natural correlation among these constructs (e.g., health-related constructs correlate with many other variables). Also, medical comorbidities were determined via participant self-report, which may not provide a complete representation of individuals’ health histories. Medical records should be obtained for verification of health history in future endeavors. Related, because we excluded individuals experiencing major medical conditions, these results may not generalize to those who experience more severe health comorbidities.

Although multiple psychosocial and health constructs were used to derive subgroups, this did not reflect an exhaustive list of protective resources. Indeed, the small sample limited our ability to examine several factors, including relevant physiological/biological markers (e.g., inflammatory cytokines) (Khan et al., 2017), or demographic characteristics such as sex and race/ethnicity. Moreover, there is a need to replicate these findings in a more diverse sample, especially given growing evidence of the impact of race/ethnicity on resilience and pain-related outcomes (Bartley et al., 2019a). Inclusion of these and other diversity variables will be important considerations for future studies. Employing additional measures of health/lifestyle factors (e.g., physical activity, sleep, diet, alcohol, and drug consumption), as well as other positive psychological (e.g., self-efficacy and sense of coherence) and social support-related indices (e.g., quantity versus quality of support) will be key to improving our understanding of resilience. Likewise, given the importance of external determinants of resilience, several other contextual and social/environmental factors, including socioeconomic status and access to healthcare, are critical areas to examine (Liu et al., 2017). And finally, it may be beneficial to use a dual-focus approach to closely examine both risk and resilience factors and how this interplay influences multisystem resiliency

in respect to pain (perhaps even considering the “degree” of negative factors, such as the impact of lower NA, less catastrophic thinking, etc.). For example, personal resources (such as optimism or PA) may broaden an individual’s coping repertoire by facilitating engagement in adaptive behaviors that mitigate the narrowing effects of pain catastrophizing (Fredrickson, 2001). This would align with predominant risk-resilience models that highlight the consideration of both vulnerability and protective mechanisms in understanding individual adaptation to pain (Sturgeon and Zautra, 2013; Goubert and Trompetter, 2017).

CONCLUSION

In sum, our findings support the contribution of protective factors in the context of pain and suggest that examining resilience from a multisystem perspective may have significant clinical utility. Importantly, homogenous subgroups emerged from psychological, social, and health-related processes, with lower disability, better functional performance, and higher psychological functioning observed among individuals with a more resilient phenotype. Consideration of the multiple resources that harness resilience, including their additive effects, may improve our understanding of adaptive function among older adults with chronic pain and ultimately facilitate the development of more targeted clinical care.

DATA AVAILABILITY

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the University of Florida Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

EB and SP contributed to the conception and design of the study, and wrote the first draft of the manuscript. EB performed the statistical analysis. All authors reviewed the text critically and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsyg.2019.01932/full#supplementary-material>

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Psychological Flexibility as a Resilience Factor in Individuals With Chronic Pain

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Resilience factors have been suggested as key mechanisms in the relation between symptoms and disability among individuals with chronic pain. However, there is a need to better operationalize resilience and to empirically evaluate its role and function. The present study examined psychological flexibility as a resilience factor in relation to symptoms and functioning among 252 adults with chronic pain applying for participation in a digital ACT-based self-help treatment. Participants completed measures of symptoms (pain intensity, and anxiety), functioning (pain interference and depression), as well as the hypothesized resilience factor psychological flexibility (measured as avoidance, value obstruction, and value progress). As expected, symptoms, functioning and resilience factors were significantly associated. Hierarchical linear regression analyses showed that psychological flexibility significantly contributed to the prediction of pain interference and depression when adjusting for age, pain and anxiety. Also, participants with low levels of psychological flexibility were more likely to be on sick leave. Furthermore, a series of multiple mediation analyses showed that psychological flexibility had a significant indirect effect on the relationship between symptoms and functioning. Avoidance was consistently shown to contribute to the indirect effect. Results support previous findings and suggest the importance of psychological flexibility as a resilience factor among individuals with chronic pain and anxiety.

Keywords: chronic pain, psychological flexibility, resilience, avoidance, values

INTRODUCTION

Resilience – the ability to adapt and function well despite significant strain – has gained increasing attention in the field of chronic pain management. Turk et al. (2008) concluded that “Living with chronic pain requires considerable emotional resilience and tends to deplete emotional reserve.” Patients often report “being stuck” or “putting life on hold” as a consequence of chronic pain, which corresponds with data showing that pain interference rather than pain intensity, predicts levels of functioning (Kemani et al., 2016). Why some patients continue to function well in day to day life after the onset of chronic pain, and others do not, is yet unclear (Reid et al., 2011; Hauser et al., 2014; Vervoort et al., 2014; Goubert and Trompetter, 2017).

Resilience is commonly defined as overcoming adversity or “effective functioning, despite the exposure to stressful circumstances, and/or internal distress” (Sturgeon and Zautra, 2013).

However, current definitions of resilience provide limited information on *how* a person recovers or maintain functioning during and following difficult life events. The concept of resilience sometimes reflects a deterministic view where resilience factors are relying on personality traits, biology, social support, past experiences, and/or innate properties such as sex. Resilience may alternatively, and in accordance with Goubert and Trompetter (2017), be conceptualized as a contextual behavioral factor, and defined as the ability to continuously engage in meaningful activities that promote current and future quality of life and health, in the presence of pain and distress (Goubert and Trompetter, 2017). This conceptualization of resilience is closely related to the construct *psychological flexibility*, defined as the ability to act in alignment with values and long-term goals in the presence of inner discomfort such as pain and distress and has been suggested as a key factor in maintaining or improving functioning among individuals with chronic pain (Hayes et al., 1999, 2006).

Lack of psychological flexibility, or psychological *inflexibility*, is commonly displayed as *avoidance* of stimuli and situations associated with pain, and related distress. From a learning perspective, avoidance is an operant under contextual control, meaning that a behavior is influenced by environmental factors (internal and external) preceding the behavior, and/or acting as consequences of that same behavior. Avoidance behaviors may be reinforced – for example resulting in short term reductions of pain or discomfort – and therefore sustained and used in similar contexts. Over time, such operant behaviors may become a default strategy in situations perceived as threatening, with increasing difficulties for the individual to respond differently (Vowles et al., 2007). Due to contextual factors, the behavior repertoire becomes increasingly narrow and less flexible. This behavior pattern is normally not associated with a corresponding decrease in pain or distress, but rather a life less stimulating and active. Avoidance may take many different forms, such as not engaging in social or physical activities, excessive opioid use (overt behaviors), thinking about situations associated with pain or refraining from planning future events (covert behaviors).

Value orientation is another key aspect of psychological flexibility, that can be defined as verbal guidelines that function to initiate and maintain behavior over time, also without the presence of obvious reinforcers (Hayes et al., 1999; Hayes, 2012). For example, clarifying the value (“being an attentive parent”) associated with an operant (“playing in the park”) can alter the context and thereby increase the likelihood that this behavior is initiated and/or maintained also in the context of potentially interfering pain. For chronic pain patients, value-based behaviors have been associated with higher levels of functioning and improved mood (Vowles et al., 2014) and adding a value aspect in an experimental intervention has been shown to elevate pain tolerance (Branstetter-Rost et al., 2009). Value orientation usually comprise components such as clarifying and engaging in value-based behaviors, including effectively dealing with obstacles to value-based behaviors (Smout et al., 2014).

The aim of the present study was to examine the role and function of psychological flexibility, assessed as avoidance, value obstruction, and value progress as resilience factors in a sample

of individuals with chronic pain. More specifically, analyses were conducted to (1) broadly characterize the relationship between symptoms, functioning and psychological flexibility, (2) assess the amount of variance in functioning (pain interference and depression) explained by psychological flexibility, (3) explore low vs. high psychological flexibility as a potential risk/resilience factor for self-reported sick leave and opioid use, and (4) examine the indirect effects of psychological flexibility in the relationship between symptoms and functioning.

MATERIALS AND METHODS

Data Collection

This study used a cross-sectional design, with data from baseline assessments for participants applying for internet-delivered Acceptance and Commitment Therapy for chronic pain (ClinicalTrials.gov identifier: NCT03105908 and NCT03344926). Participants were recruited via ads in newspapers and social media between January 30, 2017 and January 31, 2018. All questionnaires and demographic questions were completed online in a secure web platform. Participants provided written informed consent prior to enrollment in the study, the study was approved by the Regional Ethics Committee and followed the Helsinki declaration.

Eligibility criteria were pain duration over 6 months, age 18 years or older, ability to read and write in Swedish and completion of all assessments.

Measurements

Demographic variables included age, sex and educational level.

Symptom Variables

Pain variables included *pain intensity* – current and average in the past week – measured with a numeric rating scale (NRS, 0 = no pain at all, 10 = worst pain imaginable), *pain duration* (self-reported in years), and *pain location* (self-reported descriptions of localization).

To account for some of the complexity of symptoms commonly co-occurring with chronic pain, the present study used *anxiety* as measured with Generalized Anxiety Disorder-7 (GAD-7) (Spitzer et al., 2006) as a proxy for distress symptoms such as strain, worry, and restlessness.

GAD-7 measures the frequency of anxiety symptoms during the last 2 weeks are scored on a four-point Likert scale (0 = Not at all, 3 = Every day). A total score of 10 was chosen as cut-off for anxiety problems, in accordance with guidelines (Spitzer et al., 2006). Internal consistency (Cronbach's Alpha) was 0.92 in the validation study (Spitzer et al., 2006), and 0.88 in the present dataset. GAD-7 is validated for chronic pain patients with migraine (Seo and Park, 2015a).

Functioning

Pain interference was measured with the pain interference index (PII), a brief self-report questionnaire assessing the influence of pain on behavior, or to what extent pain interferes with daily functioning (Kemani et al., 2016). Six items are rated on

a seven-point Likert scale (0 = Not at all, 6 = Completely). Internal consistency (Cronbach's Alpha) in the present sample was 0.85, which corresponds to the alpha from the original validation study, which was performed on a chronic pain sample (Kemani et al., 2016).

Level of *depression* was measured using the Patient Health Questionnaire 9 items (PHQ-9) (Kroenke et al., 2001). The frequency of depressive symptoms occurring during the past 2 weeks are rated on a four-point Likert scale from 0 = Not at all to 3 = Nearly every day. Internal consistency (Cronbach's Alpha) in the present sample was 0.81, and in the original validation studies Alpha varied from 0.86 to 0.89 (Kroenke et al., 2001). PHQ-9 has been validated for chronically ill patients (Wu, 2014) and migraine patients (Seo and Park, 2015b). PHQ-9 was used as a measure of functioning in this study, with the theoretical assumption that depressive disorder is a result of a series of behaviors, not merely the occurrence of symptoms.

Sick leave during the past 2 months was assessed using self-report and classified as temporary, permanent, or none. Due to a large overlap between temporary and permanent sick leave, the variable was transformed to a binary variable (sick leave/no sick leave).

Data on *opioid use* was collected via self-report. Participants listed all current medications, which then were classified according to the Anatomical Therapeutic Chemical classification system by an anesthesiologist. Participants with opioids in their list were coded as "currently taking opioids" and participants without opioids in their list were coded as "currently not taking opioids."

Psychological Flexibility

In the present study, two aspects of psychological flexibility were assessed: avoidance and values orientation.

Avoidance was measured using a subscale of Psychological Inflexibility in Pain Scale (PIPS) (Wicksell et al., 2008b). The avoidance subscale consists of 8 items, rated on a seven-point Likert scale from 1 = never true, to 7 = always true. The avoidance subscale has consistently been shown to be a robust and valid measure in chronic pain samples (Wicksell et al., 2010a; Barke et al., 2015). In the present sample internal consistency was 0.91, which corresponds with an alpha of 0.89 in the original validation study (Wicksell et al., 2008b).

Values orientation was assessed using the subscales *value progress* and *value obstruction* from the Valuing Questionnaire (VQ) (Smout et al., 2014). Items are rated on a seven-point Likert-scale ranging from 0 = not at all true, to 6 = completely true. Higher scores on the progress subscale indicate greater progress toward value-based behavior, while higher scores on the obstruction subscale indicate greater obstruction to value-based behavior. The two-factor solution has shown adequate model fit and strong internal consistency (Cronbach's alpha = 0.87) (Smout et al., 2014). In the present study Cronbach's Alpha was 0.83 for value progress and 0.76 for value obstruction. The Valuing Questionnaire has been validated for use with chronic pain samples (Carvalho et al., 2018).

Avoidance and value obstruction are both negatively valenced measures (measuring the occurrence of something

negative) while value progress is positively valenced (measuring the presence of something positive). However, both are important aspects of the psychological flexibility model (McCracken and Morley, 2014).

Participant Characteristics

The sample consists of adults with chronic pain applying for participation in a digital ACT-based self-help treatment. The sign-up-process for the clinical trial was initiated by 266 persons, of which 253 completed the assessment. One individual was excluded due to pain duration <6 months. Thus, data from 252 participants was used in the analyses.

Participants were predominantly female (81%, $n = 204$) and born in Sweden (90%, $n = 226$) with a mean age of 47.4 years (SD 11.5, range 18–70). Two thirds (66%, $n = 166$) of the sample had some level of university education (>12 years of education), nearly one third (30%, $n = 75$) had completed upper secondary school (12 years), and a few participants (4%, $n = 11$) had completed only compulsory school (9 years). Occupational status varied, with 31% ($n = 79$) working full-time, 25% ($n = 62$) part-time, 25% ($n = 63$) being on temporary sick leave, and 23% ($n = 59$) on permanent disability.

Pain duration was on average 18.2 years (SD 12.5, range 0.5–57), and the participants reported that last week's mean pain intensity was 6.6 (SD 1.7, range 1–10). Most individuals had multiple pain locations (88%, $n = 222$), and the most common pain locations were: back (75%, $n = 188$), neck (64%, $n = 160$), and lower extremities (64%, $n = 160$). Half of the sample experienced headaches (50%, $n = 125$) and 40% ($n = 101$) experienced generalized pain. Primary pain diagnoses were classified by an anesthesiologist as nociceptive (e.g., spinal disc hernia, and rheumatoid arthritis) for 37% ($n = 93$), as nociplastic (e.g., fibromyalgia and CRPS) for 17% ($n = 44$), neuropathic (e.g., trigeminal neuralgia and nerve damage) for 8% ($n = 20$), and headaches (e.g., migraine and Horton's) for 8% ($n = 19$). A fifth (19%, $n = 47$) had no diagnosis and 12% had mixed or unclassifiable pain diagnosis. Furthermore, more than half (54%, $n = 157$) scored above cut-off for depression, and one in four (25%, $n = 64$) above cut-off for anxiety.

Statistical Analyses

Analyses were computed using SPSS version 25 and STATA version 15. The dataset was complete, and no imputation strategies were needed. In all analyses, statistical significance was set to a conservative level of $p < 0.01$, except in the criteria for inclusion of covariates where $p < 0.05$ was used.

To determine the adequate sample size for mediation and regression analyses, a power analysis was conducted using the G*power software (Faul et al., 2009). Assuming a medium effect size (f^2) of 0.15, an alpha of 0.01, a power level of 0.80 and a total of 6 predictors, the power analysis suggested a minimum of 109 participants.

Initial analyses were conducted to broadly characterize the relationships between the variables using descriptive statistics and Pearson's product moment correlations (r) (Field, 2013).

A series of hierarchical linear regression analyses were performed to investigate the amount of variance explained by

psychological flexibility (avoidance, value obstruction, and value progress) in pain interference and depression. Demographic variables were entered as step 1, symptoms were added as step 2, and psychological flexibility as step 3. For each analysis, only variables having a significant bivariate correlation ($p < 0.05$) with the dependent variable were included.

The relationships between levels of psychological flexibility and the risk of sick leave and opioid use were analyzed using maximum likelihood logistic regression models. We estimated risks, or odds ratios (OR), for sick leave and opioid use (dependent variables) with each respective measure of psychological flexibility (avoidance, value obstruction, and value progress) as independent variables. The independent variables were categorized as low (first quartile), medium (second and third) and high (fourth) of the continuous distributions in line with recommendations from van Kuijk et al. (2019), as the association between predictors and outcomes was not linear and the assumption of non-additivity between different predictors and covariates was not met (van Kuijk et al., 2019). Age and pain intensity showed bivariate correlations with the dependent variables and were therefore used as covariates in these analyses.

A series of analyses of indirect effects using PROCESS for SPSS were conducted to evaluate the importance of psychological flexibility (avoidance, value obstruction, and value progress) for the relationships between predictors (pain and anxiety) and dependent variables (pain interference and depression). In all analyses the influence of age was adjusted for (covariate). Four models were analyzed, with each predictor and dependent variable, and with multiple mediators (PROCESS model #4). PROCESS is a bootstrapping method in which samples of the original size, drawn from the original data, are generated (Hayes and Rockwood, 2017). The total effect (c) is comprised of the direct effect (c') and the indirect effect (ab). Thus, the indirect effect represents the part of the relation between the predictor and the dependent variable that can be explained by the proposed mediator. The mean value for the ab product across the bootstrapped samples provided a point estimate of the indirect effect. Confidence intervals (CI) were derived from the obtained distribution of ab , using a 99% CI level representing a significance level of $p > 0.01$. If lower and upper bounds did not contain zero, the indirect effect was significant at the specified level. Each analysis was based on 5000 bootstrapped samples, as suggested by Preacher and Hayes (2008).

RESULTS

Mean, standard deviations, and range for all self-report measures are reported in Table 1.

Bivariate Correlations Between Symptoms, Functioning, and Psychological Flexibility

Strong positive correlations were found between avoidance and pain interference ($r = 0.668$), avoidance and depression ($r = 0.514$), value obstruction and depression ($r = 0.522$) as well as

TABLE 1 | Self-report measures: Means, standard deviations, and range.

Measures	Mean (SD)	Range (possible range)
Pain intensity average	6.6 (1.7)	1–10 (0–10)
Anxiety	7.1 (5.1)	0–21 (0–21)
Pain interference	23.5 (8.3)	1–36 (0–36)
Depression	11.3 (5.7)	0–26 (0–27)
Avoidance	35.9 (9.7)	8–56 (8–56)
Value obstruction	14.2 (6.8)	0–30 (0–30)
Value progress	14.1 (6.7)	0–30 (0–30)

between anxiety and depression ($r = 0.663$). Pairwise correlations between all variables are shown in Table 2.

Amount of Variance in Functioning Explained by Psychological Flexibility

Hierarchical regression analyses were conducted to evaluate the amount of variance explained by psychological flexibility in the two dependent variables *pain interference* and *depression*. Age had a significant positive bivariate correlation with sick-leave and a significant negative bivariate correlation with pain interference and depression and was therefore entered as step 1.

Pain Interference

Psychological flexibility explained a significant amount of variance in pain interference (r^2 change = 0.44, $p < 0.0001$), when adjusting for the influence of pain and anxiety ($r^2 = 0.27$, $p < 0.0001$). Of the psychological flexibility variables solely avoidance showed a significant – and positive – beta value (b 0.52, $p < 0.0001$).

Depression

In depression, psychological flexibility explained a significant amount of variance (r^2 change = 0.11, $p < 0.0001$) when adjusting for the influence of pain and anxiety. Avoidance had a significant positive beta coefficient and value progress had a significant negative beta coefficient in the model. Results from the hierarchical regression analyses are presented in Table 3.

Odds for Sick Leave and Opioid Use in Individuals With Low vs. High Psychological Flexibility

Logistic regression analyses were conducted to evaluate the odds for sick leave and opioid use in individuals with low and high psychological flexibility.

Sick Leave

The odds of being on sick leave was four times lower in the low value progress group compared to the high value progress group (OR 0.25, $p = 0.001$). For avoidance, the high avoidance group had lower odds for being on sick leave (OR 5.23, $p < 0.0001$) compared to the low avoidance group. For different levels of value obstruction there were no significant differences in odds for sick leave.

TABLE 2 | Correlations between all variables.

Variable	Sex ¹	Edu ¹	Dur	Intensity	Anx	Interfer	Depr	Sick ¹	Opioid ¹	Avoid	Obstr	Progress
Age	−0.099	−0.060	0.300**	0.042	−0.304**	−0.166**	−0.245**	0.159*	0.012	−0.141*	−0.266**	0.127*
Sex ¹	–	0.042	−0.006	0.081	0.101	0.067	−0.004	0.036	−0.108	−0.041	−0.024	0.004
Education ¹		–	0.022	−0.124	−0.023	−0.028	−0.034	−0.053	−0.029	−0.084	−0.057	0.033
Symptoms (predictors)												
Pain duration			–	0.110	−0.045	0.010	−0.022	0.051	0.077	−0.045	−0.093	0.037
Pain intensity				–	0.180**	0.356**	0.191**	−0.002	0.141*	0.266**	0.101	−0.018
Anxiety					–	0.387**	0.663**	−0.081	−0.039	0.342**	0.555**	−0.309**
Functioning (dependent variables)												
Pain interference						–	0.594**	0.216**	0.180**	0.668**	0.439**	−0.311**
Depression							–	0.091	0.084	0.514**	0.522**	−0.422**
Sick leave ¹								–	0.178**	0.184**	0.013	−0.188**
Opioid use ¹									–	0.121	−0.006	−0.074
Psychological flexibility (Independent variables)												
Avoidance										–	0.465**	−0.425**
Value obstruction											–	−0.386**
Value progress												–

* $p < 0.05$, ** $p < 0.01$. ¹Spearman correlation used for categorical variables. Edu, education; Dur, pain duration; Intensity, pain intensity; Anx, anxiety; Interfer, pain interference; Depr, depression; Avoid, avoidance; Obstr, obstruction.

TABLE 3 | Hierarchical linear regressions: the influence of psychological flexibility on functioning.

Dependent variable	Step	Predictors	R^2	R^2 Change	F Change (df)	Sig. F Change	Standardized Beta with all variables entered		
							β	t	Sig.
Pain interference	1	Demographics	0.03	0.03**	7.10 (1, 250)	0.008			
		Age					−0.04	−0.85	0.395
	2	Symptoms	0.24	0.21**	34.9 (2, 248)	<0.0001			
		Pain intensity					0.19**	4.01	<0.0001
		Anxiety					0.10	1.78	0.077
	3	Psy flex	0.51	0.27**	45.55 (3, 245)	<0.0001			
		Avoidance					0.52**	9.58	<0.0001
Depression		Obstruction					0.11	1.83	0.068
		Progress					−0.01	−0.13	0.895
	1	Demographics	0.06	0.06**	16.03 (1, 250)	<0.0001			
		Age						−0.62	0.533
	2	Symptoms	0.45	0.39**	86.91 (2, 248)	<0.0001			
		Pain intensity					0.03	0.69	0.493
		Anxiety					0.48**	8.96	<0.0001
	3	Psy flex	0.55	0.11**	19.54 (3, 245)	<0.0001			
		Avoidance					0.24**	4.63	<0.0001
		Obstruction					0.08	1.44	0.152
		Progress					−0.14**	−2.75	0.006

Psy flex, psychological flexibility. ** $p < 0.01$.

Opioid Use

For opioid use, no significant difference in odds between high and low avoidance, value obstruction, or value progress were found. Results from the logistic regression analyses are presented in **Table 4**.

The Indirect Effect of Psychological Flexibility in the Relationship Between Symptoms and Functioning

The bootstrap method (PROCESS) with $n = 5000$ bootstrap resamples and 99% bias-corrected and accelerated confidence

intervals was used in a series of analyses conducted to evaluate the indirect effects of psychological flexibility (M1 = avoidance, M2 = progress, and M3 = obstruction) on the relationship between symptoms (average pain intensity and anxiety) and functioning (pain interference and depression).

In short, all four multiple indirect effect models, with pain intensity/anxiety (x) as predictors and pain interference/ depression (y) as dependent variables, showed a significant total indirect effect of psychological flexibility, i.e., the combined indirect effects of avoidance (M1), progress (M2), and obstruction (M3). Results from the

TABLE 4 | Odds ratios for sick leave and opioid use, respectively, for low, medium and high levels of psychological flexibility, with age and pain intensity as covariates.

Dependent variable	Predictor	Low (ref. odds)		Medium			High		
		Yes/No	OR	Yes/No	OR	OR 99% CI	Yes/No	OR	OR, 99% CI
Sick leave	Avoidance	15/45	1.0	64/64	3.54**	1.40–8.95	35/29	5.23**	1.74–15.74
	Obstruction	28/35	1.0	56/77	1.24	0.51–2.97	30/26	2.56	0.81–8.25
	Progress	43/27	1.0	51/74	0.37**	0.16–0.85	20/37	0.25**	0.09–0.72
Opioid use	Avoidance	16/44	1.0	40/88	1.30	0.52–3.26	28/36	2.30	0.79–6.70
	Obstruction	23/40	1.0	43/90	0.97	0.39–2.38	18/38	0.97	0.29–3.21
	Progress	26/44	1.0	41/84	0.78	0.34–1.79	17/40	0.60	0.21–1.67

** $p < 0.01$.**TABLE 5 |** Total, direct and indirect effect of symptoms on pain interference and depression using psychological flexibility as indirect effect.

X	Y	m	a path coefficient	b path coefficient	Total effect (c)	Direct effect (c')	Indirect effect		
							CI (99%)		
							Effect (SE)	LLCI	ULCI
Pain intensity	Pain interference	Psy flex			1.77**	0.99**	0.78** (0.20)	0.26	1.32
		Avoidance	1.55**	0.45**			0.70** (0.18)	0.26	1.21
		Obstruction	0.44	0.19**			0.08 (0.05)	−0.04	0.26
		Progress	−0.09	−0.21			0.00 (0.02)	−0.06	0.08
	Depression	Psy flex			0.68**	0.32	0.36** (0.13)	0.02	0.71
		Avoidance	1.55**	0.15**			0.24** (0.08)	0.07	0.47
		Obstruction	0.44	0.25**			0.11 (0.06)	−0.05	0.30
		Progress	−0.09	−0.15**			0.01 (0.04)	−0.09	0.14
Anxiety	Pain interference	Psy flex			0.60**	0.22	0.38** (0.09)	0.17	0.61
		Avoidance	0.63**	0.50**			0.31** (0.07)	0.14	0.51
		Obstruction	0.68**	0.12			0.08 (0.05)	−0.06	0.23
		Progress	−0.40**	0.02			−0.01 (0.03)	−0.09	0.07
	Depression	Psy flex			0.73**	0.54**	0.18** (0.05)	0.07	0.31
		Avoidance	0.63**	0.15**			0.09** (0.03)	0.04	0.18
		Obstruction	0.68**	0.07			0.05 (0.04)	−0.05	0.14
		Progress	−0.40**	−0.11**			0.04** (0.02)	0.00	0.10

All models adjusted for age. Psy flex, psychological flexibility. ** $p < 0.01$.

analyses of indirect effects are summarized in Table 5. Detailed results for each model are presented below and in Figures 1–4.

Direct and Indirect Effect of Pain Intensity on Pain Interference

The full model showed a significant indirect effect of psychological flexibility on the relationship between pain intensity and pain interference. Among the individual psychological flexibility factors only avoidance had a significant indirect effect in the full model.

In the analyses of each respective path avoidance illustrated significant coefficients in both the *a* and *b* paths. Of the value factors, obstruction showed significant *b* path.

Direct and Indirect Effect of Pain Intensity on Depression

A significant indirect effect was shown for the full model. Notably, the direct effect was not significant, which implies a strong indirect effect of psychological flexibility on the relation between pain intensity and depression. Again, avoidance individually showed a significant indirect effect in the full model. Furthermore, avoidance showed significance in both *a* and *b* paths, whereas value obstruction and value progress had significant *b* paths only.

Direct and Indirect Effect of Anxiety on Pain Interference

A significant indirect effect of psychological flexibility on pain interference was seen in the full model. The direct effect

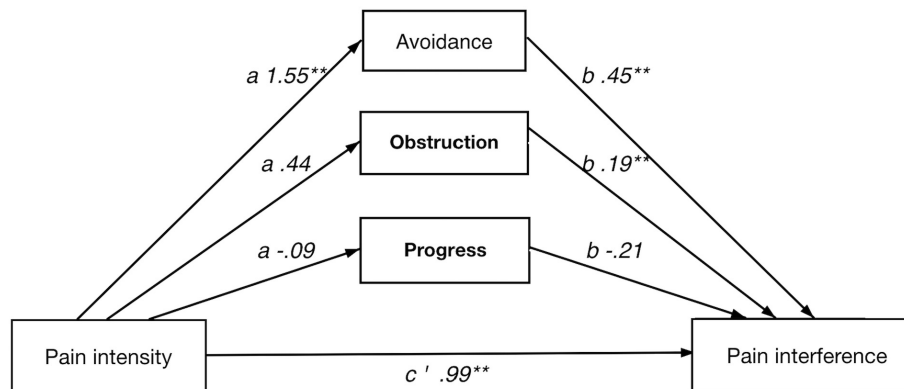


FIGURE 1 | Direct and indirect effect of pain intensity on pain interference.

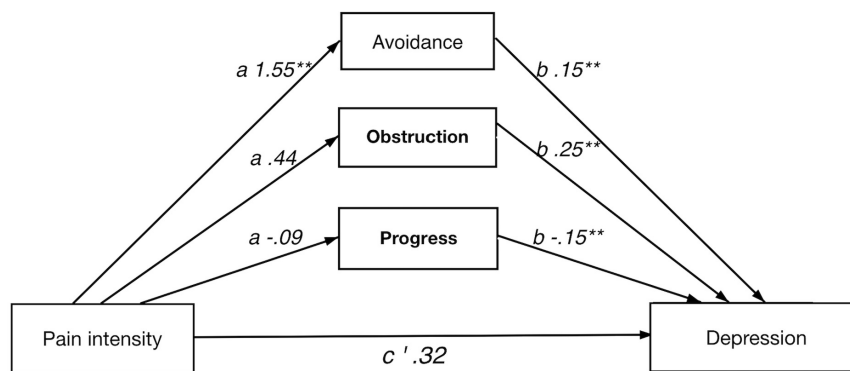


FIGURE 2 | Direct and indirect effect of pain intensity on depression.

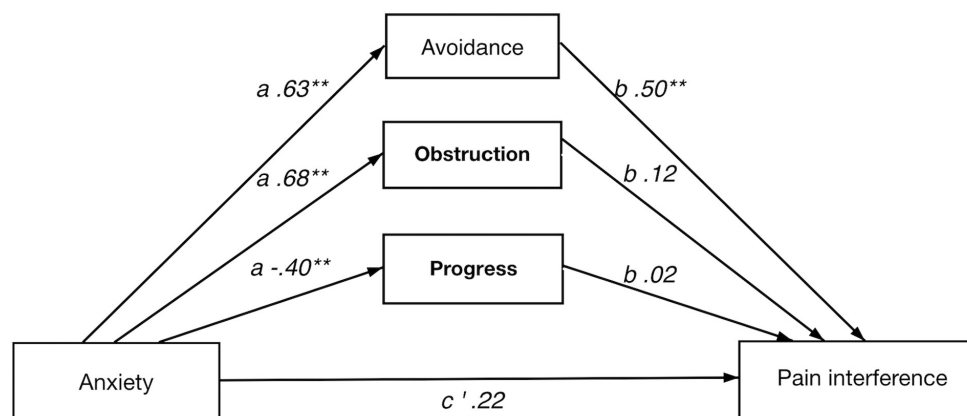


FIGURE 3 | Direct and indirect effect of anxiety on pain interference.

(anxiety on pain interference) was not significant, implying a strong indirect effect of psychological flexibility. Avoidance had an individually significant indirect effect in the full model. The analysis of individual paths showed avoidance had significant *a* and *b* paths in the model, where value obstruction and value progress showed significant *a* paths only.

Direct and Indirect Effect of Anxiety on Depression

The full model showed a significant indirect effect of psychological flexibility on depression. Among the individual psychological flexibility factors, both avoidance and value progress individually had significant indirect effect. Avoidance

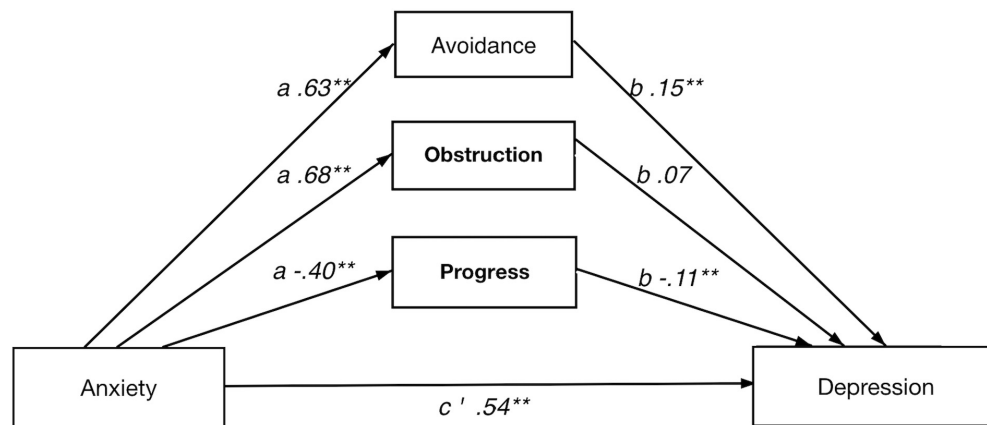


FIGURE 4 | Direct and indirect effect of anxiety on depression.

and value progress showed significant *a* and *b* paths while value obstruction only had significant *a* path.

DISCUSSION

The purpose of the present study was to examine the role and function of psychological flexibility – assessed with avoidance, value obstruction and value progress – as a resilience factor in a sample of 252 persons with chronic pain. In general, psychological flexibility was shown to be a significant factor in the relationship between symptoms (pain intensity and anxiety) and functioning (pain interference and depression).

Avoidance was shown to be particularly important for the influence of symptoms on functioning, as illustrated by significant *a*- and *b*-paths across all four models of indirect effects. The two value components, obstruction and progress, were primarily relevant in the association between anxiety and functioning, which warrants further studies to explore the relative importance of different aspects of psychological flexibility.

The results from the present study support research indicating the relevance of psychological flexibility in explaining variance in functioning in individuals with chronic pain (McCracken and Vowles, 2007; Zetterqvist et al., 2017). Previous studies have also shown that psychological flexibility is an important change mechanism in exposure-based interventions (Vowles et al., 2008, 2014; Wicksell et al., 2010b; Trompetter et al., 2015).

The conceptualization of resilience as a key factor in the relation between pain and functioning is seen also in a Scottish population-based study illustrating that resilient persons (high pain intensity and low disability score) had a higher 10-year-survival than vulnerable persons (low pain intensity, high disability score) (Elliott et al., 2014). In that study, factors associated with higher resilience were being male, lower age, higher education, owning your own home, and absence of chronic illness. Although these factors are informative of risk for higher disability, they are not directly modifiable to increase resilience in individuals with chronic pain, and thereby elevating functioning. The authors underline the importance

of identifying modifiable factors (Elliott et al., 2014). Similarly, in a recent cross-sectional study (Richardson and Jost, 2019) on development of depression and PTSD following early life trauma, the authors emphasize the importance of evaluating psychological flexibility rather than traits or personal attributes as it “seems to be more adaptable to change and is an opportunity for therapeutic intervention.” This is also consistent with Goubert and Trompetter (2017) who emphasizes the importance of focusing on resilience factors that can be changed and used to improve the ability to “ward off, buffer against and recover from disability” for chronic pain patients. In the present study, resilience is conceptualized as a contextual behavioral factor, or a set of behaviors. This conceptualization is of particular clinical relevance, since (operant) behaviors are under contextual control, which implies they can be directly changed. Chronic pain has detrimental effects on functioning for many individuals. The results in the present study support the indirect effects of psychological flexibility – avoidance in particular – and suggest it as relevant target in treatment for chronic pain to improve functioning. This corresponds with previous research showing that avoidance is associated with functioning, and that addressing avoidance in exposure-based interventions can improve functioning (Wicksell et al., 2009; Vlaeyen et al., 2016; Bonnert et al., 2018; Hedman-Lagerlof et al., 2018). The results from the present study also supports the importance of values orientation, and the specific, or incremental, utility of interventions promoting value-oriented behaviors to improve resilience should be addressed in further research.

Furthermore, resilience is more than the absence of disability (Goubert and Trompetter, 2017), which corresponds with the conceptualization in the present study. Future research should further explore the construct of resilience by evaluating the importance of related variables among individuals with chronic pain and distress. For example, future research may benefit from using a longitudinal design to examine the mediating role of psychological flexibility as a resilience factor to allow for analyses of temporal relationships. Also, studies exploring the relationships between subprocesses of psychological flexibility, such as acceptance and present-moment-awareness, as well as

other constructs relevant to the concept of resilience in chronic pain, are warranted and should ideally use behavioral measures such as task performance, in combination with self-report questionnaires. Lastly, experimental studies evaluating the effects of specific interventions on resilience are needed.

While the present study examines the role and function of psychological flexibility as a resilience factor in relation to chronic pain, it is worth noting the transdiagnostic properties of the psychological flexibility model, particularly as there is a need for psychological interventions that better meet the needs of patients with comorbid psychiatric and medical conditions (Barlow et al., 2004; Evans et al., 2005; Merikangas et al., 2007; Löwe et al., 2008). Psychological flexibility is not limited to chronic pain but a psychological skill, or set of skills, that has broad applicability and goes beyond any single mental or physical health condition (Dindo et al., 2017). Psychological inflexibility has been suggested to underlie a wide array of problems, including mental health, behavioral and comorbid complications (Hayes et al., 2006; Kashdan and Rottenberg, 2010). Psychological inflexibility has also been associated with mood- and anxiety disorders (Spinoven et al., 2016). Conversely, improvements in psychological flexibility has been found to predict improvements in depressive symptoms in patients with borderline personality disorder (Berking et al., 2009), improvements in depression and anxiety in patients at risk for vascular disease (Dindo et al., 2015), and improvements in diabetes self-care, blood glucose levels and diabetes-related acceptance in patients with diabetes (Gregg et al., 2007) to mention a few. Although more and larger studies are needed, the empirical support including the present findings, suggest that psychological flexibility is an interesting and important resilience factor across conditions. To address the transdiagnostic nature of psychological flexibility future research should address if the level and implications of psychological flexibility varies across subgroups of patients, for example diagnoses and comorbidities.

In acceptance and commitment therapy (ACT) psychological flexibility, rather than a reduction in symptoms (for example pain and anxiety), is the key therapeutic target. However, ACT is an exposure-based treatment and share several important aspects with other forms of exposure therapy, such as graded exposure based on the fear-avoidance model. Future research should further explore the unique contribution of ACT-specific components such as acceptance and values-orientation, as well as differences and similarities in change processes between exposure-based treatments.

A few limitations should be considered when interpreting the results from the present study. The use of a cross-sectional design prevents any causal inferences. More research is needed to examine psychological flexibility as a resilience factor in longitudinal studies. Also, even though avoidance, value obstruction and value progress are variables relevant to resilience, other behavioral factors of potential importance that impact pain interference and depression such as sleep or social support, were not included. Furthermore, avoidance items from the Psychological Inflexibility in Pain Scale may have some conceptual overlap with pain interference (e.g., "I avoid scheduling activities because of my pain"). The levels

of education as well as the proportion of women (81%) in the study sample is higher than the Swedish average, which may affect the generalizability of results. The sample is self-referred, which may imply limitations to the external validity. However, when compared with samples from a tertiary pain clinic the self-referred sample displays similar levels of pain, distress, and disability (Wicksell et al., 2008a, 2010b; Kemani et al., 2016).

CONCLUSION

Psychological flexibility – in this study assessed as avoidance, value obstruction and value progress – plays a significant role as a resilience factor in the relationship between symptoms and functioning among individuals with chronic pain. Psychological flexibility has been successfully improved in previous clinical trials, and the present findings thus support the utility of this as an important target in treatment.

DATA AVAILABILITY

The datasets generated for this study will not be made publicly available since the ethical permit does not allow sharing of data.

ETHICS STATEMENT

Human Subject Research: This study was reviewed and approved by the Regional Board of Ethics, Stockholm, Sweden. All participants provided written informed consent prior to participation.

AUTHOR CONTRIBUTIONS

CG and JR collected the data. CG, JR, and RW designed the study. JR and VZ prepared the data and performed the preliminary data analysis. CG, JR, and RW performed the final data analyses. CG, JR, and RW prepared the manuscript with valuable contributions from ML, LS, and VZ. All authors approved the final version of the manuscript.

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Pain-Specific Resilience in People Living With HIV and Chronic Pain: Beneficial Associations With Coping Strategies and Catastrophizing

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Objective: Chronic pain is increasingly recognized as a common and disabling problem for people living with HIV (PLWH). In a recent systematic review of psychosocial factors associated with chronic pain in PLWH, it was reported that very few studies to date have examined protective psychological factors that might help mitigate chronic pain for PLWH. The current study examined pain-specific resilience in relation to clinical and experimental pain, as well as pain coping in PLWH and chronic pain. Pain-specific resilience specifically refers to the ability to maintain relatively stable, healthy levels of psychological and physical functioning in the face of ongoing and persistent pain.

Methods: A total of 85 PLWH (mean CD4 = 643; 13% detectable viral load ≥ 200 ; 99% on antiretroviral therapy) who met criteria for chronic pain (>3 consecutive month's duration) were enrolled. Medical records were reviewed to confirm clinical data. All participants provided sociodemographic information prior to completing the following validated measures: Pain Resilience Scale (PRS), Coping Strategies Questionnaire-Revised (CSQ-R), Center for Epidemiologic Studies – Depression Scale (CES-D), and the Brief Pain Inventory – Short Form (BPI-SF). They then completed a quantitative sensory testing battery designed to assess tolerance for painful heat and cold stimuli.

Results: In adjusted multiple regression models controlling for covariates, greater pain-specific resilience was significantly associated with less pain interference ($p = 0.022$) on the BPI-SF, less pain catastrophizing ($p = 0.002$), greater use of distraction ($p = 0.027$) and coping self-statements ($p = 0.039$) on the CSQ-R, as well as significantly greater heat pain tolerance ($p = 0.009$). Finally, results of a parallel multiple mediation model demonstrated that the effect of pain-specific resilience on heat pain tolerance was indirectly transmitted through less pain catastrophizing (95% confidence interval: 0.0042 to 0.0354), but not use of distraction (95% confidence interval: -0.0140 to 0.0137) or coping self-statements (95% confidence interval: -0.0075 to 0.0255).

Conclusion: The findings suggest that pain-specific resilience may promote adaptation and positive coping in PLWH and chronic pain.

Keywords: HIV-human immunodeficiency virus, chronic pain, resilience (psychological), coping, catastrophizing

INTRODUCTION

Due to advancements in antiretroviral therapy (ART), people living with HIV (PLWH) who are connected to care and adhere to their medication regimens can achieve near normal life expectancies (Negin et al., 2012; O'Keefe et al., 2013). However, living longer with HIV is often accompanied by an increased likelihood of developing HIV-associated chronic health conditions (Pitts et al., 2005). One particularly important health condition that affects aging PLWH is chronic pain (Merlin et al., 2012). Estimates suggest that chronic pain may affect over half of all PLWH throughout their lifetimes (Parker et al., 2014). The experience of chronic pain in PLWH often comes at a high cost, such that it significantly and negatively impacts quality of life (Merlin et al., 2013, 2014). Furthermore, treatment of pain in this population can be difficult due to complicating factors including substance use and psychiatric illness (Tsao and Soto, 2009). Pharmacologic pain treatment options, including opioid medications, have limited efficacy for managing chronic pain for many PLWH (Bruce et al., 2017). Psychological approaches for chronic pain in PLWH have demonstrated initial promise (Merlin et al., 2018); however, the full potential of this treatment modality to yield positive outcomes remains underappreciated given the lack of sufficient research to date focused on psychological contributors to chronic pain in PLWH (Scott et al., 2018).

The extant literature addressing psychological contributors to chronic pain in PLWH has primarily focused on vulnerabilities and risk factors for poor outcomes. Scott et al. (2018) recently published a comprehensive review on this topic indicating that depression, psychological distress, post-traumatic stress and substance use were the psychological factors most associated with negative pain outcomes in PLWH. Only a small number of studies thus far have examined the role of protective psychological factors in relation to chronic pain for PLWH. For example, PLWH and chronic pain may possess lower optimism (Simmonds et al., 2005) and self-efficacy for disease management (Parker et al., 2017) and treatment adherence (Berg et al., 2009) relative to PLWH without chronic pain. In a study by Wadley et al. (2016), PLWH and chronic pain reported significantly lower levels of resilience compared to PLWH without chronic pain. However, in this same study resilience was not significantly associated with pain severity or interference for PLWH and chronic pain. Despite these equivocal findings, additional research investigating the association of resilience with chronic pain in PLWH appears warranted for two reasons. First, it is well documented that PLWH are often able to remain resilient despite the many hardships they often face (Dale et al., 2014; Emlet et al., 2017). Second, in non-HIV populations with chronic pain, high resilience has been associated with positive responses to pain, adaptive coping styles, and favorable health care and medication utilization patterns (Karoly and Ruehlman, 2006; Sturgeon and Zautra, 2010).

Resilience is broadly conceptualized as the ability to cope with a crisis or adversity while maintaining positive emotional and physical functioning (Joyce et al., 2018). Numerous measures have been developed to assess resilience as a general psychological

construct, and indeed these measures have proven effective for predicting adaptation to chronic pain (Ong et al., 2010; Ramírez-Maestre et al., 2012). More recently, it has been suggested that a pain-specific measure of resilience is likely to be better suited for studies examining clinical and experimental pain experiences than a general measure of psychological resilience (Slepian et al., 2016). Pain-specific resilience specifically refers to the ability to maintain relatively stable, healthy levels of psychological and physical functioning in the face of ongoing and persistent pain (Ankawi et al., 2017). In the laboratory setting, high pain-specific resilience has been shown to be associated with less sensitivity to experimental pain stimuli during quantitative sensory testing (QST) (Slepian et al., 2016). Further, a series of clinical studies conducted with chronic pain samples found that high pain-specific resilience was associated with better quality of life and lower pain intensity (Ankawi et al., 2017), as well as greater pain self-efficacy and acceptance (Slepian et al., 2018). It remains to be determined whether a measure of pain-specific resilience might also be associated with clinical and experimental pain in a sample of PLWH and chronic pain.

In studies conducted with non-HIV populations, highly resilient individuals with chronic pain have been shown to report greater positive emotions and less pain catastrophizing compared to their less resilient counterparts (Ong et al., 2010; Sturgeon and Zautra, 2013). Similarly, resilient individuals have been found to engage in more adaptive pain coping strategies, which promote efforts to control pain and to function at a high level in spite of pain (Sturgeon and Zautra, 2010). Adaptive pain coping strategies such as distraction and positive coping self-statements facilitate adaptation to chronic pain while also decreasing sensitivity to experimental pain stimuli (Roditi et al., 2009; Malloy and Milling, 2010; Verhoeven et al., 2011). Based upon the existing literature, it stands to reason that pain-specific resilience may be associated with less pain catastrophizing and greater engagement in adaptive pain coping strategies for PLWH and chronic pain, which in turn would be associated with decreased severity of clinical and experimental pain experiences.

The primary objective of the current study was to examine whether pain-specific resilience was associated with reports of clinical and experimental pain, as well as pain catastrophizing and coping strategies, in PLWH and chronic pain. Three distinct hypotheses were tested. (1) High pain-specific resilience would be significantly associated with lower clinical pain severity and interference, as well as greater tolerance for painful cold and heat stimuli during QST. (2) High pain-specific resilience would be significantly associated with less pain catastrophizing and greater use of pain coping strategies (e.g., distraction). (3) Pain catastrophizing and active pain coping strategies would significantly mediate the effect of pain-specific resilience on clinical and experimental pain.

MATERIALS AND METHODS

Study Design Overview

People living with HIV with chronic pain were recruited via posted flyers from a large, urban HIV clinic in Alabama,

United States, that provides comprehensive medical, social, and behavioral services to approximately 3,500 adults (≥ 18 years) living with HIV. Those interested in study participation were assessed for eligibility during an initial telephone screening. Medical records were then reviewed for each prospective participant to assist with eligibility determination. Eligible participants subsequently presented to the laboratory to complete a single study session. At the beginning of the study session resting blood pressure and core body temperature were recorded for each participant. Blood was then taken from each participant for determination of CD4+ count and viral load. Participants completed a QST battery designed to assess tolerance for thermal pain (heat and cold). Following QST, participants completed standardized self-report questionnaires that assessed pain severity and interference, pain-specific resilience, pain coping, and pain catastrophizing. Sociodemographic information was collected from all participants, and this information included age, natal sex, ethnicity/race, educational attainment, and poverty status. Poverty status was determined through adjusting the recorded annual household income by number of occupants through guidelines put forth by the 2017 United States Department of Health and Human Services (U. S. Federal Poverty Guidelines, 2017).

Medical Record Review

Medical record reviews were completed to ascertain rates of psychiatric diagnoses among participants, as well as determine whether participants were actively being prescribed antiretroviral therapy (ART). Medical record review also assisted with determining duration of chronic pain and whether participants were actively being prescribed analgesic medications that could affect reported pain and/or responses to QST, particularly opioids (Nieters et al., 2013). Lastly, medical record review was used to confirm participants' self-reported health history provided during telephone screening. Those PLWH and chronic pain whose medical records corroborated their self-reported health history, and who met study inclusion criteria, were deemed eligible for ongoing participation.

Participants

A total of 91 PLWH and chronic pain were enrolled into this cross-sectional study. Six participants were disqualified from further participation due to the presence of uncontrolled hypertension, which was a contraindication for the completion of QST. This resulted in a final study sample size of 85 PLWH and chronic pain. Study procedures were approved by the local Institutional Review Board and carried out in accordance with guidelines for the ethical conduct of research. Written informed consent was obtained from each participant prior to the study, and the participants were compensated for their participation.

People living with HIV with chronic pain were included in this study if they reported chronic pain that had persisted for at least three consecutive months and was present on at least half the days in the past 6 months (Treede et al., 2015). Additional inclusion criteria were: age ≥ 18 years; no evidence of uncontrolled hypertension (i.e., resting blood pressure $> 150/95$); no circulatory disorders (e.g., Raynaud's disease); no history

of cardiac events, no history of stroke, seizures, or other neurological disorders, no history of metabolic disease, no history of cancer and related treatment, and not currently pregnant. Furthermore, participants were excluded from study participation if they demonstrated signs of acute infection (i.e., core body temperature $> 37.8^\circ\text{C}$), reported any pain-alleviating surgery within the past year, or receipt of any pain intervention treatment within the past month (e.g., steroid injection).

Measures

Quantitative Sensory Testing

Previous studies have found that the relationship between resilience and responses to QST emerges during prolonged exposures to painful stimuli (Pulvers and Hood, 2013). For this reason, the QST battery in this study was designed to specifically assess tolerance for painful thermal stimuli (heat and cold). Heat pain tolerance (HPT_o) refers to the maximum heat stimulus intensity (i.e., temperature, $^\circ\text{C}$) a person is willing to tolerate before discontinuing due to pain. Similarly, cold pain tolerance (CPT_o) refers to the maximum duration (i.e., time in seconds) a person is willing to tolerate a cold stimulus prior to discontinuing due to pain. Participants prescribed analgesics including opioids were not asked to abstain from these medications prior to the completion of QST given that temporary withdrawal could alter pain responses (Mao, 2006).

Heat Pain Tolerance

HPT_o was recorded as the temperature in Celsius at which the participant discontinued the heat stimulus. HPT_o was assessed on participants' ventral forearm using a Medoc Thermal Sensory Analyzer-II (TSA) (Medoc Ltd., Ramat Yishai, Israel) with a 30×30 -mm-diameter thermode in accordance with an ascending method of limits. From a baseline of 32°C , probe temperature increased at a rate of 0.5°C/s until participants responded by pressing a button on the patient response unit to indicate when they were no longer able to tolerate the pain. Three trials of HPT_o were completed separately, and the position of the thermode was altered slightly between trials so that the site of stimulation did not overlap (though it remained on the ventral forearm). The average HPT_o across all three trials was computed for use in statistical analysis.

Cold Pain Tolerance

For the assessment of cold pain tolerance (CPT_o), participants were asked to fully immerse their non-dominant hand up to the wrist in a cold pressor for a maximum of 300 s. The water temperature was maintained at 10°C ($\pm 0.050^\circ\text{C}$) by an ARTIC A25 refrigerated bath with an SC150 immersion circulator (Thermo Fisher Scientific, United States) that constantly circulated the water to prevent local warming around the submerged hand. The water temperature was selected based upon our previous work with other PLWH cohorts demonstrating that 10°C was deemed moderately painful and resulted in the most normally distributed range of CPT_o. Participants were asked to give pain intensity ratings on a 0 (no pain) to 100 (most intense pain possible) numeric rating scale at 30 and 60 s intervals. Participants were told that they could remove their hand from

the cold pressor at any time if the pain became intolerable. The procedure lasted either the full 300 s or until the participant discontinued. Time of hand removal was recorded in seconds and included as an index of CPTo for statistical analysis.

Pain Severity and Interference

The Brief Pain Inventory – Short Form (BPI-SF) is a multidimensional pain scale used to assess the severity of pain and its impact on daily functioning (Tan et al., 2004). The questionnaire is composed of four items asking about pain intensity (worst pain, least pain, average pain, and pain right now) over the past 24 h. There are also seven items that assess the degree to which pain interferes with functioning in the following domains: general activity, mood, walking ability, normal work, relations with other people, sleep, and enjoyment of life. The BPI-SF yields two overall scores: a pain severity score and a pain interference score. The pain severity score is the average of the four items asking about worst, least, average, and current pain. Each item is scored from 0 (no pain) to 10 (worst imaginable pain). The pain interference score is the average of the nine items addressing functional impairment. Each item is scored from 0 (does not interfere) to 10 (completely interferes). Higher scores suggest great pain severity and interference. Overall, the BPI-SF in this study had excellent internal consistency (Cronbach's $\alpha = 0.95$).

Pain-Specific Resilience

The Pain Resilience Scale (PRS) is a 14-item assessment of resilience in the presence of intense or prolonged pain. The assessment has 2 subscales to measure specific domains of resilience: behavioral perseverance and cognitive/affective positivity (Slepian et al., 2016; Ankawi et al., 2017). The behavioral perseverance subscale examines an individual's ability to continue engaging in behaviors or activity when experiencing pain. The cognitive/affective positivity subscale examines an individual's ability to maintain positive thoughts and manage negative thoughts or emotions while in pain. Each item is scored from 0 (not at all) to 4 (all the time) to determine the degree to which individuals engage in resiliency resources. The total PRS score results from the summation of response to all 14 items. Higher scores are suggestive of greater pain related resilience. The PRS used in this study possessed excellent internal consistency (Cronbach's $\alpha = 0.94$).

Pain Coping and Catastrophizing

The Coping Strategies Questionnaire-Revised (CSQ-R) is a 27-item assessment that was utilized to assess participants' use of cognitive strategies to cope with pain, as well as pain catastrophizing (Rosenstiel and Keefe, 1983). The CSQ-R includes the following subscales representing six cognitive domains: distraction (five-items), ignoring pain sensations (five-items), distancing oneself from pain (four-items), coping self-statements (four-items), praying/hoping (three-items), and catastrophizing (six-items). Each item is scored from 0 (never do that) to 6 (always do that) to indicate how frequently the strategy is engaged in response to pain. Each subscale is scored separately, and higher scores indicate greater engagement in that respective

cognitive domain. The CSQ-R in this study had adequate internal consistency (Cronbach's $\alpha = 0.73$).

Depression

Depressive symptoms were assessed using the Center for Epidemiological Studies Depression Scale (CES-D). This 20-item measure assesses the frequency of experiencing depressive symptoms over the past week (0 – never or rarely, to 3 – most of the time/all the time). Symptoms of depression measured by the CES-D include negative mood, guilt/worthlessness, helplessness/hopelessness, psychomotor retardation, loss of appetite, and sleep disturbance (Radloff, 1977). This measure has been shown to be reliable and valid in general populations, as well as HIV and chronic pain populations (Geisser et al., 1997; Natamba et al., 2014). Responses are summed (0–60), with higher scores indicating greater severity of depression. The CES-D measure used in the current study had good internal consistency (Cronbach's $\alpha=0.88$).

CD4 and Viral Load

Blood was collected from each participant at the beginning of the study session and sent to the local diagnostics laboratory for quantification of CD4 helper T-cell count and viral load. Absolute CD4 helper T-cell count was quantified as cells/microliter of blood, while viral load was quantified as viruses/microliter. Participants with 200 viruses/microliter of blood or greater were considered to be “detectable.” CD4 and viral load reflect immune health and response to ART therapy, respectively. Each was included in this study to assess whether these aspects of HIV infection were associated with clinical and/or experimental pain.

Data Organization and Analysis

All data were analyzed using SPSS, version 25 (IBM; Chicago, IL, United States). All participants provided complete demographic (e.g., sex, age) and QST data; however, a small portion of missing data existed for one or more key study variables such as pain-specific resilience and pain coping ($\leq 5\%$ of the total data comprising each measure). Data appeared to be missing at random. A simple data imputation method was completed using the macro for Hot Deck imputation (Myers, 2011). This data imputation method is well validated and accepted in the statistical community and resulted in complete study data for each of the 85 study participants.

Descriptive data for the sample are presented as percentages or as means and standard deviations. Differences across categorical variables were assessed using chi-square tests, while differences on continuous variables were assessed using analysis of variance (ANOVA). Zero order relationships among all study variables were assessed using Pearson correlations. To assess the unique relationships of pain-specific resilience with pain severity and interference, HPTo and CPTo, as well as pain coping and catastrophizing, a series of linear multiple regressions was completed controlling for selected covariates. The PROCESS macro (model 4) created and described by Hayes (2013) for obtaining 95% bootstrapped confidence interval with 5,000

resamples was utilized to test whether catastrophizing and/or any of the pain coping strategies significantly mediated the associations between pain-specific resilience and pain, including pain severity, pain interference, HPTo, and CPTo.

RESULTS

Participant Characteristics

Descriptive characteristics for the 85 study participants are presented in **Table 1**. The mean age of the sample was 49 years. The study population was comprised of 67% men and 33% women. The majority of the study sample was non-Hispanic Black (74%), and lived below the poverty line (85%). The mean CD4 count was 643 cells/mm³, 13% had a detectable viral load, and 99% were actively prescribed antiretroviral therapy. Seventeen percent of the study sample were prescribed opioid medications for pain. The most frequently reported locations of chronic pain were low back/hips (46%), legs/feet (25%), widespread (2 + sites) (20%), arms/hands (6%), head (2%), and neck/shoulders (1%). Medical records indicated that 24% of the sample had a pain duration of >3 months but <1 year, 25% >1 year but <5 years, 23% >5 years but <10 years, and 28% >10 years. Average pain severity over the past 24 h was 5.8, while pain interference was 4.5 on the 0–10 numeric rating scale of the BPI-SF.

Bivariate Associations and Selection of Covariates

Zero-order Pearson correlations among continuously measured variables are presented in **Table 2**. Greater pain-specific resilience was significantly correlated with less depressive symptoms ($p = 0.011$) and less pain catastrophizing ($p < 0.001$), greater use of distraction ($p = 0.007$) and coping self-statements ($p = 0.013$), less clinical pain severity ($p = 0.042$) and pain interference ($p < 0.001$), as well as greater HPTo ($p = 0.014$) and CPTo ($p = 0.041$). Greater depressive symptom severity was significantly correlated with increased pain interference ($p < 0.001$) and more pain catastrophizing ($p < 0.001$). Greater pain catastrophizing was significantly correlated with greater clinical pain severity ($p = 0.034$) and pain interference ($p = 0.001$), as well as diminished HPTo ($p = 0.003$) and CPTo ($p = 0.031$). Longer duration of pain was significantly correlated with greater clinical pain severity ($p < 0.001$) and pain interference ($p = 0.034$), as well as less pain-specific resilience ($p = 0.038$). Results from a series of one-way ANOVAs revealed that participants actively prescribed opioids tended to report greater pain interference ($p = 0.069$) than those not receiving opioid medication. A significant sex difference was observed for HPTo ($p = 0.013$), such that males demonstrated diminished HPTo compared to females. Age, race, poverty status, and HIV clinical characteristics including CD4+ and detectable viral load, were not significantly associated with any of the key variables of interest.

Prior to completing the linear multiple regression analyses below, specific covariates were chosen based upon whether

TABLE 1 | Participant characteristics ($N = 85$).

Variable	Mean (SD) or Count (%)	Range
Demographic characteristics		
Age—Years	49 (8.3)	26–67
Sex		
Males	57 (67%)	
Females	28 (33%)	
Race		
non-Hispanic Black	63 (74%)	
non-Hispanic White	16 (19%)	
American Indian	1 (1%)	
Multiracial	5 (6%)	
Poverty		
Below Poverty Line	72 (85%)	
Above Poverty Line	13 (15%)	
Clinical characteristics		
CD4	643 (324)	62–2,491
Viral load (≥ 200 copies/mL)		
Undetectable	74 (87%)	
Detectable	11 (13%)	
Anti-Retroviral Therapy (ART)		
Actively Prescribed	84 (99%)	
Not Prescribed	1 (1%)	
Opioids		
Actively Prescribed	14 (17%)	
Not Prescribed	71 (83%)	
Depressive symptoms		
CES-D – Depressive Symptoms	21.2 (11.5)	0–53
Resilience		
PRS	36.35 (13.55)	0–56
Coping		
CSQ-R – Catastrophizing	2.5 (1.4)	0–6
CSQ-R – Distraction	2.6 (1.6)	0–6
Pain duration		
>3 months but <1 year	20 (24%)	
>1 year but <5 years	22 (25%)	
>5 years but <10 years	19 (23%)	
>10 years	24 (28%)	
Clinical pain severity and interference		
BPI-SF – Pain Severity	5.8 (2.4)	0–9.8
BPI-SF – Pain Interference	4.5 (2.8)	0–10
Experimental pain		
HPTo (°C)	48.1 (2.2)	38.6–50.5
CPTo (seconds)	163.9 (111.7)	12–300

CES-D, Center for Epidemiologic Studies-Depression Scale; PRS, Pain-Specific Resilience Scale; CSQ-R, Coping Strategies Questionnaire-Revised; BPI-SF, Brief Pain Inventory-Short Form; HPTo, heat pain tolerance; CPTo, cold pain tolerance.

they demonstrated significant associations with key variables of interest including clinical pain severity and interference, HPTo and CPTo, as well as pain coping strategies and pain catastrophizing. Participants' sex, opioid medication prescription, depressive symptom severity, and chronic pain duration were included as covariates in all study models displayed in **Tables 3–5**. The reported answer to “Pain Right Now” on the BPI-SF was included as a covariate in all analyses (except for the

TABLE 2 | Zero-order pearson correlations.

Variable	1	2	3	4	5	6	7	8	9	10	11	12
(1) PRS	–											
(2) CES-D	–0.279**	–										
(3) CSQ-R Catastrophizing	–0.453**	0.471**	–									
(4) CSQ-R Distancing	0.098	0.030	0.362**	–								
(5) CSQ-R Distraction	0.293**	–0.264*	0.036	0.486**	–							
(6) CSQ-R Ignoring	0.021	–0.037	0.313**	0.715**	0.525**	–						
(7) CSQ-R self-statements	0.267*	–0.285**	–0.094	0.215*	0.638**	0.404**	–					
(8) CSQ-R Praying/Hoping	–0.067	–0.038	0.141	0.162	0.282**	0.247*	0.304**	–				
(9) BPI-SF pain severity	–0.221*	0.076	0.230*	0.134	–0.072	0.082	–0.138	0.073	–			
(10) BPI-SF Pain Interference	–0.388**	0.374**	0.343**	0.023	–0.163	–0.066	–0.150	0.029	0.646**	–		
(11) HPTo	0.266*	–0.047	–0.317**	–0.072	0.021	–0.051	0.067	–0.144	–0.061	–0.205	–	
(12) CPTo	0.222*	–0.021	–0.235*	–0.120	0.012	–0.038	0.206	–0.080	–0.144	–0.035	0.212	–
(13) Pain duration	–0.226*	0.011	0.211	0.036	0.004	0.058	0.123	0.041	0.442**	0.264*	–0.028	–0.031

* $p < 0.05$, ** $p < 0.01$. PRS, Pain Resilience Scale; CES-D, Center for Epidemiological Studies Depression Scale; CSQ-R, Coping Strategies Questionnaire-Revised; BPI-SF, Brief Pain Inventory-Short Form; HPTo, Heat Pain Tolerance; CPTo, Cold Pain Tolerance.

TABLE 3 | Multiple regressions models demonstrating associations with pain interference and clinical pain severity.

Variables	BPI-SF Pain interference				BPI-SF Clinical pain severity			
	R^2	B	SE B	b	R^2	B	SE B	b
Variables	0.522**				0.251**			
Sex ^a		–0.380	0.477	–0.064		–0.199	0.510	–0.040
Opioid prescription ^b		0.651	0.613	0.086		0.214	0.655	0.034
CES-D		0.065	0.020	0.264**		0.006	0.022	0.031
Pain Duration		–0.047	0.125	–0.034		0.485	0.122	0.418**
Pain Right Now		0.555	0.088	0.555**		–	–	–
Pain-Specific Resilience		–0.041	0.018	–0.199*		–0.020	0.019	–0.116

^aCoded Variable (1 = Males, 2 = Females); ^bCoded Variable (1 = Prescribed, 2 = Not Prescribed). * $p < 0.05$; ** $p < 0.01$. BPI-SF, Brief Pain Inventory-Short Form. CES-D, Center for Epidemiologic Studies Depression Scale.

TABLE 4 | Multiple regressions models demonstrating associations with pain catastrophizing, distraction, and coping self-statements.

Variables	CSQ-R Catastrophizing				CSQ-R Distraction				CSQ-R self-statements			
	R^2	B	SE B	b	R^2	B	SE B	b	R^2	B	SE B	b
Variables	0.355**				0.133*				0.182*			
Sex ^a		–0.052	0.281	–0.017		0.038	0.361	0.011		0.195	0.301	0.068
Opioid prescription ^b		0.001	0.361	0.001		–0.377	0.465	–0.089		–0.359	0.388	–0.098
CES-D		0.047	0.012	0.379**		–0.024	0.016	–0.174		–0.023	0.013	–0.191
Pain Duration		0.078	0.074	0.111		0.062	0.095	0.080		0.165	0.079	0.247*
Pain Right Now		0.032	0.052	0.063		–0.077	0.067	–0.012		–0.072	0.056	–0.149
Pain-Specific Resilience		–0.033	0.010	–0.309**		0.030	0.013	0.257*		0.023	0.011	0.233*

^aCoded Variable (1 = Males, 2 = Females); ^bCoded Variable (1 = Prescribed, 2 = Not Prescribed). * $p < 0.05$; ** $p < 0.01$. CSQ-R, Coping Strategies Questionnaire – Revised. CES-D, Center for Epidemiological Studies Depression Scale.

analysis of pain severity) to control for individual differences in clinical pain severity at the time of study participation.

Associations With Clinical Pain Severity and Pain Interference

Table 3 displays the results of two multiple regression models that assessed whether pain-specific resilience was uniquely and

significantly associated with clinical pain severity and pain interference reported on the BPI-SF. On the left of Table 3, results revealed that the overall model accounted for a significant 52% of the variance in pain interference [$F(6, 78) = 14.207$, $p < 0.001$]. Greater pain-specific resilience was significantly associated with less pain interference in PLWH and chronic pain even after controlling for covariates ($\beta = -0.199$, $p = 0.022$). As seen on the right of Table 3, the overall model did account

TABLE 5 | Multiple regressions models demonstrating associations with heat pain tolerance (HPTo) and cold pain tolerance (CPTo).

Variables	HPTo				CPTo			
	<i>R</i> ²	B	SE B	b	<i>R</i> ²	B	SE B	b
Variables	0.157*				0.081			
Sex ^a		−1.350	0.491	−0.292**		−24.692	26.216	−0.104
Opioid prescription ^b		−0.119	0.632	−0.020		31.685	33.739	0.106
CES-D		0.006	0.021	0.030		0.240	1.127	0.025
Pain duration		0.118	0.129	0.110		4.238	6.891	0.077
Pain Right Now		−0.029	0.091	−0.038		−4.726	4.865	−0.119
Pain-Specific Resilience		0.049	0.018	0.302**		1.882	0.969	0.228

^aCoded Variable (1 = Males, 2 = Females); ^bCoded Variable (1 = Prescribed, 2 = Not Prescribed). * $p < 0.05$; ** $p < 0.01$. CES-D, Center for Epidemiologic Studies Depression Scale.

for a significant portion of variance in clinical pain severity [$F(5, 79) = 4.320, p = 0.002$]. However, pain-specific resilience was not significantly associated with clinical pain severity after controlling for covariates ($\beta = -0.116, p = 0.283$).

Associations With Pain Coping Strategies and Pain Catastrophizing

A series of three additional multiple linear regressions were conducted to assess whether pain-specific resilience was significantly associated with pain catastrophizing, as well as the use of two pain coping strategies (distraction, coping self-statements). Overall, the multiple regression model presented to the left in **Table 4** accounted for a significant 36% of the variance in pain catastrophizing [$F(6, 78) = 7.143, p < 0.001$]. Results revealed that greater pain-specific resilience was significantly associated with less catastrophizing about pain ($\beta = -0.309, p = 0.002$) controlling for covariates. The overall multiple regression model presented in the middle of **Table 4** accounted for a significant 13% of the variance in use of distraction [$F(6, 78) = 2.329, p = 0.050$]. Greater pain-specific resilience was found to be significantly associated with more frequent use of distraction as a pain coping technique ($\beta = 0.257, p = 0.027$). As demonstrated on the right side of **Table 4**, the overall multiple regression model accounted for a significant 18% of the variance in coping self-statements [$F(6, 78) = 2.891, p = 0.013$]. Pain-specific resilience was significantly associated with coping self-statements after controlling for covariates ($\beta = 0.233, p = 0.039$).

Associations With HPTo and CPTo

Results of two multiple regression models examining associations with HPTo and CPTo are presented in **Table 5**. As shown on the left of **Table 5**, the overall model accounted for a significant 16% of the variance in HPTo [$F(6, 78) = 2.426, p = 0.033$]. Furthermore, results revealed that pain-specific resilience was significantly associated with HPTo, such that participants with greater pain-specific resilience demonstrated higher HPTo ($\beta = 0.302, p = 0.009$). On the right side of **Table 5** it can be seen that the overall model did not account for a significant portion of the variance in CPTo [$F(6, 78) = 1.149, p = 0.342$]. Although pain-specific resilience was not significantly

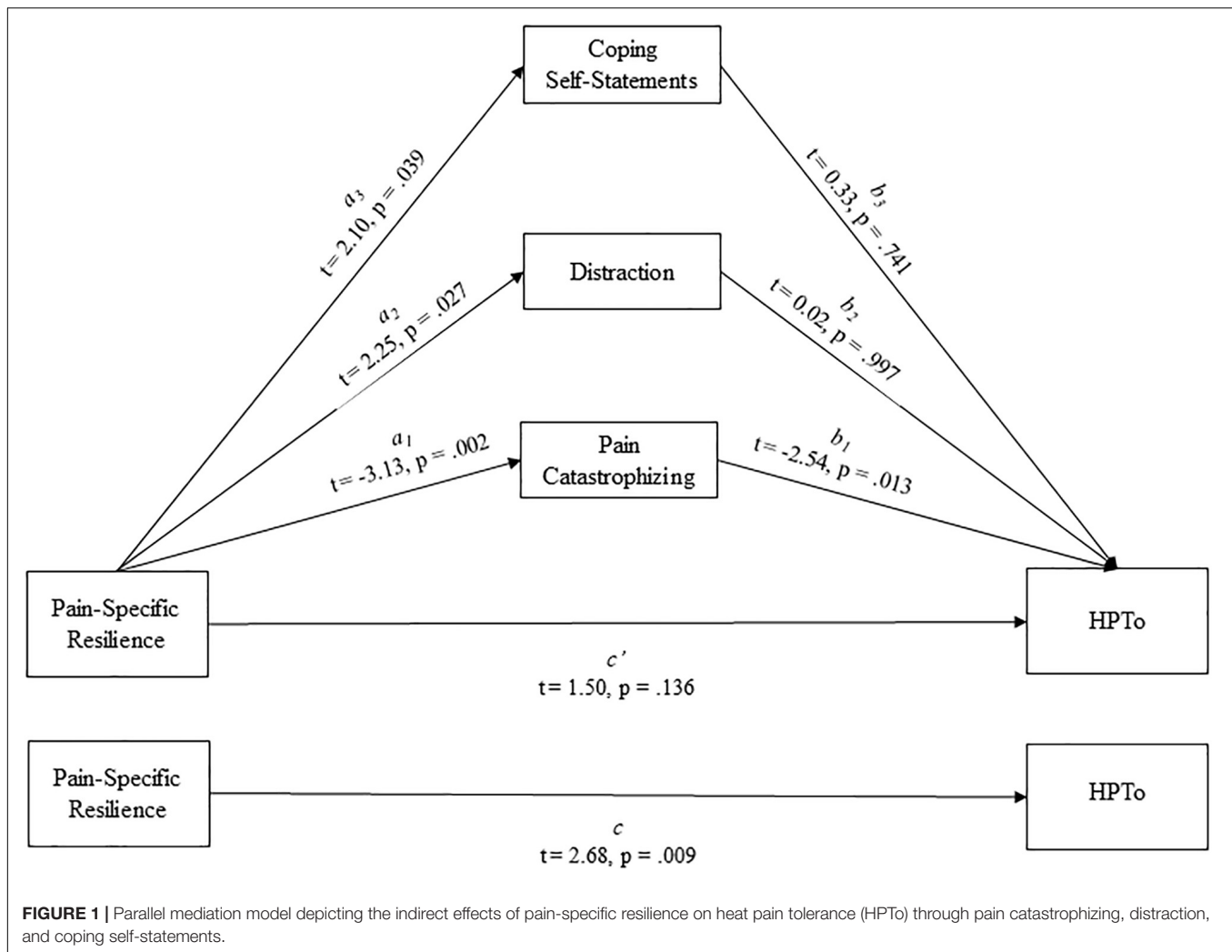
associated with CPTo ($\beta = 0.228, p = 0.056$) after controlling for covariates, there was a trend toward significance. The observed power for the multiple regression model examining associations with CPTo was 0.497.

Parallel Multiple Mediation

Whether pain catastrophizing, distraction, and/or coping self-statements significantly mediated the effect of pain-specific resilience on HPTo was examined utilizing a parallel multiple mediation model with bias-corrected bootstrapped confidence intervals (**Figure 1**). Results indicated that this overall model accounted for a significant 23% of the variance in HPTo ($R^2 = 0.229, p = 0.015$). It was revealed that pain-specific resilience was indirectly related to HPTo (i.e., mediated) through catastrophizing (indirect effect = 0.0168, 95% CI: 0.0042 to 0.0354), but not through distraction (indirect effect = 0.0001, 95% CI: −0.0140 to 0.0137) or coping self-statements (indirect effect = 0.0017, 95% CI: −0.0075 to 0.0255). More specifically, the higher HPTo shown by those with greater pain-specific resilience was partly accounted for by their less frequent engagement in pain catastrophizing. Additional parallel multiple mediation models were completed and demonstrated that neither catastrophizing, distraction, nor coping self-statements significantly mediated the effect of pain-specific resilience on CPTo or pain interference.

DISCUSSION

Resilience to pain is a conceptually complex psychological phenomenon. The previous work of Sturgeon and Zautra (2010) has provided much needed clarity for this topic by addressing important resources and mechanisms that promote pathways to resilience for those with chronic pain. Qualities of an individual and his/her social world such as optimism, perseverance, high socioeconomic status, and a helpful social support network represent resilience resources that increase the likelihood of adaptive responses to chronic pain. Resilience mechanisms refer to the helpful thoughts, affects, and behaviors utilized by individuals with chronic pain when confronting adversity. Resilience resources promote the utilization of beneficial resilience mechanisms, and together these resources



and mechanisms interactively influence resilient responses to chronic pain. In this study, the pain-specific resilience measure (Slepian et al., 2016) is arguably an index of resilience mechanisms. This is because its two constituent subscales assess the ability to continue engaging in behaviors or activity when experiencing pain (behavioral perseverance subscale), as well as the ability to maintain positive thoughts and manage negative thoughts or emotions while in pain (cognitive/affective positivity subscale). In this regard, our study demonstrates that PLWH and chronic pain possess wide ranging pain-specific resilience mechanisms that confer either relative protection or vulnerability to the deleterious effects of chronic pain. Those with high pain resilience are perhaps best equipped to cope with chronic pain.

The goal of this study was to investigate the extent to which pain-specific resilience was associated with the following aspects of clinical and experimental pain in a sample of PLWH: (1) engagement in adaptive pain coping strategies, (2) pain catastrophizing, (3) pain interference, and (4) tolerance for painful stimuli delivered in a laboratory setting. As hypothesized, findings suggest that PLWH and chronic pain who demonstrate greater pain-specific resilience may be more

likely to engage in adaptive pain coping strategies by specifically utilizing distraction techniques and coping self-statements, while concurrently refraining from catastrophizing about their chronic pain. Additionally, greater pain-specific resilience may mitigate the extent to which chronic pain interferes with daily living and the quality of life of PLWH. Similarly, our findings suggest that greater pain-specific resilience promotes the ability of PLWH and chronic pain to tolerate a painful heat stimulus, an effect which may be attributed to less engagement in pain catastrophizing. Our results are generally consistent with previous studies of chronic pain patients without HIV. For example, greater resilience was associated with better physical functioning and less pain interference in individuals with knee osteoarthritis (Wright et al., 2008). Furthermore, other positive psychological factors associated with resilience such as optimism are indirectly associated with less experimental pain sensitivity via decreased pain catastrophizing (Goodin et al., 2013; Pulvers and Hood, 2013).

Bivariate analyses initially revealed that pain-specific resilience was significantly correlated with diminished clinical pain severity and greater tolerance for a cold pain stimulus; however,

these associations were no longer statistically significant after adjustment for covariates in the multiple regression models. It appears that the adjusted multiple regression model examining the unique association between pain-specific resilience and CPTo may have lacked sufficient statistical power to detect a significant association. That the p -value was 0.056 and the observed power was 0.497 suggests that with a larger sample size of PLWH and chronic pain, it is very likely the association between greater pain-specific resilience and greater CPTo would have remained significant even after adjustment for covariates. However, future research with a larger sample of PLWH and chronic pain will be necessary to confirm this assertion. In the multiple regression model examining clinical pain severity, the strongest association was with duration of pain. Furthermore, PLWH and chronic pain with the longest pain duration (e.g., > 10 years) also reported the lowest levels of pain-specific resilience. These findings suggest that PLWH and chronic pain who have been dealing with their pain for many years may be at greatest risk for poor pain outcomes due to a lack of pain-specific resilience mechanisms.

As a matter of clinical importance, a logical extension of our work would be to address the question of whether a tailored cognitive and behavioral intervention might promote resilience mechanisms, specifically for those PLWH with long duration of pain and who demonstrate low pain resilience. Previous intervention development efforts support the likelihood of this possibility. For example, Padesky and Mooney's (2012) four-step, strengths-based cognitive-behavioral therapy model was designed to help individuals become more resilient by helping them identify and utilize their personal strengths in ways that promote self-efficacy, positive emotions, and better regulation of negative emotions in response to stress. This strengths-based approach to increasing resilience through cognitive-behavior therapy has not yet been applied to PLWH and chronic pain, to the best of our knowledge. Recent and ongoing work conducted by Merlin et al. (2018) suggests that a tailored and evidence-based behavioral intervention may facilitate adaptation to chronic pain in PLWH by promoting pain-specific resilience mechanisms. In their 12-session pain self-management intervention, "Skills TO Manage Pain (STOMP)," PLWH and chronic pain learn specific skills for coping with stress, building self-efficacy and worth, remaining appropriately engaged in valued activities of daily living. Acquisition of these resilience-building mechanisms is completed in group-based sessions that foster peer support around living with HIV and chronic pain, a resilience resource. Whether an ongoing clinical trial of STOMP will improve the chronic pain experiences of PLWH by promoting pain-specific resilience mechanisms has yet to be determined (clinicaltrials.gov NCT03692611). On balance, future psychological interventions that target pain-specific resilience may play an important role in determining whether PLWH effectively manage and cope with their chronic pain. Cognitive and behavioral-based psychological interventions designed to promote adaptive coping and resilience and specifically tailored for HIV populations may provide patients with the ability to ameliorate distress, reduce pain perception, and increase quality of life (Miller et al., 2019).

This study has several limitations that warrant consideration. First, the cross-sectional design of this study limits our

ability to form conclusions regarding whether pain-specific resilience causally yields protective effects against chronic pain in PLWH. Similarly, this study could not address the question of whether pain-specific resilience prevents the worsening of pain interference for PLWH and chronic pain over time. Future longitudinal research will be necessary to better appreciate the mechanistic pathways and processes whereby pain-specific resilience yields its pain protective effects. Second, PLWH in this study completed study questionnaires after completion of the QST battery, not prior. Timing of questionnaire completion could potentially affect how participants respond. Third, our study was not designed with specific focus on possible determinants of pain-specific resilience. Although it appears that some PLWH and chronic pain in this study were especially pain resilient, we cannot meaningfully address how such pain-specific resilience manifested. Future theoretical and applied research seems warranted in both HIV and non-HIV populations to better establish a framework for understanding how pain-specific resilience develops, can be modified, and ultimately protects against the deleterious effects of chronic pain. Whether the previous pain resilience framework previously put forth by Sturgeon and Zautra (2010) applies specifically to PLWH would be a worthwhile investigation. Fourth, the vast majority of our study sample was non-Hispanic Black PLWH who lived below the poverty line. While these sociodemographic factors closely align with the population most affected by HIV in Alabama, and the HIV clinic from which study participants were recruited, the generalizability of our findings may be limited. Future studies should examine whether this study's findings can be replicated among larger, more diverse populations of PLWH and chronic pain. Findings from our study may prove to be even stronger in subsequent research conducted with PLWH and chronic pain who possess more and better resilience resources such as higher socioeconomic status and deeper social support networks among other. Lastly, we did not specifically assess behavioral domains of pain coping such as exercise and pursuit of hobbies. The CSQ-R measure incorporated in this study asks exclusively about the cognitive domains of pain coping (e.g., distraction, use of self-statements). Therefore, at this time it remains unclear the extent to which pain-specific resilience might be associated with greater utilization of behavioral pain coping strategies in PLWH and chronic pain. Despite these limitations, the results of this study contribute to stronger understanding of how pain-specific resilience might mitigate the deleterious effects of pain for PLWH and chronic pain.

The scant amount of research to date addressing psychological contributors to chronic pain in HIV has largely focused on factors such as pain catastrophizing and depression, which confer vulnerability to negative pain-related outcomes (Scott et al., 2018). More recent studies have begun to also address protective psychological factors that promote resilience and positively influence pain-related outcomes for PLWH (Penn et al., 2019). This study helps to address a gap in the current literature pertaining to the potential impact of positive psychological factors on the experience of chronic pain in PLWH. Individuals with a high degree of pain-specific resilience are generally able to maintain behavioral engagement and appropriately regulate

their emotions and cognitions despite prolonged or intense pain (Ankawi et al., 2017). Our findings are consistent with this sentiment given that pain-specific resilience was significantly associated with less pain interference and catastrophizing, more frequent use of adaptive pain coping strategies, and higher tolerance for a painful experimental heat stimulus in a sample of PLWH and chronic pain. Low pain-specific resilience may be an important treatment target in the future for psychologically based chronic pain management. It is encouraging that our findings suggest PLWH and chronic pain may experience improved chronic pain outcomes through the strengthening of pain-specific resilience.

DATA AVAILABILITY

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of the National Institutes of Health for the responsible conduct of human subjects research with written informed consent from all subjects. All subjects gave written

informed consent in accordance with the Declaration of Helsinki. The protocol was approved by the Institutional Review Board at the University of Alabama at Birmingham.

AUTHOR CONTRIBUTIONS

All authors equally contributed to the conceptualization and design of the study. Further, each author was instrumental in helping to analyze the data and to interpret the results, as well as drafting the manuscript and providing the important edits that ultimately culminated with this submission.

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Investigating How Parental Instructions and Protective Responses Mediate the Relationship Between Parental Psychological Flexibility and Pain-Related Behavior in Adolescents With Chronic Pain: A Daily Diary Study

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Background: Parental behavior can influence how well adolescents cope with chronic pain. Previous research has largely focused on how parents negatively impact adolescent functioning. Yet more recent work suggests that parents – and particularly parental psychological flexibility – can foster better adolescent pain-related functioning. In this study we examined if parental protective responses and instructions to engage in activities in the presence of pain mediate the impact of parental psychological flexibility and acceptance of adolescent pain on adolescents' daily pain-related behavior.

Method: Fifty-six adolescents with chronic pain ($M_{\text{age}} = 14.5$ years, 86% girls) and one of their parents (93% mothers) were recruited at initial evaluation at two pediatric pain clinics in the US. Parents completed baseline questionnaires assessing psychologically flexible parenting and acceptance of adolescent pain. Next, parents and adolescents completed a 14-day self-report diary assessing adolescent activity-avoidance and activity-engagement in the presence of pain (adolescent report), and parental protective responses and instructions for their adolescent to engage in activities (parent report).

Results: Psychologically flexible parenting and acceptance of adolescent pain in parents were indirectly related to lower daily adolescent activity-avoidance, via their negative association with daily parental protective responses. Positive associations also emerged between baseline psychologically flexible parenting and overall levels of adolescent activity-engagement via its negative association with overall levels of parental protectiveness across the 14-day period. Psychologically flexible parenting and parental acceptance of adolescent pain were also indirectly related to daily decreases in adolescent activity-avoidance via their association with daily increases in parental

activity-engagement instructions. These baseline parental resilience factors were also positively related to overall levels of parental engagement instructions, a route via which an indirect association with both higher overall activity-engagement as well as higher overall activity-avoidance in the adolescent was observed.

Conclusion: Our findings suggest an (indirect) adaptive role of parental psychological flexibility on adolescent daily pain-related behavior via its impact on parental protective behavior. If our findings replicate, they would suggest that these parental behaviors could be targeted in pain treatments that include both adolescents and their parents. Future research could further examine the impact of parental instructions on pain-related behavior in adolescents with chronic pain.

Keywords: parental psychological flexibility, adolescent chronic pain, adolescent pain-related behavior, parental protective behavior, parental instructions

INTRODUCTION

Approximately one in five children and adolescents experience chronic pain (King et al., 2011), which frequently affects their physical, emotional, and social functioning (Palermo, 2000; Hunfeld et al., 2001; Palermo and Eccleston, 2009). Growing evidence suggests that parents may inadvertently and adversely impact their adolescent's functioning in the presence of that pain (Logan et al., 2012; Simons et al., 2015; Chow et al., 2016). Two lines of thought have emerged to explain how parents exert such a negative influence. The first argues that parents tend to emit protective behaviors (e.g., keeping the child home from school) when faced with their adolescent in pain, and that these behaviors directly result in heightened adolescent avoidance of pain-related activities (Palermo and Chambers, 2005; Goubert and Simons, 2013). It is this heightened and persistent avoidance which is assumed to increase risk of disability (Asmundson et al., 2012; Simons and Kaczynski, 2012). A second line of thought argues that adolescent behavior is indirectly influenced by how parents think, act, and feel. For instance, parental fear and catastrophizing about adolescent pain can indirectly influence how much their adolescent avoids pain-related activities through their impact on both parent (i.e., parent pain avoidance) and adolescent psychosocial responses to pain (e.g., adolescent pain-related fear and catastrophizing) (Vowles et al., 2010; Simons et al., 2015). Observational learning processes have been proposed to explain these indirect influences from parent to adolescent functioning (see Goubert et al., 2011; Goubert and Simons, 2013). However, *instructional learning processes* may be an alternative route through which parents may exert an indirect influence upon their adolescent's functioning. Verbal information – namely – the rich variety of pain-related instructions and rules communicated from parent to adolescent can exert a powerful influence on adolescents' pain-related behavior even in the absence of direct pain experiences (for more details, see Beeckman et al., 2019a). Parents are uniquely positioned to provide frequent verbal information regarding the potential positive or aversive outcomes of engaging in, or avoiding pain-related activities. Although theoretical work on the relationship

between verbal processes and pain has started to emerge (e.g., Bennett et al., 2015; Maeda et al., 2018; Beeckman et al., 2019a), no empirical work has examined if parental verbal instructions guide adolescent pain-related behavior, and ultimately, their functioning over time.

As we mentioned above, most work on the role of parents in the context of pediatric chronic pain has focused on their *maladaptive* influence. Yet parents might also foster resilient functioning in adolescents (i.e., “effective functioning despite stressful circumstances [such as chronic pain]”; Karoly and Ruehlman, 2006; Sturgeon and Zautra, 2010; Goubert and Trompetter, 2017). Admittedly, research supporting this idea is sparse. But several recent studies have started to identify specific parental factors that may increase adaptive functioning in adolescents with chronic pain (e.g., adaptive parenting; see also Cousins et al., 2015; Goubert and Trompetter, 2017; Feinstein et al., 2018; Ross et al., 2018). Parental psychological flexibility is thought to represent one such factor (e.g., McCracken and Gauntlett-Gilbert, 2011; Smith et al., 2015; Wallace et al., 2015; Timmers et al., 2019). Psychological flexibility refers to “being aware of, and open to unwanted and uncontrollable experiences (e.g., seeing your child suffering with chronic pain), while still having the ability to act in line with broader life values (e.g., being an encouraging parent)” (Hayes et al., 2006; McCracken and Gauntlett-Gilbert, 2011; McCracken and Morley, 2014; Vowles et al., 2014). Parents can show psychological flexibility in how they parent in general, or more specifically, for instance, in how they navigate thoughts and feelings that emerge when confronted with their adolescent's pain. Parental acceptance of adolescent pain is a sub-component of psychological flexibility and the one that has garnered the greatest attention in the pediatric pain literature (e.g., Smith et al., 2015; Feinstein et al., 2018). In contrast, the six processes that constitute psychological flexibility in parents (i.e., *acceptance, defusion, being present, self-as-context, values-based action, committed action*; see also McCracken and Morley, 2014) and the role of psychological flexibility in parent-child interactions in particular (e.g., Greene et al., 2015; Wallace et al., 2015; Brassell et al., 2016) have received far less attention. Several studies indicate that parental psychological flexibility in the context of adolescent chronic

pain is associated with lower levels of adolescent disability and depression (McCracken and Gauntlett-Gilbert, 2011; Smith et al., 2015; Wallace et al., 2015). Such work suggests that the relationship between parental psychological flexibility and adolescent functioning may itself be mediated by lower parental protective responses and higher adolescent acceptance of pain (Smith et al., 2015; Timmers et al., 2019). We build on this prior work and introduce another possibility: the verbal information that parents communicate to their adolescent (e.g., “It is important that you engage in activities that you value even though you are in pain”) may represent yet another way via which parental psychological flexibility exerts an influence on adolescent functioning.

With this in mind, the current study examined – using a diary design – the relations between parents’ psychological flexibility in the interactions with their adolescent in general and acceptance of adolescent pain, and daily activity-avoidance and activity-engagement in adolescents with chronic pain. First, we expected that higher psychologically flexible parenting and higher parental acceptance of adolescent pain would be indirectly related to lower daily adolescent activity-avoidance via lower parental protective responses displayed on a daily basis. Likewise, it was explored if psychologically flexible parenting and parental acceptance of adolescent pain would be indirectly related to higher daily adolescent activity-engagement via lower parental protective responses. Second, we explored if the type of parental verbal instructions directed at their adolescent also mediated these relationships. Our exploratory hypothesis was that higher levels of parental instructions to engage in pain-related activities would mediate the relationship between parental psychological flexibility and parental acceptance of adolescent pain on the one hand and daily adolescent behavior (i.e., lower avoidance and higher activity engagement) on the other hand. Examining the processes that underlie the influence of parental psychological flexibility on adolescent functioning on a daily basis may help to advance our understanding of its adaptive effects and help to identify (novel) targets for treatments directed at enhancing adolescent and parent functioning in the context of chronic pain in youth.

MATERIALS AND METHODS

Participants

Participants were adolescents with mixed chronic pain conditions and their primary caregiver (i.e., mother or father). Recruitment took place when they presented for initial clinical evaluation at the Pain Treatment Service at Boston Children’s Hospital (BCH) between February 2017 and December 2017, and via the Pediatric Pain Management Clinic at Stanford Children’s Health (SCH) between February 2017 and February 2018. Institutional Review Board (IRB) approval was granted at each site prior to the start of the study (BCH IRB#P0020989; Stanford IRB#39092). The present study is part of a large research project, Child Pain In Context (CP-IC), with the complete study protocol available at <http://hdl.handle.net/1854/LU-8578159>. One paper has been published already on this CP-IC research project and examined

adolescent predictors of pain-related behavior using a network analysis approach (see Beeckman et al., 2019b). The current paper focuses on parental variables impacting adolescent pain-related behavior.

Eligibility criteria for participation were [1] being 11–17 years old, [2] reporting persistent or recurrent pain for 3 months or longer, [3] having internet access at home or on an accessible smartphone, [4] absence of significant cognitive impairments (e.g., intellectual disability, severe brain injury), [5] absence of severe psychiatric or neurological conditions, and [6] availability of one primary caregiver who was also willing to participate.

Of the 84 parent-adolescent dyads who initially consented/assented to participate, 56 dyads (i.e., 67%) completed a set of baseline self-report questionnaires followed by a 14-day diary assessment period. Reasons for non-completion included: lack of interest after initial consent ($n = 19$) and no baseline parent data ($n = 9$). As specified in the CP-IC protocol, at least 50 participants (i.e., parent-adolescent dyads) should be sufficient to perform multilevel analyses (Maas and Hox, 2005; Nezlek, 2012).

Study Procedure

Informed consent was obtained on paper or online before the start of the study. Parents signed an informed consent for their own participation and that of the adolescent, and adolescents additionally gave informed assent. All study data were collected and managed using the REDCap (Research Electronic Data Capture) (Harris et al., 2009) tool hosted at BCH and Stanford University. REDCap is a secure, web-based application designed to support data capture for research studies. All communication with the participants was carried out via the parent (either via text message or e-mail).

At the start of the study participants received an online link to access the baseline self-report questionnaires. Once self-reports were completed, the diary period was scheduled to begin the following week. Automatic messages containing the diary surveys were sent to the participants each day for 14 consecutive days. Adolescents were asked to complete surveys in the afternoon and the evening, while parents completed one end-of-the-day diary. Afternoon surveys for the adolescent were sent at 2 pm and deactivated at 6 pm, and evening surveys for adolescent and parent were sent at 6 pm and deactivated at 10 am the next day. In line with the recommendation by Nezlek (2012, p. 46), all surveys completed between these time windows were treated as valid reports. If an adolescent and/or parent did not complete any of the required diary assessments on three consecutive days (despite reminder calls), the family was given the option of withdrawing from the study. If they decided to continue and the adolescent and/or parent failed to provide data on any additional days after this final reminder, their participation in the study was terminated and they received no further diary invitations. It was not possible for adolescent and/or parent to continue their participation individually.

Participants who started the 2-week diary period received one 10-dollar gift voucher (1 per family) at the end of the first week irrespective of the number of completed days. This was intended to serve both as a sign of appreciation for their participation, as well as an incentive to complete daily diaries in the second

week. Participating parent-adolescent pairs received a 20-dollar gift voucher at the end of week two unless they withdrew from the study during the first week.

Measures

Baseline Questionnaires

Adolescents and their parents completed a set of self-report questionnaires measuring demographic information and key study variables prior to the start of the diary period.

Demographic information was obtained by asking adolescents and parents to complete a short questionnaire assessing adolescent age, gender, ethnicity, race, and schooling grade. Parents were additionally asked to report on adolescent pain characteristics (i.e., pain location and duration) and parent gender, marital status, and educational level.

Adolescent pain severity was assessed by means of the child version of the Graded Chronic Pain Scale (Von Korff et al., 1992; Vervoort et al., 2014). Current and average pain intensity in the past six months were rated on a 11-point numerical rating scale (0 = *no pain*, 10 = *worst possible pain*) and used to calculate a characteristic pain intensity score. Disability was measured in terms of disability points. These points reflect a sum score of points allocated to the total number of days on which the child was prevented from carrying out usual activities in the past six months (0: <7 days; 1: ≥ 7 and <15 days; 2: ≥ 15 and <31 days; 3: ≥ 31 days) and points allocated to the degree to which pain caused difficulties in performing their usual activities in that same period (0 = *no difficulties at all*; 10 = *impossible to do activities*; 0: <3; 1: ≥ 3 and <5; 2: ≥ 5 and <7; 3: ≥ 7). Based on the scores for pain intensity and disability adolescents can be classified into 5 pain grades (0 = pain free; I = low disability [<3], low intensity [<5]; II = low disability [<3], high intensity [≥ 5]; III = moderate disability [3 or 4], regardless of pain intensity; IV = high disability [≥ 5] regardless of pain intensity) which was used to describe the sample (Vervoort et al., 2014). The GCPS has been used as a valid measure of pain severity in primary care, chronic pain, and general population samples (Von Korff et al., 1993; Smith et al., 1997; Elliott et al., 2000). The child version has shown good psychometric properties in a general population sample (Vervoort et al., 2014).

Psychologically flexible parenting was measured by the Parental Acceptance Questionnaire (6-PAQ; Greene et al., 2015). The 6-PAQ was developed to measure the six core processes that constitute psychological flexibility applied to an interpersonal, parenting context. The questionnaire consists of 18-items that are answered on a 4-point response scale (1 = *strongly disagree/never*; 4 = *strongly agree/almost always*). A total score and subscale scores for each of the six processes can be obtained. Example items for each of the six subscales are: “It is difficult to initiate/maintain routines because I don’t want to deal with my child’s reactions” (*Acceptance*); “I have negative thoughts about myself when my child behaves in a negative way” (*Defusion*); “I feel like my mind is somewhere else when I play with my child” (*Being Present*); “When parenting doesn’t go as I had planned, I feel like a failure” (*Self-as-Context*);

“My actions as a parent are consistent with my values” (*Value-based Actions*); and “My parenting behaviors are based on what matters to me as a parent rather than how I feel in the moment” (*Committed Action*). In line with previous research (see Williams et al., 2012; Beeckman et al., 2018), items were reverse-scored so that higher total scores reflect higher psychologically flexible parenting. The 6-PAQ has been shown to be a psychometrically sound measure to assess psychological flexibility in the parenting of young, healthy children (3–12 years) (Greene et al., 2015). To the best of our knowledge, this was the first study to use the 6-PAQ to assess parenting-specific psychological flexibility in parents of adolescents (11–17 years) with chronic pain. Cronbach’s alpha for the total 6-PAQ scale in the current study was 0.83.

Parental acceptance of adolescent pain was assessed by means of the Parent Pain Acceptance Questionnaire (PPAQ; Smith et al., 2015). The PPAQ consists of two subscales measuring a parent’s acceptance of pain-related thoughts and feelings [four items; e.g., “I must change my thoughts and feelings about my child’s pain before I can take important steps in my life (reverse scored)”], and a parent’s activity-engagement despite their adolescent’s pain (11 items; e.g., “I lead a full life even though my child has chronic pain”). All items were scored on a 5-point response scale (0 = *never true*; 4 = *always true*). Higher total scores reflect higher parent acceptance of child pain. The PPAQ has been validated in a sample of parents of children with chronic pain (Smith et al., 2015). In the current study Cronbach’s alpha was 0.91 for the total PPAQ scale.

Daily Diary Measures

Daily adolescent pain intensity, activity-avoidance and activity-engagement, and parental protective behavior and engagement instructions were measured by means of 14-day daily diary for adolescents and parents. Adolescents were asked to report on “the period since the previous diary entry” in the afternoon and evening assessments. Parents were asked to report on their experiences “today” in their daily diaries. All diary items were rated on a five-point response scale (unless stated otherwise) with the following labels: 0 (*not at all true*), 1 (*a little true*), 2 (*somewhat true*), 3 (*mostly true*), and 4 (*totally true*). Diary items were developed by the research team based on items of existing questionnaires that were adjusted for daily or momentary use and consequently validated using the Discriminant Content Validity (DCV) procedure of Johnston et al. (2014). As a part of this content validation procedure five psychologists with expertise in the field of pediatric pain research were asked to rate the extent to which each of the diary items measured the predetermined constructs to illuminate those that required reformulation before inclusion in the final diary. None of the items that were developed to measure the constructs used in this study required reformulation based on the results of the expert ratings. Total diary scale scores were calculated by taking the average of the single item responses (i.e., if the scale consisted of two or three items), but only if at least 75% of the items were completed. If less than 75% of the items were completed, the total scale score was not calculated and considered missing. To effectively answer the key research questions, a daily score was

calculated for each adolescent variable by taking the average of the afternoon and evening scale scores.

Daily adolescent pain intensity

Adolescents were asked about their overall level of pain in the afternoon and evening with 1 item (“Since the previous diary entry, what was your overall level of pain?”). This item was rated on a 11-point numerical rating scale from 0 (*no pain*) to 10 (*worst possible pain*), which is considered to be the golden standard for the assessment of pain intensity (Castarlenas et al., 2017).

Daily adolescent activity-avoidance

Adolescents’ avoidance of activities because of the pain in the afternoon and evening was assessed using three items that were based on the ‘Avoidance of Activities’ subscale of the Fear of Pain Questionnaire for Children [FOPQ-C (Simons et al., 2011)] and adjusted for use in the diary (“I skipped my planned activities because I expected them to trigger or increase my pain.”, “I stopped what I was doing because my pain started to get worse,” “I spent my time resting instead of doing my activities, because of my pain”). These items were selected to reflect different types of pain-related avoidance strategies in agreement with the author of the original FOPQ-C, and were evaluated as valid items by the experts during the content validation procedure. Good internal consistency ($\alpha = 0.86$) and reliability have been found for the FOPQ-C avoidance subscale in pediatric chronic pain samples (Simons et al., 2011).

Daily adolescent activity-engagement

Adolescents were asked to complete two items that assessed their engagement in activities in the presence of pain in the afternoon and evening. The items of the activity-engagement scale were only presented to those who experienced some level of pain at the same time (i.e., a pain intensity score of one or higher). This is in accordance with the operationalization of activity-engagement as a behavior which is only relevant in the presence of pain. Following items were used: “I have put effort into completing activities that I find important or fun, while I was in pain,” and “I persisted in carrying out my planned activities while I was in pain.” These daily items were developed based on items of the ‘Activity-engagement’ subscale of the Chronic Pain Acceptance Questionnaire for Adolescents (CPAQ-A) (McCracken et al., 2010) and were evaluated as valid items by the experts during the content validation procedure. The CPAQ-A has proven to be a valid and reliable measure of pain acceptance (i.e., pain willingness and activity-engagement) in youth with chronic pain (McCracken et al., 2010; Wallace et al., 2011).

Daily parental protective responses

Parents reported daily on their protective responses toward the adolescent in pain, by means of two items: “Today, I made sure that my child did not have to do certain activities (e.g., household chores) because of his/her pain” and “Today, I canceled my personal activities (e.g., job-related duties, household chores and/or hobbies) so that I could be with my child.” These daily items were constructed based on items of the ‘Solicitousness’ subscale of the Inventory of Parent/Caregiver Responses to the Children’s Pain Experience (IRPEDNA) (Huguet et al., 2008)

and were evaluated as valid items by the expert team. The IRPEDNA has shown good psychometric properties in a sample of parents of healthy children and adolescents from 6 to 16 years (Huguet et al., 2008).

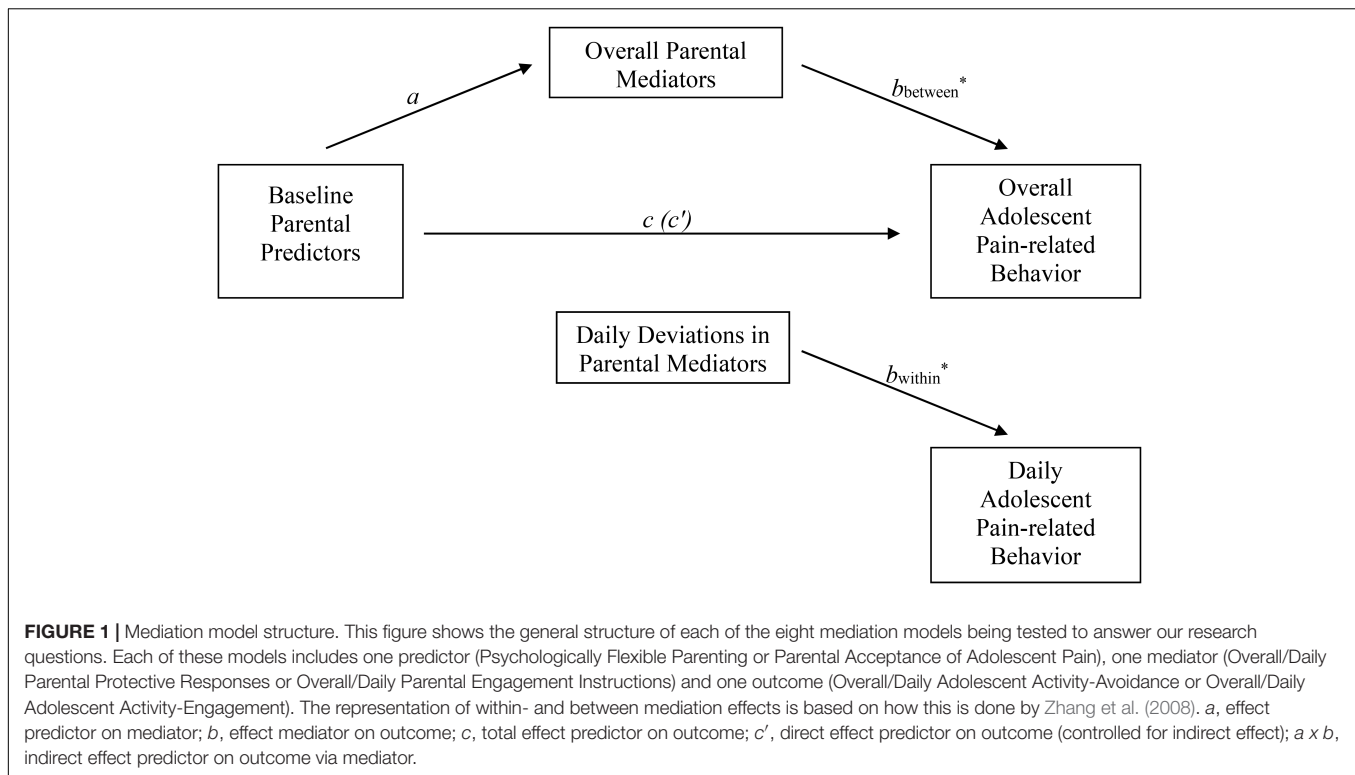
Daily parental engagement instructions

Parents were asked to report on the degree to which they provided their child with instructions to either engage in or avoid activities during the past day. The following items were used to assess this: “I told my child to stop or cancel activities when in pain” (activity-avoidance instruction) and “I told my child to keep doing fun or important activities (and other activities he/she usually does) when in pain” (activity-engagement instruction). These items were constructed by reformulating the items of the activity-engagement and avoidance scales in the adolescent diary to represent possible instructions parents might give to their children in the context of pain. We know of no other existing questionnaire measuring parental instructions in the context of pain. A relative parental activity-engagement instruction score was created by subtracting the daily activity-avoidance instruction score from the daily activity-engagement instruction score. A positive score on a given day indicates that a parent provided more instructions to their adolescent to engage in activities despite the pain than activity-avoidance instructions.

Data Analytic Strategy

Descriptive statistics, correlations, and internal consistencies of the baseline questionnaires were calculated using SPSS (v.25; IBM Statistics). Reliability of the diary scales was calculated in Mplus following a multilevel confirmatory factor analysis framework which makes it possible to estimate within- and between-level reliabilities of the scales (Geldhof et al., 2014). Pearson product-moment correlations were calculated to examine bivariate associations between adolescent age, adolescent gender, psychologically flexible parenting, parental acceptance of adolescent pain, parent and adolescent diary variables (aggregated over days). These correlations were evaluated at the 5% significance level. Multilevel mediation analyses were performed in R (v. 3.5.2; R Foundation of Statistical Computing) using the *lme4*-package (Bates et al., 2015), and 95% confidence intervals for the indirect effects were obtained using the *boot*-package (Davison and Hinkley, 1997; Canty and Ripley, 2019). Multilevel modeling can account for the hierarchical data structure (i.e., multiple observations nested within dyads) without violating the assumption of independence of observations and assumes that observations are missing at random (Snijders and Bosker, 2012).

Figure 1 presents the general structure of each of the mediation models that were fitted to answer our research questions. Predictors (psychologically flexible parenting or parental acceptance of adolescent pain), mediators (parental protective responses or parental instructions concerning activity-engagement), and outcomes (adolescent activity-avoidance or activity-engagement in the presence of pain) were entered separately into the models, resulting in eight mediation models. Adolescent age, gender, and aggregated daily pain intensity scores were explored as potential confounding variables in



each model, and were only included as control variables in the final model when they significantly correlated with both the predictor/mediator and the outcome variable. Level 2 predictors (i.e., psychological flexible parenting, parental acceptance of adolescent pain, and adolescent age) were standardized to facilitate interpretation of the coefficients. Random intercepts were allowed, while the slopes of the investigated effects were fixed.

Our longitudinal (daily diary) data allowed us to examine the aforementioned relationships both within- and between parent-adolescent dyads [we adopted this strategy based on a recommendation by Zhang et al. (2008)]. That is, we split the mediator into two independent pieces: the within-dyad deviations and the between-dyad overall means (Figure 1). Within-dyads effects were examined by analyzing the association between daily deviations from the parent-specific average and daily adolescent outcome variables within parent-adolescent dyads. Between-dyads effects were examined by analyzing the association between overall parent mediator and overall adolescent outcome variables (i.e., by taking the average of all daily observations within parents and adolescents across the 14-day diary period). Within-dyads effects reflect that part of the indirect effect of the predictor on the outcome that is explained by the daily variability in the mediator within a given parent-adolescent dyad. In other words, that part of the effect explained by “state” or momentary levels of the mediator variable, i.e., answering questions about when changes occur within the parent-adolescent dyad. Between-dyads effects reflect that part of the indirect effect explained by the variability between parent-adolescent dyads in the “trait” or characteristic level of the

mediator variable (for similar terminology see Geiser et al., 2013), i.e., answering questions about how parent-adolescent dyads differ from each other. Whereas cross-sectional data can only address between-dyads effects, the longitudinal nature of our data allowed us to disentangle mediation effects at a within-dyads and between-dyads level. The above-described analyses were focused on examining associations between parent and child variables on a daily basis, but did not examine within-day or day-to-day associations between those variables. Finally, a bootstrap procedure was used to assess the significance of the indirect effects. When weight *a* represents the effect of the predictor on the mediator, and weight *b* the effect of the mediator on the outcome, the indirect effect of the predictor on the outcome (via the mediator) is obtained as the product of weight *a* and *b* (see Figure 1). Significance is determined by inspecting the percentile-based 95% confidence intervals around this product: effects are considered to be significant if this confidence interval does not contain zero.

RESULTS

Sample Characteristics

The final sample consisted of 56 adolescent ($M_{\text{age}} = 14.50$, $SD = 1.90$) and parent (93% mothers) dyads. The majority of the sample was female (i.e., 86% adolescent girls) and Caucasian (i.e., 66%). Fifty-five percent of adolescents reported musculoskeletal pain (i.e., in the arms, shoulders, neck, or legs) as their primary pain, followed by abdominal pain (i.e., 20%), headaches (i.e., 13%), and other types of pain (i.e., 13%; e.g., pelvic pain).

About half of adolescents (i.e., 52%) reported high levels of disability (i.e., pain Grade VI; see section ‘Measures’). Detailed demographic characteristics of adolescents and parents can be found in **Table 1**.

Of a total of 784 possible daily diary observations (i.e., one observation per day/per participant for 14 consecutive days), 625 data points were available for daily adolescent activity-avoidance (i.e., 20% missing), 528 for daily adolescent activity-engagement (i.e., 32% missing), 582 for parental daily protective responses (i.e., 26% missing), and 560 for parental daily engagement instructions (i.e., 28% missing). Ninety-one percent of the daily pain intensity ratings during the 2-week period were scored at one or higher, while 62% of the daily pain ratings were scored at 4 or higher.

Descriptive Statistics

Means, standard deviations and bivariate Pearson correlation coefficients between baseline measures of adolescent age, gender, parent variables, and aggregated (adolescent and parent) diary variables can be found in **Table 2**. Correlational patterns showed a positive association between psychological flexible parenting and parental acceptance of adolescent pain ($r = 0.38, p = 0.004$). Adolescent baseline pain intensity and daily activity-avoidance behavior (aggregated over days) were positively correlated ($r = 0.27, p = 0.044$). Age, gender, and baseline pain intensity were no significant confounders of any of the investigated relations between parental variables and daily adolescent pain-related behavior and were therefore not included as control variables in the final models.

Reliability assessment showed acceptable to excellent within- and between-level reliabilities for the diary scales (see **Table 3**).

Examining the Indirect Relationship Between Psychologically Flexible Parenting/Parental Acceptance of Adolescent Pain and Adolescent Pain-Related Behavior via Parental Protective Responses

Does Psychologically Flexible Parenting Indirectly Impact Daily Adolescent Pain-Related Behavior via Parental Protective Responses?

The left column of **Table 4** shows the results of bootstrap analyses designed to test the hypothesized indirect effects from psychologically flexible parenting to daily pain-related behavior in adolescents via parental protective responses. Results showed that there was a significant indirect effect of psychologically flexible parenting on daily adolescent activity-avoidance, via daily parental protective responses, at the within-dyads level ($a \times b = -0.03$, 95% CI = -0.06 to -0.01), but not at the between-dyads level ($a \times b = -0.03$, 95% CI = -0.06 to 0.01) (also see **Table 4**). This suggests that parental psychological flexibility was predictive of lower daily parental protectiveness, and that daily decreases in parental protectiveness *within a parent-adolescent dyad* was associated with decreased levels of adolescent activity-avoidance. Critically, this mediation was not

explained by differences *between parents* in their overall level of protectiveness (across the 14-day period), but only by daily variation in protective responding. After controlling for this

TABLE 1 | Sociodemographic characteristics about adolescents and parents.

Demographic variables	M (SD) or % (N)
Adolescent characteristics	
Age (years)	14.50 (1.90)
Gender	
Female	85.7 (48)
Male	14.3 (8)
Race	
White or Caucasian	66.1 (37)
Black or African American	3.6 (2)
Asian	1.8 (1)
Multiracial	3.6 (2)
Choose to not answer	1.8 (1)
Missing	23.1 (13)
Primary Pain	
Headache	12.5 (7)
Abdominal Pain	19.6 (11)
Musculoskeletal Pain	55.4 (31)
Other	12.5 (7)
Pain duration (months)	26.59 (23.10)
Pain grades	
Grade 0	0 (0)
Grade I	10.7 (6)
Grade II	12.5 (7)
Grade III	21.4 (12)
Grade IV	51.8 (29)
Parent Characteristics	
Relation to child	
Mother	92.9 (52)
Father	7.1 (4)
Ethnic background	
Hispanic	12.5 (7)
Non-Hispanic	85.7 (48)
Missing	1.8 (1)
Marital status	
Married	71.4 (40)
Divorced	12.5 (7)
Separated	3.6 (2)
Never Married	12.5 (7)
Employment status	
Full-time	51.8 (29)
Part-time	23.2 (13)
Homemaker	17.9 (10)
Unemployed	3.6 (2)
Disabled	3.6 (2)
Education level	
High school or less	5.4 (3)
Some college/Vocational school	10.7 (6)
College degree	44.6 (25)
Graduate/Professional school	39.3 (22)

Grade 0 = pain free; Grade I = low pain intensity, low disability; Grade II = high pain intensity, low disability; Grade III = moderate disability regardless of pain intensity; Grade IV = high disability regardless of pain intensity.

TABLE 2 | Sample size, range, means, standard deviations and bivariate pearson correlation coefficients between baseline variables and aggregated daily diary scores.

Variable	N	Range	M (SD)	2	3	4	5	6	7	8	9
Baseline measures											
(1) Adolescent Age	56	11 – 17	14.50 (1.90)	0.11	0.05	0.02	0.02	0.11	−0.14	−0.07	−0.04
(2) Adolescent Gender	56	n/a	n/a	–	0.14	0.04	−0.08	−0.01	−0.03	0.16	−0.09
(3) Adolescent Pain Intensity	56	0.3 – 10	5.10 (2.31)	–	–	0.15	−0.19	0.22	−0.04	0.27*	−0.19
(4) Psychologically Flexible Parenting	56	18 – 47	28.67 (6.44)	–	–	–	0.38**	−0.15	0.17	−0.10	0.10
(5) Parental Acceptance of Adolescent Pain	56	8 – 56	34 (11.52)	–	–	–	–	−0.44**	−0.05	−0.09	0.15
Diary measures^a											
(6) Parent Protective Responses	55	0 – 4	0.52 (0.85)	–	–	–	–	–	−0.14	0.27	−0.10
(7) Parent Engagement Instructions	55	−3.25 – 3.83	1.60 (1.60)	–	–	–	–	–	–	−0.22	0.65**
(8) Adolescent Activity-avoidance	56	0 – 4	0.94 (0.86)	–	–	–	–	–	–	–	−0.30*
(9) Adolescent Activity-engagement	56	0.3 – 4	2.70 (1.01)	–	–	–	–	–	–	–	–

^aAggregated scores (over days). * $p < 0.05$, ** $p < 0.01$.

TABLE 3 | Within- and between-dyads reliabilities for the diary scales.

	Parent		Adolescent			
	Protective responses		Activity-avoidance		Activity-engagement	
	Evening	Afternoon	Evening	Afternoon	Evening	Afternoon
Within-dyads α	0.65	0.82	0.81	0.63	0.73	0.93
Between-dyads α	0.93	0.95	0.93	0.92	0.93	0.93

Reliabilities are shown for the scales (>2 items) that were used in the diary. Afternoon and evening scores were averaged to obtain a daily activity-avoidance respectively activity-engagement score. Scale reliabilities are calculated based on a procedure by Geldhof et al. (2014).

indirect effect via parental protective responses, results showed no remaining direct effect of psychologically flexible parenting on daily adolescent activity-avoidance ($c' = -0.05$, 95% CI = -0.12 to 0.05) (Table 4).

Analyses also revealed an indirect effect of psychologically flexible parenting on daily adolescent activity-engagement, via parental protective responses, but only at the between-dyads level ($a \times b = -0.03$, 95% CI = 0.01 to 0.06) (Table 4). This suggests that the indirect effect was explained by differences between parent-adolescent dyads in the overall level of protectiveness in parents and not by daily variation in parental protective responses within those dyads. There was no significant direct effect of psychologically flexible parenting on daily adolescent activity-engagement after controlling for the indirect effect via parental protective responses ($c' = 0.07$, 95% CI = -0.01 to 0.14) (Table 4).

Does Parental Acceptance of Adolescent Pain Indirectly Impact Daily Adolescent Pain-Related Behavior via Parental Protective Responses?

The hypothesized indirect effect of parental acceptance of adolescent pain on daily adolescent activity-avoidance via parental protectiveness was significant at the within-dyads level ($a \times b = -0.08$, 95% CI = -0.18 to -0.01), but not at the between-dyads level ($a \times b = -0.08$, 95% CI = -0.17 to 0.02) (see Table 4, right column). This suggests that the indirect effect was explained by daily variation in parental protective responses within parent-adolescent dyads, but not by differences between

parents in terms of their overall protectiveness across the 14-day diary period. After controlling for the indirect effect via parental protective responses, there was no significant direct effect of parental acceptance of adolescent pain on daily adolescent activity-avoidance ($c' = 0.06$, 95% CI = -0.06 to 0.17) (Table 4).

Finally, no significant indirect effect of parental acceptance of adolescent pain on daily adolescent activity-engagement via parental protective responses was observed (Table 4). The direct effect of parental acceptance of adolescent pain on daily adolescent activity-engagement was also not significant after controlling for daily parental protective behavior ($c' = 0.06$, 95% CI = -0.05 to 0.18) (Table 4).

Examining the Indirect Effect of Psychologically Flexible Parenting/Parental Acceptance of Adolescent Pain on Daily Pain-Related Behavior in Adolescents via Parental Instructions

Does Psychologically Flexible Parenting Indirectly Impact Daily Adolescent Pain-Related Behavior via Parental Engagement Instructions?

The indirect effect of psychologically flexible parenting on daily adolescent activity-avoidance via parental engagement instructions was significant, both at the within- ($a \times b = -0.06$, 95% CI = -0.13 to -0.002) and between-dyads level ($a \times b = 0.09$, 95% CI = 0.003 to 0.16) (see Table 5, left

TABLE 4 | Bootstrap tests of indirect effects of psychologically flexible parenting and parental acceptance of adolescent pain on adolescent pain-related behavior via parents' protective responses.

Effect (path)	Baseline predictor:			
	Psychologically flexible parenting		Parental acceptance of adolescent pain	
	Estimate	95% CI	Estimate	95% CI
Predictor - > parental protective responses (a-path)	-0.15	-0.20 to -0.09	-0.41	-0.47 to -0.35
Parental Protective Responses - > Outcome (b-path)				
Adolescent activity-avoidance				
Within-dyad	0.20 [†]	0.04 to 0.37	0.20 [†]	0.03 to 0.37
Between-dyad	0.18	-0.04 to 0.33	0.20	-0.05 to 0.41
Adolescent Activity-engagement				
Within-dyad	-0.11	-0.25 to 0.02	-0.11	-0.25 to 0.03
Between-dyad	-0.20 [†]	-0.36 to -0.04	-0.17	-0.37 to 0.06
Indirect effect via parental protective responses (a*b)				
Adolescent activity-avoidance				
Within-dyad	-0.03 [†]	-0.06 to -0.01	-0.08 [†]	-0.18 to -0.01
Between-dyad	-0.03	-0.06 to 0.01	-0.08	-0.17 to 0.02
Adolescent activity-engagement				
Within-dyad	0.02	-0.00 to 0.04	0.05	-0.01 to 0.10
Between-dyad	0.03 [†]	0.01 to 0.06	0.07	-0.02 to 0.15
Total effect predictor → outcome (c-path)				
Adolescent activity-avoidance	-0.04	-0.11 to 0.04	-0.01	-0.09 to 0.08
Adolescent activity-engagement	0.09	0.03 to 0.15	0.16	0.09 to 0.24
Direct effect predictor → outcome (c'-path) ¹				
Adolescent activity-avoidance	0.02	-0.05 to 0.09	0.06	-0.06 to 0.17
Adolescent activity-engagement	0.00	-0.08 to 0.09	0.06	-0.05 to 0.18

We refer to the template model in **Figure 1** for additional help in interpreting the results presented in this table. ¹ After controlling for indirect effect. Effects indicated with [†] were no longer significant when applying a conservative Bonferroni correction for multiple testing.

column). Critically, however, the direction of effect was opposite at the within and between-dyads levels. On the one hand, there was a *negative* within-dyads indirect effect of psychologically flexible parenting on daily adolescent activity-avoidance, indicating that psychologically flexible parenting was associated with less daily adolescent activity-avoidance. This was explained by increased levels of daily parental engagement instructions ($b_{\text{within}} = -0.03$, 95% CI = -0.06 to -0.001). This suggests that psychologically flexible parenting was predictive of higher daily engagement instructions, and that daily increases in engagement instructions within the parent-adolescent dyad were associated with decreased levels of adolescent activity-avoidance. On the other hand, we also found an unexpected *positive* indirect effect of parental psychological flexibility on adolescent activity-avoidance at the between-dyads level, via higher overall levels of parental engagement instructions ($b_{\text{between}} = 0.04$, 95% CI = 0.001 to 0.08). This suggests that psychologically flexible parenting was also predictive of higher overall levels of engagement instructions in parents and that these higher overall levels were associated with higher overall adolescent activity-avoidance across the 14-day period. After controlling for the indirect effects of parental psychological flexibility via daily parental engagement instructions, no significant direct

effect of parental psychological flexibility on daily adolescent activity-avoidance emerged ($c' = -0.05$, 95% CI = -0.13 to 0.04) (**Table 5**).

Finally, an indirect effect of parental psychological flexibility on higher daily adolescent activity-engagement via higher parental engagement instructions emerged, but only at the between-dyads level ($a \times b = 0.10$, 95% CI = 0.05 to 0.15) (**Table 5**). This suggests that psychologically flexible parenting predicted higher overall levels of engagement instructions in parents, and that these higher overall levels of engagement instructions were associated with higher overall adolescent activity-engagement across the 14-day period. No direct effect of parental psychological flexibility on daily adolescent activity-engagement emerged when the indirect effect was controlled for ($c' = -0.06$, 95% CI = -0.14 to 0.04) (**Table 5**).

Does Parental Acceptance of Adolescent Pain Indirectly Impact Daily Adolescent Pain-Related Behavior via Parental Engagement Instructions?

An indirect effect of parental acceptance of adolescent pain on daily adolescent activity-avoidance via daily parental engagement instructions emerged. However, the direction of this effect was opposite at the within- ($a \times b = -0.02$, 95% CI = -0.04

TABLE 5 | Bootstrap tests of indirect effects of psychologically flexible parenting and parental acceptance of adolescent pain on adolescent pain-related behavior via parental (Engagement) instructions.

Effect (path)	Baseline predictor:			
	Psychologically flexible parenting		Parental acceptance of adolescent pain	
	Estimate	95% CI	Estimate	95% CI
Predictor - > parental engagement instructions (a-path)	0.47	0.37 to 0.58	0.31	0.22 to 0.42
Parental engagement instructions - > outcome (b-path)				
Adolescent activity-avoidance				
Within-dyad	-0.03 [†]	-0.06 to -0.001	-0.06 [†]	-0.12 to -0.01
Between-dyad	0.04 [†]	0.001 to 0.08	0.08 [†]	0.003 to 0.15
Adolescent activity-engagement				
Within-dyad	0.04	-0.02 to 0.10	0.01	-0.01 to 0.03
Between-dyad	0.20	0.10 to 0.28	0.06 [†]	0.03 to 0.09
Indirect effect via parental engagement instructions (a*b)				
Adolescent activity-avoidance				
Within-dyad	-0.06 [†]	-0.13 to -0.002	-0.02	-0.04 to -0.002
Between-dyad	0.09 [†]	0.003 to 0.16	0.03 [†]	0.001 to 0.05
Adolescent activity-engagement				
Within-dyad	0.02	-0.01 to 0.05	0.04	-0.02 to 0.10
Between-dyad	0.10	0.05 to 0.15	0.19	0.10 to 0.25
Total effect predictor → outcome (c-path)				
Adolescent activity-avoidance	-0.04	-0.12 to 0.05	0.01	-0.09 to 0.08
Adolescent activity-engagement	0.09 [†]	0.03 to 0.16	0.17	0.09 to 0.24
Direct effect predictor → outcome (c'-path) ¹				
Adolescent activity-avoidance	-0.05	-0.13 to 0.04	-0.05	-0.12 to 0.05
Adolescent activity-engagement	-0.06	-0.15 to 0.04	0.07	-0.01 to 0.14

We refer to the template model in **Figure 1** for additional help in interpreting the results presented in this table. ¹ After controlling for indirect effect. Effects indicated with [†] were no longer significant when applying a conservative Bonferroni correction for multiple testing.

to -0.002) and between-dyads levels ($a \times b = 0.03$, 95% CI = 0.001 to 0.05) (see **Table 5**, right column). On the one hand, a *negative* within-dyads indirect effect emerged of parental acceptance of adolescent pain on daily adolescent activity-avoidance which was explained by daily increases in parental engagement instructions ($b_{within} = -0.06$, 95% CI = -0.12 to -0.01). This suggests that parental acceptance of adolescent pain was predictive of higher engagement instructions in parents, and that daily increases in engagement instructions were associated with daily decreases in adolescent activity-avoidance. On the other hand, a *positive* between-dyads indirect effect emerged of parental acceptance of adolescent pain on daily adolescent activity-avoidance which was explained by lower overall levels of parental engagement instructions ($b_{between} = 0.08$, 95% CI = 0.003 to 0.15). Parental acceptance of adolescent pain was predictive of higher overall parental engagement instructions, and these higher overall engagement instructions were associated with higher overall adolescent activity-avoidance across the 14-days. After controlling for these indirect effects, no direct effect of parental acceptance of adolescent pain on daily adolescent activity-avoidance emerged ($c' = 0.02$, 95% CI = -0.05 to 0.09) (**Table 5**).

Finally, an indirect effect emerged of parental acceptance of adolescent pain on higher daily activity-engagement in adolescents via parental engagement instructions. This effect

emerged at the between ($a \times b = 0.19$; 95% CI = 0.10 to 0.25) (**Table 5**) but not within-dyads level. No direct effect of parental acceptance of adolescent pain on adolescent activity-engagement emerged once this indirect effect was controlled for ($c' = 0.001$, 95% CI = -0.08 to 0.09) (**Table 5**).

DISCUSSION

Parents exert an important impact on their adolescents' functioning in the presence of persistent pain (Palermo, 2009; Palermo et al., 2014), and in certain cases, can worsen adolescent functioning (Goubert et al., 2006; Logan et al., 2012; Hechler et al., 2015; Simons et al., 2015; Chow et al., 2016). Yet parents may also positively contribute to adaptive pain-related functioning in their child. More specifically, it has recently been argued that parental psychological flexibility may be associated with beneficial adolescent outcomes (e.g., lower disability) (Wallace et al., 2015; Timmers et al., 2019). The present study further examined whether psychologically flexible parenting and parental acceptance of adolescent pain indirectly predicted daily adolescent pain-related behavior, via their respective impact on daily parental protective responses, and/or daily instructions parents provide to their adolescent.

In line with our expectations, the findings indicated that psychologically flexible parenting and parental acceptance of adolescent pain indirectly predicted lower daily adolescent *activity-avoidance* via their impact on *lower daily parental protective* responses. Such findings are consistent with previous studies showing similar adaptive effects of parental psychological flexibility on adolescent outcomes via parental protective behavior (e.g., Timmers et al., 2019). Whereas that work was based on questionnaires administered at one moment in time, we demonstrated this indirect effect with daily data collected at multiple moments. Likewise, it was found that decreases in parental daily protective responses were associated with decreases in adolescent daily activity-avoidance within those parent-adolescent dyads where parents showed higher levels of acceptance of adolescent pain.

Furthermore, as expected, psychologically flexible parenting and parental acceptance of adolescent pain also predicted adolescent activity-avoidance via their indirect impact on *parental instructions* to engage in activities. Note, however, that these indirect effects via engagement instructions showed an opposite direction at the within-dyads versus the between-dyads level. On the one hand, we found that increased daily levels of engagement instructions within these more flexible and pain accepting parents were associated with *decreased daily* levels of activity-avoidance in their adolescents. Yet, on the other hand, we found that psychologically flexible parenting and parental acceptance of adolescent pain were also related to *higher overall* levels of adolescent activity-avoidance via their association with higher overall levels of parental engagement instructions across the 2-week period. One *post hoc* explanation for these contrasting findings is that daily increases in parental instructions to engage in more activities might momentarily lower adolescent activity-avoidance but that the persistent application of those same instructions over and over again might have the opposite effect across time. It may be that overall high levels of parental engagement instructions contribute to overall high or persistent levels of adolescents' avoidance instead, which may adversely impact adolescent functioning on the long-term (Asmundson et al., 2012; Simons and Kaczynski, 2012; Chow et al., 2016).

In short, based on these exploratory findings, one could hypothesize that the adaptive effects of psychologically flexible parenting and parental acceptance of adolescent pain on lower levels of adolescent activity-avoidance may be explained by momentary or daily decreases in the level of protective responses and engagement instructions in these flexible or acceptant parents. Taking a step, this could suggest that these daily changes in parents' protective responses or engagement instructions are potentially well-adapted to the daily context (e.g., how the adolescent is feeling or what activities he/she is planning on that day). This hypothesis is consistent with the idea of psychological flexibility as one's ability to flexibly adapt behavior to the (daily) situation (Hayes et al., 2006; McCracken and Morley, 2014). Thus our findings may suggest that psychologically flexible parenting in parents of adolescents with chronic pain may be characterized by being aware of the potential consequences of being (less) protective or providing (more) engagement instructions to their adolescent.

Furthermore, this was the first study to explore the influence of psychological flexible parenting and parental acceptance of adolescent pain on adolescent *activity-engagement*. Our findings suggest that higher levels of psychologically flexible parenting indirectly contributed to higher overall activity-engagement in adolescents across the 2-week period. This indirect influence was explained by lower overall parental protectiveness on the one hand, and by higher overall engagement instructions directed at their adolescent on the other hand. Similarly, higher parental acceptance of adolescent pain indirectly influenced higher overall levels of adolescent activity-engagement across the 2-week period. However, this was only mediated by higher overall engagement instructions in parents and not by their level of protectiveness.

Finally, psychologically flexible parenting and parental acceptance of adolescent pain were only moderately related, supporting the idea that they are overlapping but unique factors (McCracken and Morley, 2014; Smith et al., 2015). We also observed little to no differences in their contribution to adolescent pain-related behavior. If anything, psychologically flexible parenting indirectly predicted adolescent activity-engagement via both protective parenting responses and engagement instructions in parents, whereas parental acceptance of adolescent pain only did so via engagement instructions.

Future Directions and Clinical Implications

Our findings have implications for future research and clinical practice. First, they contribute to the idea that parents play a meaningful role in adolescents' pain-related functioning, and in particular, how psychologically flexible parenting and acceptance of adolescent pain might serve an adaptive role in daily adolescent (avoidance) behavior and support the inclusion of parents in the study and treatment of adolescent pain (Palermo and Chambers, 2005; Palermo and Eccleston, 2009; Law et al., 2014).

This was also the first study to explore the effect of parental (engagement) instructions on adolescent functioning in the context of pain. That said, our initial findings on this effect do not lend themselves to a clear-cut interpretation. They suggest that instructions from parents to their adolescent that encourage them to keep doing fun or important activities when in pain may be adaptive in the short-term on a given day (i.e., associated with lower levels of avoidance). Yet high overall levels of instructions across days may be associated with high or persistent overall levels of activity-avoidance *and* activity-engagement in adolescents. These high overall levels of activity-avoidance or activity-engagement both have the potential to be maladaptive for the adolescent. For instance, persistent avoidance has been found to predict long-term negative outcomes (e.g., disability) in adolescents with chronic pain (Asmundson et al., 2012; Simons and Kaczynski, 2012). Moreover, one might also argue that high or persistent levels of engagement in activities may also predict long-term negative outcomes. Past work on adults with chronic pain demonstrated that persistent levels of engagement is associated with negative outcomes such as muscular overuse, hyperactivity, decreased well-being, and increased disability (e.g.,

Hasenbring and Verbunt, 2010; Crombez et al., 2012). Critically, this claim is clearly *post hoc* and awaits future replication and direct empirical testing.

Parental instructions for adolescents to engage in activities when in pain were surprisingly not associated with actual daily activity-engagement in those same adolescents. These findings therefore do not fully support the hypothesized adaptive effects of parental engagement instructions on adolescent activity-engagement. A possible explanation for these puzzling findings may be that the impact of parental instructions on their child's behavior depends on the child's developmental stage. For instance, it may be that these findings are specific to adolescents as our sample mainly consisted of adolescents aged between 11 and 17 years. Adolescence is a challenging period that puts pressure on the parent-adolescent relationship. It is a period in which adolescent behavioral autonomy and parental autonomy-support becomes increasingly important in fostering a healthy development of the adolescent (Baumrind, 1966; Grolnick et al., 1997; Gray and Steinberg, 1999; Joussemet et al., 2008). It may be that our findings reflect that adolescents simply do not want to follow any kind of instructions provided by their parents or even respond in the opposite way. Potentially, different effects may be found when examining the influence of parental instructions on the pain-related behavior of younger children. We believe that this is a promising area ripe to be explored. Next to examining the factors that moderate when instructions influence behavior (e.g., developmental stage) future work could also explore other types of parental instructions and their influence on adolescent behavior. Yet another interesting question would be to explore if parents' (in)flexibility in providing instructions (e.g., adjusted to the situation or not) differently predicts adolescent pain-related functioning (also see Beekman et al., 2019a).

Finally, upon replication, our findings may be informative for research and interventions that focus on enhancing psychological flexibility in parents to increase adolescent adaptive outcomes. Psychological flexibility is the central change process within Acceptance and Commitment Therapy (ACT), a therapy which has shown promising results for youth with chronic pain (Wicksell et al., 2005, 2007; Wicksell, 2015), and has recently been extended by incorporating parents (Kanstrup et al., 2016; Wallace et al., 2016). The current work suggests that it may be important to develop interventions directed at enhancing psychological flexibility in parents, with a specific focus on teaching parents to decrease the use of daily protective behaviors in response to adolescent pain and potentially use them in a more flexible manner. However, it was not entirely clear from our findings if these interventions should target parents' use of instructions (to engage in activities), and how this should be done. We advocate that future work is needed that examines the effects of the (in)flexible use of protective behavioral responses and instructions in daily life on adolescent functioning before incorporating this suggestion into treatment.

Strengths and Limitations

The present study has several strengths. It was the first to (a) investigate the indirect effects of psychologically flexible parenting and parental acceptance of adolescent pain on

adolescent outcomes by using daily diary methodology, (b) introduce and examine parental daily engagement instructions as an alternative route via which these factors might have effects on adolescent functioning, and (c) examine this using multi-informant data from both parent and adolescent.

Yet, the present study also had several limitations, which may inform future research. First, no temporal associations (i.e., within-day, or day-to-day) were examined at the diary level, and as such, we cannot make interpretations of the investigated indirect effects in terms of predictability or causality. All investigated daily associations between parental responses and adolescent behavior could be interpreted in the reverse direction to that reported here. It is worth noting, however, that the proposed direction of the investigated associations stems from theory and previous empirical work (e.g., Timmers et al., 2019), lending support to the idea that psychologically flexible parenting influences adolescent outcomes via parental responses. Ideally, lagged analyses should be performed to examine such temporal associations between parental responses and subsequent changes in adolescent behavior. However, this type of analyses requires larger samples than the one described in the current study (Schultzberg and Muthén, 2018). Second, due to our limited sample size we were also not able to perform more complex analyses to directly compare the relative contribution of parental protective responses versus engagement instructions in explaining the indirect effect of psychologically flexible parenting and parental acceptance of adolescent pain on adolescent outcomes. Future research in larger samples could construct more complex mediation models with multiple mediators and predictors to examine the unique contribution of each of these factors (for an example of this analytic approach see Timmers et al., 2019). Third, although it is a strength of this study that the indirect effects were disentangled in a within- and between-dyads part, we had no pre-existing hypotheses about the effects at both levels. Our interpretations of these differences are therefore exploratory and require further investigation. This is particularly true for the finding that higher overall parental engagement instructions were associated with higher overall activity-engagement and activity-avoidance in adolescents. It may be that unmeasured confounding variables are responsible for this unexpected finding. Indeed, between-dyads effects are more sensitive to potential confounders than within-dyads effects, and as such, the within-dyads effects may be interpreted with more certainty (see Talloen et al., 2016). Furthermore, our *post hoc* interpretations of between-dyads effects in terms of stable (or persistent) response styles and within-effects in terms of daily variation (or flexibility) in responding in parents are preliminary. Future work should find better ways to examine (in)flexibility in parents' responses to adolescent pain, for instance by examining statistical indicators of daily variability (for an example see Rost et al., 2016). Fourth, we used self-report measures to assess parent and adolescent behavior. Naturally these assessments are sensitive to socially desirable answering and potential memory biases. Future work could consider including observational measures to obtain a less biased, naturalistic assessment of these variables. For instance, an Electronically Activated Recorder (EAR; Mehl, 2017) may be a

useful tool to assess what parents actually say to their adolescent during the day. Fifth, our sample was predominantly female (i.e., 86% girls and 93% mothers). It may be that the parent-adolescent relationships observed in this study are typical for mother-girl dyads. Future research in samples including fathers and adolescent boys with chronic pain would be useful to examine if our findings also hold for the relationship between fathers and daughters, mothers and sons, or fathers and sons. Finally, we did not correct for multiple testing. With eight models that were being tested, there was a potential risk of inflated type I errors (i.e., false positive findings). However, it was evaluated that correction of multiple testing was not appropriate for the present study because our study did not meet any of the conditions required to make such corrections (Perneger, 1998; Rothman, 2015). First, we had priori hypotheses for each of the eight models being tested. Second, we did not repeatedly test the same model in different subsamples. Finally, we favored type I errors in favor of type II errors (i.e., false negative findings). If one would have applied a conservative *post hoc* Bonferroni correction, however, and have tested regression coefficients at the 0.00625 significance level (i.e., 0.05 divided by 8 since eight different models were tested), smaller effects would have been declared non-significant (as indicated in the footnote of **Tables 4, 5**) but the larger effects mostly remained significant. Yet, replication of our findings by future research is warranted before strong conclusions can be made.

CONCLUSION

Our findings support the claim that psychologically flexible parenting and parental pain acceptance indirectly contribute to adolescent outcomes. This was the first study to show how these parent factors predict adolescent pain-related activity-avoidance and activity-engagement on a daily basis, and suggest that this occurs indirectly via its influence on daily parental protective responses and instructions. We provided further support for the adaptive effects of psychologically flexible parenting on adolescent activity-avoidance behavior via fewer protective responses. Parents who are able to display psychological flexibility in parenting may provide their adolescent with more instructions to engage in activities (relative to instructions to avoid). Although these instructions showed short-term adaptive effects on daily adolescent avoidance behavior, our findings also suggest that parents who – on average – provide too many instructions too often might have unintended effects on adolescent behavior

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(i.e., high levels of activity-engagement *and* activity-avoidance). These findings contribute to our understanding of how parental psychological flexibility may impact adolescent functioning in the presence of pain.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Institutional Review Boards at Boston Children's Hospital (IRB#P0020989) and at Stanford University (IRB#39092). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

MB, SH, LS, and LG made substantial contributions to the conception, design, and the acquisition and interpretation of the work. MB and TL have made substantial contributions to the data analysis.

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Child and Family Adaptation to Juvenile Idiopathic Arthritis—A Systematic Review of the Role of Resilience Resources and Mechanisms

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Background: Juvenile Idiopathic Arthritis (JIA) is the most common rheumatic disease in childhood, with chronic pain being a main symptom. JIA symptoms can lead to substantial disability in children and their families. While preliminary evidence reveals the potential beneficial role of resilience in dealing with chronic pain, research on the role of resilience in how families of a child with JIA cope with pain-related symptoms is scant and dispersed.

Objectives: Using the framework of the Ecological Resilience-Risk Model, this review aims to identify (1) family characteristics that are associated with both risk and resilience in children with JIA and (2) the contribution of individual and parental resilience mechanisms and resources to resilience outcomes in children with JIA and their families.

Methods: MEDLINE, EMBASE, EBSCO, Psycharticles, and PsycINFO were systematically searched. Longitudinal, cross-sectional, and treatment studies written in English with a focus on resilience resources and/or mechanisms in families of a child (6–18 years) with JIA were included. The original search (July 2016) produced 415 articles, with a final sample of 6 articles remaining after screening. An updated search (July 2018) did not identify new articles, but identified one extra article through personal communications. The 7 articles were included in a narrative review and study quality was assessed.

Results: Limited research was available on the role of family characteristics, with just one study revealing how family dysfunction is related to reduced child resilience. Studies evaluating the role of individual resilience mechanisms and resources most commonly assessed resilience outcomes in terms of recovery and sustainability outcomes, such as health-related quality of life (HRQL) and functional disability. The findings revealed that children's psychological flexibility, self-efficacy, adherence, pain acceptance, and perceived social support contribute to resilience outcomes. Findings were inconclusive for the influence of coping strategies, such as seeking social support.

Conclusions: While our knowledge is growing, a better understanding of how familial and individual resilience resources and mechanisms influence adjustment to chronic pain as part of JIA is needed and can stimulate development of targeted interventions to enhance outcomes for children with JIA.

Keywords: resilience, chronic pain, juvenile idiopathic arthritis, children, family

INTRODUCTION

Chronic pain, defined as frequent, or recurrent pain that lasts for longer than 3 months (American Pain Society, 2001), is a common condition that occurs regardless of age, sex, or social status (King et al., 2011). In particular, chronic pain is a common symptom of Juvenile Idiopathic Arthritis (JIA), which is the most common rheumatic disease in childhood. JIA is diagnosed in children below 16 years of age when arthritis is identified in at least one joint, for a minimum of 6 weeks (Clinch and Eccleston, 2009; Stinson et al., 2012).

The trajectory of JIA is unpredictable with a wide range of physical (pain, stiffness) and emotional (anxiety, depression) symptoms that can restrict physical and social interactions, thereby potentially inducing functional disability across the lifespan (Sawyer et al., 2005). Indeed, about 30–56% of children with JIA experience continued functional limitations throughout their lifespan (Packham and Hall, 2002). Consequently, a principal aim of multidisciplinary treatment approaches for JIA is to support children in adopting effective coping mechanisms for adjusting to the condition, thereby to facilitating adaptation to JIA (Stinson et al., 2012).

Resilience may be one process that determines whether adjustment difficulties (such as post-traumatic stress symptoms) or positive adaptation (post-traumatic growth) will be observed in response to a major life event. Resilience can be defined as “a dynamic and multi-systemic progression that allows the individual to respond effectively when faced with risk or adversity (e.g. medical condition)” (Cousins et al., 2015). While the process of resilience originates within the individual, social, and environmental factors contribute substantially to the process of resilience. Resilience in the face of a pediatric chronic illness has been operationalized in various ways, all with a focus on demonstrating outcomes such as health-related quality of life (HRQL), in line with or exceeding normative development, despite being faced with managing a chronic illness (Hilliard et al., 2015). Commonly assessed concepts include post-traumatic growth, adaptation, self-esteem, self-concept, optimism, and hope (Cousins et al., 2015).

Within the context of pediatric chronic pain in particular, the recently developed Ecological Resilience-Risk Model (ERRM; Cousins et al., 2015) is based on a growing body of evidence highlighting mechanisms which optimize HRQL in children with chronic pain. The ERRM provides a framework to evaluate the interdependent role of individual and familial resilience and risk factors in adjusting to pediatric chronic pain (Cousins et al., 2015). The ERRM identifies resilience and risk factors as independent but related constructs determining the child's

pain trajectory. Importantly, the ERRM framework distinguishes between resilience mechanisms, defined as dynamic, modifiable processes children, or families engage in as a response to pain experiences (e.g., self-efficacy and pain acceptance), vs. resilience resources, defined as stable individual traits, or familial factors (e.g., optimism, and social support). The framework describes and recognize show both child and parent resilience resources and resilience mechanisms interact to promote resilience outcomes. Resilience outcomes are further categorized as recovery and sustainability (i.e., continued or resumed engagement with daily and valued activities, often assessed in terms of HRQL, and academic success) and growth (i.e., enhanced understanding of their capability, often assessed in terms of benefit finding and posttraumatic growth) (Sturgeon and Zautra, 2013; Cousins et al., 2015; Caes et al., 2018). Risk factors, such as negative affect and poor parental health, and risk mechanisms, including catastrophic thinking, and parental overprotective responses are described in the model as forces that can interfere with resilience resources and mechanisms, thereby influencing resilience outcomes. However, the presence of resilience mechanisms and resources can also buffer against the negative impact of risk factors.

Despite the development of the ERRM and the increased research attention on resilience mechanisms, the available evidence is scattered and many of the relationships suggested in the model are yet to be evaluated in the literature. For instance, the ERRM suggests that family context is an important determinant of a child's resilience outcomes. However, most research exploring the role of family resilience focuses on parental responses to pain, such as parent's psychological flexibility (Caes et al., 2018). While important, such a focus lacks the recognition that families are more than the sum of their parts (Mehta et al., 2009). To gain a true understanding of the role of family resilience on how children deal with chronic pain input from all parties involved (i.e., child, parents, and siblings) on family processes, is required.

To guide future research related to supporting resilience in families living with JIA, it is important to clarify the relationships within the ERRM that have and have not been examined in the literature. This review aims to (1) identify family characteristics that are associated with both risk and resilience in children with JIA and (2) identify the contribution of individual and parental resilience mechanisms and resources to resilience outcomes in children with JIA and their families, using the resilience-risk model for pediatric chronic pain (Cousins et al., 2015) as an organizing framework. By synthesizing existing evidence using the ERRM as an organizing framework, researchers, and intervention developers will be in a position to address gaps in the

evidence to support further research into the design of effective resilience-focused interventions. The findings will be organized and discussed according to individual and parental resilience and risk resources and mechanisms to delineate the independent and overlapping adjustment and adaptation experiences.

METHOD

Systematic Review Protocol

This systematic review of the current evidence on family resilience in JIA, was conducted and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement. The protocol for this review is registered with the International Prospective Register of Systematic Reviews (PROSPERO) database (registration number: CRD42016047226; Saetes et al., 2017).

Search Strategy

A systematic search was conducted in MEDLINE, EMBASE, EBSCO, Psycarticles, and PsycINFO, using the following search terms: (Juvenile Idiopathic Arthritis or JIA or rheumatoid arthritis or systemic-onset Juvenile Idiopathic Arthritis or psoriatic arthritis or enthesitis-related Juvenile Idiopathic Arthritis or oligoarthritis or polyarthritis), (Chronic pain or recurrent pain or pain), (Children or child or adolescence or adolescent or pre adolescence or pediatric or pediatric), (Sibling or family or family function or parent or parenting or parental or peer relationships), (Resilience or resiliency or post traumatic growth or optimism or benefit seeking or benefit finding or coping skills or coping or adjustment or adaptation or health behavior or health behavior or quality of life or hope or psychological resilience or psychosocial functioning or social support or self-concept or acceptance or self-efficacy or positive affect). Other potential sources of relevant literature, known as gray literature, was also reviewed, for example reference sections of relevant publications and conference abstracts.

Study Selection

Studies were included if they were longitudinal, cross-sectional, or treatment studies; written in English; involved young people aged 6–18 years, with a diagnosis of JIA, who were currently undergoing treatment, and were experiencing chronic pain. Studies meeting these inclusion criteria were included in the review regardless of their gender, arthritis type, and type of treatment. Studies were also included if siblings, aged 6–18 years, and parents were part of the study sample. Studies were excluded for the following reasons: review study; full text was not available; evaluation of measurement tools; sample not living with JIA; not a research study; sample outside the age range; or resilience not measured.

Studies identified by the search strategy were exported to an Endnote database for independent review by two of the authors (LH & SS). All results were reviewed by one author (SS), and 20% were reviewed by another author (LH). Duplicates were removed and studies that did not meet the inclusion criteria were excluded in three phases: review of titles,

abstracts, and full texts. After each stage of review, the two authors compared decisions and discussed disagreements until agreement was reached. A third author (LC) was consulted regarding disagreements when an additional perspective was required.

Data Extraction

Based on the review aims, a data extraction table was created to guide the systematic and standardized extraction of data from included studies. Data extraction was completed by the same two authors responsible for the search strategy. Data were extracted related to the year of publication, journal, database, sample (sample size, demographic information for child with JIA, parents & siblings), methodological aspects (study design, analysis, and measurement tools), resilience resources and mechanisms (e.g., pain acceptance, social support), findings related to the impact of resilience in families, and study limitations. The last author (LC) reviewed the extracted information to confirm the accuracy and adequacy of the data extraction process.

Quality Appraisal

The studies included in this review were appraised for quality using a method developed and utilized in a similar systematic review (Alderfer et al., 2010). For the purposes of their review, Alderfer et al. (2010) developed a 9-criteria appraisal tool with a 3-point rating scale, based on published recommendations. The criteria are: explicit scientific context & purpose; methods used; measurement reliability & statistics; statistical power; internal validity; measurement validity; external validity; appropriate discussion; contribution to knowledge. Based on reports under each criterion, included studies are rated as low (1), medium (2), or high quality (3).

Data Synthesis

A narrative synthesis of the findings extracted from the studies included in this review was chosen as the most appropriate method of analysis. This method for analyzing the findings of systematic reviews aims to identify themes and patterns across studies to present an overview of the evidence, which goes beyond description of the individual studies (Popay et al., 2006). The included studies approached the conceptualization and measurement of risk and resilience factors related to adaptation to JIA in different ways, using different designs. Therefore, meta-analysis of quantitative findings was not possible. Guidelines on narrative synthesis were used to guide the organization, analysis, and reporting of the findings in this review (Popay et al., 2006). The findings of included studies related to the review aim were summarized in the data extraction table. Using the Ecological Resilience-Risk Model (Cousins et al., 2015) as a framework, the findings were categorized. Similarities and difference across studies, and patterns of relationships between resilience resources and mechanisms, and resilience outcomes were identified and described.

RESULTS

Study Selection and Characteristics

Databases were first searched in July 2016, and 414 results were retrieved and added to the Endnote database for review. The PRISMA flowchart (**Figure 1**) presents the results of each stage of the systematic review process. After duplicates were removed, 410 results remained. As a result of review of titles and abstracts, 338 manuscripts were excluded, leaving 72 manuscripts for full text review. Comparisons of review decisions made by the two authors showed high levels of agreement (98% for title and abstract review; 100% for full text review). Review of full texts resulted in the exclusion of 66 manuscripts and six manuscripts to be included in the analysis. The search strategy

was conducted again in July 2018 but no newly published literature was eligible for inclusion in this review. However, one additional eligible manuscript was identified through another source (personal communication) in July 2018. In sum, of the 458 studies screened, seven met the inclusion criteria and were analyzed in a narrative synthesis (Timko et al., 1993; Frank et al., 1998; Sawyer et al., 2004, 2005; Connelly, 2005; Seid et al., 2014; Beeckman et al., 2018). **Table 1** provides details on study design, samples, measurement tools, and quality.

The Ecological Resilience-Risk Model (Cousins et al., 2015) was used as a framework for examining and categorizing the findings of the studies included in this review, with respect to the two primary research questions: (1) identify family characteristics that are associated with both risk and resilience in children with

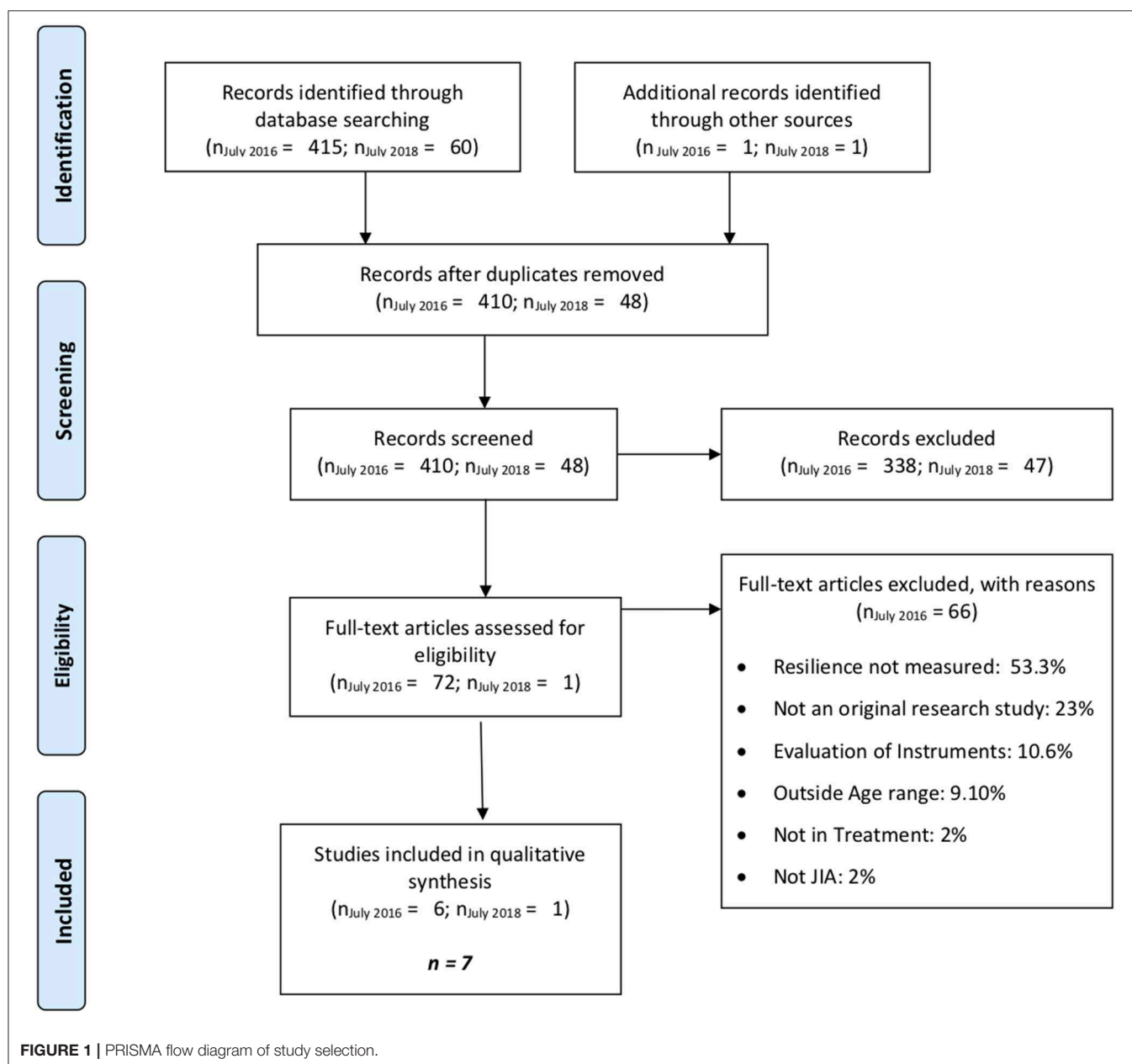


TABLE 1 | Characteristics of studies.

References	Country	Design	Sample size (% female) Mean age of pediatric sample (SD)	Resilience resources & mechanisms are measured	Outcomes related to review aims	Quality appraisal rating
(Beeckman et al., 2018)	Belgium	Cross-sectional questionnaire study	59 (61%) 13.76 year (2.67)	<i>Resources:</i> Positive & negative affect <i>Mechanisms:</i> Child general psychological flexibility and pain acceptance, parent general and pain related psychological flexibility	Individual and parent resilience mechanisms directly and indirectly associated with resilience outcomes (QoL/functioning, mood/affect), and can buffer to reduce risk associated with pain intensity.	2
(Seid et al., 2014)	United States	Prospective longitudinal cohort study	230 (69.1%) 9.42 year (4.49)	<i>Resources:</i> Social support, family climate & relationships <i>Mechanisms:</i> Symptom-related self-efficacy, parental distress, coping strategies	Proxy report HRQOL was explained by family risk mechanisms, while self-reported HRQOL was strongly predicted by family/social resilience resources and individual resilience mechanisms.	2
(Connelly, 2005)	United States	Cross-sectional questionnaire study	47 (69%) 9.8 year (1.72)	<i>Resources:</i> Family functioning, Hope <i>Mechanisms:</i> None	No relationship between individual or family resilience resources and resilience outcome of recovery/sustainability.	3
(Sawyer et al., 2005)	Australia	Prospective longitudinal study	54 (57.4%) 12.8 year (3.3)	<i>Resources:</i> None <i>Mechanisms:</i> Coping strategies	Use of individual resilience mechanisms (child pain coping strategies), have a significant impact on resilience outcome, QoL (Recovery/sustainability), but not always positive. Coping does not appear to mediate between child's experience of pain and HRQL.	2
(Sawyer et al., 2004)	Australia	Cross-sectional questionnaire study	59 (59.3%) 12.6 year (3.3)	<i>Resources:</i> None <i>Mechanisms:</i> Coping strategies	Use of more positive individual resilience mechanisms (i.e., child pain coping strategies) was associated with better resilience outcomes (QoL—Recovery/sustainability), according to parents and children.	2
(Frank et al., 1998)	United States	Longitudinal cohort study	27 (70.4%) 5.52 year (4.48)	<i>Resources:</i> Family adaptability and cohesion, child functioning <i>Mechanisms:</i> Parental distress, parental coping strategies	Parental risk mechanisms associated with child resilience outcomes (recovery/sustainability)	2
(Timko et al., 1993)	United States	Longitudinal cohort study	172 (64.5%) 12.6 year (not reported)	<i>Resources:</i> Social & community support <i>Mechanisms:</i> Positive social interaction, coping strategies	Family (mother and father distress) risk mechanism associated with poorer resilience outcomes	2

JIA and (2) identify the contribution of individual and parental resilience mechanisms and resources to resilience outcomes children with JIA and their families?

The majority of the findings extracted from the included papers related to the contribution of resilience to outcomes in children with JIA and their parents. Siblings were not included as participants in any of the studies reviewed. Therefore, no conclusions could be made on sibling resilience or the impact of resilience on outcomes such as quality of life in this group.

Quality Appraisal

The quality of the studies included varied, each having strengths and weaknesses that affect the overall quality of this review. Overarching weaknesses include generally homogenous samples, over-reliance on parent-proxy reports, and cross-sectional designs. General strengths of the individual studies include use of validated measures, some longitudinal designs (see e.g., Timko et al., 1993; Frank et al., 1998; Seid et al., 2014) and successful recruitment of fathers as well as mothers (see e.g., Timko et al., 1993), see **Table 1** for the quality assessment for each included study. In relation to this review, the most significant challenges to drawing overarching conclusions from the body of literature related to a lack of uniformity across the studies in terms of the variables measured and reported, different study designs, and differences in demographic and disease-related variables reported.

Narrative Synthesis Findings

Only one study (Connelly, 2005) reported on possible associations between family characteristics and risk or resilience in children with JIA. According to Connelly (2005), there was a negative association between family risk factors and child resilience resources. This cross-sectional study of 68 children with Juvenile Rheumatoid Arthritis (JRA), and their parents assessed family functioning (parent-proxy report, pediatric quality of life (child self-report and parent-proxy report), and children's hope (child self-report). Higher levels of parent-reported family dysfunction were significantly associated with lower levels of child-reported hope among children ($r = -0.35$, $p < 0.05$).

In relation to the second primary research question, the findings of this review provide some tentative support for aspects of the Ecological Resilience-Risk Model (Cousins et al., 2015), see **Figure 2** for a full overview. The findings suggest that child and parent resilience and risk mechanisms which were measured considerably more frequently than resources and risk factors, may influence resilience outcomes. The most common resilience outcomes measured in the studies included in this review can be characterized as recovery and sustainability outcomes, namely, HRQL and functional disability.

RESILIENCE MECHANISMS

Resilience mechanisms are generally active and dynamic cognitions and behaviors, enhanced by resilience resources, such as social support, which overcome risk factors, and risk mechanisms (Cousins et al., 2015). In this review, three studies

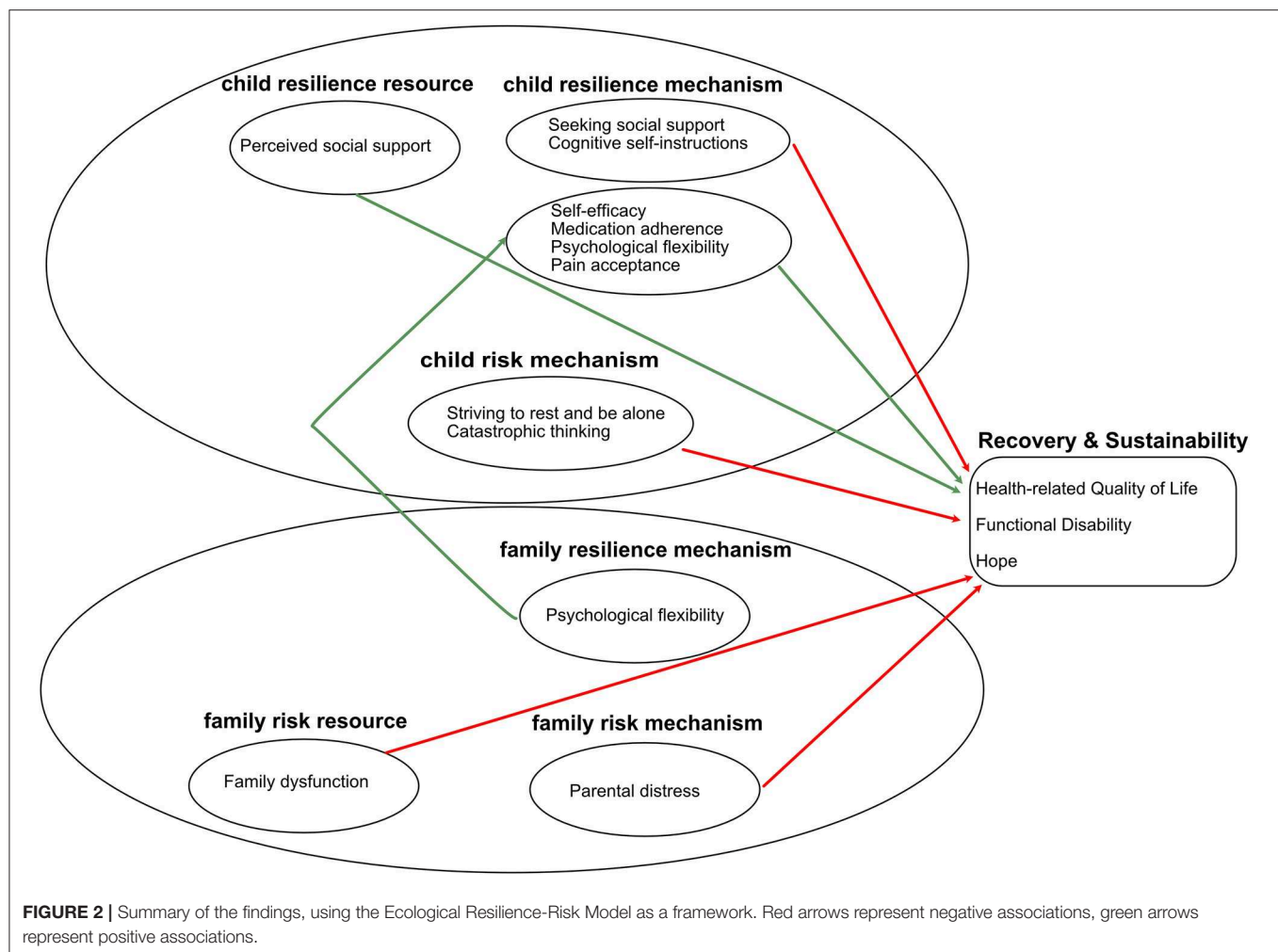
(Sawyer et al., 2004; Seid et al., 2014; Beeckman et al., 2018) reported positive relationships between resilience mechanisms, such as coping and psychological flexibility, and outcomes. All three studies included data from parents and children with JIA; however, the Beeckman et al. (2018) and Seid et al. (2014) studies collected data on parent resilience mechanisms, while Sawyer et al. (2004) focused on parent-proxy measurement of variables related to their child's resilience and outcomes.

Sawyer et al. (2004) examined child and parent reports of the use of different pain coping strategies. According to parent-proxy reporting in this study, positive pain coping strategies such as *problem solving/self-efficacy* were associated with less functional disability. As illustrated in the next section, coping strategies acted as risk mechanisms more frequently than resilience mechanisms.

According to Beeckman et al. (2018), child and parental psychological flexibility may support adaptive functioning JIA. Children's general psychological flexibility and pain acceptance were significantly associated with functional outcomes. For example, child psychological flexibility was associated with better psychosocial health (PedsQL emotional, social, and school functioning) ($B = 0.34$, $p < 0.01$) and less negative affect ($B = -0.60$, $p < 0.01$), but was not associated with better physical health or higher levels of positive affect. Child pain acceptance appeared to play an importance role in relation to resilience outcomes, including better psychosocial ($B = 0.31$, $p < 0.05$) and physical health ($B = -0.45$, $p < 0.001$) and less negative affect ($B = -0.32$, $p < 0.05$). Higher levels of pain intensity was associated with disability in this study. Higher levels of child pain acceptance, but not psychological flexibility, appeared to act as a resilience mechanism, and was associated with lower risk of disability among children with high pain intensity in this study.

The potential impact of parent resilience mechanisms on child resilience outcomes was also demonstrated in this study. Although direct effects of parent general and pain-related psychological flexibility were not found, both parent and child resilience mechanisms were indirectly related to better child resilience outcomes. For example, parent's general psychological flexibility was significantly associated with their children's psychological flexibility, which was associated with better psychosocial outcomes and positive affect. Thus, these findings underscore the complex interrelations between parent and child resilience resources and mechanisms, with parent resilience mechanisms influencing child resilience outcomes, via their association with children's resilience mechanisms (flexibility) and resources (affect) (Beeckman et al., 2018).

The focus of the study by Seid et al. (2014) was the predictive ability of non-medical variables (e.g., coping and parental distress) in relation to HRQL. The findings suggest that children rated the impact of non-medical variables on HRQL as being greater than parent-rated impact. For child self-reported HRQL, self-efficacy and adherence to medication made a positive significant contribution. Perceived social support was also positively associated with child self-reported HRQL, and is one of the few resilience resource variables measured by studies in this review. A significant association was also reported between



adherence to medication and HRQL according to parental-proxy measures.

RISK MECHANISMS

Family and individual risk mechanisms were examined in five studies included in this review (Timko et al., 1993; Frank et al., 1998; Sawyer et al., 2004, 2005; Seid et al., 2014). According to the ERRM, risk mechanisms interfere with the pathway to adaptation involving resilience resources and mechanisms, and are enhanced by risk factors.

This review suggests that parental distress is a family risk mechanism that may negatively affect child resilience outcomes (Timko et al., 1993; Frank et al., 1998; Seid et al., 2014). In one study, parental emotional distress was associated with poorer parent-proxy reported child HRQL (Seid et al., 2014). Frank et al. (1998) reported that parental distress at baseline was significantly associated with adaptation to JIA, assessed based on number of swollen joints over 18 months, an indicator of the activity or status of JIA. Higher levels of parental distress in this study were associated with a higher number of swollen joints, suggesting that

parental distress may act as a risk mechanism, hindering child adaptation to JIA. Similarly, according to Timko et al. (1993), distress in mothers and fathers was associated with higher levels of functional disability in children.

Three studies (Sawyer et al., 2004, 2005; Seid et al., 2014) reported findings suggesting negative relationships between coping strategies and outcomes. The findings of this review suggest that, when examined in detail, coping strategies frequently act as risk mechanisms. This review also demonstrates considerable disagreement between parents and children in assessments of child coping strategies for JIA.

Seid et al. (2014) reported that, according to child self-report, the use of the coping strategy “catastrophizing” by children, had a significant negative relationship with HRQL. Parental distress and report of use of catastrophizing by their children was also reported to have a significant negative relationship with child HRQL.

According to parent-proxy reporting in the Sawyer et al. (2004) study, higher usage of some pain coping strategies such as *strive to rest and be alone* are associated with poorer child HRQL. Parents in this study rated problem-solving/self-efficacy as the most common pain coping strategy used by their children.

However, parents in the study by Sawyer et al. (2005) identified “seeking social support” and “striving to rest and be alone,” as the most frequently used coping mechanisms by their children. According to parent-proxy reports, both of these pain coping mechanisms are associated with poorer physical and emotional functioning. A significant negative association was also found between parent-proxy reports of child pain coping (*seeks social support* and *striving to rest and be alone*) and the daily activities and treatment subscales of HRQL (Sawyer et al., 2005).

In contrast with parent-proxy reports of child coping, children themselves rate cognitive self-instruction (e.g., a child resilience mechanisms in which children imagine they are not in pain, or use positive self-talk related to response to pain) as the most frequently used strategy to cope with pain. Sawyer et al. (2004) reported only significant negative associations between child self-reported pain coping strategies and HRQL. For example, cognitive self-instruction, a child resilience mechanism, was associated with poorer physical functioning. Similarly, seeking social support, also considered a child resilience mechanism, was associated with poorer emotional and social functioning, as well as lower scores on the daily activities and disease-specific treatment subscales. In their 2005 study, Sawyer described similar significant negative associations between child self-reported pain coping strategies and HRQL. In comparison to parent-proxy reporting in this study, negative associations were more commonly reported by children in this study. However, after controlling for pain intensity ratings, the most consistent relationship was found with seeking social support, which was negatively associated with almost all aspects of HRQL, including daily activities, treatment, worry, and physical, emotional, and social functioning.

DISCUSSION

Using the ERRM for pediatric chronic pain (Cousins et al., 2015) as an organizing framework, this review suggests that both individual and family mechanisms are important in determining outcomes and that parents and children are having different experiences in relation to adaptation to JIA. The findings are mostly in accordance with the assumptions of the ERRM and can be summarized as follows. Limited evidence was identified with respect to possible relationships between family characteristics and resilience outcomes. The one study in the review that did address this study aim reported an association between family dysfunction and lower child resilience resources. With respect to resilience mechanisms and resources, the findings provide evidence for significant contributions of several child resilience mechanisms (i.e., self-efficacy, psychological flexibility, pain acceptance, and medication adherence) and resources (i.e., perceived social support) in explaining recovery and sustainability (i.e., HRQL and functional disability). With respect to the influence of child risk mechanisms, evidence was mixed but support was found for children’s levels of catastrophic thinking and adopting striving to rest and be alone as a coping mechanism. Furthermore, the results provide support for the important role of family resilience mechanisms (i.e., parental psychological flexibility) and risk mechanisms (i.e., parental distress) as significant contributors to their child’s recovery and

sustainability. However, different patterns of relationships were identified depending on who (parent proxy report vs. child self-report) was reporting on children’s HRQL. This lack of agreement or overlap in parent proxy or child self-report is not unique, and often identified within the literature on children’s HRQL. Indeed, parent and child agreement in relation to HRQL is moderate to low and tends to diminish as children age (Rajmil et al., 2013). The complexity of HRQL-reporting illustrated in this review and other previous research emphasizes the importance of gathering data from a range of members of a family unit to understand family resilience in the face of JIA (Mehta et al., 2009). Furthermore, most studies did not look at the full range of potential resilience outcomes but were restricted in relying on HRQL as a resilience outcome. As a result, the big picture in terms of the individual and interacting roles of resilience and risk factors in influencing resilience outcomes cannot be concluded from any one study in this review. Nevertheless, some interesting patterns of key resilience resources and mechanisms were identified.

The findings highlight that parental distress in response to child pain experiences in the context of JIA can be considered a family risk mechanism that negatively impacts child resilience outcomes. Within the broader pediatric chronic pain literature, the recent review by Palermo et al. (2014) summarizes a substantial body of evidence supporting the interrelation between child pain experiences and parental distress. For instance, this review by Palermo et al. (2014) highlights that a considerable number of parents of children with chronic pain experience clinically relevant levels of distress, which has been shown to be negatively related to child pain outcomes, such as increased pain intensity, disability, and distress (Palermo et al., 2014). The findings from our systematic review add to this growing literature by identifying that parental distress also represents a risk factor to children’s broader adjustment to JIA by negatively impacting child HRQL, number of swollen joints, and functional disability. Such evidence emphasizes the need to actively involve parents within multidisciplinary treatment approaches for JIA. Providing parents with adaptive coping mechanisms may support them to deal with their own emotional difficulties in response to their child’s diagnosis of JIA and associated symptoms. Several parent-focused interventions have been developed in the context of pediatric chronic pain, such as parental problem solving skills training (Palermo et al., 2016). Use of the ERRM for pediatric chronic pain (Cousins et al., 2015) to optimize and integrate such treatment approaches within the care plan for families of a child with JIA may play an important role in supporting child and family resilience. Our findings highlight that the ERRM can be a useful framework to guide such intervention development because it takes the individual and family into account, as well as stable and dynamic characteristics and processes relevant to promoting resilience in the context of JIA.

Although a wide range of evidence was obtained and analyzed with respect to child resilience mechanisms and resources, studies evaluating the role of child coping mechanisms provided equivocal findings. In particular, the coping strategies “seeking social support” and “cognitive self-instructions,” typically considered adaptive coping strategies or resilience mechanisms, were found to be related to poorer functioning and HRQL

in this review. A potential explanation for these unexpected findings could be that while, in principle the coping strategies are considered to be adaptive or promoting resilience, the specific relation to child's HRQL does depend on the child's developmental age and the exact way the child is engaging with this strategy. Although evidence indicates that positive peer relationships can strengthen perceived social competence and development in children with chronic pain (Forgeron et al., 2011), not all children's peer relationships are necessarily of a positive and supportive nature. Consequently, the social support a particular child receives might be dependent on the particular characteristics of each relationship, the child's context, level of adjustment or adaptation to JIA, and support needs at any one time. Similarly, to assess the effectiveness of cognitive self-instruction among children, focusing on frequency of cognitive self-instruction alone is insufficient, and the child's capacity to engage positively in this coping mechanism must also be known. These findings with respect to role of coping further support the call made by Van Damme et al. (2008) to step away from traditional categorical approaches toward pain coping strategies (i.e., adaptive vs. maladaptive coping) and adopt a motivational approach. This proposed motivational approach focuses on evaluating the function of each coping strategy in its particular context to determine to what extent the coping mechanism does or does not facilitate adaptation for each individual. Consequently, coping strategies are not inherently a resilience or risk mechanism, but their adaptive function depends on the extent the coping mechanism promotes resilience for a particular child given their unique situation and developmental capacities. Adopting such a motivational perspective on coping within the ERRM framework could allow for better insight into how child's resilience mechanisms, such as coping strategies, develop, and thereby provide more clarity on their complex impact on resilience outcomes. Such insights have the potential to guide the generation of interventions aimed at promoting resilience in a personalized and developmentally appropriate manner.

The conclusions from this review need to be interpreted in light of several limitations. Importantly, drawing strong conclusions is hampered by the limited evidence identified to include in this systematic review. Furthermore, the quality assessment of the studies included in this review highlighted issues and variations in methodological quality. In particular, the majority of the studies based their findings on cross-sectional designs in homogenous samples and were overly reliant on parent-proxy reports. Importantly, no studies were identified for inclusion in the review that investigated the impact of JIA on siblings and the role of siblings' resilience resources and mechanisms. Therefore, to expand our understanding of family resilience in the context of JIA, future studies must examine important variables from the perspectives of multiple family members, including children with JIA and their siblings. Research that is longitudinal in nature and includes heterogeneous populations are called for. Furthermore, we could only include quantitative studies in this review. A more complete understanding of the development and influence of resilience resources and mechanisms will be made possible by the use

of different approaches, including more intervention research, research using mixed methods and qualitative approaches.

Despite these limitations, the findings do provide preliminary insight into the application of the ERRM to understand which individual and family psychological processes may influence resilience in children with JIA. Given the limited empirical evidence for these resilience mechanisms and resources in the broader literature on pediatric chronic pain, a similar review with respect to resilience in the context of any pediatric chronic pain experience might be warranted to move this field forward. Our findings also highlight some limitations and challenges of the ERRM. In particular, the distinction between resilience mechanism and outcomes is not always straightforward and mostly depends on the specific operationalisation of these constructs within a study. For instance, Beeckman et al. (2018) considered affect to be a resilience outcome, whereas this is considered a resilience resource according to the ERRM. Similarly, Seid et al. (2014) considered adherence or self-management behaviors as resilience mechanisms, while appropriate adherence could also be a sign of having adapted to life with JIA and hence represent a resilience outcome. More theoretical developmental research is needed to enhance our understanding of these resilience pathways in the context of childhood chronic illness. As the ERRM framework was developed based on an adult framework, it is possible that these distinctions between mechanisms and outcomes are clearer in adulthood but are less distinct in childhood. Furthering of our theoretical understanding of family resilience and the role of both individual and familial resilience pathways is needed to inform the development and refinement of targeted interventions to enhance clinical practice and interventions aimed at fostering resilience in all family members of a child diagnosed with JIA.

AUTHOR CONTRIBUTIONS

LH assisted SS in the study selection and data extraction. In the write up of the manuscript, LH took charge of writing up the methods and results, and provided feedback on the introduction and discussion. SS took charge of designing the systematic review, the study selection process and data extraction and wrote a first draft of the methods and results section, which was used by LH to write up these sections, and also provided feedback on the full version of the manuscript. BM supervised the study selection and data extraction, conducted by SS and LH and provided feedback on the full version of the manuscript. LC supervised the study selection and data extraction, conducted by SS and LH. LC was actively involved in the write up of the data, taking charge of writing the introduction and discussion, as well as providing feedback on the write up of the methods and the results.

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Beyond Adaptive Mental Functioning With Pain as the Absence of Psychopathology: Prevalence and Correlates of Flourishing in Two Chronic Pain Samples

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Chronic pain outcomes are traditionally defined in terms of *disability* and *illness*. A definition of adaptive functioning in the context of chronic pain beyond the mere absence of negative outcomes, is the ability to *flourish* (i.e., experience emotional, psychological and social *well-being*; Keyes, 2002). We explored in two chronic pain samples the prevalence and sociodemographic, physical and psychological correlates of flourishing, and complemented this exploration with a similar examination of (*being at risk for*) *psychopathology* to help contextualize findings. Sample 1 ($n = 1498$) was a nationally representative sample. Subgroups included people with regular joint pain (1), regular joint pain and rheumatoid arthritis (2) and without chronic pain (3). Using chi-square tests we calculated the prevalence of both mental health outcomes and examined if people with or without chronic pain were more/less likely to flourish/at risk for psychopathology. Sample 2 ($n = 238$) concerned baseline data of a Randomized Controlled Trial on the effectiveness of Acceptance and Commitment Therapy for chronic pain (Trompetter et al., 2015b). We performed logistic regression analysis to identify flourishers/those at risk for depression. The Mental Health Continuum-Short Form was used to measure flourishing. The prevalence of flourishing was 34% (recurrent joint pain) and 38% (recurrent joint pain and arthritis) in sample 1, and 23% in sample 2. Compared to those without chronic pain, people with chronic pain were as likely to flourish, but more likely to be at risk for psychopathology. In sample 2, both flourishing and being at risk for depression were related foremost to *psychological* correlates. While engaged living was the most important correlate of flourishing, pain catastrophizing and psychological inflexibility were most important correlates of being at risk for depression. In conclusion, people with chronic pain *are able* to flourish. Findings suggest that positive and negative chronic pain outcomes function on two different continua, with potentially unique protective and risk factors. The Psychological Flexibility model provides pathways to explain both poor and optimal functioning in the presence of chronic pain. A better understanding of people with chronic pain who are able to flourish can be a fruitful endeavor to improve chronic pain models and interventions.

Keywords: chronic pain, positive mental health, flourishing, depression, psychological flexibility, values, fear avoidance, resilience

INTRODUCTION

Traditionally we study the effects of chronic pain on health and well-being from a medical, disease-oriented approach. Chronic pain outcomes are defined in terms of *disability*, *poor* functioning, and *illness*. It is well-established that chronic pain is associated with a variety of such negative outcomes like depression (Breivik et al., 2006; Miller and Cano, 2009; de Heer et al., 2018). Healthy, adaptive functioning with persistent pain is considered to be achieved when these negative outcomes are absent. Subsequently, we primarily study and target involved *risk* processes like pain-related fear and pain catastrophizing (Sullivan et al., 1995; Gatchel et al., 2007; Jensen and Turk, 2014). In contrast, relatively few studies examined the unique involvement of *protective* processes like optimism, positive affect, purpose in life and social support in adaptive functioning with pain (but see, e.g., Karoly and Ruehlman, 2006; Smith and Zautra, 2008; Verduin et al., 2008; Finan et al., 2013; Boselie et al., 2014). Scholars proposed that more focus on protective factors is necessary. Additionally, they suggested to develop alternative conceptualizations of chronic pain *outcomes* that define adaptive functioning as more than the mere absence of negative outcomes (Sturgeon and Zautra, 2010; Goubert and Trompetter, 2017). Based on the work of Keyes (2002) and Goubert and Trompetter (2017) proposed the study of *positive mental health* in the context of pain. Optimal levels of positive mental health entail the experience of high levels of emotional well-being (presence of positive feelings like happiness), psychological well-being (optimal functioning in life through, e.g., purpose in life and positive social relationships) and social well-being (optimal social/community functioning through, e.g., social integration). Keyes conceptualizes those functioning with optimal levels of emotional as well as psychological and social well-being as *flourishing*. He considers *complete mental health* to be achieved when someone is not suffering from mental illness and is simultaneously flourishing. To measure the construct of flourishing, Keyes developed and validated the Mental Health Continuum-Short Form (MHC-SF) (Keyes et al., 2008; Lamers et al., 2011). First studies suggest that negative (pain disability, depression) and positive (emotional, psychological well-being) chronic pain outcomes are only moderately related to each other (Mangelli et al., 2002; Schleicher et al., 2005; Huber et al., 2008). This mirrors the outcomes of a larger body of studies in general and psychiatric populations, showing that mental illness and positive mental health measured with the MHC-SF or similar instruments are associated, but *different* constructs (Huppert and Whittington, 2003; Keyes, 2005b; Westerhof and Keyes, 2009; Weich et al., 2011; Trompetter et al., 2017). This implies that positive and negative chronic pain outcomes and their underlying risk and protective factors are not two sides of the same coin (Goubert and Trompetter, 2017). Understanding if there is a subgroup of chronic pain sufferers that is able to flourish, and unraveling the factors that differentiate the flourishers from those who do not flourish might prove a fruitful endeavor. It may broaden our current theoretical models that explain chronic pain adaptation primarily in terms of unsuccessful adaptation and risk. Hereby we can expose new routes for (psychological)

treatment to support successful chronic pain adaptation. In this study we examine the prevalence and correlates of flourishing in people with chronic pain. What knowledge currently exists? Two large, representative national studies using the MHC-SF report flourishing prevalence rates of 18 and 37% respectively in the general adult population in the United States (Keyes, 2005b) and Netherlands (Schotanus-Dijkstra et al., 2016). Only two studies directly addressed *flourishing* in pain samples, and compared this to the likelihood to flourish in non-pain samples. Using data from the Canadian Community Health Survey, Gilmour (2015) identified that the people who 'usually experienced severe levels of pain/discomfort,' were significantly less likely to flourish compared to those with mild levels of pain/discomfort. Similarly, Keyes (2005a) showed that suffering from chronic physical conditions – including specific subgroups with chronic back pain and/or arthritis – was significantly and negatively associated with flourishing. First studies that assessed aspects of positive mental health like happiness or purpose in life also indicate that people with chronic pain experience lower levels of well-being than people without chronic pain (Schleicher et al., 2005; Finucane et al., 2012). To our best knowledge, no studies exist that examine the correlates of flourishing in people with chronic pain. We perform *post hoc* analyses on two existing datasets, each including different chronic pain samples. A first aim is to explore in a Dutch nationally representative sample the prevalence of flourishing in people with chronic pain, in comparison to people without chronic pain (Study 1). A second aim is to explore in another sample – that consists of people with chronic pain with different underlying etiologies seeking psychological help – the prevalence of flourishing, and identify sociodemographic, physical and psychological correlates of flourishing in the context of chronic pain (Study 2). In integration these two studies build on existing prevalence studies (Keyes, 2005a; Gilmour, 2015) by assessing different chronic pain samples and/or different cultural contexts. The second dataset provides us with the opportunity to assess key correlates from two leading theoretical models explaining chronic pain disability – the Fear Avoidance model of chronic pain (FA model: Crombez et al., 2012) and the Psychological Flexibility model of chronic pain (PF model: McCracken and Vowles, 2014) – in relation to flourishing. In *both* studies we complement the examination of the prevalence and correlates of flourishing with an examination of the prevalence and correlates of (*being at risk for*) *psychopathology*. This will help readers to contextualize the findings on flourishing as a positive chronic pain outcome, with psychopathology as a more familiar negative chronic pain outcome. Additionally, it provides an examination of the postulation that psychopathology and flourishing and their underlying correlates are related, but different. Our first hypothesis is that chronic pain is associated with a lower likelihood to flourish and higher likelihood to be at risk for psychopathology compared to people without chronic pain (Study 1: Keyes, 2005a; Miller and Cano, 2009; Gilmour, 2015; de Heer et al., 2018). We do expect, however, that a proportion of people with chronic pain *will* flourish in both studies. In Study 2, we examine the correlates of flourishing in the context of chronic pain. Overall, *psychological* correlates are expected to show stronger associations with the included

mental health outcomes than *sociodemographic* and/or *physical* correlates like pain intensity and pain disability (Keyes, 2005b; Schleicher et al., 2005; Vowles et al., 2007; Huber et al., 2008; McCracken and Gutiérrez-Martínez, 2011; Schotanus-Dijkstra et al., 2016). More specifically, *psychological* correlates that will be assessed include pain catastrophizing, psychological inflexibility, mindfulness and engaged living. Pain catastrophizing is a key construct in the FA model of chronic pain (Crombez et al., 2012). Psychological inflexibility, mindfulness and engaged living are key constructs in the PF model of chronic pain (McCracken and Vowles, 2014). These models underlie Cognitive Behavioral Therapy and Acceptance and Commitment Therapy for chronic pain, respectively (Ehde et al., 2014; Veehof et al., 2016). The FA model has been developed theoretically in line with a traditional medical view of chronic pain disability. As, furthermore (aspects of) catastrophizing thinking styles are an important risk factor for depression in- and outside the context of chronic pain (Vowles et al., 2007; Aldao et al., 2010; Crombez et al., 2012), we expect that pain catastrophizing is associated primarily to being at risk for psychopathology. In contrast, the PF model is most aligned with positive mental health of both models, through its central focus on long-term engagement with personally valued activities of the person with persistent pain (Fledderus et al., 2012; McCracken and Vowles, 2014). We therefore particularly expect that engaged living is an important correlate of flourishing in people with chronic pain. Finally, psychological inflexibility is expected to be associated with both mental health outcomes but most strongly to being at risk for psychopathology. Although it is a central process within the more positive PF model, the construct as operationalized in this study – experiential avoidance of pain and cognitive fusion with pain-related thoughts and feelings – particularly relate(s) to pain catastrophizing and negative pain-related outcomes (Aldao et al., 2010; Wicksell et al., 2010b; McCracken and Vowles, 2014; Trompetter et al., 2015a).

STUDY 1

Materials and Methods

Participants and Procedure

In this paper, we make use of data of the LISS (Longitudinal Internet Studies for the Social sciences) panel administered by CentERdata (Tilburg University, Netherlands). The LISS panel consists of a representative sample of 5000 Dutch households that were randomly selected from municipal registers in the Netherlands. We used data from a specific study module on mental health and flourishing (Lamers et al., 2011, 2015). In one third of the households, one member was selected by CentERdata to fill out this module ($n = 1662$, response rate 69%). To this dataset we linked data from a LISS core module on health that contained data on pain and relevant health-related variables, and sociodemographic variables (final $n = 1498$). All questionnaires were administered between November and December 2007. People were categorized for this study as having chronic pain when they ‘regularly suffered from back-, knee-, hip-pain or pain in any other joint’ (47.4%, $n = 710$), and compared to

those without chronic pain (52.6% of total sample, $n = 788$). As we could not ensure that all included participants in this generic pain sample would adhere to important classifications for chronic pain (e.g., a minimal duration of 3 or 6 months), we also more narrowly pinpointed this generic chronic pain sample by extracting the people that both ‘regularly suffered from back-, knee-, hip-pain or pain in any other joint’ and had ‘a diagnosis of arthritis, including osteoarthritis, rheumatism, or osteoporosis’ (7.5% of total sample, $n = 113$).

Measures

Sociodemographic and health-related characteristics

Sociodemographic characteristics were age, gender, educational level and marital status. Health-related characteristics were the number of comorbid somatic conditions/health risks (e.g., diabetes, cancer, cardiovascular disease, COPD/asthma, high cholesterol, or blood pressure) and the number of comorbid recurrent physical symptoms (e.g., fatigue, sleep problems, stomach, or intestinal problems).

Positive mental health

Positive mental health was measured with the Mental Health Continuum-Short Form (MHC-SF) (Keyes et al., 2008; Lamers et al., 2011) which consists of 14 items (Keyes, 2002). Respondents rate the frequency of every feeling in the past month on a six-point Likert scale (never, once or twice a month, about once a week, two or three times a week, almost every day, every day). Example items of the three types of well-being are: “During the past month, how often did you feel: ... interested in life? (emotional well-being, three items); ... that you had experiences that challenged you to grow and become a better person? (psychological well-being, six items); ... that you had something important to contribute to society?” (social well-being, five items). The MHC-SF showed high internal consistency in this sample ($\alpha = 0.90$). Respondents can be categorized into two groups: flourishing or not flourishing. Respondents are categorized as flourishing when they score one of the three items on the emotional well-being subscale as ‘every day’ (6) or ‘almost every day’ (5), and rate at least 6 of the 11 items on the psychological and social well-being scale as ‘every day’ (6) or ‘almost every day’ (5) (Keyes, 2002). The Dutch version has good psychometric properties in both general and psychiatric samples (Lamers et al., 2011, 2012; Franken et al., 2018).

Psychopathological symptoms

The Brief Symptom Inventory (BSI; Dutch version) is a 53-item instrument for screening and assessment of psychopathology (De Beurs and Zitman, 2005). Respondents indicate the degree to which they experienced various psychological symptoms in the past week using a five-point Likert scale, ranging from 1 (not at all) to 5 (a lot). The BSI includes nine subscales: Depression, Anxiety, Phobic Anxiety, Interpersonal Sensitivity, Psychoticism, Paranoid Ideation, Hostility, Obsessive-compulsive Complaints and Somatization. The BSI showed high internal consistency in the present study ($\alpha = 0.94$). Respondents can be categorized in two categories using a cutoff of 0.50: with or without being at risk for psychopathology.

Statistical Analyses

The Statistical Program for Social Sciences (IBM SPSS) version 24.0 was used for all statistical analyses. We used an alpha level of 0.05 for all statistical tests. Frequency distributions were calculated and descriptive analyses were performed to summarize sample characteristics of three groups beyond the total sample ($n = 1498$): People with chronic pain ($n = 597$), people with chronic pain in the context of arthritis ($n = 113$), and people without chronic pain ($n = 788$). Chi-square tests and independent t -tests were used to compare sample characteristics for the two subsamples with chronic pain with the subsample without chronic pain. Normality tests showed that three outliers were present for the variable 'number of comorbid somatic conditions/health risks' (reporting ≥ 12 comorbid somatic conditions). As findings were similar for data with these outliers removed and the full sample, only the outcomes for the full sample were reported. To examine the prevalence of *flourishing* for individuals within the two subsamples with chronic pain in comparison to individuals in the subsample without chronic pain, we first determined for each subsample the percentage of respondents in each category of positive mental health (i.e., flourishing, or not) and risk for psychopathology (i.e., at risk, or not). Hereafter, a chi-square test was performed to examine the prevalence of flourishing for individuals with chronic pain in comparison to people without chronic pain. Similarly, a chi-square test was performed to examine the prevalence of being *at risk for psychopathology* for individuals with chronic pain in comparison to people without chronic pain. All expected cell counts during chi-square tests were greater than five.

Results

Overall, Chi-square tests and independent t -tests used to compare sample characteristics for the groups with and without chronic pain (Table 1) showed that both subsamples with chronic pain significantly differed from the group without chronic pain on all included sociodemographic and health-related characteristics (all p 's < 0.05). Compared to participants without chronic pain, people with chronic pain were relatively older, more often female, received less education, were more often married, and suffered from more comorbid somatic conditions/health risks and comorbid recurrent physical symptoms. The same, but more skewed picture could be drawn for the participants with chronic pain in the context of arthritis.

Prevalence of Flourishing and Being at Risk for Psychopathology in People With and Without Chronic Pain

The prevalence of flourishing in the total sample was 34.9% (Table 2). Whereas 34.0% of the participants with chronic pain and 38.1% of the participants with chronic pain and arthritis was flourishing, the prevalence of flourishing was 35.2% for the participants without chronic pain. A chi-square test showed that the prevalence rates of flourishing did not differ significantly between the subsamples with and without chronic pain [$\chi^2(2, n = 1498) = 0.728, p = 0.695$, also the χ^2 -tests for a direct comparison of either of both subsamples with chronic pain versus the subsample without chronic pain

separately, $p > 0.05$]. The mean prevalence of being at risk for psychopathology was 22.8% in the total sample, 28.6% for those with chronic pain, 33.6% for those with chronic pain and arthritis and 16.8% for the participants without chronic pain.

TABLE 1 | Descriptive characteristics of the total sample and subsamples with chronic pain, chronic pain in the context of arthritis, or without chronic pain (Study 1).

	Chronic pain ($n = 597$) %	Arthritis ($n = 113$) %	No chronic pain ($n = 788$) %	Total sample ($n = 1498$) %
Sociodemographic				
Age (mean, sd)	*49.8 (17.1)	*61.3 (12.0)	44.9 (17.8)	48.1 (17.7)
Gender				
Female	*48.2	*77.0	45.2	49.8
Education level				
Low	*47.4	*58.0	37.6	43.0
Intermediate	41.8	33.9	48.8	44.9
High	10.8	8.1	13.6	12.1
Marital status				
Married	*57.8	*62.8	48.9	53.5
Health				
Comorbid somatic conditions ^a				
0	*49.1	*26.5	67.9	57.3
1	33.2	33.6	23.5	28.1
2	10.9	21.2	6.9	9.5
3 or more	6.8	18.7	1.7	5.1
Comorbid recurrent physical symptoms ^b				
0	*30.0	*10.6	44.4	36.1
1	28.0	20.4	26.8	26.8
2	17.4	20.4	17.9	17.9
3	10.2	18.6	6.6	8.9
4 or more	14.4	30.0	4.3	10.3

*Significant difference in comparison with subsample without chronic pain ($p < 0.05$). ^aMost prevalent comorbid somatic conditions/health risks in total sample were high blood pressure, high cholesterol, diabetes, and COPD/asthma.

^bMost prevalent comorbid recurrent physical symptoms in the total sample were fatigue, flu-related complaints, headache, and sleeping problems.

TABLE 2 | Prevalence of flourishing and being at risk for psychopathology in the total sample and subsamples with and without chronic pain (Study 1).

	Chronic pain ($n = 597$) %	Arthritis ($n = 113$) %	No chronic pain ($n = 788$) %	Total sample ($n = 1498$) %
Mental health				
Flourishing	34.0	38.0	35.2	34.9
Not flourishing	66.0	62.0	64.8	65.1
At risk for psychopathology				
At risk	28.6	33.6	16.8	22.8
Not at risk	71.4	66.4	83.2	77.2

These prevalence rates differed significantly between the three subsamples [$\chi^2(2, n = 1498) = 36.084, p < 0.001$]. *Post hoc* tests with a direct comparison of the groups showed that both the subsample with chronic pain [$\chi^2(1, n = 1358) = 29.364, p < 0.001$] and the subsample with chronic pain and arthritis [$\chi^2(1, n = 889) = 17.613, p < 0.001$] were significantly more at risk for psychopathology than the subsample without chronic pain. **Figure 1** displays the prevalence of *complete mental health* (i.e., not at risk for psychopathology and flourishing; Keyes, 2002) as well as the prevalence of being at risk for psychopathology for the different subsamples with and without chronic pain. The mean prevalence of complete mental health in the total sample and subsamples was slightly lower compared to the prevalence of flourishing (that could co-occur with being at risk for psychopathology) and ranged from 27.8% (subsample with chronic pain) to 31.9% (subsample without chronic pain). Also the prevalence of *complete mental health* did not significantly differ between the subsamples with and without chronic pain [$\chi^2(2, n = 1498) = 2.678, p = 0.262$, also the χ^2 -tests for a direct comparison of either of both subsamples with chronic pain versus the subsample without chronic pain separately, $p > 0.05$].¹

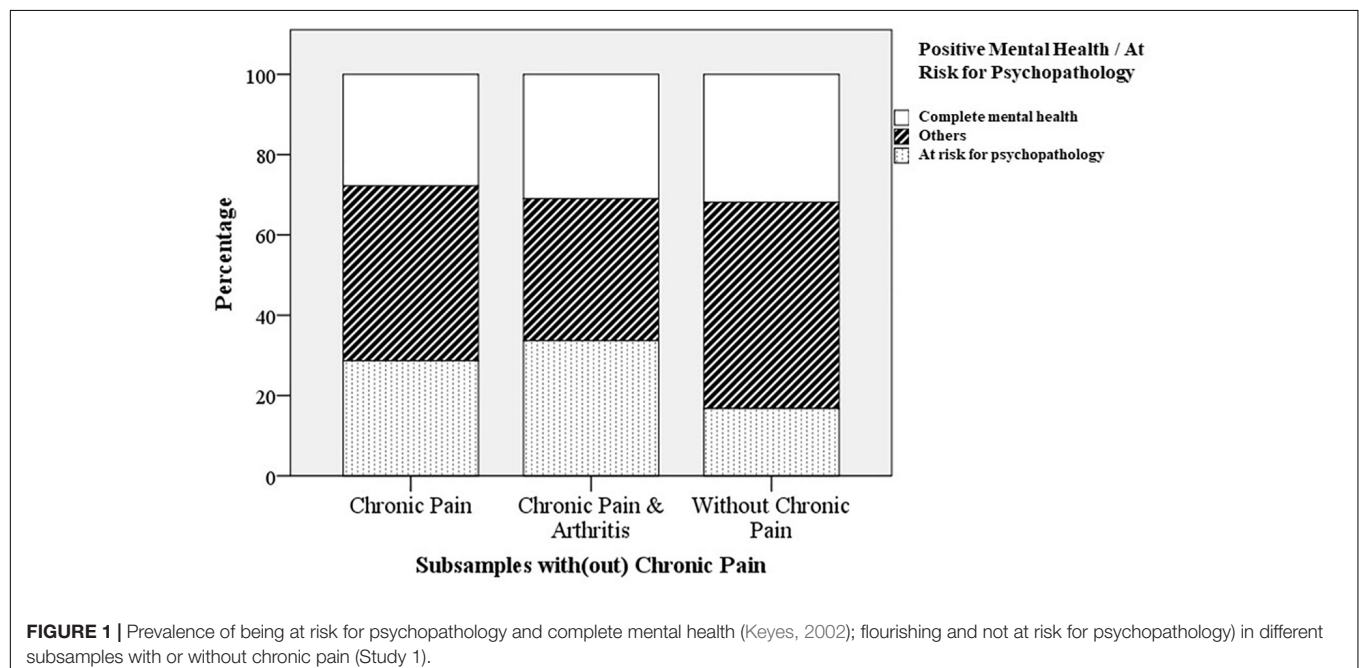
¹ Both chronic pain samples differed from the sample without chronic pain on each of the sociodemographic and health-related assessed. We thus performed *post hoc* multivariate logistic regression analyses to examine the association between chronic pain and both mental health outcomes while controlling for the included sociodemographic and health-related variables. The results were similar to the reported findings. The subsample with chronic pain, as well as the subsample with chronic pain and arthritis, were as likely to flourish as the sample without chronic pain [$B = 0.09$, S.E. (B) = 0.12, $p = 0.42$ and $B = 0.18$, S.E. (B) = 0.12, $p = 0.13$ respectively]. Simultaneously, the subsample with chronic pain as well as the subsample with chronic pain and arthritis were more likely to be at risk for psychopathology than the sample without chronic pain [$B = 0.71$, S.E. (B) = 0.14, $p < 0.01$ and $B = 0.44$, S.E. (B) = 0.14, $p < 0.01$ respectively].

STUDY 2

Materials and Methods

Participants and Procedure

This study draws on baseline data from a Randomized Controlled Trial on the effectiveness of web-based Acceptance and Commitment Therapy (ACT) for chronic pain (see Trompetter et al., 2015b, for a detailed description of study design and outcomes). The study protocol was approved by the Dutch Medical-Ethical Review Board (METC, trial number NL38622.044.11), which operates under the Dutch Central Committee for Research involving human participants (CCMO). The study has been registered in the Dutch Trial Register (Nederlands Trialregister), the primary trial register for clinical trials in Netherlands (trial number NTR3659). Participants were 238 people with chronic pain recruited in 2012 via national newspaper advertisements and online patient platforms from the Dutch general population. Diagnoses were heterogeneous and primarily included people without a chronic pain diagnosis, fibromyalgia, back pain, rheumatic diseases, and neuropathic pain. Study inclusion criteria were being 18 years or older, having a momentary pain intensity Numeric Rating Scale (11-point NRS) score > 4 , and having pain for at least 3 days per week for at least 6 months. Exclusion criteria were severe psychological distress as assessed with the Hospital Anxiety and Depression Scale (HADS; score > 24) (Zigmond and Snaith, 1983), extremely low levels of psychological inflexibility as assessed with the Psychological Inflexibility in Pain scale (PIPS; score < 24) (Wicksell et al., 2010a), participation in another Cognitive Behavioral Treatment, having no internet or e-mail address, reading problems in Dutch language, and/or an unwillingness or inability to invest time. Participants filled in the baseline data via an online



questionnaire, either during (e.g., HADS) or after checks (e.g., MHC-SF) of in- and exclusion criteria and always prior to randomization procedures.

Measures

Sociodemographic variables

Sociodemographic variables were *age*, *gender*, *educational level* (low, medium, high) and *marital status* (married/living together or not).

Physical variables

Physical variables were pain intensity, pain duration and pain disability. *Pain intensity* was measured with a 11-point Numeric Rating Scale (NRS), ranging from 'no pain' (0) to 'pain as bad as you can imagine' (10) (Dworkin et al., 2005). *Pain duration* was categorized into a dummy variable (less/more than 5 years). *Pain disability* was measured with the Pain Disability Index (PDI) (Pollard, 1984), that consists of seven items and assesses the degree to which chronic pain disables a person from performing daily activities such as work (total score range 7–70) ($\alpha = 0.81$).

Positive mental health

Positive mental health and flourishing were measured and categorized in the same way as in Study 1. Internal consistency of the MHC-SF was high in the current study ($\alpha = 0.89$).

Depressive symptoms

Depressive symptoms were measured with the depressive symptom subscale of the HADS (Zigmond and Snaith, 1983). The scale consists of seven items, and measures the presence and severity of depressive symptoms (total score range 0–21) ($\alpha = 0.79$). Individuals with a HADS score ≥ 8 are at risk for depression.

(Other) Psychological variables

Psychological variables were pain catastrophizing, psychological inflexibility, mindfulness and engaged living. *Pain catastrophizing* was measured with the Pain Catastrophizing Scale (PCS), a 13-item questionnaire (Sullivan et al., 1995). The scale measures levels of pain rumination, magnification, and helplessness (total score range 0–52) ($\alpha = 0.91$). *Psychological inflexibility* was measured with the PIPS that consists of 12 items (Wicksell et al., 2010a). The scale measures experiential avoidance of pain and cognitive fusion with pain-related thoughts and feelings (total scale range 12–84) ($\alpha = 0.87$). *Mindfulness* was measured with the Five Facet Mindfulness Questionnaire—Short Form (FFMQ-SF). The FFMQ-SF (Baer et al., 2006; Bohlmeijer et al., 2011) is a 24-item questionnaire that measures five facets of mindfulness: observing, describing, acting with awareness, non-judging and non-reactivity (total score range 24–120) ($\alpha = 0.82$). Finally, *engaged living* was measured with the Engaged Living Scale (ELS) (Trompetter et al., 2013) that consists of 16 items. The ELS measures the extent to which people know and act upon their personal values, and experience a sense of fulfillment in life as a consequence of doing so (total scale range 16–80) ($\alpha = 0.91$).

Statistical Analyses

The Statistical Program for Social Sciences (IBM SPSS) version 24.0 was used for all statistical analyses. We used an alpha level of 0.05 for all statistical tests. Frequency distributions were calculated and descriptive analyses were performed to summarize sample characteristics of the total chronic pain sample ($n = 238$). First, the prevalence of flourishing was determined as well as the prevalence of being at risk for depression. Chi-square tests and independent *t*-tests were used to compare functioning on all individual sociodemographic, physical and psychological correlates for flourishers (1) versus non-flourishers (0), and for being at risk for depression (1) versus not being at risk for depression (0). Then, we performed a multivariate stepwise logistic regression analysis with (not) flourishing (MHC-SF) as the dependent variable. Potential correlates were entered in blocks. After controlling for the level of depressive symptoms (HADS), sociodemographic (1), physical (2) and psychological variables (3) were entered. This hierarchical inclusion enabled us to assess the individual contribution to identify flourishers for each variable set. This multivariate stepwise logistic regression analysis was repeated with (not) being at risk for depression as the dependent variable. In a first step, we controlled for levels of positive mental health in this analysis. In both models we only included the correlates on which flourishers and non-flourishers, and/or those either at risk and not at risk for depression differed (marginally) significantly in previous chi-square tests and independent *t*-tests. All continuous correlates were normally distributed and did not include any outliers. Also inspection of residuals following the logistic regression analysis suggested no influential outliers in the data. Finally, outcomes of Pearson's correlation coefficients also suggested that none of the included correlates showed multicollinearity with each other (all $r < 0.57$). We reported beta's, standard errors and odds ratio's [exp (B)] for each correlate, as well as Nagelkerke R^2 as a measure of the effect size for each block of variables and the final, full model. The reported coefficients for individual correlates can be interpreted as such that – holding the other correlates constant at their mean values (continuous) or lowest values (dichotomous) – a change of one unit in this correlate will change the odds of *y* (i.e., flourishing) by a factor of exp (B). We transpose this one-unit change to a change of 1 SD for significant, continuous correlates to improve interpretation of the results.

Results

Prevalence of Flourishing and Being at Risk for Depression

The typical study participant was a middle aged, higher educated female in a relationship, whom suffered on a daily basis from pain [mean pain intensity levels: 6.2 ($SD = 1.7$)] for over 5 years (Table 3). The prevalence of flourishing was 22.8%, while the prevalence of being at risk for depression was 35.7%.

Correlates of Flourishing and Being at Risk for Depression

Chi-square tests and independent *t*-tests comparing flourishers and non-flourishers with each other on each individual

TABLE 3 | Descriptive characteristics of the sample (Study 2).

	Total sample (<i>n</i> = 238) %/ <i>M</i> (<i>SD</i>)
Flourishing (%)	22.8
At risk for depression (%)	35.7
Demographic characteristics	
Age	52.8 (12.4)
Female (%)	76.1
Education level (%)	
Low	20.2
Intermediate	35.7
High	44.1
Married/living together (%)	74.4
Physical characteristics	
Pain duration > 5 years (%)	63.0
Pain intensity (11-point NRS)	6.2 (1.7)
Pain disability (PDI)	36.2 (12.6)
Psychological characteristics	
Pain catastrophizing (PCS)	18.4 (19.8)
Psychological inflexibility (PIPS)	54.9 (11.5)
Mindfulness (FFMQ)	81.7 (10.7)
Engaged living (ELS)	50.9 (9.8)

sociodemographic, physical and psychological correlate (Table 4) revealed that both groups did not significantly differ on any of the sociodemographic variables (all p 's > 0.10). With regard to the physical correlates, flourishers ($M = 32.4$, $SD = 13.0$) scored significantly lower than non-flourishers on pain disability; $t(236) = 2.478$, $p = 0.014$. Both groups also scored significantly different on all psychological correlates. Flourishers were less

psychologically inflexible ($M = 50.4$, $SD = 11.1$) than non-flourishers ($M = 56.1$, $SD = 12.0$); $t(236) = 3.230$, $p = 0.001$, and experienced lower levels of pain catastrophizing (flourishers, $M = 16.1$, $SD = 10.0$; non-flourishers, $M = 19.1$, $SD = 9.6$); $t(236) = 1.966$, $p = 0.050$. In contrast, flourishers ($M = 86.9$, $SD = 10.4$) experienced higher levels of mindfulness than non-flourishers ($M = 80.2$, $SD = 10.4$); $t(236) = -4.173$, $p < 0.001$, and engaged living (flourishers, $M = 58.8$, $SD = 8.7$; non-flourishers, $M = 48.6$, $SD = 8.9$); $t(236) = -7.391$, $p < 0.001$. People (not) at risk for depression differed significantly on the same correlates, but with different patterns. With regard to the physical correlates, those at risk for depression scored significantly higher ($M = 39.9$, $SD = 10.6$) than those not at risk for depression ($M = 34.1$, $SD = 13.2$) on pain disability; $t(236) = -3.490$, $p < 0.001$. People at risk for depression were more psychologically inflexible ($M = 61.1$, $SD = 10.6$) than people not at risk for depression ($M = 51.4$, $SD = 10.6$); $t(236) = -6.818$, $p < 0.001$, and experienced higher levels of pain catastrophizing (at risk, $M = 23.3$, $SD = 9.2$; not at risk, $M = 15.7$, $SD = 9.0$); $t(236) = -6.146$, $p < 0.001$. In addition, those at risk ($M = 77.9$, $SD = 9.2$) experienced lower levels than those not at risk ($M = 83.8$, $SD = 11.0$) of mindfulness; $t(236) = 4.184$, $p < 0.001$, and engaged living (at risk, $M = 46.5$, $SD = 7.5$; not at risk, $M = 53.4$, $SD = 10.1$); $t(236) = 5.527$, $p < 0.001$. Both groups did not score differently on any of the sociodemographic variables. Marginally significant differences existed, however, for the distribution of men and women over both groups, with more men than women present in the group at risk for depression than the group not at risk for depression; $\chi^2(1, n = 238) = 3.199$, $p = 0.074$.

Results of the multivariate stepwise logistic regression analysis showed that the full, final model with socio-demographic,

TABLE 4 | Descriptive statistics for sociodemographic, physical and psychological variables, and comparison on these variables for flourishers and non-flourishers, and those either at risk or not at risk for depression, respectively (Study 2).

	Positive mental health		Depressive symptoms	
	Flourishing (<i>n</i> = 53) Mean (<i>SD</i>)	Not flourishing (<i>n</i> = 185) Mean (<i>SD</i>)	At risk (<i>n</i> = 85) Mean (<i>SD</i>)	Not at risk (<i>n</i> = 153) Mean (<i>SD</i>)
Sociodemographic variables				
Age	52.6 (11.3)	52.8 (12.7)	54.4 (12.0)	51.6 (12.5)
Female (%) ^a	77.4	75.7	69.4 ^X	79.7
High education level (%) ^b	41.5	44.9	41.2	45.8
Married/living together (%) ^c	83.0	71.9	80.0	71.2
Physical variables				
> 5 years pain duration (%) ^d	71.7	60.5	65.9	61.4
Pain intensity (11-point NRS)	6.5 (1.6)	6.1 (1.7)	6.5 (1.5)	6.0 (1.7)
Pain disability (PDI)	32.4 (13.0)*	37.2 (12.4)	39.9 (10.6)**	34.1 (15.7)
Psychological variables				
Pain catastrophizing (PCS)	16.1 (10.0)*	19.1 (9.6)	23.3 (9.2)**	15.7 (9.0)
Psychological inflexibility (PIPS)	50.4 (12.0)**	56.1 (11.1)	61.1 (10.6)**	51.4 (10.6)
Mindfulness (FFMQ)	86.9 (10.4)**	80.2 (10.4)	77.9 (9.2)**	83.8 (11.0)
Engaged living (ELS)	58.8 (8.7)**	48.6 (8.9)	46.5 (7.5)**	53.4 (10.1)

Chi-square and independent t-tests were performed to compare flourishers with non-flourishers (1), and to compare being at risk for depression with not being at risk for depression (2). *Significant at level $p < 0.05$; **Significant at level $p < 0.01$. ^XMarginally significant at level $p < 0.10$. ^aDummy coded, 1 = female; ^bdummy coded, 1 = high education level; ^cdummy coded, 1 = married/living together; ^ddummy coded, 1 ≥ 5 years pain duration.

physical and psychological correlates significantly explained 30% of the variance in flourishing (Omnibus $\chi^2 = 51.105$, $df = 7$, $p < 0.001$). Levels of depressive symptoms (step 1) explained 14% of the variance in flourishing (Table 5). We found that *only* the psychological correlates (step 4) were related to flourishing in the final model with an additional R^2 of 15%. Of the individual correlates, only *engaged living* ($B = 0.09$, OR = 1.10, 95% CI: 1.05–1.15) was significantly associated with flourishing, with higher levels of engaged living increasing the likelihood to flourish. The odds to flourish increase by a factor of 2.48 for each standard deviation increase in engaged living. The full, final model with socio-demographic, physical and psychological correlates significantly explained 37% of the variance in being at risk for depression (Omnibus $\chi^2 = 74.989$, $df = 7$, $p < 0.001$). Levels of positive mental health (step 1) explained 14% of the variance in being at risk for depression (Table 5). We found that *only* the psychological correlates (step 4) were related to flourishing in the final model with an additional R^2 of 18%. Of the individual correlates, *pain catastrophizing* ($B = 0.04$, OR = 1.05, 95% CI: 1.01 – 1.09), *psychological inflexibility* ($B = 0.05$, OR = 1.05, 95% CI: 1.02 – 1.09) and *engaged living* ($B = -0.05$, OR = 0.94, 95% CI: 0.91–0.99) were significantly associated with being at risk for depression. Whereas *higher* levels of pain catastrophizing and psychological inflexibility were associated with a higher likelihood to be at risk for depression, *lower* levels of engaged living increasing the likelihood to be at risk for depression. The positive coefficients for pain catastrophizing and psychological inflexibility correspond to an increase in odds of being at risk for depression of 1.54 and 1.81 for each standard deviation increase in both correlates, respectively. The size of the coefficient for engaged living means that one standard deviation increase in engaged living decreases the odds to be at risk for depression with a factor 0.60. An overall integration of the outcomes of logistic regression analyses and the descriptive patterns on the included correlates suggests that engaged living particularly sets apart flourishers from the other mental health groups, while pain catastrophizing

and psychological inflexibility particularly set apart those at risk for depression.

DISCUSSION

This study aimed to explore the prevalence and correlates of positive mental health for people with chronic pain and (being at risk for) psychopathology, and in comparison to people *without* chronic pain. The prevalence of flourishing was 34% in the general population sample with recurrent joint pain, 38% in the general population sample with arthritis, and 23% in our chronic pain sample seeking help. People with chronic pain in the general population either with or without arthritis were as likely to flourish as people *without* chronic pain. In contrast, the prevalence of being at risk for psychopathology for people with chronic pain was elevated compared to people without chronic pain. Finally, both flourishing and being at risk for depression were related foremost to psychological correlates. While engaged living was the most important correlate of flourishing, pain catastrophizing, psychological inflexibility, and engaged living were the most important correlates of being at risk for depression. Correlates of both mental health outcomes are thus partly overlapping, but simultaneously unique correlates emerged and all correlates had differential patterns of associations with both mental health outcomes. These findings support that positive and negative health- and well-being outcomes are related, but different constructs that function on two different continua (Huppert and Whittington, 2003; Schleicher et al., 2005; Huber et al., 2008; Westerhof and Keyes, 2009). Our findings on the prevalence and elevated chances of being at risk for psychopathology are in line with our hypotheses and existing research (e.g., Miller and Cano, 2009; de Heer et al., 2018). de Heer et al. (2018) reported that moderate to very severe pain was associated with a twofold risk of mood and anxiety disorders in the Dutch general population. Similarly existing research suggested that – although a significant subgroup of people with chronic pain in the general population *is*

TABLE 5 | Logistic regression coefficients associated with flourishing and being at risk for depression (Study 2).

		Flourishing		Being at risk for depression	
		B (S.E.)	OR (95% CI)	B (S.E.)	OR (95% CI)
Step 1	Depressive symptoms (HADS)/positive mental health (MHC-SF)	−0.09 (0.08)	0.92 (0.79; 1.06)	−0.19 (0.24)	0.82 (0.52; 1.31)
			$R^2 = 0.14$		$R^2 = 0.14$
Step 2	Gender ^a	−0.04 (0.44)	0.96 (0.40; 2.30)	0.64 (0.39)	1.90 (0.89; 4.09)
			$R^2 = 0.14$		$R^2 = 0.14$
Step 3	Pain disability (PDI)	−0.01 (0.02)	0.99 (0.96; 1.02)	0.02 (0.02)	1.02 (0.99; 1.05)
			$R^2 = 0.15$		$R^2 = 0.19$
Step 4	Pain catastrophizing (PCS)	0.01 (0.02)	1.01 (0.97; 1.06)	0.04* (0.02)	1.05 (1.01; 1.09)
	Psychological inflexibility (PIPS)	−0.01 (0.02)	0.96 (0.96; 1.04)	0.05* (0.02)	1.05 (1.02; 1.09)
	Mindfulness (FFMQ)	0.03 (0.02)	1.03 (0.99; 1.07)	−0.03 (0.02)	0.97 (0.94; 1.02)
	Engaged living (ELS)	0.09* (0.02)	1.10 (1.05; 1.15)	−0.05* (0.02)	0.94 (0.91; 0.99)
			$R^2 = 0.30$		$R^2 = 0.37$

Results are for the final model (step 4). Explained variance is Nagelkerke R^2 . *Significant at level $p < 0.05$. ^aDummy coded, 1 = female.

able to flourish – on average people with chronic pain are less likely flourish than people without chronic pain (Keyes, 2005a; Gilmour, 2015). We thus revealed different findings. As the sample of Gilmour (2015) resembled our broad, generic population sample, Keyes (2005a) actually included a specific chronic pain sample very similar to our subsample from the general population with arthritis. It is thus unlikely that we can attribute the differential findings between our and their studies to differences chronic pain sampling procedures. As proposed by Schotanus-Dijkstra et al. (2016), cultural factors like socio-economic advantages, and higher levels of individualism and social equality positively influence national levels of emotional well-being (Diener et al., 1995). These factors might aid people in the Dutch general population in contrast to other cultures to experience high positive mental health in the presence of chronic pain and arthritis. Individual variation in levels of positive mental health and being at risk for depression was explained primarily by psychological factors. Research and practice has long recognized the pivotal role of psychological factors in pain-related health and well-being (McCracken and Turk, 2002; Gatchel et al., 2007; Jensen and Turk, 2014). We know, for example, that pain acceptance is more important in explaining pain-related health and well-being outcomes than pain intensity levels (Viane et al., 2003; McCracken and Eccleston, 2005; Vowles et al., 2007; McCracken and Gutiérrez-Martínez, 2011). Also, behavioral interventions for chronic pain are only effective when they additionally targets psychological factors (Williams et al., 2013). Our study reveals several additional findings. Higher levels of engaged living was the most important correlate of flourishing. These findings resonate with the inherent focus of the psychological flexibility model (PF) on positive mental health. Particularly, its unique, person-oriented focus on long-term engagement with personally valued activities in the presence of persistent pain (McCracken and Vowles, 2014; Goubert and Trompetter, 2017). On the other hand, the most important correlates of being at risk for depression were pain catastrophizing, psychological inflexibility and engaged living. An overall integration of the outcomes suggests that particularly pain catastrophizing and psychological inflexibility set apart those at risk for depression. Both factors are related indeed to each other and to negative outcomes in and outside the context of chronic pain (Vowles et al., 2007; Aldao et al., 2010; Wicksell et al., 2010b; Trompetter et al., 2015a). An overall integration of findings supports that core therapeutic processes of the PF model – psychological inflexibility and engaged living – in combination provide pathways to decrease both depressive symptoms and simultaneously enhance positive mental health in chronic pain patients (McCracken and Morley, 2014; Goubert and Trompetter, 2017). Simultaneously, the FA model seems to particularly explain being at risk for depression and not flourishing. ACT interventions based on the PF model have been able to change both mental health outcomes successfully in people with mild depressive symptoms (Fledderus et al., 2012). While some people improved on both mental health outcomes, some improved on either one of both (Trompetter et al., 2017). On the contrary, the same ACT intervention reworked for people with chronic pain was able to enhance

depressive symptoms but not positive mental health (Trompetter et al., 2015b). Knowledge is fully lacking at present to explain these differential effects within and between diagnostic groups. Furthermore, positive and negative correlates and outcomes of health and well-being in the context of chronic pain are related, but different constructs that function on two different continua. It is likely that other, unique protective psychological factors exist that function as important correlates of flourishing in the context of chronic pain. For example, positive affect and optimism (Zautra et al., 2001; Finan et al., 2013; Hanssen et al., 2014; Finan and Garland, 2015). Finally, a lower percentage of people with chronic pain flourished in our second sample (23%). This is plausible as the sample was explicitly targeted for their need to help reduce the interference of chronic pain with daily life activities. Our findings do suggest that people with chronic pain are open to receive or seek help – at least via an easily accessible Internet-based intervention (Trompetter et al., 2015b) – because they experience low levels of well-being beyond high levels of negative pain-related outcomes. Improvements in well-being have proven to be at least as important as relief from symptoms of psychopathology for patient groups with major depression (Zimmerman et al., 2006; Demyttenaere et al., 2015). It is interesting to further explore chronic pain patients viewpoints on desired treatment outcomes in the future. This study has several limitations. It concerns a *post hoc*, exploratory study. This limited Study 1 primarily in our operationalization of chronic pain. We do not know if our sample with chronic (joint) pain adhered to important classifications for chronic pain like a minimal duration of 3 or 6 months. Both pain duration and intensity were, however, not associated with flourishing in Study 2, and all findings in Study 1 were similar for the sample with rheumatoid arthritis as well as the more generic pain sample. A limitation of Study 2 is that we could not include several protective psychological factors that may function as important correlates of (positive) mental health in the context of chronic pain (e.g., positive affect, optimism). A further limitation is the fact that both studies were cross-sectional. Thus, we cannot draw conclusions regarding causality between levels of positive mental health, depressive symptoms, chronic pain and included psychological correlates. Finally, both included chronic pain samples were very heterogeneous in nature. We do not know if and to what extent our findings generalize to specific diagnostic groups with chronic pain or people with clinical levels of depression. Previous studies for example showed that fibromyalgia patients, but not patients with rheumatoid arthritis, function worse on aspects of positive mental health compared to healthy subgroups (Schleicher et al., 2005; Finan et al., 2009). This study suggests that negative and positive mental health outcomes and their correlates in the context of chronic pain are related but simultaneously of a different, unique nature. Also, a relatively large group of people with chronic pain is able to maintain high levels of positive mental health. We hope that our outcomes boost further research on protective beyond risk pain-related factors and positive beyond negative pain-related outcomes, particularly the study of unique, protective mechanisms involved in the ability to maintain or achieve optimal levels of positive mental health in the presence of chronic pain and other disabilities.

DATA AVAILABILITY STATEMENT

The data for Study 1 uses LISS (Longitudinal Internet Studies for the Social sciences) panel data, administered by CentERdata (Tilburg University, Netherlands). LISS panel data is freely available for scientific purposes under specific conditions (www.lissdata.nl). Information on variables used from LISS and the dataset generated for Study 2 are available on request to the corresponding author.

ETHICS STATEMENT

All patients/participants provided their written informed consent. The original study that provided data for Study 2 was reviewed and approved by the Dutch Medical-Ethical Review Board (METC, trial number NL3622.044.11).

AUTHOR CONTRIBUTIONS

HT executed the original study that provided data for in Study 2, designed the study, performed data analysis and interpretation of the data, and drafted the manuscript. FM was involved in decisions on data analysis, data interpretation, and manuscript

writing. GW was responsible for conception of the original study that provided data for Study 1, and contributed to decisions on data analysis, data interpretation, and manuscript writing. All authors contributed to critical revisions of the manuscript and approved the final version of the manuscript.

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Using a Mindfulness-Based Intervention to Promote Subjective Well-Being, Trait Emotional Intelligence, Mental Health, and Resilience in Women With Fibromyalgia

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The objective of the present study was to experimentally assess the effects of a mindfulness-based intervention (MBI) for the improvement of subjective well-being, trait emotional intelligence (TEI), mental health, and resilience in a sample of women with fibromyalgia (FM). The sample consisted of 104 women, between 29 and 77 years old ($M = 47.59$; $SD = 5.93$). The measures used were as follows: Satisfaction with Life Scale (SWLS), Positive and Negative Affect Scale (PANAS), Trait Emotional Intelligence Questionnaire Short Form (TEIQue-SF), Mental Health Questionnaire (MH-5), and Resilience Scale (ER-14). A quasi-experimental design of repeated measures with a control group (CG) was used: before and after the application of the treatment and a follow-up assessment 6 months after the completion of the intervention. In order to assess the effect of the program, the participants were randomly assigned to the experimental and control conditions. In the pretest evaluation, a multivariate analysis of variance (MANOVA) and analysis of variance (ANOVA) were carried out. In the post-test evaluation, a multivariate analysis of covariance (MANCOVA) of the study variables as a whole was performed. Then, descriptive analyses and analysis of covariance (ANCOVA) of the post-test scores (covariate pretest score) were performed. In the follow-up evaluation, a MANCOVA of the study variables as a whole was performed. Then, descriptive and ANCOVA analyses of the follow-up scores (covariate pretest score) were performed. In addition, the effect size was calculated using partial eta-squared (η^2). The post-test results confirmed statistically significant differences in satisfaction with life (SWL), positive affect (PA), mental health, and resilience. The follow-up results showed statistically significant differences in SWL, PA, TEI, mental health, and resilience. The study provides an effective intervention tool that has been validated experimentally. The general results allow the emphasis of the importance of the implementation of MBIs framed in non-pharmacological treatments in FM.

Keywords: fibromyalgia, mindfulness, subjective well-being, chronic pain, random assignment

INTRODUCTION

According to Häuser and Fitzcharles (2018), fibromyalgia (FM) is characterized by generalized chronic pain, unrefreshing sleep, physical exhaustion, and cognitive alterations. A more comprehensive definition refers to FM as a multifaceted disease closely related to psychological discomfort and physical pain, associated with disorders such as anxiety and depression, pathologies that, when associated with this disease, cause a worsening and irremediable chronification (Revuelta-Evrard et al., 2010). Currently, there is no consensus regarding its definition. In this way, FM has been defined as a disease characterized by chronic primary pain that is associated with emotional distress or significant functional disability (interference with activities of daily living and participation in social roles) and that cannot be explained better with another chronic pain condition (International Association for the Study of Pain [IASP], 2016). On the other hand, it was included in the *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)* by the American Psychiatric Association [APA] (2013) as a somatic symptom disorder due to its relationship with physiopathological aspects of central sensitization, functional brain connectivity, and other changes of the central nervous system. Some authors define FM as a masked depression (Alciati et al., 2012); others define it as a persistent somatoform pain disorder (Häuser and Henningsen, 2014).

The FM diagnostic criteria were established in 1990 by the American College of Rheumatology (ACR) (Wolfe et al., 1990) and reviewed by Wolfe et al. (2010). FM is described as the existence of generalized pain of more than 3 months duration, absence of another causal pathology, and comorbidity with other syndromes and symptoms, such as chronic fatigue, unrefreshing sleep, cognitive deficit, and numerous somatic and emotional symptoms, such as anxiety and depression. Later, Wolfe et al. (2011) include a scale of FM symptoms. This scale adds the generalized pain index (WPI) and the severity of symptoms scale (SSS) to facilitate the diagnosis. Wolfe et al. (2016) published a new version of the severity scale of FM in which the doctor's criteria are combined with the patients' self-reports. These new criteria refined and increased the usefulness of symptom-based FM diagnosis by excluding patients with regional pain. However, they underscore the social construction of diagnosis based on symptoms and the inherent limitations in reliability and validity associated with FM criteria.

Regarding treatment, the review developed by Thieme et al. (2017), after examining the recommendations for the FM approach of some of the most influential organisms (Association of the Scientific Medical Societies in Germany, 2012; European League Against Rheumatism [EULAR], 2016), concludes that aerobic exercise, cognitive behavioral therapy, pharmacological treatment with amitriptyline, and multicomponent treatment are the most effective. In the review of recommendations for the management of FM carried out by EULAR (Macfarlane et al., 2017) through meta-analysis, relatively modest effect sizes are confirmed for most treatments. Furthermore, they add that the initial treatment must include patient education and non-pharmacological treatment, and in the case of non-effectiveness,

the rest of the additional therapies should be included. A recent review of EULAR about the guidelines for the management of FM (Arumugam and MacDermid, 2019) highlights clear support for pharmacological and non-pharmacological treatment approaches in FM, considering non-pharmacological therapy as first-line treatment. Finally, EULAR (Macfarlane et al., 2017) proposes research that clarifies the individual characteristics for the administration of certain interventions, their effect in combination, the adaptation of patients to therapies, and the organization of health systems to optimize their results.

According to the therapeutic guidelines marked by the aforementioned organisms (e.g., European League Against Rheumatism [EULAR], 2016) and to the work of different authors (Glombiewski et al., 2010; Parra-Delgado et al., 2012; Van Gordon et al., 2016), the most effective treatment strategies for FM should add and integrate non-pharmacological approaches such as mindfulness, relaxation therapy, and psycho-education. Other authors highlight the importance of investigating the effects of the therapeutic application of mindfulness for FM (Aman et al., 2018; Amutio et al., 2018; Prabhakar et al., 2019).

Considering its conceptualization, the word mindfulness is the English translation of the Pali term "sati" which implies "consciousness, attention, and remembrance" (Siegel et al., 2009). Likewise, Kabat-Zinn (1990) defines it as the ability to pay attention to the experience of the present moment with a mental attitude of receptivity and acceptance.

Despite its millennial origin, it was Kabat-Zinn (1990) who introduced it to the western world, for the treatment of psychosomatic disorders, stress, and chronic pain, being aware of the great benefits that its application could bring. From these postulates, mindfulness-based interventions (MBI) emerged, specifically the Mindfulness-Based Stress Reduction (MBSR) program designed by Kabat-Zinn (1990). In addition to MBSR, others have been added, such as the Mindfulness-Based Cognitive Therapy (MBCT) program (Segal et al., 2002), which arises from the integration of mindfulness and cognitive-behavioral therapy for depression, and others like MBEating (MBE) (Kristeller and Wolever, 2011). Many studies support the integration of mindfulness into health care as part of self-care and the management of different diseases (Greeson and Chin, 2019).

On the other hand, it has been observed that MBIs are effective in primary care, especially for patients with symptoms of stress, anxiety, or depression (Hervás et al., 2016). Authors such as Vázquez-Dextre (2016) expose clinical reasons for applying mindfulness in mental and physical health problems. Besides, the growing interest in MBI has been caused by its effectiveness in increasing the well-being of individuals by improving some physical and psychological aspects (Hervás et al., 2016). MBIs seem to enhance the use of positive reevaluation (e.g., Carmody and Baer, 2008). Also, some studies have shown that with MBI, in a population with depressive symptoms, there is a positive interrelation between positive affect (PA) [an affective component of subjective well-being (AWB)] and positive cognitions [a cognitive component of subjective well-being (CWB)] (Garland et al., 2015).

Various meta-analyses (Hilton et al., 2017; Zou et al., 2018) highlight the positive effects of mindfulness in major depression

disorder, associated with significant improvements in depression, quality of life related to physical health and mental health. Also, they recognize positive effects on health in general, in comparison to control groups (CGs). There are also works that advise the use of mindfulness in anxiety and stress disorders (Gallegos et al., 2013; De Frias and Whyne, 2015) considering these interventions as a viable protective factor to reduce the dangers of stress, reducing the reactivity to the stressor. Therefore, it could serve as a protective psychological process for health (Yat Ho Li and Bressington, 2019).

Similarly, research on the impact of the practice of mindfulness in chronic pain is increasing, and there are works with positive results in coping with it (McCracken and Vowles, 2014). Other works conclude that the practice of mindfulness improves postural awareness, causing a decrease in pain in patients with spinal pain and shoulder pain (Cramer et al., 2018) and that training in mindfulness helps relieve the suffering associated with chronic pain, improving its management and the neurobiological mechanisms involved (Brown et al., 2015).

Specifically in the field of study on FM, there is evidence that MBI is beneficial for treating certain symptoms, such as depression, anxiety, anger, and poor quality of life (Grossman et al., 2007; Sephton et al., 2007; Schmidt et al., 2011; Davis and Zautra, 2013; Amutio et al., 2015). Other authors such as Van Gordon et al. (2017) claim that in patients with FM through an 8-week MBI, they found benefits in pain perception, sleep quality, and psychological distress. Furthermore, these effects were maintained 6 months after the conclusion of the treatment. After applying an 8-week MBI in women with FM, Cash et al. (2015) conclude significant improvements in some of their associated symptoms, such as fatigue, stress, sleep, pain, salivary cortisol, and overall well-being. Also, these benefits were maintained in the follow-up evaluation.

In the same direction, Davis et al. (2015) after developing an MBI in patients with FM, with randomized study, revealed improvements in social functioning, PA, and effectiveness in coping with pain and stress. Besides, patients with depressive symptoms reported improvements in loneliness, family stress, and PA. Likewise, Schmidt et al. (2011), through a randomized clinical study in patients with FM, confirm that participants assigned to the MBSR did not report significant reductions in pain, but improvements in quality of life compared to the CG. However, these benefits were not maintained at follow-up (2 months), which suggests that MBSR does not produce stabilized improvements in the quality of life of patients with FM. Grossman et al. (2007), through a randomized clinical study in women with FM, obtained improvements in pain perception, quality of life, pain management, anxiety, depression, and somatic complaints. These results were maintained in the follow-up evaluation at 3 years. Quintana and Rincón-Fernández (2011) through an MBI with women with FM found significant improvements in quality of life, vitality, and mental health. Likewise, the participants reported improvements in the presence and intensity of pain, in coping strategies and perceptions in general health. Lakhan and Schofield (2013) conducted a systematic review of MBI in the treatment of somatic disorders (including FM) and conclude that these interventions

are effective in reducing pain, depression symptoms, and anxiety symptoms – improving the quality of life in these patients.

From other approaches, Amutio et al. (2015), through an MBI with a duration of 7 weeks in patients with FM, corroborate reductions in symptoms of anxiety, depression, and improvements in the ability of participants to regulate their anger. These results were maintained in a follow-up at 3 months. Amutio et al. (2018) showed that, compared to the control condition, MBI was effective in reducing insomnia and improving sleep quality in line with the studies of Kanen et al. (2015). It is important to note that several authors state that MBI produces more stabilized improvements in measures of pain and quality of life as a function of frequency, continuity of practice, and experience of meditation training (Quintana and Rincón-Fernández, 2011; Adler-Neal and Zeidan, 2017). They also point out that the daily average of mindfulness practice is a significant predictor of changes in all outcome variables (Van Gordon et al., 2017).

This work is projected with the idea of providing greater consistency to the application of non-pharmacological therapies in the treatment of FM, specifically the use of MBI. Thus, the purpose of the present study was to evaluate the effects of an MBI on subjective well-being, trait emotional intelligence (TEI), mental health, and resilience in women with FM. It was hypothesized that compared to the waitlist CG, women with FM that completed the mindfulness intervention would demonstrate significant improvements in the variables mentioned above, both in the post-test phase and in the follow-up phase (6 months).

MATERIALS AND METHODS

Design

A randomized experimental design was conducted with three repeated measures (pretest, post-test, and a 6-month follow-up). The participants were randomly assigned either to the experimental group (EG) or to the usual treatment of the CG. The usual treatment of the CG was focused on psychoeducation and included information on common symptoms in FM and advice on self-care. This treatment was performed by the psychologist of the FM association. All this was done in addition to the pharmacological treatment for pain indicated by specialists.

Participants

A total of 132 women from the Association of Relatives and Affected by Fibromyalgia of the province of Ciudad Real (Spain) voluntarily participated in the investigation. To participate in the study, participants had to meet three *inclusion criteria*: (a) be diagnosed with FM syndrome (e.g., via a letter from a doctor of pain consultant), (b) commit to the daily practice of mindfulness, and (c) not be currently receiving mindfulness training. Two *exclusion criteria* were also established: (a) be diagnosed with a mental disorder and (b) receive individual psychological therapy. An adjusted sample size of 102 participants was estimated using GPower 3.1.9.2 software (Faul et al., 2007) for the hypothesis contrast for independent samples ($p < 0.05$).

The 117 participants who met the proposed criteria were randomly assigned to the EG of MBI ($n = 59$) or the usual treatment of the CG ($n = 58$) using the Random Number Generator program (Deville, 2004). The therapy was completed by 104 of the 117 patients. The dropout rate in the EG was 16.95% if it is considered that the experimental mortality is the lack of attendance of at least 50% of the sessions of the program. Participant flow is displayed below (refer to **Figure 1**).

The sample consists of 104 women, aged 29–57 ($M = 47.59$; $SD = 5.93$). A non-probabilistic sampling was used, but subjects were assigned at random to the experimental ($n = 53$) and control ($n = 51$) condition. The age differences in the two conditions were not significant, $\chi^2 = 1.79$, $p > 0.05$.

Regarding the demographic characteristics; 87.5% were married, 10.5% divorced, and 2% were widows. Concerning the level of studies of the participants; 63.1% had

completed primary education, 23.2% secondary education, and 13.7% university studies; 15.2% of patients took antidepressants, 25.4% took anxiolytics, and 11.9% took both types of medication.

Measures

In this study, we used well-established measures with appropriate psychometric properties (refer to **Table 1**).

Satisfaction With Life Scale (SWLS) (Diener, 1984)

In this study, the version adapted to Spanish has been used (Vázquez et al., 2013). This scale is composed of five items, in which participants are expected to indicate the degree of agreement with each statement using a seven-point Likert scale (from 1 = *strongly disagree* to 7 = *strongly agree*). Diener (1984) states that this scale evaluates the cognitive CWB referring to the

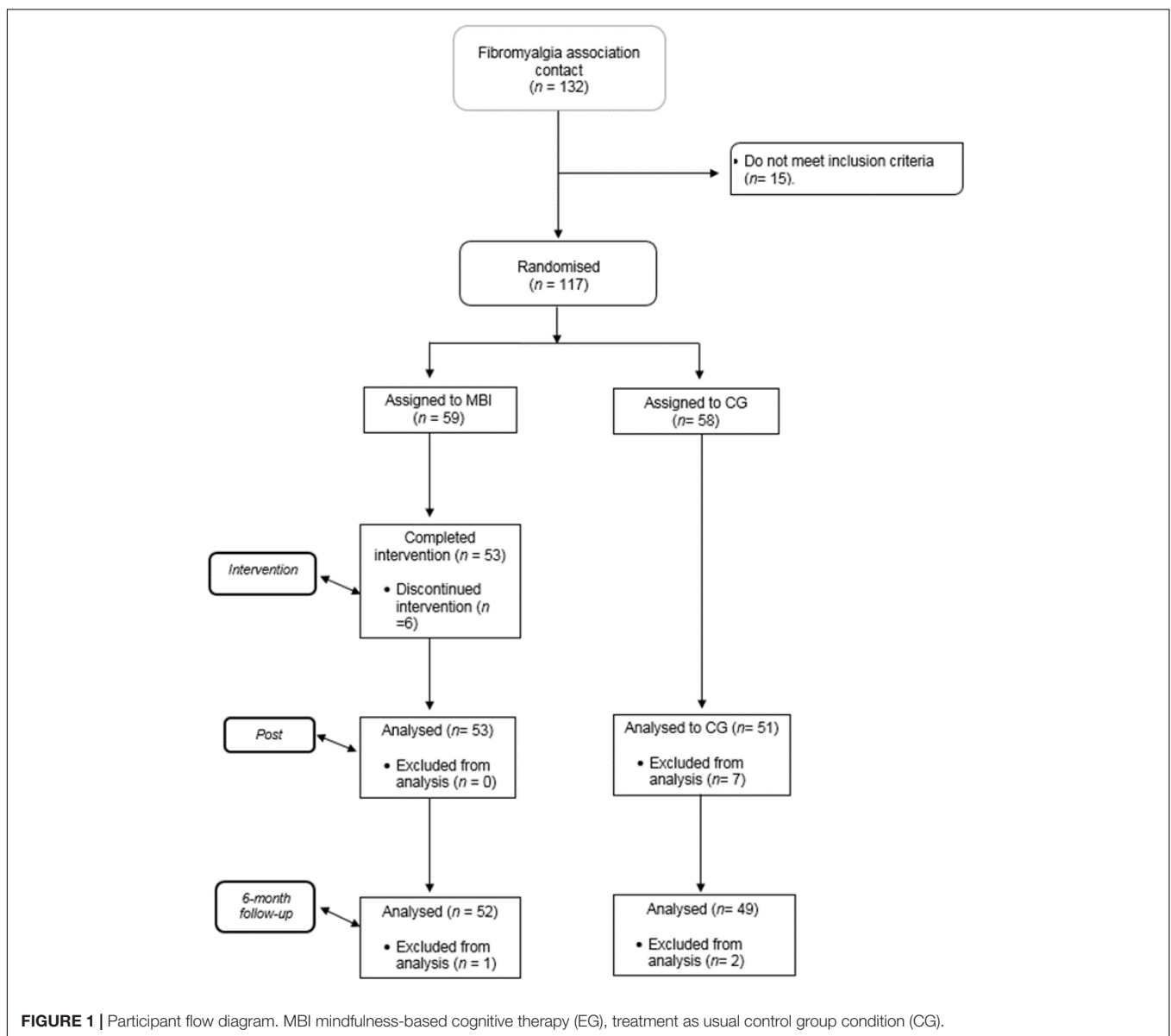


TABLE 1 | Reliability evidence.

	α	CR	AVE	Ω
SWLS	0.90	0.88	0.659	0.92
Positive affect	0.89	0.84	0.541	0.84
Negative affect	0.81	0.77	0.533	0.81
TEIQue-SF	0.88	0.92	0.617	0.91
MH-5	0.78	0.83	0.597	0.80
ER-14	0.77	0.79	0.502	0.78

α , Cronbach's alpha; CR, composite reliability; AVE, average variance extracted; Ω , McDonald's omega index.

result of the evaluation of the processing of the information that people make of their lives.

Positive and Negative Affection Scale (PANAS) (Watson et al., 1988)

A Spanish version (Sandín, 2003) was used. PANAS is a self-reported adjective checklist designed for the assessment of 20 different feelings and emotions. It contains two subscales each with 10 items, representing two constructs: PA and negative affect (NA). Participants used a five-point scale (from 1 = *very slightly or not at all* to 5 = *extremely*). This scale evaluates the AWB. According to Diener (1984), the AWB implies an individual hedonistic balance, that is, the frequency with which people experience emotions of a positive and negative nature.

Trait Emotional Intelligence Questionnaire Short Form (TEIQue-SF) (Petrides, 2009)

A Spanish version (Pérez, 2003) was used. The TEIQue-SF is a self-report inventory designed to measure global TEI with 30 items using seven-point Likert scale response options (from 1 = *completely disagree* to 7 = *completely agree*). This measure provides a total score that is obtained by adding the scores from the 30 items.

Mental Health Scale (MH5) (Ware and Sherbourne, 1992; Adapted to Spanish by Alonso et al., 1995)

The Mental Health-5 (MH-5) is one of the subscales of the SF-36 health questionnaire by Ware and Sherbourne (1992). The MH-5 is composed of five items on emotional well-being. A high score on this scale is associated with better mental health. The questions included are similar to: “*During the past four weeks, how long were you very nervous?*” The answers are encoded using a six-point Likert scale (from 1 = *always* to 6 = *never*).

Resilience Scale (ER-14) (Wagnild, 2009; Adapted to Spanish by Sánchez-Teruel and Robles-Bello, 2015)

It consists of 14 items. The answers are coded through a seven-point Likert scale (from 1 = *strongly disagree* to 7 = *strongly agree*). It measures the degree of individual resilience, considered a positive psychological characteristic that allows the individual to adapt to adverse situations. Also, this scale presents negative and significant correlations with depression and anxiety (e.g., Nishi et al., 2010).

Procedure

The study followed a quasi-experimental design of repeated measures (pretest and post-test) including a CG, where the following variables were assessed: satisfaction with life (SWL), PA, and NA, mental health (MH), TEI, and resilience (RS).

Ethical Considerations

All participants gave their informed consent, and the study was approved by the board of the association involved. Confidentiality and anonymity were guaranteed in order to comply with the Law on Protection of Personal Data of the Ethics Committee for Research on Human Beings (CEISH). The international guidelines for studies with human subjects described in the Nuremberg Code and the Declaration of Helsinki were applied. After completing all phases of the evaluation, the MBI was carried out with the CG participants.

Training Program Description

An MBI was carried out based on some previous works (e.g., Parra-Delgado et al., 2012; Amutio et al., 2015). This MBI is structured in two parts, in line with some of the principles of MBSR (Kabat-Zinn, 1990; Schmidt et al., 2011): (a) formal practice carried out in groups during face-to-face sessions and (b) the informal practice carried out individually at home through audio-guide. Therefore, we can say that the present intervention is MBSR-adapted.

The formal practice consisted of 20 group sessions. These sessions were held in 20 weeks (that is, one per week), with a duration of 1 h. The training program included three content blocks – (1) mindfulness meditation techniques according to some previous works (Kabat-Zinn, 1990; Schmidt et al., 2011), (2) exhibition and debate on the exercises used in the formal practical session, and (3) Vipassana meditation (Hart, 1994) – that promote values such as impermanence, compassion, acceptance, forgiveness, and detachment with reference to the work done by Amutio et al. (2015).

Similarly, the present MBI attempts to conform to the criteria of Van Gordon et al. (2017) on Interventions Based on Second Generation Comprehensive Care (SG-MBIs). These interventions emphasize and recognize the spiritual aspect of mindfulness. In addition, Van Gordon et al. (2015) highlight the possible positive effects of SG-MBIs in the treatment of FM.

The main objective of this MBI is to improve the relationship of participants with pain by taking perspective on the intrusive, ruminant, persistent, and egodystonic thoughts that usually accompany pain, focusing attention toward more adaptive emotional states in order to accept the experience of pain.

Each weekly session presents the following structure: (1) reflection on the practical exercises carried out during the previous week for 10 min, (2) body scan (10 min), (3) presentation of the exercises and explanation of their meaning for each session (20 min), and (4) meditative practice for 20 min.

The informal practice was carried out at home with the help of an audio-guide. Also, the participants were encouraged to

practice body scan for 5 min and attention focused on breathing (15 min). The informal practice was carried out during active treatment, as well as during the follow-up phase.

The program was designed and developed by an instructor with extensive experience in the practice and teaching of mindfulness techniques in the field of health. Program sessions are detailed in **Table 2**.

Statistical Analysis

Initially, the normality of the study variables was tested with a Kolmogorov–Smirnov test. All the variables were adjusted to the assumption of normality. The analyses were conducted with a confidence interval of 95%. After, reliability coefficient Cronbach's alpha (α), composite reliability (CR), average variance extracted (AVE), and McDonald's omega coefficient (Ω) were calculated to obtain reliability evidence. First, a multivariate analysis of variance (MANOVA) was performed with total pretest scores from the variables included in the study in order to confirm the possible pretest difference in the variables, as a whole, between EG participants and CG participants. Second, in order to determine the program's effect, descriptive analysis (mean and standard deviations) and repeated-measures analysis of variance (ANOVA) were carried out with each one of the scores obtained for the instruments used during the pretest phase. Third, having confirmed the homogeneity of the two groups *a priori*, and in order to determine whether the change was significantly different in the EG versus CG participants, a multivariate analysis of covariance (MANCOVA) was performed on the study's variables as a whole.

TABLE 2 | Intervention program sessions.

Session	Objectives	Contents
1–3	Know mindfulness techniques	<ul style="list-style-type: none"> Definition and analysis of the term by different authors Mindfulness technique, status, and via
4–9	Know/develop the types of practice; formal and informal	<ul style="list-style-type: none"> Explanation of the types of formal and informal practice with attitude (curiosity, openness, and no judgment) Commitment to practice Postural adjustments: lying, sitting, standing, and walking
10–14	Practice calm/relaxation	<ul style="list-style-type: none"> Awareness about the body (weight/calmness)
15–18	Identify and observe breathing	<ul style="list-style-type: none"> Start, breathing characteristics, frequency, changing character, and parts (inspiration, expiration, and apneas) Awareness of weight, associated with calm and breathing
19–20	Cultivate full attention	<ul style="list-style-type: none"> Attention on breathing Focused attention Open monitoring Vipassana meditation (compassion, acceptance, forgiveness, and detachment)

Besides that, descriptive analyses and analyses of covariance were performed on post-test scores [post-test analysis of covariances (ANCOVAs) covarying for pretest scores]. In the follow-up evaluation, a MANCOVA of the study variables as a whole was performed. Then, descriptive and covariance analyses of the follow-up scores were conducted (follow-up ANCOVAs, covarying for pretest scores). The effect size (μ^2) of the differences was calculated using partial eta-squared (Tabachnick and Fidell, 2007). The effect size was analyzed based on four ranges: 0–0.009, *negligible*; 0.010–0.089, *low-effect size*; 0.090–0.249, *medium-effect size*; and >0.250, *big-effect size*.

RESULTS

Results obtained in the pretest or basal evaluations are first presented, followed by the results for evaluating the impact of the MBI in the variables studied (post-test). Finally, the results obtained in the follow-up are collected.

The pretest MANOVA results did not reveal statistically significant differences between the groups prior to the intervention, Wilks' Lambda, $\Lambda = 0.739$; $F(5,99) = 0.628$; $p = 0.249$, with a small effect size ($\mu^2 = 0.030$, $r = 0.11$). The results of the ANOVA in the pretest phase (refer to **Table 3**) showed that before starting the program, there were no statistically significant differences in any of the variables dependent on the study (refer to **Figure 2**).

Post-test Evaluation

Results from the pretest–post-test MANCOVA revealed significant differences between the two conditions, Wilks' lambda, $\Lambda = 1.526$; $F(5,99) = 3.214$; $p = 0.007$, with an average effect size ($\mu^2 = 0.199$, $r = 0.33$).

Effects on Subjective Well-Being

The results showed significant improvements in life satisfaction in favor of EG (refer to **Table 3**). The size of the effect (eta-squared) reported low-effect size ($\mu^2 = 0.015$).

The results of the analyses showed a significant increase in PA scores in favor of the EG. The size of the effect was a low-effect size for PA ($\mu^2 = 0.032$). However, a significant decrease in NA is not confirmed. It is necessary to indicate that there is no significant relationship between PA and NA ($r = -0.125$, $p = 0.144$).

Effects on Trait Emotional Intelligence

Regarding the variable TEI, the results did not confirm significant improvements in favor of the EG.

Effects on Mental Health

Regarding the variable mental health, the results analyzed showed significant improvements in favor of the EG. The size of the effect was low-effect size ($\mu^2 = 0.021$).

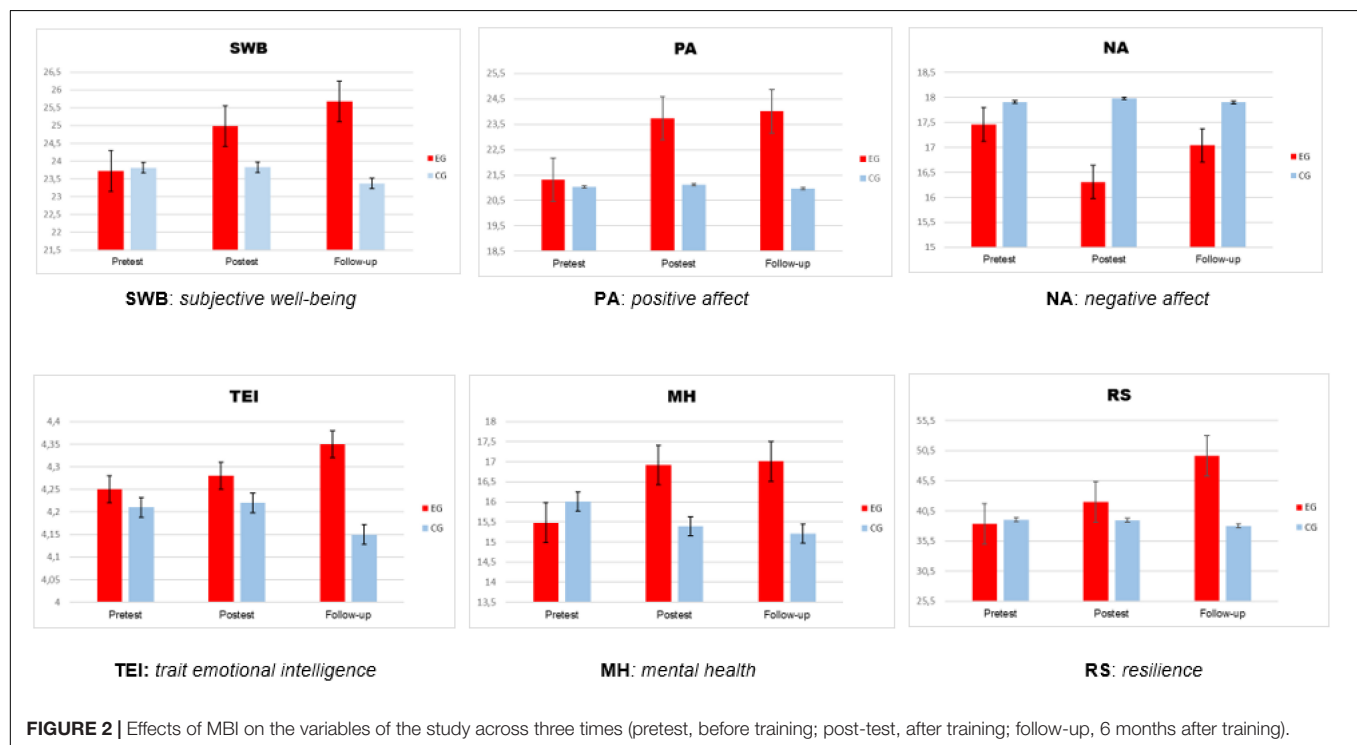
Effects on Resilience

Finally, regarding the resilience variable, the results analyzed showed significant improvements in favor

TABLE 3 | Averages and standard deviations in the variables under study (subjective well-being, trait emotional intelligence, mental health, and resilience) in the experimental and control groups.

	Pretest					Post-test					Follow-up				
	EG M (SD)	CG M (SD)	F	p	μ^2	EG M (SD)	CG M (SD)	F	p	μ^2	EG M (SD)	CG M (SD)	F	p	μ^2
SWL	23.72 (5.91)	23.81 (5.85)	0.265	0.469	0.003	24.98 (5.62)	23.82 (5.96)	1.384	0.041	0.015	25.67 (5.02)	23.37 (6.03)	2.984	0.009	0.143
PA	21.32 (5.04)	21.03 (5.24)	0.071	0.981	0.001	23.73 (4.92)	21.12 (5.61)	2.127	0.026	0.032	24.01 (4.63)	20.96 (6.12)	2.872	0.041	0.028
NA	17.46 (4.56)	17.91 (4.76)	0.184	0.719	0.001	16.31 (4.32)	17.98 (4.81)	0.631	0.139	0.014	17.04 (5.21)	17.90 (5.11)	0.995	0.532	0.009
TEI	4.25 (0.85)	4.21 (0.89)	0.228	0.641	0.000	4.28 (0.74)	4.22 (0.87)	1.831	0.241	0.003	4.35 (0.62)	4.15 (0.81)	4.126	0.037	0.083
MH	15.48 (5.29)	16.01 (4.99)	2.372	0.077	0.002	16.92 (3.32)	15.39 (4.95)	2.516	0.028	0.021	17.01 (3.42)	15.21 (4.99)	2.351	0.037	0.022
RS	38.53 (7.12)	39.04 (7.01)	0.993	0.164	0.005	41.98 (6.96)	38.92 (6.84)	1.749	0.012	0.043	49.65 (8.01)	37.99 (7.03)	3.768	0.003	0.128

SWL, satisfaction with life; PA, positive affect; NA, negative affect; TEI, trait emotional intelligence; MH, mental health; RS, resilience; μ^2 , eta squared effect size; EG, experimental group (n = 59); CG, control group (n = 58); M, means; SD, standard deviation.



of the EG. The size of the effect was low-effect size ($\mu^2 = 0.043$).

Follow-Up (6 Months)

Results from the pretest follow-up MANCOVA revealed significant differences between the two conditions, Wilks' lambda, $\Lambda = 1.168$; $F(5,96) = 3.581$; $p = 0.009$, with an average effect size ($\mu^2 = 0.298$, $r = 0.27$).

Effects on Subjective Well-Being

The results showed significant improvements in SWL in favor of EG (refer to Table 3). The size of the effect was medium-effect size ($\mu^2 = 0.143$).

Results showed a significant increase in PA scores in favor of EG, without showing a decrease in NA. The size of the effect in PA was low-effect size ($\mu^2 = 0.028$).

Effects on Trait Emotional Intelligence

Concerning the variable TEI, the results confirmed significant improvements in favor of the EG. The size of the effect was low-effect size ($\mu^2 = 0.083$).

Effects on Mental Health

Regarding the variable mental health, the results analyzed showed significant improvements in favor of the EG. The size of the effect was low-effect size ($\mu^2 = 0.022$).

Effects on Resilience

Finally, regarding the resilience variable, the results analyzed showed significant improvements in favor of the EG. The size of the effect was medium-effect size ($\mu^2 = 0.128$).

DISCUSSION

The present study analyzes the effects of an MBI on subjective well-being, TEI, mental health, and resilience in a sample of women with FM. In this sense, Van Gordon et al. (2015) emphasize that, despite the increasing amount of literature on the effects of MBI, it is necessary to continue investigating the effectiveness of these interventions in FM.

The results showed statistically significant improvements among the women who received the intervention compared to those who did not participate. The effects of the MBI in the post-test, on the EG in comparison with CG show the following results: (1) significant increase in SWL (CWB), (2) significant improvement of PA but without a decrease in NA (AWB), (3) no improvement in TEI score, (4) significant increase in mental health, and (5) significant increase in resilience. On the other hand, the effects of the MBI in the follow-up show the following results: (1) significant increase in SWL (CWB), (2) significant improvement of PA but without a decrease in NA (AWB), (3) improvement in the score TEI, (4) significant increase in mental health, and (5) significant increase in resilience.

First, the results show partial improvements in some of the components of subjective well-being. In this sense, the results in the post-test and in the follow-up show an improvement in the SWL, that is to say, according to Diener (1984), improvement of the evaluation of the processing of information that people make of their lives (CWB). The results are consistent with those found in other studies that have demonstrated the efficacy of MBI to improve subjective well-being (Grossman et al., 2007; Quintana and Rincón-Fernández, 2011; Schmidt et al., 2011; Lakhan and Schofield, 2013; Cash et al., 2015; Davis et al., 2015). These results may likely be due to MBI modifying the perception that people with FM have of the negative emotional symptoms caused by pain, facilitating greater subjective well-being (Grossman et al., 2007; Sephton et al., 2007). Concerning the AWB, the results in the post-test and in the follow-up corroborate an improvement in PA. However, a decrease in NA is not obtained in congruence with various studies (e.g., Sephton et al., 2007; Davis and Zautra, 2013; Davis et al., 2015). In this sense, these results reinforce the conclusions of Watson et al. (1988) in that both factors (PA and NA) constitute two independent dimensions of affect and, therefore, are not correlated with each other. These improvements likely derive from the strong relationship between PA and quality of life related to health (e.g., Ong, 2010). A possible explanation for these results could be that PA presents more strong relationships than NA and, therefore, assumes a more relevant position in physical and psychological health (e.g., Fredrickson, 2001; Cohen et al., 2006). Likewise, MBI can be effective in disconnecting the NA response that accompanies pain, promoting subjective well-being (Sephton et al., 2007). The improvement of PA after an MBI is consistent with the results found by other larger studies with a non-clinical population (e.g., Orzech et al., 2009) as well as in the clinical population (e.g., Geschwind et al., 2011). On the other hand, one of the contents of this MBI is compassion. We consider that it can be an

important factor to explain the reduction of certain negative symptomatology as well as the improvement of subjective well-being (Hervás et al., 2016).

Second, the results confirm that there are no significant improvements in the post-test concerning TEI. On the contrary, the results confirm an improvement of TEI in the follow-up. From our point of view, it is likely that the direct relationship between the practice of mindfulness and some of the dimensions of emotional intelligence, such as clarity and emotional repair, may have some influence (De la Fuente et al., 2010), as well as emotional regulation (Huang et al., 2019). On the other hand, some authors (Rodríguez-Ledo et al., 2018) have found positive relationships between the capacities of mindfulness and emotional intelligence. Rodríguez-Ledo et al. (2018) point out that individuals who practice mindfulness present higher scores in intrapersonal competences (e.g., emotional self-perception or emotional self-regulation) and interpersonal competences (e.g., empathy, emotional regulation of others or coping with life's adverse situations) that compound the emotional intelligence construct. In our opinion, an interesting aspect of the results obtained is the appearance of an improvement in the TEI in the follow-up. Thus, if TEI is defined as a constellation of traits related to the typical way an individual processes information of an emotional nature and react to emotional situations, it is likely that individuals need some time to recognize these typical patterns in their habitual behavior.

Third, the results show improvements in mental health in the post-test and in the follow-up. Our results are in line with other studies that have studied the effects of MBI, mainly in depression and anxiety, negatively related to mental health (Sephton et al., 2007; Quintana and Rincón-Fernández, 2011; Parra-Delgado et al., 2012; Lakhan and Schofield, 2013; Amutio et al., 2015; Van Gordon et al., 2017). The positive effects of MBI on mental health can be explained by the influence of these interventions on the improvement that takes place in the automatic processing of emotion, that is, a lower emotional reactivity to negative emotions (Hervás et al., 2016). In this sense, it is likely that the MBI seems to promote the use of more adaptive emotional regulation strategies, such as positive reevaluation, thus improving the mental health of individuals (e.g., Carmody and Baer, 2008). In addition, MBIs seem to reduce the use of maladaptive emotional regulation strategies, such as rumination and catastrophism, that negatively impact the mental health of individuals (Ortner et al., 2007). An unexpected result was that the present MBI did not result in a decrease in NA while it did improve the mental health of the participants. In this regard, it is pertinent to indicate that the results of some studies show stronger relationships between PA and mental health than the relationships between NA and mental health (Vera-Villaruel and Celis-Atenas, 2014). Furthermore, the evidence suggests that PA influences pain and adaptive coping strategies, over and above the influence of NA (Finan and Garland, 2015).

Finally, as regards the resilience variable, the results show an improvement in resilience in the post-test and in the follow-up. As we have previously stated, resilience is defined as a positive personality characteristic that allows the individual to adapt to

adverse situations (Wagnild, 2009). In this sense, we share the idea that among the processes that explain these positive changes in resilience is the influence on acceptance training (included in the present MBI), since it cushions the individual against the impact of life's difficult situations (e.g., Hervás, 2011). We agree with some authors (Hervás et al., 2016) that MBI offers participants a broader repertoire of cognitive and behavioral resources to be effective in situations of a stressful nature.

Limitations and Future Directions

It is important to note some limitations of the present investigation; first, regarding the generalization of the results, since the sample is composed only of women affected by FM, it would be necessary to investigate the effects of MBI on men who are diagnosed with FM due to the unique characteristics they have in these interventions (Kanen et al., 2015). On the other hand, it would be necessary to replicate the experience with more heterogeneous socially and culturally samples. In summary, with these promising results, additional research with MBI is required to extend these findings and test their application. Second, it is necessary to point out that only self-report measures have been used; it would be necessary to support these findings with other measures. However, the data collected with self-reports are related to neuroimaging measures, of a more objective nature (Brewer et al., 2011).

Third, another limitation of the present study, and in general of the MBI, is the difficulty to make comparisons between the results of the different studies due to the heterogeneity of the different MBI (Kanen et al., 2015). This implies that research to promote the effectiveness of MBI should be interpreted with caution given the existing differences in its design, development, and evaluation.

Regarding the future lines of research, the evaluation of the effect of MBI on other variables can be suggested, for example, perception of pain. In this sense, some studies have shown improvements in the perception of pain (e.g., Grossman et al., 2007; Davis et al., 2015); however, other studies do not support these findings (e.g., Schmidt et al., 2011). On the other hand, research should continue on the optimal level of the duration of the MBI, the contents that include the different MBI (Kanen et al., 2015) or the training of MBI instructors (Hervás et al., 2016). Nevertheless, it is necessary to investigate the possible adverse effects of MBI, as some authors warn (e.g., Shapiro, 1992; Didonna and Gonzalez, 2009). Furthermore, we believe that future research should include the analysis of the effects of MBI on the impact of FM on functional capacity and quality of life, through specific instruments such as the Fibromyalgia Impact Questionnaire (FIQ) (Martín et al., 2014).

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CONCLUSION

Given these results, we wish to emphasize that the results obtained in the present investigation suppose empirical support to the use of MBI in women with FM. Also, it is necessary to highlight the importance of implementing MBI in patients with FM to promote subjective well-being, TEL, mental health, and resilience that can act as protective psychological resources that help people cope effective daily demands. We share with some authors that these interventions present a relatively low risk, and therefore, the FM treatment protocols should include this line of treatment (Cash et al., 2015).

Also, the present study can enrich the research on the effects of MBI in people with FM, since maintaining the structure of the MBSR (Kabat-Zinn, 1990; Schmidt et al., 2011) intends to make an adaptation to the characteristics of this collective.

DATA AVAILABILITY STATEMENT

All datasets generated for this study are included in the article/supplementary material.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee for Research on Human Beings, University of Castilla-La Mancha. Patients/participants gave written informed consent to participate in this study. The animal study was reviewed and approved by the National Institutes of Health Animal Care and Use Guidelines.

AUTHOR CONTRIBUTIONS

Each author has made substantial contributions to the work. JC, F-JG-C, and AM-G conceived or designed the work. RF was responsible for audio-guide design. JC and F-JG-C collected data and drafted the manuscript. JC and PL were responsible for data analysis and interpretation. PL, DR-R, and RF were responsible for critical revision of the manuscript. JC, F-JG-C, PL, DR-R, RE, and AM-G approved the final version of the manuscript to be published.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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