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## **Changes in pain-related beliefs, coping, and catastrophizing predict changes in pain intensity, pain interference, and psychological functioning in individuals with Myotonic Muscular Dystrophy and Facioscapulohumeral Dystrophy**

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### **Abstract**

**Objectives**—The primary aim of this study was to test hypothesized associations between changes in psychological variables (i.e., pain beliefs, catastrophizing and coping strategies) and changes in pain intensity and related adjustment (i.e., pain interference and psychological functioning) in individuals with Myotonic Muscular Dystrophy (MMD) and Facioscapulohumeral Muscular Dystrophy (FSHD).

**Methods**—A sample of 107 adults with a diagnosis of MMD or FSHD, reporting pain in the past three months, completed assessments at two time-points, separated by about 24 months.

**Results**—Results showed that changes in pain-related psychological variables were significantly associated with changes in psychological functioning, pain intensity and pain interference. Specifically, increases in the belief that emotion influences pain, and catastrophizing were associated with decreases in psychological functioning. Increases in the coping strategies of asking for assistance and resting, and the increases of catastrophizing were associated with increases in pain intensity. Finally, increases in pain intensity and asking for assistance were associated with increases in pain interference.

**Discussion**—The results support the utility of the biopsychosocial model of pain for understanding pain and its impact in individuals with MMD or FSHD. These findings may inform

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the design and implementation of psychosocial pain treatments for people with muscular dystrophy and chronic pain.

### Keywords

Myotonic dystrophy; Facioscapulohumeral muscular dystrophy; pain; psychological factors

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## INTRODUCTION

Myotonic Muscular Dystrophy (MMD) and Facioscapulohumeral Muscular Dystrophy (FSHD) are two common subtypes of muscular dystrophy<sup>1</sup>. Pain is a widespread problem in persons with both types of muscular dystrophy<sup>2,3</sup>. For example, a recent study by Jensen et al.<sup>4</sup> reported that 71% of a sample of patients with MMD or FSHD had experienced bothersome pain in the last three months. The average pain intensity in their sample was in the moderate range (4.45 on a 0-10 numerical rating scale; NRS), while 25% of participants with pain experienced severe pain intensity (rated 7 or higher on a 0-10 NRS). Patients also reported that pain significantly interfered with their daily activities, primarily with recreational activities and mobility.

Although pain in these populations is likely related to underlying physiological processes, there is consistent evidence indicating that psychological and social factors also play an important role in the experience and impact of pain<sup>5,6</sup>. Specifically, biopsychosocial models propose that pain is best viewed as the product of a complex interaction of physical, cognitive, emotional, behavioral, and social factors<sup>7</sup> that, together, influence the experience of pain and psychological well-being. This model is supported by prior research on numerous patient populations with pain secondary to physical disability, including spinal cord injury<sup>8-10</sup>, amputation<sup>11,12</sup>, cerebral palsy<sup>13</sup>, and, more recently, neuromuscular disease<sup>14</sup>. The model has also provided an important theoretical basis for the design of multidisciplinary pain treatments that seek to address the interplay and effectiveness of these aforementioned factors<sup>15-19</sup>.

In a recent study, specific to neuromuscular disease, Miró et al.<sup>14</sup> analyzed three biopsychosocial domains, including pain beliefs, pain-related catastrophizing, and perceived social support in a sample of persons diagnosed with MMD or FSHD. The results of this study were consistent with the tenets of the biopsychosocial model of pain, and showed that 34% of the variance in psychological functioning in the sample was explained by patients' pain beliefs, catastrophic thinking and perceived social support, above and beyond that explained by pain intensity alone. Moreover, patients' pain beliefs, catastrophic thinking, coping strategies and perceived social support explained 19% of the variance in interference of pain in their daily activities, also above pain intensity.

The findings of this study add to a broader literature supporting the use of a biopsychosocial framework to better understand pain and functioning in persons with degenerative and acute illnesses, as well as those diagnosed by MMD or FSHD. However, a common limitation of the current research is that results are often based on cross-sectional analyses, thus limiting our ability to ascertain whether psychological and social factors are the consequence of functioning or, likewise, if such psychosocial factors directly impact pain-related functioning. In order to better understand the possible direction of these relationships, studies are needed that move beyond the analysis of cross-sectional data, and implement longitudinal designs to investigate whether changes in the predictive factors are also associated with changes in the outcomes.

The purpose of the current study is to investigate the relationship between changes in pain-related psychological variables (i.e., pain beliefs, catastrophizing and pain coping strategies) and changes in pain intensity and related adjustment (i.e., pain interference and psychological functioning) in a sample of individuals with muscular dystrophy (in this case, samples of individuals with MMD or FSHD). We hypothesized that changes in pain-related psychological variables would be significantly associated with changes in pain adjustment; specifically that changes in catastrophizing as well as pain beliefs and coping strategies deemed maladaptive would be associated negatively with psychological functioning and positively with pain intensity and pain interference over time, and that pain beliefs and coping strategies deemed adaptive would show the opposite pattern to measures of pain and patient functioning.

## MATERIALS AND METHODS

### Participants

The participants in the current study consisted of adults with a primary diagnosis of neuromuscular disease (NMD) completing assessments at two time-points (termed initial and 24-month). Participants were recruited from the NIH-funded National Registry of Myotonic Dystrophy and Facioscapulohumeral Muscular Dystrophy Patients and Family Members (<http://www.urmc.rochester.edu/nihregistry/>), and from the University of Washington NMD Clinic. To be eligible to participate in this study, participants must have had a diagnosis of either FSHD or MMD, be at least 18 years old, be able to read and write English, and report pain in the past 3 months (other than occasional headaches and menstrual cramps).

A total of 395 questionnaires were mailed to potential participants for the initial survey. Of these, 97 were not returned (2 because the participant no longer lived at the address on record, 6 were deceased, 5 were returned as ineligible -no MMD diagnosis or less than 18 years of age-, and 84 were not returned for unknown reasons). Thus, a total of 298 surveys were completed and returned, although data from 5 of these could not be analyzed and were, therefore, excluded from further analysis. One hundred eleven of the respondents were not eligible to participate in this study (75 did not report pain and 36 did not have diagnosis of NMD or FSHD). The final sample consisted of 182 individuals, and the data from these participants was used in the analyses of the initial survey (see Miró et al.<sup>14</sup> for more details of the participating sample).

Two years following the initial survey, a second one was mailed to those individuals who had completed the initial survey and reported pain ( $n = 182$ ). One hundred and seven participants completed and returned the 24-month survey (58.8% of the 182 persons that participated in the initial assessment). There were no significant differences in age, pain severity at the initial survey, sex, or education level (all  $ps > .05$ ) between participants who did and those who did not completed both initial and follow-up surveys. The University of Washington Human Subjects Review Committee approved all study procedures. All participants provided informed consent.

Among the 107 respondents with pain included in the current analyses, 61 were female (57.0%) with a mean age of 50.22 years ( $SD = 12.30$ , range = 20 - 83). The majority of the participants were Caucasian (96.3%), married (70.1%), and unemployed (85.9%). All participants had at least a high school education or had obtained their general educational development (GED). Of the 107 respondents, 65.4% were diagnosed with FSHD and 34.6% with MMD. The participants averaged 16.48 years ( $SD=12.16$ , range= 10 months-49 years) since their initial MMD diagnosis. Only 6.5% of the participants had no mobility limitations. Likewise, 48.6% were using some form of assistance for ambulatory circulation.

## Measures

**Outcome Variables—*Pain interference*** was measured using the Brief Pain Inventory (BPI)<sup>20,21</sup>, which includes 7-items asking respondents to indicate the extent to which pain interferes with certain activities (general activity, mood, walking ability, normal work, relations with other people, sleep, and enjoyment of life). For the purposes of assessing pain interference in the current study population, we modified the original scale to: (1) expand the interference domains assessed to three additional activity areas (self-care, recreational activities, and social activities) and (2) ask about interference with *mobility* instead of *ability to walk*, because there are individuals with MD who are unable to walk because of their disability (see the following for more details regarding the modified BPI Interference scale:<sup>11, 14, 22</sup>). The modified BPI Interference scale score (see Table 1) can range from 0 to 10 with a higher score indicating a higher degree of pain-related interference. Our modified 10-item version has evidenced excellent internal consistency (Cronbach's alpha=.89-.95) and validity in previous research examining secondary pain in persons with cerebral palsy<sup>23</sup>, limb loss<sup>11</sup>, and persons with SCI<sup>24</sup>.

**Psychological Functioning:** The 5-item SF-36 Mental Health scale from the SF-36 was used to assess psychological functioning<sup>25</sup>. The SF-36 Mental Health scale is a widely used measure of psychological functioning and has demonstrated good reliability, as evidenced by high internal consistency coefficients (0.81 – 0.95) and test re-test stability coefficients (0.75 – 0.80)<sup>25</sup>. It has also demonstrated validity as a measure of psychological functioning by its association with other measures of psychological functioning<sup>26</sup>. Scores on the SF-36 Mental Health scale range from 0 to 100, with higher scores indicating better psychological functioning. The same 5-item SF-36 Mental Health scale from the SF-36 was administered both at the initial and 24-month surveys (See Table 1).

**Pain Intensity:** Participants were asked, at the time of the initial survey, the following question regarding current pain: “Are you currently experiencing, or have you in the past three months experienced any pain (other than occasional headaches or menstrual cramps)?” Participants who reported pain (those included in this study) were then asked to rate the intensity of their average overall pain in the past week on a 0-10 Numerical Rating Scale, with “0” indicating “no pain” and a “10” indicating “pain as bad as it could be.” This same scale was used at the 24-month survey. Numerical rating scales evidence a strong association with other measures of pain intensity and stability over time thus demonstrating their validity and reliability as measures of pain<sup>27</sup>. The mean and standard deviations of the 0-10 NRS pain intensity measure at each time point, as well as average change scores from the initial to 24-month surveys, are presented in Table 1.

### Process Variables

**Pain Cognitions:** For the initial survey, the standard 57-item Survey of Pain Attitudes was used (SOPA)<sup>28</sup>, while the shorter, 14-item, version of the SOPA was administered at the time of the 24-month survey<sup>29</sup>. Both versions of the SOPA include 7 subscales that measure seven pain-related beliefs, including Control, Disability, Harm, Emotion, Medication, Solicitude, and Medical Cure. The 57-item SOPA has evidenced good internal consistency (Cronbach's alpha=.71-.81), test-retest reliability, and validity<sup>28,30</sup>. The shorter version of the SOPA has also demonstrated excellent validity through its relationship with parent scales, as well as strong correlations with measures of depression, disability, and pain intensity<sup>29</sup>. In order to calculate change scores in the SOPA between the two time periods, only the overlapping items between the two scales were used (see Table 1).

**Pain-Related Catastrophizing:** Pain-Related Catastrophizing was assessed at the initial survey with the 6-item Catastrophizing subscale of the Coping Strategies Questionnaire

(CSQ)<sup>31</sup>, that has evidenced excellent internal consistency reliability<sup>32-36</sup> and adequate validity<sup>12,22,37-40</sup>. For the 24-month survey, the Pain Catastrophizing Scale (PCS)<sup>41</sup> was used. This scale includes 13 statements on catastrophic thinking about pain, and has shown adequate psychometric properties in non-clinical populations<sup>42,43</sup> and in different clinical populations,<sup>44-47</sup>. In the initial survey, the 6-item CSQ catastrophizing scale was used to minimize assessment burden, as many questionnaires were administered for that survey. However, the number of measures was fewer in the second survey, which allowed us to administer the longer measure of catastrophizing that is now used in most of the studies on catastrophizing (the PCS).

In order to standardize these measures across the two assessment points, thus rendering them useful for calculating change scores, we computed scores based only on the five overlapping items between the two measures. In order to ensure that the 5 overlapping items had adequate psychometric properties, and are therefore adequate for assessing catastrophizing, we examined the internal reliability and validity of these items from each assessment. Internal consistency was excellent in both assessments (Chronbach's alpha = .90 for the initial assessment, and .87 for the 24-month assessment) and were strongly correlated with one another ( $r = .54, p < .001$ ), supporting both their reliability and validity as measures of catastrophizing. Finally, we standardized the scales for comparison, as they were each measured on different Likert scales, using the strategy implemented for the SF-36<sup>25</sup>. With this procedure both measurements were converted to the same 0 to 100 scale (see Table 1).

**Coping:** The standard 70-item Chronic Pain Coping Inventory (CPCI)<sup>48</sup> was administered at the time of the initial survey, while a shorter 12-item version of the CPCI<sup>29</sup> (this included a one-item version of each scale, except for Pacing, which, in the version administered for this study, included all original 5 items, as a brief version of the CPCI Pacing scale has not yet been developed) was included in the 24-month survey. Both the standard and brief versions of the CPCI assess the frequency with which participants use nine coping strategies for pain management, including Guarding, Resting, Asking for Assistance, Relaxation, Task Persistence, Seeking Social Support, Coping Self-Statements, Exercise/Stretch and Pacing. However, the Exercise/Stretch scale items were not included in the 24-month survey due to an oversight. The validity of the 70-item CPCI is demonstrated by strong correlations between patient and significant other versions of the scales, as well as with measures of depression and adjustment to pain<sup>49,50</sup>. The shorter 12-item version of the CPCI has also demonstrated robust validity, evidenced by strong correlations between the two-item subscales and parent scales, as well as correlations with measures of depression, disability, and pain intensity<sup>29</sup>.

For the purposes of this paper we only examined the overlapping CPCI items administered at each assessment point. Specifically, we examined the single items that overlapped and assessed Guarding, Resting, Asking for Assistance, Relaxation, Task Persistence, Seeking Social Support, and Coping Self-Statements. Because all 5 original items for the Pacing Scale were available in both surveys, we utilized the mean score for these five items from both time-points (see Table 1).

## Data Analyses

We first performed a series of analyses (zero-order correlations, independent samples t-tests, and ANOVAs) to examine the extent to which demographic characteristics (i.e., age and education) and change in pain intensity (e.g., pain intensity in previous week the time of the 24-month survey minus pain intensity at the time of the initial survey) were related with the outcome variables of interest, including change in psychological functioning, change in pain interference, and change in pain intensity, in order to determine which variables might be needed as control variables in the planned multivariate analyses. Change in average pain

intensity from the initial to the 24-month assessment was significantly positively correlated with change in psychological functioning ( $r = .22, p < .05$ ) and change in pain interference ( $r = .42, p < .01$ ). Thus, change in pain intensity was entered first in subsequent regression analyses to control for this variable. No other potential control variables were significantly related to any of the outcome variables.

We then subjected the CPCI and SOPA subscale change scores to principal components analyses (PCA) in order to reduce the number of predictor variables in the planned regression equations. Direct Oblimin was chosen to allow correlations between different components. We used the scree plot test, Kaiser criterion, amount of variance explained, and the coherence of the structure to determine the number of factors for each component analysis.

The results of the PCA of the CPCI subscales showed a clear two-factor structure which accounted for 40.49% of the variance. Four scale scores loaded on the first component, including Task Persistence (component loading = .63), Asking for Assistance (.53), Seeking Social Support (.72), and Relaxation (.54). Three scale scores loaded on the second component, including Coping Self-Statements (.59), Guarding ( $r = .55$ ) and Resting (.69). Pacing was partially loaded onto both component 1 (.41) and component 2 (.36).

The results of the PCA of the SOPA showed a three-factor structure that accounted for 59.38% of the variance. The subscale scores that loaded on the first component included Control (component loading =  $-.80$ ), and Harm (.79) scale scores. The second component included the Disability (.76), and Emotion (.73) scales. The third component included only the Solicitude (.72) scale. Medication Cure loaded on both component 2 (.49) and component 3 (.56). Likewise, Medical Cure loaded equally on component 1 ( $-.53$ ) and component 3 (.53).

To test the study hypotheses, we performed a series of three regression analyses, including component scores from the factor analyses of the SOPA and CPCI scales, and scores from the catastrophizing scale (representing pain cognitions and coping), as the primary predictors. Change in pain intensity was entered in the first step of the regression analyses predicting change in pain interference and change in psychological functioning, to control for its effect on these criterion measures. To help better understand the specific factors that contributed to the significant effects found in the regression analyses, we also examined the univariate relationships between specific psychosocial variables, including change scores of each subscale of the CPCI, SOPA and catastrophizing scales, and the outcome variables of interest (change in pain interference, pain intensity, and psychological functioning) using correlation analyses.

Due to the large number of predictive variables and statistical tests performed, we used an alpha of .01 to balance the needs to control for both Type I, as well as Type II, errors<sup>26</sup>.

## RESULTS

### Associations Between Psychosocial Variables and Change in Psychological Functioning

Change in pain intensity demonstrated a non-significant trend ( $p < .05$ ) to be associated with change in psychological functioning, explaining 5% of the variance. After controlling for change in pain intensity, cognitions and coping strategies as a whole also demonstrated a non-significant trend ( $p < .05$ ) to be associated with change in psychological functioning, and accounted for an additional 12% of the variance. However, none of the pain cognitions and coping variables uniquely predicted psychological functioning (see Table 2).

In the univariate analyses, no CPCI subscale emerged as significantly associated with psychological functioning. However, changes in the SOPA Emotion subscale ( $r = .27, p < .01$ ) were associated with changes in psychological functioning; larger initial to 24-month increases in this subscale were associated with worsening of psychological functioning. Catastrophizing change scores were also positively associated with changes in psychological functioning ( $r = .25, p < .01$ ). Finally, there was a non-significant trend between increases in pain intensity and worsening of psychological functioning ( $r = .22, p < .05$ ; see Table 3).

### **Associations Between Psychosocial Variables and Change in Pain Interference**

Initial to 24-month changes in pain intensity showed a positive non-significant trend ( $p < .05$ ) to be associated with changes in pain interference, explaining 18% of the variance in this criterion. Changes in the cognition and coping variables as a whole accounted for an additional 6% of the variance in changes in pain interference scores, after controlling for change in pain intensity, but as a block did not evidence a significant contribution to the variance of the outcome. However, the SOPA component 2 made a significant and unique contribution to the prediction of the criterion, with more change in this component associated with more change in pain interference ( $p < .01$ ) (See Table 4).

The univariate analyses indicated that Asking for Assistance was significantly and positively associated with change in pain interference ( $r = .33, p < .01$ ). Change scores of the SOPA Medication subscale, as well as the change in Catastrophizing, also evidenced a non-significant positive trend with change in pain interference (each  $r = .19, ps = .05$ ). Finally, increased pain intensity over time was strongly associated with an increase in pain interference over time ( $r = .42, p < .01$ ) (see Table 4).

### **Associations Between Psychosocial Variables and Change in Pain Intensity**

In the regression analysis predicting change in pain intensity, the cognitions and coping variables as a whole showed a non-significant trend ( $p < .05$ ) to be associated with the criterion, accounting for 16% of the variance. Change in Catastrophizing made a unique and significant contribution to the prediction of the criterion ( $p < .01$ ), such that increases in Catastrophizing ( $p < .01$ ) were associated with increases in pain intensity (See Table 5). There was also a non-significant positive trend towards the relation between CPCI Component 1 and pain intensity ( $p < 0.05$ ).

The results of the univariate analyses indicated that change in the CPCI Resting ( $r = .29, p < .01$ ) and Asking for Assistance ( $r = .28, p < .01$ ) subscales were significantly and positively correlated with change in pain intensity. Finally, consistent with the multivariate analyses, change in Catastrophizing was positively and significantly associated with change in pain intensity ( $r = .29, p < .01$ ) (see Table 3).

## **DISCUSSION**

In general, the most important contribution of this longitudinal study is the finding that, as hypothesized, changes in psychosocial factors predicted changes in the experience of, and adjustment to, pain in persons with MMD or FSHD, thus providing support for a biopsychosocial model to understand pain and its impact in this patient population. These results add to a growing literature supporting the use of a biopsychosocial model to better understand pain in numerous patient populations, as well as the potential efficacy of psychosocial pain treatments to help individuals with MD and chronic pain. More specifically, our multivariate analyses revealed that changes in catastrophizing significantly predicted changes in pain intensity, and that changes in SOPA component 2 significantly predicted changes in pain interference after controlling for changes in pain intensity.

However, in the regression analyses, no block or individual predictor was significantly associated with psychological functioning.

Taken together, these findings are in accordance with our previous cross-sectional study<sup>14</sup> in which concurrent scores in psychological variables (i.e., beliefs, catastrophizing and coping) and social support were significantly related with concurrent pain interference. Similar results have been found in the recent study by Hanley et al.<sup>8</sup> that also assessed the effects of changes of psychological variables in a sample of persons with pain secondary to cerebral palsy, as well as other populations with disabilities and chronic pain, including spinal cord injury<sup>9,10</sup>, and amputation<sup>11,12</sup>.

While the primary goal of the current longitudinal study was to further establish the importance of a biopsychosocial model, we also examined the univariate relationships between specific psychosocial variables (including change scores of each subscale of the CPCI and SOPA, and catastrophizing), and the outcome variables of interest. Concerning coping strategies (as measured by subscales of the CPCI), increases in “asking for assistance” was associated with increases in pain intensity and pain interference over time; and increases in the use of “resting” was related with increases in pain intensity. Because these findings are correlational, it is not possible to conclude that the coping responses of “asking for assistance” and “resting” contribute to increases in pain, if increases in pain contribute to a greater frequency of use of these coping strategies, or if there is a third variable or confound that influences both and is responsible for the significant associations found. However, because correlation is a necessary (but not sufficient) criterion for causality, the findings are consistent with the possibility that use of these coping responses might contribute to greater pain over time; for example, via a mechanism by which greater reliance on others (asking for help with a chore or task) and increased resting in response to pain contribute to decreased activity and deconditioning. Future research is needed to determine if experimental manipulation of these coping strategies (e.g., random assignment to conditions that (a) have little impact on rest vs. (b) decrease the use of rest as a coping response) have a causal impact on pain intensity over time. Certainly, however, these results are consistent with our previous cross-sectional study<sup>14</sup> and previous literature showing the potential importance of coping strategies in chronic pain<sup>51</sup>, especially the negative associations between both resting and asking for assistance and pain intensity<sup>9,52-54</sup>.

Concerning beliefs and cognitions (as measured by the SOPA), univariate analyses showed that increases in the belief that emotions influence pain was related with decreases in psychological functioning over time. However, this result should be interpreted with caution, because none of the predictors in the multivariate regression analysis evidenced a significant ( $p < .01$ ) association with changes in psychological functioning. Also, even if this finding were reliable, it is not possible to determine the direction of causation from these analyses. To the extent that future research shows that beliefs in the effects of emotions on pain make one more vulnerable to psychological distress this would support the need to develop treatments that target these beliefs, specifically.

The univariate results also revealed that catastrophizing was one of the variables most widely related with the outcomes assessed in our study, consistent with a large body of research supporting the importance of catastrophizing in many other samples of chronic pain conditions, including samples of individuals with physical disabilities<sup>9-13,22, 55,56</sup>. Increases in catastrophic thinking were related with worsening pain intensity in both the univariate and multivariate analyses, and with mental health in the univariate analyses. Besides the limitations associated with correlational analyses previously discussed, this finding adds to the growing literature regarding the importance of catastrophizing in the context of numerous pain populations<sup>6,57,58</sup>, and are also consistent with our previous

cross-sectional study of the same sample<sup>14</sup>. Altogether, these data suggest the central role that catastrophic thinking may exert in the experience and impact of pain and calls for additional research to examine more closely the causal direction of this relationship.

As mentioned earlier, the findings from this study may inform relevant therapeutic alternatives for patients with MMD and FSHD diseases. Treatments should be developed on the basic tenets of the biopsychosocial model, including cognitive and behavioural interventions aimed at changing maladaptive thoughts, such as catastrophizing, and teaching as well as encouraging the use of more adaptive coping strategies. A substantial amount of evidence supports the use of these treatments for other chronic pain populations<sup>15-19</sup>. Moreover, several studies have also shown that treatment-related changes in coping strategies and cognitions are related with improvements in pain adjustment<sup>26,59</sup>. Cognitive-behavioral approaches for pain treatment should be tested in persons with NMD and FSHD in order to evaluate its effectiveness and further unravel the direction and importance of the aforementioned variables identified as important correlates of changes to the outcomes in the current study.

Several limitations of this study should be noted. First, the sample size was relatively small and there was no follow-up data for almost half of the sample, thus bringing into question the external validity of our findings. Moreover, we cannot be sure that our sample is representative of the population of persons with FSHD and/or MMD. Research testing the associations between variables examined in this study in additional samples of individuals with FSHD and NMD is needed to establish their reliability. Second, at the 24-month follow-up we used brief measures for coping strategies and beliefs in order to reduce assessment burden. Although these measures have shown adequate psychometric properties<sup>29</sup>, this presented unique challenges in the process of standardizing the measures at both time-points and possible differences in the psychometric properties of these measures between assessment periods may have influenced the results in ways that are not easily assessed. Third, although we used a longitudinal design, which is somewhat more reliable for testing causal relationships than cross-sectional designs, it must be acknowledged that results are still correlational. Therefore, we cannot determine the direction of causality in the associations found. Studies using experimental methodology are needed to truly examine the potential causal relationships. Also, we did not measure a number of psychological and social variables that have been supported as significant predictors of patient functioning in previous studies. One of these, related with pain adjustment in our previous study<sup>14</sup>, is social support, which has evidenced an important relationship with similar outcomes in other studies, as well<sup>10-12</sup>. Finally, we did not assess a number of important disease-related variables (for example, disease severity or progression), which would have allowed us to determine how these variables are associated with the criterion variables studied, as well as the extent to which the psychosocial variables examined contributed to the prediction of the criterion variables over and above disease-related factors. Future researchers should assess these variables in order to help build a more thorough biopsychosocial understanding of the factors that may contribute to pain, psychological functioning, and disability.

In sum, the current study contributes to the increasing evidence that the biopsychosocial model provides a useful framework for understanding pain and pain adjustment in populations of individuals with physical disabilities, including persons with MMD and FSHD. The findings also suggest that interdisciplinary pain treatments that have proven efficacy in other samples of patients with pain should be tested in samples of patients with muscular dystrophy and chronic pain, and that such treatments should continue to focus on a broad range of biopsychosocial factors (e.g., cognitions and beliefs, coping responses) as each one of these has the potential to exert a role in the experience of pain and its impact on quality of life.

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## References

1. Hugues MI, Hicks EM, Nevin NC, et al. The prevalence of inherited neuromuscular disease in northern Ireland. *Neuromusc. Disord.* 1996; 6:69–73. [PubMed: 8845721]
2. Bushby KMD, Pollitt C, Johnson MA, et al. Muscle pain as a prominent feature of facioscapulohumeral muscular dystrophy (FSHD): four illustrative case reports. *Neuromuscul Disord.* 1998; 8:574–579. [PubMed: 10093064]
3. George A, Schneider-Gold Ch, Zier S, et al. Musculoskeletal pain in patients with myotonic dystrophy type 2. *Arch Neur.* 2004; 61:1938–1942.
4. Jensen MP, Hoffman AJ, Stoelb BL, et al. Chronic pain in persons with myotonic dystrophy and facioscapulohumeral dystrophy. *Arch Phys Med Rehab.* 2008; 89:320–328.
5. Gatchel RJ, Peng YB, Peters ML, et al. The Biopsychosocial Approach to Chronic Pain: Scientific Advances and Future Directions. *Psychol Bull.* 2007; 133:581–624. [PubMed: 17592957]
6. Keefe FJ, Rumble ME, Scipio CD, et al. Psychological aspects of persistent pain: current state of the science. *J Pain.* 2004; 5:195–211. [PubMed: 15162342]
7. Turk, DC.; Meichenbaum, D.; Genest, M. *Pain and Behavioral Medicine: A cognitive-behavioral perspective.* Guilford Press; New York: 1983.
8. Hanley MA, Raichle K, Jensen M, et al. Pain catastrophizing and belief predict changes in pain interference and psychological functioning in persons with spinal cord injury. *J Pain.* 2008; 9:863–871. [PubMed: 18550442]
9. Molton IR, Stoelb BL, Jensen MP, et al. Psychosocial factors and adjustment to chronic pain in spinal cord injury: replication and cross-validation. *JRRD.* 2009; 1:31–42.
10. Raichle KA, Hanley M, Jensen MP, et al. Cognitions, coping, and social environment predict adjustment to pain in spinal cord injury. *J Pain.* 2007; 8:718–29. [PubMed: 17611163]
11. Hanley MA, Jensen MP, Ehde DM, et al. Psychosocial predictors of long-term adjustment to lower-limb amputation and phantom limb pain. *Disabil Rehab.* 2004; 26:882–893.
12. Jensen MP, Ehde DM, Hoffman AJ, et al. Cognitions, coping and social environment predict adjustment to phantom limb pain. *Pain.* 2002; 95:133–142. [PubMed: 11790476]
13. Jensen MP, Engel JM, Schwartz L. Coping with cerebral palsy: a preliminary longitudinal study. *Pain Med.* 2006; 7:30–37. [PubMed: 16533194]
14. Miró J, Raichle KA, Carter GT, et al. Impact of biopsychosocial factors on chronic pain in persons with myotonic and facioscapulohumeral muscular dystrophy. *Am J Hosp Palliat Med.* 2009; 26:308–319.
15. Eccleston C, Malleson PN, Clinch J, et al. Chronic pain in adolescents: evaluation of a programme of interdisciplinary cognitive behaviour therapy. *Arch Dis Child.* 2003; 88:881–885. [PubMed: 14500306]
16. Flor H, Fydrich T, Turk DC. Efficacy of multidisciplinary pain treatment centers: a meta-analytic review. *Pain.* 1992; 49:221–230. [PubMed: 1535122]
17. Palermo TM, Eccleston C, Lewandowsky AS, et al. Randomized controlled trials of psychological therapies for management of chronic pain in children and adolescents: An updated meta-analytic review. *Pain.* 2010; 148:387–393. [PubMed: 19910118]
18. Scascighini L, Toma V, Dober-Spielmann D, et al. Multidisciplinary treatment for chronic pain: a systematic review of interventions and outcomes. *Rheumatology.* 2008; 47:670–678. [PubMed: 18375406]
19. Turk DC, Okifuji A. Efficacy of multidisciplinary pain centres: an antidote to anecdotes. *Baillieres Clin Anaesthesiol.* 1998; 12:103–119.
20. Cleeland CS, Ryan KM. Pain assessment: global use of the brief pain inventory. *Ann Acad Med.* 1994; 23:129–138.

21. Daut RL, Cleeland CS, Flanery RC. Development of the Wisconsin brief pain questionnaire to assess pain in cancer and other diseases. *Pain*. 1983; 17:197–210. [PubMed: 6646795]
22. Turner JA, Jensen MP, Warms CA, et al. Catastrophizing is associated with pain intensity, psychological distress, and pain-related disability among individuals with chronic pain after spinal cord injury. *Pain*. 2002; 98:127–134. [PubMed: 12098624]
23. Tyler EJ, Jensen MP, Engel JM, et al. The reliability and validity of pain interference measures in persons with cerebral palsy. *Arch Phys Med Rehab*. 2002; 83:236–239.
24. Raichle KA, Osborne TL, Jensen MP, et al. The reliability and validity of pain interference measures in persons with spinal cord injury. *J Pain*. 2006; 7:179–186. [PubMed: 16516823]
25. Ware, J.; Snow, K.; Kosinski, M., et al. *Sf-36 health survey: Manual and interpretation guide*. Quality Metric Incorporated; Lincoln, RI: 2000.
26. Jensen MP, Turner JA, Romano JM. Changes in beliefs, catastrophizing and coping are associated with improvement in multidisciplinary pain treatment. *J Consul Clin Psych*. 2001; 69:655–662.
27. Jensen MP, Turner JA, Romano JM, et al. Comparative reliability and validity of chronic pain intensity measures. *Pain*. 1999; 83:157–162. [PubMed: 10534586]
28. Strong J, Ashton R, Chant D. The measurement of attitudes towards and beliefs about pain. *Pain*. 1992; 48:227–236. [PubMed: 1534165]
29. Jensen MP, Keefe FJ, Lefebvre JC, et al. One- and two-item measures of pain beliefs and coping strategies. *Pain*. 2003; 104:453–469. [PubMed: 12927618]
30. Jensen MP, Turner JA, Romano JM, et al. Relationship of pain-specific beliefs to chronic pain adjustment. *Pain*. 1994; 57:301–309. [PubMed: 7936708]
31. Roth RS, Lowery JC, Hamill JB. Assessing persistent pain and its relation to affective distress, depressive symptoms, and pain catastrophizing in patients with chronic wounds: a pilot study. *Am J Phys Med Rehab*. 2004; 83:827–834.
32. Gil KM, Abrams MR, Phillips G, et al. Sickle cell disease pain: relation of coping strategies to adjustment. *J Consul Clin Psych*. 1989; 57:725–731.
33. Hill A, Niven CA, Knussen C. The role of coping in adjustment to phantom limb pain. *Pain*. 1995; 62:79–86. [PubMed: 7478711]
34. Keefe FJ, Caldwell DS, Queen K, et al. Osteoarthritic knee pain: A behavioral analysis. *Pain*. 1987; 28:309–321. [PubMed: 2952935]
35. Keefe FJ, Caldwell DS, Queen KT, et al. Pain coping strategies in osteoarthritis patients. *J Consult Clin Psychol*. 1987; 55:208–212. [PubMed: 3571674]
36. Rosenstiel AK, Keefe FJ. The use of coping strategies in chronic low back pain patients: relationship to patient characteristics and current adjustment. *Pain*. 1983; 17:33–44. [PubMed: 6226916]
37. Jensen, MP.; Karoly, P. Self-report scales and procedures for assessing pain in adults. In: Turk, DC.; Melzack, R., editors. *Handbook of pain assessment*. Guilford Press; New York: 1992. p. 15-34.
38. Keefe FJ, Williams DA. A comparison of coping strategies in chronic pain patients in different age groups. *J Gerontol*. 1990; 45:161–165.
39. Sullivan MJ, D'Eon JL. Relation between catastrophizing and depression in chronic pain patients. *J Abnorm Psych*. 1990; 99:260–263.
40. Tan G, Jensen MP, Robinson-Whelen S, et al. Coping with chronic pain: a comparison of two measures. *Pain*. 2001; 90:127–133. [PubMed: 11166978]
41. Sullivan MJL, Pivik J. The pain catastrophizing scale: development and validation. *Psychological Assessment*. 1995; 7:524–532.
42. D'Eon JL, Harris CA, Ellis JA. Testing factorial validity and gender invariance of the pain catastrophizing scale. *J Behav Med*. 2004; 27:361–372. [PubMed: 15559733]
43. Osman A, Barrios FC, Kopper BA, et al. Factor structure, reliability, and validity of the pain catastrophizing scale. *J Behav Med*. 1997; 20:589–605. [PubMed: 9429990]
44. Chibnall J, Tait RC. Confirmatory factor analysis of the pain catastrophizing scale in african american and caucasian workers' compensation claimants with low back injuries. *Pain*. 2005; 113:369–375. [PubMed: 15661446]

45. Miró J, Nieto R, Huguet A. The Catalan version of the Pain Catastrophizing Scale: A useful instrument to assess catastrophic thinking in whiplash patients. *J Pain*. 2008; 9:397–406. [PubMed: 18226964]
46. Osman A, Barrios FC, Gutierrez PM, et al. The pain catastrophizing scale: further psychometric evaluation with adult samples. *J Behav Med*. 2000; 23:351–365. [PubMed: 10984864]
47. Van Damme S, Crombez G, Bijttebier P, et al. A confirmatory factor analysis of the Pain Catastrophizing Scale: invariant factor structure across clinical and non-clinical populations. *Pain*. 2002; 96:319–324. [PubMed: 11973004]
48. Jensen, MP.; Turner, JA.; Romano, JM., et al. *Chronic Pain Coping Inventory: Professional Manual*. Psychological Assessment Resources; Florida: 2008.
49. Jensen MP, Turner JA, Romano JM, et al. The Chronic Pain Coping Inventory: development and preliminary validation. *Pain*. 1995; 60:203–216. [PubMed: 7784106]
50. Nielson WR, Jensen MP, Hill ML. An activity pacing subscale for the chronic pain coping inventory: Development in a sample of patients with fibromyalgia syndrome. *Pain*. 2001; 89:111–115. [PubMed: 11166466]
51. Boothby, JL.; Thorn, BE.; Stroud, MW., et al. Coping with pain. In: Gatchel, RJ.; Turk, DC., editors. *Psychosocial factors in pain*. The Guilford Press; New York: 1999. p. 343-359.
52. Ersek M, Turner JA, Kemp CA. Use of the chronic pain coping inventory to assess older adults' pain coping strategies. *J Pain*. 2006; 11:833–842. [PubMed: 17074625]
53. Tan G, Nguyen Q, Anderson KO, et al. Further validation of the chronic pain coping inventory. *J Pain*. 2005; 6:29–40. [PubMed: 15629416]
54. Truchon M, Côté D. Predictive validity of the Chronic Pain Coping Inventory in subacute low back pain. *Pain*. 2005; 116:205–212. [PubMed: 15927382]
55. Osborne TL, Jensen MP, Edhe DM, et al. Psychosocial factors associated with pain intensity, pain-related interference, and psychological functioning in persons with multiple sclerosis and pain. *Pain*. 2007; 121:52–62. [PubMed: 16950570]
56. Engel JM, Jensen MP, Schwartz L. Coping with chronic pain associated with cerebral palsy. *Occup Ther Int*. 2006; 13:224–233. [PubMed: 17623374]
57. Leeuw M, Goznes ME, Linton SJ, et al. The fear-avoidance model of musculoskeletal pain: current state of scientific evidence. *J Behav Med*. 2007; 30:77–94. [PubMed: 17180640]
58. Sullivan MJL, Thorn B, Haythornthwaite JA, et al. Theoretical perspectives on the relation between catastrophizing and pain. *J Pain*. 2001; 17:52–64.
59. Turner JA, Whitney C, Dworkin SF, et al. Do changes in patient beliefs and coping strategies predict temporomandibular disorder treatment outcomes? *Clin J Pain*. 1995; 11:177–88. [PubMed: 8535036]

**Table 1**

## Descriptive Statistics of Study Measures

Measures	Initial Survey Mean (SD)	24-Month Survey Mean (SD)	Change Scores (24-Month - Initial scores) Mean (SD)	Range of change scores
0 – 10 NRS	4.74 (2.39)	4.85 (2.31)	.11 (2.30)	–6.00 – 8.00
BPI Interference scale	3.14 (2.40)	3.23 (2.23)	.09 (2.07)	–6.33 – 6.17
SF-36 MHS	31.36 (18.39)	32.88 (19.81)	1.56 (16.37)	–57.00 – 63.00
Catastrophizing (CSQ, PCS)	21.56 (20.13)	18.92 (22.12)	–2.63 (20.54)	–66.67 – 76.67
SOPA				
Control	1.87 (.77)	2.26 (.95)	.38 (1.13)	–2.50 – 4.00
Disability	2.00 (1.11)	1.93 (1.14)	–.08 (1.85)	–4.00 – 4.00
Harm	1.83 (.94)	2.20 (.98)	.37 (1.39)	–3.00 – 4.00
Emotion	1.70 (1.14)	1.88 (1.22)	.17 (1.29)	–3.00 – 3.50
Solicitude	1.74 (1.10)	1.88 (1.14)	.14 (.99)	–3.00 – 2.50
Medication	1.53 (.78)	2.81 (1.02)	1.30 (1.14)	–2.00 – 4.00
Medical Cure	2.04 (.79)	1.28 (1.05)	–.75 (1.30)	–4.00 – 3.00
CPCI				
Guarding	4.72 (2.76)	4.65 (2.72)	–.20 (2.98)	–7.00 – 7.00
Resting	3.87 (3.04)	2.70 (2.72)	–1.17 (3.22)	–7.00 – 7.00
Asking for Assistance	3.62 (2.58)	3.08 (2.43)	–.53 (2.57)	–7.00 – 7.00
Relaxation	1.97 (2.42)	2.38 (2.64)	.41 (3.12)	–7.00 – 7.00
Task Persistence	4.09 (2.60)	3.69 (2.60)	–.40 (3.03)	–7.00 – 7.00
Coping Self-Statements	2.00 (2.56)	2.20 (2.54)	.20 (2.88)	–7.00 – 7.00
Seek Social Support	1.15 (2.01)	2.08 (2.59)	.93 (2.93)	–7.00 – 7.00
Pacing	3.60 (2.02)	4.16 (2.05)	.56 (2.20)	–4.40 – 7.00

Note: 0 – 10 NRS = 0 to 10 Numerical Rating Scale of pain intensity; BPI = Modified Brief Pain Inventory Pain Interference scale; SF-36 MHS = Mental Health Scale; SOPA = Survey of Pain Attitudes; CSQ = Coping Strategies Questionnaire; CPCI = Chronic Pain Coping Inventory; PCS = Sullivan's Pain Catastrophizing Scale.

Note: 0 – 10 NRS = 0 to 10 Numerical Rating Scale of pain intensity; BPI = Modified Brief Pain Inventory Pain Interference scale; SF-36 MHS = Mental Health Scale; SOPA = Survey of Pain Attitudes; CSQ = Coping Strategies Questionnaire; CPCI = Chronic Pain Coping Inventory; MSPSS = Multi-Dimensional Scale of Perceived Social Support.

**Table 2**

Multiple Regression Analyses Predicting Change in Psychological functioning from Change in Cognitions & Coping (n = 101)<sup>Δ</sup>

Step and Variables	Total R <sup>2</sup>	R <sup>2</sup> change	F change	Beta
1. Pain intensity	.05	.05	5.67*	.23*
2. Cognitions and Coping	.17	.12	2.25*	
Catastrophizing				.18
SOPA Component 1				.10
SOPA Component 2				.17
SOPA Component 3				.15
CPCI Component 1				-.19
CPCI Component 2				.03

\* p<.05,

\*\* p< 0.01,

<sup>Δ</sup>Total n < 107 due to missing data

**Table 3**

Zero-Order Correlation Coefficients between Subscale Change Scores of the Psychosocial Variables with Change Scores of Psychological functioning, Brief Pain Inventory and Pain Intensity Scores (24-month - Initial)

Cognition/coping change scores	Outcome Measures (Change Scores)		
	BPI Interference Scale	SF-36 Mental Health	Pain Intensity
<u>CPCI subscales</u>			
Guarding	.00	-.03	.06
Resting	-.01	-.01	.29*
Asking for Assistance	.33*	-.04	.28*
Relaxation	.03	-.04	.13
Coping Self-Statements	-.20	.04	-.15
Seek Social Support	.02	-.04	.09
Task Persistence	-.10	-.08	.04
Pacing	-.07	.04	-.07
<u>SOPA subscales</u>			
Disability	.15	.14	.02
Harm	-.02	.04	-.01
Medication	.19 <sup>†</sup>	.04	.08
Medical Cure	-.06	-.01	-.09
Emotion	.16	.27*	.01
Solicitude	-.06	.15	.06
Control	-.19	-.11	-.11
Catastrophizing	.19 <sup>†</sup>	.25*	.29*
Pain Intensity	.42*	.22 <sup>†</sup>	NA

<sup>†</sup> non-significant trend,

\*  $p < 0.01$ , two-tailed.

Note. These correlations and significant levels are presented for descriptive purposes. Given the large number of correlations performed on related variables we used an alpha cut-off of .01 to control for both Type I and Type II error. SOPA = Survey of Pain Attitudes; CPCI = Chronic Pain Coping Inventory

**Table 4**

Multiple Regression Analyses Predicting Change in Pain Interference from Change in Cognitions & Coping (n=102)<sup>Δ</sup>

Step and Variables	Total R <sup>2</sup>	R <sup>2</sup> change	F change	Beta
1. Pain Intensity	.18	.18	21.16*	.42*
2. Cognitions and Coping	.24	.06	1.23	
Catastrophizing				.05
SOPA Component 1				.05
SOPA Component 2				.21**
SOPA Component 3				-.03
CPCI Component 1				-.06
CPCI Component 2				-.07

\* p<.05,

\*\* p< 0.01,

<sup>Δ</sup>Total n < 107 due to missing data.

**Table 5**

Multiple Regression Analyses Predicting Change in Pain Intensity from Change in Cognitions & Coping (n=102)<sup>A</sup>

Step and Variables	Total R <sup>2</sup>	F change	Beta
Cognitions and Coping	.16	2.92*	
Catastrophizing			.31**
SOPA Component 1			.03
SOPA Component 2			-.02
SOPA Component 3			-.01
CPCI Component 1			.23*
CPCI Component 2			.06

\* p<.05,

\*\* p< 0.01,

<sup>A</sup>Total n < 107 due to missing data