

Pain coping and acceptance as longitudinal predictors of health-related quality of life among people with hemophilia-related chronic joint pain

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Abstract

Background: Interventions based on coping and acceptance can be adapted for people with different painful conditions. Evidence about baseline characteristics that predict improved outcomes is informative for matching people to interventions, whereas evidence about changes that predict improved outcomes is informative about the processes that interventions should target.

Methods: Participants in a low-intensity program to promote self-management of hemophilia-related chronic joint pain (n=101) reported pain intensity, coping, acceptance and quality of life at baseline and 6-month follow-up. Baseline and change measures of pain intensity, coping and acceptance were used to predict follow-up quality of life, taking account of baseline quality of life.

Results: Changed (reduced) pain intensity predicted better physical quality of life, independently of age, hemophilia severity, baseline pain intensity and baseline physical quality of life. Lower baseline passive coping and changed (increased) pain acceptance predicted better mental quality of life, independently of age, severity, and baseline mental quality of life. Increased activity engagement but not pain willingness predicted better mental quality of life when pain acceptance was decomposed. Changed (reduced) negative thoughts also predicted better mental quality of life when separate acceptance subscales were used. Active pain coping did not predict physical or mental quality of life.

Conclusions: Initially high levels of passive coping may be an obstacle to improving mental quality of life. Acceptance rather than coping may be a more useful behavioral change target, but more research is needed about the meanings and therapeutic implications of different elements of pain acceptance.

Keywords: Hemophilia; joint pain; coping; acceptance; quality of life

What is already known about this topic: Quality of life among people with chronic pain conditions can be affected by pain coping, negative thinking and acceptance.

What this study adds: Those factors could also be targets for behavior change in interventions adapted for people with chronic pain secondary to other conditions, such as hemophilia.

1. Introduction

Recent psychotherapeutic approaches for chronic pain have developed from acceptance and commitment therapy, which focuses on the context and function of distressing experiences, rather than their content (Hayes, 2004). Examples include acceptance-based treatment (McCracken et al., 2005), acceptance and commitment therapy (Vowles et al., 2011; Wetherell et al., 2011), acceptance and exposure (Wicksell et al., 2007), contextual cognitive behavioral therapy (McCracken et al., 2007a), and values-based action (Vowles and McCracken, 2008). A common aim is to increase pain acceptance, or “willingness to experience continued pain without attempting to control, avoid or change it” (McCracken, 1999, p. 93).

Acceptance-based approaches developed as alternatives to coping-based approaches, which aimed to increase coping strategies and reduce negative thoughts, using techniques from cognitive behavioral therapy (Thorn, 2004). Coping-based approaches have been broadly effective (Hoffman et al., 2007), but are limited because when pain is chronic, attempts to control or avoid it can inevitably be unsuccessful and counter-productive.

The relative utility of coping-based and acceptance-based approaches will differ between individuals, groups and settings, especially as interventions developed for people with chronic pain as a primary diagnosis are adapted for people with chronic pain that is secondary to other conditions. These include hemophilia, in which bleeding into joints causes arthropathy and chronic joint pain (Riley et al., 2011; Witkop et al., 2011).

Studies comparing the relative influence of coping and acceptance on physical and psychological outcomes can inform the application and development of interventions. Several cross-sectional studies showed that acceptance was a better predictor than coping of functioning, disability, distress, emotional adjustment and quality of life (Elander et al., 2009; McCracken and Eccleston, 2006; Rodero et al., 2011). Another showed that acceptance influenced functional status whereas coping influenced emotional distress (Esteve et al., 2007).

In a longitudinal study, baseline ‘control-oriented’ coping predicted worse functioning at follow-up, and baseline ‘acceptance-oriented’ coping predicted better functioning, but neither was a significant predictor when the baseline measure of functioning was included as a predictor. However, *change* in control-oriented coping from baseline to follow-up added significantly to the prediction of certain aspects of functioning (increases in control-oriented coping predicted poorer functioning), whereas change in acceptance-oriented coping did not add to the prediction of any functioning measures (McCracken et al., 2007b).

Evidence about baseline measures that predict improved outcomes is informative for matching individuals to interventions, whereas evidence about changes that predict improved outcomes is informative about the processes that interventions should target. In this longitudinal study, we examined both baseline and change measures of pain intensity, coping and acceptance as predictors of quality of life at follow-up among men with hemophilia during a low-intensity self-management program. The aims were to identify characteristics that could make people with hemophilia more or less likely to benefit from initiatives to improve quality of life, and to identify change processes associated with improvements in quality of life that could be targeted in initiatives designed specifically for people with hemophilia.

2. Methods

2.1. Participants and recruitment

The participants were 101 men with hemophilia in a low intensity psycho-educational program to promote readiness to self-manage chronic joint pain. Participant details are given in table 1. Participant recruitment was through the membership and registration list of the Haemophilia Society UK. Inclusion criteria were diagnosed hemophilia, Society membership or registration with good mailing status, and age over 18 years. Exclusion criteria were medical conditions that complicate pain self-management (such as Alzheimer's disease) or previously indicated unwillingness to participate in research. All the participants were male.

All information about participant identity and contact details was retained by the Haemophilia Society, which undertook the mailings to participants. At baseline, participants were sent an invitation, questionnaire booklet and return envelope. A reminder was sent to those who did not return a completed questionnaire within four weeks. Six months later, participants were sent a follow-up questionnaire booklet and return envelope.

Table 1: Participant details

N	101
Mean age (SD, range)	50.3 (12.2, 26-84)
<i>Type of disorder</i>	
Hemophilia A	78 (77.2%)
Hemophilia B	20 (19.8%)
Not known	3 (3.0%)
<i>Hemophilia severity</i>	
Mild	19 (18.8%)
Moderate	11 (10.9%)
Severe	71 (70.3%)
<i>Marital status</i>	
Single	12 (11.9%)
Married/cohabiting	75 (74.3%)
Divorced/separated	10 (9.9%)
Other	4 (4.0%)
<i>Educational level</i>	
High school only ¹	38 (37.6%)
Post-16 education ²	35 (34.7%)
Higher education ³	24 (23.8%)
Other/not known	4 (4.0%)

1. Left school at 16 years

2. High school to 18 years or Further Education

3. University undergraduate or postgraduate study

2.2. The psycho-educational program

Following informed consent and baseline data collection, all participants received an information booklet and a randomly selected half also received a DVD. The booklet and DVD were designed to increase knowledge, awareness and motivation to self-manage chronic joint pain, and were informed by the self-management model of chronic pain (Jensen et al., 2003). They did not provide instruction in any specific self-management skills or techniques. The booklet included information about types of pain in hemophilia; the impact of pain on emotions

and other aspects of life; positive and negative ways of using pain medications; and the benefits of active coping and exercise. The DVD was a 25-minute film featuring personal stories told by people with hemophilia about their experiences of living with joint pain, reinforced by information delivered by health professionals. More information about the booklet and DVD are given by Elander et al. (2011).

2.3. Measures

Age, hemophilia type and hemophilia severity were recorded at baseline. Pain intensity in the last month was rated at baseline and 6-month follow-up on a 10cm visual analogue scale labeled 'no pain' to 'worst pain possible'. Pain coping, acceptance and health-related quality of life were measured at baseline and 6-month follow-up.

2.3.1. Pain coping

The Hemophilia Pain Coping Questionnaire (HPCQ) is a 27-item condition-specific adaptation of more generic pain coping measures, and gives scores for three scales based on factor analysis (Elander and Robinson, 2008). 'Active coping' comprises 10 items about diverting attention from pain, ignoring pain sensations, reinterpreting pain sensations, increasing behavioral activities when in pain, and coping self-statements. 'Negative thoughts' comprises 9 items about catastrophizing, anger, fear, and seeking isolation when in pain. 'Passive adherence' comprises 6 items about resting, using pain medications, and using ice when in pain. Internal reliability (active coping 0.80; negative thoughts 0.86; passive adherence 0.76) and test-retest reliability (active coping 0.70; negative thoughts 0.73; passive adherence 0.64) were good, and validity was demonstrated by differential relationships with other responses to pain (Elander and Robinson, 2008).

2.3.2. Pain acceptance

The Chronic Pain Acceptance Questionnaire (CPAQ) is a 20-item measure of how well individuals can desist from attempts to avoid or reduce their chronic pain (McCracken et al., 2004). The 'activity engagement' subscale comprises 11 items about engaging in activities when in pain, and the 'pain willingness' subscale comprises 9 items about recognizing that avoidance and control are often unworkable methods of adapting to chronic pain. A pain acceptance total score is obtained by adding one subscale score to the other. A recent review concluded that there was more positive evidence about the reliability and validity of the CPAQ than for any comparable measure of pain acceptance (Reneman et al., 2010).

2.3.3. Health-related quality of life

The RAND-36 (SF-36) is a measure of health-related quality of life that gives scores for 8 subscales and two summary scales, one for physical and one for mental health (Hays and Morales, 2001). The summary scales are computed as weighted aggregates of standardized subscale scores, and are then transformed to T scores (Ware et al., 1994). These have good psychometrics and the measure is frequently used in hemophilia research (Fischer et al., 2003), and was recommended for use with people with chronic pain (Angst et al., 2008).

2.4. Data analysis

SPSS 18.0 for Windows was used for the statistical analysis. In a small number of cases, mean values were substituted for missing values of questionnaire items where more than half the items in a scale were present. Residualized change scores were computed by regressing baseline scores onto follow-up scores (with baseline scores as predictors and follow-up scores as dependent variables), to produce change measures uncorrelated with baseline scores. Higher change scores indicate increases from baseline to follow-up.

The relative influence of baseline and change measures of pain coping and acceptance on quality of life was tested in multiple linear regression analyses with follow-up physical and mental health summary scores as the dependent variables. Predictor variables were added to the regression model in successive blocks: first age, hemophilia severity and DVD group, using the 'enter' method to control for these variables in all subsequent models; then baseline quality of life; then baseline measures of pain intensity, coping and acceptance; then change measures of pain intensity, coping and acceptance. After the first block, predictor variables were added and removed using the stepwise method, with criteria for entry and removal of $p < 0.05$ and $p > 0.10$ respectively.

When both baseline and change measures were independently predictive, we added interaction terms in a final block. Interaction terms were computed as standardized baseline score times standardized residualized change score. We also ran alternative analyses with separate subscale scores for activity engagement and pain willingness instead of the pain acceptance total score.

We assessed independence of errors with the Durbin-Watson statistic, which should be close to 2, with values less than 1 and greater than 3 indicating potential non-independence (Field, 2009, p. 221). We assessed the normality of residuals by examining the histograms of standardized residuals. We assessed non-linearity and homoscedasticity by examining the scatter plots of standardized residuals against standardized predicted values. Scatter plots with curvature are indicative of non-linearity, and those with funnelling, where the range of values of standardized residuals increases across the range of predicted values, are indicative of heteroscedasticity (Field, 2009, p. 247-248; Tabachnick and Fidell, 1996, p. 136). We also examined tolerances and variance inflation factors (VIFs) for all predictor variables, including those excluded from final models. Multicollinearity is indicated by tolerances below 0.2 and VIFs greater than 10.0 (Field, 2009, pp. 224, 242).

When regression coefficients for a predictor variable were reduced by more than 10% by including another predictor, we tested indirect effects with Sobel tests (Sobel, 1982), using the SPSS macro provided by Preacher and Hayes (2004). This tests the mediation of one variable by another, which occurs when the mediating variable "accounts for the relationship between the predictor and the criterion" (Baron & Kenny, 1986, p. 1176). This would occur when a predictor variable is confounded with a 'mediating' variable that accounts for the relationship between the predictor and the criterion (dependent) variable.

3. Results

Of 209 individuals who completed baseline assessment, 144 returned follow-up questionnaires and 101 had complete baseline and follow-up data for all study measures. Compared with the remainder of the initial sample of 209, the study sample had lower baseline

physical quality of life (mean 30.31, SD 11.17, compared with 34.22, SD 13.82, $t = 2.17$, $p = 0.032$), but did not differ in age, hemophilia type or severity, or any other baseline measures. Compared with the remainder of those who returned follow-up questionnaires but were not included in the analyses, the study sample did not differ on any of the baseline measures.

Table 2 shows descriptive statistics for study variables at baseline and follow-up, together with tests of changes from baseline to follow-up, and correlations between baseline and follow-up measures. There were significant increases over time in active coping, negative thoughts, pain acceptance, and physical quality of life, and highly significant positive correlations over time for all the measures.

Table 2. Descriptive statistics for study measures

	Items	Baseline		Follow-up		Difference (follow-up minus baseline)		Baseline/follow-up correlation
		Mean (SD)	Alpha	Mean (SD)	Alpha	Mean (SD)	T	
Pain intensity	1	5.31 (2.90)	-	5.15 (2.62)	-	-0.16 (2.03)	0.78	0.73**
Active coping	10	2.36 (1.11)	0.81	2.73 (1.16)	0.85	0.37 (0.82)	4.58**	0.74**
Negative thoughts	9	1.84 (1.36)	0.87	2.07 (1.28)	0.87	0.23 (1.01)	2.29*	0.71**
Passive adherence	6	3.32 (1.44)	0.79	3.13 (1.31)	0.76	-0.19 (1.14)	1.67	0.66**
Pain acceptance total	20	65.82 (15.95)	0.83	68.15 (16.73)	0.86	2.33 (10.12)	2.31*	0.81**
Activity engagement	11	40.50 (10.18)	0.78	41.17 (10.53)	0.84	0.67 (7.76)	0.87	0.72**
Pain willingness	9	25.32 (9.21)	0.80	26.98 (9.71)	0.83	1.66 (6.99)	2.38*	0.73**
Physical quality of life	8	30.31 (11.17)	0.86	31.68 (11.39)	0.86	1.38 (6.37)	2.17*	0.84**
Mental quality of life	8	47.09 (11.60)	0.86	47.28 (11.27)	0.86	0.19 (9.85)	0.20	0.63**

* $p < .05$; ** $p < .001$

Table 3 shows correlations among the measures. A number of predictor variables were correlated with quality of life. Age, hemophilia severity and baseline pain intensity were all negatively correlated with both baseline and follow-up physical quality of life. Baseline pain acceptance, activity engagement and pain willingness were all positively correlated with both baseline and follow-up physical quality of life. Change in pain intensity was negatively correlated with follow-up but not baseline physical quality of life. Baseline passive adherence was negatively correlated with baseline but not follow-up physical quality of life. Change in passive adherence was negatively correlated with follow-up but not baseline physical quality of life.

Baseline negative thoughts was negatively correlated with both baseline and follow-up mental quality of life. Baseline pain acceptance, activity engagement and pain willingness were positively correlated with both baseline and follow-up mental quality of life. Change in activity engagement was positively correlated with follow-up but not baseline mental quality of life.

There were numerous correlations among baseline measures of predictor variables, the largest of which, apart from between pain acceptance total and subscale scores, was between negative thoughts and pain acceptance total (-0.57). Age was correlated negatively with severity and pain willingness. Severity was correlated positively with pain intensity. Pain intensity was correlated negatively with pain acceptance, activity engagement and pain willingness. Active coping was correlated positively with pain acceptance and activity engagement. Negative thoughts was correlated positively with passive adherence and negatively with pain acceptance, activity engagement and pain willingness. Passive adherence was correlated negatively with

Table 3. Correlations among study measures

1. Age	1.00									
2. Severity	-0.45**	1.00								
3. Baseline pain intensity	-0.11	0.40**	1.00							
4. Change in pain intensity	0.08	-0.07	0.00	1.00						
5. Baseline active coping	-0.07	-0.06	-0.10	0.10	1.00					
6. Change in active coping	-0.08	0.07	-0.07	-0.03	0.00	1.00				
7. Baseline negative thoughts	-0.08	-0.10	0.15	0.11	0.05	-0.05	1.00			
8. Change in negative thoughts	0.07	0.05	0.09	0.22*	-0.13	0.28*	0.00	1.00		
9. Baseline passive adherence	0.00	-0.05	0.14	0.04	-0.19	0.02	0.37**	0.13	1.00	
10. Change in passive adherence	0.01	-0.06	0.04	0.14	-0.05	0.04	0.06	0.24*	0.00	1.00
11. Baseline pain acceptance total	-0.16	0.04	-0.35**	-0.09	0.26*	0.05	-0.57**	-0.14	-0.34*	-0.21*
12. Change in pain acceptance total	0.02	-0.04	-0.02	-0.25*	0.14	0.06	0.13	-0.33*	-0.13	-0.18
13. Baseline activity engagement	-0.05	-0.01	-0.28*	0.02	0.39**	0.17	-0.39**	-0.04	-0.29*	-0.14
14. Change in activity engagement	0.01	-0.05	0.01	-0.19	0.08	0.08	0.07	-0.22*	-0.12	-0.13
15. Baseline pain willingness	-0.21*	0.08	-0.29*	-0.17	0.01	-0.11	-0.56**	-0.20*	-0.26*	-0.21*
16. Change in pain willingness	-0.02	-0.00	-0.12	-0.18	0.18	0.02	-0.01	-0.28*	-0.13	-0.17
17. Baseline physical quality of life	-0.22*	-0.32**	-0.56**	-0.13	0.10	-0.11	-0.09	-0.17	-0.20*	-0.17
18. Follow-up physical quality of life	-0.26*	-0.25*	-0.47**	-0.24*	0.04	-0.04	-0.12	-0.11	-0.13	-0.24*
19. Baseline mental quality of life	-0.11	0.07	-0.09	-0.13	0.00	-0.06	-0.50**	-0.04	-0.04	0.02
20. Follow-up mental quality of life	-0.09	0.11	0.03	-0.12	0.11	-0.02	-0.41**	-0.25*	-0.25*	0.00
	1	2	3	4	5	6	7	8	9	10

* $p \leq .05$; ** $p \leq .001$

Table 3 continued

1. Age									
2. Severity									
3. Baseline pain intensity									
4. Change in pain intensity									
5. Baseline active coping									
6. Change in active coping									
7. Baseline negative thoughts									
8. Change in negative thoughts									
9. Baseline passive adherence									
10. Change in passive adherence									
11. Baseline pain acceptance total	1.00								
12. Change in pain acceptance total	0.00	1.00							
13. Baseline activity engagement	0.84**	0.08	1.00						
14. Change in activity engagement	0.02	0.75**	0.00	1.00					
15. Baseline pain willingness	0.80**	-0.09	0.35**	0.03	1.00				
16. Change in pain willingness	0.20*	0.65**	0.32*	0.02	0.00	1.00			
17. Baseline physical quality of life	0.35**	0.09	0.25*	0.10	0.34**	0.11	1.00		
18. Follow-up physical quality of life	0.33*	0.08	0.25*	0.11	0.30*	0.07	0.84**	1.00	
19. Baseline mental quality of life	0.48**	-0.11	0.32*	0.02	0.47**	-0.08	-0.06	0.07	1.00
20. Follow-up mental quality of life	0.38**	0.18	0.27*	0.24*	0.37**	0.08	-0.04	-0.07	0.63**
	11	12	13	14	15	16	17	18	19

* $p \leq .05$; ** $p \leq .001$

pain acceptance, activity engagement and pain willingness. Activity engagement was correlated positively with pain willingness.

There were fewer correlations among change measures, but the largest was again between negative thoughts and acceptance total (-0.33). Change in pain intensity was correlated positively with change in negative thoughts and negatively with change in pain acceptance. Change in active coping was correlated positively with change in negative thoughts. Change in negative thoughts was correlated positively with change in passive adherence and negatively with changes in pain acceptance, activity engagement and pain willingness.

There were even fewer correlations between baseline and change measures. Baseline pain acceptance and activity engagement were correlated positively with change in pain willingness. Baseline pain willingness was correlated negatively with changes in negative thoughts and passive adherence.

The results of the regression analyses are given in table 4. Age and hemophilia severity were significant negative predictors of physical quality of life, but not when baseline quality of life was included in the model. Change in pain intensity was the only other significant predictor, such that reduced pain intensity predicted greater physical quality of life.

The F ratio for the final model was 52.35 (df 5, 95; $p < 0.001$), indicating a good fit, and the Durbin-Watson statistic was 1.90, indicating independence of errors. The residuals were normally distributed and the scatter plots showed no curvature or funnelling. The lowest tolerance was 0.53 and the highest VIF was 1.66, indicating no multicollinearity.

Baseline physical quality of life reduced the coefficients for age and severity to almost zero, so we tested indirect mediation effects. These showed that baseline physical quality of life mediated the effects of age (Sobel = -0.17, 95% CIs -0.32 to -0.02, $p = 0.03$) and severity (Sobel = -3.82, 95% CIs -6.15 to -1.50, $p = 0.001$).

We repeated the regression analysis with activity engagement and pain willingness subscales in place of pain acceptance. This produced exactly the same results. Neither activity engagement nor pain willingness was predictive, and the only significant predictors were baseline physical quality of life and change in pain intensity.

The results of the analysis with follow-up mental quality of life as the dependent variable are also given in table 4. Age and severity were not predictive in any model. DVD group was predictive on entry, but not when other predictors were added. Baseline quality of life was again the strongest predictor, but baseline passive adherence and change in pain acceptance were also significantly predictive in the final model. Lower baseline passive adherence and greater increases in pain acceptance predicted greater quality of life. We repeated the analysis with the baseline passive adherence x change in pain acceptance interaction as an additional predictor, but it was not significant (final model $\beta = 0.09$, $p = 0.21$).

The F ratio for the final model was 16.62 (df 6, 94; $p < 0.001$) and the Durbin-Watson statistic was 1.90. The residuals were normally distributed and the scatter plots showed no curvature or funnelling. The lowest tolerance was 0.58 and the highest VIF was 1.73.

Because DVD group was a significant predictor in block 1 but not subsequently, we tested indirect mediation effects. Neither baseline quality of life, nor baseline passive adherence, nor change in pain acceptance accounted individually for the DVD effect, although DVD group was not significantly predictive independently of those factors.

With separate activity engagement and pain willingness subscales in place of pain acceptance, baseline quality of life (final model $\beta = 0.61$, $p < 0.001$) and passive adherence (final model $\beta = -0.16$, $p = 0.030$) were again significant predictors. Change in activity engagement was also a significant predictor (final model $\beta = 0.17$, $p = 0.024$), and so also was change in negative thoughts (final model $\beta = -0.150$, $p = 0.048$). Increased activity engagement and reduced negative thoughts predicted improved mental quality of life. We repeated the analysis with the interaction terms for baseline passive adherence x change in activity engagement, and baseline passive adherence x change in negative thoughts included as predictors, but neither was significant (final model $\beta = 0.09$, $p = 0.25$; $\beta = 0.05$, $p = 0.50$, respectively).

The F ratio for the final model was 14.80 (df 7, 93; $p < 0.001$), and the Durbin-Watson statistic was 1.88. The residuals were normally distributed and the scatter plots showed no curvature or funnelling. The lowest tolerance was 0.58, and the highest VIF was 1.74.

Table 4. Standardized regression coefficients (β), changes in R^2 (ΔR^2), total and adjusted R^2 from hierarchical multiple regression analyses predicting follow-up physical and mental quality of life^a

Block and predictor	Entry β	Final β	ΔR^2	Total R^2	Adj. total R^2
<i>Follow-up physical quality of life</i>					
1. Age	-0.46**	-0.10			
Severity	-0.46**	-0.06			
DVD group	-0.04	-0.06	0.23**		
2. Baseline quality of life	0.81**	0.78**	0.48**		
3. N/A ^b	-	-	-		
4. Change in pain intensity	-0.14*	-0.14*	0.02*	0.73	0.72
<i>Follow-up mental quality of life</i>					
1. Age	-0.04	0.02			
Severity	0.10	0.07			
DVD group	0.21*	0.14	0.06		
2. Baseline quality of life	0.62**	0.64**	0.38**		
3. Baseline passive adherence	-0.19*	-0.17*	0.04*		
4. Change in pain acceptance	0.22*	0.22*	0.05*	0.52	0.48

Notes:

a) In each analysis, age, hemophilia severity and DVD group were entered simultaneously in block 1 and retained in all subsequent models. In the next blocks, the baseline measure of the dependent variable, then baseline measures of predictor variables, then change measures of predictor variables were added, using the stepwise method (probability of F to enter < 0.05 ; probability of F to remove > 0.10).

b) No baseline measures of predictor variables were retained in the block 3 model.

* $p < .05$; ** $p < .001$

4. Discussion

Reduced pain intensity predicted better physical quality of life, independently of other factors. Lower baseline passive adherence and increased pain acceptance predicted better mental quality of life, independently of other factors. Activity engagement rather than pain willingness was the predictive component of pain acceptance, and reduced negative thoughts also predicted better mental quality of life when separate pain acceptance subscales were used. Active pain coping did not predict either physical or mental quality of life.

Previous studies reported that indices of morbidity like arthropathy and joint complications were the main predictors of physical quality of life in hemophilia (Fischer et al., 2005; Solovieva, 2001), but in our data the influence of hemophilia severity (and age) was mostly explained by baseline quality of life. This does not mean that severity and age are not important influences on quality of life, just that they have static effects that contribute to continuity rather than change.

Previous studies of people with chronic pain conditions also identified pain intensity as a key influence on physical functioning (Viane et al., 2003; Esteve et al., 2007; Vowles et al., 2007). In one study, changes in acceptance and/or catastrophizing (a construct similar to negative thoughts) also influenced disability independently of pain (Vowles et al., 2007), but others found no psychological influences on physical functioning, including one that used the same quality of life measure as the present study (Viane et al., 2003).

Physical quality of life therefore tracks pain intensity quite closely, suggesting that efforts to improve functioning and quality of life should focus mainly on reducing pain. That is not easy, of course, especially in chronic pain conditions where pain is already a primary focus of treatment, but in hemophilia there is greater scope for improving pain management (Riley et al., 2011), which should also improve physical quality of life.

The key predictors of mental quality of life were also consistent with research on people with chronic pain conditions, which found that acceptance influenced mental quality of life (Viane et al., 2003) and depression and anxiety (Vowles et al., 2007). Our findings suggest that improving pain acceptance could also be a focus for initiatives to improve mental quality of life for people with chronic joint pain related to hemophilia.

When pain acceptance was separated into two components, activity engagement rather than pain willingness was predictive of mental quality of life. That is consistent with most other evidence about the relative predictive value of CPAQ subscales (with some exceptions, e.g. McCracken et al., 2004), leading some authors to propose that the pain willingness subscale is not robust and should be discarded (Nicholas and Asghari, 2006).

This might suggest that activity engagement should be the main target for efforts to improve mental wellbeing for people with chronic pain, including hemophilia-related joint pain. However, it is not really clear what an activity engagement intervention would look like, and in fact most 'acceptance-based' interventions have a focus that is broader than the attitudes and behaviors represented by CPAQ subscales. For example, one acceptance-based treatment included physical exercises, awareness raising exercises, exposure exercises, habit reversal training, mindfulness meditation exercises, relaxation exercises, and sensation focusing exercises (McCracken et al., 2005).

People with hemophilia may not distinguish acute from chronic joint pain (Witkop et al., 2011), but acute joint pain signals bleeding and should be promptly treated with clotting factors.

Acceptance-based interventions designed for people with chronic pain conditions should therefore be adapted very carefully to ensure they are safe and appropriate for people with hemophilia.

People with hemophilia are also quite varied and might be matched to different types of exercise or program. Baseline passive pain coping (relying on resting and taking pain medications) was a negative predictor of mental quality of life, so people with less initial passive coping might be selected for interventions on the basis that they have better prospects of improvement. Alternatively, different interventions might be developed for individuals with less and more passive coping, or phased programs could be developed that targeted first passive coping, then pain acceptance.

Active coping was not a significant predictor in any of the analyses, so perhaps attempts to improve active pain coping are not justified, at least in hemophilia. The relative importance of coping and acceptance probably varies between conditions, and in hemophilia there may be less scope for ignoring, re-interpreting or diverting attention from pain, for acute joint pain is a sign of current bleeding that should be promptly treated. Given the effects of passive coping described above, it is also possible that reducing passive coping is more important than increasing active coping, at least in hemophilia.

Change in negative thoughts predicted mental quality of life independently of changes in activity engagement and pain willingness, but not independently of change in pain acceptance total score. This aspect of the findings is difficult to interpret, and highlights the need for further research on the meanings of CPAQ scores, to address questions like whether the processes involved in pain acceptance and negative thinking should be targeted separately or in combination. Rather speculatively, our data might suggest that activity engagement is a more behavioral construct and pain willingness is more motivational or emotional. For example, activity engagement but not pain willingness was significantly correlated with active coping, whereas pain willingness was more strongly correlated with negative thoughts.

The study has a number of potential limitations. First, the study sample was a subset of a larger group. As previously reported, the initial sample of 209 represented only 37% of 568 invited to take part, but compared with eligible non-participants, differed only in containing more individuals with severe hemophilia (Elander et al., 2009). Compared with the national UK population of adults with hemophilia, the initial sample was representative in terms of proportions with hemophilia A and B, but contained higher proportions of people aged over 40 and people with severe hemophilia (Elander et al., 2009). Hemophilia-related joint pain affects older people and those with severe hemophilia more than others, so the study sample is more representative of people with hemophilia who are most affected by chronic joint pain, who would be the target population for applications of the findings.

Second, the program did not constitute a full treatment intervention. Participants all received written materials, and half received a DVD that did not differentially improve quality of life. There was no personal contact or interaction, so it was a very low intensity intervention that in some ways resembled a public health promotion program. Active coping, pain acceptance and physical quality of life all increased over time, so the program did have some benefits, but pain intensity and mental quality of life did not, so for those variables, change scores represent individual rather than group changes.

For some variables, therefore, it could be argued that change scores might just reflect unreliability or lack of temporal consistency. However, the high correlations between baseline and follow-up measures (table 2) showed that there was considerable temporal consistency among the sample as a whole, and the correlations among change scores (table 3) appear to reflect systematic, meaningful patterns of change. Most importantly, the regression analyses showed that changes in pain intensity and pain acceptance were more predictive of quality of life than would be expected by chance variation.

Third, six months is a relatively long follow-up period, and fluctuations within that period would not be captured in our measures. We chose a six month follow-up because the materials were designed for participants to use at their own pace, and aimed to prompt participants to reflect on, monitor and review their responses to pain over an extended period.

Fourth, the analyses involved concurrent changes in predictor and outcome variables, and a more rigorous approach would be to use changes during one period as predictors of changes during a subsequent period. However, this would require multiple follow-up points, which would increase participant attrition and missing data. There was already considerable attrition from baseline to follow-up, although those followed-up and not followed-up did not differ on any of the predictor variables.

Fifth, we tested the effects of only a few interaction terms, and other combinations of baseline and change measures could potentially have interacting influences. However, testing each possible interaction would have involved at least 25 (5 x 5) interaction terms in each of the two analyses, even before considering separate acceptance subscales. Without any clear basis for predictions, that would have amounted to a trawl for interaction effects.

In conclusion, the findings suggest that influences on quality of life among those affected by haemophilia-related chronic joint pain are similar to those with chronic pain conditions, so there should be scope to adapt the content of existing acceptance-based interventions, provided that care is taken not to compromise the prompt treatment of acute bleeding episodes. However, more research is needed on conditions where pain is secondary to other features, in order to understand better the interplay between factors that influence outcomes in similar ways across conditions, and those that affect outcomes in ways specific to each condition. More research is also needed on the meanings of specific elements or aspects of pain acceptance, and this may become more important as acceptance-based interventions are developed for different conditions. Greater initial passive coping may be an obstacle to improving quality of life, so there may be scope for selecting people for interventions based on their degree of passive coping, or for developing phased interventions that aim first to reduce passive coping and then to promote pain acceptance and/or reduce negative thoughts.

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