



Using the common-sense model to understand health outcomes for medically unexplained symptoms: a meta-analysis

Lisa M. McAndrew^{a,b}, Marcus Crede^c, Kieran Maestro^b, Sarah Slotkin^b, Justin Kimber^b and L. Alison Phillips^c

^aWar Related Illness and Injury Study Center, Veterans Affairs New Jersey Healthcare System, East Orange, NJ, USA; ^bDepartment of Educational and Counseling Psychology, University at Albany, Albany, NY, USA; ^cDepartment of Psychology, Iowa State University, Ames, IA, USA

ABSTRACT

Consistent with the common-sense model of self-regulation, illness representations are considered the key to improving health outcomes for medically unexplained symptoms and illnesses (MUS). Which illness representations are related to outcomes and how they are related is not well understood. In response, we conducted a meta-analysis of the relationship between illness representations, self-management/coping, and health outcomes (perceived disease state, psychological distress, and quality of life) for patients with MUS. We reviewed 23 studies and found that threat-related illness representations and emotional representations were related to worse health outcomes and more negative coping (moderate to large effect). Generally, increases in negative coping mediated (with a moderate to large effect) the relationship of threat/emotional illness representations and health outcomes. Protective illness representations were related to better health outcomes, less use of negative coping and greater use of positive coping (small to moderate effect). The relationship of protective illness representations to better health outcomes was mediated by decreases in negative coping (moderate to large effect) and increases in positive coping (moderate effect). Perceiving a psychological cause to the MUS was related to more negative health outcomes (moderate to large effect) and more negative emotional coping (small effect). The relationship of perceiving a psychological cause and more negative health outcomes was mediated by increases in negative emotional coping. Improving our understanding of how illness representations impact health outcomes can inform efforts to improve treatments for MUS. Our results suggest behavioural treatments should focus on reducing threat-related illness representations and negative coping.


ARTICLE HISTORY


Received 6 February 2018
Accepted 4 September 2018

KEYWORDS

Illness representations; common-sense model; medically unexplained symptoms; chronic fatigue; chronic pain; illness beliefs; physical symptoms

Medically unexplained symptoms and syndromes (MUS), or persistent physical symptoms, cause more disability than other chronic conditions (McAndrew, Chandler, et al., 2016; McAndrew, Helmer, et al., 2016), and are considered by primary care provider among the most difficult medical conditions to treat (Hahn, Thompson, Wills, Stern, & Budner, 1994; Hinchey & Jackson, 2011; Jackson & Kroenke, 1999; Steinmetz & Tabenkin, 2001). MUS is an umbrella term that refers to conditions defined by multiple chronic physical symptoms that cause functional impairment. Growing research suggests that the actual cause of MUS is multi-deterministic (Burton, 2003), leading to calls to change the name of MUS although none of the proposed terms have yet to become widely accepted.

CONTACT Lisa M. McAndrew  lisa.mcandrew@va.gov

 Supplemental data for this article can be accessed here <https://doi.org/10.1080/17437199.2018.1521730>.

© 2018 Informa UK Limited, trading as Taylor & Francis Group

Illness representations are considered key to understanding health outcomes for MUS (Moss-Morris, 2011). Illness representations are the patient's understanding of their MUS, including the identity (e.g. I have fibromyalgia), timeline (e.g. I will have it the rest of my life), cause (e.g. exposure to vaccines caused my symptoms), consequences (e.g. I cannot work), control (e.g. I need to rest) and coherence (e.g. my fibromyalgia is confusing) (Leventhal, Leventhal, & Breland, 2011). Illness representations also include the patient's emotional beliefs about the health condition (e.g. my fibromyalgia is upsetting to me).

Illness representations account for 30–40% of the variance in MUS health outcomes, such as quality of life (Hunt, Richardson, Engel, Atkins, & McFall, 2004; Moss-Morris, 2011). Negative illness representations are prospectively related to greater healthcare costs (Frostholm, Petrie, Ørnbøl, & Fink, 2014), higher utilisation (Frostholm et al., 2010), declining health (Sharpe et al., 2010), poorer patient provider satisfaction (Frostholm et al., 2010) and poorer mental and physical health function (Frostholm et al., 2007) among patients with MUS. They also predict who develops chronic MUS from acute health episodes [e.g. stomach virus, concussion (Hou et al., 2012; Moss-Morris, Spence, & Hou, 2011; Spence, Moss-Morris, & Chalder, 2005; Spence & Moss-Morris, 2007; Whittaker, Kemp, & House, 2007)]. Illness representations are believed to be stronger predictors of outcomes for patients with MUS as compared to patients with other better understood chronic conditions [e.g. heart disease (Moss-Morris & Wrapson, 2003)].

Behavioural interventions for MUS, including cognitive behavioural therapy and reattribution treatment, seek to change patient's illness representations to improve health outcomes. While these treatments improve outcomes for some patients, their efficacy is lower than behavioural treatments for other illnesses, such as anxiety and depression, suggesting room for improvement (Gask, Dowrick, Salmon, Peters, & Morriss, 2011; Hofmann, Asnaani, Vonk, Sawyer, & Fang, 2012). A better understanding of *which* illness representations best predict health outcomes, and *how* they predict health outcomes, is needed to improve treatments for MUS.

Which illness representations impact health outcomes for MUS?

It is not known which illness representations best predict health outcomes for patients with MUS. For patients with any chronic condition, *threat-related* illness representations (perceiving more severe consequences, perceiving more symptoms to be attributed to the condition, and perceiving a longer timeline) and emotional illness representations are related to worse outcomes (Hagger, Koch, Chatzisarantis, & Orbell, 2017). Moss-Morris (2005) has suggested that threat (or negative) illness representations are also related to worse outcomes among patients with chronic fatigue syndrome (a type of MUS) with perceiving serious consequences and perceiving more symptoms to be attributed to the condition (identity) to be the strongest predictors of negative outcomes. Because there has never been a meta-analysis, the effect size of threat-related MUS illness representations to health outcomes is unknown, as is the differential impact of threat-related illness representations as compared to other types of illness representations (such as protective illness representations).

Protective illness representations may be related to health outcomes for patients with MUS, but there has been little research on this topic. Generally, among patients with chronic conditions, greater perceived control over the health condition and perceiving coherence (i.e. the chronic condition is understandable) is associated with positive health outcomes. Perceiving greater control may not be protective for patients with MUS, as there is no cure for MUS (Hagger et al., 2017). Counter to this argument, while there is no cure, patients can learn management strategies to improve their quality of life, suggesting that greater perceived control could be related to better outcomes. There is mixed empirical evidence of a relationship between potentially protective illness representations and health outcomes for patients with MUS (Hou et al., 2012; Moss-Morris et al., 2011; van Wilgen, van Ittersum, Kaptein, & van Wijhe, 2008). One reason for the mixed evidence is that individual studies do not always have enough subjects to detect small to moderate effects. For example, van Wilgen et al. (2008) surveyed 51 patients with fibromyalgia

and found a correlation of .20 and .22 between control beliefs and outcomes, but these relationships did not reach statistical significance.

Finally, *causal* illness representations are thought to be important predictors of health outcomes. There is a long-standing belief that perceiving the cause of MUS to be physical may lead patients to pressure providers to provide medical intervention (e.g. surgery, medication) which is ineffective at improving MUS and leads to worse outcomes. Perceiving the cause to be psychological has been considered beneficial as it leads patients to focus on reducing stress and improving self-management (Brown, 2004; Czachowski, Piszczek, Sowińska, & olde Hartman, 2011; Deary, Chalder, & Sharpe, 2007; Kirmayer, Young, & Robbins, 1994; Moss-Morris, 2005; Rief & Broadbent, 2007). Recent evidence, however, questions this assumption. Research finds that patients do not regularly pressure providers for medical intervention (Salmon, Ring, Dowrick, & Humphris, 2005); therefore, it is unknown if, by extension, causal beliefs are not especially connected to health outcomes. Moss-Morris (2005) suggest that for patients with CFS, the specific causal belief may be less important than the strength of the belief that the MUS are generally a sign of physical illness. They also suggest that external attributions of MUS to illness may be associated with worse disability but less psychological distress. To date, however, no study has systematically reviewed the literature to develop an empirically based understanding of the relationship of causal beliefs to health outcomes for patients with MUS. This is important because changing causal beliefs is a focus of behavioural interventions and, as yet, the relationship of causal beliefs to health outcomes for patients with MUS is 'less clear than expected' (Rief & Broadbent, 2007).

How do illness representations impact health outcomes for MUS?

How illness representations influence MUS-related health outcomes is also poorly understood. According to Leventhal's common-sense model of self-regulation (CS-SRM), illness representations are used to guide self-management and coping such as seeking medical care, exercising, or focusing on acceptance (Brandes & Mullan, 2014; Hagger & Orbell, 2003; Hale, Treharne, & Kitas, 2007; McAndrew et al., 2017; McAndrew et al., 2008). Few studies, however, have examined if self-management/coping mediates the relationship between MUS illness representations and MUS health outcomes.

A recent meta-analysis on the relationships between patients' explicit illness beliefs across all chronic health conditions found a direct impact of illness representations on health outcomes and an indirect impact through coping (Hagger et al., 2017). This, and meta-analyses of specific chronic conditions (e.g. heart disease), find that threat-related illness representations (more severe consequence, greater number of symptoms, longer timeline) and emotional representations are related to greater use of negative coping strategies and poorer health outcomes. Protective representations, including greater perceived control and coherence (feeling their illness makes sense) are related to more positive, self-management/coping and better health outcomes (Brandes & Mullan, 2014; Broadbent et al., 2015; Dempster, Howell, & McCorry, 2015; Foxwell, Morley, & Frizelle, 2013; French, Cooper, & Weinman, 2006; Hagger et al., 2017; Hagger & Orbell, 2003; Hudson, Bundy, Coventry, & Dickens, 2014; Mc Sharry, Moss-Morris, & Kendrick, 2011). It is not known if the relationships between illness representations and self-management and health outcomes for patients with MUS are similar to those found in other health conditions.

Understanding how illness representations, self-management/coping and health outcomes are related specifically for patients with MUS is important, because these relationships may be different for patients with MUS as compared to better understood chronic conditions. MUS are one of the only conditions where illness representations are part of the conceptualisation of the etiology of the condition. In cognitive behavioural conceptualizations of MUS, illness representations are believed to be a bridge between susceptibility to symptoms (e.g. increase genetic risk) to long-term outcomes. Specifically, an over focus on the threat of the symptom and over attribution of the symptom as a sign of physical illness are conceptualised to increase attention to symptoms, maintain heightened arousal and lead to poor coping with the symptoms. Overtime, this response is thought to contribute to a chronic presentation of MUS (Rief & Broadbent, 2007).

Current study

To address the gaps in our understanding of *which* illness representations predict health outcomes and *how*, we conducted a meta-analysis of the relationships between illness representations, self-management/coping and health outcomes for patients with MUS. We hypothesised that illness representations have a direct impact on health outcomes and an indirect impact through self-management/coping. We further hypothesised that threat-related illness representations (greater consequences, more symptoms attributed to MUS (identify), longer timeline) and emotional representations would be related to more negative self-management/coping and worse health outcomes. We hypothesised that protective illness representations (greater control beliefs and more coherence) would be related to more positive self-management/coping and better health outcomes. We did not have specific hypotheses about causal beliefs.

Method

Literature search

Potential sources for inclusion in this review were identified via keyword searches of the MEDLINE, Web of Science, PsychINFO, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and ProQuest databases. Keywords included in the search were terms used to identify articles with discussion of the CSM (e.g. 'illness representation', 'illness perception', 'illness beliefs', 'health beliefs') and MUS (e.g. 'medically unexplained illness', 'gulf war illness', 'fibromyalgia', 'chronic fatigue syndrome'). These databases were selected in consultation with a psychology research librarian for their focus on psychology, health, and medical science literature. A complete list of the search terms and scripts used in each database is included in Appendix A; there was not a protocol. The search was last updated on April 26, 2016.

Inclusion and exclusion criteria

An initial abstract review and final article review were conducted. Articles were included if they reported: 1) data on adult patients who had been diagnosed with MUS (e.g. chronic pain, fibromyalgia, irritable bowel syndrome); and 2) the article reported the correlation between any combination of the independent or dependent variables described in [Table 1](#), or reported the data in a way which allowed such a correlation to be computed. Articles were excluded if: 1) patients were not diagnosed with MUS, 2) patients were not adults, and 3) neither an illness representation/illness belief nor a health outcome was measured and reported on. All samples, irrespective of national origin were included although all coded articles were written in English.

Coding procedure

The initial screening of articles (760 articles) was conducted by three authors (JK, SS, KM) and two masters students in counselling psychology. Each abstract was reviewed by two coders, and any discrepancies were resolved through discussions in a weekly meeting with the first author (LMM). One hundred and eighty-two articles were moved forward for full article review. Full articles were reviewed by two of the authors (JK, SS, KM) and the data was entered into a database. The first author re-reviewed all included articles and re-entered data to ensure accuracy.

We coded the articles using pre-determined coding categories which are included in Appendix D. For illness representations we used Hagger et al.'s (2017) codes which included: consequences, timeline, identity, emotional representations, control and coherence. These categories are based off of the most used illness representations measure, the Illness Perception Questionnaire (also brief version and revised version) (Broadbent, Petrie, Main, & Weinman, 2006; Broadbent et al., 2015;

Table 1. Meta-analytic estimates of the relationships between independent variables and dependent variables.

Independent Variable	Dependent Variable	k	N	r_{obs}	SD_{obs}	ρ	2.5%CI	97.5%CI	SD _p	10%CV	90%CV	%Var
Cause ¹	Psych. Distress	3	422	0.02	0.00	0.02	-0.07	0.11	0.00	0.02	0.02	100
Cause	Quality of life	4	589	-0.09	0.00	-0.15	-0.18	-0.12	0.00	-0.15	-0.15	100
Coherence	Disease State	6	762	-0.06	0.05	-0.07	-0.16	0.02	0.05	-0.14	0.00	76
Coherence	Psych. Distress	2	326	-0.10	0.00	-0.12	-0.17	-0.07	0.00	-0.12	-0.12	100
Coherence	Quality of life	9	1,428	0.10	0.04	0.11	0.05	0.18	0.04	0.06	0.12	82
Consequence	Disease State	9	1,098	0.30	0.10	0.36	0.26	0.46	0.11	0.21	0.50	45
Consequence	Psych. Distress	7	969	0.41	0.07	0.50	0.41	0.59	0.08	0.40	0.60	57
Consequence	Quality of life	14	2,100	-0.46	0.08	-0.55	-0.61	-0.48	0.09	-0.66	-0.43	48
Cyclical Timeline	Disease State	3	387	-0.08	0.00	-0.08	-0.17	0.01	0.00	-0.08	-0.08	100
Cyclical Timeline	Psych. Distress	2	184	0.11	0.15	0.12	-0.16	0.39	0.16	-0.09	0.32	31
Cyclical Timeline	Quality of life	4	723	0.03	0.04	0.03	-0.06	0.12	0.04	-0.03	0.08	75
Emot. Rep.	Disease State	6	762	0.23	0.09	0.31	0.29	0.32	0.12	0.16	0.46	48
Emot. Rep.	Psych. Distress	2	184	0.64	0.00	0.88	0.81	0.94	0.00	0.88	0.88	100
Emot. Rep.	Quality of life	10	1,342	-0.42	0.09	-0.56	-0.66	-0.47	0.12	-0.71	-0.41	40
Identity	Disease State	7	888	0.28	0.19	0.34	0.15	0.53	0.23	0.05	0.63	17
Identity	Psych. Distress	6	854	0.26	0.03	0.32	0.24	0.40	0.02	0.29	0.35	96
Identity	Quality of life	9	1,517	-0.36	0.09	-0.44	-0.53	-0.35	0.11	-0.58	-0.30	40
Internal Control	Disease State	9	1,087	-0.09	0.03	-0.12	-0.19	-0.04	0.04	-0.16	-0.07	90
Internal Control	Psych. Distress	5	687	-0.15	0.00	-0.18	-0.27	-0.10	0.00	-0.18	-0.18	100
Internal Control	Quality of life	13	2,009	0.21	0.00	0.26	0.21	0.31	0.00	0.26	0.26	100
Psychological Cause	Disease State	2	210	0.17	0.00	0.21	0.14	0.27	0.00	0.21	0.21	100
Psychological Cause	Psych. Distress	5	663	0.32	0.00	0.39	0.38	0.41	0.00	0.39	0.39	100
Psychological Cause	Quality of life	6	801	-0.21	0.10	-0.26	-0.38	-0.13	0.12	-0.41	-0.11	45
Timeline	Disease State	6	762	0.18	0.06	0.21	0.11	0.32	0.07	0.13	0.30	69
Timeline	Psych. Distress	5	698	0.19	0.13	0.23	0.06	0.40	0.16	0.03	0.43	29
Timeline	Quality of life	11	1,674	-0.15	0.07	-0.18	-0.26	-0.11	0.08	-0.28	-0.08	60

Note: k = number of studies, N = number of subjects, r_{obs} = sample size weighted mean observed correlation, ρ = true score correlation, 2.5%CI and 97.5%CI = lower and upper bound of 95% confidence interval, SD_{ρ} = standard deviation of true score correlation, 10%CV and 90%CV = lower and upper bound of 80% credibility intervals, %Var = percentage of variance in observed effect sizes that can be explained by study artifacts, ¹=cause refers to perceiving a cause of the MUS being something other than psychological.

Moss-Morris et al., 2002; Weinman, Petrie, Moss-Morris, & Horne, 1996). Consistent with previous research, we coded for psychological causal beliefs (MacLeod, Haynes, & Sensky, 1998) and cause other than psychological (e.g. illness, environmental hazard). Cyclical timeline was included as a separate code. Self-management/coping was coded as problem-focused (directly coping with the MUS) or emotional coping (coping with the emotional consequences of MUS) and either negative (considered harmful e.g. catastrophising) or positive [considered beneficial e.g. acceptance] (Gray & Rutter, 2007; Moss-Morris & Wrapson, 2003; Spence et al., 2005)]. Health outcomes were coded as perceived disease state (e.g. symptom severity), psychological distress (e.g. depression) and quality of life.

For each effect the following information was coded: 1) the correlation (r) between two variables (illness representation, self-management, or outcomes), 2) the sample size (N) associated with that correlation, 3) the independent variable (e.g. timeline), 4) the local reliability estimate for the independent variable, 5) the dependent variable (e.g. quality of life), 6) the local reliability estimate for the dependent variable, 7) the mediator variable (e.g. positive problem-focused coping), and 8) the local reliability estimate for the mediator variable. A variety of other study characteristics (e.g. gender, age, study design) were also coded as potential moderators of effect sizes but the literature was not sufficiently large enough to allow exploration of moderators.

Final database

Twenty three articles were included in the meta-analysis. The final database includes 64 correlations from 24 independent samples representing 3,407 patients with MUS. The final database and output are publically available (<https://osf.io/gm329/>; DOI 10.17605/OSF.IO/GM329 | ARK c7605/osf.io/gm329) and a list of all included articles is attached in Appendix C and E. There was little variation in the quality of the studies. All studies were cross-sectional or reported on the cross-sectional relationship within a larger study. All studies used appropriate measures and analyses (correlation). Information about the study samples and measures are included in Appendix C.

Analytical approach

Whenever more than one correlation for a particular relationship was obtained from a single sample, those correlations were combined into a single estimate using the approach described by Ghiselli, Campbell, and Zedeck (1981). When the information necessary to compute composites was not present, we computed simple averages. We also computed Mosier reliability estimates for composites when possible and if not possible, we calculated simple averages of local reliability estimates.

We computed meta-analytic estimates of relationships using the Schmidt and Le software package (Schmidt & Le, 2004) which is based on Schmidt and Hunter's (2014) interactive psychometric method. This approach was preferred over other approaches for two reasons. First, it is based on a random-effects model and therefore provides a direct estimate of the degree to which effect sizes are characterised by greater variability than that which would be expected as a result of sampling error and study artefacts such as measurement error. This, in turn, provides information regarding the degree to which relationships are characterised by heterogeneity and therefore likely to be moderated by study or sample characteristics. Second, it allows researchers to correct for unreliability in the measurement of both the independent and dependent variables, even if the individual studies do not report local reliability estimates for measured variables. That is, we used the reported reliability information for each variable to construct reliability distributions (see Appendix B), and then used these reliability distributions to correct the distribution of observed correlations for measurement error.

We report eight statistics for each meta-analytic estimate of a relationship: (1) the number of independent samples on which the estimate was based (k), (2) the total sample size across those independent samples (N), (3) the sample-size weighted mean observed correlation (r_{obs}), (4) the estimate of the population correlation disattenuated for unreliability in the measurement of both variables (ρ), (5)

the lower and upper bound of the 95% confidence interval of effects sizes (2.5%CI and 97.5%CI), (6) the estimate of the standard deviation in effect sizes after removing the variability that can be attributed to sampling error and variability in the reliability of measurement across studies (SD_{ρ}), (7) the lower and upper bound of the 80% credibility interval of effect sizes (10%CV and 90%CV), and (8) the proportion of the total observed variance in effect sizes that can be accounted for by study artefacts (%Var).

The last three of these values speak to the absence or presence of moderators; large values for SD_{ρ} , wide credibility intervals, and small values for %Var all indicate the likely presence of moderators of the relationship. Schmidt and Hunter (2014), suggest the use of the 75% rule: if examined study artefacts can account for at least 75% of the observed variance in effect sizes then it is likely that there are no substantive moderators; the reasoning being that unaccounted for study artefacts would likely account for the remaining amount of variance. Similarly, a credibility interval that contains zero would indicate that there are situations in which the relationship would be zero.

Consistent with previous literature, effect sizes for the main effects correlations above .10 (Hagger et al., 2017; Seaton, Marsh, & Craven, 2010) for which the 95% confidence interval did not include zero were interpreted. We interpreted .10–.20 as a small correlation, .20–.30 as a moderate correlation and .30 as a large correlation consistent with a meta-analysis of effects in social psychology literature (Richard, Bond & Stokes-Zoota, 2003). The direct effects are summarised in Figure 1.

Mediation analysis

We conducted simple mediation analysis for all possible combinations of a single independent variable, single mediator variable, and single dependent variables. One hundred and eight combinations

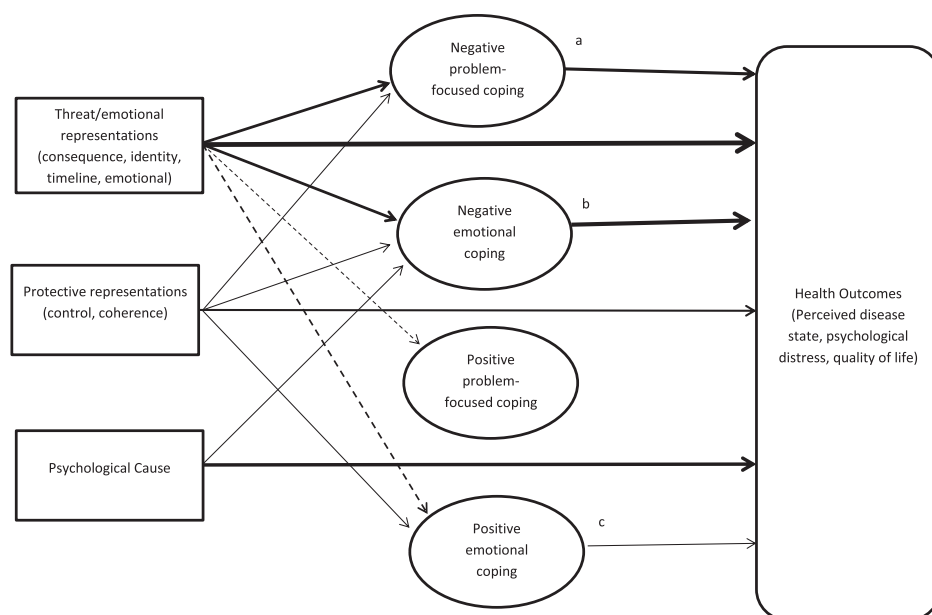


Figure 1. Direct relationships between illness representations, self-management/coping and health outcomes.

Notes: 1. Cyclical timeline and perceiving a cause other than psychological are not depicted in this figure as there were few effects for these illness representations. 2. Thicker lines indicate larger effects and dashed lines indicate inconsistent effects. a. Negative problem-focused coping had a consistent direct effect on health outcomes, a consistent mediational effect between illness representations and quality of life, and an inconsistent mediational effect between illness representations and perceived disease state. b. negative emotional coping had both a consistent direct effect and consistent mediational effect (with one exception) on the relationship between illness representations and health outcomes. c. positive emotional coping had a consistent direct effect on health outcomes and an inconsistent mediational effect. Positive emotional coping inconsistently mediated the relationship between threat/emotional representations and quality of life and consistently mediated the relationship between protective illness representations and quality of life. Not shown in this figure, positive emotional coping also mediated the relationship between perceiving a cause other than psychological and quality of life.

of such variables were possible and the necessary meta-analytic estimates of correlations among any set of three variables was available for 58 of these combinations. In each combination we constructed a meta-analytic correlation matrix among the three variables and then used this correlation matrix in conjunction with the harmonic mean of the sample sizes to compute four standardised regression coefficients: (1) the coefficient a for the prediction of the mediator from the independent variable, (2) the coefficient c for the prediction of the dependent variable from the independent variable, and (3) the coefficients b and c' when the mediator and independent variable jointly predict the dependent variable.

For each mediation analysis we then computed the size of the completely standardised indirect effect ab , also referred to as the index of mediation (Preacher & Hayes, 2008; Preacher & Kelley, 2011) to reflect the size of the indirect effect. This index of mediation reflects the standard deviation change in the dependent variable that results from a standard deviation increase in the independent variable via the mediator. If one views a standardised regression coefficient of .30 as having a large effect size then an indirect effect of .09 or $(.30 \times .30)$ might be considered as a large effect size for indirect effects. We interpreted mediation effects .04 or higher $(.20 \times .20)$, a moderate effect size. We also compute the ratio of the indirect effect to the total effect (ab/c) to represent the proportion of the total effect that is mediated, although it should be remembered that this value can be large even when the total effect is very small. Instances in which this ratio is greater than one represent likely suppression and were not interpreted.

Results

Association of illness representations to health outcomes

There was a moderate to large effect of threat-related (consequences, identity, timeline) and emotional illness representations being related to worse outcomes ($|\rho| = .18$ to $.88$; Table 1). Greater severity of consequences, more symptoms attributed to MUS (identity), longer timeline, and more emotional representations were each related to worse perceived disease state (greater number and severity of symptoms), worse psychological distress, and poorer quality of life.

Protective illness representations (control and coherence) had a small to moderate relationship with better disease state, lower psychological distress and better quality of life ($|\rho| = .11$ to $.26$). Perceiving greater control was related to lower perceived disease state, less psychological distress, and greater quality of life. Perceiving greater coherence was related to less psychological distress and greater quality of life (coherence had no relationship with perceived disease state).

Perceiving a non-psychological cause of MUS was related to lower quality of life ($\rho = -.15$) but was not related to psychological distress. Greater perception of a psychological cause of MUS was related to worse disease state, greater psychological distress and lower quality of life ($|\rho| = .21$ to $.39$). Cyclical timeline was not related to health outcomes.

Relationship of illness representations to self-management/coping

Threat-related representations (consequence, identity, timeline) and emotional illness representation each had a moderate to large relationship with greater use of negative problem-focused coping [e.g. all or nothing activity ($\rho = .28$ to $.34$; Table 2)] and negative emotional coping [e.g. catastrophising ($\rho = .20$ to $.55$)]. The only exception was there was no relationship between timeline and negative problem-focused coping.

Threat-related/emotional illness representations had an inconsistent relationship with positive problem-focused (e.g. problem-solving) and positive emotional coping (e.g. acceptance). Perceiving more consequences predicted more positive problem-focused coping ($\rho = .15$). More emotional representations predicted less positive emotional coping ($\rho = -.65$). Otherwise, positive problem-focused coping was not predicted by attributing more symptoms to MUS (identity), timeline,

Table 2. Meta-analytic estimates of the relationships between independent variables and mediators.

Independent Variable	Mediator	k	N	r _{obs}	SD _{obs}	ρ	2.5%CI	97.5%CI	SD _p	10%CV	90%CV	%Var
Cause ¹	-Emotional	2	385	0.02	0.00	0.03	0.02	0.05	0.00	0.03	0.03	100
Cause	+ Problem-focused	2	385	0.01	0.00	0.02	-0.13	0.16	0.00	0.02	0.02	100
Cause	+ Emotional	2	385	-0.08	0.00	-0.15	-0.32	0.02	0.00	-0.15	-0.15	100
Coherence	- Problem-focused	2	252	-0.09	0.00	-0.12	-0.15	-0.09	0.00	-0.12	-0.12	100
Coherence	- Emotional	3	402	-0.15	0.09	-0.18	-0.34	-0.01	0.10	-0.30	-0.05	48
Coherence	+ Problem-focused	4	645	0.02	0.00	0.02	-0.04	0.09	0.00	0.02	0.02	100
Coherence	+ Emotional	2	326	0.13	0.00	0.16	0.09	0.23	0.00	0.16	0.16	100
Consequences	- Problem-focused	3	485	0.21	0.00	0.28	0.20	0.36	0.00	0.28	0.28	100
Consequences	- Emotional	8	1,297	0.32	0.14	0.40	0.27	0.54	0.16	0.19	0.61	25
Consequences	+ Problem-focused	7	1,184	0.13	0.11	0.15	0.04	0.26	0.14	-0.03	0.32	33
Consequences	+ Emotional	5	865	-0.06	0.26	-0.08	-0.40	0.25	0.35	-0.52	0.37	8
Emot. Rep.	- Problem-focused	2	252	0.20	0.00	0.33	0.32	0.35	0.00	0.33	0.33	100
Emot. Rep.	- Emotional	4	556	0.39	0.20	0.55	0.25	0.85	0.28	0.19	0.91	13
Emot. Rep.	+ Problem-focused	5	606	0.01	0.08	0.02	-0.14	0.18	0.12	-0.13	0.18	54
Emot. Rep.	+ Emotional	2	247	-0.42	0.16	-0.65	-1.04	-0.27	0.25	-0.97	-0.34	18
Identity	- Problem-focused	3	408	0.22	0.00	0.34	0.29	0.39	0.00	0.34	0.34	100
Identity	- Emotional	5	924	0.16	0.10	0.21	0.07	0.35	0.13	0.04	0.37	34
Identity	+ Problem-focused	7	1,243	0.09	0.14	0.12	-0.04	0.27	0.19	-0.12	0.36	21
Identity	+ Emotional	4	691	0.00	0.20	0.00	-0.31	0.31	0.27	-0.34	0.34	13
Internal Control	- Problem-focused	2	252	-0.10	0.00	-0.15	-0.23	-0.07	0.00	-0.15	-0.15	100
Internal Control	- Emotional	7	1,200	-0.20	0.16	-0.26	-0.43	-0.10	0.20	-0.52	-0.01	19
Internal Control	+ Problem-focused	6	1,087	0.13	0.14	0.17	0.00	0.34	0.19	-0.07	0.41	21
Internal Control	+ Emotional	4	768	0.15	0.06	0.22	0.09	0.34	0.08	0.11	0.32	60
Psych. Cause	- Emotional	4	577	0.14	0.02	0.18	0.13	0.24	0.00	0.18	0.18	100
Psych. Cause	+ Problem-focused	3	482	0.05	0.11	0.07	-0.14	0.28	0.15	-0.12	0.26	33
Psych. Cause	+ Emotional	2	385	-0.01	0.00	-0.02	-0.13	0.09	0.00	-0.02	-0.02	100
Timeline	- Problem-focused	3	485	0.07	0.08	0.11	-0.08	0.30	0.12	-0.04	0.27	48
Timeline	- Emotional	5	844	0.15	0.12	0.20	0.04	0.35	0.15	0.01	0.38	32
Timeline	+ Problem-focused	5	854	0.01	0.04	0.01	-0.09	0.11	0.05	-0.05	0.07	82
Timeline	+ Emotional	3	535	-0.08	0.13	-0.11	-0.34	0.12	0.18	-0.33	0.12	25

Note: k = number of studies, N = number of subjects, + = positive, - = negative, r_{obs} = sample size weighted mean observed correlation, r = true score correlation, 2.5%CI and 97.5%CI = lower and upper bound of 95% confidence interval, SD_p = standard deviation of true score correlation, 10%CV and 90%CV = lower and upper bound of 80% credibility intervals, %Var = percentage of variance in observed effect sizes that can be explained by study artifacts. ¹=cause refers to perceiving a cause of the MUS being something other than psychological

emotional representation; and greater positive emotional coping was not predicted by consequence, identity or timeline.

Protective illness representations (control and coherence) each had a small to moderate relationship with less use of negative problem-focused coping ($\rho = -.12$ and $-.28$) and negative emotional coping ($\rho = -.18$ and $-.26$) and greater use of positive emotional coping ($\rho = .16$ to $.22$). Protective illness representations were not related to positive problem-focused coping.

Generally causal beliefs did not predict self-management/coping. The exception to this was greater perception of a psychological cause to MUS was related to greater negative emotional coping ($\rho = .18$). Perceiving a cause other than psychological had no relationship with negative emotional coping. Perceiving a psychological or non-psychological cause was not related to positive problem-focused or emotional coping. There were not enough studies to examine the relationship of causal beliefs to negative problem-focused coping.

There were not enough studies to examine the relationship of cyclical timeline to any self-management/coping.

Relationship of self-management/coping to health outcomes

Negative coping had a consistent moderate to large relationship with worse health outcomes. Negative problem-focused coping (e.g. all or nothing activity) predicted worse perceived disease state and lower quality of life ($|\rho| = .21$ to $.41$; Table 3). Negative emotional coping (e.g. catastrophizing) predicted worse perceived disease state, greater psychological distress and lower quality of life ($|\rho| = .44$ to $.53$).

There were few studies on positive coping and health outcomes and these showed inconsistent results. Positive problem-focused coping (e.g. problem-solving) was not related to perceived disease state or quality of life. Positive emotional coping (e.g. acceptance) was related to better quality of life ($\rho = .46$).

There were not enough studies to examine the relationships between negative problem-focused coping and psychological distress, positive problem-focused coping and psychological distress, positive emotional coping and perceived disease state, or positive emotional coping and psychological distress.

Mediation

Negative problem-focused coping

Negative problem-focused coping (e.g. all or nothing activity) consistently mediated the relationship between illness representations and *quality of life*. These relationships can be interpreted as greater threat/emotional representations were associated with more negative problem-focused coping, which was associated with lower quality of life ($ab = .04$ to $.10$, Table 4); greater protective representations were associated with less negative problem-focused coping which was associated with better quality of life ($ab = .06$ and $.08$).

Negative problem-focused coping was an inconsistent mediator of the relationship between illness representations and *perceived disease state*. Negative problem-focused coping mediated the relationship between emotional representations and perceived disease state ($ab = .04$) and identity and perceived disease state ($ab = .04$). This can be interpreted as greater emotional representations and identity were each associated with more negative problem-focused coping which was associated with more disease state. Negative problem-focused coping did not mediate the relationship between consequence, timeline, control or coherence to *perceived disease state*.

Negative emotional coping

With only a few exceptions, negative emotional coping (e.g. catastrophizing) had a large mediational effect on the relationship between illness representations and health outcomes. Negative emotional coping mediated the relationship between *threat-related/emotional* illness representations and

Table 3. Meta-analytic estimates of the relationships between mediators and dependent variables.

Mediator	Dependent Variable	k	N	r_{obs}	SD_{obs}	ρ	2.5%CI	97.5%CI	$SD\rho$	10%CV	90%CV	%Var
- Problem-focused	Disease State	2	252	0.15	0.00	0.21	0.04	0.37	0.00	0.21	0.21	100
- Problem-focused	Quality of life	2	252	-0.29	0.00	-0.41	-0.43	-0.38	0.00	-0.41	-0.41	100
- Emotional	Disease State	2	245	0.45	0.00	0.53	0.46	0.59	0.00	0.53	0.53	100
- Emotional	Psych. Distress	2	382	0.44	0.24	0.52	0.11	0.92	0.28	0.16	0.87	10
- Emotional	Quality of life	6	816	-0.38	0.04	-0.44	-0.52	-0.36	0.04	-0.49	-0.39	85
+ Problem-focused	Disease State	2	252	-0.08	0.00	-0.09	-0.20	0.01	0.00	-0.09	-0.09	100
+ Problem-focused	Quality of life	4	525	-0.08	0.22	-0.10	-0.38	0.18	0.26	-0.43	0.24	14
+ Emotional	Quality of life	3	423	0.36	0.21	0.46	0.14	0.79	0.27	0.12	0.80	11

Note: k = number of studies, N = number of subjects, + = positive, - = negative, r_{obs} = sample size weighted mean observed correlation, p = true score correlation, 2.5%CI and 97.5%CI = lower and upper bound of 95% confidence interval, $SD\rho$ = standard deviation of true score correlation, 10%CV and 90%CV = lower and upper bound of 80% credibility intervals, %Var = percentage of variance in observed effect sizes that can be explained by study artifacts.

Table 4. Summary of mediation analysis results.

IV	Mediator	DV	a	b	c	c'	ab	ab/c
Cause ¹	—Emotional	Quality of Life	0.03	-0.44	-0.15	-0.14	-0.01	0.10
Cause	—Emotional	Psych. Distress	0.03	0.52	0.02	0.00	0.02	0.71
Cause	+ Problem-focused	Quality of Life	-0.15	-0.10	-0.15	-0.15	0.00	0.01
Cause	+ Emotional	Quality of Life	-0.15	0.45	-0.15	-0.08	-0.07	0.46
Coherence	—Problem-focused	Disease State	-0.12	0.21	-0.07	-0.05	-0.02	0.36
Coherence	—Problem-focused	Quality of Life	-0.12	-0.40	0.11	0.06	0.05	0.42
Coherence	—Emotional	Disease State	-0.18	0.54	-0.07	0.03	-0.09	Supp.
Coherence	—Emotional	Quality of Life	-0.18	-0.43	0.11	0.03	0.08	0.67
Coherence	—Emotional	Psych. Distress	-0.18	0.52	-0.12	-0.03	-0.09	0.77
Coherence	+ Problem-focused	Disease State	0.02	-0.09	-0.07	-0.07	0.00	0.03
Coherence	+ Problem-focused	Quality of Life	0.02	-0.10	-0.07	-0.07	0.00	-0.02
Coherence	+ Emotional	Quality of Life	0.16	0.45	0.11	0.11	0.00	0.63
Consequences	—Problem-focused	Disease State	0.28	0.12	0.36	0.33	0.03	0.09
Consequences	—Problem-focused	Quality of Life	0.28	-0.28	-0.55	-0.47	-0.08	0.14
Consequences	—Emotional	Disease State	0.40	0.46	0.36	0.18	0.19	0.52
Consequences	—Emotional	Quality of Life	0.40	-0.26	-0.55	-0.45	-0.11	0.19
Consequences	+ Problem-focused	Psych. Distress	0.40	0.38	0.50	0.35	0.15	0.31
Consequences	+ Problem-focused	Disease State	0.15	-0.15	0.36	0.38	-0.02	-0.06
Consequences	+ Emotional	Quality of Life	0.15	-0.02	-0.55	-0.52	0.00	0.00
Consequences	+ Emotional	Quality of Life	-0.08	0.42	-0.55	-0.52	-0.03	0.06
Emot. Rep.	—Problem-focused	Disease State	0.33	0.12	0.31	0.27	0.04	0.13
Emot. Rep.	—Problem-focused	Quality of Life	0.33	-0.25	-0.56	-0.48	-0.08	0.15
Emot. Rep.	—Emotional	Disease State	0.55	0.52	0.31	0.03	0.28	0.92
Emot. Rep.	—Emotional	Quality of Life	0.55	-0.19	-0.56	-0.46	-0.10	0.19
Emot. Rep.	+ Problem-focused	Psych. Distress	0.55	0.05	0.88	0.85	0.03	0.03
Emot. Rep.	+ Problem-focused	Disease State	0.02	-0.10	0.31	0.31	0.00	-0.01
Emot. Rep.	+ Problem-focused	Quality of Life	0.02	-0.09	-0.56	-0.56	0.00	0.00
Emot. Rep.	+ Emotional	Quality of Life	-0.65	0.17	-0.56	-0.45	-0.11	0.19
Identity	—Problem-focused	Disease State	0.34	0.11	0.34	0.30	0.04	0.11
Identity	—Problem-focused	Quality of Life	0.34	-0.29	-0.44	-0.34	-0.10	0.23
Identity	—Emotional	Disease State	0.21	0.48	0.34	0.24	0.10	0.30
Identity	—Emotional	Quality of Life	0.21	-0.36	-0.44	-0.36	-0.08	0.17
Identity	+ Problem-focused	Psych. Distress	0.21	0.47	0.32	0.22	0.10	0.31
Identity	+ Problem-focused	Disease State	0.12	-0.13	0.34	0.36	-0.02	-0.05
Identity	+ Problem-focused	Quality of Life	0.12	-0.05	-0.44	-0.43	-0.01	0.01
Identity	+ Emotional	Quality of Life	0.00	0.46	-0.44	-0.44	0.00	0.00
Internal Control	—Problem-focused	Disease State	-0.15	0.20	-0.12	-0.09	-0.03	0.26
Internal Control	—Problem-focused	Quality of Life	-0.15	-0.38	0.26	0.20	0.06	0.22
Internal Control	—Emotional	Disease State	-0.26	0.54	-0.12	0.02	-0.14	Supp.
Internal Control	—Emotional	Quality of Life	-0.26	-0.40	-0.12	0.16	0.11	0.40
Internal Control	—Emotional	Psych. Distress	-0.26	0.51	-0.18	-0.05	-0.13	0.74

Internal Control	+ Problem-focused	Disease State	-0.07	-0.12	-0.11	-0.01	0.11
Internal Control	+ Problem-focused	Quality of Life	-0.15	0.26	0.29	-0.02	-0.09
Internal Control	+ Emotional	Quality of Life	0.42	0.26	0.17	0.09	0.35
Psych. Cause	-Emotional	Disease State	0.51	0.21	0.12	0.09	0.45
Psych. Cause	-Emotional	Quality of Life	-0.41	-0.26	-0.19	-0.07	0.29
Psych. Cause	-Emotional	Psych. Distress	0.47	0.39	0.31	0.09	0.22
Psych. Cause	+ Problem-focused	Disease State	-0.11	0.21	0.22	-0.01	-0.04
Psych. Cause	+ Problem-focused	Quality of Life	-0.08	-0.26	-0.25	-0.01	0.02
Psych. Cause	+ Emotional	Quality of Life	0.46	-0.26	-0.25	-0.01	0.04
Timeline	-Problem-focused	Disease State	0.19	0.21	0.19	0.02	0.10
Timeline	-Problem-focused	Quality of Life	-0.40	-0.18	-0.14	-0.04	0.24
Timeline	-Emotional	Disease State	0.51	0.21	0.11	0.10	0.47
Timeline	-Emotional	Quality of Life	-0.42	-0.18	-0.10	-0.08	0.45
Timeline	-Emotional	Psych. Distress	0.49	0.23	0.13	0.10	0.43
Timeline	+ Problem-focused	Disease State	-0.09	0.21	0.21	0.00	0.00
Timeline	+ Problem-focused	Quality of Life	-0.10	-0.18	-0.18	0.00	0.01
Timeline	+ Emotional	Quality of Life	0.45	-0.18	-0.13	-0.05	0.26

Note: +=positive, -=negative, a = standardised regression coefficient for the prediction of the mediator from the independent variable, c = standardised regression coefficient for the prediction of the dependent variable from the independent variable, b and c' = standardised regression coefficients for the mediator and independent variable when jointly predicting the dependent variable, ab = index of mediation, ab/c = proportion of total effect explained by indirect effect. =cause refers to perceiving a cause of the MUS being something other than psychological.

health outcomes (disease state, psychological distress and quality of life; $|ab| = .08$ to $.28$), such that greater threat/emotional representations were associated with greater negative emotional coping which was associated with worse outcomes. The exception was there was only a small (non-interpreted) mediational effect ($ab = .03$) of negative emotional coping on the relationship between emotional representations to psychological distress.

Negative emotional coping mediated all the relationships between *protective* illness representations (control and coherence) and health outcomes ($|ab| = .08$ to $.29$) such that more protective illness representations were associated with less negative emotional coping, which was associated with better health outcomes. Negative emotional coping also mediated the relationship between perceiving a *psychological cause* of MUS and health outcomes ($|ab| = .07$ to $.09$), such that greater perception of a psychological cause was associated with more negative emotional coping and worse health outcomes. Negative emotional coping did not mediate the relationship between perceiving a *cause other than psychological* and health outcomes.

Positive problem-focused and emotional coping

Positive *problem-focused* (e.g. problem-solving) coping did not (or $ab < .04$) mediate any of the relationships between illness representations and health outcomes.

Positive *emotional* coping (e.g. acceptance) inconsistently mediated the relationship between threat/emotional illness representations and health outcomes. Positive emotion focused coping mediated the relationship between timeline ($ab = .05$) and emotional representations ($ab = .11$) with quality of life, such that greater timeline and more emotional representations were associated with less positive emotional coping which was associated with lower quality of life. Positive emotional coping did not mediate the relationship between consequence or identity representations and quality of life.

Positive emotional coping consistently mediated the relationship between protective illness representations and quality of life ($ab = .09$ and $.07$), such that more protective illness representations were associated with more positive emotion focused coping which was associated with greater quality of life.

Positive emotional coping mediated the relationship between perceiving a cause other than psychological and quality of life, such that greater perception of a cause other than psychological was associated with less positive emotional coping which was associated with poorer quality of life. Positive emotional coping did not mediate the relationship between perceiving a psychological cause and quality of life.

Bias

We used Egger's Test of Funnel Plot Asymmetry (Egger, Smith, Schneider, & Minder, 1997) to examine if there was publication bias, where studies with non-significant effects were not published. Here, the standard normal deviate of the effect size estimate for studies is regressed against the precision of the effect size estimate. Because of the relatively small number of unique samples, we conducted the Egger's Test on the meta-analytic estimate with the largest number of independent samples (i.e. the consequence – quality of life relationship). For these fourteen effect sizes, there was no statistically significant evidence of publication bias ($a = .64$, $p = .68$). While the statistical power to detect publication bias is low for this number of studies, there is no strong evidence that publication bias would result in upwardly biased meta-analytic effect size estimates.

Discussion

Illness representations are considered the key to understanding medically unexplained symptoms, and have been the basis of interventions to improve the quality of life of patients with MUS. Despite this, there has not been a comprehensive analysis of *which* illness representations are

important or an analysis of *how* they are related to outcomes. To address this gap in the literature, we conducted a meta-analysis of the relationship of illness representations and health outcomes and tested if self-management/coping mediated this relationship. We examined the relationships between threat-related illness representations, protective illness representations and causal illness representations to self-management/coping and health outcomes.

We reviewed 23 studies and found that threat-related illness representations (e.g. consequence, identity and timeline) and emotional representations were related to worse health outcomes and more negative coping (moderate to large effect). Generally, increases in negative coping mediated (with a moderate to large effect) the relationship of threat/emotional illness representations and health outcomes. Protective illness representations (control and coherence) were related to better health outcomes, less use of negative coping and generally related to greater use of positive coping (small to moderate effect). The relationship of protective illness representations to better health outcomes was generally mediated by decreases in negative coping (moderate to large effect) and increases in positive coping (moderate effect). Perceiving a psychological cause to the MUS was related to more negative health outcomes (moderate to large effect) and more negative emotional coping (small effect). The relationship of perceiving a psychological cause and more negative health outcomes was mediated by increases in negative emotional coping (moderate to large effect).

Our finding that perceiving a psychological cause is related to negative outcomes is important. Difficulties in the communication between patients with MUS and providers are thought to stem from patients perceiving the cause of MUS to be physical while providers perceive the cause to be psychological. Qualitative studies with providers document that providers feel that patients pressure them to provide a medical explanation and medical treatment for their MUS symptoms (Casiday, Hungin, Cornford, de Wit, & Blell, 2009; Czachowski et al., 2011; Dixon-Woods & Critchley, 2000). In response, reattribution treatment, in part, teaches physicians skills to help patients perceive the cause of MUS to be psychological. Cognitive behavioural treatments for MUS similarly teach patients the relationship of stress to their MUS. Recently, however, the efficacy of these treatments have been questioned. A recent review concluded that there is little evidence for reattribution treatment in the current form (Gask et al., 2011). Similarly, the effect sizes of cognitive behavioural treatments for MUS are smaller than those for patients with other disorders [e.g. depression) (Hofmann et al., 2012)]. The findings of this meta-analysis suggest that helping patients reattribute their illness representations from a physical to a psychological cause may not be beneficial.

Reducing threat-related illness representations may be more important to improving health outcomes for patients with MUS. We found that threat-related illness representations accounted for up to 77% of the variance in health outcomes. It is unlikely that challenging threat-related illness representations (top-down approach) will be beneficial. Patients believe that MUS cause significant consequences, last a long-time (timeline), cause many symptoms (identity) and cause emotional consequences (emotional representation), because they do. For many patients, MUS cause more disability than other better understood health conditions (Bombardier & Buchwald, 1996; Hotopf et al., 2003; Moss-Morris & Chalder, 2003; White, Speechley, Harth, & Ostbye, 1999), they are highly comorbid with mental health conditions (Henningsen, Zimmermann, & Sattel, 2003), and many people with a MUS condition will have it their entire life (olde Hartman et al., 2009).

A better approach is likely a bottom up approach of addressing the behaviours patients enact in response to their illness representations. Our data suggests that threat-related illness representations are associated with poorer outcomes, in part, because they are associated with more negative coping, such as all or nothing behaviour or catastrophising. Behavioural techniques such as teaching patients to engage in moderate-paced new activities (as opposed to all or nothing activity spurts) or mindfulness to reduce catastrophic thinking can teach patients a better response to threat-related illness representations. Overtime, these strategies may reduce threat-related illness representations. For example, as patients use moderate activity to slowly reengage with their life they may perceive

fewer and less severe consequences from their MUS and perceive less of an emotional response to MUS.

Consistent with previous literature of other health conditions (Hagger et al., 2017), we found a smaller relationship for protective illness representations with health outcomes as compared to threat-related illness representations with health outcomes. Two recent meta-analyses across chronic conditions found a larger relationship between threat-related illness representations (consequences, identity and emotional response) and outcomes as compared to protective illness representations [coherence and control; (Broadbent et al., 2015; Hagger et al., 2017)]. Our finding that illness representations have a stronger relationship with negative coping as compared to positive coping is also confirmed by previous literature (Brandes & Mullan, 2014; French et al., 2006).

When comparing the effect sizes found in the current meta-analysis of MUS to effect-sizes found in reviews of patients with better understood chronic conditions, we found many effect sizes were relatively similar, with some being larger, for the relationship of illness representations to health outcomes (Hagger et al., 2017) and potentially larger effect sizes for the relationship of illness representations with self-management/coping. This interpretation should be viewed cautiously because there are differences in how self-management/coping is coded between the current study and Hagger et al. It has been hypothesised that illness representations may be particularly important for MUS (Moss-Morris & Wrapson, 2003), as a result the primary behavioural treatments for MUS have focused on changing these illness representations. Our results suggest for patients with MUS, illness representations are sometimes better predictors of how patients with self-manage/cope with their condition and sometimes better predictor of health outcomes as compared to patients with other chronic conditions.

There are other differences in our meta-analysis as compared to meta-analyses for other health conditions. Hudson et al. (2014) for example found that cyclical timeline was a strong predictor of poorer emotional health for patients with diabetes, while we found that it was not related to outcomes for patients with MUS. Broadbent et al. (2015) found smaller relationship with timeline to outcomes and coherence to outcomes across chronic conditions as compared to the effects found in the current study, suggesting that a chronic timeline might be more threatening and coherence more protective for patients with MUS as compared to patients with better understood chronic conditions. Finally, we found that illness representations may have a stronger relationship with perceived disease state for MUS as compared to better understood chronic conditions (Hagger et al., 2017), likely because disease state of MUS is typically self-report of symptom severity while disease state for other chronic conditions is often an objective measure (e.g. HbA1c). The current study was the first meta-analysis to examine the relationship of causal beliefs to outcomes for any health condition.

When comparing the mediational effects of the current study to Hagger et al. (2017) meta-analysis of all chronic conditions, it is notable that Hagger et al found that positive problem-focused coping was a consistent mediator of the relationship of many different illness representations to health outcomes. In contrast, positive problem-focused coping (e.g. problem-solving) did not mediate any of the relationships of illness representations to health outcomes for MUS. Positive problem-focused coping may be less impactful when the best self-management strategies are unknown, as is the case for MUS. Hagger et al. also found more complex mediational relationships. The current review found that the associations between threat/emotional illness representations and health outcomes were mediated by more negative coping; the associations between protective illness representations and health comes were mediated by less negative coping and more positive coping. Hagger et al. found that the relationship between consequences and identity were related to health outcomes through multiple pathways, some positive (e.g. problem-solving) and some negative (e.g. avoiding). Further, unexpectedly, Hagger et al found that coherence was associated with more positive outcomes through the greater use of negative coping. The more complex mediational relationships may be due to differences in statistical analysis approaches (i.e. Hagger et al. used multiple regression) or due to there being more complex relationships when examining all chronic conditions.

A limitation of the current study is that the reviewed studies used standardised measures of illness representations, such as the Illness Perception Questionnaire. We, and others, have argued that one of the most critical illness representations for patients with MUS is moving from the belief that one must find the cause of the symptoms – to a belief that one must focus on managing symptoms (McAndrew et al., 2017). This belief is not captured in standardised measures. The reviewed studies also used general coping measures. We may have found even larger effect sizes if illness representations and coping measures were specific to MUS. By using standardised measures there may also have been some conceptual overlap such as with identity and perceived disease status. Another limitation is the studies were cross-sectional. We interpreted the direction of the associations consistent with common-sense model and previous studies which have found that illness representations predict the onset of MUS (Hou et al., 2012) and improvements in outcomes (Chilcot & Moss-Morris, 2013). We could not, however, test the direction of these relationships in this study.

Conclusions

The goal of this meta-analysis was to determine *which* illness representations are related to health outcomes for patients with MUS and *how* they are related to health outcomes. Illness representations are considered important for MUS and changing illness representations has been a focus of behavioural interventions. A better understanding of how illness representations are related to health outcomes has the potential to improve interventions for MUS. There have been calls to develop interventions to address illness representations for other conditions. Improving treatments for MUS may help us develop these treatments for other conditions.

Overall, we found that illness representations are important predictors of health outcomes and self-management/coping for patients with MUS. We did not find that causal beliefs were especially related to outcomes, as has been previously suggested. Instead, threat related illness representations were the largest predictors of outcomes. The results of this meta-analysis suggest that future treatment efforts should focus on reducing threat/emotional representations and negative coping of patients with MUS.

Disclosure statement

No potential conflict of interest was reported by the authors.

Funding

This study was supported by grants from the Department of Veterans Affairs, Health Services Research & Development Service (IK2 HX001369-01A1 to L.M. McAndrew).

References

- Bombardier, C. H., & Buchwald, D. (1996). Chronic fatigue, chronic fatigue syndrome, and fibromyalgia: Disability and health-care use. *Medical Care*, 34, 924–930. doi:10.1097/00005650-199609000-00005
- Brandes, K., & Mullan, B. (2014). Can the common-sense model predict adherence in chronically ill patients? A meta-analysis. *Health Psychology Review*, 8, 129–153. doi:10.1080/17437199.2013.820986
- Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research*, 60, 631–637. doi:10.1037/t10379-10000
- Broadbent, E., Wilkes, C., Koschwanez, H., Weinman, J., Norton, S., & Petrie, K. J. (2015). A systematic review and meta-analysis of the brief illness perception questionnaire. *Psychology & Health*, 30, 1361–1385. doi:10.1080/08870446.2015.1070851
- Brown, R. J. (2004). Psychological mechanisms of medically unexplained symptoms: An integrative conceptual model. *Psychological Bulletin*, 130(793). doi:10.1080/08870446.2015.1070851
- Burton, C. (2003). Beyond somatisation: A review of the understanding and treatment of medically unexplained physical symptoms (MUPS). *British Journal of General Practice*, 53, 231–239.

- Casiday, R. E., Hungin, A., Cornford, C. S., de Wit, N. J., & Blell, M. T. (2009). GPs' explanatory models for irritable bowel syndrome: A mismatch with patient models? *Family Practice*, 26, 34–39. doi:10.1093/fampra/cm088
- Chilcot, J., & Moss-Morris, R. (2013). Changes in illness-related cognitions rather than distress mediate improvements in irritable bowel syndrome (IBS) symptoms and disability following a brief cognitive behavioural therapy intervention. *Behaviour Research and Therapy*, 51, 690–695. doi:10.1016/j.brat.2013.07.007
- Czachowski, S., Piszczek, E., Sowińska, A., & Olde Hartman, T. C. (2011). Challenges in the management of patients with medically unexplained symptoms in Poland: A qualitative study. *Family Practice*, 29, 228–234. doi:10.1093/fampra/cm065
- Deary, V., Chalder, T., & Sharpe, M. (2007). The cognitive behavioural model of medically unexplained symptoms: A theoretical and empirical review. *Clinical Psychology Review*, 27, 781–797. doi:10.1016/j.cpr.2007.07.002
- Dempster, M., Howell, D., & McCorry, N. K. (2015). Illness perceptions and coping in physical health conditions: A meta-analysis. *Journal of Psychosomatic Research*, 79, 506–513. doi:10.1016/j.jpsychores.2015.10.006
- Dixon-Woods, M., & Critchley, S. (2000). Medical and lay views of irritable bowel syndrome. *Family Practice*, 17, 108–113. doi:10.1093/fampra/17.2.108
- Egger, M., Smith, G. D., Schneider, M., & Minder, C. (1997). Bias in meta-analysis detected by a simple, graphical test. *BMJ*, 315, 629–634. doi:10.1136/bmj.315.7109.629
- Foxwell, R., Morley, C., & Frizelle, D. (2013). Illness perceptions, mood and quality of life: A systematic review of coronary heart disease patients. *Journal of Psychosomatic Research*, 75, 211–222. doi:10.1016/j.jpsychores.2013.05.003
- French, D. P., Cooper, A., & Weinman, J. (2006). Illness perceptions predict attendance at cardiac rehabilitation following acute myocardial infarction: A systematic review with meta-analysis. *Journal of Psychosomatic Research*, 61, 757–767. doi:10.1016/j.jpsychores.2006.07.029
- Frostholm, L., Oernboel, E., Christensen, K. S., Toft, T., Olesen, F., Weinman, J., & Fink, P. (2007). Do illness perceptions predict health outcomes in primary care patients? A 2-year follow-up study. *Journal of Psychosomatic Research*, 62, 129–138. doi:10.1016/j.jpsychores.2006.09.003
- Frostholm, L., Ørnboel, E., Hansen, H. S., Olesen, F., Weinman, J., & Fink, P. (2010). Which is more important for outcome: The physician's or the patient's understanding of a health problem? A 2-year follow-up study in primary care. *General Hospital Psychiatry*, 32, 1–8. doi:10.1016/j.genhosppsych.2009.08.004
- Frostholm, L., Petrie, K., Ørnboel, E., & Fink, P. (2014). Are illness perceptions related to future healthcare expenditure in patients with somatoform disorders? *Psychological Medicine*, 44, 2903–2911. doi:10.1017/s003329171400035x
- Gask, L., Dowrick, C., Salmon, P., Peters, S., & Morriss, R. (2011). Reattribution reconsidered: Narrative review and reflections on an educational intervention for medically unexplained symptoms in primary care settings. *Journal of Psychosomatic Research*, 71, 325–334. doi:10.1016/j.jpsychores.2011.05.008
- Ghiselli, E. E., Campbell, J. P., & Zedeck, S. (1981). *Measurement theory for the behavioural sciences*. San Francisco: WH Freeman and Company.
- Gray, S. E., & Rutter, D. R. (2007). Illness representations in young people with chronic fatigue syndrome. *Psychology & Health*, 22, 159–174. doi:10.1080/14768320600774595
- Hagger, M. S., Koch, S., Chatzisarantis, N. L. D., & Orbell, S. (2017). The common sense model of self-regulation: Meta-analysis and test of a process model. *Psychological Bulletin*, 143, 1117–1154. doi:10.1037/bul0000118
- Hagger, M. S., & Orbell, S. (2003). A meta-analytic review of the common-sense model of illness representations. *Psychology & Health*, 18, 141–184. doi:10.1080/088704403100081321
- Hahn, S. R., Thompson, K. S., Wills, T. A., Stern, V., & Budner, N. S. (1994). The difficult doctor-patient relationship: Somatization, personality and psychopathology. *Journal of Clinical Epidemiology*, 47, 647–657. doi:10.1016/0895-4356(94)90212-90217
- Hale, E. D., Tretharne, G. J., & Kitas, G. D. (2007). The common-sense model of self-regulation of health and illness: How can we use it to understand and respond to our patients' needs? *Rheumatology*, 46, 904–906. doi:10.1093/rheumatology/kem060
- Henningsen, P., Zimmermann, T., & Sattel, H. (2003). Medically unexplained physical symptoms, anxiety, and depression: A meta-analytic review. *Psychosomatic Medicine*, 65, 528–533. doi:10.1097/01.psy.0000075977.90337.e7
- Hinchey, S. A., & Jackson, J. L. (2011). A cohort study assessing difficult patient encounters in a walk-in primary care clinic, predictors and outcomes. *Journal of General Internal Medicine*, 26, 588–594. doi:10.1007/s11606-11010-1620-1626
- Hofmann, S. G., Asnaani, A., Vonk, I. J., Sawyer, A. T., & Fang, A. (2012). The efficacy of cognitive behavioural therapy: A review of meta-analyses. *Cognitive Therapy and Research*, 36, 427–440. doi:10.1007/s10608-10012-9476-9471
- Hotopf, M., David, A. S., Hull, L., Nikalaou, V., Unwin, C., & Wessely, S. (2003). Gulf War illness—better, worse, or just the same? A cohort study. *BMJ*, 327, 1370. doi:10.1136/bmj.327.7428.1370
- Hou, R., Moss-Morris, R., Peveler, R., Mogg, K., Bradley, B. P., & Belli, A. (2012). When a minor head injury results in enduring symptoms: A prospective investigation of risk factors for postconcussional syndrome after mild traumatic brain injury. *Journal of Neurology, Neurosurgery, and Psychiatry*, 83, 217–223. doi:10.1136/jnnp-2011-300767
- Hudson, J. L., Bundy, C., Coventry, P. A., & Dickens, C. (2014). Exploring the relationship between cognitive illness representations and poor emotional health and their combined association with diabetes self-care. A systematic review with meta-analysis. *Journal of Psychosomatic Research*, 76, 265–274. doi:10.1016/j.jpsychores.2014.02.004

- Hunt, S. C., Richardson, R. D., Engel Jr., C. C., Atkins, D. C., & McFall, M. (2004). Gulf War veterans' illnesses: A pilot study of the relationship of illness beliefs to symptom severity and functional health status. *Journal of Occupational and Environmental Medicine*, *46*, 818–827. doi:10.1097/01.jom.0000135529.88068.04
- Jackson, J. L., & Kroenke, K. (1999). Difficult patient encounters in the ambulatory clinic: Clinical predictors and outcomes. *Archives of Internal Medicine*, *159*, 1069–1075. doi:10.1001/archinte.159.10.1069
- Kirmayer, L. J., Young, A., & Robbins, J. M. (1994). Symptom attribution in cultural perspective. *The Canadian Journal of Psychiatry*, *39*, 584–595. doi:10.1177/070674379403901002
- Leventhal, H., Leventhal, E. A., & Breland, J. Y. (2011). Cognitive science speaks to the “common-sense” of chronic illness management. *Annals of Behavioral Medicine*, *41*, 152–163. doi:10.1007/s12160-12010-9246-9249
- MacLeod, A. K., Haynes, C., & Sensky, T. (1998). Attributions about common bodily sensations: Their associations with hypochondriasis and anxiety. *Psychological Medicine*, *28*, 225–228. doi:10.1017/s0033291797005849
- Mc Sharry, J., Moss-Morris, R., & Kendrick, T. (2011). Illness perceptions and glycaemic control in diabetes: A systematic review with meta-analysis. *Diabetic Medicine*, *28*, 1300–1310. doi:10.1111/j.1464-5491.2011.03298.x
- McAndrew, L. M., Chandler, H. K., Serrador, J. M., Quigley, K. S., Natelson, B. H., & Lange, G. (2016). Comparison of the functional health limitations of veterans deployed to Iraq or Afghanistan to veterans deployed to desert shield/storm with chronic fatigue syndrome. *Military Behavioral Health*, *4*, 299–306. doi:10.1080/21635781.2016.1175980
- McAndrew, L. M., Helmer, D., Phillips, L., Chandler, H., Ray, K., & Quigley, K. (2016). Iraq and Afghanistan veterans report symptoms consistent with chronic multisymptom illness one year after deployment. *Journal of Rehabilitation Research and Development*, *53*, 59. doi:10.1682/jrrd.2014.10.0255
- McAndrew, L. M., Martin, J. L., Friedlander, M. L., Shaffer, K., Breland, J. Y., Slotkin, S., & Leventhal, H. (2017). The common sense of counseling psychology: Introducing the common-sense model of self-regulation. *Counselling Psychology Quarterly*, 1–16. doi:10.1080/09515070.2017.1336076
- McAndrew, L. M., Musumeci-Szabó, T. J., Mora, P. A., Vileikyte, L., Burns, E., Halm, E. A., ... Leventhal, H. (2008). Using the common sense model to design interventions for the prevention and management of chronic illness threats: From description to process. *British Journal of Health Psychology*, *13*, 195–204. doi:10.1348/135910708x295604
- Moss-Morris, R. (2005). Symptom perceptions, illness beliefs and coping in chronic fatigue syndrome. *Journal of Mental Health*, *14*, 223–235. doi:10.1080/09638230500136548
- Moss-Morris, R. (2011). Are illness representations key? *The Psychologist*, *24*, 899–899.
- Moss-Morris, R., & Chalder, T. (2003). Illness perceptions and levels of disability in patients with chronic fatigue syndrome and rheumatoid arthritis. *Journal of Psychosomatic Research*, *55*, 305–308. doi:10.1016/s0022-3999(03)00013-00018
- Moss-Morris, R., Spence, M., & Hou, R. (2011). The pathway from glandular fever to chronic fatigue syndrome: Can the cognitive behavioural model provide the map? *Psychological Medicine*, *41*, 1099–1107. doi:10.1017/s003329171000139x
- Moss-Morris, R., Weinman, J., Petrie, K., Horne, R., Cameron, L., & Buick, D. (2002). The revised illness perception questionnaire (IPQ-R). *Psychology & Health*, *17*, 1–16. doi:10.1080/08870440290001494
- Moss-Morris, R., & Wrapson, W. (2003). Representational beliefs about functional somatic syndromes. In L. D. Cameraon, & H. Leventhal (Eds.), *The self regulation of health and illness behaviour* (pp. 119–137). London: Routledge.
- olde Hartman, T. C., Borghuis, M. S., Lucassen, P. L., van de Laar, F. A., Speckens, A. E., & van Weel, C. (2009). Medically unexplained symptoms, somatisation disorder and hypochondriasis: Course and prognosis. A systematic review. *Journal of Psychosomatic Research*, *66*, 363–377. doi:10.1016/j.jpsychores.2008.09.018
- Preacher, K. J., & Hayes, A. F. (2008). Asymptotic and resampling strategies for assessing and comparing indirect effects in multiple mediator models. *Behavior Research Methods*, *40*, 879–891. doi:10.3758/brm.40.3.879
- Preacher, K. J., & Kelley, K. (2011). Effect size measures for mediation models: Quantitative strategies for communicating indirect effects. *Psychological Methods*, *16*, 93–115. doi:10.1037/a0022658
- Richard, F. D., Bond Jr, C. F., & Stokes-Zoota, J. J. (2003). One hundred years of social psychology quantitatively described. *Review of General Psychology*, *7*, 331–363. doi:10.1037/1089-2680.7.4.331
- Rief, W., & Broadbent, E. (2007). Explaining medically unexplained symptoms-models and mechanisms. *Clinical Psychology Review*, *27*, 821–841. doi:10.1016/j.cpr.2007.07.005
- Salmon, P., Ring, A., Dowrick, C. F., & Humphris, G. M. (2005). What do general practice patients want when they present medically unexplained symptoms, and why do their doctors feel pressurized? *Journal of Psychosomatic Research*, *59*, 255–260. doi:10.1016/j.jpsychores.2005.03.004
- Schmidt, F. L., & Hunter, J. E. (2014). *Methods of meta-analysis: Correcting error and bias in research findings*. Los Angeles: Sage publications.
- Schmidt, F. L., & Le, H. (2004). *Software for the hunter-schmidt meta-analysis methods (versions 1.0 and 1.1)*. Iowa City: University of Iowa, Department of Management & Organizations.
- Seaton, M., Marsh, H. W., & Craven, R. G. (2010). Big-fish-little-pond effect: Generalizability and moderation—Two sides of the same coin. *American Educational Research Journal*, *47*, 390–433. doi:10.3102/0002831209350493
- Sharpe, M., Stone, J., Hibberd, C., Warlow, C., Duncan, R., Coleman, R., ... Cavanagh, J. (2010). Neurology out-patients with symptoms unexplained by disease: Illness beliefs and financial benefits predict 1-year outcome. *Psychological Medicine*, *40*, 689–698. doi:10.1017/s0033291709990717

- Spence, M. J., & Moss-Morris, R. (2007). The cognitive behavioural model of irritable bowel syndrome: A prospective investigation of patients with gastroenteritis. *Gut*, *56*, 1066–1071. doi:10.1136/gut.2006.108811
- Spence, M., Moss-Morris, R., & Chalder, T. (2005). The behavioural responses to illness questionnaire (BRIQ): A new predictive measure of medically unexplained symptoms following acute infection. *Psychological Medicine*, *35*, 583–593. doi:10.1017/s0033291704003484
- Steinmetz, D., & Tabenkin, H. (2001). The 'difficult patient' as perceived by family physicians. *Family Practice*, *18*, 495–500. doi:10.1093/fampra/18.5.495
- van Wilgen, C. P., van Ittersum, M. W., Kaptein, A. A., & van Wijhe, M. (2008). Illness perceptions in patients with fibromyalgia and their relationship to quality of life and catastrophizing. *Arthritis & Rheumatism*, *58*, 3618–3626. doi:10.1002/art.23959
- Weinman, J., Petrie, K. J., Moss-Morris, R., & Horne, R. (1996). The illness perception questionnaire: A new method for assessing the cognitive representation of illness. *Psychology & Health*, *11*, 431–445. doi:10.1080/08870449608400270
- White, K. P., Speechley, M., Harth, M., & Ostbye, T. (1999). The London fibromyalgia epidemiology study: Comparing the demographic and clinical characteristics in 100 random community cases of fibromyalgia versus controls. *The Journal of Rheumatology*, *26*, 1577–1585.
- Whittaker, R., Kemp, S., & House, A. (2007). Illness perceptions and outcome in mild head injury: A longitudinal study. *Journal of Neurology, Neurosurgery & Psychiatry*, *78*, 644–646. doi:10.1136/jnnp.2006.101105