Cognitive-behavioural and social factors do not predict recurrent secondary healthcare use in patients with fibromyalgia: a longitudinal study

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ABSTRACT

Objective. *Healthcare use in fibromyalgia (FM) is relatively high. Besides disease-related variables, cognitive-behavioural factors have been concurrently associated with healthcare use. It is unknown whether cognitive-behavioural and social factors also predict future healthcare use. The aim of this study was to identify cognitive-behavioural and social factors predicting recurrent secondary healthcare use in FM.*

Methods. *Using self-reported questionnaires, healthcare use, cognitive-behavioural, social, sociodemographic and disease-related variables including comorbidities were collected in 199 patients with FM, in a prospective longitudinal cohort spanning 18 months. Patients were recruited after receiving their diagnosis and protocolled treatment advice by a rheumatologist. Univariate and multivariate logistic regression models examined whether and which variables were predictors for recurrent secondary healthcare use. Internal validation was performed to correct for over-fit of the final multivariate model.*

Results. *Recurrent secondary healthcare use was lower than initial secondary healthcare use. Univariate analysis showed that having at least one comorbidity, depressive feelings, severe consequences of FM, low personal control and a high severity of fibromyalgia predicted recurrent secondary healthcare use. In the multivariate model, having at least one comorbidity was the only remaining predictor for recurrent secondary healthcare use.*

Conclusion. *Our results suggest that the existence of comorbidities as communicated by the patient is the strongest warning signal for recurrent secondary healthcare use in FM. There seems no value in using cognitive-behavioural and social factors for early identification of patients with FM at risk for recurrent secondary healthcare use.*

Introduction

Fibromyalgia (FM) is a chronic, musculoskeletal pain disorder of largely unknown aetiology, characterised by widespread pain and the occurrence of a wide range of symptoms including fatigue, sleep disturbance, functional disability, stiffness, cognitive impairment, and psychological distress (1, 2). FM strongly affects patient's daily functioning and work participation (3, 4).

Healthcare use, and related healthcare costs in FM are quite substantial (5-12). Research shows that healthcare use remains relatively stable at a considerably high level after diagnosis, comparable to the level before diagnosis (6, 13-15). High cost categories in healthcare are found in secondary healthcare, such as medical specialist's consultations, admissions to healthcare institutions, and multimodal rehabilitation programs (16). The repeated use of secondary healthcare resources, can reflect unsatisfactory health benefits of previous care, and likely also reflects overuse and unnecessary expenditure in healthcare.

After patients' FM diagnosis, patients are advised to attend healthcare, according to recommendations for the management of FM (17). In our clinic, patients receive an appropriate and tailored stepped-care treatment in the consecutive year after diagnosis, which in the long-term could contribute to better personal management of the pain disorder. Improved personal management of the illness seems noticeable in a reduction of the healthcare consumption (18). However, some patients with

FM recurrently use secondary healthcare, and it is useful to get insight into their characteristics. Identifying these patients at an early stage gives the opportunity to develop and apply strategies to prevent recurrent secondary healthcare use.

Cross-sectional research has consistently identified cognitive-behavioural factors (*e.g.* coping abilities, self-efficacy, and illness perceptions) associated with healthcare use and costs in FM (19-24). These factors are relevant for clinical practice, as they are modifiable targets in the intervention of patients with FM (17, 25). Promising results have been found in cognitive-behavioural interventions in chronic pain, resulting in decreased healthcare use (26). Besides cognitive-behavioural factors, social factors (*e.g.* partner and family responses) could influence patient's healthcare use (24, 27). Patient's perceived responses from the environment could aggravate or reduce their symptoms. Lack of social support is associated with mental health problems (28), and invalidation by family, and partner's solicitous and punishing responses are associated with more symptoms and limitations (28, 29). These health problems could in turn contribute to higher healthcare use.

Whether cognitive-behavioural and social factors can predict recurrent secondary healthcare is unknown. For this, longitudinal studies are needed yielding the opportunity to identify patients with FM at risk for becoming recurrent secondary healthcare users. Preventive strategies targeting these cognitive, behavioural, or social factors could be offered to patients at an early stage of the stepped care approach (*e.g.* as part of patient education in the initial management of FM) (17).

The aim of this longitudinal cohort study was to identify cognitive-behavioural and social factors predicting recurrent secondary healthcare use in FM 18 months after diagnosis.

Methods

Study design

Within the protocolised diagnosis-and-treatment approach for patients with FM, a longitudinal cohort study was set up. All patients in the cohort received a patient-tailored treatment advice. Treatment options in the protocol were referral to a general practitioner, physiotherapist, psychologist, or a multimodal rehabilitation program.

Healthcare use, and its potential predictors were examined in patients with FM, newly referred to the Sint Maartens kliniek rheumatology outpatients clinic, location Nijmegen and Woerden, the Netherlands, between December 2011 and May 2013.

In this study, the baseline and 18 months follow-up data of the prospective longitudinal cohort study were analysed.

Patients

Consecutive patients were included in the cohort after being classified as having FM by certified rheumatologists. They were 18 years or older at time of diagnosis, were able to read and write Dutch language, and gave informed consent. The Institutional Review Board of the Radboud University Medical Centre, Nijmegen concluded that the Medical Research Involving Human Subjects Act did not apply to this study (protocol number: 2011/271).

Measurements

Sociodemographic and disease-related data were collected using self-report questionnaires, including gender, age, education level (low/middle/high), employment status (yes/no), work absence (yes/no) and comorbidity (*i.e.* at least one comorbidity, yes/no). A list of 20 common comorbidities was given, including pulmonary diseases, sinusitis, cardiac diseases, high blood pressure, cardiovascular accident, stomach ulcer, chronic bowel dysfunction, diabetes mellitus, thyroid dysfunction, epilepsy, vertigo, migraine, severe skin disease, malignant disease, depression, personality disorder, anxiety disorder, attention deficit disorder, bipolar disorder and eating disorder. Respondents indicated which of each of the 20 comorbid conditions they had and could add additional comorbidities.

Outcome measure

– *Healthcare use:* Healthcare data were collected using self-reported registration forms with a 6 months recall format and comprised consultations with healthcare providers and complementary practitioners, secondary healthcare resources, and medication use (yes/no; prescription- and over-the-counter medication).

Specifically, secondary healthcare use included the following categories: 1) consultation with medical specialists, 2) diagnostic procedures, 3) admission to healthcare institutions, and 4) multimodal rehabilitation programs. Consultation with medical specialist was assessed by 3 questions: 1) whether a medical specialist was consulted (*i.e.* "In the past 6 months, did you visit a medical specialist?" [yes/no] as answering options), and if patients responded positively; 2) what kind of specialist they visited (a predefined list with 8 different specialists was used (rheumatologist, orthopaedist, cardiologist, neurologist, internist, rehabilitation physician, psychiatrist and otolaryngologist), and additional specialists could be listed), and 3) the number of visits to each medical specialist they consulted. Diagnostic procedures, admission to and treatment in healthcare institutions, were assessed in the same way as the consultation with a medical specialist (*i.e.* use or non-use, and if indicated the type of use and quantity of use). For the diagnostic procedures, the predefined list comprised of 5 different diagnostic procedures (blood sample, x-ray, ultrasound scan, MRI, CT-scan), and additional diagnostic procedures could be listed.

Primary outcome was recurrent secondary healthcare use at 18 months follow-up defined as the use or non-use for each of the 4 categories of secondary healthcare.

Potential predictors

Besides sociodemographic and disease-related variables (*i.e.* comorbidities, severity of FM, depressive and anxious symptoms), potentially relevant cognitive-behavioural and social predictors for recurrent secondary healthcare use were collected using existing and validated questionnaires. Specifically, data were collected on illness perceptions of FM, generic ill-

ness cognitions (*i.e.* helplessness, acceptance, and perceived benefits), pain coping, coping flexibility, invalidation by family, spousal responses to patient pain behaviours and well behaviours (see Supplementary Table I, which provides detailed information about the questionnaires administered).

Severity of fibromyalgia
The Fibromyalgia Impact Questionnaire (FIQ) is a 10-item instrument for assessing health status in FM (30). In this study the total FIQ score was used. The Cronbach's alpha was 0.82.

Illness perceptions
Illness perceptions of FM were assessed with the revised Illness Perception Questionnaire (IPQ-R-FM), a valid and reliable instrument for measuring illness perceptions in patients with FM (31). The following 7 dimensions were included: acute/chronic timeline, cyclical timeline, consequences, personal control, treatment control, illness coherence and emotional representation. In this study the Cronbach's alpha ranged from 0.74 to 0.87 for the 7 dimensions.

Anxiety and depression
The Hospital Anxiety and Depression Scale (HADS) is a 14-item widely used self-report screening instrument to assess levels of anxiety and depression in an medical out-patients clinic (32). It has shown good sensitivity and specificity for identification of anxiety disorders and depression in various clinical populations (33). In this study the Cronbach's alpha for the anxiety subscale and depression subscale were 0.81 and 0.82 respectively.

Illness cognitions (helplessness, acceptance, and perceived benefits)
Illness cognitions were assessed with the Illness Cognition Questionnaire (ICQ), a generic 18-item instrument assessing different ways of cognitively (re)evaluating the inherently aversive character of a chronic disease. The ICQ comprises 3 subscales helplessness, acceptance and perceived benefits (34). The ICQ has good psychometric properties in rheumatic diseases (34). In this study Cronbach's alpha for helplessness, acceptance and perceived benefits were 0.86, 0.89, and 0.85 respectively.

Active and passive pain coping
Pain coping was assessed with the Pain Coping Inventory (PCI), a 33-item questionnaire comprising 6 scales: pain transformation, distraction, reducing, retreating worrying and resting (35). The PCI scales can be grouped into active (transformation, distraction, reducing demands) and passive (retreating, worrying, resting) pain coping dimensions. In this study the Cronbach's alpha ranged from 0.70 to 0.79 for the 6 dimensions. The composite standardised mean score for active and passive pain coping was used in the statistical analyses.

Coping flexibility
Coping flexibility was assessed with the Coping Flexibility Questionnaire (COFLEX), a 13-item questionnaire comprising 2 subscales: versatility and reflective coping (36). Versatility assesses the ability to flexibly use a variety of coping strategies in accordance with personal goals and changing circumstances. Reflective coping assesses the ability of generating and considering coping options, and appraising the suitability of a coping strategy in a given situation. Reliability and preliminary validity has been reported to be adequate (36). In this study Cronbach's alpha for versatility and reflective coping were 0.93 and 0.81 respectively.

Invalidation by the family
Invalidation by the family was assessed with the 8-item family scale of the Illness Invalidation Inventory (3*I) (37). The 3*I measures the occurrence of invalidation by 5 different sources (spouse, family, medical professionals, work environment, and social services). The family scale comprises 2 subscales: discounting and lack of understanding. The 3*I has good validity, internal consistency, and measurement invariance (28, 37). In this study the Cronbach's alpha for discounting and lack of understanding were 0.89 and 0.83 respectively.

Spousal responses to pain behaviour and well behaviour
The Spouse Response Inventory (SRI) assesses spousal responses to patient pain and well behaviour. Responses to patient pain behaviour comprises 2 subscales: solicitous responses to pain behaviour and negative response to pain behaviour.
Responses to patients well behaviours comprises 2 subscales: facilitative responses to well behaviour and negative responses to well behaviour. The SRI has shown good validity and reliability (29, 38). In this study Cronbach's alpha's ranged from 0.87 to 0.92 across the four dimensions.

Spouse solicitous, punishing and distracting responses to pain behaviour
To assess spouse solicitous, punishing and distracting responses to pain behaviour, the 14-item 'Significant Other Response Scale' of the West Haven-Yale Multidimensional Pain Inventory (WHYMPI) was used (39, 40). The Significant Others Response Scale of the WHYMPI has good validity and internal consistency (39). In this study the Cronbach's alpha for the punishing subscale, the solicitous subscale, and the distracting subscale were 0.83, 0.79, and 0.65 respectively.

Statistical analyses
Descriptive statistics were computed for sociodemographic and disease-related variables at baseline. Based on total FIQ score, patients were categorised into severity groups according to established cut-off points (41). Complete-case analysis was chosen as less than 5% of the patients had missing values on one or more variables. Prior to constructing and validating the prediction models, relevant assumptions were tested (42). As a rule of thumb 10 to 15 cases per predictor variable in logistic regression analyses will suffice for a regression model. Potential differences in baseline characteristics of responders and non-responders were examined using *t*-tests. Healthcare use of patients with FM (yes/no) was computed at baseline and 18 months follow-up (Table I). The McNemar test was used to compare baseline and follow-up healthcare use. The

number of visits (if at least one) to the healthcare providers between baseline and follow-up was compared using the Wilcoxon signed-rank test (Table I).

Secondary healthcare use was, first, summed separately for each of the 4 categories of secondary healthcare. This resulted in a total score and if applicable a range for each category (Table I). Subsequently, the data of these categories were dichotomised (0 = not using healthcare from this category, and 1 = using healthcare from this category at least once). A patient was considered a recurrent secondary healthcare user, if secondary healthcare from at least one of the 4 categories was used in the past 6 months. Thus, the primary outcome measure being a dichotomised variable (0 = non-secondary healthcare user at follow-up, and 1 = recurrent secondary healthcare user).

In the main analyses, first, in order to examine the association between the potential predictors (*i.e.* sociodemographic variables, disease-related variables, cognitive-behavioural and social variables) and primary outcome, univariate logistic regression analyses were conducted. Subsequently, variables with $p<0.157$ (*i.e.* Akaike information criterion) (42) in the univariate association model were entered into the multivariate regression model. Second, multivariate regression modelling with backward selection was conducted. Third, to internally validate the final prediction model, a bootstrapping technique (500 samples) was used in order to check for overfitting of the model (43) and the regression coefficients (*i.e.* the slope value) of the final model were adjusted, if appropriate. The area under the curve was calculated to check the discriminative ability of the model (*i.e.* C-index). Furthermore, the Hosmer en Lemeshow goodness-of-fit test was calculated to check the overall performance of the model. A *p*-value 0.05 was considered to be statistically significant. Statistical analyses were performed using Stata 13.0 and R Studio (v. 1.0.143).

Table I. Health care use (HCU) at baseline and at 18 months follow-up in patients with fibromyalgia (n=199).

Health care use	Baseline		18 months follow-up	
	HCU in past 6 months n (%)	Number of visits, if at least one Median (p25-p75)	HCU in past 6 months n (%)	Number of visits, if at least one Median (p25-p75)
Secondary Health Care	196 (98)		122 (61)	
Medical specialist	183 (92)	2 (1-4)	85 (43)#	2 (2-5)
Admission Health Care Institution	16 (8)		11 (5)	
Multimodal rehabilitation program	17 (9)		14 (7)	
Diagnostic Procedures	165 (83)		105 (53)#	
Miscellaneous Care				
General Practitioner	183 (92)	4 (2-6)	136 (68)#	3 (2-5)#
Health professional*	129 (65)	11 (6-19)	110 (55)#	11 (5-24)
Complementary Practitioner	54 (27)	5 (3-10)	42 (21)	5 (3-8)
Medication use	188 (94)		177 (89)#	

*Health professional included: physical therapist, manual therapist, exercise therapist, occupational therapist, psychologist, social worker, (psychosomatic) nurse, podiatrist, and dietitian. #Significant difference ($p<0.05$) between baseline and follow-up.

Results

Sample

Of the 280 patients returning the baseline questionnaire, 199 patients (71.1%) completed follow-up measurements at 18 months. Most frequent dropout reasons were lack of energy or concentration (39%), and lack of time (27%); several patients did not specify a reason (21%). Responders did not differ from non-responders on any of the baseline characteristics. After diagnosis confirmation at baseline, 38% (n=76) of the patients were advised to receive further treatment in primary care, and the remaining 62% (n=123) of the patients were advised to follow a multimodal rehabilitation program. Participants were mostly female n=190 (95%), with a mean age of 43 (range 18–72); 106 participants (53%) were employed, and 109 participants (55%) finished middle or high education. Twenty four patients were classified with mild complaints (12%), 78 patients (39%) with moderate complaints and 97 patients (49%) with severe complaints of FM at baseline. Between baseline and follow-up the number of patients with at least one comorbidity decreased significantly, from 153 (77%) to 134 (67%).

Healthcare use

In general, the number of patients that used healthcare decreased between baseline and 18 months follow-up (Table I). Significantly fewer patients visited healthcare providers, and less patients used medication and had diagnostic procedures carried out, at 18 month follow-up compared to baseline. However, if patients did visit one of the healthcare providers, the median number of visits did not change between baseline and follow-up, except for the visits to the general practitioner where a small decrease in visits was noticeable.

Predictors of recurrent secondary healthcare use

Table II shows the results of the univariate and multivariate logistic regression analyses. Of the investigated predictors of recurrent secondary healthcare use, 8 predictors were included in the multivariate model : comorbidity, severity of FM, anxiety, depression, consequences of FM, personal control, helplessness, and active coping. See Table, Supplementary file 2 for the results of the univariate regression analyses with all potential predictors of recurrent secondary healthcare use.

In the final (multivariate) model, comorbidity was the only remaining significant variable predicting recurrent secondary healthcare use (odds ratio = 2.47, CI: 1.22–5.01). After correction for overfitting, the explained variance (Nagelkerke's R^2) of the model was 4%.

Discussion

This study is one of the few longitudinal studies into healthcare use in pa-

Table II. Univariate (top) and multivariate (bottom) logistic regression analyses of predictors of recurrent secondary health care use.

Univariate logistic regression analyses*		
	odds ratio [95% CI]	*p*
Disease-related variables		
Comorbidity	2.60 [1.33 – 5.10]	0.005
Severity of fibromyalgia	1.02 [1.00 – 1.04]	0.04
Mood		
Anxiety	1.06 [0.99 – 1.14]	0.09
Depression	1.10 [1.02 – 1.19]	0.02
Cognitive-behavioural variables		
Illness perceptions		
Consequences	1.08 [1.01 – 1.16]	0.02
Personal control	0.91 [0.83 – 0.99]	0.03
Pain coping		
Active coping	0.84 [0.71 – 1.00]	0.05
Cognitions		
Helplessness	1.08 [1.00 – 1.17]	0.05
Multivariate logistic regression analyses‡		
	odds ratio [95% CI]	*p*
Comorbidity	2.47 [1.22 – 5.01]	0.01
Model Performance	Model	Corrected
Explained variance (Nagelkerke's R^2)	0.12	0.04
c-index	0.67	0.62
Calibration		
Hosmer and Lemeshow	X^2 (8)= 6.88	$p = 0.55$
Slope value	0.70	

*Only predictors according to the Akaike information criterion ($p<.157$) were selected.
‡Only statistically significant predictors ($p<.05$) are displayed.

tients with FM. A decrease of (secondary) healthcare use was observed over a period of 18 months. The investigated cognitive-behavioural and social factors such as illness and cognitions, coping styles, perceived spousal responses to pain behaviours, and illness invalidation, after taking demographic and disease-related variables such as severity of FM and mood, did not significantly predict recurrent secondary healthcare use in FM, 18 months after diagnosis. Only having at least one comorbidity turned out to be a predictor of recurrent secondary healthcare use.

In agreement with previous studies that demonstrated a concurrent and longitudinal association between comorbidity and healthcare costs (10, 22, 44), the current study shows that comorbidity predicts recurrent secondary healthcare use in patients with moderate to severe FM. A detailed look into comorbidities reported at baseline revealed that the vast majority of comorbidities were symptoms attributable to FM, such as chronic bowel dysfunction, anxiety and depressive symptoms and migraine. Patients reporting one or more comorbidities seem to reflect a subgroup of patients with more symptomatology. Additional diagnostic testing as a means to reassure the patient is most often of no added value, and might even lead to an increase instead of decrease of worries and undesirable healthcare costs (45, 46). An implication of this study is that a reported comorbidity could be used in general practice or rheumatology as a warning signal that the patient is at risk for becoming a recurrent healthcare user. Provision of adequate education about FM symptomatology including the comorbidities commonly observed in this population by a general practitioner or specialised nurse after FM diagnosis is warranted.

None of the hypothesised modifiable psychosocial factors turned out to be unique predictors of recurrent secondary healthcare use at 18 months follow-up. In cross-sectional research, cognitive-behavioural factors (20, 47) have been found to be associated with healthcare use, and social factors (28, 29) have been found to be associated with health outcomes. The current study does not disconfirm the existence of a cross-sectional relation indicating that cognitive-behavioural and social factors may affect the intensity of healthcare use and should be a included in education and management of FM. However, although targeting these cognitive-behavioural and social factors may reduce symptoms in patients with FM, the findings in this longitudinal study suggest that there is no value in using these factors in tools for early identification of patients with FM at risk for recurrent secondary healthcare use.

Determinants of healthcare use can be distinguished on 3 levels: personal level, the healthcare provider level, and the level of the healthcare system (48). The observation that none of the personal factors examined in this cohort study contributed to recurrent secondary healthcare use, does not rule out the importance of other factors. Using health-care or not is the result of a mutual, shared decision making process between the healthcare provider and the patient. Therefore, in the actual use of secondary healthcare, there is likely a role for the general practitioner who in some countries refers the patient to secondary healthcare (49). The influence of the referral policy on secondary healthcare use was not investigated in this study.

The findings should be interpreted in light of the strengths and limitations of this study. Using a prospective cohort of patients with a relatively large sample size, measuring a wide range of potentially modifiable predictors to healthcare use over a longer period of time are strengths of this study. A first limitation of this study is that it remains unclear if these patients followed their advised treatment, and how this (non) adherence could influence recurrent secondary healthcare use. A second

limitation might be the operationalisation of the primary outcome measure. We choose to compose one measure out of 4 categories of secondary healthcare *i.e.* 1) consultations with medical specialist, 2) diagnostic procedures, 3) admission to healthcare institutions, and 4) multimodal rehabilitation programs. A patient was considered a recurrent secondary healthcare user, if secondary healthcare from at least one of the 4 categories was used in the past 6 months. Although this is a face valid definition of recurrent secondary healthcare use, it needs further empirical support and replication in future studies. A third limitation was the use of patient-reported questionnaires to measure healthcare use and comorbidity. Although self-report methods are prone to response bias, such as social desirability and recall bias, they also reflect the subjective perception of patients that was indicated to be more crucial for health in FM than objective observations (50). Although validation of patient-reported data against data from medical or administrative records is a preferred method, research shows a good concordance between self-reported and registered utilisation of healthcare (51).

For future research we suggest that recurrent secondary care users should be questioned on their reasons for returning to secondary healthcare. Perhaps revealing other potentially relevant variables for recurrent secondary healthcare use not investigated in this study. Additionally, future studies should examine the referral policy of the general practitioner in patients with FM, to clarify potential healthcare provider predictors of secondary healthcare use.

Overall, our results suggest that the existence of comorbidities as communicated by the patient is the strongest warning signal for to recurrent secondary healthcare use in FM. While cognitive-behavioural and social factors have been shown to be associated with concurrent healthcare use or health outcomes, there is no value in using these variables for early identification of patients with FM who have an additional risk at recurrent secondary healthcare use.

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