Persistent medically unexplained symptoms in primary care

The patient, the doctor and the consultation

Tim C. olde Hartman

Colophon

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Promotor:	Dhr. prof. dr. C. van Weel
Copromotoren:	Dhr. dr. P.L.B.J. Lucassen Dhr. dr. H.C.A.M. van Rijswijk
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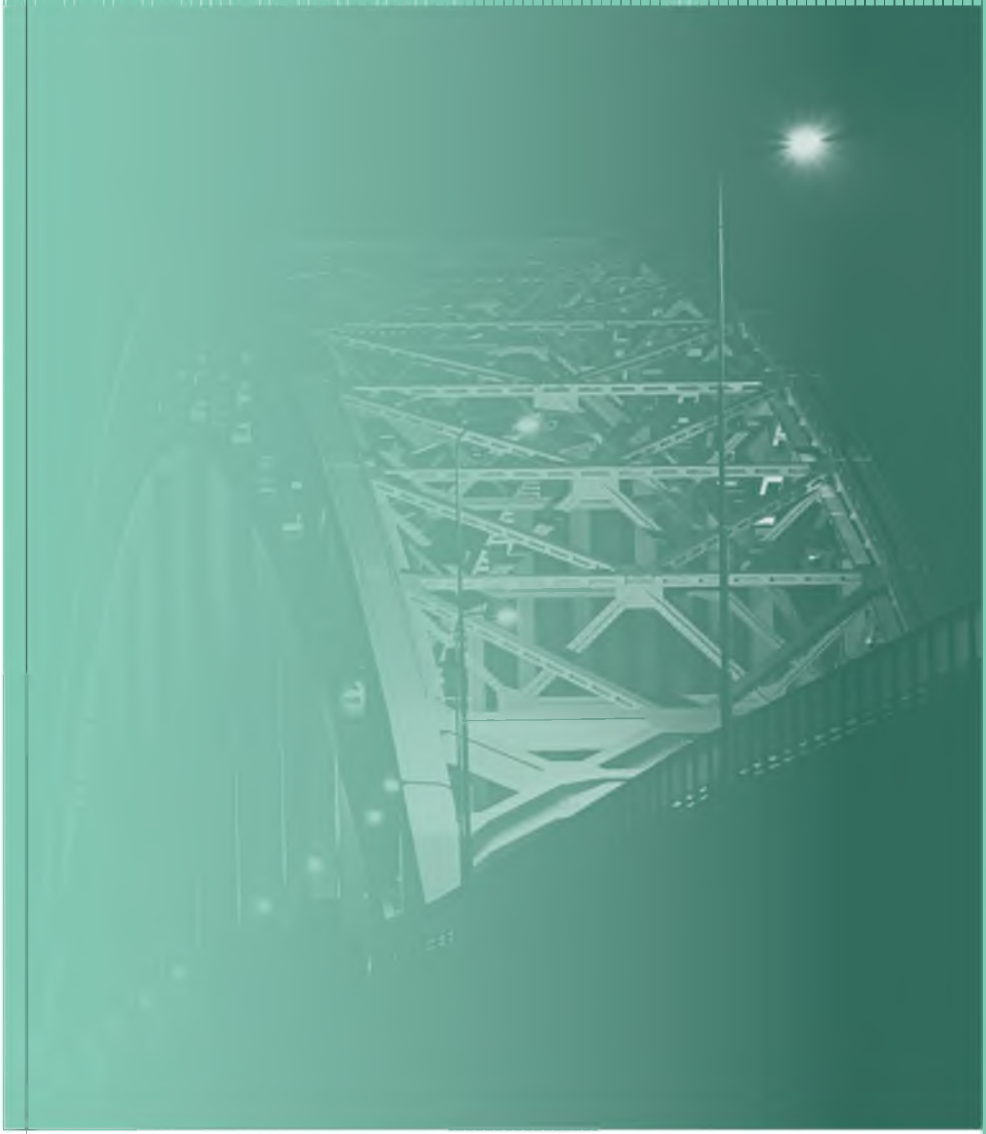
GENERAL INTRODUCTION



Tim olde Hartman

This chapter is based on:
Somatic fixation. New life for an old concept [Somatische fixatie. Een nieuw leven voor een oude begrip]
P.L.B.J. Lucassen, T.C. olde Hartman, M. Borghuis
Huisarts en Wetenschap 2007; 50(1):11-15

Medicine is not only a science; it is also an art. It does not consist of compounding pills and plasters; it deals with the very processes of life, which must be understood before they may be guided. (Paracelsus (c. 1493-1541))

Background

Physical symptoms such as headache, back pain, dizziness and fatigue are common in the general population. Two thirds of men and four fifth of women report at least one physical symptom in the last two weeks.[1,2] However most people do not contact professional medical care for these symptoms.[3,4] When people contact their General Practitioner (GP) for these symptoms 80% remain restricted to one doctor-patient contact,[5] suggesting that most of these symptoms are transient.

In about 25-50% of all symptoms presented in primary health care, no evidence can be found for an underlying physical disease, and should be considered as medically unexplained symptoms (MUS).[6,7] In specialist care these percentages are even higher, ranging from 30-70%.[8,9] MUS usually disappear spontaneously in the course of time or as a consequence of the physician's management. A recent Dutch study found that only 2.5% of the patients in general practice presenting with medically unexplained symptoms meet criteria for chronicity.[10] However, this minority represent a major problem in health care for the following reasons:

- **a.** patients with *persistent* MUS suffer from their symptoms, are functionally impaired, and are at risk for potentially harmful additional testing and treatment;
- **b.** these patients are a burden for the health care system as they are responsible for high, often unnecessary, health care costs;
- **c.** patients with *persistent* MUS are often dissatisfied with the medical care they receive during their illness;
- **d.** these patients often cause feelings of frustration and irritation in their physicians; and
- **e.** the suitability, applicability and effectiveness of specific interventions towards patients with *persistent* MUS in primary care are limited.

The problems described above are at the basis of the studies reported in this thesis, which together aimed at gaining more insight into the care patients with persistent MUS expect *(the patient)*, the way GPs experience the care they deliver for these patients *(the doctor)* and the care GPs deliver during encounters with these patients *(the consultation)* in order to guide new intervention strategies for these patients.

Confusing terms

Definitions in literature

In the scientific literature there is discussion and debate for the best term to describe physical complaints of patients when the aetiology is unclear. Lipowski defined *somatisation* as the tendency to experience and communicate psychological distress in the form of somatic symptoms that the patient misinterprets as signifying serious physical illness.[11] In DSM-IV *somatisation disorder* is defined as a history of many physical complaints (four pain symptoms, two gastrointestinal symptoms, one sexual symptom and one pseudoneuro-logical symptom) beginning before the age of 30 that occur over a period of several years. Escobar introduced the concept of *abridged somatisation*, a construct for less severe forms of somatisation characterized by four or more unexplained physical symptoms in men and six or more unexplained physical symptoms in women.[12] In *functional somatic syndromes* a set of unexplained symptoms is clustered into a syndrome. In this way each medical specialty has his own functional somatic syndrome. For example Chronic Fatigue Syndrome (CFS) in internal medicine, Irritable Bowel Syndrome (IBS) in gastroenterology and Fibromyalgia (FM) in rheumatology. In Nijmegen the term *chronic nervous functional somatic symptoms* has been developed.[13] Patients meet this diagnostic category when they repeatedly present physical symptoms that remain medically unexplained after adequate examination, in combination with the (presumed) presence of psychosocial problems or psychological distress. This diagnostic category has been included in the Continuous Morbidity Registry (CMR) system which was introduced by the Nijmegen Department of Family Medicine in 1971.[13-16] To our knowledge, the CMR is the only classification system with a separate category for patients with persistent MUS. In a further elaboration of chronic nervous-functional symptoms, the theory of 'somatic fixation' was developed. *Somatic fixation* is the consequence of continuous one-sided emphasis on the somatic aspects of symptoms and health problems resulting in people becoming more and more entangled in and dependent of health care.[17;18] This theory explicitly acknowledges the role of the physician in the development of persistent MUS.

Which term will we use in this thesis?

A number of terms and definitions are used for persistent symptoms without obvious pathology. *Somatisation* has been criticized, as it suggests that physical symptoms originate from psychosocial distress. Furthermore this term assumes pathogenic processes and tendencies, such as illness behaviour, on the part of the patient, without considering at all the role of the physician.[19] Concerning *functional somatic syndromes*, researchers argue that the existing definitions of these individual syndromes are of limited value because: (1) the substantial overlap between the individual syndromes and (2) the similarities between them outweigh the differences.[20] Furthermore, there is some confusion regarding its core concept as it may refer to a functional disturbance of organs or the brain systems or it may refer to the function of symptoms within the framework of secondary gain.[21] This also applies for the term *chronic nervous-functional somatic symptoms*. It has been criticized for suggesting (1) the translation of mental distress into physical symptoms, (2) the implication that patients hold on to somatic attribution of symptoms, and (3) a 'function' of the symptoms in expressing psychological distress.[22] However, although doctors may think the term 'functional' is pejorative, patients do not perceive it as such.[23] The term *persistent medically unexplained symptoms* has gained popularity during recent years among general practitioners. Although it defines patient's symptoms by what they are not, rather than by what they are, and although it reflects dualistic thinking, this term is purely descriptive and the most neutral as it does not indicate an underlying causal mechanism or interpretation.[24] However, it has negative connotations for patients.[23]

These various terms and definitions reflect the difficulties in the concept of unexplained bodily complaints which is not unequivocally defined.[25] As a consequence of conceptual problems doctor-patient communication and symptom explanation during consultation is hampered.[23] Furthermore, it complicates research in this population as a appropriate selection of the study population, a accurate definition of reference standards and a useful definition of outcome measures are lacking.[26]

In the light of the foregoing, we decided to use the term *persistent medically unexplained symptoms* (persistent MUS) in this thesis, because we think this is the most neutral term and because this term is widely accepted in today's scientific community and in primary care.

The patient, the doctor and the consultation

In 1979 researchers of the department of Family Medicine of the Radboud University Nijmegen Medical Centre in the Netherlands started a study of somatic fixation.[17] This study culminated in the book '*To heal or to harm. The prevention of somatic fixation in general practice*' by Richard Grol et al.[18] The concept of somatic fixation connected a theory of health and illness with consultation skills. Somatic fixation assumes a tendency within patients to persistently experience symptoms, frequently as a consequence of psychological problems. Patients who are inclined towards somatic fixation also tend to inadequate help seeking behaviour and dependence on health care professionals. The result of these tendencies is that the patient focuses on his or her body and denies the relation between bodily symptoms and psychosocial problems.

According to that theory, the GP has powerful tools to prevent somatic fixation and with it persistent MUS. These tools – consultation and communication skills – comprise a goal-oriented and systematic approach, effective management of the doctor-patient relationship and adequate treatment of both somatic and psychosocial symptoms. However, despite the use of these tools, a small group of patients ends up with persistent MUS. Research on the management by GPs of those functionally impaired and suffering patients and regarding the care these patients expect is still limited.

Elaborating on the theory of somatic fixation and the role of the patient, the doctor and the consultation we will (1) explore the knowledge regarding the problems arising when GPs meet patients who have developed persistent MUS in daily practice, and (2) indicate knowledge gaps regarding the care GPs deliver and the care patients expect when encountering with persistent MUS.

The patient

Patients with persistent MUS often have the feeling that doctors do not acknowledge the legitimacy of their symptoms, and that they constantly have to oppose their doctor's skepticism.[27] Many patients feel that their doctors label them as 'psychological cases'.[28;29] Furthermore, patients have the feeling that GPs don't take them seriously because GPs often tell them 'it is nothing' or 'you do not have a disease'.[27;30-33]

Patients with distinct functional syndromes are often dissatisfied with the medical care they receive during their illness.[28;34] They sense the need to fight to gain recognition and acceptance from their GP. Often, these patients consider their doctor incompetent and themselves as experts regarding their own symptoms.[35] Patients with persistent MUS use strategies to keep up medical attention when they meet an atmosphere of distrust in the consultation. These include somatizing (i.e. persisting in bodily explanations), claiming under cover (i.e. referring to other authorities such as TV or a neighbour who is a doctor), and pleading (crying and begging) to catch the doctor's interest.[32;36]

Which care do patients with persistent MUS expect?

The frequently expressed dissatisfaction by patients with the medical care received during illness might originate from the mismatch between what patients want and what they actually receive from their GP. Analysis of videotaped consultations showed this mismatch clearly during the *initial* presentation of MUS in primary care. Salmon et al. showed that patients with MUS wish to have a convincing, legitimating and empowering explanation for their symptoms which, unfortunately, is not given by their GP.[37-40] Furthermore they indicated that they want more emotional support from their GP.[30] However, research towards preferences of patients with *persistent* MUS regarding the care they expect and receive in primary care is still lacking.

The doctor

Research pointed out that doctors often experience patients with persistent MUS as difficult to manage.[27;41] Furthermore, they indicate that effective management strategies are lacking.[42] In fact, many GPs think that persistent MUS are associated with personality or psychiatric disorders.[42;43] Furthermore, many doctors regard persistent MUS as an expression of psychological distress with patients failing to see the connection between the physical symptoms and the psychological distress.[41;44] According to many GPs, the physical symptoms are not the real problem.[44] In addition, many doctors remain skeptical about physical symptoms that cannot be explained by a physical disease.[45] Compared with patients with 'real' diseases, patients with persistent MUS do not have much prestige in the medical arena.[46] In fact, research has shown that doctors' judgments regarding the intensity of pain felt by patients is associated with the presence or absence of supporting medical evidence.[47] When there is objective medical evidence for the pain, doctors were more inclined to accept the patient's claim regarding the pain intensity. Furthermore, doctors tend to believe that there is incongruence between the presentation of MUS and the actual burden in this case.[45]

When facing patients with persistent MUS, many GPs feel pressurized to offer somatic interventions.[44;48] This feeling of being pressurized has been widely attributed to patients' belief that symptoms are caused by physical disease and to their rejection of psychological help.[42;49]
In conclusion, GPs' subjective feelings of being pressurized for somatic interventions as well as GPs' skepticism regarding persistent MUS helps to explain their dissatisfaction with consultations with patients with persistent MUS and the widespread labelling of them as 'heartsink' or 'difficult' patients.[42;50-53] However, despite this frustration and skepticism, many GPs believe that patients with persistent MUS should be managed in primary care as providing reassurance, counselling and acting as a 'gatekeeper' to prevent inappropriate investigations are considered important roles for GP management.[42]

How do GPs experience the care they deliver for these patients ?

As can be expected from the aforementioned difficulties with patients with persistent MUS, GPs experience difficulties in their management of these patients. Previous studies revealed that these difficulties are mainly associated with the communication and the doctor-patient relationship with these patients.[27;45;54-59] However, most studies did not focus specifically on GPs' experiences of the care they deliver to patients with persistent MUS. As patients with persistent MUS will keep attending the consulting hours of their GPs with their symptoms, GPs have to find strategies to specifically deal with the communication and the doctor-patient relationship during the encounters with these chronic patients. Unfortunately, however, knowledge about these strategies and GPs' experiences, which are built over time during the continuing process of GPs' health care delivery for these patients, is still lacking.

The consultation

A number of studies regarding the doctor-patient communication during consultations with MUS patients have been published.[38;60;61] The results of these studies are important because they demonstrate that GPs communicate with MUS patients more poorly than previously thought.

In contrast to consultations with patients with explained symptoms, GPs did explore the symptoms, feelings, concerns, opinions and expectations of the patient less adequately in consultations with patients with MUS. It seems that the doctor-patient communication in patients with MUS is less patient-centred compared to patients with explained symptoms. Although patient-centred communication is of major importance, the results demonstrated that doctors communicated inadequately in precisely those consultations where patient-centred communication is most desired and advantageous.[38]

These studies also showed that patients with MUS did not request somatic interventions more frequently than patients with medically explained symptoms. Additionally, patients with MUS did not ask for an explanation or require reassurance more often than patients with medically explained symptoms. These findings refute the GPs' subjective feeling of being pressurized. The only difference between patients with explained symptoms and patients with unexplained symptoms was that patients with MUS desired more emotional support from their GP.[60] However, GPs were less empathic toward these patients. Furthermore, in consultations with patients with MUS, GPs are more inclined to offer a prescription than that patients asked for a prescription themselves (70% versus 58%). Similar results were found with respect to proposals for additional tests (35% versus 13%) and suggestions for referrals (20% versus 14%).

During most consultations (more than 95%) with patients with MUS, patients indicated one or more psychosocial problems. Furthermore they suggested that the problem(s) may cause or influence their symptoms. However, most GPs do not seem to respond to these cues.[62;63]

What care do GPs deliver during encounters with patients with persistent MUS?

Doctor-patient communication in patients with MUS is a complex phenomenon. Most studies described above studied the doctor-patient interaction during the *initial* presentation of MUS and did not focus on patients with *persistent* MUS.[38;60-61] However, in general most of the time MUS are transient and improve without further interventions after one or two consultations. When symptoms evolve into a chronic and disabling condition (i.e. *persistent* MUS), encounters as well as doctor-patient communication become more complicated. However, knowledge of the doctor-patient communication in patients with *persistent* MUS is still lacking.

Rationale for this thesis

As described above, there is already substantial knowledge regarding the problematic interaction when GPs meet patients during the presentation of MUS. However most of this research did not focus specifically on patient with *persistent* MUS. The initial presentation of

MUS does not represent a large problem for health care, as most of these symptoms are transient and have a good prognosis. The main problem with MUS patients rises when these symptoms develop towards a chronic condition in which patients *persistently* present MUS to their GP. From this point on the GP has to find a way to manage these patients in order to improve patients' subjective well-being, symptom reduction and quality of life, to prevent potential harmful investigations and referrals, and to prevent GPs own dissatisfaction and frustration with these patients.[7;38;64;65]

To improve the quality of care for patients with *persistent* MUS, we need insight into the care GPs deliver and the care patients expect. This will lead to a better understanding of strategies for the management of patients with persistent MUS by GPs. Furthermore, given the limited suitability, applicability and effectiveness of specific interventions in primary care (such as anti-depressants, cognitive behavioural therapy and reattribution therapy) for patients with *persistent* MUS,[66-70] knowledge of patients opinions, GPs' views and doctor-patient communication might guide new intervention strategies for patients with *persistent* MUS in primary care and enhance the satisfaction of GPs as well as patients while encountering daily GP practice.

Objectives of this thesis

The aim of this thesis was to obtain more insight in what happens when GPs encounter patients with *persistent* MUS and which problems then arise. Specifically we were interested in the care GPs deliver and the care patients expect when visiting the GP with *persistent* MUS. Therefore, we designed a study to evaluate the three essential parts in the care for these patients: the patient, the doctor and the consultation.

Regarding *the patient* we aimed to answer the following research questions:

1. What are the characteristics of patients who present persistent MUS in primary care?
2. What is the course of MUS and which factors influence its course?
3. What are patients' opinions about encounters in which they present persistent MUS?

Regarding *the doctor* we aimed to answer the following research questions:

1. What are GPs' perceptions about encounters with patients with persistent MUS?

Regarding *their consultations* we aimed to answer the following research questions:

1. How do GPs talk to patients with persistent MUS during encounters in which patients present persistent MUS?

We furthermore studied the literature aiming to find *starting points for improving the management* of patients with persistent MUS. We therefore aimed to answer the following research questions:

1. What are, according to experts in the field, important and effective elements in the treatment of MUS in primary care?
2. Which explanatory models for MUS are described in the literature?

Outline of this thesis

The patient

In Chapter 2 we present data about comorbidity, referrals, diagnostic tests, and hospital admissions over a period of 10 years prior to the diagnosis chronic functional somatic symptoms in four general practices participating in the Continuous Morbidity Registry (CMR) of the university of Nijmegen.

Chapter 3 contains a systematic review and best-evidence synthesis of the literature on the course of MUS, somatisation disorder, hypochondriasis, and related prognostic factors.

Chapter 4 includes patients' opinions about encounters in which they present persistent MUS. As we consider the doctor-patient relationship as a key factor in the management of patients with persistent MUS, we specifically explored patients' opinions about the doctor-patient relationship.

The doctor

Chapter 5 describes GPs' perceptions about encounters with patients with persistent MUS. We focused on perceptions about explaining MUS to patients and GPs' perceptions about how relationships with these patients evolve over time in daily practice.

The consultation

Chapter 6 describes a consultation with a patient with unexplained palpitations during vacuuming which changed my personal communication skills during encounters with patients with persistent MUS.

Chapter 7 provides insights in how GPs talk to patients with persistent MUS during their encounters. We focused primarily on GPs' exploration of patients' symptoms and problems and GPs distribution of the available time on different stages in the persistent MUS consultations.

Starting points for improving the management

In Chapter 8 we present the results of a qualitative analysis of narrative reviews and scientific editorials to explore experts opinions regarding effective management strategies for patients with MUS.

Chapter 9 provides an overview of explanatory models (i.e. models of explaining the nature of the symptoms) of MUS described in the scientific literature that may be of use in daily general practice.

Chapter 10 contains the results of a symposium and workshop on MUS in primary care held at the Wonca World Conference 2007 in Singapore. During this meeting we focused on detecting knowledge gaps in MUS and establishing priorities in MUS research.
In Chapter 11 the results of this thesis are critically reviewed, and recommendations for clinical practice, medical education and future research are given.

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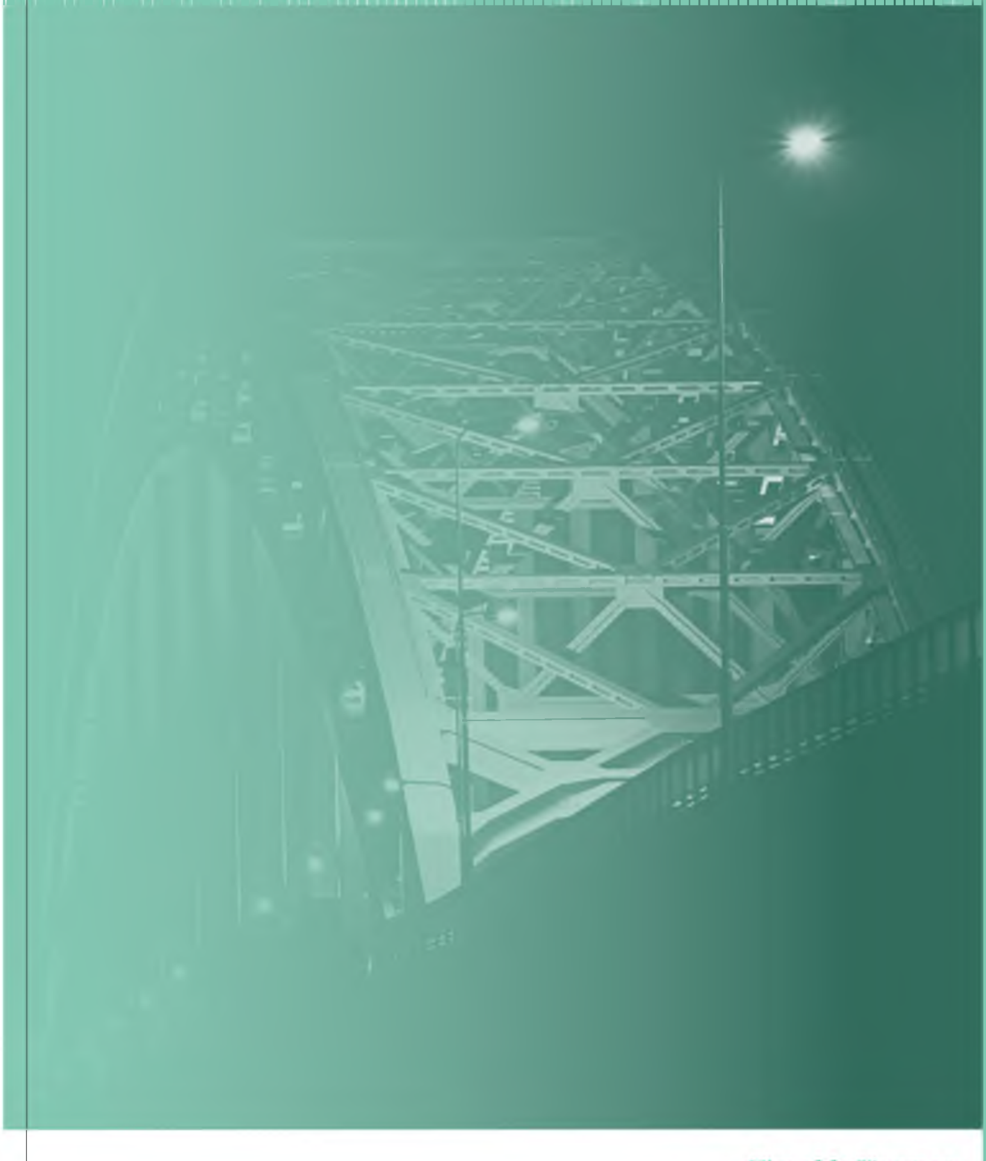
CHAPTER 2

CHRONIC FUNCTIONAL SOMATIC SYMPTOMS: A SINGLE SYNDROME?



Tim olde Hartman
Peter Lucassen
Eloy van de Lisdonk
Hans Bor
Chris van Weel

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Abstract

Background. Reliable longitudinal data of patients with functional somatic symptoms in general practice are lacking.

Aim. To identify distinctive features in patients with chronic functional somatic symptoms, and to determine whether these symptoms support the hypothesis of the existence of specific somatic syndromes.

Design. Observational study, with a comparison control group.

Setting. Four primary care practices affiliated with the University of Nijmegen in The Netherlands.

Methods. One hundred and eighty-two patients diagnosed between 1998 and 2002 as having chronic functional somatic symptoms and 182 controls matched by age, sex, socioeconomic status, and practice were included. Data on comorbidity, referrals, diagnostic tests, and hospital admissions over a period of 10 years prior to the diagnosis were collected. Medication use and number of visits to the general practitioner (GP) were extracted from the moment computerized registration was started.

Results. In the 10 years before the diagnosis chronic functional somatic symptoms, significantly more patients than controls present functional somatic symptoms in at least two body systems, and used more somatic and psychotropic drugs. They visit the GP twice as much, statistically had significantly more psychiatric morbidity, and were referred more often to mental health workers and somatic specialists. The number of patients undergoing diagnostic tests was higher for patients with chronic functional somatic symptoms than for controls, but hospital admissions rates were equal.

Conclusion. Patients with chronic functional somatic symptoms have a great diversity of functional somatic symptoms. They use more somatic and psychotropic drugs than controls in the years before diagnosis. Moreover, they show high rates of referrals and psychiatric morbidity. The diversity of symptoms of patients with chronic functional somatic symptoms supports the concept that symptoms do not cluster in well-defined distinct syndromes. Therefore, patients with chronic functional somatic symptoms should preferably not be classified into medical subspecialty syndromes.

Introduction

Medically unexplained symptoms are common, and account for one in five new consultations in primary care.[1;2] In 20-25% of all primary care visits, no serious medical (that is, organic) cause is found to explain the patient's presenting symptom, and 20-40% of the patients seen by medical specialists do not receive a clear diagnosis.[3;4] The presented symptoms are then referred to as 'medically unexplained' or 'functional'.[5] Functional, or rather medically unexplained, somatic symptoms are ranked second on the list of the 10 most common physical symptoms in primary care and have an incidence rate of 70 per 1000 patient years in the Netherlands.[6]

Although an occasional visit to the general practitioner (GP) for a functional somatic symptom seems natural, repeated consultations because of these symptoms represent a serious problem. Patients who do this are often diagnosed as having 'chronic functional somatic symptoms'. Psychological distress or psychosocial problems are presumed to be the underlying causes.[7] As such, diagnosing chronic functional somatic symptoms requires the patient to repeatedly present physical symptoms that remain medically unexplained after adequate examination, and indications, from the patients' personal circumstances of presumed psychosocial problems or psychological distress.

As patients with chronic functional somatic symptoms are functionally impaired, have high rates of comorbid psychiatric disorders, and are at risk of unnecessary diagnostic procedures and treatments,[1;4;8-11] a correct diagnosis is of paramount importance. However, most research on this topic has been performed either on unselected population-based samples,[12;13] or in selected patients referred to secondary care.[14;15] Moreover, most of these studies make use of questionnaires in which patients have to recall a variety of symptoms existing for a considerable amount of time.[10;16;17] This method has been shown to produce unstable results in which lifetime symptoms present at baseline are not remembered at follow-up.[18] Research on patients from primary care settings in whom the diagnosis had been made on reliable longitudinal data is generally lacking.

Moreover, there is considerable debate regarding the question of whether functional somatic symptoms cluster in well defined distinct syndromes, such as fibromyalgia, chronic fatigue syndrome, or tension headache, or whether these specific somatic syndromes are largely an artefact of medical specilization.[7;19] In this debate reliable data on primary care patients are also needed, whereas most research on this topic is performed in referred populations[20;21] concentrating on specific symptoms,[22-24] or in community samples[25] using questionnaires in which inconsistencies of recall may have a great effect on the assessment of the ultimate diagnosis.[18;26] The aims of this study, therefore, are to explore with longitudinal data:

- how and how often patients with chronic functional somatic symptoms present to their GP and other medical institutions,
- whether patients with chronic functional somatic symptoms indeed present more functional somatic symptoms in the years before the diagnosis,
- if symptoms presented by patients with chronic functional somatic symptoms support the existence of specific somatic syndromes.

Methods

Continuous Morbidity Registration database

This study uses data from the Continuous Morbidity Registration (CMR) database, a project of the department of Family Medicine of the University of Nijmegen the Netherlands.[27-30] This project was started in 1971 in four practices in and around Nijmegen[31] and monitors a population of approximately 12 000 patients, representative of the Dutch population with regard to age and sex. Every episode of illness seen by, or reported to, the GP is registered as soon as it is established using an adapted version of the E-list.[32] Diagnoses and codes are corrected when necessary. Over many years, monthly meetings of all GPs involved are held to discuss classification problems, to monitor the application of diagnostic criteria, and to discuss coding problems of hypothetical case histories. As well as medical data, the following information is available: age, sex, socioeconomic status (low, middle and high), and marital status. In the beginning the registration was performed on the medical chart; since 1994 a computerized registration has been used.

Patients with chronic functional somatic symptoms

We selected all patients from the CMR database in whom chronic functional somatic symptoms were diagnosed for the first time between 1998 and 2002 ($n = 182$). For a period of 10 years before this diagnosis, the following variables had been collected: sociodemographic characteristics; morbidity data; and data on referrals, diagnostic tests, and hospital admissions. Use of medical facilities was assessed by the number of contacts with the GP. Data on medication use could be extracted from when computerized registration started, and medication data were transformed into the prescribed daily dose by using the Anatomical Therapeutical Chemical Classification/Defined Daily Doses (ATC/DDD) system.[33] As a proxy of somatic morbidity, we assessed three prevalent categories of chronic disorders (diabetes mellitus, asthma/chronic obstructive pulmonary disease [COPD], and cardiovascular diseases) and three prevalent categories of self-limiting disorders (skin disorders, musculoskeletal, and airway) in order to study the hypothesis that patients with chronic functional somatic symptoms are at risk for somatic morbidity.[37]

We allocated the registered functional somatic symptoms to specific body systems; for example, gastrointestinal or musculoskeletal, as described by Escobar *et al.*[10] Irritable bowel syndrome and hyperventilation syndrome, sometimes regarded as medically unexplained symptoms, are not included in this study.

Controls

For each patient with chronic functional somatic symptoms, a control matched by age, sex, socioeconomic status, and practice was drawn from the CMR population. The only exclusion criterion in the control group was the diagnosis chronic functional somatic symptoms. Patients who were controls had to have had at least one registered episode of illness during the period they had been on the practice list. For controls, the same information as described for patients with chronic functional somatic symptoms was obtained from 1990-2000.

Statistical methods

Our analysis primarily involved comparing patients with chronic functional somatic symptoms with their matched controls. Statistical analyses were conducted using SPSS 9.0. Descriptive statistics were calculated for all variables. The data on specific body systems were analysed using exploratory factor analysis, and then simplified by varimax rotation. The χ^2 test and student's *t*-test were used for comparing means of consultations and medication use in both groups. Odds ratios (ORs) and 95% confidence intervals (CIs) were used as the main measurement for associations, particularly with regard to functional somatic symptoms, comorbidity, referrals, diagnostic tests and hospital admissions. All *P*-values are two-tailed.

Results

Characteristics of subjects

Of the 182 included patients with chronic functional somatic symptoms included in the study, 141 (77.5%) were women; the mean age of all patients was 42.0 years (range = 10-85 years). Most subjects were of low (44.5%) or middle (42.9%) socioeconomic class.

Functional somatic symptoms

The incidence rate of patients with chronic functional somatic symptoms was 3.5 per 1000 patient years, whereas the prevalence of patients known to have chronic functional somatic symptoms is established on 68.8 per 1000 patient years.

The presented functional somatic symptoms in various body systems in patients and controls is displayed in Table 1. For each symptom group, patients and controls differ significantly

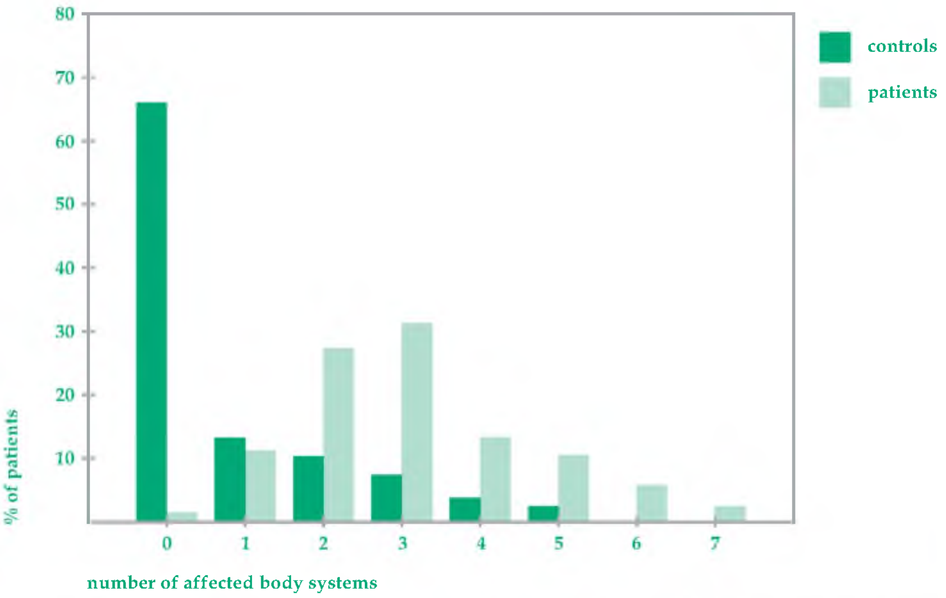
Table 1. Distribution of functional somatic symptoms in the various body systems (n = 182)

	Patients (%)	Controls (%)	Odds ratio (95% CI)[a]
Pseudoneurological	54 (29.7)	13 (7.1)	5.5 (2.8 to 11.1)
Gastrointestinal	69 (37.9)	15 (8.2)	6.8 (3.6 to 13.1)
Musculoskeletal	58 (31.9)	6 (3.3)	13.7 (5.5 to 36.5)
Cardiorespiratory	74 (40.7)	16 (8.8)	7.1 (3.8 to 13.5)
Headache and other pain	80 (44.0)	15 (8.2)	8.7 (4.6 to 16.6)
Pseudopsychiatric	150 (82.4)	48 (26.4)	13.1 (7.7 to 22.4)
Others	66 (36.3)	10 (5.5)	9.8 (4.6 to 12.2)
Unknown	67 (36.8)	0 (0)	

[a] Statistical significant difference between patients and controls.

(*P*<0.05). It is remarkable that many patients had symptoms in various body systems – a finding that is supported by the factor analysis (Figure 1) – as it is often considered that there are a number of well-defined distinct functional somatic syndromes, clustering around physical symptoms of one body system. Moreover, factor analysis suggests that, on the one hand, gastrointestinal, cardiorespiratory, and pseudopsychiatric symptoms are linked and, on the

Figure 1. Symptom diversity in patients with chronic functional somatic symptoms and their matched controls



other, that pseudoneurological symptoms, musculoskeletal symptoms, and headache and other pain, are linked with each other. Significantly more patients than controls presented symptoms in two or more body systems (87.9 versus 19.8%; OR = 29.5, 95% CI = 16.0 to 54.9). Of all the patients, 25% presented symptoms in four or more body systems.

Half of the patients had three or more episodes of functional somatic symptoms before he or she was diagnosed as having chronic functional somatic symptoms; 25% of the patients had five or more episodes before chronic functional somatic symptoms were diagnosed.

Comorbidity: somatic and psychiatric

Patients with chronic functional somatic symptoms had significantly more psychiatric disorders in comparison with controls (OR = 2.4) (Table 2). Patients did not have a much higher rate of chronic and self-limiting somatic comorbidity, and they had only slightly more episodes of self-limiting airway problems than controls.

Consultations, referrals, diagnostic tests and hospital admissions

The number of consultations in patients with chronic functional somatic symptoms is significantly higher (n = 9.8 versus n = 4.2, P<0.001), as the number of subjects referred for diagnostic testing (n = 156 versus n = 140, OR = 1.8). The number of home visits was equal in both groups. About three-quarters of patients had been referred to somatic specialists, compared with about half of the controls. About one-third of the patients had been referred to mental health sources compared with less than 10% in controls. Hospital admissions were the same. These data are outlined in Table 2.

Medication use

The results regarding medication use are detailed in Table 3. We found that patients with chronic functional somatic symptoms used a significant more somatic medication (2.6 versus 1.5, P<0.001) and psychotropic drugs (0.4 versus 0.05, P<0.001) per year compared with controls. The number of patients using antidepressants and benzodiazepines is statistically different in both groups (35.6% versus 5.9%; 52.5% versus 12.7% respectively, P<0.001).

In patients using medication, antidepressants were used for a mean of 20 days a year and benzodiazepines for 9 days a year compared with 5 and 4 days, respectively, in controls. However, these findings do not reach statistical significance. Moreover, there is no difference in prescribed daily dose for patients and controls.

Table 2. Number of consultations, comorbidity, referrals, diagnostic tests, and hospital admissions in patients and controls (n = 182)

	Patients	Controls	*P*-value	Odds ratio (95% CI)
GP consultations in one year[a] (mean [range])				
Practice visits	9.8 (2.5-26.8)	4.2 (0-15.1)	<0.001[b]	-
Home visits	0.2 (0-3.5)	0.3 (0-7.1)	0.53	-
Comorbidity (*n* [%])				
Somatic chronic:				
Diabetes Mellitus	5 (2.7)	4 (2.2)	-	1.3 (0.3 to 5.7)
Asthma/COPD	20 (11.0)	10 (5.5)	-	2.1 (0.9 to 5.0)
Cardiovascular	16 (8.8)	8 (4.4)	-	2.1 (0.8 to 5.5)
Somatic self-limiting:				
Skin	96 (52.7)	82 (45.1)	-	1.4 (0.9 to 2.1)
Musculoskeletal	130 (71.4)	115 (63.2)	-	1.5 (0.9 to 2.3)
Airway	136 (74.7)	113 (62.1)	-	1.8 (1.1 to 2.9)[b]
Psychiatric[c]	41 (22.5)	20 (11.0)	-	2.4 (1.3 to 4.4)[b]
Referrals (*n* [%])				
Somatic: Medical	130 (71.4)	97 (53.3)	-	2.2 (1.4 to 3.5)[b]
Somatic: Paramedical[d]	129 (70.9)	87 (47.8)	-	2.6 (1.7 to 4.2)[b]
Mental Health	59 (32.4)	15 (8.2)	-	5.3 (2.8 to 10.3)[b]
Diagnostic test[e] (n[%])	156 (85.7)	140 (76.9)	-	1.8 (1.0 to 3.2)[b]
Hospital admissions (*n* [%])				
Somatic	57 (31.3)	51 (28.0)	-	1.2 (0.7 to 1.9)
Psychiatric	1 (0.5)	1 (0.5)	-	1.0

[a] n = 118: one general practice did not use the computerised registration, so consultation could not be established in this practice. [b] Statistically significant difference between patients and controls $P<0.05$. [c] Including schizophrenia, depression, psychoses, hysteria, phobia, neuroses, post-traumatic stress disorder, alcoholism, use of street drugs. [d] Including physiotherapist, dietician. [e] Including hematological tests, x-ray examinations, ultrasonography, electrocardiography.

Discussion

Strengths and limitations of this study

The strength of the present study is that the patients who were included were those who consulted their GP, irrespective of the presented symptoms. Therefore, a threshold of relevance of the symptoms for the patient was established and we were able to analyse all symptoms presented. Most population-based studies assess all symptoms irrespective of the perceived need for help.[10;16;17] Moreover, in population-based studies, interviewing patients repeatedly does not lead to a consistent classification of somatoform disorders,[18] whereas our classification of the presented morbidity is based on very stable data,[29;34] in which longitudinal research is allowed and recall bias will not occur.

Table 3. Medication use in patients and controls[a] (n = 118)

	Patients	Controls	P-value	Odds ratio (95% CI)
Number of somatic medication per year (mean [range])	2.6 (0–18.0)	1.5 (0–7.5)	<0.001[b]	-
Number of psychotropic drugs per year (mean [range])	0.4 (0–9.0)	0.05 (0–0.62)	<0.001[b]	-
Number of patients using psychotropic drugs (n [%])				
Antidepressants	42 (35.6)	7 (5.9)	-	7.5 (3.1to18.9)[b]
Benzodiazepines	62 (52.5)	15 (12.7)	-	5.8 (3.0to11.1)[b]
Others	3 (2.5)	3 (2.5)	-	1.0
Days of psychotropic drug use per year (median [range])	19.5 (1.9–297.0)[c]	5.0 (1.62–91.7)[d]	-	-
Antidepressants	8.9 (0.4–322.5)[c]	4.2 (1.1–90.8)[d]	-	-
Benzodiazepines	34.3 (1.7–58.2)[c]	3.8 (0.2–25.1)[d]	-	-
Others				
Prescribed daily dose (mean [range])				
Antidepressants	0.76 (0.20–1.37)[c]	0.77 (0.13–2.00)[d]	-	-
Benzodiazepines	0.62 (0.06–1.50)[c]	0.61 (0.20–1.54)[d]	-	-
Others	0.51 (0.25–0.75)[c]	0.31 (0.25–0.40)[d]	-	-

[a] n = 118: one general practice did not use the computerised registration, so medication could not be established in this practice. [b] Statistically significant difference between patients and controls $P<0.05$. [c] For patients with antidepressants, benzodiazepines and other psychotropic drugs, n = 42, 62, and 3, respectively. [d] For controls with antidepressants, benzodiazepines and other psychotropic drugs, n = 7, 15 and 3, respectively.

The limitations of the study are the retrospective use of data in existing medical records and the possible interdoctor variation of the diagnosis of chronic functional somatic symptoms.[35] The interdoctor variation is partly a consequence of not having explicitly stated criteria for chronic functional somatic symptoms in the CMR. This subjectivity will possibly always exist because diagnosing chronic functional somatic symptoms remains an interpretation of the symptoms, and is influenced by foreknowledge and context.[27] However, it is known from the literature that the GP's judgement on somatisation seems valid in daily practice. Moreover, additional validation of clinical judgement is possible through longitudinal follow-up.[36] The subjectivity of the diagnosis and the doctor-patient relationship also make important contributions to the genesis and persistence of functional somatic symptoms.[37] The doctor's knowledge of the patient's complaints is an important issue and is associated with a better outcome.

Of all variables described, only consult frequency is directly linked with the diagnosis of chronic functional somatic symptoms – as such, the higher frequency of GP visits was to be expected a priori.[38]

Summary of main findings

This is the first observational study using longitudinal data describing patients in whom consulting the GP for functional somatic symptoms has become a regular way of presenting. During the 10 years before the diagnosis of chronic functional somatic symptoms is established by the GP, patients consult their GP twice as much, use much more somatic and psychotropic medication, have more psychiatric morbidity and are more often referred to mental health workers than controls. During these 10 years, the number of diagnostic tests is slightly higher in patients and the number of hospital admissions is equal in comparison with controls. Patients with chronic functional somatic symptoms are more likely to present symptoms in two or more body systems and they present a higher number and greater diversity of symptoms to the GP than control patients. GPs in this study appear to classify patients as having chronic functional somatic symptoms after three episodes of presenting with functional somatic symptoms.

Comparison with the existing literature

The finding that patients could be recognized as having chronic functional somatic symptoms after they had experienced three episodes of functional somatic symptoms presented in two or more body systems, is an important one. Functional somatic symptoms are often recognized after having excluded other possible diagnosis. This may be associated with unnecessary and possibly harmful diagnostic strategies and may promote somatic fixation. Early identification of these patients could prevent somatic fixation, and enables the GP to modify his/her proceedings.[39] Additionally, this finding was confirmed using factor analysis and shows that functional somatic symptoms probably do not cluster in well defined specific somatic syndromes. It also suggests that symptom variation is great in these patients.

The concept of patients with functional somatic syndromes presenting symptoms in many body systems has also been supported by recent studies.[20] Therefore, the existence op specific somatic syndromes should be challenged. With a broad-based approach, the GP might be the appropriate practitioner to diagnose and treat these patients by emphasizing the biomedical as well as the psychosocial factors involved in symptom production and perception.[40]

The finding that patients with chronic functional somatic symptoms did not have a higher rate of chronic and self-limiting somatic comorbidity is remarkable because it is stated in the literature that somatisation with more frequent examination may increase the chance for chronic diseases to be discovered.[36] We found that more frequent consultation did not lead to more diagnosed chronic and self-limiting diseases in patients with chronic functional somatic symptoms.

The diagnosis chronic functional somatic symptoms is not recorded as such in the DSM-IV classification. It no doubt exists as part of the spectrum somewhere between somatisation disorder and somatoform disorder not otherwise specified. The condition resembles the concept of 'abridged somatization',[10] but is not based on the number of symptoms. Both 'abridged somatization' and chronic functional somatic symptoms presume underlying psychological distress. The prevalence of somatisation disorder according to the DSM-IV in primary care is low because of the stringent criteria.[40;41] On the other hand, less severe forms of somatization have a major impact on quality of life and on the use of health services, and are more prevalent.

Implications for further research and clinical practice

Patients with chronic functional somatic symptoms may be considered as persistent complainers and consequently labelled as 'difficult' patients. The condition may indeed reflects a greater propensity to complain; however, as is apparent from the excess of psychiatric comorbidity, patients with chronic functional somatic symptoms also have more reason to complain. With regard to these patients, it seems that consulting the GP for functional somatic symptoms has become a regular way of presenting, but it might also be that patients who attend more often are at higher risk of being considered as chronic functional somatic symptoms. Moreover, the diagnosis might relate to frustrated doctors as a consequence of lack of understanding, or failures in the communication between doctor and patient.

Chronic functional somatic symptoms are a major cause of morbidity and deserve further investigation to estimate the importance of the doctor-patient relationship, the feasibility of treatments, and the understanding of the aetiology of functional symptoms to identify patients who are likely to become persistent complainers and develop the behavioural pattern of patients with chronic functional somatic symptoms.

Also, the overlap of chronic functional somatic symptoms with the various DSM-IV diagnoses should be studied. This is important because the validity of the classification of mental disorders is useful, but also questionable.[42] Thinking in narrow syndromes might hinder an appropriate interpretation of the patient's syndromes.

Patients repeatedly presenting functional somatic symptoms to the GP in two or more body systems, particularly when combined with psychological complaints, should be regarded as candidates for the diagnosis of chronic functional somatic symptoms. It seems that the presented functional somatic symptoms are part of a single syndrome and that symptom variation is great in these patients. Therefore, the GP, who is considered as being knowledgeable about underlying psychosocial problems, should diagnose, treat, and accompany these patients.

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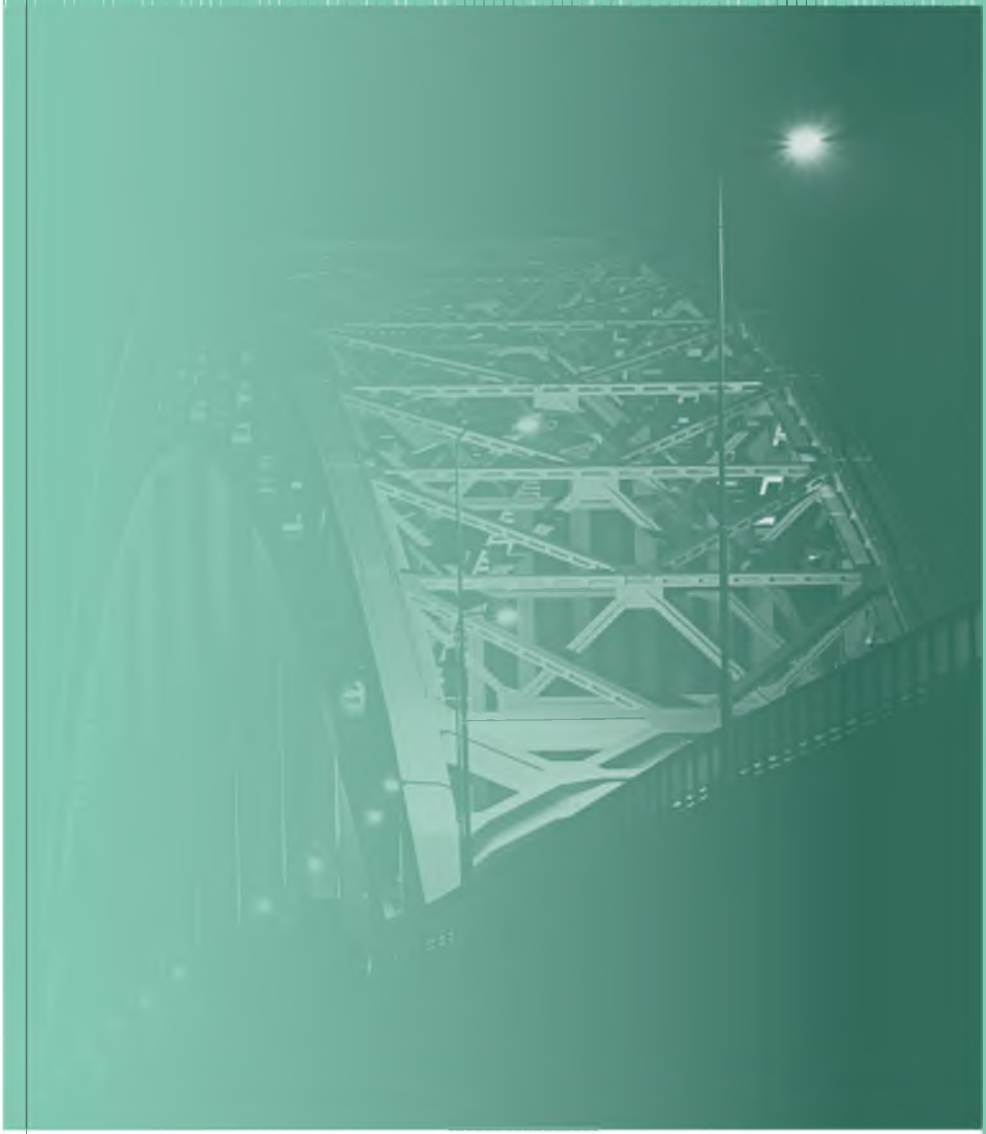
CHAPTER

3

MEDICALLY UNEXPLAINED SYMPTOMS, SOMATISATION DISORDER AND HYPCHONDRIASIS: COURSE AND PROGNOSIS. A SYSTEMATIC REVIEW



Tim olde Hartman
Machteld Borghuis
Peter Lucassen
Floris van de Laar
Anne Speckens
Chris van Weel

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Abstract

Objective. To study the course of medically unexplained symptoms (MUS), somatisation disorder and hypochondriasis and related prognostic factors. Knowledge of prognostic factors in patients presenting persistent MUS might improve our understanding of the naturalistic course and the identification of patients with a high risk of a chronic course.

Methods. A comprehensive search of Medline, PsycInfo, CINAHL and EMBASE was performed to select studies focusing on patients with MUS, somatisation disorder and hypochondriasis and assessing prognostic factors. Studies focusing on patients with single-symptom unexplained disorder or distinctive functional somatic syndromes were excluded. A best-evidence synthesis for the interpretation of results was used.

Results. Only six studies on MUS, six studies on hypochondriasis, and one study on abridged somatisation could be included. Approximately 50 to 75% of the patients with MUS improve, whereas 10 to 30% of patients with MUS deteriorate. In patients with hypochondriasis recovery rates vary between 30% to 50%. In studies on MUS and hypochondriasis we found some evidence that the number of somatic symptoms at baseline influences the course of these conditions. Furthermore, the seriousness of the condition at baseline seemed to influence the prognosis. Comorbid anxiety and depression do not seem to predict the course of hypochondriasis.

Conclusions. Due to the limited numbers of studies and their high heterogeneity, there is a lack of rigorous empirical evidence to identify relevant prognostic factors in patients presenting persistent MUS. However, it seems that a more serious condition at baseline is associated with a worse outcome.

Introduction

Medically unexplained symptoms (MUS) are common in primary care.[1] In 25-50% of all primary care visits, no somatic cause is found to explain the patient's presenting symptoms.[2] It is generally believed that persistent presentation of MUS is a chronic and disabling disorder.[3] However, in many patients MUS are transient and have a good prognosis. A recent Dutch study found that only 2,5% of the attendees in general practice presenting with such symptoms meet criteria for chronicity.[4]

In a recent review, researchers stated that in population-based and primary care samples, MUS is the common characteristic of the *DSM-IV* and *ICD-10* somatoform disorders including somatisation and hypochondriasis.[5-7] Somatisation is characterized by recurrent and frequent presentation of MUS whereas hypochondriasis is characterized by excessive worry about illness and the belief of having an undiagnosed physical disease.

Despite the low prevalence of persistent MUS, it represents a serious problem in primary care. Patients are functionally impaired, have high rates of comorbid psychiatric disorders and are at risk for unnecessary, potentially harmful diagnostic procedures and treatments.[2,3,8] Moreover, part of the burden to GPs are the difficulties in explaining the symptoms, finding a shared understanding necessary to reach reassurance and acceptation of the symptoms and the lack of treatment options.[9,10] Often, GPs label these patients as 'heartsink patients' or 'helpoholic patients'.[11] For patients, as well as GPs and the health care system it is important to prevent persistent MUS. Therefore, GPs should be able to recognize patients with a high risk of persistent MUS. However, GPs experience difficulties in distinguishing self limiting MUS from persistent MUS.[12] Knowledge of prognostic factors may improve our management of patients with MUS, as patients with a good prognosis can be reassured about the favourable spontaneous recovery rates, whereas a more intensive approach including some form of reattribution or cognitive behavioural therapy might be indicated from the beginning in the high-risk groups. The aim of our study is to gain insight in the course of MUS and in factors influencing its course.

Method

Data sources and search strategy

We systematically reviewed prospective cohort studies in primary, secondary or tertiary care on patients with MUS, somatoform disorders and hypochondriasis. We studied somatisation disorder, MUS and hypochondriasis together because they appear to have much in common: medically unexplained symptoms, typical illness and sick role behaviour, disproportionate disability and preoccupation with health and illness.[7]

Although there are many other general terms to describe physical symptoms without an organic explanation, we use the term 'medically unexplained symptoms' as none of these terms are ideal and this is the most neutral description.[13,14]

We did not include clinical trails in this review as the patients recruited into trails are often not representative of the population with the disorder.[15,16] Moreover, participating in a trial can influence the natural course of the symptoms as participating in a trial can be considered as an intervention in itself.

We searched in the MEDLINE database for publications published between 1965 and 1 June 2006, in PsycINFO between 1967 and 1 June 2006, in CINAHL between 1982 and 1 June 2006 and in EMBASE between 1965 and 1 June 2006. We obtained additional references from the reference lists of review articles and retrieved original papers. We used the following keywords: somatoform disorder, hypochondriasis, neurasthenia, conversion disorder, psychophysiological disorder, functional somatic sympt* and medically unexplained*.

We combined this search using the Boolean operator AND with the sensitive MEDLINE search for clinical studies on prognosis.[17] The search strategy is shown in Appendix A. There were no limitations regarding the language of publication. We tested the search strategy on 30 publications about medically unexplained symptoms in our own database and found the search strategy to be sensitive as all known articles were found.

Study selection

ToH and MB independently screened the titles and abstracts of all identified citations to identify eligible articles. When we could not decide on inclusion, we consulted the full publication.

If after studying the complete manuscript disagreement persisted, we consulted a third reviewer (FvdL). We used Cohen's kappa statistic (κ) to assess agreement between the two reviewers.[18] Inclusion criteria were: prospective cohort design, focus on prognosis of patients with medically unexplained symptoms and a follow-up of 3 months or more.

We excluded studies that focused primarily on patients with medical or psychiatric disease (except somatoform disorders and hypochondriasis). We also excluded studies that focused on patients suffering from single-symptom unexplained disorder (tension headaches, dysmenorrhoea) or patients suffering from distinctive functional somatic syndromes (irritable bowel syndrome, chronic fatigue syndrome) because we were interested in the course and prognosis of undifferentiated medically unexplained symptoms. We focused on undifferentiated MUS as we assume that these are more difficult to handle for the physician than single symptom unexplained disorders and distinctive functional syndromes. After all, the latter give more opportunity to explain the symptoms to patients. Finally, there is evidence that the name of a condition influences prognosis.[19] Studies on children and adolescents (age < 18 years) and studies on specific groups of patients such as refugees, street prostitutes etc, were excluded. Case-control studies, cross-sectional studies and case studies were also excluded.

Data extraction

Two reviewers (ToH and MB) independently scored the methodological quality of the included studies. We used a standardized checklist of predefined criteria (see Appendix B), which has been used in previous prognostic reviews.[20,21] The list is based on theoretical considerations and methodological aspects described by Hudak et al.[22] and Altman.[23] We modified these checklists according to new insights.[24] We tested the quality assessment checklist in a pilot assessment. A detailed explanation of each criterion is given in Appendix C. Each criterion was scored positive (+), negative (-) or unclear (?). The total quality score is the sum of all the criteria that are scored positive. The maximum quality score is 21. We calculated the quality of a study as the percentage of the maximum score.

We discussed disagreements in the scoring of quality items in a consensus meeting. In a case of persisting disagreement between the two reviewers a third reviewer (FvdL) made the final decision.

We categorized quality criteria into four major forms of bias: selection bias, completeness of follow-up, information bias and confounding. Furthermore, we defined studies with a quality score of 60% or higher as studies with high quality.[25]

The two reviewers (ToH and MB) independently extracted the information from the selected papers by using standardized and pre-tested data-extraction forms. The extracted information involved data on study population, diagnostic criteria, inclusion and exclusion criteria, setting, type of prognostic factors, duration of follow-up, outcomes and data on associations. In a case of disagreement, we reached consensus after discussion with a third reviewer (FvdL).

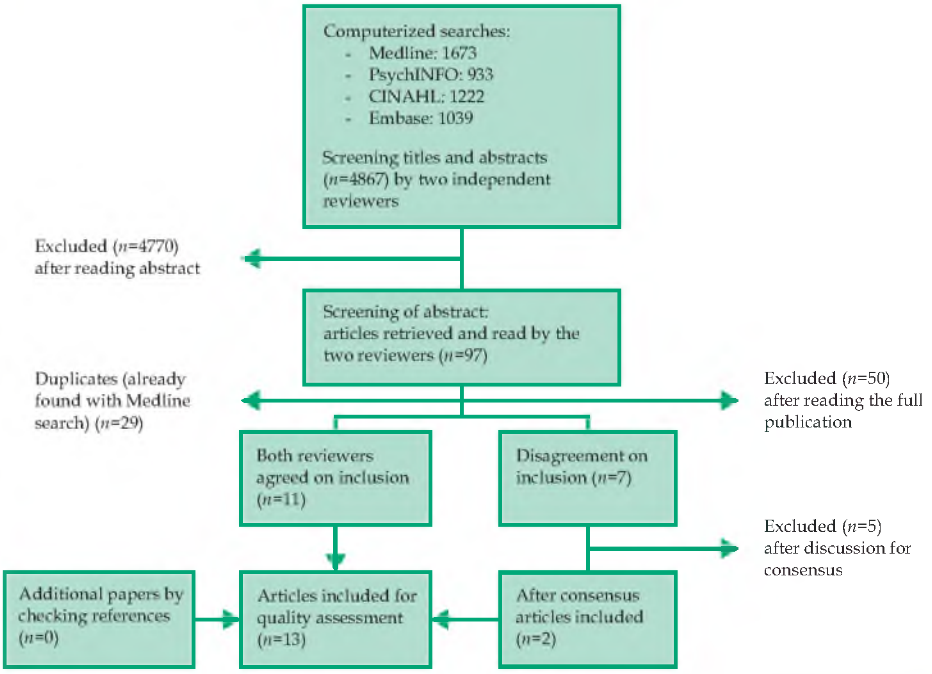
Data synthesis

We did not plan statistical pooling as we anticipated considerable heterogeneity. Therefore, a qualitative analysis (best evidence synthesis) was performed to summarize the value of the prognostic indicators. Furthermore, we considered the strength of evidence regarding a prognostic factor as strong, moderate, weak or inconclusive depending on consistency of the findings and on quality of the study:[26,27]

- strong: consistent findings (≥ 75% of the studies reporting on a factor showed the same direction of the association) in at least two high quality studies
- moderate: consistent findings (≥ 75% of the studies reporting on a factor showed the same direction of the association) in one high quality cohort and at least one low quality study
- weak: findings of one high quality cohort or consistent findings (≥ 75% of the studies reporting on a factor showed the same direction of the association) in at least three or more low quality studies
- inconclusive: inconsistent findings irrespective of study quality, or less than three low quality studies available

We only present prognostic factors which in at least one study showed a statistically significant association. Preferably, we derived the associations from the multivariate results. If only univariate results were presented in the original study, we used these univariate associations to determine the strength of evidence.
We present results of the studies on MUS, somatisation disorder and hypochondriasis separately.

Figure 1. Selection of studies



Results

We retrieved a total of 4867 publications from searches of the various electronic bibliographies (1673 Pubmed, 933 Psychinfo, 1222 CINAHL and 1039 EMBASE) (see Figure 1). After screening the titles and abstracts, 68 abstracts seemed to fulfil the inclusion criteria. After assessing the full publication, 13 articles fulfilled all inclusion criteria and were included in our review.[28-40] Major reasons for excluding papers were: focus not on patients with medically unexplained symptoms (n=30) and no study of prognostic factors (n=14). The reference lists of the retrieved papers did not reveal any relevant publication. Six studies reported on MUS,[28-31,36,37] six studies on hypochondriasis[32-35,38,39] and one study on abridged somatization.[40] The abridged definition of somatisation required the presence of four symptoms in males and six symptoms in females.[41] We did not find any prospective cohort studies on *DSM-IV* somatoform disorders.
The interobserver agreement for inclusion between the two reviewers (ToH, MB) was $\kappa = 0{,}73$ (95%-CI: 0.59 – 0.87). We considered the strength of agreement to be 'good'.[42]

Study characteristics

We found 6 studies on MUS. Table 1 gives the data of the quality assessment of the included studies. The quality score of MUS publications ranged from 62% to 86%. As none of the included studies described a treatment subsequent to inclusion in the study cohort (item L), we cannot decide on whether the natural course was studied or course during treatment (as usual). Loss to follow-up ranged from 0 to 27%.

We found one prospective cohort study on abridged somatisation. The quality of this publication scored 67%.

We included 6 studies on hypochondriasis. The methodological quality score of hypochondriasis publications ranged from 57% to 76%. (see Table 1) As in the MUS studies, in these six hypochondriasis studies application of treatments was not described. So whether the natural course or course during treatment was studied cannot be concluded. Selection bias and confounding was present in all studies and in four of the six studies information bias was presented.[32,33,38,39] (see Table 1) Loss to follow-up ranged from 4% to 36,6%.

A summary of the study characteristics is presented in Table 2, including population, setting, diagnostic criteria, follow-up and baseline characteristics.

Four of the six studies on MUS are performed in The Netherlands. Studies reporting on MUS defined MUS as symptoms that could not be attributed to a clear organic cause according to the physician's judgment after a thorough physical examination including laboratory tests. So, physician's judgment was often the most important diagnostic instrument.

Table 1. Results of the methodological quality of prognostic cohort studies on MUS, abridged somatisation and hypochondriasis

	A	B	C	D	F	G	I	J	N	O	P	L	Q	R	E	H	K	M	S	T	U		
Quality criteria	Selection bias						Completeness of follow-up		Information bias			Confounding			Descriptive items							Quality score[a]	Score (%)
MUS																							
Kooiman et al., 2004[30]	+	+	+	+	-	+	+	+	+	+	+	-	+	+	-	+	+	+	+	+	+	18	86.0
De Gucht et al., 2004[29]	+	+	+	+	+	+	+	+	+	-	+	-	-	+	+	-	+	+	+	-	+	16	76.2
Speckens et al., 1996[36]	+	+	+	+	?	?	+	+	+	+	+	-	+	+	-	+	+	+	+	+	-	16	76.2
Carson et al., 2003[31]	+	+	+	+	?	+	-	+	+	+	+	-	+	-	-	-	+	+	+	+	-	14	66.6
Speckens et al., 1996[37]	+	+	+	+	-	-	+	-	+	-	+	-	+	-	-	+	+	+	+	-	+	13	61.9
Henningsen et al., 2005[28]	+	+	+	+	?	-	-	+	+	+	+	-	?	+	-	-	+	+	+	+	-	13	61.9
Abridged somatisation																							
Gureje and Simon, 1999[41]	+	-	+	+	-	+	-	+	+	-	+	-	+	+	+	+	+	+	+	-	-	14	66.6
Hypochondriasis																							
Noyes et al., 1994[35]	+	+	+	+	-	-	+	-	+	+	+	-	+	+	-	+	+	+	+	+	+	16	76.2
Barsky et al., 1998[33]	+	+	+	+	?	?	-	+	+	-	+	-	+	+	-	+	+	+	+	+	-	14	66.7
Barsky et al., 2000[34]	+	+	+	+	?	?	+	+	+	-	+	-	-	+	-	+	+	+	+	-	-	13	61.9
Barsky et al., 1993[32]	+	-	+	?	-	-	-	-	+	+	+	-	+	-	-	+	+	+	+	+	+	12	57.1
Fernandez et al., 2005[38]	+	+	+	+	-	-	+	-	+	-	+	-	+	-	-	-	+	+	+	+	-	12	57.1
Simon et al., 2001[39]	+	-	+	+	-	+	?	+	+	-	+	-	-	-	-	+	+	+	+	+	-	12	57.1

[a] Total '+'.

In these six MUS studies we found high levels of heterogeneity regarding clinical setting (primary care, secondary care and tertiary care), numbers enrolled in the cohort (80 to 377 patients), duration of follow-up (6 to 15 months) and loss to follow-up (0 to 27%). Only two studies reported on the duration of symptoms at baseline.[28,36] Speckens et al.[36] (1996) reported a median duration of symptoms of 7,8 months (range 0-168), whereas Henningsen et al.[28] (2005) reported a mean duration of 70 ± 94 months (median 26).

The study on abridged somatisation was performed in primary care from 15 sites in 14 countries and enrolled 1596 patients into the cohort. Abridged somatisation was diagnosed according to the Somatic Symptoms Index (SSI). Duration of symptoms at inclusion was not reported.

All studies on hypochondriasis used a formal diagnostic interview to diagnose patients with hypochondriasis as stated in the *DSM-III-R*. (see Table 2) Fernandez et al.[38] (2005) included patients with health anxiety. These patients share many characteristics with patients suffering from hypochondriasis.[38] Despite the use of formal diagnostic interviews, there was considerable heterogeneity in the six included studies. Duration of follow-up (one to five years), numbers enrolled in the cohort (50 to 129 patients) and loss to follow-up (4 to 37%) vary considerable between the included studies. Only two studies reported on duration of symptoms at baseline. Noyes et al.[35] (1994) included patients with a median duration of symptoms of 19 (range 2-144) months. Fernandez et al.[38] (2005) reported that worries on health started more than 5 years ago in 36% of the patients, whereas in 12% of the patients these worries started in the last 6 months.

Course of MUS, somatisation disorder and hypochondriasis

Five out of 6 articles on MUS reported on the course of the symptoms (see Table 2). Based on prevalence, the typical MUS patient in our review is female, between 35 and 45 years old and consulted a primary care practice or secondary care outpatient clinic. Irrespective of the clinical setting, the majority of the patients with MUS (50 to 75%) improve during follow-up. However, about 10% to 30% of the patients deteriorate.

Five out of six studies on hypochondriasis reported on the course of hypochondriasis. Based on prevalence, the typical hypochondriasis patient is again female and between 35 and 45 years old. Fifty to 70% of the patients with hypochondriasis did not recover. Only Simon et al.[39] found a recovery rate in hypochondriasis patients of 85%.

The only study on somatisation disorder studied a modified concept.[40] Recovery rates were comparable with the data found in studies on MUS patients.

Prognostic factors of MUS, somatisation disorder and hypochondriasis

In Table 3 a summary of outcomes measures, prognostic factors and (strength of) significant associations is given. Apart from the high heterogeneity in study characteristics, we also found considerable heterogeneity in prognostic factors and outcome measures in the 13 studies included in this review.

Four of the six publications on MUS studied potential prognostic factors on the outcome 'symptom change'.[29,30,36,37]

There is some evidence that the number of symptoms at baseline predict the course of MUS. (see Table 4) Moreover, it seems that the more serious the condition at baseline, the more unfavourable the prognosis. This is represented by the factors General Health Perception

Table 2. Summary of main study characteristics and course of MUS, abridged somatisation, and hypochondrias

First author	Study quality (%)	Setting/country	Number enrolled in cohort	Criteria for diagnosis	Duration of follow-up [months (range)]	Loss to follow-up [*n* ,(%)]	Gender (M/F) and age (years±S.D.) at baseline	Course
MUS								
Kooiman et al., 2004[30]	81	General internal medicine outpatient clinic/The Netherlands	127	Judgement investigators on base of internist's final conclusion	14.1 (12.2-17.8)	0	43:84 40.2±12.7	62% improved; 38% not improved
De Gucht et al., 2004[29]	76	Primary care practices/ The Netherlands	377	Judgment FP	6	59 (16.0)	103:274 43.5±12.2	53.1% decrease of symptoms; 33.6% increase of symptoms; 9.7% same[a]
Speckens et al., 1996[36]	76	General medical outpatient clinic/ The Netherlands	81	Judgement investigators on base of medical records	15.2±4.0	0	28:53 39.4±10.7	30% recovered; 46% improved; 13% same; 11% worse
Carson et al., 2003[31]	67	General neurology outpatients/UK	90	Judgment neurologist	8	24 (27.0)	24:42[b] 42[b]	46% improved; 41% same; 14% worse[c]
Speckens et al., 1996[37]	62	General medical outpatient clinic/ The Netherlands	87	Judgment physician	11.6±0.8	5 (6.0)	Not given Not given	20% recovered; 51% improved; 18% same; 11% worse
Henningsen et al., 2005[28]	62	Tertiary care clinics/ Germany	186	Judgment of 2 physicians	6	43 (23.0)	84:102[b] 42.1±13.5[b]	Not given
Abridged somatisation								
Gureje and Simon, 1999[40]	67	Primary care/Europe, South America, US	1596	Diagnostic interview	12	525 (32.9)	Not given Not given	51.3% remitted; 48.7% unremitted
Hypochondriasis								
Noyes et al., 1994[35]	76	Medicine clinic/US	50	Diagnostic interview	13.8 (12.6-20.3)	2 (4.0)	10:38[b] 39.6±0.9[b]	33% remitted; 67% unremitted
Barsky et al., 1998[33]	67	Primary care clinic/US	C_1:60 C_2:60	Diagnostic interview	C_1:64.7±6.8 C_2:50.2±5.0	C_1:13 (15.0) C_2:22 (13.3)	22:63[b] 53.5[b]	36.5% remitted; 63.5% unremitted
Barsky et al., 2000[32]	62	Primary care clinic/US	60	Diagnostic interview	50.2±5.0	22 (36.6)	11:27[b] 48.0±15.2[b]	34.2% remitted; 65.8% unremitted
Barsky et al., 1993[34]	57	Primary care clinic/US	28	Diagnostic interview[d]	22.2 (12-35)	6 (21.4)	Not given Not given	Not given
Fernandez et al., 2005[38]	57	Primary care health centre/Spain	25	Semi-structured interview and questionnaires[e]	11.3	4 (16.0)	6:19 40.1	52% remitted; 48% unremitted[e]
Simon et al., 2001[39]	57	Primary care/Europe, South America, US	129	Diagnostic interview	12	Not given[f]	Not given Not given	84.5% remited; 15.5% unremitted

C_1: cohort 1.

C_2: cohort 2.

[a] Number of MUS; 3.6 % missing.

[b] Baseline characteristics only calculated for patients who completed the follow-up period.

[c] According to the clinical global improvement scale.

[d] Transient hypochondriasis.

[e] Health anxiety.

[f] Loss to follow-up only calculated for the whole cohort of the World Health Organization's Psychological Problems in General Health Care (PPGHC) study.[67]

Table 3. Prognostic factors

First author	Outcome measures	Prognostic factors	Direction of significant associations	Strength of association[a]
MUS				
Kooiman et al., 2004[30]	(1) Symptom change (2) Change in general health perception (3) Medical consumption (4) Psychiatric pathology	(a) Alexithymia (b) Sociodemograpic characteristics (c) Medical history (d) Mental problems (e) Illness behaviour (f) Symptom characteristics (g) Attribution	Symptom change - Longer duration of the symptom: absence of improvement - Higher number of physical symptoms: absence of improvement Change in general health perception (GHP) - Lower initial GHP at baseline: poor GHP at follow-up - Higher number of physical symptoms: poor GHP at follow-up - Less pain: poor GHP at follow-up	β=0.01 (0.005), P<.05 β=0.05 (0.02), P<.05 β=-0.04 (0.01), P<.01 β=0.07 (0.03), P<.05 β=-0.03 (0.01), P<.05
De Gucht et al., 2004[29]	(1) Changes in number of MUS (2) Symptom persistence or recurrence	(a) Neuroticism (b) Alexithymia (c) Negative or positive affective state (d) Sociodemographics	Number of MUS - Negative affect increase from T1 to T2: increase - Positive affect decrease from T1 to T2: increase Presence of a consistently high number of MUS - Female: increase - Consistently high negative affect: increase - Difficulty in identifying feelings (dimension of alexithymia): increase	OR=1.78 (1.33 to 2.39) OR=0,71 (0.54 to 0.94) OR=2.29 (1.14 to 4.62) OR=2.77 (1.46 to 5.27) OR= 1.08 (1.02 to 1.14)
Speckens et al., 1996[36]	(1) Change in symptoms (2) Change in functional impairment	(a) Gender (b) Age (c) Number and duration of symptoms (d) Psychiatric disorders	Change in symptoms - Female gender: absence of improvement - Higher number of symptoms: absence of improvement Change in functional impairment (FI): (b, 95% CI) - Higher FI at baseline: higher FI at follow-up - Higher age: higher FI at follow-up	OR=2.7 (1.01 to 7.4) β=1.0 (0.1 to 1.9) β=0.30 (0.17 to 0.43) β=0.03 (0.01 to 0.05)
Carson et al., 2003[31]	(1) Change in global clinical improvement	(a) Age (b) Gender (c) Health status (d) Mental state	Change in global clinical improvement[b] - Less physical function: absence improvement	P<.02
Speckens et al., 1996[37]	(1) Recovery of symptoms (2) Change in medical care utilization	(a) Hypochondriasis (questionnaire; WI) (b) Hypochondriasis (interview) (c) Illness attitude (d) Somatosensory amplification	Recovery of symptoms[b] - Higher scores on hypochondriasis questionnaire (WI): less recovery Change in medical care utilization (number of medical visits) - Higher scores on illness behaviour subscale of the illness attitude scale: increase of number of medical visits	β=-0.89 (-1.58 to -0.20) β=0.31 (0.09 to 0.52)
Henningsen et al., 2005[28]	(1) Affective and cognitive symptoms (2) Somatoform symptoms (3) Hypochondriasis (4) Quality of life	(a) Attribution	Affective and cognitive symptoms - Organic causal attribution: more depressive symptoms Quality of life - Organic causal attribution: less quality of life	P<.03 P<.01
Abridged somatisation				
Gureje and Simon, 1999[40]	(1) Persistence of abridged somatisation	(a) Gender (b) Self-rated poor health (c) Occupational disability (d) Physician-rated poor physical health (e) Depression (f) Generalized anxiety disorder (g) Age (h) Number of current symptoms at baseline	Persistence of abridged somatisation - Self-rated poor overall health: persistence - Moderate/severe occupational disability: persistence	OR=1.82 (1.32 to 2.52) OR=1.55 (1.17 to 2.06)

Table 3. Prognostic factors *(continued)*

First author	Outcome measures	Prognostic factors	Direction of significant associations	Strength of association[a]
Hypochondriasis				
Noyes et al., 1994[35]	(1) Remission of hypochondriasis (2) Levels of hypochondriacal symptoms[c]	(a) Demographics (b) Health care utilization (c) Social adjustment (d) Overall functioning (e) Duration and scores of hypochondriacal symptoms (f) Health perception (g) Sensitivity to bodily sensations and environmental stimuli (h) Personality, neuroticism, extroversion (i) Comorbid depression or anxiety	Remission of hypochondriasis[b] - Lower scores of hypochondriacal symptoms on WI: remission - Lower scores of hypochondriacal symptoms on SSI: remission - Lower mean rating of hypochondriasis: remission - Shorter mean duration of illness: remission - Higher level of overall functioning: remission Level of hypochondriacal symptoms[c] - More unrealistic fear of illness: higher - Higher scores on SSI: higher - Higher somatosensory amplification: higher - Higher scores on somatisation (SCL-90): higher - Higher level of neuroticism: higher - Older age: higher - More social adjustment: higher	*P*<.05 *P*<.05 *P*<.05 *P*<.05 *P*<.05 *r*=0.4; *P*=.01 *r*=0.4; *P*=.01 *r*=0.39; *P*=.01 *r*=0.38; *P*=.02 *r*=0.36; *P*=.02 *r*=0.34; *P*=.02 *r*=0.34; *P*=.02
Barsky et al., 1998[33]	(1) Remission of hypochondriasis	(a) Hypochondriacal symptoms (WI and SSI) (b) Hypochondriacal somatic complaints (26-items SSI) (c) Symptoms amplification (d) Functional status (e) Psychiatric comorbidity (f) Medical comorbidity	Remission of hypochondriasis - Decreases in hypochondriacal somatic complaints: remission	*P*<.05
Barsky et al., 2000[32]	(1) Remission of hypochondriasis	(a) Hypochondriacal somatic complaints (26-items SSI) (b) Somatosensory amplification (c) Normative beliefs about health and sickness	Remission of hypochondriasis - The 3-way interaction of health norms x hypochondriacal somatic complaints x amplification significantly increased the likelihood of a diagnosis of hypochondriasis af follow-up	OR=0.98
Barsky et al., 1993[34]	(1) Hypochondriacal symptoms (2) Somatisation (3) Disability	(a) Age (b) Gender (c) Personality disorder (d) Health status (e) Somatosensory amplification (f) Hypochondriacal symptoms (WI) (g) Hypochondriacal somatic complaints (SSI) (h) Intermediate activities of daily living	Number of hypochondriacal symptoms - Higher somatosensory amplifications: more hypochondriacal symptoms	*P* not given
Fernandeze et al., 2005[38]	(1) Persistent health anxiety	(a) Depression / anxiety (b) Negative affectivity (c) Somatic discomfort (d) Personal and family experiences related to illness throughout childhood (e) Current stress and illness (f) Sociodemographics (g) Satisfaction with medical attention (h) Evaluation of state of health (i) Degree of health anxiety	Persistent health anxiety[b] - Less positive medical self- evaluation of health problems: persistence - Greater degree of self-judged health anxiety: persistence	*P*=.031 *P*=.049
Simon et al., 2001[39]	(1) Persistence of hypochondriasis	(a) Anxiety (b) Depressive disorder	No significant association found	

WI: Whitely Index; SSI: Somatic Symptom Inventory; SCL-90: Symptom Checklist-90.

[a] Adjusted estimates and 95% CI.

[b] Only univariate results available (crude estimates and 95% CI, significant differences of associations).

[c] The measure of hypochondriacal symptoms at follow-up was the sum of the Whitely Index x 5.6 + the Somatic Symptom Inventory.

(GHP), degree of pain, physical functioning and illness behaviour. It is unclear whether female gender predicts an unfavourable course of MUS as two studies found that gender was of prognostic significance, whereas one study found that gender was not of prognostic significance.[31] Studies on comorbid mental health problems such as affective state and alexithymia showed conflicting results.[29,30,36]

We found weak evidence that poor self-evaluation of overall health and for occupational disability at baseline predicts persistence of abridged somatisation.[40]

Potential prognostic factors on recovery of hypochondriasis were studied in four publications (see Table 3).[32,33,35,39] We found some evidence for the number of somatic complaints on the Somatic Symptom Inventory (SSI) at baseline predicting recovery of hypochondriasis. A higher score predicts persistence of hypochondriasis (Table 4).[33,35] Furthermore we found weak evidence for the prognostic value on the course of hypochondriasis of symptoms scores on the Whitley Index, rate of severity of hypochondriasis, duration, level of functioning and degree of unrealistic fears of illness. Again, it looks like that the more serious the condition at baseline, the more unfavourable the outcome (i.e. persistence of hypochondriasis). Psychiatric comorbidity seems not to influence the course of hypochondriasis,[33,35,38,39] whereas somatosensory amplification seemed to influence the outcome of hypochondriasis in two studies.[34,35]

Discussion

Main results

Although a lot of research is done on the epidemiology of, and interventions for medically unexplained symptoms, we are not aware of a systematic review of the literature that focuses on the course and the prognosis of medically unexplained symptoms. Creed and Barsky[7] performed a systematic review of the epidemiology of somatisation disorder and hypochondriasis to examine the characteristics and associated features of these disorders. However, they did not systematically search and study prognostic factors.[7] So, this is the first systematic review which systematically searched for studies on prognostic factors in this area.
Generally, the included studies were of good quality. However, the heterogeneity between those included studies regarding clinical setting, numbers enrolled in the cohort, duration of follow-up, loss to follow-up, prognostic factors and outcome measures used is considerable. This limits direct comparability of the studies and makes it difficult to draw reliable conclusions.

The studies on MUS and abridged somatisation showed improvement rates of 50% or more. This is better than we expected. However, 10 to 30% of patients with MUS deteriorate. Given the large

Table 4. Strength of evidence of prognostic factors with a significant influence on outcome in multivariate analysis

Prognostic factor	Outcome	QS>60%	QS<60%	Strength of evidence
MUS				
Affective state/depressivity	Symptom change	½ (50%)	-	Inconclusive
Female gender	Symptom change	2/3 (66%)	-	Inconclusive
Alexithymia	Symptom change	1/2 (50%)	-	Inconclusive
Symptom duration	Symptom change	1/2 (50%)	-	Inconclusive
Number of symptoms	Symptom change	2/2 (100%)	-	Strong
Hypochondriasis questionnaire (WI)	Symptom change	1/3 (33%)[a]	-	Inconclusive
Initial GHP	Change in general health perception	1/1 (100%)	-	Weak
Number of physical symptoms	Change in general health perception	1/1 (100%)	-	Weak
Pain	Change in general health perception	1/1 (100%)	-	Weak
Physical function[b]	Change in global clinical improvement	1/1 (100%)	-	Weak
Age	Difference in functional impairment	1/1 (100%)	-	Weak
Illness behaviour subscale of IAS[b]	Change in medical care utilization	1/1 (100%)	-	Weak
Attribution	Quality of life	-	1/1 (100%)	Inconclusive
Abridged somatisation				
Self-rated overall health	Persistence of abridged somatisation	1/1 (100%)	-	Weak
Occupational disability	Persistence of abridged somatisation	1/1 (100%)	-	Weak
Hypochondriasis				
Health norms x somatisation x amplification[c]	Remission of hypochondriasis	1/1 (100%)	-	Weak
Hypochondriacal somatic complaints (SSI)	Remission of hypochondriasis	2/2 (100%)[d]	-	Strong
Hypochondriacal symptoms (WI)[b]	Remission of hypochondriasis	1/1 (100%)	-	Weak
Rating of hypochondriasis[b]	Remission of hypochondriasis	1/1 (100%)	-	Weak
Duration of illness[b]	Remission of hypochondriasis	1/1 (100%)	-	Weak
Level of overall functioning[b]	Remission of hypochondriasis	1/1 (100%)	-	Weak
Unrealistic fear of illness	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Hypochondriacal symptoms (SSI)	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Somatosensory amplification	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Somatisation (SCL-90)	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Neuroticism	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Age	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Social adjustment	Hypochondriacal symptom level	1/1 (100%)	-	Weak
Somatosensory amplification	Number hypochondriacal symptoms	-	1/1 (100%)	Inconclusive
Self-evaluation of health problems[b]	Change in health anxiety	-	1/1 (100%)	Inconclusive
Degree of self-judged health anxiety[b]	Change in health anxiety	-	1/1 (100%)	Inconclusive

Only factors are presented which scored significant associations in at least one study.
QS: Quality score; IAS: Illness Attitude Scale.

[a] Significant association only in one study with univariate analysis.

[b] Only univariate analysis available

[c] Only the three-way interaction significantly improved the model and increased the likelihood of a diagnosis of hypochondriasis at follow-up.

[d] Significant association in one study with multivariate analysis and in one study with univariate analysis.

numbers of patients presenting with MUS in primary and secondary care, deterioration of one third of these patients still means that large numbers of patients with MUS are going to get worse. The studies on hypochondriasis showed a less optimistic picture: the majority of these patients (50 tot 70%) do not recover during follow up. This might be due to the definition of hypochondriasis which requires patients to have symptoms for six months or more.

We did not find any prospective study on course or prognostic factors in patients with *DSM-IV* somatoform disorders. As the evidence for the number of symptoms at baseline in MUS as a prognostic factor originate from only two of the included MUS study, we conclude that, there is some evidence that the number of symptoms at baseline predicts the course of MUS. In the studies on hypochondriasis we found some evidence that the somatic symptom score on the SSI at baseline predicts the course of hypochondriasis. Furthermore, the condition of patients with MUS at baseline, represented by health perception and physical functioning, and the condition of patients with hypochondriasis at baseline, represented by rating of severity, physical functioning and duration of illness, showed a weak association with the outcome of MUS and hypochondriasis. So, we conclude that there is some evidence that the seriousness of the conditions of patients with MUS or hypochondriasis at baseline might be of prognostic significance.

We found only weak evidence for many other prognostic factors. Evidence on gender to be of prognostic significance was inconclusive. Remarkably, we found no evidence to support the influence of psychiatric comorbidity and personality traits on the course of MUS, abridged somatisation and hypochondriasis.

Comparison with the literature

Although only a minority of the MUS presented during consultation result in a chronic condition, patients with MUS are problematic in health care.[4] Physicians perceive these patients as difficult and demanding.[43,44] They also believe that patients with MUS increase health care costs due to sickness absence and service use, that they are at risk for unnecessary diagnostic procedures. Physicians express the need to prevent somatic fixation in these patients.[45-47] However, we found that the prognosis of MUS in primary and secondary care is more favourable then expected, as the majority of the patients with MUS improve. A possible explanation for this finding is that improvement of symptoms is partly caused by regression to the mean because symptoms are on their worst when selecting patients during primary or secondary care clinic visits.

However, our finding that the majority of the patients with hypochondriasis do not recover is supported by the literature in which hypochondriasis is considered to be a chronic condition.[3,48,49] Although, according to the literature, spontaneous recovery of hypochondriasis is rare, we found recovery rates of 35% to 50%. A possible explanation for this finding might be the procedure as required for inclusion in the study cohorts. This procedure is an extensive clinical assessment consisting of diagnostic interviews and additional testing and might in itself be of therapeutic importance.[50,51]

Giving the many factors hypothesized to be prognostic for a chronic course of MUS, somatisation disorder and hypochondriasis, there is not much evidence on these factors. Although personality traits, including neuroticism and alexithymia,[52,53] and psychiatric comorbidity, including anxiety and depression[54-56] have been demonstrated to be associated with MUS and hypochondriasis, only a limited number of studies have examined their prognostic value. In this review we did not find evidence for their prognostic value.[29,30,33,35-39] However, in well defined medically unexplained syndromes such as chronic fatigue syndrome and irritable bowel syndrome the evidence on prognostic factors is much stronger.[57-61] Cairns et al. found that less fatigue severity at baseline, a sense of control over symptoms and not attributing illness to a physical cause were associated with a good outcome.[57] Their findings of the prognostic significance of the fatigue severity at baseline is in line with our findings.

Strengths and limitations

In this systematic review, we used an extensive search strategy to identify relevant studies. We added rigor to our study by pre-testing the search strategy on publications about MUS in our own database and by searching all relevant databases without language restriction. Moreover, we had good interobserver agreement for in- and exclusion. Finally, we independently extracted data and assessed the quality of included studies with a validated checklist.

Because the quality of the individual study influences outcomes, we presented our results together with a quality score of each study. So, we visualize the susceptibility of each study for bias. Currently, no standardized method is available to assess the quality of prognostic studies. Therefore, we used a checklist of predefined criteria which has been used in previous prognostic reviews.[20,21]

The median number of participants enrolled in the cohorts of the included studies in this review is 87. Only one study on MUS, one study on abridged somatisation and none of the studies on hypochondriasis enrolled more than 200 patients into the cohort.[29,40] These low numbers of participants in the cohorts limits the strength of the evidence concerning outcome and prognostic factors.

Only a minority of the included studies presented sufficient data on the duration of symptoms at baseline. Therefore, it is not clear whether the study patients were all included at a similar point in the course of their disease. Studies reporting duration of symptoms at baseline showed a considerable range of duration of symptoms. This also limits the interpretation of our results.

Another limitation of this review is the absence of a detailed description of treatments during follow-up. The results of our study apply to the course of MUS, hypochondriasis and somatisation disorder in the medical system. We assume that during the studies in all patients some kind of treatment has been applied, although no study reported on this.

As statistical pooling was not possible because of the high heterogeneity of study populations, prognostic factors and outcome measures among included studies, we performed a best evidence synthesis. Although such a qualitative analysis is not as objective as a meta-analysis, we were able to summarize the value of prognostic indicators which takes the methodological quality into account.[62]

Implications for further research and clinical practice

The pessimistic views of GPs and their worries about the development of somatic fixation in patients with MUS and abridged somatisation might not always be justified as the majority of these patients generally have a favourable prognosis. However, the majority of the patients with hypochondriasis do not recover suggesting that hypochondriasis is a more severe condition.

Establishing the number of somatic symptoms and seriousness of the condition in patients with MUS or hypochondriasis during the first consultations might help GPs to value the risk of persistence and may guide GPs whether to offer only reassurance about the favourable prognosis or, for the high-risk patients, a more intensive approach such as reattribution. However, due to its heterogeneity, the data collated in this systematic review on prognostic factors are inadequate to identify predictors of the course of MUS, somatisation disorder and hypochondriasis. Therefore, it is difficult to advise clinicians how to distinguish between patients with low and high risks of persistence.

Although it is widely accepted that personality traits and comorbid depression and anxiety are associated with MUS, somatisation disorder and hypochondriasis, studies examining their prognostic value show conflicting results. As a consequence of the paucity of current research, there is need for more well conducted prospective cohort studies with a reasonable number of patients (>200 patients), in which assessment of treatments during follow-up and inclusion of patients at a similar point in the course of their disease are important topics.

Although we know for long that the doctor-patient relationship effects the outcome of consultations and can be therapeutic, none of the included studies took the doctor-patient relationship into account.[63-65] The more non-specific aspects of consultation such as described in the patient-centred clinical method needs attention in future research.[66]

Appendix A. Search strategy

(somatoform disorder [mesh] OR somatization [tw] OR somatisation [tw] OR hypochondriasis [mesh] OR neurasthenia [mesh] OR conversion disorder [mesh] OR somatoform disorder* [tw] OR hypochondriasis [tw] OR neurasthen* [tw] OR conversion disorder* [tw] OR psychophysiological disorder [Mesh] OR psychosomatic medicine [Mesh] OR psychophysiological disorder* [tw] OR psychosomat* [tw] OR psychosomatic medicine [tw] OR functional somatic sympt* [tw] OR functional somatic syndrom* [tw] OR functional syndrom* [tw] OR unexplained sympt* [tw] OR medically unexplained [tw] OR unexplained medical sympt* [tw] OR psychogen* [tw] OR non-organ* [tw] OR non-specific complain* [tw] OR non-specific sympt* [tw]) **AND** (incidence[MeSH:noexp] OR mortality[MeSH Terms] OR follow up studies[MeSH:noexp] OR prognos*[Text Word] OR predict*[Text Word] OR course*[Text Word]) **AND** (((Prospective studies [mesh] OR cohort studies [mesh] OR follow-up studies [mesh] OR observational stud* [tw] OR prospective stud* [tw] OR cohort stud* [tw] OR follow-up stud* [tw])) OR ((randomized controlled trial [pt] OR controlled clinical trial [pt] OR randomized controlled trials [mh] OR random allocation [mh] OR double-blind method [mh] OR single-blind method [mh] OR clinical trial [pt] OR clinical trials [mh] OR "clinical trial" [tw] OR ((singl* [tw] OR doubl* [tw] OR trebl* [tw] OR tripl* [tw]) AND (mask* [tw] OR blind* [tw])) OR "latin square" [tw] OR placebos [mh] OR placebo* [tw] OR random* [tw] OR research design [mh:noexp] OR comparative study [mh] OR evaluation studies [mh] OR follow-up studies [mh] OR prospective studies [mh] OR cross-over studies [mh] OR control* [tw] OR prospectiv* [tw] OR volunteer* [tw]) NOT (animal [mh] NOT human [mh])))

Appendix B. Criteria list for assessing the methodological quality of prognostic cohort studies on chronic medically unexplained symptoms

Criteria	Score
Study population	
A. Description of inception cohort	+ / - / ?
B. Description of study population	+ / - / ?
C. Description of relevant inclusion and exclusion criteria	+ / - / ?
D. Definition of chronic functional somatic symptoms	+ / - / ?
E. Number of subject in study population ≥ 200	+ / - / ?
Response	
F. Response rate ≥ 75%	+ / - / ?
G. Information about non-responders versus responders	+ / - / ?
Follow-up (extend and length)	
H. Follow-up of at least 12 months	+ / - / ?
I. Loss-to-follow-up < 20%	+ / - / ?
J. Information about completers versus those loss-to-follow-up	+ / - / ?
K. Prospective data collection	+ / - / ?

Treatment	
L. Description of possible treatment in cohort	+ / - / ?
Outcome	
M. Clinically relevant outcome measures	+ / - / ?
N. Standardized assessment of symptom outcome	+ / - / ?
O. Standardized assessment of functional outcome	+ / - / ?
Prognostic factors	
P. Standardized assessment of potential prognostic factors	+ / - / ?
Analysis	
Q. Appropriate univariate crude estimates	+ / - / ?
R. Appropriate multivariate analysis techniques	+ / - / ?
Data presentation	
S. Frequencies of most important outcome measures presented	+ / - / ?
T. Frequencies of most important prognostic factors presented	+ / - / ?
U. Influence of prognostic factors presented	+ / - / ?

+, positive (design or conduct adequate);
-, negative (design or conduct inadequate);
?, unclear (insufficient information)

Appendix C. Explanation of the criteria of the checklist for methodological quality

A. Description of inception cohort
Positive if it is described in what setting the subjects were recruited (i.e. general population, patients attending the general practitioner, inpatient or outpatient setting).

B. Description of study population
Positive if it is described which subjects from the inception cohort are recruited and if age and sex are described.

C. Description of relevant inclusion and exclusion criteria
Positive if it is described how subjects were identified with chronic functional somatic symptoms (CFSS) or somatization.
+ = CFSS or somatization diagnosed by the general practitioner or standardized diagnostic interview
- = CFSS or somatization diagnosed by a (standardized) self-administered symptom checklist
? = not clear

D. Definition of chronic functional somatic symptoms
Positive if the definition is described of CFSS or somatization.

E. Number of subjects in study population ≥ 200
Positive if the number of subjects with CFSS or somatization in the study population was at least 200 at baseline.

F. Response rate ≥ 75%
Positive if response rate is at least 75%. Response rate: the number of patients in the study population, divided by the number of subjects in the inception cohort.

G. Information about non-responders versus responders
Positive if demographic or clinical information (such as age and sex) was presented for responders and nonresponders, or if there was no selective response, or no nonresponse.

H. Follow-up of at least 12 months
Positive if the follow-up period was at least 12 months.

I. Loss-to-follow-up < 20%
Positive if total number of patients with CFSS or somatization was at least 80% at the end of follow-up compared to the number of participants with CFSS or somatization at baseline. Loss to follow-up: the number of patients in the study population at baseline minus the number of patients at the main health status measurement for the main outcome measure at the end of follow-up, divided by the number of patients in the study population at baseline.

J. Information about completers versus those loss-to-follow-up / dropouts
Positive if demographic or clinical information (such as age and sex, disease characteristics and other potential prognostic predictors) was presented for completers with CFSS or somatization and those lost to follow-up at the main moment of outcome measurement, or if there was or no selective loss-to-follow-up, or no loss-to-follow-up.

K. Prospective data collection
Postive if main outcome measures on potential prognostic predictors was collected prospectively.

L. Description of possible treatment in cohort
Positive if treatment subsequent to inclusion in cohort is fully described or standardized. Also positive if no treatment is given.

\+ = treatment/multivariate correction for treatment in analysis, or no treatment given
\- = different treatment regimens, not clear how outcome is influenced by it
? = not clear if any treatment is given

M. Clinically relevant outcome measures
Positive if at least one of the following outcome measures is presented: CFSS / somatization diagnosis, symptoms, remission or recurrence, functional status, social functioning, lost days of work, quality of life, impairment, mortality.

N. Standardized assessment of symptom outcome
Positive if standardized questionnaires or objective outcome measurements of at least one of the following three outcome measures were used for each follow-up measurement:
a. CFSS / somatization diagnosis
b. Symptoms
c. Remission or recurrence

O. Standardized assessment of functional outcome
Positive if standardized questionnaires or objective outcome measurements of at least one of the following six outcome measures were used for each follow-up measurement:
a. functional status
b. social functioning
c. lost days of work
d. quality of life
e. impairment
f. mortality

P. Standardized assessment of potential prognostic factors
Positive if standardized questionnaires or objective measurements were used at baseline of at least 4 of the following 18 potential prognostic factors:
a. sex
b. age
c. marital status
d. family history of CFSS/somatization
e. race
f. social economic status (SES)
g. education level
h. number of episodes of CFSS/somatization
i. sick leave
j. functional impairment
k. comorbidity (i.e. anxiety disorder or chronic disease)
l. duration of symptoms
m. social support
n. stressful life events
o. difficult doctor-patient relationship
p. coping strategy
q. perception of symptoms (i.e. illness attitude, somatosensory amplification)
r. personality traits

Q. Appropriate univariate crude estimates
Positive if separate univariate (repeated measures) analysis of variance were calculated for each dependent measure.

R. Appropriate multivariate analysis techniques
Positive if multivariate (repeated measures) analysis of variance were calcultated for changes among the dependent measures occuring during the follow-up interval.

S. Frequencies of most important outcome measures presented
Positive if frequency, percentage or mean, median (interquartile range) and standard deviation/confidence intervals are reported of the most important outcome measures.

T. Frequencies of most important prognostic factors presented
Positive if:
a. frequency of percentage is reported, or
b. mean and standard deviation or standard error are reported, or
c. median and interquartile range are reported, or
d. if the influence of each separate factor is reported

U. Influence of prognostic factors presented
Positive if the influence of each separate prognostic factor on the natural course of CFSS or somatization is presented.

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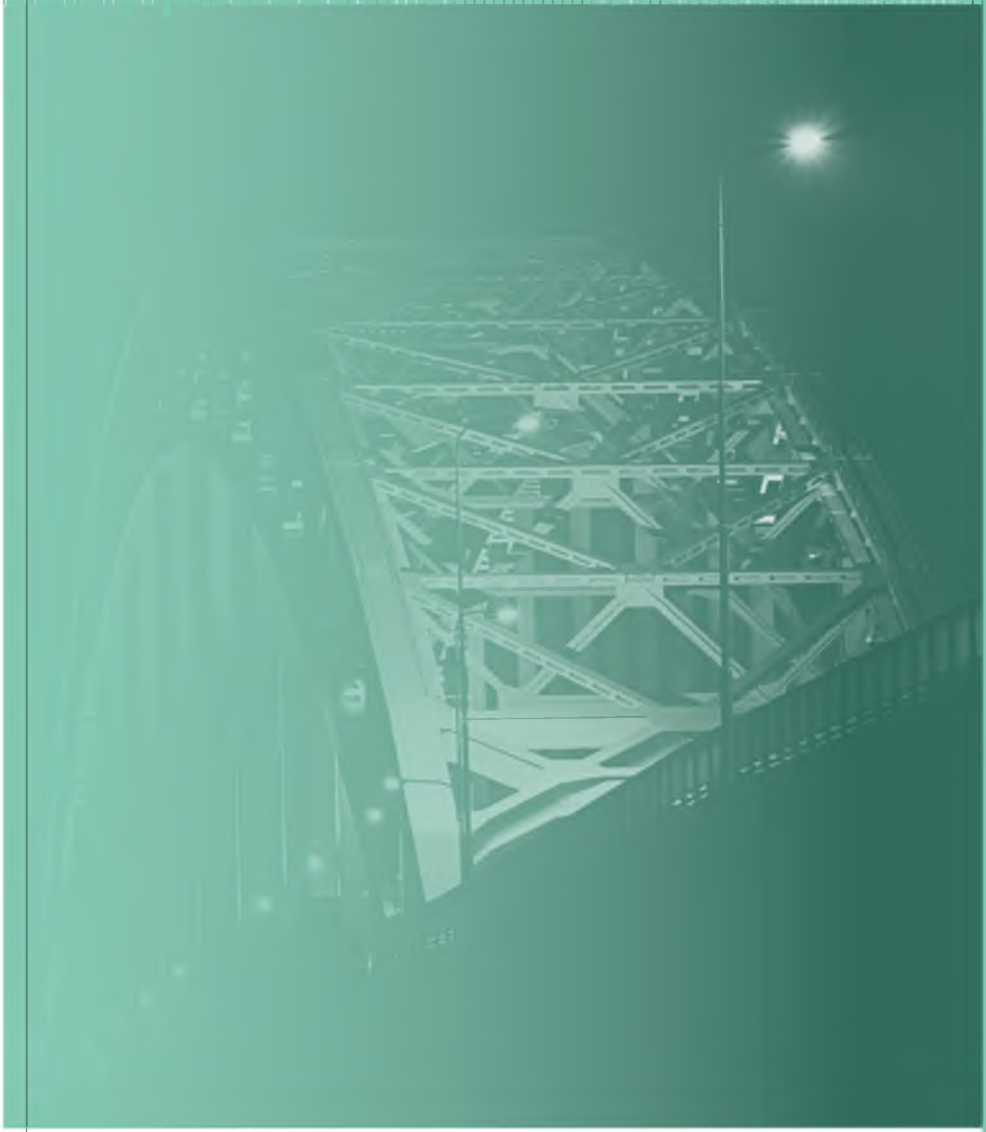
CHAPTER

4

THE DOCTOR-PATIENT RELATIONSHIP FROM THE PERSPECTIVE OF PATIENTS WITH PERSISTENT MEDICALLY UNEXPLAINED SYMPTOMS. AN INTERVIEW STUDY



Tim olde Hartman
Hiske van Ravesteijn
Eric van Rijswijk
Juke Nijenhuis
Peter Lucassen
Chris van Weel

Submitted for publication

Summary

Purpose. The interaction between FPs and patients with persistent medically unexplained symptoms (persistent MUS) is described as complicated. Although perspectives of patients with an initial presentation of MUS are studied, research on perspectives of patients with persistent MUS are lacking. Knowledge of opinions of patients with a long history of presenting MUS might guide interventions to improve the care for these patients.

Methods. A qualitative approach, interviewing 17 patients with persistent MUS. Data were analyzed using an iterative process according to the principles of constant comparative analysis.

Results. Patients with persistent MUS stressed the importance of a personal continuing doctor-patient relationship. Such a relationship is built on physician attitude, medical competence, availability and shared authority. Patients want to be taken serious in a non-judgmental open communication style. They appreciate a thorough exploration and a comprehensible explanation of the symptoms. Furthermore, they want a competent FP who is easy accessible and who engage them as partners in the consultation and the decision making process.

Conclusions. Patients with persistent MUS value a healing FP-patient relationship. They appreciate a patient-centered communication and orientation to care in which personal continuity and continuity of the relationship are important elements.

Introduction

Physical symptoms such as headache, back pain, dizziness and fatigue are common in the general population and most people do not contact professional medical care for these symptoms.[1-3] In those who do present these symptoms, physicians often do not find an organic cause (i.e. medically unexplained symptoms).[4;5] Fortunately, medically unexplained symptoms (MUS) have a beneficial course whether or not as a result of physicians' management.[6] Only a minority of patients will develop *persistent* MUS. Exactly these patients represent a serious problem in health care as they suffer from their symptoms, are functionally impaired, and are at risk for potentially harmful additional testing and treatment.[5;7]

The interaction between FPs and patients with *persistent* MUS is described as complicated. Encounters between FPs and these patients often leave both with frustration and confusion.[8-10] While the FP perspective on *persistent* MUS has been studied,[11;12] research on patients' views and experiences regarding the care they receive is still limited. Knowledge in this area is necessary in order to improve the care for these patients.

Existing knowledge regarding patient perspectives on MUS originates from patients with an *initial* presentation of unexplained symptoms.[11;13-15] This research pointed out that patients often feel stigmatized and not taken serious,[16] that they often wish to have a convincing, legitimating and empowering explanation for their symptoms,[17-20] and that they want emotional support from their FP.[21] Furthermore, analysis of videotaped consultations in primary care revealed a mismatch between what patients with *initial* MUS want and what they actually receive from their FP.[13;14] This mismatch might explain why patients frequently express their dissatisfaction with the medical care received during their illness. However, it is unclear to what extent these results can be generalized to patients with *persistent* MUS in primary care. Studies in patients with the distinctive functional syndromes chronic fatigue syndrome (CFS) and irritable bowel syndrome (IBS) concluded that patients feel dissatisfied because of the delay of or confusion over the diagnosis, the attitude of the doctor and the inadequate and often conflicting information given by their doctors.[22;23]

In order to improve the management of patients with *persistent* MUS in family medicine, we performed an interview study focusing on the opinions of patients with a long history of presenting MUS to their FP.

Method

Study sample

We conducted 17 semi-structured interviews with patients with persistent MUS who recently attended the FP. We selected 12 patients from the Continuous Morbidity Registration (CMR) project and 5 patients from three practices of the authors.

CMR project

We selected patients who were diagnosed with *persistent* MUS for the first time between 2006 and 2008 from the CMR database. We invited patients aged > 18 years, who were longer than one year on the practice list, without language barriers, psychiatric diseases (anxiety disorder, depressive disorder, PTSS, drug or alcohol abuse) or cognitive disabilities according to their FP (n = 59). These patients were sent a letter and if necessary a reminder in which we asked them to participate in an interview study regarding the quality of care they receive from their FP. A total of 31 patients responded to this letter and 12 patients volunteered to participate.

In the CMR project of the Department of Primary and Community care at the Radboud University Nijmegen Medical Centre every episode of illness seen by, or reported to, the FP is registered as soon as it is established using an adapted version of the E-list.[24-27] As far as we are aware, the CMR project is the only morbidity registration system with a structural possibility to classify patients with *persistent* MUS.[28;29]

Table 1. Patient characteristics

Characteristic	*n*
Age in years (mean (range))	50.4 (27-76)
Sex	
- Women	15
- Men	2
Level of education[a]	
- Low	5
- Middle	6
- High	6
Situation of living	
- With partner	9
- With partner and children	4
- Single	4
Time on practice list	
- < 1 year	0
- 1-3 years	2
- 3-5 years	2
- > 5 years	13

[a] Education level was classified as low (primary and lower secondary education), middle (upper secondary education, until age 17-18), and high (pre-university, higher vocational training, and university)

Authors practices

The 5 patients selected from the authors' practices were used to test the interview guide and to train the interview technique. They were selected as they persistently consulted with symptoms that could not be attributed to a clear organic cause (as described in the CMR project). We included these patients in the analysis as the interviews provided rich data.

Sociodemographic details (age, sex, marital status, level of education, and time on the practice list) were obtained from the participants. A summary of patients characteristics of the 17 interviewed patients is shown in Table 1.

Semi-structured interviews

One of the authors (JN) conducted the semi-structured interviews at the patient's home. These interviews were recorded, transcribed verbatim, and entered into Atlas.ti, a software program for the analysis of qualitative data. The interviewer was guided by a list of topics, based on important topics highlighted in the literature regarding patients' views on MUS. (see Table 2) Open questions were used to encourage patients to communicate their views on important aspects of the care they receive for their persistent MUS.

As the collection of data and the analysis in qualitative research is an iterative process, two researchers (ToH, PL) added relevant topics to the interview topic list after a preliminary analysis of the first five interviews. Ideas and thoughts that emerged in primary stages of the analysis were brought back to subsequent interviews as the study proceeded.

Analysis

The interviews were analysed independently by two researchers (ToH, HvR) after reading the transcripts several times to familiarize themselves with the data. The two researchers coded the transcripts and compared and discussed these codes with each other. Codes in each interview were compared with those in other interviews. Additional codes, which emerge from discussions were also applied to the transcripts. Concepts and categories emerged through this iterative process of coding, analysis and discussion (constant comparative qualitative analysis).[30;31] The results of the analysis were discussed with a third researcher (PL). Data collection continued until saturation was reached and no new themes emerged.[32]

Table 2. Interview guidebook

Understanding, recognition and support - Do you think your FP understands your symptoms and problems? - Are you satisfied with the help and support you get from your FP? - Does your FP take your symptoms serious?
FPs' consulting behaviour - Did you discuss your own thoughts and/or concerns regarding your symptoms with your FP? - Did you get enough space to communicate your own feelings and emotions during the consultations? - Did your FP do additional diagnostic tests, and what do you think of that? - Did your FP give you the advice to come back for a new, possibly longer consultation?
Time and attention - Do you think you get enough space and time to discuss your symptoms and problems? - Do you think your FP has enough attention during the consultation? - Did you feel hurried during the consultation?
Knowledge and interest in the patient's context - Does your FP know about your personal circumstances? - Did your FP discuss the influence of your symptoms/problems on your daily life? - Did you discuss ways of coping with your symptoms/problems during consultation?
Explanation and reassurance - How did your FP explain the symptoms to you? - Did your FP give you a diagnosis for your symptoms? - What did you think of the explanation of your symptoms, did it satisfy you? - Did your FP give you advice on how to handele your symptoms?
Shared decision making - What did you expect your FP was going to do (referral, additional diagnosic tests, etc.) with your symptoms? - Did your FP explain/discuss the treatment plan to/with you? - Did you and your FP make a shared decision on the treatment plan?
Trust - How do you characterize/perceive the doctor-patient relationship? - Did the doctor-patient relationship change over time?
Quality of care - Are you satisfied with the quality of care you received from your FP for your symptoms/problems? - Which elements in the quality of received care are important and which are less important for you?

Results

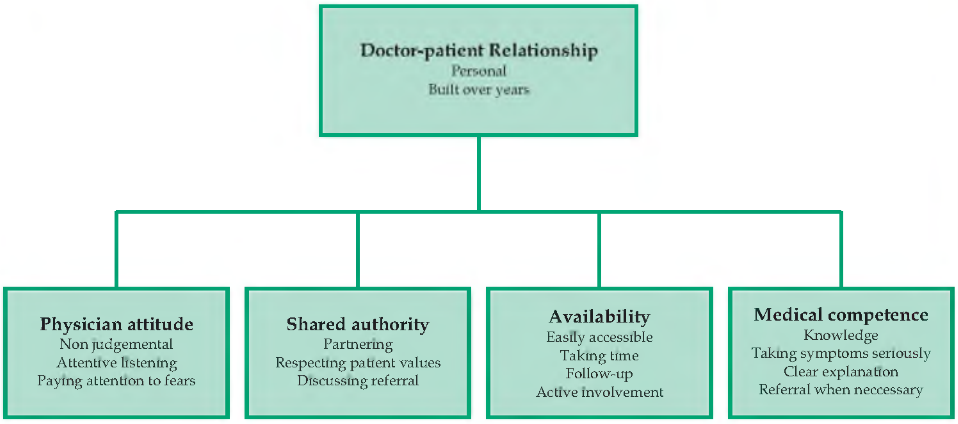
FP-patient relationship

Patients indicate that they want a warm, personal continuing FP-patient relationship. Such a relationship makes it easier for patients to discuss symptoms and problems with their FP. Patients mention that there has to be a match between the FP and the patient. Furthermore, they state that the FP-patient relationship is built over time.

> *'it used to be that the distance between the doctor and the patient was really big. Now, the distance is much smaller. You're basically on the same footing now [...] I find that much nicer'*
> (P1: female, age 59)
> *'I have the sense that it clicks. I feel good and safe there. I don't feel like that there are certain things I would be reluctant to say'* (P13: female, age 28)
> *'I've had the same FP since I was 19 so you really do know each other, also because, before all the vague complaints started, I would go to my FP for this or that, a sore throat and what not. And, actually, my history of vague health complaints is already so long that we've come to know what to expect from each other and how to play into each other.'* (P3: female, age 33)

We could distinguish four key-themes on which, according to the interviewed patients, a satisfying FP-patient relationship is build: physicians attitude, medical competence, availability and shared authority. These components are described in Figure 1.

Figure 1. Doctor-patient relationship model according to patients with persistent MUS



1. Physician attitude

Patients state that they want an FP with an open mind using a non-judgmental communication style. Such a communication makes patients feel at ease and free to tell their story. Furthermore, it makes patients feel free to visit their FP. Patients expect an FP to be an attentive listener showing a lot of understanding.

> *'and then he says, have a seat. And then I start to talk, of course (…) and he listens. (…) he let's me first tell my story (…) I can say whatever I want to say. So, that's nice.'* (P4: female, age 29)
> *'she has an active way of listening. She knows me (...) then responds and her voice is so understanding. It's like, Okay, I understand that is very frustrating for you.'* (P3: female, age 33)
> *'You just notice that she really listens. She is not thinking, what am I going to eat this evening?'* (P13: female, age 28)

Patients appreciate FPs who take the symptoms as well as the person seriously. When patients notice that the FP does not take them seriously, they hesitate or even postpone to go to the FP. Furthermore, they feel powerless, according to the interviewed patients. Knowing that the FP takes him or her seriously is reassuring. Some patients fear that frequently visiting the FP will have a negative influence on the FP's attitude. Patients state that it can take quite some time before they have the feeling of being taken seriously.

> *'I'm always reluctant to go to my FP because I don't feel like I am taken seriously. (…) It's really humiliating and it makes you really feel sad because you have pain and you can't explain it. You can't just say, I do have pain! I need more tests.'* (P5: female, age 76)
> *'I: Do you have any idea how long it took before you got the impression that he does take you seriously, that he believes you? P: Well, I think that it took about a year.'* (P9: male, age 75)*
> * *I = interviewer; P = patient*

Patients appreciate it if the FP pays attention to emotions. When the FP is not paying attention to the patient's fears, this can result in a feeling of not being taken seriously. Patients state that talking about the fear is not always enough, sometimes a referral for reassurance is necessary.

> *'so that more attention is paid to your feelings? Yeah, I'd definitely appreciate that. Yes.'* (P1: female, age 59)
> *'I: And were you able to then discuss that fear with your FP? P: Yes, I did do that. I said, I am scared. And he dealt with that well. He said, What are you scared of? I am scared that I have some kind of cancer in my bowels. Then he examined my belly and he started to talk about stress (…) I really appreciated the fact that he did that but, in the back of my mind, I thought (…) I think that he kind of felt that and so he said, Well, I'll refer you to your specialist.'* (P6: female, age 60)

2. Medical competence

Patients want a skillful and competent FP. Someone who, from the first presentation, takes a close look at the symptoms to find out what is wrong and who gives good advice. They want a doctor who reacts quickly, sharply and adequately to the symptoms presented during the consultation. Furthermore, the FP's attitude has to be positive and supporting.

> *'I'll take care of that for you. I know we'll make it through this.'* (P1: female, age 59)
> *'that it's properly examined to determine if something is wrong or if it is serious or if it just requires some rest, that someone just takes a look to find out what's going on and just gives you good advice.'* (P13: female, age 28)

When the FP and the patient together get stuck with the symptoms, some patients stress that referral to specialty care is necessary.

> *'sometimes, I think, send someone to the specialist earlier. All too often, it's we'll try this medication first. If it doesn't work, we'll try another kind of medication. Then you can keep coming back and only after a really long time do you get a referral.'* (P5: female, age 76)

Some patients stress the importance of getting a diagnosis for their symptoms. Patients state that such a diagnosis is important both for themselves and for their FP. However, patients realize and accept the difficulty FPs face when searching for a diagnosis for their vague complaints.

> *'on the one hand, it changes absolutely nothing. On the other, it allows me to label it and then I can look for a solution more effectively. And I think that it is also important for my FP. Then he can also be more focused and effective in looking for a solution together with me.'* (P3: female, age 33)

All patients stressed the need for an explanation for their symptoms in comprehensible language. Different explanations from different doctors confuse patients. Patients expect explanations about what is going on in their body. They want to know where their symptoms come from and how their body functions.

> *'Then she explains it to me in a really simple fashion. She says, this and that…and that's why your body reacts differently and that's why you have those complaints. (…) Now she is the first that has given me that kind of concrete information'* (P8: female, age 27)

Patients also mention examples of explanations which contain a link between physical and psychological processes.

> *'He explained that the mind and the body are connected, and that maybe, yeah, if (...) the psychological pressure were off (…) then maybe things would improve. So, I totally understood the explanation.'*
> (P8: female, age 27)

If the FP does not know the origin of the symptoms, patients appreciate the doctor mentioning this.

Experiences of the past are taken into account when patients judge the FP's medical competence. Patients regard a good past performance of the FP in diagnosing and treating symptoms as very important: it strengthens the FP's medical competence and patients' trust in the doctor.

> *'earache, ear drops - you walk around with this for a week and only then do they send you to the hospital. You end up with a double ear infection that has to be lanced …yeah, then I'm angry!'*
> (P5: female, age 76)
> *'usually he [the FP] tries to figure it out himself He suspected that there was something in my knee because of strain or overuse. And what did it turn out to be? Indeed, it was strain. He was right!'*
> (P4: female, age 29)

Sometimes patients question the medical competence as a result of stories of other patients in whom something went wrong.

3. Availability

Patients have experienced quite some frustrations concerning the access to their FP. They mention that their 'own' FP is not available during evenings, nights and weekends. Moreover, not all FPs work fulltime. Some patients find it difficult that they have to wait several days before they can see their FP. Furthermore, patients dislike discussions with the practice nurse before they are allowed to have an appointment with their FP.

> *'He only works 2 or 3 days a week. So, usually, when you call to make an appointment, he's already booked. The first available appointment is normally about 2 weeks away. I think that's really unfortunate.'* (P11: female, age 52)
> *'Whenever you call for something or about something, the assistant always screens the calls and won't put you through. I want to speak to the doctor himself about something. Then you need to do a lot to be put through, (…) I say, I want to speak to him directly, and then the assistant says, But what is it about? And, it's not possible…and it's constant blocking'* (P11: female, age 52)

Patients state that they need an FP who takes time for them. The limited consultation time sometimes has a negative influence on the contact with the FP, many patients consider the standard of 10 minutes consultation time too short. Some mentioned the long time they spend in the waiting room and the feeling of hurry which this causes during the consultation.

> *'It's not like, you're in, you're out. He really does take the time for you. He lets you finish talking.'* (P4: female, age 29)
> *'Yeah, you always have to wait. You're never seen on time (...) I feel the time pressure and I see it clearly in him [the FP]. (…) Obviously, that's not good.'* (P11: female, age 52)

Patients appreciate follow-up appointments, especially when initiated by the FP, and the possibility of longer consultation time. Active involvement from the FP, for example visiting a patient spontaneously after a major event, is highly appreciated by the patients.

> *'I had to come back on regular intervals - every three or four weeks or so. Not that they send you out on your own to figure it out. No, it's nice that they keep tabs on you.'* (P4: female, age 29)
> *'I think that they are really concerned and involved and empathic. We have experienced this over the past couple of months [as my father was being cared for prior to his passing] in many different ways. (…) The FP was wonderful. We got phone calls asking how he was. I am really happy with the guidance and help we received from the FP.'* (P1: female, age 59)

4. Shared authority

Patients state that they want to be engaged as partners in the consultation, to look and search together for possibilities and solutions. Patient appreciate the FP for initiating the search for solutions and for explaining why a certain solution is preferable. However, some patients want to make their own choice based on the provided information. They expect the FP to respect their decision.

> *'we think about things together. It's not one-way like that I have to think about things on my own. Together, we come up with the best approach and determine what's possible.' [...] Together, we try to find the best solution, and then I trust that she [the FP] is the one who takes the lead. That's best.'*
> (P10: female, age 52)
> *'she [the FP] helps me make decisions by providing the information needed to make the decision'*
> (P3: female, age 33)

When patients themselves provide solutions, for example in complementary or alternative medicine, they expect support and a positive reaction from their FP. Some patients stress the importance of clarifying their own ideas, thoughts, wishes and needs to the FP.

> *'the FP wasn't open to that. In any event, he didn't suggest it. And when I then said, maybe this is something for a homeopath?, he said, Yeah, that's up to you. I don't really have a lot of faith in that. So, obviously, I'm not really encouraged to explore those options. I don't think that's very good.'*
> (P11: female, age 52)
> *'Well, then, at a certain point in time, I just decided to go see a craniosacral therapist. He thought that was odd. It's crazy that she [the FP] didn't tell me that that also exists.'* (P1: female, age 59)

Patients stress their problems with FPs resistance for referral. But when an FP immediately agrees with the requested referral or additional test, patients mentioned to be surprised. A positive response to a request for referral is not always necessary as long as the FP has a proposal for further inquiry.

> *'then you have to practically beg: Can you please refer me to the neurologist?'* (P7: female, age 49)
> *,I said, at home, to my husband, I am going to demand a bowel examination. I was incredibly surprised that he [the FP] was okay with that. I expected that he would say that that wouldn't be necessary.'*
> (P6: female, age 60)
> *'He [the FP] always tries to find a way to help you himself I really appreciate that. It's better than being referred to all sorts of other practitioners and agencies.'* (P4: female, age 29)

Discussion

Summary of main findings

Patients with persistent MUS stressed the importance of a personal continuing FP-patient relationship. According to these patients, such a relationship is built on four components: physician attitude, medical competence, availability and shared authority. Patients want to be taken serious by an FP with an open mind who uses a non-judgmental communication style. Furthermore they want a skillful and competent FP who is supporting and offers a comprehensible explanation for their symptoms. Patients with persistent MUS also want easy access to their 'own' FP who takes time to explore and discuss the patients' symptoms and/or problems. Finally, patients want to be engaged as partners in the consultation in a way they can make their own choices based on the provided information.

Comparison with existing literature

Studies on opinions of *persistent* MUS patients in primary care regarding the doctor-patient relationship are scarce. Studies on expectations of patients during the *initial* presentation of MUS suggest that physician attitude and medical competence are important at the beginning of an episode of unexplained symptoms. However, our patients with *persistent* MUS considered *also* the availability of the doctor and shared authority of major importance. This is probably due to the fact that during the course of *persistent* MUS personal continuity (i.e. the importance of seeing a personal doctor) and continuity of the relationship with the FP becomes more and more important.

Shared authority reflects the patients' expectations concerning their FP to listen to their treatment proposals and to go through a process of shared decision making. Churchill interviewed fifty practitioners, who were identified as "healers" by their peers. Sharing authority appeared to be one of the eight healing skills. This skill is described as sharing the responsibility for healing at the very beginning of the consultation, recognizing the patient as a 'fellow expert', with a particular level of expertise, and having and showing confidence in the relationship with the patient.[33] Scott conducted in-depth interviews about healing relationships with FPs and their patients. A key process which emerged from these interviews was 'appreciating power'. Engaging patients as partners in decisions about diagnosis and treatment was seen as quintessential. These FPs reported that most often they work to increase patients' power.[34]

Patients opinions on the doctor-patient relationship in our study (Figure 1) show great overlap with 'the healing relationship model' of Scott et al. and 'the eight healing skills' of Churchill et al.[33;34] However, the patients interviewed by Scott et al. were not selected on basis of their illnesses, but they suffered from different chronic illnesses. Although we selected patients based on the presence of *persistent* MUS, their perspective upon the relationship with the doctor does not seem to differ from the other patients with chronic conditions. Furthermore patients' opinions regarding the doctor-patient relationship in our study seem to be in line with the opinions of a specific group of practitioners regarding healing skills. Churchill described the importance of taking time for the patients and demonstrating that there is space for their story. This fits perfectly with the wish of the patients with persistent MUS: they want their FP to be mindfully present, listening to their story and giving them room to tell their story.

Strengths and limitations

This is the first study on opinions of patients with *persistent* MUS about the doctor-patient relationship. Most of the patients interviewed in our study know their FP for more than 5 years. Furthermore they are included in the study because they visit their FP frequently with MUS and because they are all currently receiving care for persistent undifferentiated MUS from their FP. Therefore, our results reflect the opinions of patients who have had recent and relatively many contacts with the health care system and their FP. These patients have built their opinions about the doctor-patient relationship on a broad experience with medical services. As the attitude of FPs towards patients with MUS is often negative,[11] one could expect that the interviewed patients are more likely to have experienced their consultations as unsatisfactory and stressful and therefore they might be more critical about the doctor-patient relationship than others. On the other hand, most patients included in this study know their FP already for more than 5 years, which possibly implicates satisfaction with their FP.

The qualitative method has been recommended as the best method to explore and clarify patients' opinions.[35] By using a cyclical and interactive way of collecting and analyzing data, 'progressive focusing' and exploration of patients' opinions in depth was possible.[36] Transcribing the interviews verbatim, entering the full texts into Atlas.ti, and coding and re-organizing data by two independent researchers, strengthens our findings. Although small, the number and characteristics of participants included in our study are considered adequate for capturing an optimal variety of opinions.[37] The findings presented here, however, reflect the perceptions of a small group of patients and may not represent the views of patients in general. The validity of our analysis is enhanced by the diversity of training and experience of the analysis team (experienced FP and a psychiatrist) and reflexivity (reflecting on our own experiences).[38]

A weakness of the study is that patients were recruited by sending them a letter and a reminder to ask for participation in a study regarding the quality of care they receive from their FP. Only 12 of the 31 patients (39%) agreed to participate. This might have caused response bias as patients who are satisfied with their FP are possibly more inclined to participate in such a study. Therefore, certain specific viewpoints could have been left out of sight in this study.

Interviewed patients did not mention any expectation about receiving information about the prognosis of the symptoms. None of the patients made remarks about discussing the duration of symptoms. This might be due to the fact that almost all interviewed patients had chronic complaints and assume that the doctor will not be able to predict the duration of the symptoms.

The doctor-patient relationship is built over time and over many consultations. Interviewing patients regarding their opinions on the doctor-patient relationship gives only a snapshot in time. Patients may change their perspective over time as a response to the symptoms, diseases, life events, and/or other circumstances.

Implications for further research and clinical practice

Like patients presenting *initial* MUS to their FP, patients with *persistent* MUS want an medical competent FP with an open non-judgemental attitude. However, patients with *persistent* MUS *also* want personal continuity and a therapeutic doctor-patient relationship.[39] An easily accessible FP who is willing to see the patient as a partner in the decision making process contributes to a patient-centered orientation to care which is highly valued by these patients.

The doctor-patient relationship largely depends on the communication style of the FP. Further research should focus on studying and maximizing the therapeutic effects of FPs' communication to achieve better health outcomes in patients with persistent MUS. As patients appreciate a non-judgmental and attentive listening style, educational programs in health communication interventions as mindful communication might be of help.[40;41] Furthermore, studying the effects of the (improvement of the) quality of the FP-patient relationship on symptoms, impairment and satisfaction of patients will gain significant insights into the how FP-patient relationships contribute to healing and well-being of patients with *persistent* MUS.

Acknowledgements

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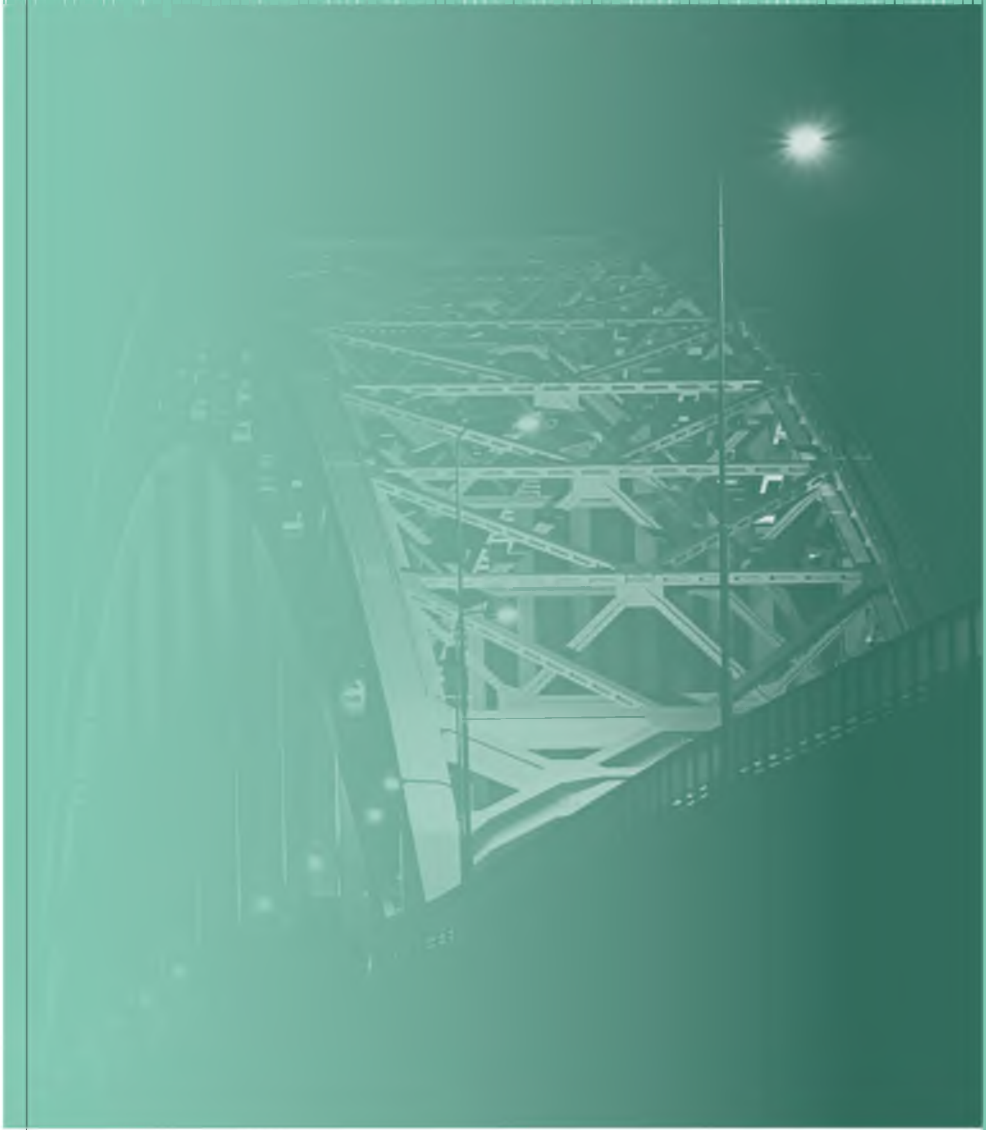
CHAPTER

5

EXPLANATION AND RELATIONS. HOW DO GENERAL PRACTITIONERS DEAL WITH PATIENTS WITH PERSISTENT MEDICALLY UNEXPLAINED SYMPTOMS: A FOCUS GROUP STUDY



Tim olde Hartman
Lieke Hassink-Franke
Peter Lucassen
Karel van Spaendonck
Chris van Weel

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Abstract

Background. Persistent presentation of medically unexplained symptoms (MUS) is troublesome for general practitioners (GPs) and causes pressure on the doctor-patient relationship. As a consequence, GPs face the problem of establishing an ongoing, preferably effective relationship with these patients. This study aims at exploring GPs' perceptions about explaining MUS to patients and about how relationships with these patients evolve over time in daily practice.

Methods. A qualitative approach, interviewing a purposive sample of twenty-two Dutch GPs within five focus groups. Data were analyzed according to the principles of constant comparative analysis.

Results. GPs recognise the importance of an adequate explanation of the diagnosis of MUS but often feel incapable of being able to explain it clearly to their patients. GPs therefore indicate that they try to reassure patients in non-specific ways, for example by telling patients that there is no disease, by using metaphors and by normalizing the symptoms. When patients keep returning with MUS, GPs report the importance of maintaining the doctor-patient relationship. GPs describe three different models to do this; mutual alliance characterized by ritual care (e.g. regular physical examination, regular doctor visits) with approval of the patient and the doctor, ambivalent alliance characterized by ritual care without approval of the doctor and non-alliance characterized by cutting off all reasons for encounter in which symptoms are not of somatic origin.

Conclusion. GPs feel difficulties in explaining the symptoms. GPs report that, when patients keep presenting with MUS, they focus on maintaining the doctor-patient relationship by using ritual care. In this care they meticulously balance between maintaining a good doctor-patient relationship and the prevention of unintended consequences of unnecessary interventions.

Background

In 25 to 50 percent of the contacts patients present medically unexplained symptoms (MUS) to the general practitioner (GP).[1,2] Although it is only a minority (2.5 percent) of these contacts that will result in a chronic condition associated with recurrent consultations, extensive investigations and referrals, this minority of chronic patients represents a serious problem in primary care.[3] Persistent presentation of MUS is troublesome for the GP because many GPs experience difficulties in the communication and the relation with these patients.[4-11]

There is evidence that the difficulties in communication may result from misperceptions of patients' needs and worries by GPs.[12] GPs feel pressurized by patients to apply biomedical interventions but they do not have much to offer in a strictly biomedical way.[13,14] At the same time, most patients with MUS do not overtly insist on additional somatic interventions. They primarily want to be understood and seek emotional support, which doctors do not provide.[15-18] Moreover, the lack of a biomedical explanation hinders GPs in adequately telling patients what is wrong.[19] As a consequence patients' needs are unmet, reassurance will be hampered and relief of symptoms will be complicated.[20,21]

Recent research has shown that the current management of patients with MUS should consist of: communicating to the patients that the symptoms are real, making patients feel understood, engagement of the GP to establish common ground with the patient, offering a detailed explanation about the nature of the complaints, and if necessary symptomatic relief.[22-25]

By definition, patients with persistent MUS will keep attending the consulting hours of their GPs with MUS. However, they will also face other health care problems, for example serious somatic disease.[26] Given the importance of a good relation with the patient in the light of continuity of care, GPs have to find strategies to specifically deal with these patients.[10] To our knowledge it is not known how GPs do this and how, according to GPs, this relationship evolves over time.[27] Furthermore, we need to know GPs' opinions about explaining the nature of unexplained symptoms to patients with persistent MUS during consultations, in order to develop more effective interventions for these patients.

In this qualitative exploratory study we focus on GPs' perceptions of giving explanations to patients with persistent MUS and on GPs' perceptions about the doctor-patient relationship and how the relationship evolves over time.

Methods

We conducted five focus groups with 22 Dutch GPs to study their views on MUS. Each focus group consisted of 4 to 5 GPs. We used a purposive sampling strategy to increase the external validity of our results with respect to the variety of existing views among GPs. From the literature, we considered the following characteristics as relevant for this variety: age, gender, working experience, number of listed patients, 'academic working career', geographical location of practice (city versus rural) and site of education.[28-31]

Each focus group was homogenous for the characteristics 'academic working career' and 'geographical location' (Table 1); participants otherwise represented a variety of the listed characteristics (Table 2).

We chose focus groups rather than individual interviews to use group interaction which stimulates participants to explore and clarify their views into more depth.[32] Discussions were facilitated by a skilled moderator, and lasted for approximately one and a half hour.

Following the guidelines for conducting focus groups, the moderator used a discussion guide to direct the discussion and to fulfil the research aims (Table 3). The discussion guide was mainly based on important topics highlighted in the literature.

The discussions were tape-recorded with the participants' consent and completely verbatim transcribed. Data collection and analysis proceeded as an iterative process. Two researchers (ToH, LH) added relevant and new topics to the discussion guide after a preliminary analysis of each session. In this way, ideas and thoughts that emerged in primary stages of the analysis were brought forward in subsequent focus groups as the study proceeded.

Finally, the first author verified the transcription and entered all data into Atlas.ti, a software program used to support the analysis of qualitative data.

Table 1. Focus group charcteristics

	characteristic
Focus group 1	GPs with an academic working career in Radboud University Nijmegen Medical Center
Focus group 2	GPs without an academic working career working in a rural area
Focus group 3	GPs without an academic working career working in a rural area
Focus group 4	GPs with an academic working career in VUmc Amsterdam or AMC University Amsterdam
Focus group 5	GPs without an academic working career working in a city

Table 2. Key characteristics of purposive sample of general practitioners

	Number of general practitioners
Gender	
Male	14
Female	8
Working hours	
Full time*	10
Part time	11
Not practicing at the moment	1
Type of practice	
Solo	1
Duo	2
Group	17
Variable	1
Not practicing at the moment	1
Urbanization	
Rural	3
Suburban	6
Urban	11
Variable	1
Not practicing at hthe moment	1
Age in years (range)	47 (31-58)
Experience as a GP in years (range)	15 (0-30)

* full time: 80% to 100% full time

Table 3. Focus group interview guidebook

What are the characteristics of patients with persistent MUS?	- Regarding patient characteristics? - Regarding symptom characteristics? - Do you have problems to recognize these patients?
How do you call patients with persistent MUS?	- Which terms do you use to characterize these patients? - Which terms do you tell to your patients?
What's the aetiology of persistent MUS?	- What is the nature of these symptoms? - When do patients experience these symptoms? - Why do these symptoms persist for such a long time?
Do you explain the diagnosis persistent MUS to your patients?	- Do you think explanation is important in consultations with these patients? - How do you explain the diagnosis persistent MUS to the patient? - Which specific words do you use during explanation of the symptoms?
How do you manage patients with persistent MUS?	- How do you deliver health care to them? - What do you do with requests for additional research? - Which problems do you face in the management of these patients? - How do you manage diagnostic uncertainty? - Do you feel capable to manage these patients?
How do you describe the doctor-patient relationship with those patients?	- Is the doctor-patient relationship important, and why? - Do you experience problems in the doctor-patient relationship?
How do you experience the MUS consultations?	- Which problems do you face during the MUS consultation?

Analysis

Analysis followed the principles of constant comparative analysis in which transcripts are subsequently thematically coded.[33] The main aim of this analysis is to organize responses by theme and explore similarities and differences in and between groups.

Two researchers (ToH, LH) read all interviews several times to familiarize themselves with the data. They independently made a first categorization by coding meaningful sentences. Initial codes were discussed, seeking agreement on their content. In the event of disagreement, the opinion of a third researcher (PL) was sought. We grouped the codes into themes to identify key features of GPs' views on MUS. Recurrent and important themes were frequently discussed and refined as part of an ongoing iterative process.[34] During the entire analysis we constantly matched the developing themes with the transcripts and with available scientific literature on this subject. Therefore, these repeated themes are grounded in the data and not imposed onto the data by the researcher. We also checked our developing themes for inconsistencies with the transcripts.

Data collection continued until saturation was reached with no new major themes arising from analysis of the fifth focus group.

The validity of our findings were explored by checking our results in an independent group of GPs who had no specific interest in MUS. They judged the results to be consistent with their perceptions and experiences.[35]

Results

The GPs in this study considered the explanation of the nature of the symptoms as well as maintaining the doctor-patient relationship as a difficult but important task in helping patients with persistent MUS.

GPs with an academic working career discussed more about the classification and current theories about patients with persistent MUS. GPs without an academic working career had a more clear focus on the difficulties they experience in daily practice working with these patients. We could not find further major differences between the perspectives of academic and non-academic GPs. Furthermore, we could not find differences in perspective between rural and urban working GPs.

GPs' perceptions of giving explanations

Importance of explanations and difficulties in explaining were recurrent themes in the focus group discussions. The first focus group discussion (GPs with an academic working career in Nijmegen) revealed that difficulties in a good explanation was an important topic in consultations with MUS patients. During the focus groups with GPs without an academic working career (focus group 2, 3 and 5) we discussed in depth the ways of explaining MUS to the patient. Both GPs with an academic working career (focus group 4) as GPs without an academic working career stressed the importance of a clear explanation. In focus group 5, no new themes on the importance of explanation came up.

GPs stressed the importance of a clear explanation of the symptoms. An adequate explanation was regarded as important in both reassuring patients that there is no serious disease, and in helping patients to accept that there is not always a medical explanation for physical symptoms.

> *"GP13 (male, 5 years GP working experience): But you need to explain damned well. GP12 (male, 29 years GP working experience): The doctor has the monopoly of truth, so you need to be very clear about the cause of the symptoms. Don't be vague because otherwise a patient will return home muddled which make things worse"* [FG 3]

GPs stated that adequate formulated explanations may help patients understand the connection between their psychosocial life and the symptoms. According to the GPs, patients' family members and patients' colleagues also wish an explanation of the symptoms too, especially when patients have benefits of being ill.

Although GPs firmly agreed on the importance of a clear explanation of the symptoms they experience difficulties in doing this. GPs have difficulties indicating from which conditions the symptoms originate. We see this from the vague and avoiding answers of the GPs to the questions of the moderator and the long silence after a question of the moderator on this topic.

> *"GP4 (male, 26 years GP working experience): I explain to patients which symptoms are bothering them and I leave the diagnosis in the middle. I accept the symptoms as such and ask about the consequences of the symptoms. GP3 (female, 8 years GP working experience): Yes, I avoid diagnostic terms too and I confine myself to the particular symptom and I explain that it could be anything. Moderator: But which terms do you use? (moments of silence) GP2 (male, 17 years GP working experience): I discuss with them a different way of coping with their symptoms which may relieve them."* [FG 1]

> *"GP5 (male, 7 years GP working experience): In these patients there is often no connection between symptoms and problems in daily life. At least I can't see one. GP7 (male, 25 years GP working experience): I often tell them that we are not yet knowledgeable. Particularly about patients who really have difficulties with their symptoms, yes, you have to respond differently. GP5: But, when you ask me 'where exactly do the symptoms come from, that chronic fatigue', then I don't have an answer"* [FG 2]

Our analysis revealed three approaches, according to GPs, to explain the unexplained symptoms. First, GPs indicate that they *tell patients there is no disease*. However, GPs highlighted the dilemma of how to communicate the finding of a patient suffering from symptoms without evidence of any physical anomaly. They describe that they try to reassure patients with statements that 'nothing is wrong'.

> *"GP9 (female, 19 years GP working experience): Yes, I always say: I don't know it either. It is not your heart, not your lungs, we did not find any abnormality. GP6 (female, 1 year GP working experience): Yes, I recognize what [GP9] is saying: at least we can conclude that it's nothing serious. We have examined a lot, but the question as to what you are actually suffering from is difficult to answer. GP5 (male, 7 years GP working experience): Yes, these symptoms are not caused by a disease"* [FG 2]

Secondly, GPs indicate that they *use metaphors* to give patients some insight in the hypothesized interactions between symptoms and psychosocial life. GPs reported that they use metaphors – often a tangible physical mechanism indicating some kind of imbalance between load and capacity – that reflect their tacit beliefs and ideas about the nature of MUS. According to the GPs, sometimes the metaphor facilitates a discussion of psychological or social problems.

> *"GP5 (male, 7 years GP working experience): Every human being has a weak spot and if there's something wrong you feel it there. GP9 (female, 19 years GP working experience): I always tell them to compare it with a heavily overloaded elevator. GP7 (male, 25 years GP working experience): I recognize your story, I always tell patients that everyone has a backpack and this backpack can be too heavy."* [FG 2]

Thirdly, GPs indicate that they *normalize the symptoms* of the patients, telling patients that having symptoms is a part of normal life. GPs reported that they explain to the patient that the symptoms are within a common, acceptable range, that they are not dangerous and that diagnostic procedures or treatment are not necessary.

> *"GP10 (female, 1 year GP working experience): I normalize. I mean, I explain to patients that it is normal, that it's not strange. I try to normalize as much as possible. GP12 (male, 29 years GP working experience): Yes. GP10 : saying that it is part of normal life. GP12: Yes"* [FG 3]

GPs' perceptions of the evolving doctor-patient relationship

In all focus groups GPs discussed the importance of the doctor-patient relationship. The difficulties arising in the relationship was a recurrent theme. Focus groups with GPs without an academic working career (focus group 2, 3 and 5) discussed the way of dealing with the doctor-patient relationship in patients with persistent MUS. Furthermore, focus group 4 and 5 GPs discussed the difficulties they face in the relationship with these patients.

GPs intend to clarify the link (as supposed by them) between somatic experiences and psychosocial circumstances of the patient. In other words, they indicate to try to *change the agenda*. When talking about psychosocial circumstances is not successful, GPs reported that they suggest and discuss a range of activities: doing some sports, giving more frequent consultations for the symptoms, using a symptom diary, taking medication or referring to a social worker.

When changing the agenda doesn't work out well, GPs reported that they focus on *dealing with the doctor-patient relationship*. Within this strategy we could distinguish three different doctor-patient relationship models.

A first doctor-patient relationship model can be characterized as *mutual alliance*. This alliance is realized by some sort of ritual care. GPs stated that they use rituals with seemingly mutual approval and that these rituals emerge gradually after many consultations.

> *"GP9 (female, 19 years GP working experience): At a certain moment in your approach of the patient, when a patient has had all diagnostic procedures and many referrals, then comes the moment when one realizes: this is the only way this patient can live. Consequently I let him consult me sporadically, even without complaints [...] And when he feels such a ritual is sufficient, examining his heart, lungs and blood pressure, reassurance is reached to keep him happy for some time. [...] GP7 (male, 25 years GP working experience): Finally, you have created some kind of relationship, some kind of game in which patients are quite satisfied with little. Someone listening [...] a pat on the back."* [FG2]

Examples of those rituals are regular physical examination, referral to a physiotherapist, prescribing medication or performing additional investigations, all with preserving a good relationship with the patient and keeping in mind the unintended consequences of unnecessary interventions.

> *"GP16 (male, 23 years GP working experience): Sometimes I just wait and see, and take care not to cause any damage in these patients"* [FG4]

GPs said that these rituals are connected with requests or wishes of the patients and that they primarily aim at reaching agreement. GPs reported that they provide this kind of care with warmth and empathy and that they assume that patients are satisfied with it.
The second doctor-patient relationship model is *ambivalent alliance*. As stated by the GPs, this model is characterized by the same rituals as in the first model, but in fact the GPs do not agree with the rituals and are unhappy with the situation. There is often a negative colouring in GPs' utterances about this method.

> *"GP14 (male, 15 years GP working experience): Nowadays I ask patients to undress and I practice all sorts of complicated physical examinations I can think of, and when I have done all the physical examinations; then I say: everything is all right, you are healthy and then they go home satisfied […] A patient visits me six times for a referral note, yes, when he or she comes for the third time, then I agree with a referral, inevitable, otherwise you have an argument and in my working experience that does not work at all […]"*[FG4]

GPs reported that the ambivalent alliance indicates a disagreement with supposed requests for medical interventions as medical necessity of these interventions is doubtful. They reported that in these situations the patient is in control of the situation.

The third relationship model appearing from the discussions is *non-alliance*. GPs reported that this model rarely occurs in daily practice. They stated that this model is characterized by cutting off all reasons for encounter in which symptoms are not of somatic origin by taking a cool, objectifying medical gaze.

> *"GP12 (male, 29 years GP working experience): sometimes it is easier to be very short, in a way of 'you have to find out for yourself'. Go to a social worker and do not bother me with that problem again. Just be practical. Then it does not bother me at all. The patient is the one with the problem, it is not my problem, it is your problem, and you have to solve that with the social worker."* [FG3]

In this non-alliance model, GPs reported that in case of an absence of a somatic explanation for the symptoms, they communicate this negative finding directly to the patient and at the same time give the message that the patient should not consult with these kinds of problems.

Analysis in and between the focus group discussions revealed that each GP has a preferred way of handling the relationship problem. GPs who stressed the importance of the relationship preferred the mutual alliance model, whereas GPs who stressed the importance of changing the agenda seemed to rely on the ambivalent alliance model. Non-alliance was not frequently mentioned. One GP stated that he coped with patients with persistent MUS in such a way during his GP residency. One GP told that he used this non-alliance model to cope with these patients during out-of-our-services.

Discussion

GPs are aware of the importance of explaining the diagnosis of MUS adequately to patients with persistent unexplained symptoms. However, they face difficulties in explaining the nature of the symptoms during the encounter with these patients. GPs state that they use three different approaches to explain the symptoms to the patients; normalization of symptoms, telling patients that there is no disease, and using metaphors. According to the literature, normalization of symptoms and telling patients that they don't have a disease is not effective and may even result in more health-care seeking.[19,36] This might contribute to the fact that a small but relevant proportion of MUS patients become persistently impaired and keep attending the GP.[37] Metaphors, on the other hand, can be useful in reaching shared understanding between patient and doctor because they are tangible and non-blaming, although there is limited evidence for their effectiveness.[36,38] Seemingly, there is a paradox in arguing that physicians should provide explanations for a problem that they themselves describe as unexplained. However, most complaints presented in primary care remain at the level of a symptom diagnosis and never result in the diagnosis of a disease.[39] The connotation 'unexplained' in medically unexplained symptoms indicates that the symptoms are not explainable from the reductionist disease framework.[40] However, these symptoms are frequently explainable in other terms given by models as the somatosensory amplification model or the cognitive-perceptual model.[41] Apparently, GPs lack the competence to use these available models adequately in patients presenting with persistent MUS. However, searching for a symptom explanation together with the patient is an important task of GPs in daily practice as it gives them the opportunity to establish common ground on which they can jointly understand and manage the patients' needs.[42]

GPs realize the usefulness and importance of a good relation in encounters with these patients, although they face difficulties in putting this into practice when explaining and removal of symptoms is not feasible. When patients keep presenting MUS, GPs report that the doctor-patient relationship evolves into three different models characterized by the presence or absence of mutual understanding and a careful balance between maintaining the doctor-patient relationship and preventing unintended consequences of the interventions.

Although GPs recognize the limitations and difficulties of establishing an ongoing and preferably effective relationship with these persistent MUS patients in daily practice, they seems to take responsibility to build and maintain such a relationship. Taking this responsibility fits into the philosophy of primary care in which a long-term and continuous relationship in general practice is emphasized.[20,43] These relationships evolve over time and are built on regular consultations as well as other shared experiences.[20,26] It is known from the literature that building on and establishing an effective and satisfactory doctor-patient relationship has an appreciable impact on health outcomes for patients.[44]

GPs stated that different relationship models with patients with persistent MUS develop over time: mutual alliance, ambivalent alliance and non-alliance. These relationship models are, as far as we know, not described elsewhere. The *mutual alliance* model, and to a lesser degree the *ambivalent alliance* model, can be conceptualized as comprising a positive relationship and collaboration with mutual approval between patient and doctor.[45] The goal of this collaboration is to maintain the doctor-patient relationship by providing emotional support through some kind of ritual care. In this strategy GPs keep in balance the doctor-patient relationship and the unintended consequences of interventions. Reaching mutual alliance corresponds with findings that patients with MUS seek a high level of emotional support rather than somatic interventions.[17,46]

Chew-Graham pointed out that GPs who experience difficulties in the relationships with some groups of patients felt that concentrating on maintaining the doctor-patient relationship make them to collude with patients and their symptoms.[47] It is possible that GPs in the '*ambivalent alliance*' model are hindered by this collusion and feel unhappy with the ritual care.

One could argue that the presented models for the doctor-patient relationship are doctor-centred. This may be the result of the aim of our study to examine GPs' perceptions. We asked the GPs in the focus groups how they manage patients with persistent MUS. In other words, we asked for their own GP perspective. In response they described how they struggle to preserve their relation with the patient. In this respect the GPs are working patient-centred. Moreover, the mutual alliance strategy as well as the ambivalent alliance strategy incorporate certain elements of patient-centredness such as finding common ground regarding management and enhancing the doctor-patient relationship.[48] However, a more patient-centred approach in which an exploration of the patients' needs and expectations in order to support patients' self management and coping with the symptoms, did not come up in the focus groups. GPs did not introduce several other aspects of patient-centredness such as disclosing patients' concerns and suffering, and focussing on patients' self management and coping.[49] We assume that this can be explained by the fact that during our focus group interviews we focussed on situations in daily practice in which GPs felt that they get stuck with these patient. Possibly, strategies as disclosing concerns, relief of suffering, and focussing on self management and coping had failed in an earlier stage of the doctor-patient contact.

Strengths and limitations of this study

The qualitative method is appropriate to explore and clarify what GPs think about these patients and what they experience in the consultations with these patients.[32] However this method does not provide insight in the GPs actual behaviour. By using a cyclical and interactive way of collecting and analyzing data, 'progressive focusing' and exploration of GPs' perceptions in depth were realized.[50] In focus group discussions the participants influence each other by listening and discussing. Such group dynamics may silence individual contrasting opinions and result in the articulation of group norms or early consensus before all views were fully expressed.[32] However, the goal is not to reach consensus. Instead, our goal was to study how GPs as professionals think about patients with persistent MUS. It would nevertheless be interesting to analyze how individual participants influenced each other during the discussion, but this was not the aim of our study. To reach an optimal variety of opinions, we used a purposive sampling strategy. Although small, the number of participants is considered adequate for this purpose.[32] By using a purposive sampling strategy we have captured the variety of opinions present in the population of GPs.

As MUS are not equally distributed among men and women and men and women have different expectations and experiences of clinical encounters,[6] a gender perspective may enhance understanding. However, we did not study the differences in thinking about patients with persistent MUS between female versus male GPs, as in this study we focussed on eliciting GPs' perceptions of giving explanations and their perceptions about the doctor-patient relationship.

Although we know from recent research that there are cultural differences in the distribution of MUS and the meaning and significance of a symptom depends on the perceived relationship with diseases in a culture,[51-53] GPs did not spontaneously introduce their opinions on cultural aspects of persistent MUS during the focus group discussions.

This qualitative study examines GPs' perceptions and not actual behaviour. We deliberately chose to study the perceptions because actual behaviour may result from perceptions to a certain degree. Moreover, this study is part of a larger project in which we examine actual behaviour and communication strategies of GPs in a video registration study and the patient perspective in a qualitative interview study.

We describe our results by using the phrase 'relationship models'. Although we are aware of the overlap with communication strategies, we think that 'relationship models' is a more appropriate term in the context of our findings as it reflects the opinions of the GPs that maintaining the relationship with those patients is of major importance. GPs indicate that they maintain the relationship not only by the way they communicate but also by the way they take care for these patients.

Tape-recording the discussion, multiple coding during analysis and our triangulation strategy of asking independent GPs to judge our results to be consistent with their own perceptions and experience, added to the rigor of the study.

Further studies using a mixed method methodology may reveal effective methods of explaining symptoms to patients with MUS. Moreover, it would be useful to study the effects of the three relationship models reported by the GPs on outcomes and satisfaction in patients. In research, as well as education we should face the challenge of explaining the unexplained symptoms and building a truly effective doctor-patient relationship with these fascinating patients. With the results of further research we would like to address the challenge of explaining unexplained symptoms adequately and building effective doctor-patient relationships with these patients, preferably by educating doctors with tools how to do so.

Conclusion

GPs are aware of the importance of explaining MUS adequately to their patients, however they have difficulties in doing so. GPs report that, when patients keep presenting with MUS, they focus on maintaining the doctor-patient relationship by using ritual care. These relationships evolves into three different models characterized by the presence or absence of mutual understanding and a careful balance between maintaining the doctor-patient relationship and preventing unintended consequences of the interventions.

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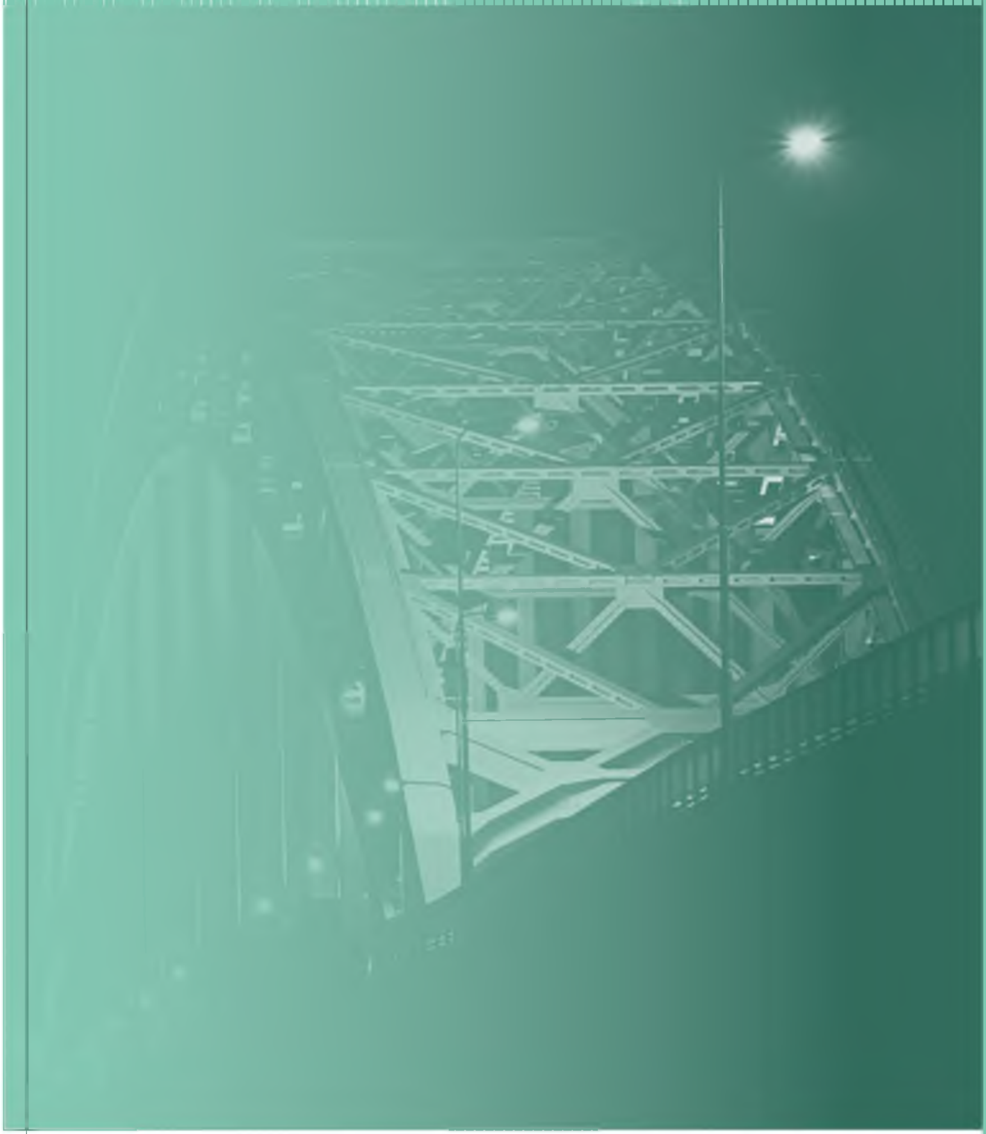
CHAPTER
6

'WELL DOCTOR, IT IS ALL ABOUT HOW LIFE IS LIVED': CUES AS A TOOL IN THE MEDICAL CONSULTATION



Tim olde Hartman
Hiske van Ravesteijn

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Abstract

Introduction. During consultations, the perspective of the patient and the family physician come together. In order to reach a shared view about the symptoms it is important to know the agenda of the patient. Cues (i.e. non-explicit remarks that can enclose a special meaning) can serve as a tool to clarify the agenda.

Case Report. In this article, we describe a patient with unexplained palpitations during vacuuming. During one of the following consultations she provided an important psychosocial cue which changed my perspective on her palpitations, resulting in a deeper understanding of her symptoms.

Discussion. Recognition and exploration of cues is important for reaching mutual understanding of doctors and patients about the symptoms. Moreover, it enhances the therapeutic relationship and improves illness outcomes and patient satisfaction.

Conclusion. Noticing cues in the medical consultation helps to understand the patient's real worries. It gives us, as doctors, a better understanding of the patient's perspective.

Introduction

An important task for family physician (FPs) is listening to patients.[1,2] The patient's symptoms are discussed by the patient and the doctor together during consultation. Patients have their own ideas, worries and expectations about their symptoms and so has the FP, partly because of what he was taught in medical education, but mainly because of his experience.[3,4] During consultation the perspectives of the patient and the FP come together.

One of the most important tasks of the FP is to come to a mutual understanding with the patient: a shared view about the symptoms.[5] To reach this agreement, the doctor needs to know the agenda of the patient. This can be achieved by exploring the patient's expectations, cognitions and emotions.[5-7] Through this working style, the doctor enhances the patient's satisfaction, adherence and health.[8,9]

In daily practice it appears to be difficult to get to know the agenda of the patient.[10] In approximately half of all consultations the doctor does not reveal the reason for encounter and the worries of the patient.[9] In about 20% of all consultations the patient has an unvoiced biomedical or psychosocial agenda.[11]

During a consultation patients often give cues. Cues are non-explicit remarks that can enclose a special meaning. They can point towards ideas, worries or expectations the patient has not shared before.[3] Noticing and exploring the patient's 'cues' is helpful, it can serve as a 'tool' to clarify the agenda of the patient.[3,5] However, doctors appears to have great difficulty in detecting and responding to more indirect forms of communication such as cues.[12] The following case changed my (ToH) communication skills and illustrates the importance of noticing and exploring a cue.

Case

The first consultation

A 79 year old woman visits me (ToH) with symptoms of fatigue and palpitations. The symptoms started while vacuuming; she frequently had to stop doing it just to recover for a while. She is very active, but the years seem to start counting. She asks me why she suddenly developed palpitations. She is worried because she never had them before. Further history taking does not give any indication for an underlying cause of the palpitations. Because she has palpitations during exertion, I want to exclude a cardiac problem. With the patient's approval I decide to make an electrocardiogram and test her blood to exclude cardiac and other causes for the palpitations.

The following consultations

The electrocardiogram is normal, the blood test reveals a hypothyroidism. This could explain her fatigue, but not her palpitations. I reassure her, and after discussing this we decide together that I will prescribe her thyrax. We settle a couple of appointments for follow up consultations. During these consultations I adjust the thyrax dosage and also have a chat with her. Gradually I get to know her a little better. She tells me more about her history and background. I enjoy these consultations. With the thyrax the thyroid function normalises and the fatigue disappears almost completely.

A special consultation

Six weeks after the first consultation, she comes for a follow-up visit again. Her blood has been tested and her thyroid function is stable. She is happy and satisfied with this news. Then she brings up her palpitations again. She wants to know whether it is possible that they are caused by her thyroid problem. I ask her if they have increased since the start of the thyrax, which she denies. I explain to her that the palpitations are probably unrelated to her hypothyroidism and probably benign. She seems to accept this and says: 'Well doctor, it is all about how life is lived'.

'What a remarkable sentence' I think. I show my wonder and ask: 'Well then, how is life lived?' And in front of me, there and then a story unfolds that had been unknown to me. At the age of 16 she went into a convent. Initially she had a good time there, but this changed when a new mother superior was appointed with whom she could not get on. Mother superior allotted her all the nasty tasks and she degraded her to being a cleaner. For a long time she did not do anything but scrubbing floors. She was teased and cold-shouldered by her fellow sisters. Years of bullying followed. A couple of times she tried to bring up this subject, but they did not seem to hear her. During this period she often had physical symptoms, as a result of which she could not perform her cleaning tasks. After living in the convent for 15 years she had to leave – cast out from the convent.

With astonishment I listen to her story and she says: 'Gosh, doctor, I have never told this to anybody' and 'maybe that is why I get these palpitations during vacuuming. I will think about it at home'. We talk some more and she leaves my consultation room noticeably relieved, with the words 'thanks for listening doctor'.

Half a year later

During the follow-up visit for her thyroid problem the woman is cheerful. I realize that it has been half a year since the day that she told me that special story. I ask her about her palpitations. She tells me that she has had them a couple of times since the last consultation, but she is hardly bothered by them anymore. Vacuuming has become easier now and she feels better too. She says everything is fine now.

Reflection on the case

During the first consultation I (ToH) collected data by asking open ended questions, summarising and asking directive questions. Furthermore, I aimed at searching for a biomedical diagnosis of her palpitations. I looked at her palpitations from a biomedical point of view and wanted to exclude a cardiac cause. When I found a hypothyroid function with the blood test I mainly focused on the medical policy and management plan, without reconsidering the palpitations and without legitimising patient's feelings.

Looking back on this case I realised that initially I hadn't dwelled on the fact that the palpitations came up especially during vacuum cleaning. Why of all times did she get them during vacuum cleaning? At this stage I should have shown my curiosity. Did she get palpitations because of the exercise or could it be linked to vacuum cleaning in particular? A better exploration of her cognitions and worries about the palpitations would have been helpful. However, the following consultations gave me the opportunity to ask questions regarding patient's social situation and history and build the doctor-patient relationship. Thanks to the remark: 'Well doctor, it's all about how life is lived', I was capable of leaving my biomedical point of view. Together with the patient I found a deeper meaning in her symptoms.

Did my consultation skills change since this case? Yes, they certainly have. Through this case I became keener on picking up cues patients give during the consultation. Cues are an easy tool in doctor-patient communication and very useful in daily practice. Of course, I do not know for sure if there is a causal relationship between telling the story and her improvement. But the patient seemed to benefit from discussing the possible relationship, especially considering her relief at the end of the consultation.

Cues in the consultation

In primary care there is a tradition of paying attention to cues and their meaning. Recognition of cues and exploration of their meaning is important for the mutual understanding of the doctor and the patient. With the patient-centeredness movement at the end of the 1980s, paying attention to the significance of cues became in vogue again.[5]

Responding to patients' cues is one of the most important tools for a successful consultation.[13] Cues are described in different ways by different authors. Gask and Usherwood refer to verbal and nonverbal expressions of the patient that hint at psychosocial or social problems.[13] Livinson *et al* describe 'cues' as direct or indirect expressions with information about patients' life and feelings.[14] Balint uses the word 'offers' for expressions by the patient about the significance of their symptoms and for expressions about the reason for encounter with the FP.[15] Branch and Malik describe cues as 'windows of opportunity' for the doctor to show empathy.[16] It is important to detect and respond to cues at the time they are offered by the patients.[13] Not addressing cues during clinical encounters may inhibit patients from further disclosures. Berkatis *et al* reported a significant relationship between the doctor's response to emotional cues and the patient's disclosure.[17] Moreover, cues enable better understanding of patients' life, cognitions and emotions.[18,19] Recognition and exploration of them has another advantage. It shows that the FP is listening carefully, wants to understand the meaning of the symptoms and is interested in the patient.[3] By picking up cues and exploring them, the FP enhances the therapeutic relationship and, as a consequence, improves illness outcomes and patient satisfaction.[17,20]

We know that doctors have difficulties recognising cues. Livinson *et al* examined how patients presented cues and how FPs reacted to them.[21] In more than half of the consultations, cues were present (average of 2.6 cues per consultation). Patients initiated 71% of the cues themselves and 29% were initiated by the FP asking open questions. In a majority of the consultations (79%), FPs missed the opportunity to react to the cues given in the consultation. Moreover, these consultations were of significantly longer duration. Butow *et al* found the same results in their study of verbal cues in cancer patients: oncologists did not consistently detect and address cues for emotional support. Consultations in which oncologists responded to higher proportions of patients' cues did not last longer than other consultations.[22] Cegala analysed videotaped primary care consultations of 16 doctors with 32 patients, and found that doctors rarely provide information in the absence of a direct patient question.[12] Moreover, patients' indirect cues of informational and emotional needs are far more common than direct patient questions. Thus, a focus on cues of needs in the clinical encounter is important.

Salmon *et al* studied consultations about medically unexplained symptoms and showed that most patients gave explicit cues about emotional or social problems.[23] Most FPs reacted to these cues by either blocking or facilitating a discussion about psychosocial issues. The FPs who blocked a discussion did not pay attention to the cues, they refocused on the symptoms and normalised the worries of the patient or stressed patients' own responsibility. The FPs who facilitated a discussion about psychosocial issues did so by asking questions about the experiences of the patient and by exploring the problems of the patient. However, when the FPs gave an explanation of the symptoms, the FPs rarely took the psychosocial problems into account. Consequently, patients' concerns were not addressed and reassurance failed.

Conclusion

Most people interpret their symptoms in the context of their personal, family and life experiences. Active listening and noticing cues, as described in this case report, can help FPs to get to know their patients.[1]

Discovering the patients' real worries and getting to know their stories gives us, as doctors, a better understanding of their own world.[24] In addition, mutual understanding leads to a higher satisfaction with care for both patient and doctor and it strengthens the doctor-patient relationship.[3,25,26] Remarkable words, strange clauses, sentences that you do not directly understand – ask for their meaning. It brings the patient's and the doctor's world closer together.

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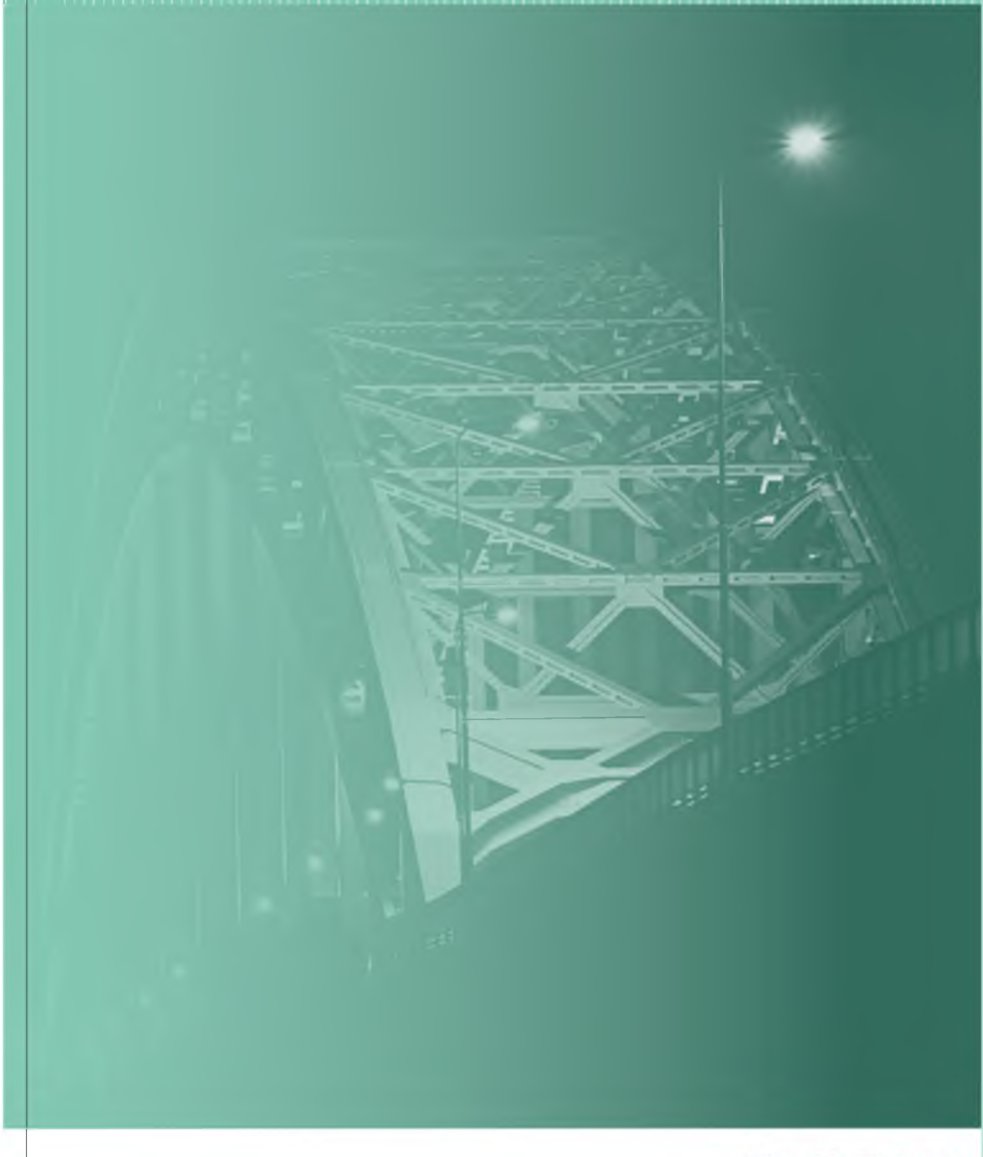
CHAPTER

7

HOW PATIENTS AND FAMILY PHYSICIANS COMMUNICATE ABOUT PERSISTENT MEDICALLY UNEXPLAINED SYMPTOMS. A QUALITATIVE STUDY OF VIDEO-RECORDED CONSULTATIONS



Tim olde Hartman
Eric van Rijswijk
Sandra van Dulmen
Evelyn van Weel-Baumgarten
Peter Lucassen
Chris van Weel

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Summary

Objectives. To study doctor-patient interaction styles in consultations with patients presenting persistent medically unexplained symptoms (MUS) and to study on which stages of the consultation patients and doctors focus within the available time.

Methods. Exploratory, qualitative analysis of transcripts of 20 videotaped consultations between family physicians (FP) and persistent MUS patients.

Results. Patients presented many symptoms in a rather unstructured way. However, FPs hardly used structuring techniques such as agenda setting and summarizing. Patients with persistent MUS got much opportunity to tell their story, but the reasons for encounter, their beliefs and concerns were not discussed in a structured manner. Although consultations were focused on these issues, mostly patients themselves initiated discussion of their ideas, concerns and expectations. FPs' extensive explanations of the origin of the symptoms often did not take patients' beliefs and concerns into account.

Conclusions. Due to patients' multiple symptom presentation and the absence of FPs' structuring techniques, consultations of persistent MUS patients proceed rather unfocused. However, patients got ample opportunity to tell their story.

Practice implications. Persistent MUS patients might benefit from structured consultations focused on the exploration of the reason for encounter.

Introduction

Patients complaining of physical symptoms in the absence of physical disease are common in primary care. These symptoms are often described as medically unexplained symptoms (MUS).[1,2] In patients with MUS, symptoms or impairment improve when consultations meet the patients' expectations or when patients feel understood.[3] Recently, two studies showed that physicians' communication during the *initial* presentation of MUS is hampered.[4,5] Epstein et al. concluded that physicians face lack of time and do not explore and validate the MUS patient's reason for visit, their ideas, expectations and concerns.[4] Kappen and Van Dulmen concluded that family physicians (FPs) explore patients' concerns mainly medically.[5] Despite these physicians' communication barriers during the initial presentation of MUS,[6] only a minority (2.5 percent) of the patients will evolve into a chronic disabling condition of persistent MUS.[7] From this moment communication is often the only tool FPs have in handling these patients.[4,8] However, doctor-patient interaction styles in consultations with patients with *persistent* MUS have not been well studied, and may bring important insights to improve the quality of care of these patients.

Consultations between doctors and persistent MUS patients are not straightforward but can be considered as complex consultations.[9] As symptoms are medically unexplained, the link between cause and symptom is unclear, and FPs are uncertain about the way forward.[10] Persistent MUS patients are aware of the complex nature of their problems.[11-13] They have to present complex and multifaceted reasons for consulting, discuss concerns about the symptoms and problems, and choose whether or not to present emotional aspects of their problems, in a 10-15 minutes consultation.[13-15] It is understandable that both patients and FPs report insufficient time to deal effectively with persistent MUS during consultations.[16,17] However, it is still not clear how patients and FPs reach their goals during the consultations and on which stages in the consultation they focus.

Therefore, the aim of this study is to analyze (1) how patients present and how FPs explore the patients' symptoms and problems during consultations and (2) on which stages of the consultation they focus within the available time.

Methods

Data source: Dutch National Survey of General Practice

Data for the present study were drawn from the Second Dutch National Survey in General Practice (DNSGP-2).[18] This survey is a large-scale research project carried out in the Netherlands between May 2000 and April 2002 and studied a representative sample of 104 family practices with 195 FPs and 399,068 listed patients. The survey comprised an epidemiologic study about the work of FPs and a video observation study of consultations in which each participating FP consented to video tape approximately 20 clinical encounters on two arbitrary days. A total of 142 FPs (72.8%) agreed to participate in the video observation study.[19] Prior to the consultation, a research assistant informed the patients and asked informed consent about the video observation. A total of 2784 patients participated in the video-observation study, 377 patients (11.9%) refused. Before and after the videotaped encounter patients completed a questionnaire about sociodemographic characteristics, presented symptoms and general health status. Immediately after the consultation, FPs completed a questionnaire with items on characteristics of the FP, questions about how familiar the FP was with the patient and how the FP valued the psychosocial and work related aspects of the presented symptoms. Furthermore, FPs rated on a 5-point Likert-type scale (extremes labeled as 'not at all' and 'very much') whether they believed psychosocial factors played a role in the problem presented during the consultation. FPs coded patients' symptoms and diagnoses according to the International Classification in Primary Care (ICPC) system.[20,21]

Selection of patients with medically unexplained symptoms

We selected from the video-observation study all videotaped consultations in which medically unexplained symptoms were the main subject of the encounter. Inclusion criteria were: (1) patients consulting their own FP, (2) consultations for physical symptoms for which the patient had consulted the FP before and in which, according to the FP, the symptoms were related to psychosocial factors ('much'/'very much' on 5-point Likert-type scale), (3) age of the patient ≥ 18 years and (4) no psychiatric diagnosis and/or social diagnosis according to the ICPC during this consultation. Exclusion criteria were: bad sound-quality and consultations by one of the authors. When more than one video consultation of a FP appeared in our selection, only the first consultation was included. Two independent researchers (ToH and SvD or EvR) looked at the video recordings and established whether medically unexplained symptoms were the main topic. Encounters were included in the final sample when both researchers agreed. In case of disagreement, we excluded the consultation. We used Cohen's kappa statistic (κ) to assess agreement between the two researchers.[22]

Data analysis

Data analysis started by using qualitative methods to develop a coding scheme of FPs' exploration of the patients' symptoms and problems.[23] The theoretical framework guiding the analysis was the broad concept of the biopsychosocial model.[24] This model proposes illness to be viewed as a result of interacting mechanisms at the biomedical, interpersonal and environmental levels. The model is widely used in primary care consultations. Using this model in consultations implies that patients' symptoms, illness beliefs, anxiety, concerns, illness behavior and social environment are addressed.[25]

The included videotaped consultations were completely and anonymously verbatim transcribed and entered into Atlas.ti. The qualitative analyses were executed with this software program, a package for detailed coding in qualitative data analysis.

We analyzed the transcripts using the principles of constant comparative analysis.[26] During this analysis transcripts are subsequently thematically coded. The main aim of this analysis is to organize utterances by theme and to explore similarities and differences between consultations.

Two researchers (ToH, EvR) read all transcripts several times to familiarize themselves with the data. They independently made a first categorization by applying codes to meaningful words and sentences in the transcripts. These codes were discussed and refined during consensus meetings. When additional codes emerged these were discussed and applied to the transcripts. Concepts and categories emerged through this iterative process of coding, analysis and discussion. Codes appearing from the utterances of the participants in the consultations are presented in Table 1. During the iterative process of qualitative analysis we noticed the complex structure of the consultations. To gain a better understanding of this complex structure we decided to code the stages of the consultation, apart from the utterances of the participants during the consultation. These stages were derived from the Dutch FPs' communication skills training program[15,27,28] (Table 2). After 8 videotaped consultations, the first results of the analysis were discussed with a senior researcher (PL). Data collection continued until no significant new themes emerged (saturation).[29] This was achieved after 15 consultations. To quantify on which stages of the consultation patients and FPs focus within the available time of the consultation, we calculated, as a proxy, the percentage of text in the transcript (the number of text lines of a particular stage of the consultation divided by the total number of text lines of the consultation) spent on each of the different stages.

Ethical approval

The study was carried out according to Dutch privacy legislation rules. The privacy regulation was approved by the Dutch Data Protection Authority.

Results

Sample characteristics

The total number of video consultations in the Second Dutch National Survey of General Practice (DNSGP2) was 2784. Fifty nine of these video consultations met our inclusion criteria and were screened by one of the authors (ToH). In 14 cases more than one video consultation of a FP appeared in the selection, 4 video consultations had a bad sound quality and in 1 video consultation one of the authors (PL) was the FP. These 19 video consultations were excluded. Therefore, a total of 40 video consultations could be included for screening by two independent researchers (ToH and SvD or EvR). Because MUS was not the main topic of the consultation in 20 of these video consultations, a total of 20 video consultations could be included for further analysis (Figure 1). The interobserver agreement for inclusion was κ = 0.79 (95%-CI: 0.59 – 0.99). We considered this level of agreement to be 'good'.

Eight (40%) of the patients involved in the final video consultation sample were men, aged between 25 and 80 (mean = 47) years. The 20 FPs consisted of 15 (75%) men, aged from 34 to 61 (mean = 45) years with an average of 15.7 (range 3 – 30) years of FP working experience. Mean (and median) duration of consultation was 13.5 (12.1) minutes; ranging from 8.1 to 37.0 minutes.

Within the available time of the consultation, patients and doctors focus most on the story of the patient, discussion and exploration of patients' beliefs and concerns, and on explaining the symptoms (respectively 21.8, 18.7 and 16.8% of the total amount of text is spent on these stages) (Table 3). Agenda setting, summarizing and evaluation of the consultation was limited.

Figure 1. Selection of persistent MUS video consultations

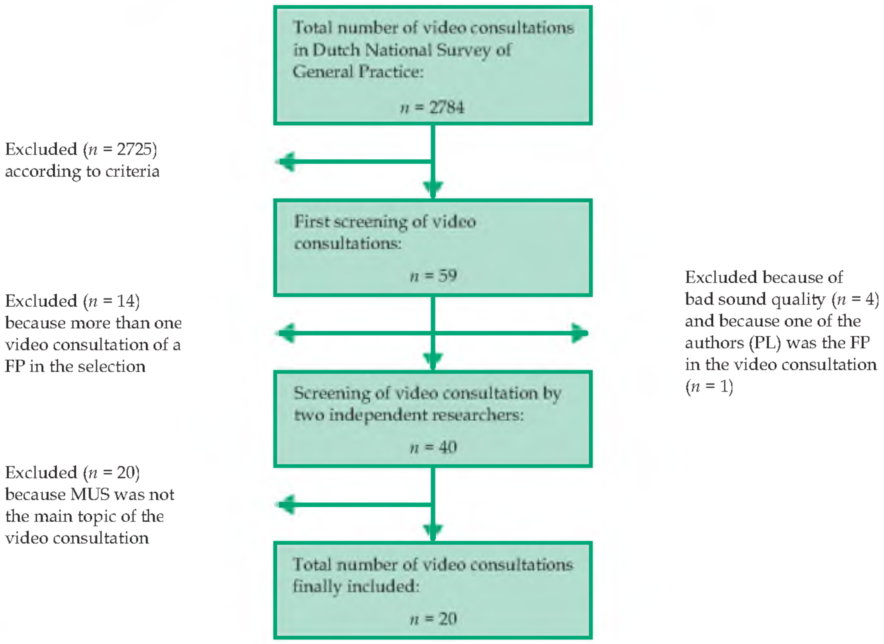

Table 1. Codes appearing from utterances of the participants of the persistent MUS consultations

Codes
Story of the patient (narrative)
Symptoms
Patients’ beliefs and expectations
Patients’ fears and concerns
Consequences on patients’ daily activities and illness behavior
Consequences on patients’ social environment
Explanation

Table 2. Stages of the consultation

Story of the patient (narrative)	The retelling, in patients' own words, of a series of unfolding events regarding the symptoms, representing the patient individual viewpoint
Agenda setting	Agreement (between patient and FP) of the issues which should be discussed during the consultation
Discussion / exploration of the symptoms	Discussion and exploration of the characteristics of the symptoms (nature, location, intensity, frequency and duration)
Discussion / exploring of the reason for encounter (RFE)	Discussion and exploration of the reason for encounter, including the patients' expectations regarding the actions of the doctor
Discussion / exploring of patients’ beliefs and concerns[a]	Discussion and exploration of patients' ideas regarding the symptoms, such as patients' own explanations, symptom attribution, patients' own influences on the symptoms (self-efficacy), concerns and (negative) emotions regarding the symptoms
Discussion / exploration of the consequences on patients' daily activities, social environment and illness behavior[b]	Discussion and exploration of patients' behaviour in response to the symptoms, the way patients cope with their symptoms, the consequences of the symptoms on patients' daily activities, the influences of the symptoms on patients' social life, and reactions and opinions of the people in patients' surroundings.
Summarizing	FPs' summary of the symptoms, reason for encounter, beliefs, concerns and consequences of the symptoms presented during the consultation
Physical examination	The process by which the FP performs a physical examiniation
Explanation	FPs' explanation of the origin of the symptoms presented during the consultation
Evaluation of the consultation (including making a follow-up appointment)	Evaluation of the consultation, including the question whether the consultation has been helpful, explanation and/or advices, whether the reason for encounter has been answered, and whether follow-up appointments have been made.

[a] Matches with the codes “patients' beliefs and expectations” and “patients' fears and concerns” that appeared from the utterances of participants of persistent MUS consultations.

[b] Matches with the codes “consequences on patients' daily activities and illness behavior” and “consequences on patients' social environment” that appeared from the utterances of participants of the persistent MUS consultations.

Table 3. The percentage of text in the transcripts spent on each of the different stages of the consultation

	Persistent MUS patient Mean
Story of the patient (narrative)	21.8
Agenda setting	limited
Symptom exploration	7.9
Reason for encounter (RFE)	
Initiated by the FP	0.6
Initiated by the patient	2.1
Patients' beliefs and concerns	
Initiated by the FP	4.8
Initiated by the patient	13.9
Consequences on patients' daily activities, social environment and illness behavior	
Initiated by the FP	1.1
Initiated by the patient	2.7
Summarizing	limited
Physical examination[a]	-
Explanations	16.8
Evaluation of the consultation	limited
Other[b]	28.3

[a] Quantifying physical examination is not possible by using the proxy percentage of text spent.

[b] Talk/discussion regarding medically explained symptoms (for example: presentation of a pneumonia, discussion on therapies for asthma, discussion and management of hypertension, etc.).

Patients' symptom presentation

Most (95%) persistent MUS patients presented more than one symptom. Three quarters of the patients presented medically unexplained symptoms as well as medically explained symptoms. The average number of symptoms presented was 3.6 (range 1 – 5). The average number of MUS presented was 2.4 (range 1 – 5). The most common symptoms were musculoskeletal (n = 15), gastrointestinal (n = 8) and general and unspecified symptoms (n = 11), such as feeling ill, weakness/tiredness, sweating or swelling.

Persistent MUS patients presented multiple symptoms and seemed to switch from one symptom to another during all stages of the consultation.

They often started to discuss new symptoms and concerns after the FP finished history taking, physical examination and explanation. **(Quotation 1)**

Quotation 1 (P11 116-12;023-111)

[Patient's story, symptom 1]*

P: It's either painful gas or it's a lot of pain in my belly. And my bowel movements happen or don't *happen but they look kind of weird. But it will go away. It's always gone away eventually.*

D: Okay, so you have had these complaints for years.

[Medical exploration]

D: Abnormal bowel movements. No blood or mucus?

P: No. No.

D: Firm and pulpy, no diarrhea?

P: No, I don't have that.

[No physical examination]

[Explanation]

D: Okay, it seems to me that you have irritable bowel syndrome - sensitive intestines - and that can cause cramps. About 20% of the population has this kind of bowel problem to some extent. It's not malignant and there is no infection. It's just that the intestines don't function optimally. And so it's important that you get enough fiber and that you take the powders (movicolon).

[Patient's story, symptom 2]

P: And the itch. It won't go away and it's awful.

D: So, we have to deal with the itch.

P: Yes, it's ridiculous but it's sometimes really bad on my face and on my back, it's the worst – it's always the same place. I've had it for a really long time.

* the words between brackets represent the codes given by the researchers and used in the constant comparative analysis

Patients' attempts to address their needs

In most encounters, the patients' reason for seeking help remained unclear. Although clarifying the *reason for encounter* (RFE) is an important task for the FP in order to reach a more focused communication, in 13 consultations (65%) there was no exploration of the reason for encounter at all. The absence of FPs' exploration of the reason for encounter might be the reason why patients themselves try to initiate such a discussion. It appeared from the data that during consultations in which the reason for encounter was discussed (n = 7), most of the time the patients themselves initiated talking about this subject (n = 5). **(Quotation 2)** Only in two consultations the doctor initiated the discussion on the reason for encounter.

Discussions on the reason for encounter took 2.7% (range 0 – 15.7) of the text in the transcripts. (Table 3)

Quotation 2 (P8 C10208;24-31)

P: And when I go to bed, I feel very restless. I have these heart palpitations.

D: Yes,

P: So I was wondering – I read about something called beta-blockers. I don't know if that applies to my situation but -

> *D: Hmm*
> *P: Someone told me about propanolol. I was wondering if that was something for me.*
> *D: Yeah, okay.*

In all but two video consultations patients started talking about their beliefs and concerns. However, in the two encounters in which no discussion on beliefs and concerns took place, the patient tried to initiate such a discussion but the FP refrained from responding **(Quotation 3)**. In the encounters in which beliefs and concerns were discussed, patients mostly initiated such a discussion: in 16 consultations (89%) patients made attempts to initiate discussion, whereas in 9 consultations (50%) FPs did. This is also reflected in the amount of text spent in the transcripts of the consultations on this topic. The total amount of text spent in the transcripts regarding discussing patients' beliefs and concerns initiated by the patient or by the FP is 13.9% (range 0 – 40.1), respectively 4.8% (range 0 – 17.8). (Table 3)

It is noteworthy that in the majority of the consultations (10 out of 18) in which patients' beliefs and concerns were discussed, discussion of these beliefs and concerns only took place for a limited number of symptoms presented during the consultation.

> **Quotation 3 (P1 C2601;132-140)**
> *P: It's not good at all. I feel myself going so unbelievably downhill.*
> *D: This is something you need to discuss with a neurologist. You can make an appointment with one. I'll write a referral letter and you can pick that up at my assistant's desk. But your legs: that's also an issue for you?*
> *P: Yes, I know I have a lot of weight to carry but, oh my, that's not easy. It's like something is broken in my brain. I think every time, 'I have to lose weight, I have to lose weight,' and, at the very same moment, I stuff myself. It just doesn't work that way up here (P points to head).*
> *D: You need to lose weight but you eat too.*

The consequences of the symptoms on patients' daily activities, social environment and illness behavior was less well discussed. In 9 consultations (45%) there was no discussion of these themes at all. Again, most of the time discussion of these consequences of the symptoms was initiated by the patients namely in 7 consultations; in one consultation this discussion was initiated by the FP as well as the patient and in three consultations the FP initiated the discussion. **(Quotation 4)** Almost 3% (2.7; range 0 – 12.3) of the total amount of text in the transcripts of the consultations spent on talking about the consequences of the symptoms on daily activities, social environment and illness behavior was initiated by the patient against 1.1% (range 0 – 7.4) initiated by the FP. (see Table 3)

Quotation 4 (P2 C070-13;34-45)
D: How are you sleeping?
P: At night, I sleep okay now, but during the day, I try not to spend time in bed. It's nice to know that I can go to bed but then I restrain myself with, 'No, I can't stay in bed all day.' And then I try to do some stuff around the house – the dishes, vacuuming, dusting.
D: So you do that?
P: And then I try to sleep at night. And when I wake up in the morning, I have those puffy eyes. And those chewable pills, I don't take them one after another anymore. Or should I keep taking them? Because sometimes I still have - especially when I have to bend over, I still see stars.
D: But you eat well?
P: Yes, if I know that I need some kind of feeling or is that not necessary?

In nearly all consultations there was much opportunity for explanation of the symptoms. The total amount of text in the transcripts about explanations is 16.8% (range 0 – 52.9). (Table 3) Although almost one fifth of the text in the transcripts was spent on explaining the symptoms, patients responded on FP explanations with new remarks about their symptoms, beliefs and concerns.

Furthermore, most of the time, explanations given by the FPs did not incorporate beliefs and concerns that patients presented during the consultation. Moreover, FPs attempts to reassure patients were often not focused on the patients' concerns. **(Quotation 5)**

Quotation 5 (P4 C077-04:4-9;21-26;80-85;109-11)
[Patient concerns]
P: I'm having problems with my throat, esophagus again. I feel it when I am doing exercise. It doesn't have anything to do with my heart, does it? I have it every time I roll over at night.
D: Yes
P: I don't know but I think that it's lower. It's like something is stuck there.
[Reason for encounter and patient concerns]
P: I've also had chest pains so I wanted to ask if you would be willing to take a look. I'm a little concerned. I'd just like to know for sure that there's nothing going on.
D: If we know that it's your throat and not your heart, then that's good.
P: Yes.
[Explanation]
P: I was also really busy earlier this week. I've been feeling kind of hurried the last little while so I took a seresta pill and the pain in my throat went away.
D: Yes, that could mean that the tension that you feel inside is coming out through your throat, like your throat is literally being choked.
P: Yes, I do have that kind of feeling.

[Patient response]
P: But my heart has nothing to do with it?
D: No, your heart has nothing to do with it.

FPs' structuring behavior

In the video consultations, FPs gave patients with persistent MUS much opportunity to tell their story (21.8% (range 5.1 – 80.7) of the text in the transcripts). However, FPs did not do an in-depth inquiry of the symptoms (7.9% (range 0 – 39.4) of the text in the transcripts). In 50% of the consultations (n = 10) there was no in-depth inquiry of the presented symptoms at all. In half of the consultations in which an in-depth inquiry of the symptoms was performed, the FP did not explore all medically unexplained symptoms presented during that consultation. Furthermore, it appeared from the video consultations that FPs hardly use structuring techniques, such as agenda setting, announcing and performing physical examination and summarizing the information obtained during the consultation. Agenda setting was explicitly performed in one consultation . Physical examination was performed in eight consultations and a summary was given in only one consultation.

Discussion and conclusion

Discussion

Our findings of the difficulties of discussing the reason for encounter and patients' beliefs and concerns regarding the symptoms during the persistent MUS consultations are in line with the findings of Epstein et al.[4] Furthermore, it is known from direct observation of patients' presentations of MUS that almost all patients provide opportunities for FPs to address psychosocial issues, psychosocial concerns.[5,30] Our study adds rigor to these findings as we studied doctor-patient communication in persistent MUS consultations. Our results indicate that doctor-patient communication can be improved by focusing on the exploration of patients' beliefs and concerns and incorporating these into FPs' explanatory and reassuring strategies. These findings are important as discussion of patient's ideas and concerns, shared understanding and clarifying the reason for encounter contribute to a more satisfactory consultation according to patients.[31]

The observations of our study confirm that the chaotic structure of most MUS consultations reflect the chaotic narrative of the people who live with MUS.[32] Nettleton highlighted that the narratives of MUS patients shared many features of chaos narratives.[33] These narratives are characterized by confusion and uncertainty in the absence of a diagnosis and prognosis.[32] Moreover, these narratives are difficult to 'listen' to and difficult to 'hear', because it reminds us of our own vulnerability and limitations.[34] This might explain why FPs have difficulties in structuring the persistent MUS consultations and why patients present multiple symptoms in a rather unstructured way. Although giving patients time for their story is an important element of working patient-centered, persistent MUS patients might benefit from more focused patient-centered interaction style in which FPs structure the consultation and explore needs, concerns and beliefs. The ample opportunity patients get to tell their story and present their symptoms possibly reflect FPs' commitment with these patients which is in line with research on FPs perceptions about patients with persistent MUS.[10] However, this commitment is worth to yield more effect.

This is the first study in patients with heterogeneous undifferentiated persistent MUS using consultations between doctor and MUS patients in which neither the doctor, nor the patient was aware of the subject of study. Doctor-patient communication studied in this way represents daily practice reality. Furthermore, FPs and patients in these consultations already built a doctor-patient relationship as they knew each other for a long time and had discussed the symptoms before. However, in most cases, a single consultation is not the beginning or the end of the story. Each new consultation carries over memories of previous ones, which might have influenced the videotaped consultations.[35] This might explain why we found limited medical exploration of the presented symptoms and no physical examination in most consultations. However, we did not find utterances referring to the content of previous consultations on the symptoms presented.

Video-recording has been recommended as the best method for researching doctor-patient communication during consultations.[36] According to Coleman, there is little evidence that video-recording influences the behavior of either the FPs or patients (i.e. Hawthorne effect), but it may cause bias in the characteristics of doctors and patients who agree to participate.[37] However, with a response rate of 89% in the DNSFP-2 and the attendance of a representative sample of family practices in The Netherlands, participation bias in our study will be limited. Furthermore, we found 2.1% (59 out of 2784) of the total number of video consultations in the DNSGP-2 concerned consultations with patients with persistent MUS. This is in line with the findings of Verhaak et al. that 2.5% of the patients in primary care present with persistent MUS.[7]

By quantifying on which stages of the consultation patients and FPs focus within the available time of the consultation, we studied the doctor-patient communication in persistent MUS consultations on different levels which improved our understanding of the role of the doctor-patient communication.[23,38] We chose to quantify as measure of focus the number of text lines. We decided to this approach as it had in our view, face validity, but it is important to formally validate it in further study. Besides, we do realize that the FP's and patient's speaking rate, the duration of silences within the consultation and other non-verbal characteristics can make the number of text lines as a proxy of speech focus less reliable. As our goal was not to study doctor-patient interaction on micro level, we choose not to use conversation analysis techniques.[39]

The small sample size and the cross sectional nature of the qualitative analysis preclude definitive conclusions. Our findings should be confirmed in a larger, prospective qualitative study that could track doctor-patient communication regarding the unexplained symptoms over time.

Conclusion

Patients' showed a rather unfocused and fragmented presentation of multiple symptoms during all stages of the persistent MUS consultation. However, ways of giving structure to the consultation such as agenda setting and summarizing were hardly ever used by the FPs. Patients had ample opportunity to tell their story, but the reason for encounter, patients' beliefs and concerns were not discussed in a structured manner. Mostly, patients themselves initiated discussion on the reason for encounter, their beliefs and concerns and the consequences on daily activities, social environment and illness behavior. Furthermore, the extensive explanation of the origin of the symptoms they received from their FP was often not focused on their beliefs and concerns.

Although consultations with persistent MUS patients seemed quite patient-centered as patients have much opportunity for telling their story, patients might benefit more from a structured consultation focused on the exploration of their ideas, concerns and expectations.

Practice implication

Exploration and validation of patients' experiences of illness, patients' distress and patients' concerns and incorporating these items into explanations and reassurance may improve the care of patients with persistent MUS.

Educational interventions in graduate and advanced professional training, aiming at enhancing a systematic symptom exploration and reason for encounter, improving FPs symptom explanation and reassurance during the persistent MUS consultation should be developed and might result in a more focused patient-centered approach which can enhance the wellbeing of patients with MUS.

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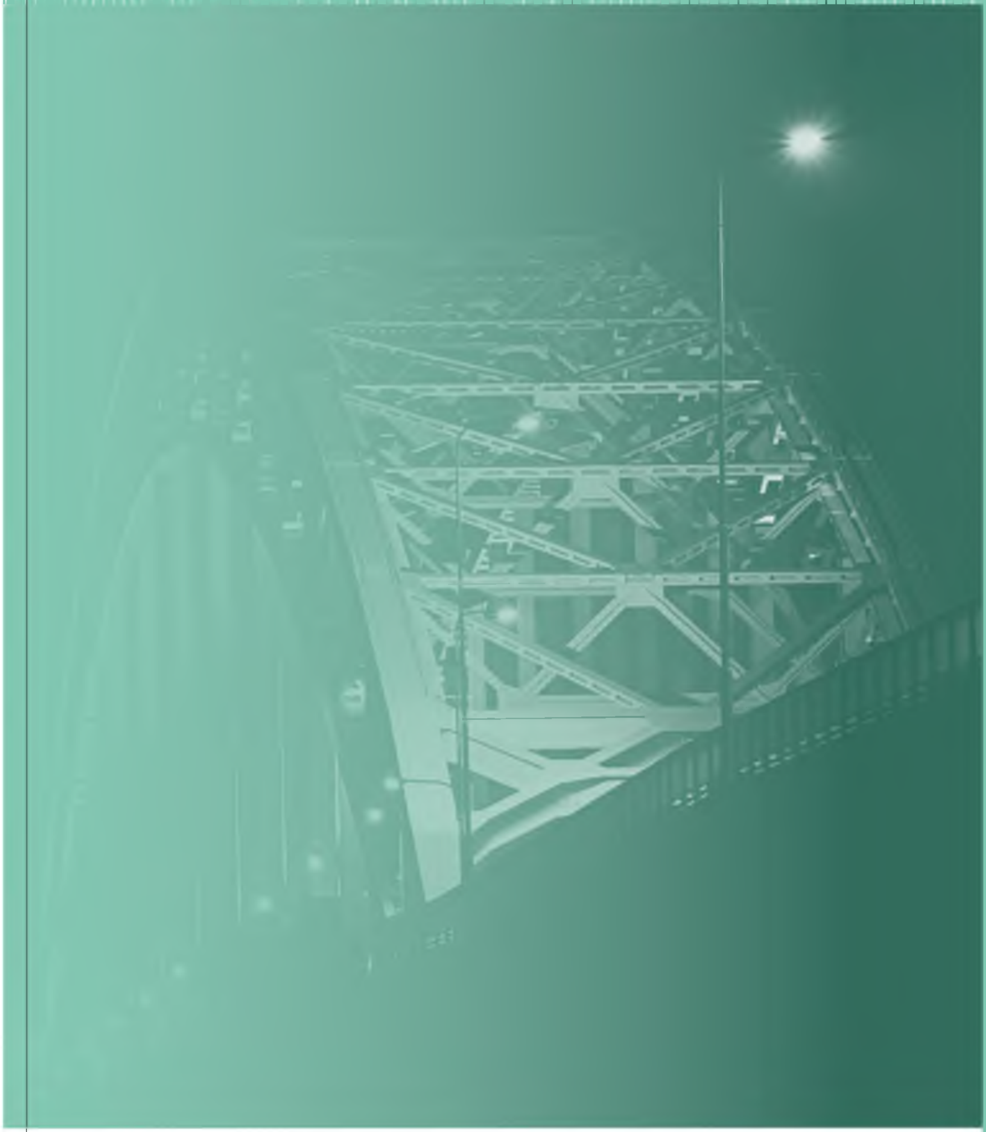
CHAPTER
8

EXPERTS' OPINIONS ON THE MANAGEMENT OF MEDICALLY UNEXPLAINED SYMPTOMS IN PRIMARY CARE. A QUALITATIVE ANALYSIS OF NARRATIVE REVIEWS AND SCIENTIFIC EDITORIALS



Mieke Heijmans
Tim olde Hartman
Evelyn van Weel-Baumgarten
Christopher Dowrick
Peter Lucassen
Chris van Weel

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Summary

Background. The feasibility as well as the suitability of several therapies for medically unexplained symptoms (MUS) in primary care applied by the family physician (FP) appeared to be low. FPs need effective and acceptable strategies to manage these functionally impaired patients.

Objective. To review important and effective elements in the treatment of patients with MUS in primary care according to experts in MUS research.

Methods. We performed a systematic search of narrative reviews and scientific editorials in Medline and PsycINFO and triangulated our findings by conducting a focus group with MUS experts.

Results. We included 7 scientific editorials and 23 narrative reviews. According to MUS experts the most important elements in the treatment of MUS are creating a safe therapeutic environment, generic interventions (such as motivational interviewing, giving tangible explanations, reassurance and regularly scheduled appointments) and specific interventions (such as cognitive approaches and pharmacotherapy). Furthermore, MUS experts indicate that a multi-component approach in which these three important elements are combined are most helpful for patients with MUS. In contrast to most specific interventions, opinions of MUS experts regarding generic interventions and creating a safe therapeutic relationship seem to be more based on theory and experience than on quantitative research.

Conclusions. MUS experts highlight the importance of generic interventions and doctor-patient communication and relationship. However, studies showing the effectiveness of these elements in the management of MUS in primary care is still scarce. Research as well as medical practice should focus more on these non-specific aspects of the medical consultation.

Introduction

Medically unexplained symptoms (MUS) are somatic symptoms that cannot be attributed to a clear organic cause after appropriate medical assessment.[1] MUS are a common and important problem in primary care. In 19% – 50% of all symptoms seen in primary health care, no evidence can be found for any physical disease (i.e. MUS).[2-4] Most of the time MUS are transient and self limiting and do not need further medical attention after one or two consultation(s). A recent Dutch study found that only 2.5% of the attendees in general practice presenting with MUS meet criteria for chronicity.[5;6] However, this minority of patients represent a major problem in health care. These patients suffer from their symptoms, are functionally impaired and are at risk of unnecessary and possibly harmful tests, referrals and treatment.[4;7] Moreover, scarce healthcare resources are wasted without clinical benefit.[2] This leads to frustration for both doctor and patient.

There is an often complex overlap between MUS and common mental health problems, both longitudinally and cross-sectionally.[8] For example, somatic symptoms are common in many psychiatric conditions, such as anxiety or depressive disorders, and of primary care patients with a diagnosable psychiatric disorder, 50% – 70% initially present with somatic symptoms.[9;10]

Several treatments for patients with MUS have been described, with considerable recent research focused in primary care. Some studies show that antidepressants and cognitive behavioural therapy (CBT) are effective in the treatment of persisting MUS, improving symptoms and functional status and reducing psychological distress.[11;12] Reattribution therapy, a structured intervention to provide an explanation of the mechanism of patients' symptoms through negotiation and patient-centred communication,[13] is probably not effective as three of four trials do not show any benefit.[14] Moreover, in one RCT reattribution training by FPs was associated with decreased quality of life.[15] While family physicians (FPs) face a considerable workload from patients with MUS, the applicability of CBT is limited because many patients do not accept CBT as they do not consider their complaints to be 'psychological'. Thereby a coherent and integrative model of disease mechanisms combining predisposing, precipitating and perpetuating factors is lacking.[16]

Moreover, the application of medication is generally seen as less suitable as it is a passive form of treatment.[17] In conclusion, the evidence in this field is that specific interventions for patients with MUS are at best of limited help for FPs.

Therefore, to improve the care for patients with MUS, it seems valuable to consider expert opinions on effective management strategies for patients with MUS. Scientific editorials and narrative reviews are an important resource to learn about the opinions of leaders in the field.[18]

We decided to study the elements experts consider important and effective in the management of MUS in primary care.

Methods

We performed a systematic review with qualitative analysis of editorials and narrative reviews. Both types of papers are usually written by experts in the field. Scientific editorials allow leaders of research and clinical communities to communicate with each other and are a forum for the expression of widely shared expert beliefs and opinions.[18] Narrative reviews tell us what is known about therapies for patients with MUS according to experts in the field. The information obtained from the analysis of the systematic review was triangulated by a focus group with Dutch experts in the field.

Data sources and search strategy

In October 2009 we performed a systematic search in Medline and PsycINFO for narrative reviews and scientific editorials about MUS. We used two search strings and combined these with the Boolean operator AND. The first string consisted of terms indicating somatization (for example: somatization, somatoform disorders, functional somatic syndrome, symptom, medically unexplained). The second search string included terms for treatment (for example therapy, intervention). The search strategy for Medline (see Appendix 1) was adapted for PsycINFO. We pretested the search strategy on five important articles that should be included in our study. Furthermore, we obtained additional references from the reference list of retrieved articles by systematically checking these.

After reading several articles we found that ideas and statements published in articles in the years before 2004 were reviewed and discussed in more recent articles. Therefore, we limited our search strategy to articles published in the last 5 years.

Selection of studies

Two reviewers (MH, physician with an interest in MUS; TCoH, FP with an interest in MUS) independently read all titles and abstracts for inclusion. The full texts of the included abstracts were read by one reviewer (MH) who once again checked for inclusion and exclusion criteria before definitive inclusion. When in doubt she consulted the other reviewer (TCoH). Inclusion criteria were narrative reviews or scientific editorials focussing on the management of patients with MUS. We excluded papers that focused primarily on diagnosis or classification. As we were interested in important elements in the management of undifferentiated MUS and not in the management of specific symptoms or syndromes (for instance melatonin for fibromyalgia or probiotics for IBS), we excluded articles about specific syndromes or single unexplained symptoms. We focused on undifferentiated MUS as we assume that these are more difficult to handle for the physician than single symptom unexplained disorders and functional syndromes: the latter give more opportunity for guideline-based management or a specific referral to a medical specialist with specific interest regarding functional syndromes. Papers on children and adolescents (age <18 years) and papers on specific groups of patients (for example refugees, commercial sex workers) were excluded as well. We calculated the inter-reviewer agreement in article selection based on title and abstracts with kappa statistics.[19] Disagreements were resolved during a consensus meeting.

Analysis

We qualitatively analysed the included scientific editorials and narrative reviews to explore expert opinions about important elements in the management of MUS. Analysis followed the principles of constant comparative analysis, in which included studies are subsequently thematically coded.[20]

Two reviewers (MH and TCoH) independently read two articles (one editorial and one narrative review) to develop a coding scheme. Initial codes were discussed, seeking agreement on their content. After the two reviewers agreed on the coding scheme, one reviewer (MH) coded one editorial and one narrative review. This initial coding was checked by the second reviewer (TCoH). Since no significant discrepancies were discovered, the first reviewer (MH) proceeded to code the entire data set. In the event of doubt or ambiguity the first reviewer (MH) sought the opinion of the second reviewer (TCoH). During such a consensus meeting the coding scheme was reviewed and if necessary modified. Subsequently the transcripts were recoded with the modified coding scheme. We used Atlas.ti qualitative data analysis software for coding and recoding the transcripts. We grouped the codes into themes to identify key features of experts opinion. Recurrent and important themes, identified by the researchers, were frequently discussed and refined as part of an ongoing iterative process.[21] During the entire analysis we constantly matched the developing themes with the transcripts. Therefore, these repeated themes are grounded in the data an not imposed onto the data by the researchers.

To triangulate the results of our qualitative analysis we conducted a focus group meeting. We invited FPs who are participating in the guideline committee on MUS in primary care of the Dutch College of General Practitioners. The participants' characteristics are listed in Table 1. All five have a specific interest in managing patients with MUS in primary care. Moreover, three participants did their PhD in this field.[22-24] Following the guidelines for conducting focus groups, we used an interview guide to direct the discussion and to fulfill the research aims. This interview guide was based on the key themes we identified during the analyses of the articles (Table 2). The discussion was facilitated by a moderator (MH) and lasted for ~1 hour. We audiotaped the discussions in the focus group, transcribed the text and entered it into Atlas.ti. Next, we analysed with two reviewers (MH and TCoH) the text according to the principle of constant comparative analysis and compared the results with the findings from the systematic review.

Table 1. Characteristics of participating FPs in the focus group discussion

Characteristics	Number of family physicians
Gender - Male - Female	 1 4
Working hours - Full time - Part time	 0 5
Type of practice - Solo - Duo - Group	 0 2 3
Age in years (range)	51,4 (48-56)
Experience as a FP in years (range)	18,6 (10-25)

Table 2. Focus group interview guide

Opening	- What are important elements in the management of patients with MUS? - How do you implement this in a consultation?
Creating a safe therapeutic environment	- What are important elements in the management of patients with MUS that create a safe environment? - How do you implement this in a consultation?
Generic interventions	- What are important elements in the management of patients with MUS that belong to this theme? - How do you implement this in a consultation?
Specific interventions	- What are important specific treatments for patients with MUS? - How do you implement this in a consultation?
Multi-component approach	- What are important steps in the management of patients with MUS? - How do you implement this in a consultation? - When do you take which step?
End	- Would anyone like to add elements in the management of patients with MUS which are not discussed today?

Results

We retrieved 960 articles from the electronic databases (572 Medline and 388 PsycINFO). A total of 74 articles found with PsycINFO had already been found in the Medline search (Fig. 1). After screening the titles and abstracts, 53 papers fulfilled the inclusion criteria. The inter-reviewer agreement was κ = 0.89 (95% confidence interval: 0.83-0.96). The full text of 7 of the 53 articles was not available in the Nijmegen library and the authors (living outside The Netherlands) had to be asked for a copy. As after 9 months no response came, these papers could not be included in this study. After reading the full publication, we included 30 of the 46 articles in our analysis. These articles concerned 23 narrative reviews and 7 scientific editorials.[25-54]

During the analysis of the included articles we distinguished four key themes describing the important elements in the management of MUS in primary care according to opinion leaders in the field: (i) creating a safe therapeutic environment, (ii) generic interventions, (iii) specific interventions and (iv) multi-component approach. These themes will be discussed below.

Creating a safe therapeutic environment

According to experts in the field, a doctor has to actively create a safe therapeutic environment before he/she starts a therapy. In such a safe therapeutic environment, the patient should have the opportunity to talk freely about the symptoms and problems that bother him/her. Experts state that a good doctor-patient relationship and good communication are necessary to create such an environment.

Doctor-patient relationship

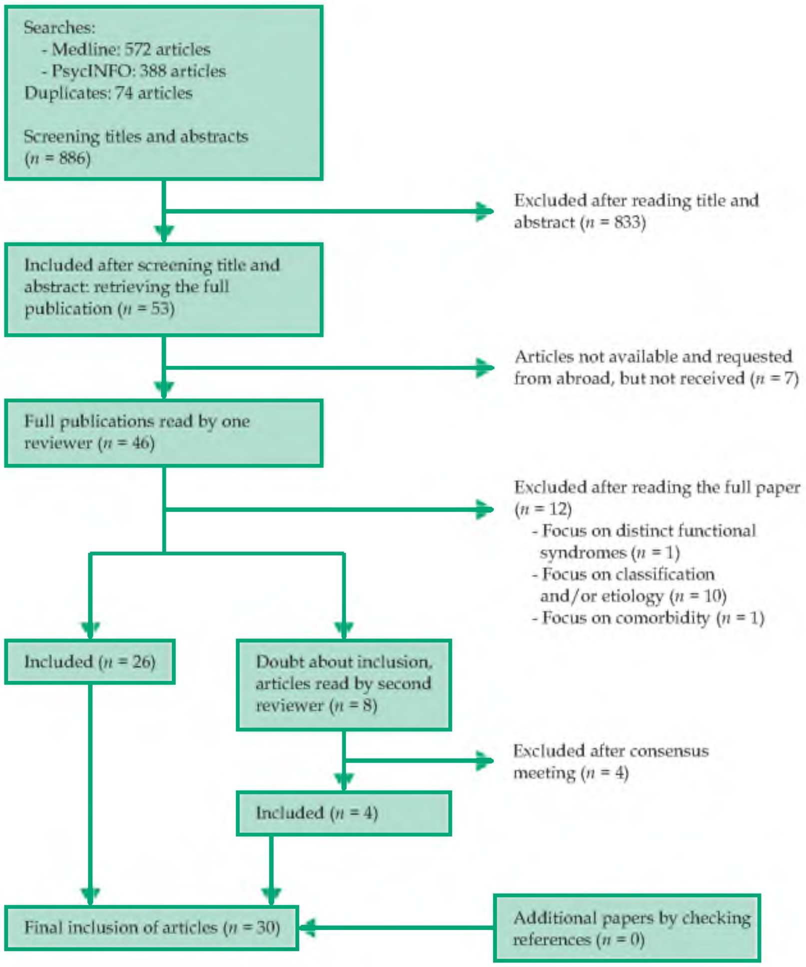
In 17 of the 30 included papers, the importance of the doctor-patient relationship is stressed. However, only one expert referred to quantitative evidence from a randomized clinical trial which studied the effectiveness of a patient-centred method to establish a good patient-provider relationship.[35]

Experts suggest that a good doctor-patient relationship is necessary for a treatment to be effective. A doctor can achieve this by being empathic, by showing the patient that he/she takes the problems and symptoms seriously and that he/she is willing to help the patient.

> *Conversations in the primary care setting usually take place in the context of long-standing, trusting doctor-patient relationships. Such relationships have been shown to be an important factor in the healing process.*[44]

> *The doctor legitimized the patient's suffering, removed blame, and created a therapeutic alliance. The symptom and emotion were thereby linked.*[43]

Figure 1. Selection of studies



A good doctor-patient relationship is not only important at the start of the therapy, according to experts, but also during the course of the treatment. Doctors should find a way to deal with the doctor-patient relationship problems which they face in the contact with these patients.

> *During the course of treatment, a relationship of mutual trust with the patient should be maintained, but if some problem occurs, it should be addressed directly with the patient in a descriptive, yet non-judgmental manner.*[54]

Doctor-patient communication

In 18 of the 30 included narrative reviews and editorials experts discussed the doctor-patient communication. However, none of the MUS experts described quantitative evidence for the effectiveness of the doctor-patient communication.

According to experts, clear and focused communication is an important element in creating a safe therapeutic environment. This means that a doctor should listen carefully to his patient and question the patient extensively about the symptoms, the consequences of the symptoms for daily life and what the symptoms mean to the patient. It is also important to ask the patient about his/her cognitions, emotions, fears and concerns regarding the symptoms. The doctor has to try to understand the patient's beliefs, sources of information and knowledge gaps.

> *Better communication has been associated with higher satisfaction in a number of studies, as well as greater adherence and lower rates of litigation, but few studies have found a relationship between communication and disease or symptom outcomes.*[35]

> *Successful management of patients with MUS has to address the subjective illness perceptions, possibly underlying illness fears and information-processing biases. However, this requires not only knowledge about the patient, but also behavioural skills in the doctor.*[32]

MUS experts stressed that a clear and focused communication between doctor and patient can enhance the doctor-patient relationship and results in a more patient-centred explanation and management of the symptoms. Furthermore, when the influence of psychosocial factors has been elicited in an early stage of the consultation the relevance of psychological factors becomes more acceptable for the patient.

> *To provide the patient with a qualifying explanation, it is necessary to thoroughly explore the patient's illness beliefs and symptom worries. Identification of the patient's dysfunctional beliefs and behaviours lends the possibility of helping the patient to modify them.*[27]

> *Moreover, early recognition and communication of the fact that symptoms may not result from organic disease, and early appreciation of the role of psychosocial factors, may improve outcomes.*[34]

In two papers, experts mentioned, without providing quantitative evidence, that giving a summary during MUS consultations may be important.[32;34] This summary should include relevant physical, psychological, social factors and possible links between them. According to the MUS experts, giving a summary is a way of showing that the doctor is an attentive listener and is interested in the patient's symptoms and problems. Furthermore, it helps the doctor to uncover the patient's opinions and expectations and whether or not the patient agrees with the treatment plan.

Generic interventions

Motivational interviewing

In 13 of the 30 included papers, MUS experts mention motivational interviewing to stimulate patients' motivation and to enhance the efficacy of specific interventions. However, the experts do not refer to quantitative evidence for motivational interviewing. According to the MUS experts, doctors should, for example, encourage appropriate activating behaviour. Furthermore, they have to give patients practical and positive advice for lifestyle changes, which they can apply straight away. Examples are recommendations for (graded) exercise, dietary advice, sleep routine, stress reduction and relaxation.

> *The essence of these recommendations is: to convey to the patient that his symptoms are real, to offer positive advice and treatment and to engage the patient in an active role in alleviating the often chronic symptoms.[…] The evidence of non-pharmacological passive treatments, be they invasive or non-invasive, seems to be weaker than the evidence of non-pharmacological treatments that involve active patients' cooperation.*[33]

> *Encourage patients to bring about change in lifestyle and diet, such as exercising, maintaining regular hours and stopping use of alcohol, caffeine, nicotine and so forth.*[51]

Furthermore, MUS experts stressed the importance of involving patient's allies (family, friends, etc.) in the management of MUS in primary care. In this way, patients would be more motivated to make important lifestyle changes.

Explanation

The importance of explanation of the symptoms in the management of MUS is mentioned in 22 of the 30 included papers. In none of the papers, MUS experts referred to publications quantitatively studying the effectiveness of explanations. According to the experts, a doctor should be able to give the patient a tangible explanation for his/her symptoms, which links the physical complaints with contextual factors and psychosocial influences. Giving the patient a positively formulated explanation with practical advice for management would enhance treatment outcome. They state that explanations should be person centred and adjusted to the patient's cognitions and illness beliefs. However, MUS experts do not give clear examples of explaining the symptoms to patients.

> *Explanations should integrate psychological and biological factors and provide patients and doctors with a model for managing the condition.*[26]

Reassurance

In sixteen of the 30 included papers, the importance of reassurance is highlighted. None of the MUS experts described the effect of reassurance quantitatively. In one narrative review, an expert described that the effect of diagnostic testing depends on what patients think a normal result means.[26]

MUS experts suggest that doctors should explain, educate, give advice and communicate in positive terms, in order to reassure the patient. Sometimes additional tests or referrals will be necessary to reassure the patient. They suggest that prior to the diagnostic tests, the doctor has to explain what a normal test result will mean. Moreover, the doctor should explain what the next step will be if the results are normal and the symptoms persist. Furthermore, while making the choice for further tests or referral, a doctor should consider the risk of iatrogenic harm caused by the additional investigation or referral, according to the MUS experts.

> *Discuss the planned examinations and their consequences with the patient as early as possible. Anticipate when you will stop with medical investigations. Avoid unnecessary medical investigations and petty diagnoses.*[34]

Some MUS experts mention that normalizing symptoms and test results are likely to be more beneficial.[27,34]

Regularly scheduled appointments

In 9 of the 30 included narrative reviews and editorials, MUS experts indicated that regularly scheduled appointments should have a place in the management of patients with MUS in primary care. One expert stated that evidence from randomized controlled trials suggests that regularly scheduled appointments; performing a brief physical examination at each visit, to look for signs of disease rather than relying on symptoms and avoiding investigations and hospital admissions, unless clearly indicated, decrease health service use and increase physical functioning.[26] However, the effect of counselling is not described quantitatively. According to MUS experts, these regularly scheduled appointments enhance the doctor-patient relationship.

> *A schedule of regular, brief follow-up office visits with the physician is an important aspect of treatment. This maintains the therapeutic alliance with the physician, provides a climate of openness and willingness to help, allows the patient an outlet for worry about illness and the opportunity to be reassured repeatedly that the symptoms are not signs of a physical disorder, and allows the physician to confront problems or issues proactively. Scheduled visits may also prevent frequent and unnecessary between-visit contacts and reduce excessive health care use.*[29]

Specific interventions

In the included publications, MUS experts commented about the specific treatments: (i) cognitive approaches, (ii) pharmacotherapy, (iii) activating therapy and (iv) complementary and alternative medicine.

Cognitive approaches

Almost all experts (in 28 of 30 included papers) stress the importance of cognitive approaches in the management of MUS. Of these cognitive approaches, they most often mention CBT. Although they had different opinions about the magnitude of the effect of CBT, many experts described the evidence quantitatively. For example, one expert stated that the results of 31 CBT controlled trials for treatment of somatoform disorders showed that with CBT, patients improved more than controls in 71% of the studies.[54] Another expert stated that 82% of patients with MUS receiving CBT and 64% of control subjects had improved or recovered at 6-month follow-up and that this difference was maintained at 12-month follow-up.[43] However, a third expert pointed out that the results of the effectiveness of CBT were no longer significant after controlling for covariates.[44] Furthermore, Henningsen described moderate evidence for the effectiveness of CBT in patients with MUS or somatoform disorder.[33] Interpretation of the effectiveness of CBT seems complicated as most of the time different variants of CBT are studied. Moreover, it is not clear which specific elements make the CBT effective.

> *In CBT, the therapist structures the patient's social and physical environment to promote appropriate behaviour (in this case, healthy social and personal adjustment without somatisation) and discourage inappropriate behaviour (that is, illness behaviour and preoccupation with physical symptoms).*[43]

Other cognitive therapies studied in the literature are relaxation training, reattribution, biofeedback, body mentalization therapy and other forms of psychotherapy. Most experts state that their contents and results are very heterogeneous, which hampers drawing conclusions regarding their effectiveness.

Pharmacotherapy

In 23 of the 30 included papers, MUS experts discussed the importance of pharmacotherapy in the management of MUS. They suggested that antidepressants can be helpful and provided quantitative evidence for its effectiveness. For example, one expert stated that a systematic review of 94 RCTs with a total of 6595 patients with MUS found that antidepressants significantly improved symptoms (number needed to treat four).[26] However, one expert stated that a literature search did not reveal any published controlled studies evaluating the efficacy of pharmacotherapy for MUS (either the full or abridged somatization disorder diagnosis).[45]

According to MUS experts, antidepressants can reduce symptoms of often co-morbid depression of anxiety disorders. Furthermore, they might also be helpful in relieving symptoms, like pain, in the absence of a co-morbid psychiatric disease. However, the doctor-patient relationship and communication may also play a role in the effectiveness of antidepressants, according to the experts.

> *A literature review including a qualitative comparison of information on understanding and treatment of medically unexplained somatic symptoms was carried out by Burton (2003). He found that CBT and anti-depressant drug are both effective treatments, but their effects may be greatest when the patient feels empowered by the doctor to tackle his or her problem.*[55]

We found some comments on other pharmacotherapeutic agents being studied in the literature, including anxiolytics. However, according to MUS experts, there is not much evidence for their effectiveness in the management of MUS.

Activating therapy

Although in 11 included papers MUS experts described the possibility of activating therapy, none of them describe quantitative evidence for the effectiveness of this therapy. A lot of different activating therapies like graded exercise, physiotherapy and revalidation are mentioned by the experts. The experts suggest that these therapies can be beneficial in some functional somatic syndromes when combined with other therapies. According to MUS experts, patients should agree with the activity. Furthermore, the activity should be person centred and relevant to the individual situation and be structured so that it gradually increases. Doctors also need to tell the patient that he/she might feel temporarily worse but that there will be benefits in the long term.

Expressive therapies like creative therapy or writing disclosure are also mentioned by MUS experts. However, they state that these therapies, like exercise therapies, seem mostly beneficial in combination with other therapies.

> *Cognitive techniques, psycho-education and attention training are suggested to alter cognitive-perceptual factors, and should be combined with the modification of illness behaviour and graded activity.*[47]

Complementary and alternative medicine

In four papers, MUS experts discussed the application of complementary or alternative medicine in the management of MUS. Quantitative evidence was not mentioned. One expert argued that St John's Wort showed excellent efficacy on standardized assessment instruments and outcome measures.[42] Some experts mentioned hypnotherapy for the management of MUS. However, the experts stated that it is not clear which specific element of these therapies is effective.

> *A couple of interesting placebo controlled trials have recently been published reporting on the efficacy of St John's wort in the treatment of somatization disorder, undifferentiated somatoform disorder and somatoform autonomic dysfunction (Volz, Murck, Kasper & Moller, 2002; Muller, Mannel, Murck & Rahlfs, 2004). A Set of standardized assessment instruments and outcome measures were used in both studies and the data showed excellent efficacy, tolerability and safety of St John's wort, independent of any existing depressive symptomatology.*[42]

Multi-component approach

In most scientific editorials and reviews experts indicate that management of MUS should consist of a multi-component approach whereby creating a safe therapeutic environment, and general and specific interventions are combined. MUS experts often mentioned the stepped care model. In this model, severity and chronicity of the symptoms guide the management.

> *Such findings lead to recommendations for stepped care as a basis for routine care:*[28]
> *Step 1: Reassurance, advice, and explanation in the medical clinic;*
> *Step 2: Reassessment, more extended CBT-based discussion and encouragement of self-help;*
> *Step 3: Reassessment, sessions of CBT or other specialist care.*

Results of focus group of experts in the field

The experts in the focus group discussion were inclined to discuss the importance of a safe therapeutic environment (clear and focused doctor-patient communication) and generic interventions (reassurance and explanations).

> *... most important is that people feel they have been taken seriously. Therefore attention and providing the opportunity to discuss all questions and concerns. (FP5, female, 25 years FP working experience)*

Furthermore, participants suggested the importance of a thorough exploration of patients' somatic symptoms, beliefs and concerns, and consequences of these symptoms on patients' daily activities, social environment and illness behaviour in order to reach a better understanding of the patients' symptoms and problems.

> *All symptoms have certain dimensions and the physical dimension is just one of them. However each symptom results in emotions, cognitions and illness behavior. I think that all these*

> *dimensions are important to explore. To look at all these dimensions together with the patient. Sometimes, most of the time, you will find a starting-point for an intervention in one of these dimensions. (FP4, female, 24 years FP working experience)*

Some participants used a symptom diary during the MUS consultations as a tool to explore the cognitions and emotions of the patient. In this symptom diary the patient should write the moment of occurrence of the symptoms and his/her thoughts, fears and actions at that moment.

> *I ask patients to write down their own thinking, especially the thoughts not directly related to disease. (FP3, female, 10 years FP working experience)*

The participants also indicated the importance of giving the patient a positive tangible explanation and practical advices. However, they did not give examples of such explanations and advices. Furthermore they stressed the value of discussing psychosocial factors influencing the symptoms at an early stage in the management of these patients.

> *You have to explain patients at an early stage that you will use a somatic as well as a psychosocial pathway during the management of their unexplained symptoms. (FP1, 15 years FP working experience.)*

The participants mentioned CBT, reattribution and referrals to psychiatrist, psychologist or physiotherapist as specific interventions. They also stressed the importance of a good working relationship with these caregivers.

> *I try to teach the patient cognitive techniques, relaxation exercises, or I refer them to a physiotherapist. (FP 2, 18 years FP working experience)*

The participants agreed that the management of MUS should consist of a multi-component and step-wise approach. The severity of the symptoms and problems makes the FP decide what the next step in the management will be.

> *It is nonsense to think that there should be one management for MUS. It really depends on the type of symptoms, the type of patients and all dimensions that influence these symptoms. (FP4, female, 24 years FP working experience)*

When we compare the results from our literature review with the results of the focus group discussion, we conclude that no additional therapeutic elements were found. However, the participants of the focus group discussion explicitly emphasized the importance of a safe therapeutic environment and generic interventions. Furthermore, participants of the focus group discussion indicated that the management of patients with MUS should consist of a multi-component approach in which creating a safe therapeutic environment, generic and specific interventions are combined.

Discussion

Summary of main findings

According to MUS experts in the field, the most important elements in the management of MUS in primary care are: (i) creating a safe therapeutic environment through a good doctor-patient communication and an effective doctor-patient relationship, (ii) generic interventions such as motivational interviewing, giving tangible explanations and reassurance and (iii) specific interventions such as cognitive approaches and pharmacotherapy. However, in contrast to most specific interventions, experts rarely describe the effects of generic interventions, doctor-patient communication and relationship quantitatively in their scientific editorials and narrative reviews. MUS experts indicate that a multi-component approach in which these three important elements are combined are most helpful for patients with MUS.

Comparison with the literature

MUS experts stress the importance of generic interventions, clear and focused communication, preserving the doctor-patient relationship and other non-specific aspects of the consultation such as described in the patient-centred clinical method in order to affect the outcome of consultations and to reach a therapeutic consultation.[56;57] However, they do not describe the effects of these interventions quantitatively. Although the management of MUS (especially in secondary care) as well as research in this area focus on planned approaches and specific treatments, we assume that these non-specific aspects of the consultation elements, although important for all medical problems, are specifically relevant for the management of MUS because of the paucity of effective interventions.

In recent years, several treatments of persistent unexplained symptoms have been introduced in primary care such as CBT, reattribution therapy, disclosure, group psychotherapy, psychiatric consultation, etc.[23;58-61] However, their effectiveness is questionable and sometime these interventions may be counterproductive.[62] Experts' opinions about the importance of the therapeutic environment, the doctor-patient relationship and communication and the importance of generic interventions indicate an important focus for practicing physicians to manage patients with MUS. These factors fit well into the domain and the principles of primary care.[63] These elements should be applied in routine daily practice, regardless of the origin of the symptoms. The doctor as medicine, as Balint stated years ago, might still be the most important and effective intervention for patients with MUS.[64]

Recent studies suggest that doctors and patients have very different perspectives on MUS and doctors' communication skills.[65] They suggest a mismatch between what patients with MUS want and what they actually receive from their FP. Salmon et al.[66] showed that patients with unexplained symptoms often present opportunities for FPs to address psychological needs. FPs' engagement with these cues, however, seems limited. Furthermore, some FPs provide reassurance without a clear explanation of the symptom, while patients wish to have a convincing, legitimating and empowering explanation.[67-70] Finally, FPs generally showed less empathic responses towards patients with MUS. [71;72] These findings might explain why an effective and clear communication with patients with MUS might not be as straight forward as it seems and why implementation of the results of this study in daily practice may be complex.

Although our study revealed important elements in the management of MUS in primary care, we did not study the effectiveness of these elements. The effectiveness of these separate elements is still not well known. Research in the future should address these issues in order to improve the care for all patients in primary care, especially those with MUS.

Strengths and limitations of this study

This study gives a broad overview of important elements in the treatment of MUS according to opinion leaders. Our findings regarding the specific interventions like cognitive approaches and pharmacotherapy were expected on beforehand. However, as these specific interventions do not help the FP much in daily practice, our findings regarding the more generic interventions are even more important. The fact that we found limited references and quantitative descriptions of the effect of creating a save therapeutic environment and generic interventions reflects the necessity to study the effects of these non-specific aspects of the medical consultation. Although such studies might face methodological problems of measuring the effect of these elements on patients' outcome, there are a view good examples of these kind of studies in primary care. Thomas[73], for example, showed the importance of being positive during consultations with patients with MUS in primary care, whereas van Os et al.[74] examined the effect of depression treatment, empathy and support, and their interaction on patient outcomes for depression in primary care.

By performing our search in only two databases (PubMed and PsycINFO), we might have missed some important editorials regarding the management of MUS. However, most important and high quality peer reviewed journals will be presented in our search. Furthermore, we pretested our search strategy on important publications about MUS in our own database and we could retrieve all of them by searching in PubMed and PsycINFO. Our good inter-observer agreement for inclusion and exclusion enhanced the quality of our literature search. Furthermore, by developing a coding scheme by two independent reviewers and checking the coding process of two papers, we concluded that one reviewer (MH) was able to code the entire data set. Finally, by conducting a focus group discussion in addition to the qualitative analysis of the literature study, we were able to triangulate our findings with opinions of experts in the field.

One could argue that there is some overlap between the key themes that we could distinguish in this study. For example, creating a safe therapeutic environment is interconnected to most generic interventions. Furthermore, some generic interventions, such as motivational interviewing, could also be considered as a specific intervention. However, after an in-depth discussion during the iterative process of analysis we decide to categorize the different element into one of the key themes: creating a safe therapeutic environment, generic interventions or specific interventions.

As we only included scientific editorials and narrative reviews, we could not check whether or not the opinions and statements mentioned in the papers were in concordance with the findings of the original studies. For example, some experts stated that normalizing symptoms is likely to be beneficial while recent literature suggested that this is controversial.[69] Furthermore, MUS experts did not give clear examples of how to explain the symptoms to patients while examples of explanations in primary care research do exist.[69] However, by conducting a focus group discussion in addition to the literature study, we were able to check if experts in the field of MUS agreed with the opinions found in the narrative reviews and scientific editorials. One could argue that by only including narrative reviews and scientific editorials published in the last 5 years and not including original research, important elements in the management of MUS (such as the narrative medical approach) have been missed. However, the validity of our findings was explored by checking our results during a focus group discussion with experts in the field. No new elements in the management of MUS appeared from this discussion. Furthermore, they judged the results to be consistent with their perceptions and experiences.[73]

Conclusions

The experts' opinions on management of MUS seem to be more based on theory and experience, than on high quality research. Although opinion leaders do not describe the evidence regarding its effectiveness quantitatively, they emphasize the importance of creating a safe therapeutic environment and other generic interventions. Furthermore, in accordance with the evidence regarding the effectiveness of specific interventions (i.e cognitive approaches and pharmacotherapy), experts indicate specific interventions as important elements in the management of patients with MUS. Creating a safe therapeutic environment and generic interventions such as motivational interviewing, explanation, reassurance and regularly scheduled appointments might be key to improving the management of patients with MUS in primary care.

Acknowledgments

I would like to thank the FPs who participated in the focus group discussion for their contribution in this study.

Appendix 1. Search strategy

(somatoform disorders[mesh] OR somatization[tw] OR somatisation[tw] OR hypochondriasis[mesh] OR neurasthenia[mesh] OR conversion disorder[mesh] OR somatoform disorder*[tw] OR hypochondriasis[tw] OR neurasthen*[tw] OR conversion disorder*[tw] OR psychophysiologic disorders[Mesh] OR psychosomatic medicine[Mesh] OR psychophysiological disorder*[tw] OR psychosomat*[tw] OR psychosomatic medicine[tw] OR functional somatic sympt*[tw] OR functional somatic syndrom*[tw] OR functional syndrom*[tw] OR unexplained sympt*[tw] OR medically unexplained[tw] OR unexplained medical sympt*[tw] OR psychogen*[tw] OR non-organ*[tw] OR non-specific complain*[tw] OR non-specific sympt*[tw]) AND (("therapy"[Subheading] OR "therapy"[tw] OR "treatment"[tw] OR "therapeutics"[MeSH Terms] OR "therapeutics"[tw]) OR ("therapeutics"[tw]) OR ("management"[tw]) OR "intervention"[tw] OR interventions[tw] OR ("therapies"[tw]) OR ("therapeutic"[tw]) OR ("treatments"[tw]))

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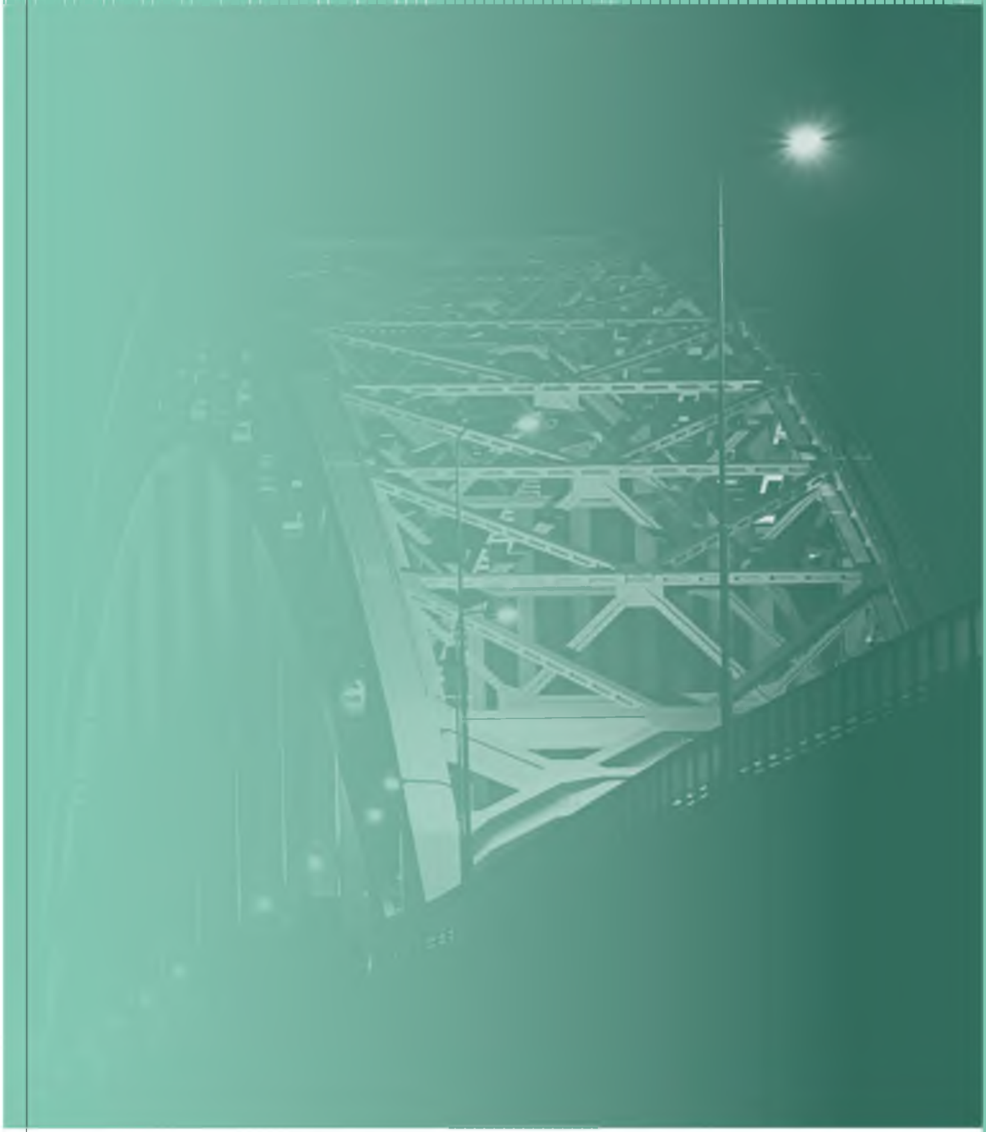
CHAPTER
9

EXPLANATORY MODELS OF MEDICALLY UNEXPLAINED SYMPTOMS: A QUALITATIVE ANALYSIS OF THE LITERATURE



Janine van Ravenzwaaij
Tim olde Hartman
Hiske van Ravesteijn
Eric van Rijswijk
Peter Lucassen

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Abstract

Background. Medically unexplained symptoms (MUS) are common in primary health care. Both patients and doctors are burdened with the symptoms that negatively affect patients' quality of life. General practitioners (GPs) often face difficulties when giving patients legitimate and convincing explanations for their symptoms. This explanation is important for reassuring patients and for maintaining a good doctor-patient communication and relationship.

Objective. To provide an overview of explanatory models for MUS.

Study design. We performed a systematic search of reviews in PsycINFO and PubMed about explanatory models of MUS. We performed a qualitative analysis of the data according to the principles of constant comparative analysis to identify specific explanatory models.

Results. We distinguished nine specific explanatory models of MUS in the literature: somatosensory amplification, sensitisation, sensitivity, immune system sensitisation, endocrine dysregulation, signal filter model, illness behaviour model, autonomous nervous system dysfunction and abnormal proprioception. The nine different explanatory models focus on different domains, including somatic causes, perception, illness behaviour and predisposition. We also found one meta-model, which incorporates these four domains: the cognitive behavioural therapy model.

Conclusion. Although GPs often face difficulties when providing explanations to patients with MUS, there are multiple explanatory models in the scientific literature that may be of use in daily medical practice.

Introduction

Medically unexplained symptoms (MUS) have a high prevalence in health care. Physical symptoms such as headache, backache, pain in muscles and joints and fatigue are common. In the general population two-thirds of men and four-fifths of women report at least one of these complaints in the previous two weeks.[1] In about 25-50% of symptoms seen in primary health care, no evidence can be found for any physical disease.[2,3] In specialist care these percentages are even higher, ranging from 30 to 70%.[4,5]

MUS can become chronic. Patients with persistent MUS are at risk for extensive investigations and referrals, therefore becoming a great burden on health care.[6,7] Doctors and patients are both burdened by the phenomenon of symptoms without disease. Bodily symptoms with unknown physical pathology have a great impact on patient functioning. Such patients suffer greatly from the symptoms and their quality of life is negatively affected.[8,9]

Unexplained physical symptoms are often confusing for both doctor and patient.[10,11] Many general practitioners (GPs) feel powerless and irritated when patients repeatedly visit their practice with these symptoms.[12] Patients often feel disbelieved and not taken seriously by their doctor.[13] Although it is often suggested that GPs are pressured by patients with MUS to deliver somatic interventions, Ring *et al* pointed out that patients with MUS request somatic interventions less often than physicians offer them.[14] Moreover, patients seek emotional support and a legitimate and convincing explanation for their symptoms.[15-17]

GPs recognise the importance of explaining the diagnosis of MUS adequately to patients with persistent MUS. However, they often face difficulties in explaining the nature of the symptoms during the clinical encounter with these patients.[18] Therefore, we searched and analysed the literature for explanatory models for MUS. Providing an overview of such models can improve the knowledge and communication of GPs, thus enhancing the quality of care for patients with MUS.

Methods

Data sources and search strategy

We performed a qualitative analysis of systematic and narrative reviews on the topic of medically unexplained symptoms using the databases PubMed and PsycINFO. We decided to search for reviews, as in this type of article views of MUS and explanatory models are frequently discussed. Our search strategy consisted of two search strings which we combined with the

Boolean operator AND. The first string contained keywords regarding MUS, combined with the Boolean operator OR. The second string of our search strategy contained terms for explanatory models, combined with OR (see Figure 1). This search string was limited to reviews, the English and Dutch languages, articles published in the last five years, and age over18 years. We limited our search strategy to articles published in the last five years as most articles about explanatory models of MUS published before 2005 have been reviewed in more recent reviews.

We tested the accuracy of our search strategy by checking whether or not five key papers on explanatory models in MUS were included in the results.

Figure 1. Search strategy

("2005/01/01"[Publication Date] : "3000"[Publication Date]) AND (((((Model [tw] OR models [tw] OR conceptual*[tw] OR concept [tw] OR concepts OR pathophysiolog*[tw] OR physiopatholog*[tw] OR mechanism* [tw] OR causal* [tw] OR cause [tw] OR explanat* [tw] OR etiology [tw] OR aetiology [tw] OR aitiology [tw]) AND (somatoform disorder[mesh] OR somatization[tw] OR somatisation[tw] OR hypochondriasis[mesh] OR neurasthenia[mesh] OR conversion disorder[mesh] OR somatoform disorder*[tw] OR hypochondriasis[tw] OR neurasthen*[tw] OR conversion disorder*[tw] OR psychophysiological disorder[Mesh] OR psychosomatic medicine[Mesh] OR psychophysiological disorder*[tw] OR psychosomat*[tw] OR psychosomatic medicine[tw] OR functional somatic sympt*[tw] OR functional somatic syndrom*[tw] OR functional syndrom*[tw] OR unexplained sympt*[tw] OR medically unexplained[tw] OR unexplained medical sympt*[tw] OR psychogen*[tw] OR non-organ*[tw] OR non-specific complain*[tw] OR non-specific sympt*[tw])))))

Study selection

Two researchers (JvR, ToH) independently performed inclusion and exclusion of articles, studying title and abstract. In case of doubt they consulted the full paper. Disagreements on inclusion were discussed in a consensus meeting. All disagreements were easily resolved. We calculated inter-rater agreement for inclusion with kappa statistics.[19]

We excluded studies that focused primarily on patients suffering from single-symptom unexplained disorder (tension headaches, dysmenorrhoea) and distinctive functional somatic syndromes (irritable bowel syndrome, chronic fatigue syndrome) because we were interested in explanatory models of undifferentiated MUS in the literature. We focused on undifferentiated MUS as we assume that these are more difficult to explain than single symptom unexplained disorders and distinctive functional syndromes.[20] We also excluded studies that focused primarily on patients with medical or psychiatric disease (except somatoform disorders). Studies on children and adolescents (age less than 18 years) and studies on specific groups of patients such as refugees, street prostitutes etc. were also excluded.

Data-analysis

We analysed the included reviews for explanatory models describing the cause of MUS. The publications were fully entered into a computer database (Atlas.ti) suitable for qualitative processing. The collection and analysis of data from the included reviews was performed both parallel and cyclic, thus mutually influencing each other. First, two researchers (JvR and ToH) independently read the articles in which many different models were assembled, to develop a coding scheme of explanatory models. Initial coding was discussed to seek agreement on content. The coding was improved, adjusted, explicated and specified by applying the constant comparative method.[21] One researcher (JvR) thematically coded the included articles in Atlas.ti according to the final coding scheme.

Results

We retrieved 710 articles from the search in the electronic databases (480 PubMed and 230 PsycINFO). Sixty-five papers were duplicates. After two independent researchers screened title and abstract, 24 papers fulfilled the inclusion criteria (Figure 2). The inter-rater agreement (kappa) was 0.65 (95% CI: 0.51- 0.79), which was considered 'good'. Two articles were not available in the Netherlands and were therefore excluded. After reading the full text, 19 out of 22 articles were included in our study.[13,22-39] The three articles that were excluded reported on therapy/diagnosis or somatic disease and one turned out to be a review of a book.

We could distinguish nine different explanatory models (somatosensory amplification, sensitisation, sensitivity, immune system sensitisation, endocrine dysregulation, signal filter model, illness behaviour model, autonomous nervous system dysfunction and abnormal proprioception) and one meta-model (the cognitive behavioural therapy model) that contains components of these nine different explanatory models. Each model is described, including citations and comments from the reviews.

Explanatory models

A. Somatosensory amplification theory

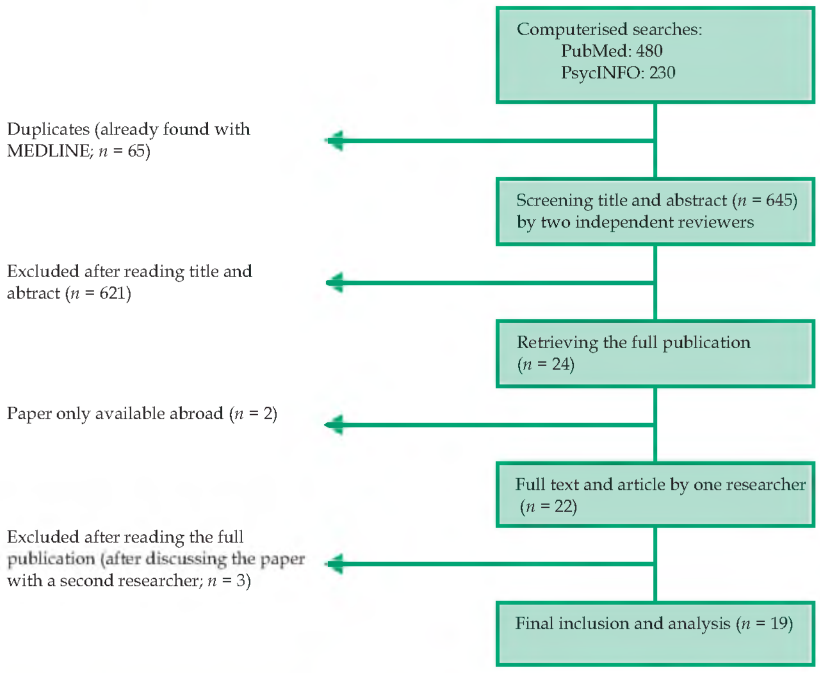
The process described as somatosensory amplification suggests that a physical sensation arises and that as a consequence, patients focus their attention on this sensation. They develop certain cognitions and attributions which further amplify the perception of these physical signals. This amplification results in a vicious circle in a way that symptoms are reinforced by patients' thoughts and concerns. As a result patients with MUS experience a range of feelings as more severe, more damaging, and more alarming.

> *'The strength of this model is its simple formulation, and it can even be used to explain the disorder to patients. The basic mechanisms used in this model, such as attention, perception, and attribution processes have some empirical validation, although the model neglects many other well-validated factors, or offers only indirect explanations for them.' (p. 837)*[22]

> *'Petrie and Weinman (2003) have called for more attention to be given to symptom appraisal and we would widen this by calling for more attention to attention in general. The theoretical literature and some of the empirical literature supports this mechanism as being an important part of the cycle maintaining MUS.' (p. 791)*[23]

> *'Amplification has, in general, been found to be related to reporting of somatic symptoms. However, there are conflicting reports on whether this is an independent effect or whether this is mediated by such factors as anxiety, depression and negative affect/neuroticism. Findings suggest that somatosensory amplification can only partially account for somatization, and that other mechanisms may also be important in this process.' (p. 28)*[26]

Figure 2. Selection of studies



B. Sensitisation theory

Sensitisation means having an enhanced somatic response to sensations as a result of former experiences of these sensations. In patients with MUS, repeated experiences of pain and symptoms can lead to memory traces at a neuronal level which increase sensitivity for future stimulation. This could result in normal benign stimuli being perceived as pain. A patient's body reacts stronger to stimuli when it has become more sensitive by earlier and repetitive encounters. The process of sensitisation has, besides a neural and sensory part, also a psychological component. In MUS in general, a larger memory complex may play a role. Experiencing a single symptom would not only sensitise this sensation, but would also activate a wider memory trace. This in turn, can result in the experience of other physical symptoms. Therefore, sensitization may cause a wide range of symptoms. Furthermore, expectations also play a role in further sensitisation.

> *'The development of symptom memories can be associated with cerebral restructuring. This has been shown for single pain symptoms, where already 24 hours of pain perception can cause neuronal reorganization (neural plasticity) that will facilitate and intensify further symptom perceptions (Arnstein, 1997). For the phenomenon of multiple physical complaints, a general symptom memory matrix can be postulated.' (p. 830)*[22]

> *'The repeated perception of physical signals in combination with uncertainty about the origin of the sensations can hinder the habituation that would ordinarily be expected.' (p. 1000)*[27]

C. Sensitivity theory

This theory suggests that some individuals are more vulnerable to develop MUS. This vulnerability can be based on personality traits, such as negative affect and neuroticism. Furthermore, patients with MUS seem to have difficulty in experiencing the relationship between bodily signals and emotions and thoughts. Catastrophic thinking may also play a part in the vulnerability of pain in these patients. There is little evidence for genetic influences, but many researchers suggest that early childhood experiences, such as abuse, insecure attachment and parental influence, play an important role in the development of MUS.

> *'Viewing the MUS from the perspective of underlying developmental influences that affect the function of a variety of organs based on familial (genetic and environmental) predispositions rather than from traditional viewpoint of isolated organ-originated diseases has at least two important implications. First, it provides a more parsimonious explanation for many findings that have been quite difficult to account for. (...) Second, and more importantly, it invites investigation of new areas of therapy that may otherwise escape consideration.' (p. 142)*[28]

> *'Studies within the framework of attachment theory have provided clear evidence that insecure attachment patterns, and in particular an insecure dismissing attachment pattern, are associated with an avoidant style of affect regulation.' (p. 21)*[29]

D. Immune system sensitization theory

The brain has a cytokine system that reacts to the immune system. It monitors danger in parts of our body and coordinates the responses to these threats. The brain cytokine system is activated by the immune system and mediates the subjective, behavioral and physiological components of sickness, in a reversible way. It can be sensitised in response to activation during early stages of development, repetitive stimulation or prior exposure to immunological stimuli. The brain cytokine system, when sensitised, reacts very fast and is less likely to shut down after eliminating the initial stimulus. Furthermore the brain cytokine system can be triggered by non-immunological stimuli. In patients with MUS, a chronic immune activation with production of cytokines can act as a motivation for the brain to change priorities in face of the presented threat (such as stress or trauma) resulting in a feeling of being sick.

> *'The brain cytokine system also plays a key role in the experience of pain that is associated with danger, to the point that it has been proposed that pain is actually the main determinant of sickness behaviour rather than just a component of it (Watkins and Mayer, 2000).' (p. 951)*[30]

> *'The main medical implication of this view is that many somatization symptoms including depressed mood, fatigue, and pain may represent the expression of a previously sensitized brain cytokine system that is reactivated by infectious or noninfectious trauma.' (p. 853)*[31]

> *'A growing body of evidence suggests that pathophysiological processes explain some of the aspects of illness behaviour that are typically viewed as psychological in origin. The experience of general malaise or feeling sick has a physiological basis, mediated by centrally acting proinflammatory cytokines such as interleukin and tumour necrosis factor.' (p. 56)*[32]

E. Endocrine dysregulation theory

In the hypothalamus pituitary adrenal (HPA) axis, feedback loops exist to regulate the body's response to acute and chronic stress. Dysregulation of this axis has been found in patients with MUS. One interpretation is that prolonged activation has led to a 'burnout' response and a down regulation of HPA activity in MUS. Another suggestion is that hypocortisolism may in fact be a protective response of the body. Hypercortisolism has been found in patients with MUS. Early traumata during pregnancy or childhood can have long lasting effects on the stress sensitivity of the HPA axis which may be associated with increased prevalence of MUS.

> *'The link so far found between central nervous system processes, such as the HPA axis, and immunological processes are intriguing but far from conclusive; the causal relationships are unclear, as are the nature of the change in these systems in different conditions at different stages. There is however already sufficient data to propose hypotheses about some of the important links, for example, between life events, HPA axis and immune functioning, that could be tested in prospective studies.' (p. 791)*[23]

> *'We can conclude that the relevance of the HPA-axis for the somatization syndrome is still unclear. HPA-activity definitely plays a role; however, this role might be unspecific, course depending, and multi-directional.' (p. 998)*[27]

F. Signal filter theory

There is a permanent sensory stimulation from the body sending information to the brain. In healthy individuals, however, this 'sensory noise' is filtered, in order to ensure that the brain is not over-stimulated by information from physiological processes. In patients with MUS 'faulty filtering' leads to the inability of these patients to differentiate between information from physiological process (produced by the body) and information from pathophysiological processes (produced externally). Patients with MUS experience both types of information. Therefore, the number of physical sensations experienced by these patients is increased.

> *'The perception-filtering-model is in line with the findings on the relevance of memory processes and expectation, two empirically well-founded mechanisms not directly included in the other models. Further strength of this model is the close relationship to the neuronal process of perception. Therefore they offer a link between psychological and psychobiological findings on MUS.' (p. 837)*[22]

> *'The effect of distraction on pain perception was demonstrated by Bantick et al., who found that distraction leads to reduced activity in pain-associated centers (Bantick et al., 2002), again supporting a signal-filter-model as presented.' (p. 999)*[27]

G. Illness behaviour theory

This theory hypothesises that patients' believes influence their behaviour. This behaviour can in turn affect physiology and symptoms, resulting in a vicious circle and maintaining symptoms. Avoidance of physical, social or mental activity can result in more symptoms. For example, when a patient with chronic fatigue believes she will get more tired by doing sports, she will stop all physical activity. This may result in an increase of bodily attention and physical deconditioning, ending in more awareness and susceptibility of physical symptoms. Therefore symptoms can be sustained because of patients' behaviour.

> *'There is actually relatively little literature concerning illness responses, despite a clinically prevalent belief that 'all or nothing coping' and avoidance behaviours are important in the onset and perpetuation of syndromes such as CFS. More longitudinal work of this nature is needed to clarify the role of behaviour in the development of MUS.' (p. 787)*[23]

> *'Behavioral aspects are also important in operant conditioning of illness behavior, coifirmation of health attitudes, and the development of physical deconditioning. While these aspects could be of major importance for this patient group, their role has been insufficiently investigated in scientific trials.' (p. 836)*[22]

> *'Cognitive, emotional and behavioural factors have the capacity to relieve symptoms and even change the brain.' (p. 994)*[33]

H. Autonomic nervous system dysfunction theory

Autonomic nervous system (ANS) dysfunction is a potential mechanism connecting psychosocial stress to MUS. In healthy controls, the change from attention tasks to rest periods is associated with a substantial decrease in heart rate activity (`recovery response'). This reduction of physiological activity after mentally distressing tasks is not present in patients with MUS. It is hypothesised that this is a result of a parasympathetic nerve system dysfunction, resulting in a long lasting increased heart rate and stress burden in these patients.

> *'To summarize the results on autonomic physiological activity, we can conclude that only few studies have addressed this question so far. Only small differences have been found, although there is some consistency indicating the involvement of the cardiovascular system.' (p. 998)*[27]

> *'We conclude that current available evidence is not adequate to firmly reject or accept a role of ANS dysfunction in functional somatic disorders and it would therefore be misleading to provide a definitive summary estimate.' (p. 108)*[34]

I. Abnormal proprioception theory

Increased or abnormal proprioception can be a cause of physical symptoms in patients with MUS. It is suggested that patients with MUS demonstrate more exact and sensitive perception of their body than healthy individuals. In patients with MUS, minimal changes in muscle tension would lead to an enhanced feeling of abnormality. Therefore, benign physiological sensations (small changes in their body) can be interpreted as signs of a physical disease.

> *'If patients with MUS perceive physical sensations more precisely, this could lead to increased likelihoods of perceiving even minor physical symptoms, although these differences could also be due to higher distraction by external stimuli in healthy controls.' (p. 828)*[22]

J. Cognitive behavioural therapy model

This meta-model proposes that the cause of MUS is a self-perpetuating multi-factorial cycle, with interaction of different factors in several domains. This model provides a framework to incorporate patients' own personal perpetuating factors as well as predisposing and precipitating factors. Each factor can result in physical symptoms and/or distress. Doctor and patient together have to search for the patient's personal circumstances that might contribute to the distress. Furthermore, this meta-model incorporates processes from at least five different theories described above: sensitivity, sensitisation, somatosensory amplification, endocrine dysregulation and illness behaviour model.

> *'This is the explicit purpose of the CBT assessment: to form a coherent multi-factorial case conceptualization that forms the rationale for treatment.' (p. 789)*[23]

> *'The biopsychosocial perspective becomes increasingly sophisticated, thus allowing the formation of a tight chain of findings from psychology to specific disease processes playing a role in the etiology and maintenance of illness conditions.' (p. 182)*[39]

> *'As such the autopoietic explanation of MUS as proposed by the CBT model both fits the current data and could form a theoretically coherent basis for further research. More generally, the research bears out the over-arching CBT hypothesis that the autopoietic interaction of distinct but linked systems could serve to produce physical symptoms in the absence of physical pathology.' (p. 789)*[23]

Discussion

Summary of main findings

This review illustrates a considerable number of explanatory models of MUS, grounded in the scientific literature. We could distinguish nine different explanatory models of MUS in the literature: somatosensory amplification, sensitisation, sensitivity, immune system sensitisation, endocrine dysregulation, signal filter model, illness behaviour model, autonomous nervous system dysfunction and abnormal proprioception. Furthermore, we found one meta-model, the cognitive behavioural therapy model.

Some of the models aim at a physical explanation, such as the immune system sensitisation theory, the endocrine dysregulation theory, the autonomic nervous system dysfunction theory and the abnormal proprioception theory. Other models aim at a psychological explanation, such as the somatosensory amplification theory and the sensitivity theory. And some models combine a physical and psychological explanation, such as the sensitisation theory, the signal filter theory and the illness behavior model.

The nine different explanatory models seek an explanation in different domains, including somatic causes, perception, illness behaviour and predisposition. The meta-model integrates these four domains.

Medical explanations in clinical practice

Current medical training focuses on acting (diagnosing and treating patients) instead of listening, explaining and reflecting. Several studies pointed out that patients seek legitimacy for their symptoms.[16,40-42] They want to feel that the doctor accepts that the symptoms are real and warrant the doctor's attention.[25] Therefore, good and relevant doctor consultation skills, including explaining symptoms, are needed. Plenty doctors think in terms of action and reaction, while the explanation of symptoms in itself might be the most important intervention for patients with MUS.[43] Such explanations might prevent patients from extending or elaborating symptoms and doctors from providing investigations or somatic treatment.[3] Explanation as a consultation skill in its own right is rarely addressed in the literature and teaching programs. As education on explaining and explanatory models is limited in today's clinical education programs, medical students and GPs have little knowledge of theories and models which they can use during consultation. This might explain part of the difficulties GPs experience in giving an adequate and tangible explanation to patients with MUS. However, GPs indicate that they build their own explanatory models of medically unexplained symptoms based on their experience in daily practice.[44] Furthermore, building acceptable and effective (i.e. reassuring) explanations together with the patient needs a mutual understanding of patients' beliefs, concerns and expectations regarding their symptoms.[45,46] Knowledge of explanatory models of MUS, together with this mutual understanding and daily practice experiences can facilitate the doctor-patient communication and strengthen the doctor-patient relationship with these patients. Furthermore, mutual understanding between GP and individual patients on the aetiology of MUS might result in greater reassurance, patient satisfaction and commitment to the proposed interventions.[25]

Strengths and limitations of this study

In this qualitative analysis of the literature, we used an extensive and systematic search strategy to identify relevant reviews. Including the full text papers and having them coded by two independent researchers added rigour to our study. Moreover, we had good inter-rater agreement for inclusion and exclusion.

By using a cyclical way of analysing data, we were able to focus and explore explanatory models in depth.[47] Entering the full text of included studies into Atlas.ti and using constant comparative method to code and reorganise data strengthened our findings.

We limited our literature search to the past five years. It seems, however, that we have captured most explanatory models in the literature as the reviews included in our study also discussed and summarised explanatory models described in earlier literature. Although across cultures many systems of medicine provide sociosomatic explanations linking problems in family and community with bodily distress, we did not find culturally based explanatory models in our literature search.[48]

A qualitative analysis of the literature is not as objective as a meta-analysis. However, we were able to summarise the range of explanatory models grounded in the current scientific literature. As studying the scientific evidence of the different models was not the goal of our study, we are not able to draw conclusions on the degree of evidence of the explanatory models found in the literature.

Implications for future practice and research

This review illustrates quite a number of different explanatory models of MUS described in literature. Most theories are based on symptom perception, somatic causes, illness behaviour and predisposition. On the other hand, more progress has to be made towards a fuller understanding of the complex aetiology of MUS.

Further studies using in-depth interviews with GPs may reveal new explanatory models based on experiences in daily medical practice. This qualitative analysis of the literature examines explanatory models of MUS and not the usefulness of these models in clinical practice. Therefore, new research has to clarify the usefulness of the different explanatory models in daily practice. In addition, studies using a mixed method methodology have to point out patient preferences and the effectiveness of the explanatory models individually in family practice.

As persistent MUS are present in all medical specialties, these explanatory models should be integrated in the educational programs of all medical doctors in order to improve the quality of care for patients with persistent MUS.

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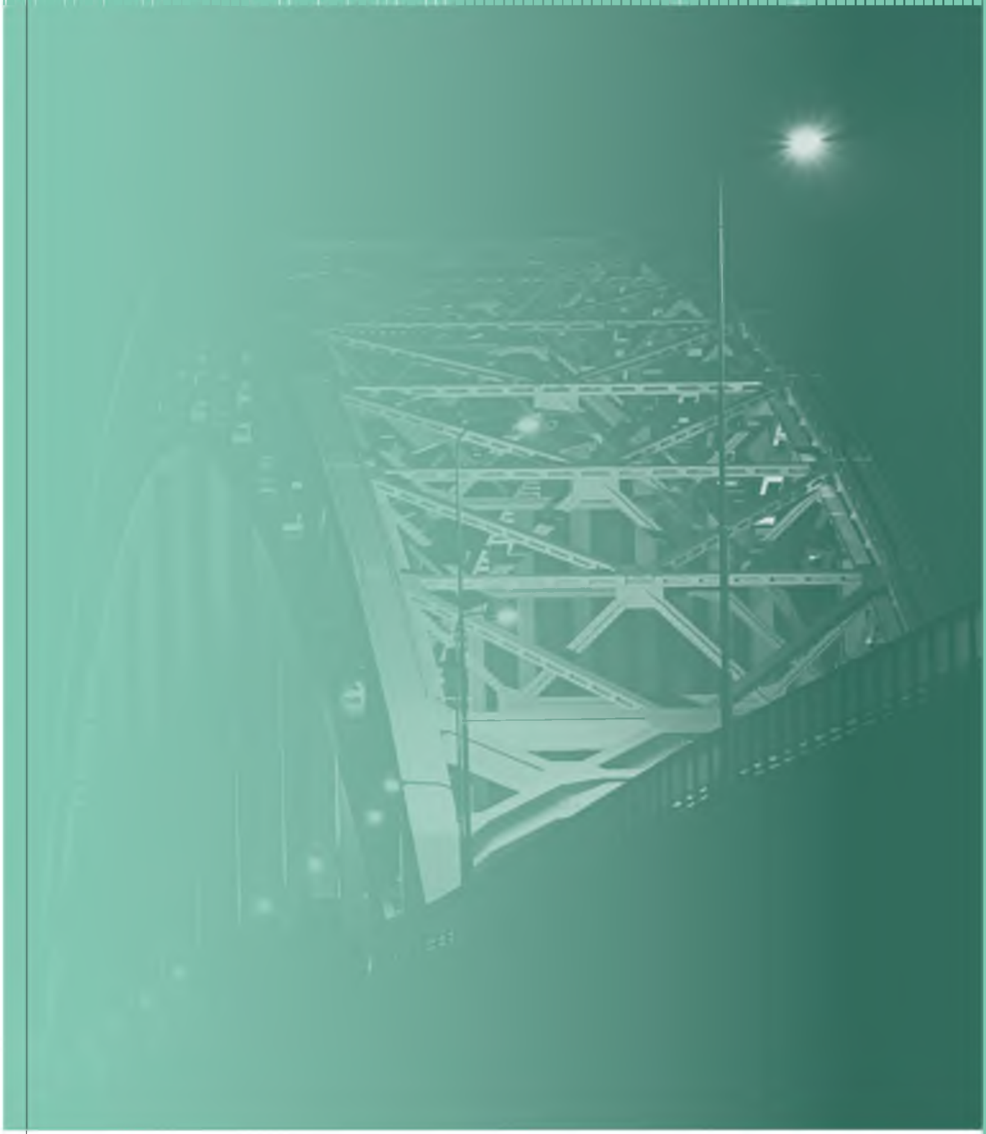
CHAPTER

10

MEDICALLY UNEXPLAINED SYMPTOMS IN FAMILY MEDICINE: DEFINING A RESEARCH AGENDA. PROCEEDINGS FROM WONCA 2007



Tim olde Hartman
Lieke Hassink-Franke
Christopher Dowrick
Sandra Fortes
Cindy Lam
Henriette van der Horst
Peter Lucassen
Evelyn van Weel-Baumgarten

Family Practice 2008; 25(4):266-71

Abstract

Background. Medically unexplained symptoms (MUS) are frequently presented in primary care. Unfortunately, knowledge of these patients and/or symptoms in primary care is still limited. Available research comes mainly from Europe and North America, while the perspectives of cultures such as Africa, Asia and South America are relatively unknown. To bring cultural perspectives together, a symposium and workshop on MUS in primary care was held at the WONCA World Conference 2007 in Singapore.

Objective. Main goals of this symposium and workshop – apart from presenting ongoing research and bringing together experts in MUS – were to detect knowledge gaps in MUS and to establish priorities in MUS research. This publication focuses on the proposed research agenda.

Methods. Using a nominal group technique we generated research topics and set priorities. Research topics were grouped into research themes.

Results. Participants' (66 researchers and GPs from 29 nationalities) most important research topics were 'formulating a broadly accepted definition of MUS', 'finding a strategy to recognize MUS better and at an earlier stage', 'studying the value of self-management and empowerment in patients with MUS' and 'finding predictors to decide which strategy will best help the individual patient with MUS'. Priorities in research themes of MUS are: (i) therapeutic options for patients with MUS and (ii) problems in consultations with these patients.

Conclusions. More research on MUS in primary care is needed to improve the consultations with and management of these patients. Internationally primary care conferences are excellent for exchanging ideas and formulating central issues of research.

Introduction

Acute medically unexplained symptoms (MUS) are common in family medicine. Twenty to fifty per cent of all contacts between patient and GPs concern unexplained physical symptoms.[1,2] Mostly, these symptoms resolve or do not need further medical attention after one consultation. However, there is a group of patients who frequently consult the GP with unexplained symptoms. In a Dutch survey of GP-patient contacts, the prevalence of patients with at least four contacts per year for MUS was 2,5%.[3] Frequent attenders with MUS have a high risk of extensive investigations and referrals.[4] Only about a quarter of patients frequently presenting MUS met the criteria of one of the DSM-IV somatoform categories (i.e. somatisation or undifferentiated somatoform disorder).[5] This means that the majority of patients with MUS in primary care are different from patients meeting DSM-IV criteria for somatoform disorder. Consequently, this relevant group of patients in primary care deserve to be studied seperately.[6] However, study and hence knowledge of many aspects of this group of MUS patients is scarce. For instance, we are still debating the exact definition and classification of MUS which is strongly related to cultural differences.[7] Then, we know little about the prognosis of the symptoms, and we have limited knowledge about factors predicting the prognosis or its course of MUS over time. Furthermore, there is doubt about the best approach to and treatment of these patients.

In the traditional approach of patients who repeatedly present MUS, GPs try to reassure them by giving some – but often inadequate – explanation about the symptoms. Furthermore, concerns of patients with MUS are less likely to be explored and facilitation of patients' talk about psychosocial problems is limited.[8-10] As many patients (and doctors) are not satisfied by this traditional approach, in recent years some research has been done on cognitive behavioural therapy (CBT)-based interventions. These studies show conflicting results. Most studies show no effect of CBT interventions on well-being, sick leave and limitations due to the symptoms.[11-14] Several explanations may be offered for these disappointing results: the training of the GPs might have been insufficient or important issues such as the anxiety of the patients might not have been adequately addressed. Furthermore, an adequate explanation of the symptoms and non-specific effects caused by mutual trust, empathy and shared understanding have not been studied.[15] Alternatively, one might ask whether the outcomes, such as number and duration of symptoms, well-being, sick leave and limitations that doctors or researchers usually study, are the outcomes that patients would also prefer. We must conclude that there are still many uncertainties about this group of patients.

Although MUS is a universally common phenomenon, most research on MUS comes from Europe and North America. The perspectives of other cultures such as Africa, the Asia Pacific and South America are relatively unknown. It is known from recent research that there are cultural differences in the distribution of MUS and the meaning and significance of a symptom depends on the perceived relationship with diseases in a culture.[7,16] Moreover, cultural and social norms determine whether a person with a particular MUS can adopt the sick role: cultures such as the Chinese reject psychological symptoms and therefore cause psychosomatization.[17] Bringing these international perspectives together might help to fill the gaps in knowledge about MUS.

We therefore organized a symposium and a workshop about MUS in primary care during the WONCA World Conference 2007 (Singapore, July 26). Our aim was to gather topics for research on MUS from discussions with researchers and GPs from all over the world and to prioritize these topics in order to establish a global research agenda.

Methods

During the symposium 'MUS in family medicine: the state of the art', GP researchers presented ongoing research in MUS in different countries all over the world. The two main themes were the classification and cultural variation of MUS and the process of consultation with MUS patients.[18-20] The symposium was also meant as an introduction to the subsequent workshop. The aim of this workshop 'MUS in family medicine: where should we go?' was to bring together GPs with a broad cultural variety interested in MUS to discuss ideas about research topics in the field of MUS and to make choices on which ideas needed priority.

Participants in the workshop

In order to discuss the various perspectives of MUS research and bring together GPs from different cultural backgrounds to look at the broader picture of MUS, we invited researchers and interested GPs from different nationalities. The nationality of each participant in the workshop was registered by one of the organizers (TCoH).

Nominal group technique

In the workshop, we used the nominal group technique to generate a research agenda. In the nominal group technique, participants are brought together for discussion. The procedure usually comprises four stages: generating ideas, recording of those ideas, clarification and priority voting.[21] This method encourages contributions from everyone by allowing equal participation among participants. Moreover, it is a structured, transparent and replicable way of synthesizing and generating ideas.[22]

During the workshop, we generated ideas by stimulating discussion: we allowed 10 minutes of brainstorming in small groups to discuss what they thought to be the most relevant MUS research topics. Each small group made a shortlist of their most important research topics. We continued with an inventory of all relevant topics arising from these discussions. We wrote the topics onto flip charts. If necessary, the discussion leaders (LJAH-F, EMvW-B) asked for clarification. After this inventory, each participant was asked to choose five topics with the highest priority from this list and rank them. Finally, we asked all the participants to vote, allocating the highest score (5) to the most important research topic, then 4 to the next most important statement and so on, by sticking coloured cards on the flip charts. The organizers of the workshop and preceding symposium circulated the room during this task, giving assistance where needed.

The research topics with the highest votes were presented to the audience and discussed briefly to clarify possible research methodologies suitable to answer the research question.

Data analysis

One of the researchers (TCoH) summed the votes for each research topic that was brought up. According to the voting scores, topics were ranked, producing a list of the participants' 'priority research topics'. Identical scores were given the same ranking number; research topics with higher number of individual votes were listed higher.[23] Finally, research topics were grouped into research themes.

Results

Baseline characteristics of workshop attendees

A total of 66 researchers and GPs from 29 nationalities from 6 different continents attended our workshop. (Table 1) Fourteen participants (21%) only attended the introduction of the workshop.

Most important topics in MUS research

The nominal group technique resulted in a total of 29 research topics. In Table 2, the ranking of these research topics is shown.

Almost half (48%) of the audience agreed that the most important problem is that MUS are not clearly defined. There is need for an unambiguous definition of MUS that can be applied globally. Without a clear definition, research outcomes and their meaning remain muddled and uncertain. As research about a definition is not possible, discussion and reaching consensus is the best way to solve this. A common consensus building method is the Delphi technique.[24] The purpose of this technique is to obtain consensus on the opinions of 'experts' through question rounds using structured questionnaires. The audience emphasized their preference of working as soon as possible on a definition of MUS.

Workshop participants chose as second most important research question 'what is the best strategy to recognize MUS in primary care?' Patients with MUS are functionally impaired and at risk for unnecessary diagnostic procedures and treatments. Recognition of patients with a high risk of persistent MUS is therefore of paramount importance. However, in the absence of a clear definition, studying this research question is difficult.

Thirdly, research about effective interventions in patients with MUS, especially self-management and patient empowerment, should have priority according to 40% of the workshop audience. We should take the cultural background and patient perspectives into

Table 1. Nationality of participants in the workshop on MUS

Asia	**21**
- Korea	1
- Singapore	3
- India	1
- Malaysia	5
- China	3
- Hong Kong	3
- Philippines	2
- Mongolia	1
- Taiwan	1
- Thailand	1
Middle East	**6**
- Dubai	1
- Qatar	2
- Israel	1
- Bahrain	1
- Saudi Arabia	1
Australia	**3**
Africa	**7**
- South Africa	2
- Nigeria	5
Europe	**22**
- UK	**4**
- The Netherlands	10
- Slovenia	1
- Turkye	2
- Italy	2
- Spain	1
- Ireland	1
- Portugal	1
North America	**4**
- US	2
- Canada	2
South America	**1**
- Brazil	1
Total number of attendees	**66**
Total number of participants	**52**[a]

[a] Fourteen participants only attended the introductory presentation of the workshop.

Table 2. Ranking of research topics in the field of MUS

Rank	Research topic	Total of individual votes	Total score
1	What is the definition of MUS?	25	91
2	What is the best strategy / tool to recognize MUS in primary care and when do I have to stop searching for a rare disease?	18	69
3	What is the effect of self-management and/or patient empowerment in patients with MUS?	21	58
4	How does the patient explain his symptoms and what do patients do with the diagnosis MUS?	14	44
5	What do doctors in primary care do wrong and right in the consultations with patients with MUS?	13	44
6	What is the aetiology of MUS?	11	40
7	Which interventions in primary care are effective in patients with MUS?	11	35
8	What is the natural course of MUS in primary care?	13	34
9	Are there cultural differences in diagnosing MUS?	9	34
10	At what point should we intervene to prevent patients becoming chronic presenters of MUS?	13	32
11	What is the effect of (the teaching of) communication skills of the doctor in consultations with patients with MUS?	12	30
12	What is the effect of a comprehensive assessment of global aspects (spiritual, psychosocial, contextual, family view) of patients with MUS?	9	27
13	What alternative/complementary medicine approaches/interventions are effective in patients with MUS?	9	23
14	What is the value of the use of metaphors by patients and doctors in consultations on MUS	6	20
15	What method can we use to measure muscle tension in rest?	9	16
16	What is the influence of MUS on the QOL of the patient and the doctor?	5	14
17	Is there a relation between MUS prevalence/presentation and different health care systems (payment of care by insurance or by patients themselves)?	6	13
18	What is the efficacy of a stepped-care model in the therapy of patients with MUS?	4	12
19	What is the influence of the role of the GP (gatekeeper - non-gatekeeper) on the outcome of MUS?	3	12
20	Is there a need for a better categorisation of patients with MUS?	5	10
21	What is the effect of a community and/or family approach in therapies on the outcome of MUS?	4	10
22	What and to what extent should GPs explain to patients about MUS?	4	9
23	What is the relation between MUS and 'heartsink' patients?	4	8
24	What is the difference between MUS and hysteria?	2	8
25	How do patients develop chronic MUS?	2	7
26	What is the influence of the dimensions of the biopsychosocial model on the development of chronic MUS?	1	4
27	What is the difference between MUS and hypochondria?	1	3
28	What are the views of patients on MUS?	1	2
29	Are patients with MUS satisfied with the consultation?	0	0

Table 3. Themes of research topics in the field of MUS

Research topic	Rank	Total score[a]
Therapeutic options		**195**
- What is the effect of self-management and/or patient empowerment in patients with MUS?	3	
- Which interventions in primary care are effective in patients with MUS?	7	
- What is the effect of (the teaching of) communication skills of the doctor in consultations with patients with MUS?	11	
- What is the effect of a comprehensive assessment of global aspects (spiritual, psychosocial, contextual, family view) of patients with MUS?	12	
- What alternative/complementary medicine approaches/interventions are effective in patients with MUS?	13	
- What is the efficacy of a stepped-care model in the therapy of patients with MUS?	18	
- What is the effect of a community and/or family approach in therapies on the outcome of MUS?	21	
Problems in consultations: The doctors' perspective		**174**
- What is the best strategy / tool to recognize MUS in primary care and when do I have to stop searching for a rare disease?	2	
- What do doctors in primary care do wrong and right in the consultations with patients with MUS?	5	
- At what point should we intervene to prevent patients becoming chronic presenters of MUS?	10	
- What is the value of the use of metaphors by patients and doctors in consultations on MUS?	14	
- What and to what extent should GPs explain to patients about MUS?	22	
Definition		**120**
- What is the definition of MUS?	1	
- Is there a need for a better categorisation of patients with MUS?	20	
- What is the relation between MUS and 'heartsink' patients?	23	
- What is the difference between MUS and hysteria?	24	
- What is the difference between MUS and hypochondria?	27	
Aetiology and course		**115**
- What is the etiology of MUS?	6	
- What is the natural course of MUS in primary care?	8	
- What method can we use to measure muscle tension in rest?	15	
- What is the influence of MUS on the QOL of the patient and the doctor?	16	
- How do patients develop chronic MUS?	25	
- What is the influence of the dimensions of the biopsychosocial model on the development of chronic MUS?	26	
Cultural influences		**59**
- Are there cultural differences in diagnosing MUS?	9	
- Is there a relation between MUS prevalence/presentation and different health care systems (payment of care by insurance or by patients themselves)?	17	
- What is the influence of the role of the GP (gatekeeper – non-gatekeeper) on the outcome of MUS?	19	
Patient perspective		**46**
- How does the patient explain his symptoms and what do patients do with the diagnosis MUS?	4	
- What are the views of patients on MUS?	28	
- Are patients with MUS satisfied with the consultation?	29	

[a] Sum of the scores of each theme.

account. Probably, more than one intervention is needed to help patients with MUS. Mixed method qualitative research with doctors and patients may be the key to develop an effective intervention for these patients.

Finally, we discussed what doctors in primary care might do wrong, and what they do well, in consultations with patients with MUS. For instance, medical investigations can reassure some patients but can worsen symptoms in other patients. It would be helpful to study predictors, in order to decide which strategy will help the individual patient with MUS most.

Themes in MUS research

In Table 3, we grouped the research topic into six themes: (i) definition of MUS, (ii) aetiology and prognosis of MUS, (iii) problems in the consultation with patients with MUS, (iv) therapeutic options for patients with MUS, (v) patient perspectives and (vi) cultural differences. As appears from this table, the most important research themes worth studying were 'therapeutic options for patients with MUS' and 'problems in the consultation with patients with MUS' (total score 195 and 174, respectively). Research about the definition of MUS got a total score of 120 and research about aetiology and course 115. The research themes 'cultural differences' and 'the patient perspective' were regarded as less important (total score of 59 and 46, respectively).

Discussion

Bringing together experts of MUS research and discussing the state of the art in MUS research internationally was of great value: we could share the global perspective on MUS and discuss ideas about future developments on MUS research. Although cultural differences will play a role in the classification of patients with MUS, and doctors' perspectives and management of patients with MUS, we did not find many prominent cultural differences during our symposium and workshop. Doctors from all over the world face more or less the same problems in consultations with patients with MUS. The high attendance rate and the number of nationalities in our symposium and workshop reflect that MUS are present all over the world and attract great attention of GPs and researchers.

The problem in MUS that was recognized most was the indistinct and ambiguous definition of MUS. There is need for an unambiguous definition of MUS because without a clear definition, research outcomes and their meaning remain muddled and unclear. This need is even more urgent because in the next versions of ICD-11 and DSM-V the end of the somatoform disorder category is being considered.[25,26] Difficulties arising in defining MUS are the connection between MUS and prevalent mental health disorders such as depression and anxiety and functional somatic syndromes such as fibromyalgia and Irritable Bowel Syndrome.[27,28] Therefore, integrated research in primary care and specialized services is needed.

Important research themes worth studying appeared to be 'therapeutic options in patients with MUS' and 'problems within the consultation with patients with MUS'. Recent research already focus on these themes.[20,29,30] There is need for more patient-oriented research, e.g. qualitative research and mixed method research, to learn more about the needs of patients with MUS.[31] Together with studying the course of MUS, this will provide GPs with instructions and advices for the best therapeutic approaches towards these patients.[12,13] At best, the results of these studies should be combined with the development and investigation of suitable and effective interventions in these patients.

As far as we know, this is the first time that a research agenda in MUS has been defined by GPs from all over the world. The participation of 29 nationalities from 6 different continents extends the validity of the findings to cultures other than the Western culture. The strength of the nominal group technique as a method for defining the research agenda is its structured, detailed discussion. However, it may also lead to unrepresentative, unreliable judgements and contextual differences, such as differences in national health care systems. Furthermore, the lack of anonymity can make participants feel reserved to articulate their opinion.

We observed an interesting discrepancy: results of research as presented in the symposium focussed on miscommunication between doctors and patients as reason for persisting MUS, whereas in the workshop participants discussed MUS as if it were a real disease with its own aetiology and course. Patients often want to discuss psychosocial problems but doctors do not do so and choose a somatic pathway. This difference between patient's and doctor's perspective is regarded as a cause of persistence of symptoms. Consequently, these symptoms have a 'course' and the workshop participants were particularly interested in that course. Only some MUS patients turn out to have a "real" somatic or psychological disease during follow-up. Turning all persistent unexplained symptoms into a disease is medicalising and not very helpful for patients, because they actually want to be heard and discuss their psychosocial problems. If doctors would facilitate this, more MUS might resolve.

Another interesting finding of our workshop was the fact that research on patient views was not considered as priority. Knowledge of patients' expectations is necessary to answer patients needs in consultations of MUS and enhances patients' satisfaction, adherence and health.[32] The discussion on MUS as a real disease and the fact that only doctors participated in defining the research agenda might explain the lack of priority of patient views. Moreover, it shows that patient participation in defining a research agenda is needed in the future.

It is clear from the literature, that more research on MUS in primary care is needed in order to provide an improvement of the management of these patients and a better understanding of the problems we face during consultations with patients with MUS. Moreover, it is important to exchange ideas and coordinate research efforts on MUS in primary care at an international level, in order to prevent redundancy. Future WONCA conferences, as well as other primary care oriented conferences, offer excellent opportunities for this purpose.

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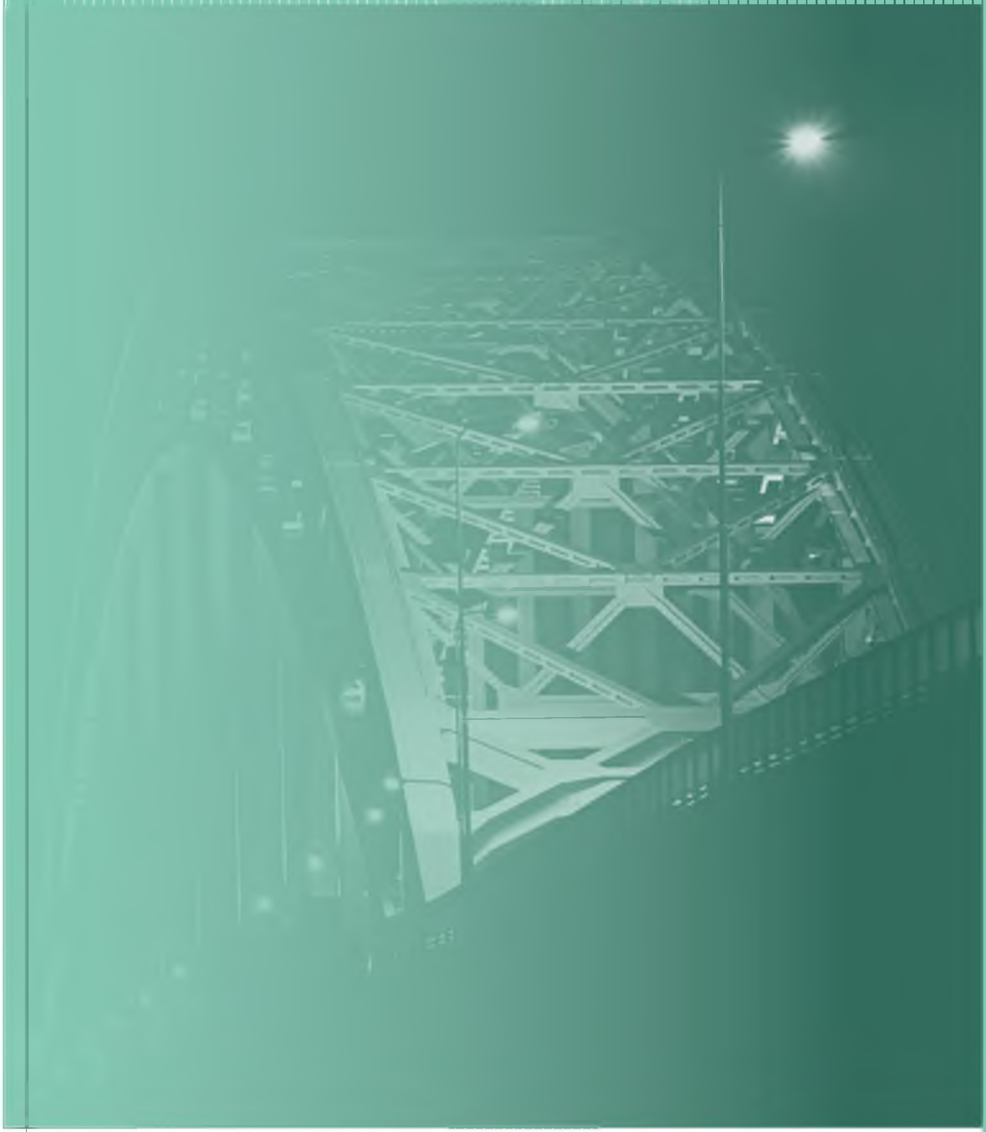
CHAPTER
11

GENERAL DISCUSSION



Tim olde Hartman

The aim of this thesis was to explore the care GPs deliver and the care patients expect when visiting the GP with persistent medically unexplained symptoms (MUS) in order to guide new feasible and effective intervention strategies for these patients in primary care. In this final chapter, the results of the previous chapters will be brought together for discussion and will be related to existing literature. Subsequently, methodological considerations will be discussed. Finally, implications and recommendations for intervention strategies, future research and primary care development will be considered.

Summary of main findings

Analyzing the results of the studies of this thesis, we could distinguish three main themes: course and prognosis of MUS, the mismatch in the doctor-patient communication, and the importance of the doctor-patient relationship. Before we will outline and summarize these themes, we have to notice that doctor-patient communication and doctor-patient relationship are strongly interconnected as they highly influence each other. However, we assume that the distinction between those two is worthwhile as doctor-patient communication focuses more on interaction styles within a single consultation whereas doctor-patient relationship covers a more longitudinal concept which is shaped throughout multiple consultations. For example, the doctor-patient relationship is build over multiple consultations whereas the doctor-patient communication is shaped in each single consultation.

Course and prognosis of MUS

In the first two studies **(chapter 2 and 3)** we examined the course and prognosis of MUS. Simultaneously we searched for patient characteristics which can help GPs in daily practice to identify patients who are at risk for persistent MUS.

By using the CMR registration project, we were able to determine patient characteristics during the ten years before the diagnosis of chronic functional somatic symptoms (CFSS). The CMR registration project has two unique features: (1) longitudinal registration which makes it possible to follow the development towards persistent MUS and (2) a defined code for MUS and persistent MUS (CMR classifies persistent MUS as chronic functional somatic symptoms (CFSS)). Patients with CFSS appeared to consult their GP twice as much, to use much more somatic and psychotropic medication, to have more psychiatric morbidity and to be more often referred to mental health workers then patients without CFSS. Furthermore, they are more likely to present symptoms in two or more body systems and to present a higher number and greater diversity of symptoms to the GP then patients without CFSS.

To identify relevant prognostic factors in patients presenting persistent MUS we also performed a systematic literature review on prognostic factors. The review showed that patients with MUS and abridged somatisation (the abridged definition of somatisation which requires the presence of four symptoms in males and six symptoms in females) had improvement rates of 50% or more. However, 10-30% of patients with MUS symptoms deteriorated during the 6 to 15 months follow-up period. Furthermore, the majority of patients with hypochondriasis did not recover during follow-up. We found that the seriousness of the condition at baseline (i.e. number of symptoms and severity of symptoms) might be of prognostic significance. We found no evidence to support the influence of psychiatric comorbidity and personality traits on the course of hypochondriasis.

Conclusion

The number, diversity and severity of symptoms are important characteristics in the course and prognosis of MUS. Establishing the number of presented symptoms and the seriousness of the condition in patients with MUS might help GPs to be aware of an increased risk of persistence of MUS.

The mismatch in the doctor-patient communication

Analysis of video consultation of patients with persistent MUS **(chapter 7)** indicated that patients present many symptoms in a rather unstructured way and GPs hardly used structuring techniques such as agenda setting and summarizing. Furthermore patients with persistent MUS got much opportunity to tell their story, but the reasons for encounter, their beliefs and concerns were not discussed in a structured manner. The extensive explanation of the origin of the symptoms given by the GP often did not take patients' beliefs and concerns into account. In the interview study, patients indicate **(chapter 4)** that they appreciate a non-judgmental and attentive listening style, a thorough exploration and a comprehensive explanation of this symptoms. So, based on our qualitative analysis of the doctor-patient communication, we conclude that there seems to be a mismatch between what patients with persistent MUS want and what GPs deliver to them. This mismatch emerges most obvious in the domains: 'exploration' and 'explanation'.

Exploration

MUS experts **(chapter 8)** state that GPs should listen carefully to the patient and question them extensively about the symptoms, the consequences of the symptoms for daily life and the meaning of the symptoms for the patient. Furthermore they state that it is important to ask the patient about his cognitions, emotions, fears, and concerns regarding the symptoms. So, this concurs with what patients want as assessed in our interview study **(chapter 4)**. Contrary to these recommendations, GPs seldom explore patients' ideas, concerns and expectations in a structured manner given their actual behaviour during the encounters with patients with persistent MUS **(chapter 7)**. We showed in a case report that recognizing and exploring cues (non-explicit remarks) **(chapter 6)** can be a helpful tool for this exploration.

Explanation

GPs are aware of the importance of explaining the symptoms to the patient **(chapter 5)**. This is in line with the opinions of MUS experts **(chapter 8)**, who stress the importance of providing patients with a tangible explanation for his/her symptoms. However, GPs indicate that they face difficulties to explain the nature of the presented unexplained symptoms **(chapter 5)**. They state that they use three different approaches to explain the symptoms to the patients; normalization of symptoms, telling patients that there is no disease, and using metaphors. The findings of **chapter 9** offered a number of templates around which GPs could further develop and specify their explanations. Some of these models aimed at a physical level of explanation, such as the endocrine dysregulation theory. Other models aimed at a more psychological level of explanation, such as the somatosensory amplification theory. And some models combined a physical and psychological levels of explanation, such as the sensitization theory. Finally, one model combined predisposing, precipitating and perpetuating factors such as somatic causes, perception, and illness behaviour.

The relevance of more focused explanation came from **chapter 7** in which we found that GPs' extensive explanations of the origin of the symptoms often did not take patients' beliefs and concerns into accounts. This might be a result of the limited exploration of these issues. However, experts indicated the importance of linking the physical complaints with patients' cognitions and illness beliefs. Therefore, understanding of patients' ideas, concerns and expectations together with knowledge of the different explanatory models might help GPs to improve their communication with patients with persistent MUS.

The importance of the doctor-patient relationship

According to patients with persistent MUS **(chapter 4)**, a good and effective doctor-patient relationship depends on: (1) a GP with an open mind who uses a non-judgmental communication style; (2) a skillful and competent GP who is supportive and who offers a comprehensible explanation for the symptoms; (3) easy access to their 'own' GP who takes time to explore and to discuss the patients' symptoms and/or problems; (4) being taken seriously; and (5) engagement as partners in the consultation in a way they can make their own choices based on the provided information. This means that patients value a personal continuing GP-patient relationship built on physician attitude, medical competence, availability and shared authority. These findings are in line with the opinions of GPs **(chapter 5)**, who indicate the usefulness and importance of a good doctor-patient relationship in encounters with patients with persistent MUS. They are also in line with the opinions of MUS experts **(chapter 8)**, who state that the doctor-patient relationship is an important tool in order to create a safe therapeutic environment for these patients. However, although patients, GPs and MUS experts recognize the importance of the doctor-patient relationship, GPs indicate **(chapter 5)** that they face difficulties in preserving the doctor-patient relationship during these encounters.

Although GPs realize the usefulness and importance of a good doctor-patient relation in encounters with patients with persistent MUS, they face difficulties in putting this into practice. This is in particular the case when explaining what is wrong or relieving of symptoms is problematic. GPs try to overcome this by securing the relationship with some sort of ritual care (e.g. regular physical examination, regular doctor visits). Some GPs do this wholeheartedly, while others feel rather ambivalent with this ritual **(chapter 5)**. Hereby, GPs carefully balance between the maintenance of the doctor-patient relationship, their inability in explaining MUS, and avoiding damage from drug therapy and interventions.

Comparison with the existing literature

Course and prognosis

The 'ecology of medical care' model explains that most people do not contact professional medical care for most of their complaints.[1] Only a small minority of people contact a health care professional when they feel ill.[1,2] Twenty to thirty percent of the encounters of this small minority concern symptoms that can not be explained by any physical disease.[3-5] However, most of these episodes of illness remain restricted to one doctor-patient contact.[6] Therefore we can conclude that most of the symptoms are transient and disappear spontaneously in the course of time or patients find a way to cope with their symptoms.[7] This is in line with our findings. We have found that the prognosis of MUS in primary and secondary care is more favourable than expected, as the majority of the patients with MUS improve.

Creed and Barsky performed a systematic review of the epidemiology of somatisation disorder and hypochondriasis to examine the characteristics and associated features of these disorders.[8] Although they did not systematically search and study prognostic factors, they concluded that the number of longitudinal studies providing data concerning natural history and predictors of outcome is inadequate. Furthermore, they raised the hypothesis that abridged somatisation, somatisation disorder and hypochondriasis are closely connected to anxiety and depressive disorders.[8] However, we could not confirm their hypothesis as in our review comorbid anxiety and depression did not have prognostic value. Personality traits, including neuroticism and alexithymia, have been demonstrated to be associated with MUS and hypochondriasis.[9,10] However, our review did not find evidence for their prognostic value.

Our studies pointed out that the number, diversity and severity of symptoms are important characteristics associated with the course of MUS. Patients with persistent MUS are more likely to report higher number of symptoms, greater diversity of symptoms or more severe symptoms. This is in line with the finding that patients with distinct functional somatic syndromes often have symptoms overlapping with other functional somatic syndromes indicating a high number and great diversity of symptoms in these patients.[11] Furthermore, patients with distinct functional somatic syndromes often present symptoms in many body systems.[12] Due to the substantial overlap between the individual functional somatic syndromes the existence of distinct specific somatic syndromes should be questioned. Taking the fundamental unity of those syndromes into account, the GP, is the most appropriate practitioner to diagnose and treat these patients as (1) they use a broad-based and generic approach; and (2) they try to prevent iatrogenic harm of unnecessary interventions by using this approach.

In contrast to the limited numbers and considerable heterogeneity of studies focusing on the prognosis of patients with undifferentiated MUS, distinct functional somatic syndromes are studied more meticulously. Cairns and Hotopf for example, found 28 papers describing the clinical follow-up of patients after the diagnosis chronic fatigue syndrome (CFS).[13] The full recovery rate in these patients was only 5% and only 40% of the patients improved. Lower fatigue severity at baseline, a sense of control over symptoms, and not attributing illness to a physical cause were associated with a good outcome. Their findings of the prognostic significance of the fatigue severity at baseline are in line with our findings.

The mismatch in the doctor-patient communication

An important task for GPs is listening to patients,[14,15] as patients seek understanding for their symptoms[16-19] and want to feel that the GP accepts the symptoms as real.[20] To achieve this, consultations skills, including exploration and explanation, are needed. However, we found that exploration was only practiced to a limited extend by GPs when encountering patients with persistent MUS. This is in line with the findings of others. According to Stewart et al. GPs do not explore the reason for encounter and the worries of the patients in approximately half of the consultations.[21] Furthermore, in about 20% of all consultations the patient has an biomedical or psychosocial agenda which is not addressed in the consultation.[22] An important tool to explore patients' expectations and concerns is responding to patients' cues.[23,24] Cues are non-explicit remarks that can enclose a special meaning and point towards ideas, worries or expectations. Discussing these cues enable better understanding of the reason for encounter, cognitions and emotions.[25,26] However, Salmon et al found that during the initial presentation of MUS, GPs' engagement with explicit cues about emotional or social problems was limited.[27] This is in line with our findings, as we found that GPs' exploration of patients' reason for encounter, beliefs and concerns is also limited in persistent MUS consultations. Not exploring patients' beliefs and expectations has several disadvantages. Firstly, patients might think that the GP is not listening carefully, does not want to understand the meaning of the symptoms or is not interested in the patient.[28] Secondly, contrary to what is expected, these consultations will last significantly longer. For example, Levinson found that in a majority of the consultations (79%), GPs missed the opportunity to react to the cues given in the consultation resulting in a significant longer duration of these consultations.[29] Thirdly, not building a shared understanding regarding the symptoms hampers effective explanation.[20,30-32] This might explain why GPs provide reassurance without a clear explanation of the symptoms[27]; they simply lack information. For example, normalization of symptoms and telling patients that they don't have a disease without providing tangible mechanisms grounded in patients' concerns is not effective and results in more demand for health-care.[32] Finally, patient's satisfaction, adherence and health are reduced.[21,33,34] However, what we also have found in the video consultation study was that GPs gave patients with persistent MUS ample opportunity to tell their story and this is exactly what these patients want. In the interview study we found that patients want their GP to be mindfully present, to listen to their story and to give them room to tell their story. Churchill described the importance of taking time for the patients and demonstrating that there is space for their story.[35] Furthermore, giving patients time for their story is one of the important elements of working in a patient-centred way.[36] However, we think that GPs' patient-centred working skills can further be improved by a more focused communication, including enhancing GPs' exploratory behaviour.

Prior research has indicated that doctor-patient communication is associated with patient health outcomes.[37-39] Studies in cancer care revealed that words can be therapeutic because when patients feel understood and reassured resulting in positive psychosocial health outcomes.[40,41] Thomas showed the importance of being positive during consultations with patients with MUS.[42] He studied patients receiving a positive consultation (firm diagnosis and confidence of a good prognosis) or a non-positive consultation (no firm assurance and unsure about the prognosis). Two weeks after the consultation, patients who received the positive consultation were significantly more satisfied. Furthermore, 64% of those patients got better, compared with 39% of those who received a negative consultation. The role of patient- provider communication was also studied in patients with osteoarthritis of the knee receiving traditional Chinese acupuncture and sham acupuncture.[43] Patient receiving a 'high expectations' communication style (acupuncturist conveyed positive expectations of improvement about his/her treatment) experienced statistically significant improvement in pain (effect size 0.25) and satisfaction (effect size 0.22) compared with patients receiving a 'neutral expectations' communication style (acupuncturist conveyed neutral expectations of improvement about his/her treatment). Cals et al. evaluated the effect of doctor-patient communication on antibiotic prescribing for lower respiratory tract infection and on patient recovery.[44] They found that GPs who received a training in enhanced communication skills prescribed significantly fewer antibiotics than those not trained in these communication skills (27% vs 54%, P<0.01).

The three studies described above show that positive doctor-patient communication is essential for the effectiveness of treatments in health problems in primary care. One of the proposed pathways through which doctor-patient communication can contribute to improved health is a shared understanding of illness.[45] Although reaching a shared understanding in patients with persistent MUS is often difficult[15] and treatment for these patients are not easily available, a positive doctor-patient communication is an important tool in the management of these patients.

The importance of the doctor-patient relationship

GPs in our study recognized the importance of establishing an ongoing and effective doctor-patient relationship with patients with persistent MUS. Furthermore, they took responsibility to build and maintain such a relationship. Taking this responsibility fits well in the values of family medicine and general practice.[30,46] Important indicators of the quality of the doctor-patient relationship include mutual trust among GPs and patients, personal continuity, empathy, compassion and the patient's perception of feeling respected and being cared for.[45,47]

It is known from the literature that the quality of the doctor-patient relationship affects health outcomes by promoting patient's emotional well-being, enhancing continuity of care, patient satisfaction, and patient commitment to treatment plans.[48-50] Furthermore, in psychotherapy outcome research therapist-client relationship factors have been estimated to contribute 30% of patient outcomes.[51] Di Blasi et al. reviewed the empirical evidence of the therapeutic effect of the doctor-patient relationship in medicine.[52] They identified 25 trials studying the effects of the patient-practitioner relationship on patients' health status. GPs who attempted to form a warm and friendly relationship with their patients were found to be more effective than practitioners who kept their consultations impersonal and formal.

Some studies have established the association between doctor-patient relationship and patient health outcomes in mental health. Van Os et al. examined the effect of depression treatment, empathy and support, and their interaction on patient outcomes for depression in primary.[53] They found that only the combination of depression treatment according to the guidelines and empathy and support resulted in an effective treatment of depression in primary care. Kaptchuk et al. evaluated the effect of the doctor-patient relationship in patients with irritable bowel syndrome (IBS).[54] They found that augmented interaction (a warm, empathetic, and confident patient-practitioner relationship) was more effective than limited interaction (patients reporting adequate symptom relieve: respectively, 61% vs 53%). Smith et al. studied the effect of explicitly establishing a good patient-provider relationship in high-utilizing patients with MUS in primary care.[55,56] They found an absolute 14% improvement (48% improved vs 34% improved) on the mental composite score of the SF-36 among those receiving the intervention, compared with those who did not. Furthermore, depression and disability scores, as well as satisfaction improved.

The three studies described above as well as the review of Di Blasi et al.[52] show that the doctor-patient relationship is an important and potent therapeutic component which should not be ignored. Furthermore, the role of the doctor-patient relationship should be explicitly addressed in the treatment of (persistent) MUS in primary care. However, the doctor-patient relationship should not be a primary goal of the consultation as, according to Chew-Graham, this may result in GPs feelings of powerlessness to resolve the symptoms and problems presented and collusion with patients' illness behaviour that maintain chronic incapacity.[57] Therefore, the doctor-patient relationship should always be a means in stead of an arm to provide effective care.

Methodological considerations

The quantitative methods

To identify patient characteristics, course and prognosis of patients with persistent MUS we performed (1) an observational study with a comparison control group using CMR longitudinal data **(chapter 2)** and (2) a systematic review of the literature regarding prognostic factors in patients presenting persistent MUS **(chapter 3)**. Here we will describe the strengths and limitations of the research methods we used in these two chapters.

Strengths

The CMR registration project is, as far as we are aware, the only morbidity registration system with a structural possibility to classify patients with chronic functional somatic symptoms (i.e. persistent MUS). The validity of morbidity registration is enhanced by monthly meetings of all GPs involved to discuss classification problems, to monitor the application of diagnostic criteria and to discuss coding problems of hypothetical case histories.[58] Longitudinal research in the field of MUS is mostly done by using questionnaires.[59,60] However, questionnaires have been shown to produce unreliable results due to recall bias. For example, the Psychological Problems in General Health Care (PPGHC) study examined the stability of somatisation disorder and somatisation symptoms and concluded that somatisation disorder showed considerable instability during 12 months.[61] In our review and our CMR registration study, MUS and persistent MUS was diagnosed by the GPs or investigators themselves in patients experiencing a certain threshold of relevance of symptoms as they seek medical care for them meaning that the diagnosis of (persistent) MUS was not related to the recall of 'lifetime' symptoms.[62]

Our systematic review is the first which systematically searched for studies on prognostic factors in patients with persistent MUS. We used an extensive search strategy to identify relevant studies and we independently assessed the quality of included studies with a validated checklist. Furthermore, we searched all relevant databases, in- and excluded studies with two researchers independently and did not use language restrictions. Although the heterogeneity of included studies was high, we were able to perform a best-evidence synthesis to summarize the value of the different prognostic indicators taking methodological quality into account.

Limitations

In the CMR registration there might be an interdoctor variation in the diagnosis chronic functional somatic symptoms as diagnosing these patients will always remain an interpretation of the symptoms in which GPs will use their knowledge of the context of the specific patients.[21,58,63] One might argue that the higher frequency of GP visits, referrals and diagnostic test was to be expected in advance, as the diagnosis chronic functional somatic symptoms can only be made after repeated consultations for physical symptoms that remain medically unexplained after adequate examination.

There were some limitations regarding the quality of included studies of the review. Firstly, the numbers of participants enrolled in the cohorts is limited. Secondly, only a minority of the included studies presented sufficient data on the duration of symptoms at baseline. Thirdly, a detailed description of treatments during follow-up was absent although we assume that all patients received some kind of treatment during the follow-up.

The qualitative methods

Most chapters of this thesis used qualitative research methods to explore the care GPs deliver to patients with persistent MUS and to explore the care patients with persistent MUS expect. The qualitative research methods used in this thesis are: qualitative interviews **(chapter 4)**, focus group discussions **(chapter 5)**, consultation analysis **(chapter 7)**, qualitative analysis of the literature **(chapter 8 and 9)** and nominal group technique **(chapter 10)**. Here we will discuss the strengths and limitations of the research methods used in these different chapters.

Strengths

The qualitative methods used in the interview study, the focus group study, the two literature studies and nominal group technique has been recommended as the best method to explore and clarify opinions.[64] For researching the doctor-patient communication during consultations, video-recording has been recommended as the best method.[31] In all qualitative studies we used a cyclical and interactive way of collecting and analyzing data in order to progressively refine the focus and explore the data in depth.[65] Furthermore, we transcribed the data verbatim, entered the full texts into Altas.ti, and coded and re-organized data by two researchers independently using the principles of constant comparative analysis.[66] The constant comparative method constitute the core of qualitative analysis in qualitative research.[67]

Our patient interview study is the first study on opinions and expectations of patients with documented, persistent MUS and reflects opinions of patients who have had recent and relatively frequent encounters with the healthcare system and their GP. Therefore, their opinions are built on a broad and recent experience with medical services. By using a purposive sampling strategy and a triangulation strategy of asking independent GPs to judge our results we were able to capture and verify the variety of opinions present in the population of GPs. As the focus group study examined GPs' perceptions and not actual behaviour, we performed a video consultation study. As neither the GP nor the patient was aware of the subject of study and as there is little evidence that video-recording influences the behaviour of either the GPs or patients, the Hawthorne effect (the effect that participants improve or modify their behaviour in response of the fact that they are being studied) in our video consultation study was limited. The qualitative analysis of the literature regarding explanatory models of MUS and expert opinions on the management of MUS gave a broad overview of this field. We used an extensive and systematic search strategy to identify relevant papers and we included and coded the full text papers by two independent researchers to minimize biases.

In this thesis we integrated more qualitative research techniques for data collection and analysis, also referred to as mixed method research. Therefore, we were able to benefit from combining the strengths of the different research methods resulting in a more holistic, valid and robust exploration and description of the difficulties arising when GPs encounter patients with persistent MUS.[68-70] Furthermore, this helped us to gain more insight into the complex phenomenon of persistent MUS in primary care.

Limitations

Qualitative research methods are not suited to quantify a certain phenomenon or variable, or to generalize results from a small sample to a larger population. Therefore the results of our interview study, focus group study, video consultation study, two literature studies and nominal group technique should not be interpreted with a quantitative view. The methods allow the in-depth exploration of the lived experience or actual behaviour of individuals or a small group of people and put these into the context of the day-to-day reality. Generalization towards GPs or towards patients with persistent MUS in general is only possible after quantitative research in random samples of patients and/or GPs, based on hypotheses resulting from qualitative analyses.

Patient interviews about opinions on the doctor-patient relationship give only a snapshot in time, whereas this relationship is built over time and over many consultations. Patients' perspectives might have changed over time as a response to their experiences with the GP and/or the health care system. The same counts for the video consultation study, as each new consultation carries over memories of previous ones which might have influenced the videotaped consultation. These longitudinal influences on the doctor-patient communication and relationship were not captured in these studies. Furthermore, although participation bias in our video consultations study was limited (response rate 89%), in our interview study this kind of bias might have influenced our findings (response rate 39%) as patients who are satisfied with their GP are possibly more inclined to participate in the interview study. In contrast to one-to-one interviews, during focus group discussions group dynamics might result in the articulation of group norms or consensus in stead of provoking individual contrasting opinions. By using a purposive sampling strategy and a skilled moderator during the focus group discussions, we tried to provoke and capture the full variety of GPs' opinions. In our two literature studies we did a qualitative analysis of the literature. Although such an analysis is not as objective as a meta-analysis, we were able to summarize the range of explanatory models and important elements in the interventions described in the scientific literature nowadays. This summary and overview might help to guide future intervention strategies for patients with persistent MUS. However, we are not able to draw conclusions on the degree of evidence and the effectiveness of the explanatory models and important elements in the interventions for patients with persistent MUS as we did not study the evidence regarding these models and elements.

Recommendations for clinical practice

The worries of GPs regarding the development of persistent MUS are mostly not justified as the majority of these patients generally have a favourable prognosis. Establishing the number of somatic symptoms and the seriousness of the condition during the first consultations might help GPs to value the risk of persistence and may guide GPs whether to offer only reassurance about the favourable prognosis or a more intensive approach. Furthermore, it seems important to communicate this favourable and positive prognosis with the patient during the MUS consultation.

To improve the outcomes for patients with persistent MUS, GPs have to focus on the consultation itself, the presented symptoms, and the words they use during these consultations. GPs have to improve their communication towards a more clear and focused communication style by (1) the exploration of patients' needs, beliefs, concerns and expectations, (2) the exploration of the reason for encounter, (3) active listening and noticing cues, (4) agenda setting and summarizing the information obtained during the consultation, (5) providing tangible, positively formulated explanations incorporating patients' beliefs and concerns. Furthermore, a positive and continuous doctor-patient relationship is necessary for a consultation to be effective. Therefore, GPs have to focus on (a) taking patients seriously, (b) showing empathy, (c) understanding the context of an individual patient with his or her illness and (d) mutual trust. Regularly scheduled appointments can serve as a tool for this.

Creating a safe therapeutic environment shaped by a clear and focused communication and a therapeutic doctor-patient relationship might be key to improve the health care delivered to patients with persistent MUS.

Recommendation for future research

Although this thesis makes an important contribution to a more patient-oriented way of studying the possibilities for improving the health care delivered to patients with persistent MUS, more research is needed to provide GPs with instructions and advices for the best individualized therapeutic approaches towards these patients. As our research agenda in MUS, defined by GPs from all over the world, indicated, more research is needed on (1) aetiology and course, (2) problems in the consultation, and (3) therapeutic options.

Regarding aetiology and course, there is need for more well-conducted prospective cohort studies with a reasonable number of patients (>200 patients), in which assessment of treatments during follow-up and inclusion of patients at a similar point in the course of there disease are important topics. These studies should incorporate the quality of the communication and the doctor-patient relationship, in order to reveal which nonspecific aspects of the clinical encounter affects help-seeking behaviour in these patients, and the outcome of the consultations with these patients.

Regarding the problems in the consultation, there is need for further examination of the efficacy of improving the exploration of patients' ideas, concerns and expectation and the structure of the persistent MUS consultation. Furthermore, our findings should be confirmed in a larger, prospective qualitative study that could track doctor-patient communication regarding MUS over time. Such studies should reveal whether patients wish and accept a more in-depth exploration and structure of the consultation.

Regarding therapeutic options for patients with persistent MUS, mixed method methodology may reveal effective methods of explaining the symptoms. Furthermore, this research should study the effects of the (improvement of the) quality of the GP-patient relationship on symptoms, impairment, disability and satisfaction in these patients.

Recommendations for medical education

Medical students and GP residents have to realize that they have powerful tools to effectively manage and treat patients with persistent MUS. These students and residents have to be convinced of this powerful tools, for example by presenting them the available evidence, about the importance of a positive consultation and communication style and the therapeutic effects of the doctor-patient relationship. Furthermore, we have to teach them about patients' preferences and experts opinions regarding the management of persistent MUS in primary care.

Given that ideas in doctors' minds play such an important role, it is imperative that something should be done about the way doctors are trained. In particular, we need to pay special attention to the side-effects of the monolithic position of the biomedical approach. We need to teach our students that the biomedical model is only one of many perspectives. We also need to focus on the fact that doctors usually view symptoms as expressions of disease and on the fact that doctors consider symptoms with a clear underlying pathology to be more real than symptoms that cannot be objectively measured.

Renewed attention for the consultation behaviour is desirable. This extra attention means paying attention to symptoms, expectations, ideas and feelings regarding presented symptoms in order to attain a good understanding of the symptoms, finding common ground and building mutual trust. Generic elements such as attentive listening, explaining symptoms to patients and ways to reassure patients needs further attention

Conclusion

The majority of patients with MUS have a favourable prognosis. Only a minority ends up in a chronic condition. Establishing the number, diversity and severity of the symptoms during the first consultations might help GPs to value the risk of persistence of the symptoms.

There seems to be a mismatch between what patients with persistent MUS want and what GPs deliver to them. Patients want a tangible and positive formulated explanation for their symptoms, whereas GPs face difficulties in doing so. Furthermore, patients with persistent MUS want GPs to pay attention to their ideas, concerns and expectations, whereas GPs actual behaviour during the encounters with these patients show the opposite (i.e. limited exploratory behaviour).

Patients with persistent MUS and GPs agree on the importance of a good doctor-patient relationship. They both value a personal continuing GP-patient relationship built upon physician attitude, medical competence, availability and shared authority. However, at the same time GPs indicate that they often face difficulties in preserving the doctor-patient relationship during encounters with these patients. They try to balance between the maintenance of the relationship and the prevention of unintended consequences of their interventions.

Based on these findings we conclude that there seems to be room for improving the care and outcomes for patients with persistent MUS. For example by establishing a more clear and focused communication style in which patients' ideas, concerns and expectations are explored and taken into account. Active listening, noticing cues, agenda setting, summarizing the information obtained during the consultation, exploring the reason for encounter, and giving tangible and positively formulated explanations can help with that. Furthermore, a positive doctor-patient relationship in which patients are taken seriously, GPs show empathy and try to understanding the context of an individual patient, and both patients and GPs work on mutual trust, might contribute to improve the care and outcomes for these patients.

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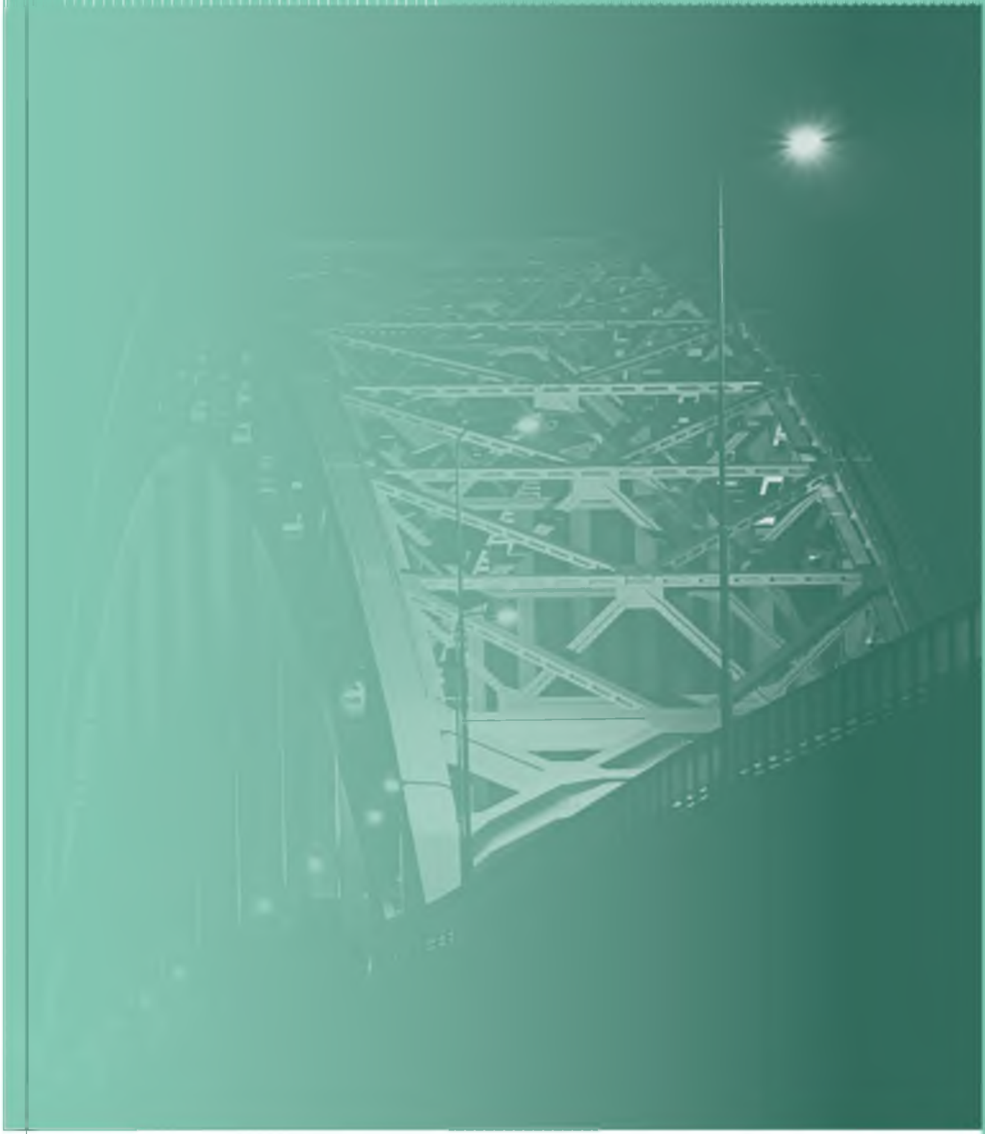
SUMMARY / SAMENVATTING



Tim olde Hartman

DOCTOR
PATIENT
CONSULTATION

Summary

This thesis explored (1) the care patients with persistent MUS expect when visiting the GP, (2) the care GPs deliver to patients with persistent MUS and (3) the way GPs experience the care they deliver to their patients with persistent MUS. Furthermore, we present starting points for intervention strategies for patients with persistent MUS in primary care in order to improve the care for these, often functionally impaired, patients.

Chapter 1. General introduction.

In this chapter the rationale, aims and outline of this thesis are described.

Medically unexplained symptoms (MUS) are common and most of these symptoms are transient. However, a minority of the patients will keep visiting the doctor because of MUS (i.e. persistent MUS). This minority represent a major problem in health care as they are responsible for high, often unnecessary, health care costs. Furthermore, these patients suffer from their symptoms and are functionally impaired.

We explored the knowledge regarding the problems arising when GPs meet patients with persistent MUS in daily practice. Furthermore, we indicated knowledge gaps regarding the care GPs deliver and the care patients expect when encountering with persistent MUS.

Patients with persistent MUS often feel the skepticism of their doctors and have the feeling that the GPs don't take them serious. They are often dissatisfied with the medical care they receive. Doctors experience patients with persistent MUS as difficult to manage. They tend to believe that (1) these patients fail to see the connection between physical symptoms and psychological distress, and that (2) there is incongruence between the presentation of MUS and the actual burden. Furthermore, they feel pressurized to offer somatic interventions. Consultations between GPs and patients with MUS seems to be less patient-centred. In these consultations patients with MUS did not pressurize for somatic interventions, but desired more emotional support.

Most research regarding the problematic interaction between GPs and patients with MUS did not specifically focus on patients with persistent MUS. During the chronic condition in which patients persistently present MUS to their GP, GPs have to find a way to manage these patients in order to improve patients' health.

The patient

Chapter 2. Chronic functional somatic symptoms: a single syndrome?

In this chapter we aimed to identify distinctive features in patients with persistent MUS by studying the Continuous Morbidity Registration database. This is the only morbidity registration system in the world with a structural possibility to classify patients with persistent MUS (CMR classifies persistent MUS as chronic functional somatic symptoms (CFSS)). We included 182 patients with persistent MUS and 182 matched controls from this registration system and collected data on comorbidity, referrals, diagnostic tests, hospital admissions, medication use and number of visits to the GP over a period of 10 years prior to the diagnosis. We found that during the 10 years prior to the diagnosis persistent MUS patients presented MUS in at least two body systems and used more somatic and psychotropic drugs compared to controls. Furthermore, they visited the GP twice as much and had more psychiatric morbidity, and were referred more often to mental health workers and somatic specialists. The number of patients undergoing diagnostic test was also higher. Hospital admission rates were equal. We concluded that patients with persistent MUS have a great diversity of unexplained symptoms, use more somatic and psychotropic drugs in the years before diagnosis and show high rates of referrals and psychiatric morbidity compared with controls.

Chapter 3. Medically unexplained symptoms, somatisation disorder and hypochondriasis: course and prognosis. A systematic review.

In chapter 3 we studied the course of MUS, somatisation disorder and hypochondriasis and related prognostic factors. We searched the relevant databases of the medical literature to select studies focusing on patients with MUS, somatisation and hypochondriasis and to assess prognostic factors. We performed a best-evidence synthesis for the interpretation of the results. We could include six studies on MUS, six studies on hypochondriasis and one study on abridged somatisation and found that approximately 50 to 75% of the patients with MUS improved, whereas 10-30% of patients with MUS deteriorate. In patients with hypochondriasis recovery rates varied between one third to 50%. In studies on MUS and hypochondriasis we found some evidence that the number of somatic symptoms and the seriousness of the condition at baseline influenced course and prognosis. Comorbid anxiety and depression did not seem to predict the course of MUS, abridged somatisation or hypochondriasis. We concluded that due to the limited number of studies and their high heterogeneity, there is a lack of empirical evidence to identify relevant prognostic factors in patients with persistent MUS. However, it seems that a more serious condition at baseline is associated with a worse outcome.

Chapter 4. The doctor-patient relationship from the perspective of patients with persistent medically unexplained symptoms. An interview study.

In Chapter 4 we studied the perspectives of patients with persistent MUS regarding their expectations of GPs' health care. By conducting qualitative semi-structured interviews with 17 patients with persistent MUS we were able to reveal opinions of patients with a documented long history of presenting MUS. All patients stressed the importance of a personal continuing doctor-patient relationship. They indicate that such a relationship is built on physician attitude, medical competence, availability and shared authority. Patients want to be taken serious in a non-judgmental open communication style. They appreciate a thorough exploration and a comprehensible explanation of the symptoms. Furthermore, they want a competent. GP who is easily accessible and who engages them as partners in the consultation and decision making process. We conclude that patients with persistent MUS appreciate a patient-centred communication and orientation to a form of care in which personal continuity and continuity of the relationship are important elements.

The doctor

Chapter 5. Explanation and relations. How do general practitioners deal with patients with persistent medically unexplained symptoms: a focus group study.

This chapter explored GPs' perceptions about explaining MUS to patients with persistent MUS and about how relationships with these patient evolve over time in daily practice. We used a qualitative focus group approach in which twenty-two GPs within five focus groups were interviewed. GPs indicated that they recognise the importance of an adequate explanation of the diagnosis of MUS but often feel incapable of being able to explain it clearly to their patients. Therefore, they try to reassure patients in non-specific ways, for example by telling patients that there is no disease, by using metaphors and by normalizing the symptoms. When patients keep returning to the GP with MUS, GPs stress the importance of maintaining the doctor-patient relationship. GPs describe three different models to do this: mutual alliance characterized by ritual care (e.g. regular physical examination, regular doctor visits) with approval of the patient and the doctor, ambivalent alliance characterized by ritual care without approval of the doctor and non-alliance characterized by cutting of all reasons for encounter in which symptoms are not of somatic origin. We conclude that GPs experience difficulties in explaining the symptoms and that they meticulously balance between maintaining a good doctor-patient relationship and the prevention of unintended consequences of unnecessary interventions.

The consultation

Chapter 6. 'Well doctor, it is all about how life is lived': cues as a tool in the medical consultation.

In this chapter we described a patient with unexplained palpitations during vacuuming. During one of the consultations she provided an important psychosocial cue which changed my perspective on her palpitations, resulting in a deeper understanding of her symptoms. We experienced that this non-explicit remark that enclosed a special meaning served as a tool to clarify the patient's agenda. This resulted in a mutual understanding about the symptoms, enhanced the therapeutic relationship and improved the illness outcome. We concluded that noticing cues in the medical consultation helps the doctor to understand the patient's real worries and gives a better understanding of the patient's perspective.

Chapter 7. How patients and family physicians communicate about persistent medically unexplained symptoms. A qualitative study of video-recorded consultations.

This chapter describes an exploratory, qualitative analysis of 20 videotaped consultations between GPs and patients with persistent MUS in order to study doctor-patient interaction styles and to study on which stages of the consultation patients and doctors focus within the available time. We found that patients presented many symptoms in a rather unstructured way. However, GPs hardly use structuring techniques such as agenda setting and summarizing. Patients with persistent MUS got much opportunity to tell their story, but the reasons for encounter, their beliefs and concerns were not discussed in a structured manner. Most of the time patients themselves had to initiated discussion of their ideas, concerns and expectations. However, GPs' extensive explanations of the origin of the symptoms often did not take patients' beliefs and concerns into account. We concluded that due to patients' multiple symptom presentation and the absence of GPs' structuring techniques, consultations of patients with persistent MUS in this study proceed rather unfocused. However, patients got ample opportunity to tell their story. Furthermore, patients with persistent MUS might benefit from structured consultations focused on the exploration of the reason for encounter.

Starting points for improving management

Chapter 8. Experts' opinions on the management of medically unexplained symptoms in primary care. A qualitative analysis of narrative reviews and scientific editorials.

This chapter reviewed important and effective elements in the treatment of patients with MUS in primary care according to experts in MUS research in order to explore effective and acceptable strategies to manage these functionally impaired patients. We performed a systematic search of narrative reviews and scientific editorials in the most important databases of the medical literature and triangulated our findings by conducting a focus group interview with MUS experts. We included 7 scientific editorials and 23 narrative reviews. According to MUS experts the most important elements in the treatment of MUS are: creating a safe therapeutic environment; generic interventions (such as motivational interviewing, giving tangible explanations, reassurance and regularly scheduled appointments); and specific interventions (such as cognitive approaches and pharmacotherapy). However, in contrast to most specific interventions, experts rarely describe the effects of generic interventions, doctor-patient communication and relationship quantitatively in their scientific editorials and narrative reviews. MUS experts indicate that a multi-component approach in which these three important elements are combined are most helpful for patients with MUS. We concluded that, in contrast to most specific interventions, opinions of MUS experts regarding generic interventions and creating a safe therapeutic relationship seem to be more based on theory and experience, than on quantitative research. Studies showing the effectiveness of these elements in the management of MUS in primary care are still scarce. Research as well as medical practice should focus more on these non-specific aspects of the medical consultation.

Chapter 9. Explanatory models of medically unexplained symptoms: a qualitative analysis of the literature.

This chapter provides an overview of explanatory models for MUS in order to provide GPs with explanatory models known from the scientific literature that may be useful in daily medical practice. Therefore we performed a systematic search of reviews in the relevant databases of the scientific literature and analyzed the data qualitatively according to the principles of constant comparative analysis to identify specific explanatory models. We distinguished nine specific explanatory models of MUS in the literature: somatosensory amplification, sensitization, sensitivity, immune system sensitization, endocrine dysregulation, signal filter model, illness behaviour model, autonomous nervous system dysfunction and abnormal proprioception. The nine different explanatory models focus on different domains, including: somatic causes, perception, illness behaviour and predisposition. We also found one meta-model, which incorporates these four domains: the cognitive behavioural therapy model. We concluded that although GPs often face difficulties when providing explanations to patients with MUS, there are multiple explanatory models in the scientific literature that may be of use in daily medical practice.

Chapter 10. Medically unexplained symptoms in family medicine: defining a research agenda. Proceedings from WONCA 2007.

In this chapter we detected knowledge gaps in MUS and established priorities in MUS research by bringing together experts of MUS research and discussing the state of the art in MUS research internationally. By using a nominal group technique, we generated research topics and themes, set priorities in MUS research and proposed a research agenda. Participants' (66 researchers and GPs from 29 nationalities) most important research topics were 'formulating a broadly accepted definition of MUS', 'finding a strategy to recognize MUS better and at an earlier stage', 'studying the value of self-management and empowerment in patients with MUS' and 'finding predictors to decide which strategy will best help the individual patient with MUS'. Priorities in research themes of MUS were: (i) therapeutic options for patients with MUS and (ii) problems in consultations with these patients. We concluded that more research on MUS in primary care is needed to improve the consultations with and management of these patients. Furthermore, internationally primary care conferences seemed to be excellent for exchanging ideas and formulating central issues of research.

Chapter 11. General discussion.

This final chapter considers the results described in this thesis together with some methodological issues, and ends with implications and recommendations for intervention strategies and suggestions for further research and general practice.

We conclude that (1) the majority of the patients with MUS have a favourable prognosis and that establishing the number, diversity and severity of the symptoms during the first consultation might help GPs to value the risk of persistence of the symptoms, (2) there seems to be a mismatch between what patients with persistent MUS want and what they receive from their GP, and (3) patients and GPs agree on the importance of a good doctor-patient relationship. Therefore, exploring and taking into account patients' ideas, concerns and expectations, together with building a positive doctor-patient relationship, might contribute to improve the care and outcomes for patients with persistent MUS.

The most important recommendations for clinical practice are that we have to focus on the consultation itself, the presented symptoms, and the words we use during these consultations. Furthermore, we have to focus on creating a safe therapeutic environment shaped by a clear and focused communication and a therapeutic doctor-patient relationship.

The most important recommendations for future research are that future MUS research has to take the doctor-patient communication and relationship into account in order to reveal which nonspecific aspects of the clinical encounter affects the outcome of these consultations.

Furthermore, effective methods of explaining symptoms to patients with persistent MUS have to be tested.

Medical students and GP residents have to be taught that a positive consultations and communication style and a good doctor-patient relationship are therapeutic in itself and therefore a powerful and important tool in order to effectively manage and treat patients with persistent MUS.

Samenvatting

Dit proefschrift beschrijft (1) de zorg die patiënten met persisterende somatisch onvoldoende verklaarde lichamelijke klachten (SOLK) verwachten van hun huisarts, (2) de zorg die huisartsen leveren aan patiënten met persisterende SOLK en (3) de ervaringen van huisartsen met de zorg voor patiënten met persisterende SOLK. Vervolgens presenteren we uitgangspunten voor de behandeling en begeleiding van deze patiënten in de huisartsenpraktijk met als doel de zorg te verbeteren voor deze patiënten die vaak erg beperkt zijn in hun functioneren.

Hoofdstuk 1. Algemene inleiding.

In dit hoofdstuk beschrijven we de achtergronden, doelen en opbouw van dit proefschrift. SOLK komen veel voor en meestal zijn deze klachten van voorbijgaande aard. Echter, een minderheid van de patiënten blijft de huisarts bezoeken met SOLK omdat de klachten aanhouden (i.e. persisterende SOLK). Deze minderheid vormt een groot probleem voor de gezondheidszorg omdat ze verantwoordelijk is voor hoge, vaak onnodige, gezondheidszorgkosten. Bovendien hebben deze patiënten vaak veel last van hun klachten en ondervinden ze beperkingen in hun dagelijks leven ten gevolge van de klachten. Consulten met deze patiënten in de huisartspraktijk verlopen vaak niet zonder problemen.

We beschrijven de reeds bestaande kennis over de problemen die ontstaan wanneer huisartsen en patiënten met persisterende SOLK elkaar ontmoeten in de dagelijkse praktijk. Daarnaast geven we aan waar meer kennis nodig is over de zorg die huisartsen leveren en de zorg die patiënten verwachten op het moment dat ze het spreekuur bezoeken met persisterende SOLK.

Patiënten met persisterende SOLK voelen vaak de scepsis van de verschillende dokters en hebben vaak het gevoel dat de huisarts de klachten niet serieus neemt. Ze zijn vaak ontevreden over de medische zorg die ze krijgen. Dokters vinden patiënten met persisterende SOLK vaak moeilijk te behandelen. Ze hebben de neiging te denken dat (1) deze patiënten de link tussen lichamelijke klachten en psychosociale problemen niet zien, en dat (2) de klachtpresentatie en de daadwerkelijke ziektelast niet met elkaar in overeenstemming zijn. Daarnaast voelen huisartsen zich vaak onder druk gezet om somatische interventies aan te bieden. Consulten tussen huisartsen en patiënten met SOLK lijken weinig patiëntgericht. Bovendien blijkt dat patiënten met SOLK helemaal geen druk uitoefenen voor het krijgen van somatische interventies, maar dat ze vooral emotionele ondersteuning van hun huisarts willen.

Het meeste onderzoek naar de moeizame interactie tussen huisartsen en patiënten met SOLK is niet specifiek gericht op patiënten met persisterende SOLK. Wanneer de klachten chronisch worden en patiënten het spreekuur van de huisarts blijven bezoeken met SOLK, moet de huisarts een manier vinden om deze patiënten te behandelen en te begeleiden om zo de gezondheid van deze patiënten te verbeteren.

De patiënt

Hoofdstuk 2. Chronisch functionele klachten: één syndroom?

Het doel van dit hoofdstuk is om, vanuit de Continue Morbiditeit Registratie (CMR), onderscheidende kenmerken van patiënten met persisterende SOLK te identificeren. De CMR database is het enige registratie systeem in de wereld dat structureel patiënten met persisterende SOLK in de huisartspraktijk registreert. De CMR classificeert persisterende SOLK als chronisch nerveus-functionele klachten. We includeerden 182 patiënten met persisterende SOLK en 182 controlepatiënten vanuit de CMR database en we verzamelden data over een periode van 10 jaar voorafgaand aan de diagnose betreffende comorbiditeit, verwijzingen, diagnostische testen, ziekenhuisopnames, medicatiegebruik en het aantal bezoeken aan de huisarts. We vonden dat gedurende de 10 jaar voor de diagnose persisterende SOLK patiënten onverklaarde klachten presenteerden in minstens twee delen van het lichaam en dat deze patiënten meer somatische medicatie en meer psychofarmaca gebruikten dan de patiënten zonder persisterende SOLK. Bovendien bezochten patiënten met persisterende SOLK hun huisarts twee keer zo vaak, hadden ze meer psychiatrische comorbiditeit en werden ze vaker verwezen naar de geestelijke gezondheidszorg en somatisch medisch specialisten. Het aantal patiënten dat een diagnostische test onderging was ook hoger. Ziekenhuisopnames waren gelijk. We concluderen dat patiënten met persisterende SOLK een grote diversiteit aan onverklaarde klachten presenteren, meer somatische medicatie en psychofarmaca gebruiken in de jaren voor de diagnose, dat ze vaker verwezen worden en meer psychiatrische aandoeningen hebben in vergelijking met controlepatiënten.

Hoofdstuk 3. Somatische onvoldoende verklaarde lichamelijke klachten, somatisatiestoornis en hypochondrie: beloop en prognose. Een systematische review.

In hoofdstuk 3 beschrijven we het beloop van SOLK, somatisatiestoornis en hypochondrie en hun prognostische factoren. We doorzochten de medische literatuur en selecteerden onderzoeken gericht op SOLK, somatisatie en hypochondrie en hun prognostische factoren. We voerden een best-evidence synthese uit om de resultaten te interpreteren. We konden zes onderzoeken naar SOLK, zes onderzoeken naar hypochondrie en één onderzoek naar de verkorte vorm van somatisatiestoornis (i.e. abridged somatisation) includeren en vonden dat ongeveer 50 tot 70% van de patiënten met SOLK verbeterden en 10 tot 30% van de patiënten met

SOLK verslechterden. Bij patiënten met hypochondrie vonden we herstelpercentages van 30 tot 50%. In de onderzoeken naar SOLK en hypochondrie vonden we enig bewijs dat het aantal klachten en de ernst van de klachten op baseline het beloop en de prognose beïnvloeden. Comorbiditeit van angst of depressie lijkt het beloop van hypochondrie niet te beïnvloeden. Onderzoeken naar de invloed van angst of depressie op het beloop van SOLK laten tegenstrijdige resultaten zien. We concluderen dat door het beperkt aantal onderzoeken en de grote heterogeniteit van die onderzoeken het onvoldoende mogelijk is om relevante prognostische factoren bij patiënten met persisterende SOLK te identificeren. Wel lijkt de ernst van de klachten op baseline geassocieerd met een slechter beloop.

Hoofdstuk 4. De arts-patiënt relatie volgens patiënten met persisterende SOLK. Een interview studie.

In hoofdstuk 4 onderzoeken we de verwachtingen die patiënten met persisterende SOLK hebben van de huisartsenzorg. We namen bij 17 patiënten met persisterende SOLK een semi-gestructureerd interview af om de meningen en opvattingen van deze patiënten boven tafel te krijgen. Alle patiënten benadrukten het belang van een persoonlijke langdurige arts-patiënt relatie. Patiënten gaven aan dat een dergelijke relatie gebaseerd is op de attitude, de medische competentie, de beschikbaarheid van de dokter en het hebben van een gedeelde verantwoordelijkheid. Patiënten willen serieus genomen worden door een huisarts die een niet-veroordelende open manier van communiceren gebruikt. Ze waarderen een nauwgezette uitdieping van hun klachten en een begrijpelijke uitleg. Bovendien willen ze een competente huisarts die gemakkelijk toegankelijk is en die hen betrekt als partners in het consult en het besluitvormingsproces. We concluderen dat patiënten met persisterende SOLK een patiëntgerichte communicatie waarderen waarbij ze een persoonlijke langdurige arts-patiënt relatie erg belangrijk vinden.

De dokter

Hoofdstuk 5. Uitleg en relatie. Hoe gaan huisartsen om met patiënten met persisterende SOLK: een focusgroep onderzoek.

Dit hoofdstuk belicht de meningen en opvattingen van huisartsen over de uitleg van de klachten aan patiënten met persisterende SOLK en hoe de relatie met deze patiënten zich ontwikkeld in de loop van de tijd. We gebruikten een kwalitatieve focusgroep benadering waarin we 22 huisartsen verdeeld over 5 focusgroepen interviewden. Huisartsen geven aan dat ze het belang van een goede uitleg van de persisterende SOLK onderkennen, maar dat ze vaak grote moeite hebben met het geven van een dergelijke uitleg. Daarom proberen ze patiënten gerust te stellen op een niet-specifieke manier, bijvoorbeeld door te zeggen dat er geen ziekte is, door metaforen te gebruiken en door de klachten te normaliseren. Wanneer patiënten terug blijven komen bij de huisarts met persisterende SOLK, is het volgens huisartsen belang om de arts-patiënt relatie goed in stand te houden. Ze beschrijven drie verschillende modellen om dit te doen: (1) onderlinge alliantie gekenmerkt door zorgrituelen (zoals regelmatig lichamelijk onderzoek, regelmatig contact met de huisarts) met goedkeuring van zowel de patiënt als de dokter; (2) ambivalente alliantie gekenmerkt door zorgrituelen zonder dat de dokter deze eigenlijk goedkeurt; (3) non-alliantie gekenmerkt door het afkappen van alle consulten waarin klachten worden gepresenteerd zonder duidelijk lichamelijke oorzaak. We concluderen dat huisartsen moeilijkheden ervaren bij het uitleggen van de klachten en dat ze nauwgezet balanceren tussen het in stand houden van de arts-patiënt relatie en het voorkomen van de negatieve consequenties van onnodige interventies.

Het consult

Hoofdstuk 6. 'Ach dokter, het leven wordt nu eenmaal zo geleefd': het gebruik van hints in het consult.

In dit hoofdstuk beschrijven we een patiënt met onverklaarde hartkloppingen tijdens het stofzuigen. Tijdens een van de consulten gaf ze een belangrijke psychosociale hint die mijn perspectief op haar hartkloppingen veranderde en resulteerde in een beter begrip van haar klachten. Ik ondervond dat deze niet expliciete maar toch bijzondere opmerking behulpzaam was voor het verder uitdiepen van het verhaal van de patiënt. Dit resulteerde in een gezamenlijk begrip van de klachten, versterkte de therapeutische relatie en bevorderde het ziektebeloop. We concluderen dat het oppikken van hints in het consult de dokter kan helpen om de mening, opvattingen, verwachtingen en angst en zo het perspectief van de patiënt beter te begrijpen.

Hoofdstuk 7. Hoe praten patiënten en huisartsen over persisterende SOLK? Een kwalitatief onderzoek van video consulten.

Dit hoofdstuk beschrijft een explorerende, kwalitatieve analyse van 20 consulten opgenomen op videotape tussen huisartsen en patiënten met persisterende SOLK. Hierin belichten we de arts-patiënt communicatie en onderzoeken we op welke onderdelen in het consult patiënten en dokters zich richten. Patiënten blijken meerdere klachten te presenteren kris-kras door het consult heen. Huisartsen gebruiken echter nauwelijks gesprekstechnieken, zoals het bepalen van de agenda en samenvatten, om deze consulten te structureren. Patiënten met persisterende SOLK krijgen ruim de mogelijkheid om hun verhaal te vertellen. De reden van komst, hun ideeën en angsten worden echter niet structureel besproken. Meestal initiëren patiënten zelf een discussie over hun eigen ideeën, angsten en verwachtingen. De uitgebreide uitleg die huisartsen geven over de persisterende SOLK houdt vaak geen rekening met deze ideeën en angsten. We concluderen dat doordat patiënten meerdere klachten presenteren en huisartsen deze consulten niet actief structureren, consulten met patiënten met persisterende SOLK weinig gericht zijn op het structureel uitdiepen van de klachten, ideeën, opvattingen en verwachtingen van patiënten. Wel krijgen patiënten ruim de mogelijkheid om hun verhaal te vertellen. Patiënten met persisterende SOLK hebben waarschijnlijk baat bij gestructureerde consulten gericht op het uitdiepen van de reden van komst.

Uitgangspunten voor het verbeteren van de behandeling en begeleiding

Hoofdstuk 8. Meningen van experts over de behandeling en begeleiding van patiënten met SOLK in de huisartspraktijk. Een kwalitatieve analyse van reviews en editorials.

In dit hoofdstuk gaan we op zoek naar belangrijke en effectieve elementen in de behandeling en begeleiding van patiënten met SOLK in de huisartspraktijk. Daarvoor inventariseerden we de meningen van wetenschappelijke experts op het gebied van SOLK. We voerden een systematische zoekactie uit naar reviews (overzichtsartikelen) en editorials (hoofdredactionelen) in de medische literatuur. We controleerden onze bevindingen aan de hand van een focus groep interview met wetenschappelijke experts in Nederland. We includeerden 7 editorials en 23 reviews. Volgens de wetenschappelijke experts zijn de meest belangrijke elementen in de behandeling en begeleiding van patiënten met SOLK: het creëren van een veilige therapeutische omgeving; generieke interventies (zoals motiverend interviewen, het geven van een duidelijke uitleg, geruststelling en regelmatige vervolgconsulten); en specifieke interventies (zoals cognitieve behandelingen en farmacotherapie). In tegenstelling tot de meeste specifieke interventies beschrijven wetenschappelijke experts bijna nooit de kwantitatieve effecten van de generieke interventies, de arts-patiënt communicatie en de arts-patiënt relatie. Wetenschappelijke experts geven aan dat een meervoudige benadering waarin deze drie belangrijke elementen worden gecombineerd het beste helpt bij patiënten met SOLK. We concluderen dat, in tegenstelling tot de meeste specifieke interventies, de meningen en opvattingen van wetenschappelijke experts over generieke interventies en het creëren van een veilige therapeutische omgeving meer gebaseerd lijken te zijn op theorie en persoonlijke ervaring, dan op kwantitatief onderzoek. Onderzoeken die de effectiviteit van deze elementen in de behandeling en begeleiding van SOLK onderzoeken zijn schaars. Onderzoek en de dagelijkse praktijk moet zich meer richten op deze non-specifieke aspecten van het consult.

Hoofdstuk 9. Verklaringsmodellen van SOLK: een kwalitatieve analyse van de literatuur.

Dit hoofdstuk geeft een overzicht van de verklaringsmodellen voor SOLK beschreven in de medisch wetenschappelijke literatuur. Kennis van deze verklaringsmodellen kan behulpzaam zijn voor de dagelijkse praktijk van huisartsen. We verrichtten een systematische zoekactie in de medische literatuur en analyseerden de data kwalitatief volgens de principes van de constant vergelijkende analyse. We konden negen specifieke verklaringsmodellen voor SOLK identificeren in de literatuur: somatosensorische amplificatie, sensitisatie, overgevoeligheid, gevoeligheid van het immuunsysteem, endocriene dysregulatie, signaal-filter theorie, ziektegedrag model, dysfunctie van het autonoom zenuwstelsel en abnormale proprioceptie. De negen verschillende verklaringsmodellen zijn gericht op verschillende domeinen, te weten: lichamelijke oorzaken, perceptie, ziektegedrag en aanleg. We vonden ook een overkoepelend model die de vier domeinen incorporeert: het cognitieve gedragsmodel. We concluderen dat er meerdere verklaringsmodellen in de medische literatuur te vinden zijn die eventueel gebruikt kunnen worden in de dagelijkse praktijk.

Hoofdstuk 10. SOLK in de huisartsgeneeskunde: het definiëren van een onderzoeksagenda. Uitkomsten van WONCA 2007.

In dit hoofdstuk gaan we op zoek naar ontbrekende kennis op het gebied van SOLK om vervolgens prioriteiten in het SOLK onderzoek aan te geven. Dit deden we door wetenschappelijk experts op het gebied van SOLK uit meerdere landen bijeen te brengen en de stand van zaken van het wetenschappelijk onderzoek naar SOLK te bespreken. We gebruikten een nominale groepstechniek om onderzoeksonderwerpen en thema's te genereren en prioriteiten voor het SOLK onderzoek aan te brengen. Hieruit werd een onderzoeksagenda opgebouwd. De deelnemers bestonden uit 66 wetenschappelijk onderzoekers en huisartsen uit 29 verschillende landen. Zij gaven aan dat 'het formuleren van een breed gedragen en geaccepteerde definitie van SOLK', 'het vinden van een manier om SOLK in een eerder stadium te herkennen', 'het bestuderen van de waarde van zelf-management en empowerment bij patiënten met SOLK' en 'het vinden van voorspellers om te bepalen welke behandeling het meest effectief is bij de individuele patiënt met SOLK' de belangrijkste onderzoeksonderwerpen zijn. Ze gaven de hoogste prioriteit aan de volgende onderzoeksthema's: (i) therapeutische opties voor patiënten met SOLK en (ii) problemen in het consult met deze patiënten. We concluderen dat er meer onderzoek naar SOLK in de huisartsgeneeskunde nodig is om de consulten met en de behandeling en begeleiding van deze patiënten te verbeteren. Bovendien zijn internationale huisartsgeneeskundige congressen een uitstekende mogelijkheid om ideeën uit te wisselen en onderzoeksthema's te bespreken.

Hoofdstuk 11. Algemene discussie.

Dit laatste hoofdstuk plaatst de resultaten van dit proefschrift in z'n perspectief, bespreekt enkele methodologische kwesties, geeft implicaties en aanbevelingen voor de behandeling en begeleiding van patiënten met persisterende SOLK en geeft suggesties voor verder onderzoek en de dagelijkse praktijk.

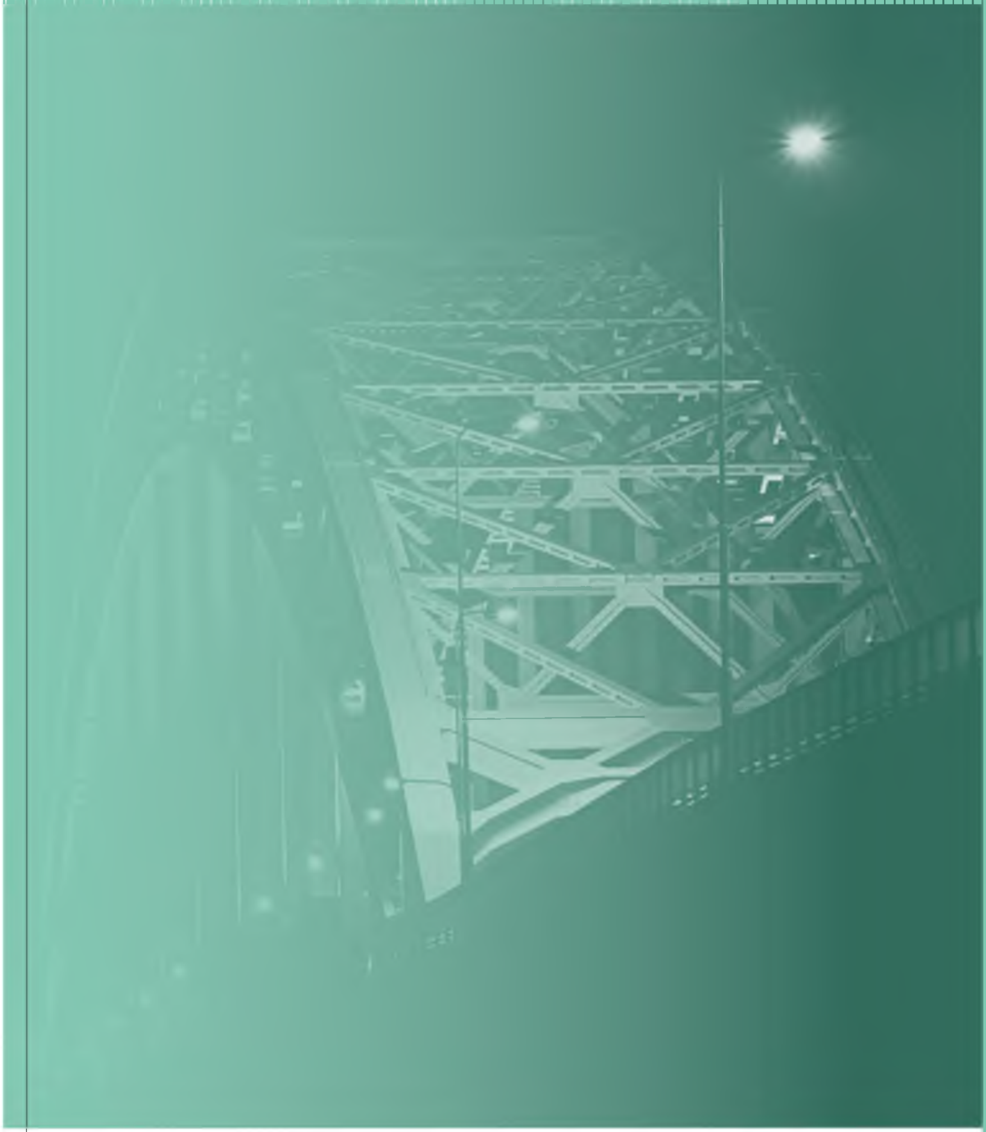
We concluderen dat (1) de meerderheid van de patiënten met SOLK een goede prognose heeft en dat het vaststellen van het aantal, de diversiteit en de ernst van de klachten tijden het eerste consult huisartsen kan helpen om het risico van persisteren van klachten in te schatten, (2) er een mismatch is tussen wat patiënten met persisterende SOLK willen en wat ze daadwerkelijk krijgen van hun huisarts, en (3) patiënten en huisartsen het eens zijn over het belang van een goede arts-patiënt relatie. Het uitdiepen van de ideeën, angsten en verwachtingen van de patiënt tezamen met het opbouwen van een positieve arts-patiënt relatie dragen bij aan het verbeteren van de zorg en haar uitkomsten voor patiënten met persisterende SOLK.

De belangrijkste aanbevelingen voor de dagelijkse praktijk zijn dat we ons moeten richten op de consultvoering, de klachten die de patiënt presenteert en de woorden die we gebruiken in deze consulten. Daarnaast moeten we ons richten op het creëren van een veilige therapeutische omgeving voortkomend uit een heldere en gerichte communicatie en een therapeutische arts-patiënt relatie.

De belangrijkste aanbevelingen voor toekomstig onderzoek is dat onderzoek naar SOLK zich moet richten op de communicatie en relatie tussen arts en patiënt, om zodoende te achterhalen welke non-specifieke aspecten van het consult de uitkomsten van deze consulten beïnvloeden. Daarnaast is er behoefte aan onderzoek naar welke manieren effectief zijn om klachten uit te leggen aan patiënten met persisterende SOLK.

We moeten medisch studenten en huisartsen in opleiding leren dat een positief consult, positieve communicatie en een goede arts-patiënt relatie therapeutisch in zichzelf zijn en daarmee een erg krachtig en belangrijk instrument om patiënten met persisterende SOLK te behandelen en te begeleiden.

DANKWOORD



Tim olde Hartman

DOCTOR
PATIENT
CONSULTATION

Dank!

In 2002 begon ik met dit project. In 2011 dan uiteindelijk de afronding. Sommigen van u zullen wel denken, had dat niet wat sneller gekund? Ach ja, vast wel, zou mijn antwoord zijn, maar er zijn ook zoveel andere leuke dingen: de huisartsopleiding, redactielidmaatschap bij Huisarts en Wetenschap, internationale congressen en samenwerkingsverbanden, meeschrijven aan nationale (multidisciplinaire) richtlijnen, een eigen praktijk starten en opbouwen en natuurlijk de persoonlijke patiëntenzorg. En veel belangrijker nog: trouwen met mijn jeugdliefde, een huis kopen, twee prachtige zoons krijgen en een leuk sociaal leven met familie en vrienden. Werk en privé in een continu samenspel, en dat maakt de afronding van dit proefschrift ook zo mooi en bijzonder.

Voor een succesvol samenspel is natuurlijk de inzet van meerdere personen nodig. Zij krijgen dan ook een welverdiende plek in mijn dankwoord. Zo'n dankwoord is altijd gevaarlijk omdat ik natuurlijk mensen zal vergeten te noemen. Daarom wil ik bij deze alle mensen bedanken die hun steentje hebben bijgedragen aan al het leuks dat ik in de afgelopen 10 jaar heb mogen doen.

Peter Lucassen. Peter, jij bent de geestelijk vader van dit project, maar bovenal een goede vriend. Je hebt het project bedacht en uitgeschreven. Onze eerste kennismaking was in 2002 toen ik me meldde bij de toenmalige afdeling Huisartsgeneeskunde (HAG) van het UMC St. Radboud. Ik wilde mijn huisartsopleiding graag combineren met een onderzoek, het liefst een klinische trial, over een duidelijk somatische aandoening. En jij kwam met een kwalitatief onderzoek naar onverklaarde lichamelijke klachten! Hoe vaag en pseudowetenschappelijk wil je het hebben, dacht ik toen nog. Toch besloot ik, hoewel jij dat niet verwacht had, toe te happen. Achteraf de beste keus die ik had kunnen maken. Samen hebben we de ontdekkingreis door kwalitatief onderzoek en onverklaarde klachten ondernomen. Het was prachtig! Mede door de frequente koffiediscussies werd mij steeds helderder waar het echt om gaat bij patiënten met onverklaarde klachten en daarmee in de huisartsgeneeskunde. De persoonlijke zorg die jij aan je patiënten geeft, geef je ook aan je promovendi. Je bent een groot voorbeeld voor mij.

Chris van Weel. Beste Chris, ik ben enorm trots dat je mijn promotor bent. Hoewel je veel in het buitenland was, heb je mijn vorderingen in onderzoeksland van dichtbij gadegeslagen. Je vaak subtiele commentaren op mijn stukken waren heerlijk om te verwerken. De stukken werden er zienderogen beter van. Altijd had je oog voor de kern van het huisartsenvak die door moest klinken in wat ik schreef. Ik heb er graag gebruik van gemaakt. Net als van je formidabele internationale netwerk. Daar kreeg ik pas echt goed zicht op tijdens de WONCA World en de NAPCRG annual meeting in 2004 in Orlando, Florida. De manier waarop je mij gevraagd en ongevraagd voorstelde aan en in contact bracht met al die internationaal bekende huisartsonderzoekers heb ik als een groot voorrecht ervaren. Ik raad alle jonge huisartsonderzoekers op de afdeling aan nu het nog kan, gretig gebruik te maken van je naamsbekendheid en enorme internationale netwerk.

Eric van Rijswijk. Eric, ik ben erg blij dat je mijn tweede copromotor bent. In de tijd dat ik begon op de afdeling was jij bezig met de afronding van je eigen proefschrift. Ondertussen heb je een geheel eigen onderzoekslijn palliatieve zorg opgebouwd. De enorme snelheid waarmee je dat doet vind ik bewonderenswaardig. Het samen stukken schrijven, medisch studenten begeleiden en natuurlijk de gezellige congresbezoeken maakten mijn promotietraject tot een feest.

Lieke Hassink-Franke. Onderzoeksmaatje! We begonnen ongeveer gelijktijdig met ons traject en zaten jaren op dezelfde kamer. In Orlando sliepen we zelfs op dezelfde kamer (schuld van Twanny!). Wat is het heerlijk om met iemand met zoveel positieve energie en enthousiasme te mogen samenwerken. We hebben onze ganggenoten vaak gestoord met ons harde gelach. En dat zal de komende tijd nog wel zo blijven, want we zullen de komende jaren samen de NMP-kar gaan trekken. Lieke, ik ben ontzettend blij dat je mijn paranimf wilt zijn.

Evelyn van Weel – Baumgarten. Beste Evelyn, samen met Peter, Eric en Lieke hebben we op menig internationaal congres ons GGZ gedachtegoed gepresenteerd en verder uitgewerkt. Presentaties, workshops, forums en posters, niets was je te gek. Veelal zat jij deze bijeenkomsten op een interactieve en levendige manier voor. Je onderwijsachtergrond kwam daar geweldig bij van pas. Door jouw Mental Health netwerk ontstonden er internationale samenwerkingsverbanden en hebben we de GGZ onderzoekslijn internationaal op de kaart kunnen zetten. De aanwezigheid van Chris Dowrick in mijn manuscriptcommissie heb ik daarmee vooral aan jou te danken.

Mark van der Wel. Mark, ik weet niet of je verwacht had dat ik je zo prominent in mijn dankwoord zou noemen. Toch doe ik het. De laatste jaren zijn we, tot mijn grote plezier, steeds intensiever gaan samenwerken. Je scherpzinnigheid, doorvragen en immer kritische blik dwingen me vaak tot nadenken. Onze trip naar Japan (we konden niet eens languit liggen in de business class), de hot tub op Vancouver Island (gelukkig hadden we bier en rendiersnacks) en de dance events tijdens de NAPCRG (wij sprongen het hoogst) waren hoogtepunten naast het harde werken wat we daar deden. Ik hoop, en ga er van uit, dat we nog veel meer zullen gaan samenwerken in de toekomst.

Huub Meijerink. Amice, vriend van het eerste uur, paranimf. We ontmoetten elkaar in de collegezaal in Rotterdam, september 1995, en zijn daarna samen opgetrokken in het medische wereldje. Van studie naar co-schap, van basisarts tot specialist, van Rotterdam naar Nijmegen, van doctorandus naar doctor. Hoe moet dat nou nu jij naar Heerenveen vertrekt om daar als orthopeed te gaan werken? Graag een huis met aanlegsteiger en een boot, dan komen wij wel logeren!

De huisartsgeneeskunde leer je niet vanuit boeken, leer je niet vanuit onderzoek, maar leer je eigenlijk alleen maar door het dagelijks in praktijk te brengen:

Huisartsopleiding Nijmegen (VOHA). Ben Bottema bedankt voor het faciliteren van mijn gecombineerde onderzoeks- en opleidingstraject. Rudy en Marijke Kleerekooper-Corsten, bedankt dat ik mijn eerste huisartsopleidingsjaar bij jullie in Elst heb mogen doen. De gezellige diensten zonder huisartsenpost (!) en met macaroni (van Leonard) in m'n haar zal ik niet snel vergeten. Jaap Schreuder, mijn derdejaars opleider in Malden, bedankt dat je mij een kijkje in de keuken van het lokale, regionale en landelijke bestuur van de huisartsgeneeskunde hebt gegeven. Destijds in de huisartsopleiding een ondergeschoven kindje, maar voor mij, dankzij jou, een enorme aanvulling op mijn opleiding.

Huisartspraktijk Mesker-Niesten. Jeanne en Pierre, bij jullie kwam ik als net afgestudeerde huisarts om als waarnemer ervaring op te doen. Eigenlijk heb ik bij jullie een vervolgopleiding in de kernwaarden van de huisartsgeneeskunde gekregen. Elke woensdag aan het eind van de middag een patiëntenbespreking met veel discussies over onverklaarde klachten, contextgeneeskunde en persoonlijke patiëntenzorg. Door de vier jaar die ik bij en met jullie heb gewerkt ben ik een andere en ik denk veel betere huisarts geworden. Ik vind jullie een groot voorbeeld voor de nieuwe generatie huisartsen.

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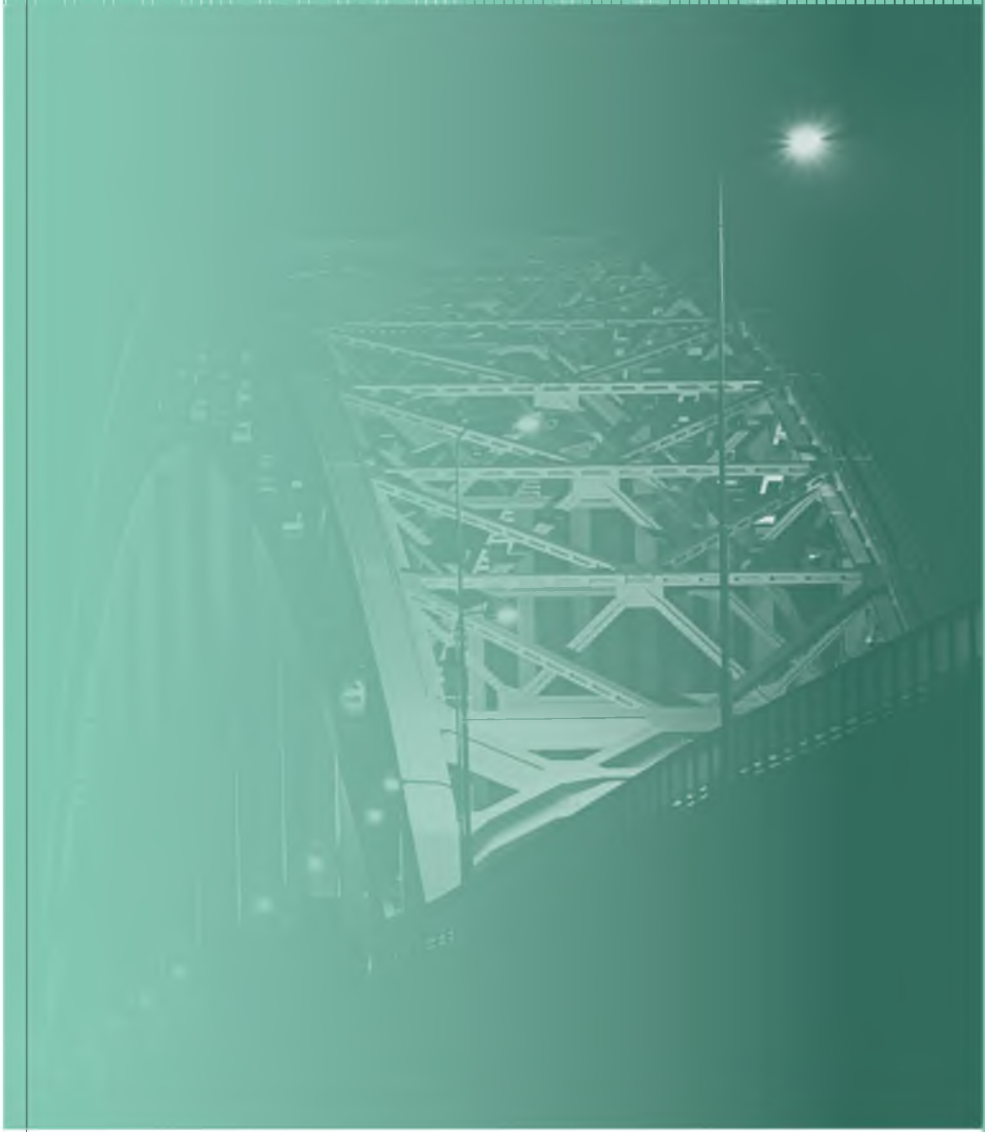

Tim olde Hartman

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Medically unexplained symptoms

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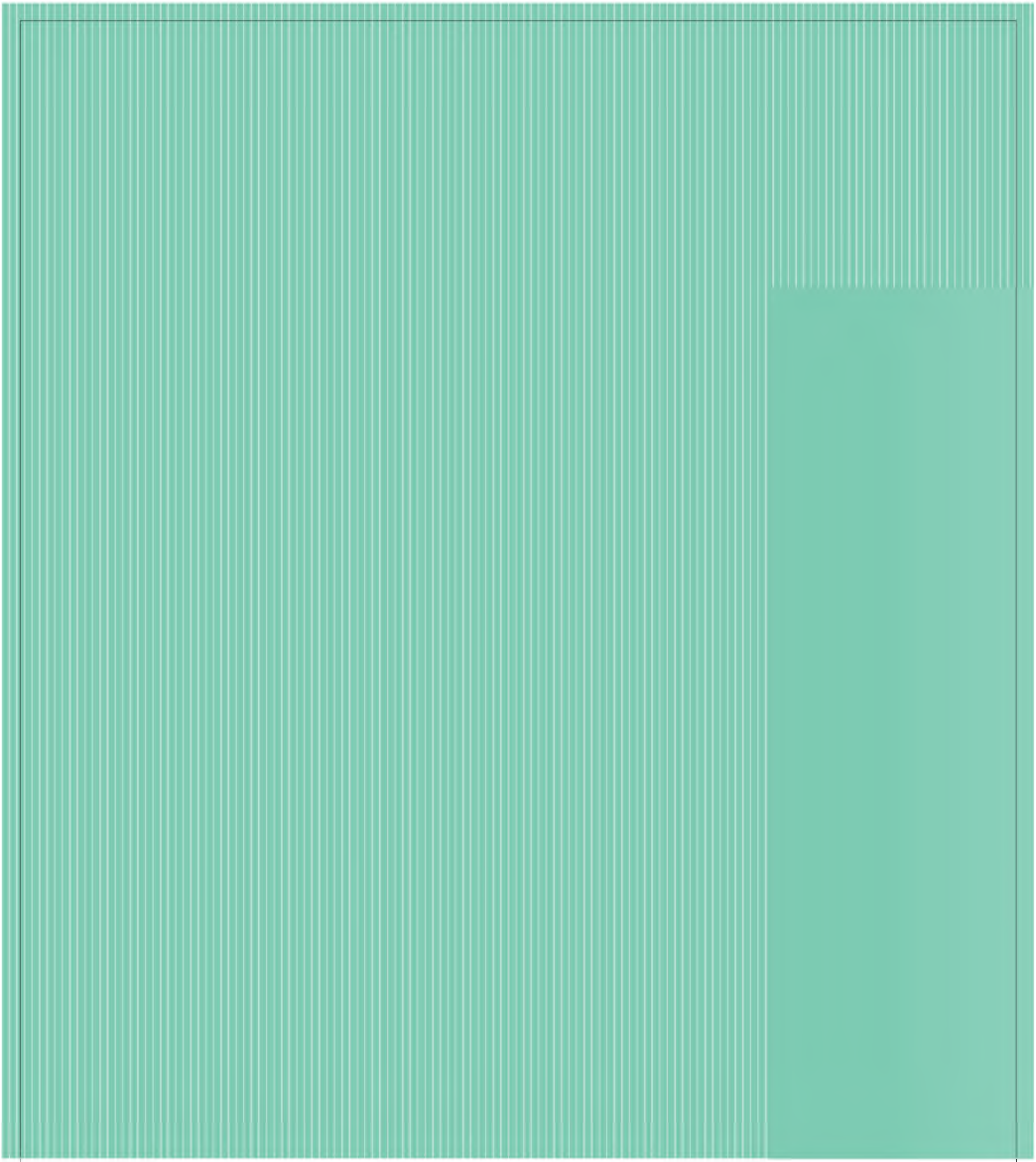
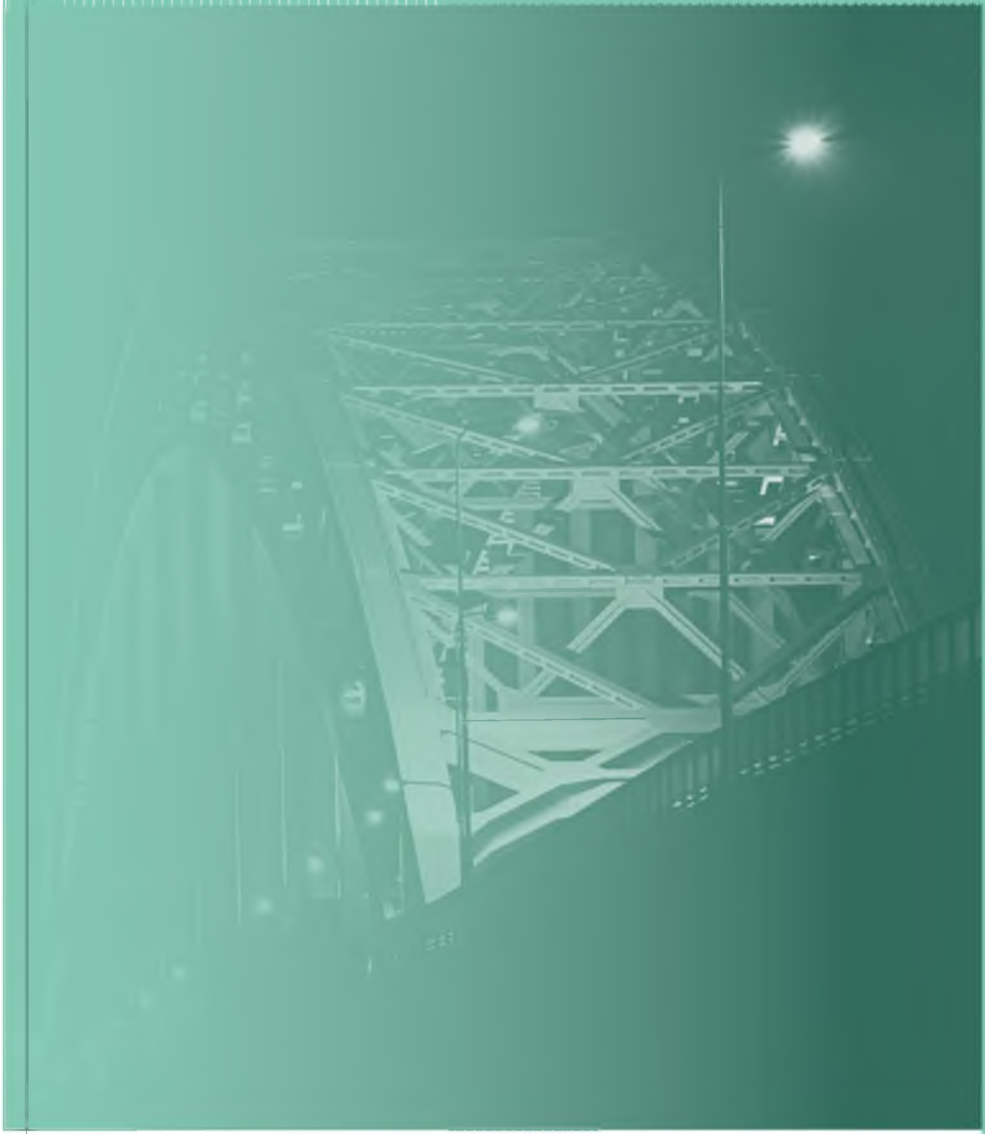


CURRICULUM VITAE



Tim olde Hartman

DOCTOR PATIENT CONSULTATION

Curriculum Vitae

Tim Christian olde Harman was born on March 13th 1977 in Almelo (The Netherlands) as son of Gerrit olde Hartman and Annie olde Hartman-Droste. He grew up with his twin brother Coen and brother Frank in Tubbergen. He completed his high school (VWO) in 1995 at the Pius X College in Almelo and was allowed to enter medical school at the Erasmus University Rotterdam in the same year. In 1999 he conducted a research elective at the department of general practice at the Erasmus Medical Centre Rotterdam, supervised by Dr. Marjolein Berger. During this period his enthusiasm for scientific research and general practice increased. He graduated as a medical doctor (cum laude) in 2001 and started to work at the department of Emergency Medicine in hospital Zevenaar. In 2002 he started his PhD project on persistent medically unexplained symptoms in primary care at the department of primary and community care at the Radboud University Nijmegen Medical Centre. The results of this project are described in this thesis. At the same time he started general practice (GP) residency training at the Radboud University Nijmegen Medical Centre in order to combine research with vocational training (the combined residency and research training programme - 'AIOTHO-programme'). He worked as GP resident in the practices of Rudy and Marijke Kleerekooper-Corsten (Elst, Gelderland) and Jaap Schreuder (Malden). During this period he was member of the editorial board of the Journal of the Dutch college of General Practitioners (Huisarts en Wetenschap) and member of the PhD Student Training Committee (PTC) of the Netherlands School of Primary Care Research (CaRe). In 2005 he finished GP training and started to work as GP in the practice of Jeanne and Pierre Mesker-Niesten. He became member of the editorial board of the international peer-reviewed journal Mental Health in Family Medicine and participated in the national multidisciplinary guideline committee on medically unexplained symptoms and somatoform disorders. In 2007 he started his own private practice in Oosterhout (Nijmegen) in the academic health care centre Oosterhout supported by the other practicing GPs in Oosterhout (Charles Verhoeff, Erna van Ewijk-van der Wielen, Han Beekwilder and Inge Nobacht-Wagenvoort). In 2010 he continued his academic practice in close collaboration with Annemarie Uijen.

In recent years he, together with Greg Irving (UK), Karen Falloon (New Zealand) and Mark van der Wel (The Netherlands), founded the MINERVA (Montreal INitiative de Education et Recherche Vocationale Academique) initiative, which aims at promoting combined residency and research training programs in family medicine globally. Last year he became chair of the Nijmegen Monitoring Project (NMP) a practice-based research network recording the process and outcome of care of chronic diseases in general practice. Furthermore he participates in the national GP guideline committee on medically unexplained symptoms of the Dutch college of General Practitioners. After his PhD defence he will continue his research activities as a post-doctoral researcher at the department of primary and community care at the Radboud University Nijmegen Medical Centre.

Tim is happily married to Marjolein olde Hartman-Hofsté, who works as an oncology nurse at the department of surgery of the Radboud University Nijmegen Medical Centre. They are the proud parents of two sons, Jop (2009) and Guus (2010).