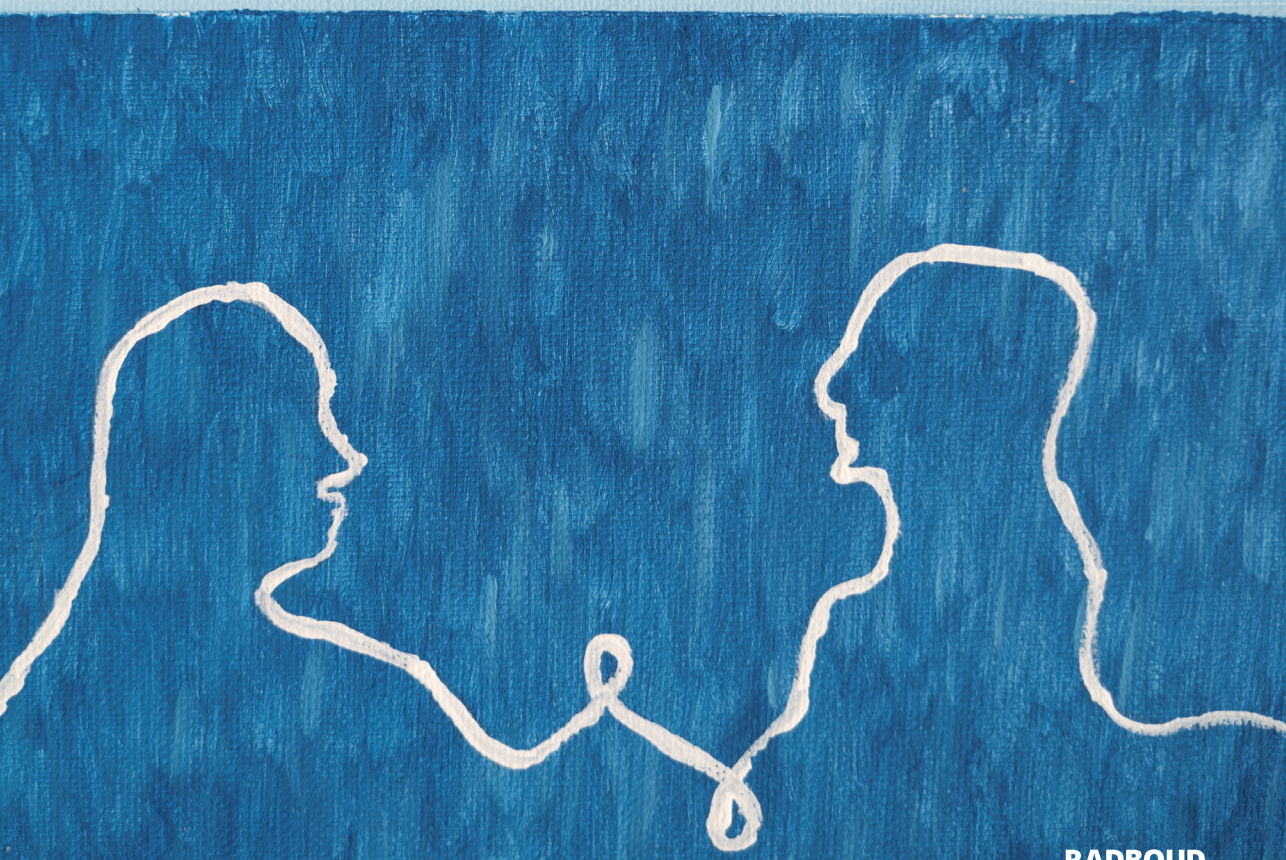
Stigmatisation of people with persistent somatic symptoms by healthcare professionals

Measurement and manifestations



Brodie McGhie-Fraser

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Stigmatisation of people with persistent somatic symptoms by healthcare professionals

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Chapter 1

General introduction

1. Introduction

Experiencing somatic (physical) symptoms such as fatigue, aches, pains or dizziness are common in our lived human experience. In most cases people do not consult a healthcare professional for their symptoms (1). Of people presenting symptoms to a general practitioner, about half of these will result in a symptom diagnosis (not meeting the criteria for a disease classification). Most of these resolve quickly. However, for approximately one in six people with a symptom diagnosis, these symptoms persist for more than a year (2).

Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months (3). These include single symptoms such as chronic pain, combinations of symptoms, or syndromes meeting the criteria for functional disorders such as fibromyalgia or irritable bowel syndrome. While they can be present in patients with well-established diseases such as cardiovascular disease (4) and in patients with symptoms without well-established biomedical pathology (5), PSS are seen as disproportionate to currently recognised pathology. These symptoms are known to have a high personal impact on daily functioning and quality of life (6-8), work participation (9), and healthcare costs (8). People with PSS report that they are often not satisfied with the quality of the healthcare they receive, often passed between specialities without a clear explanation of their symptoms or management plan (10). There is also the risk of iatrogenic harm due to high number of interventions (11).

For people living with PSS, they do not only face distress and suffering caused by the symptoms themselves. People with PSS have reported experiencing stigmatising attitudes and behaviours from healthcare professionals while seeking medical help (12-17). Specifically, people have reported that their symptoms are not taken seriously, dismissed as emotional problems, or outright fabrication, and their truthfulness and accuracy in describing symptoms is questioned (12-16). Stigmatisation is a dynamic process where elements of labelling, stereotyping, separation, status loss, and discrimination occur in the context of power (18). There is a considerable health outcome impact to this stigma. The stigmatisation of people with PSS is associated with decreased wellbeing (19), increased depression and anxiety (20), and treatment non-adherence (21).

A key component to stigma in healthcare is the personal beliefs, attitudes and behaviours of healthcare professionals. It is well reported that many healthcare professionals find the care of people with PSS difficult (22, 23), and that there are many barriers to providing healthcare to people with PSS (24). Healthcare professionals in particular report feeling insecure about communication (25). This includes providing adequate explanations for PSS, addressing psychosocial factors that can play a role in symptoms, or applying a more person-centered communication style (26). This lack of perceived confidence and competence has negative effects, for example avoiding interactions with patients where possible (27), or compensating by ordering inappropriate diagnostic procedures and interventions (28-30).

Underpinning the stigma that affects both patients and healthcare professionals is a false belief (or stereotype) that PSS are less legitimate compared to similar symptoms with more established biomedical pathological mechanisms. This is reflected in the diagnostic criteria for these symptoms that mostly rely on the exclusion of other diagnoses, and lack of explicit training for managing PSS (31, 32). But this stereotype also reflects a deeper structure of medicine that divides illness and disease into those that are somatic and others which are psychosocial (33). This binary is not just an abstract concept. It affects the type of educational material and training that is available for healthcare professionals, the trajectories of care available for patients, the financing of healthcare, and in some cases (especially in secondary care) even the buildings themselves where care is provided. To reduce stigma in PSS then, we must consider structural as well as interpersonal interventions.

In recent years there has been increased attention to how healthcare professionals work with people with PSS. This includes many examples of stigmatisation by healthcare professionals towards people with PSS, qualitative exploration of why this happens, and the development of educational interventions. However, little attention has been paid to how we measure stigma in this context. By establishing a valid and reliable measurement instrument, we would be able to provide a foundation for future stigma intervention development and evaluation.

This thesis studies stigmatisation by healthcare professionals towards people with PSS. This includes the development of a valid and reliable measurement instrument that can be used to measure stigmatisation by

healthcare professionals. This aims to provide a foundation to develop and evaluate future stigma interventions, as well as providing a more specific understanding of stigma in the context of PSS. We explore manifestations of stigma in a clinical context, the experience of stigma for patients, and the quality of available online health information.

In this introduction, we first describe the concept of stigma, introducing influential models and characteristics of stigmatised statuses (known as stigma dimensions). Second, we describe contributing factors to stigmatisation in the context of PSS, providing some understanding of *how* and *why* stigmatisation occurs by healthcare professionals. Third, we consider the terminology and diagnostic categories used to describe PSS and provide a current understanding of aetiological mechanisms. Fourth, we describe how stigmatisation has been measured in the context of PSS, and the need to evaluate stigma reduction interventions. Lastly, the rationale for this thesis and outline of each chapter is described in more detail.

2. Conceptualising stigma

The concept of stigma is increasingly salient in research, policy, clinical practice and everyday interactions. While this increased attention is commendable, it underlines the importance of using and defining a clear stigma construct. From its original meaning in Greek of a physical marker or brand onto a person, stigma was described by Goffman as the negative evaluation and discrediting of a person due to an undesired characteristic (34). This acknowledges the heavy burden that stigma carries, reducing a person who is stigmatised from a "*whole and usual person to a tainted, discounted one.*" (34)(p3).

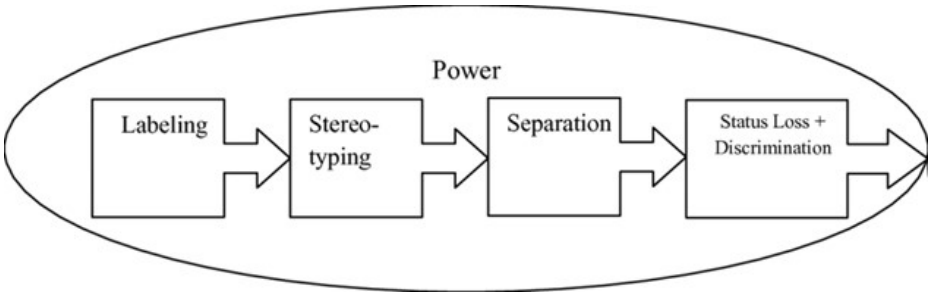
Throughout this thesis, we wanted to consider both general stigma models that can describe the stigmatisation process and impact, but also the qualities of stigma that are unique to the context of healthcare professionals managing people with PSS. We are particularly influenced by three models throughout the following chapters: 1) a sociological model of stigma (18); 2) an interpersonal model of stigma (35); and 3) a model focusing on the particular *dimensions* that characterise different types of stigma (36).

2.1. Sociological model of stigma

An influential sociological model by Link and Phelan describes stigma as starting when a person is labelled based on particular characteristics, such as having PSS. The labelled people are placed in distinct categories, which enables the effective separation of “us” from “them”. They are then linked to undesirable characteristics (such as stereotypes about patients), and devalued or excluded through status loss or discrimination (18).

This model is particularly important as it acknowledges the role and context of power. Power can be approached in absolute and relative terms. In many countries, (including the Netherlands and the UK) a general practitioner acts as a gatekeeper to secondary care (31). Therefore, they hold an element of absolute power over the trajectory, diagnosis and management plan of patients (37). In relative terms, power can also be expressed by patients. Healthcare professionals have often reported feeling powerless in their discussions with patients: feeling dominated in the conversation, or feeling responsible without being able to meaningfully improve the symptoms of the patient (38).

Figure 1: Sociological model of stigma (reproduced from Link & Phelan, 2001) (18)

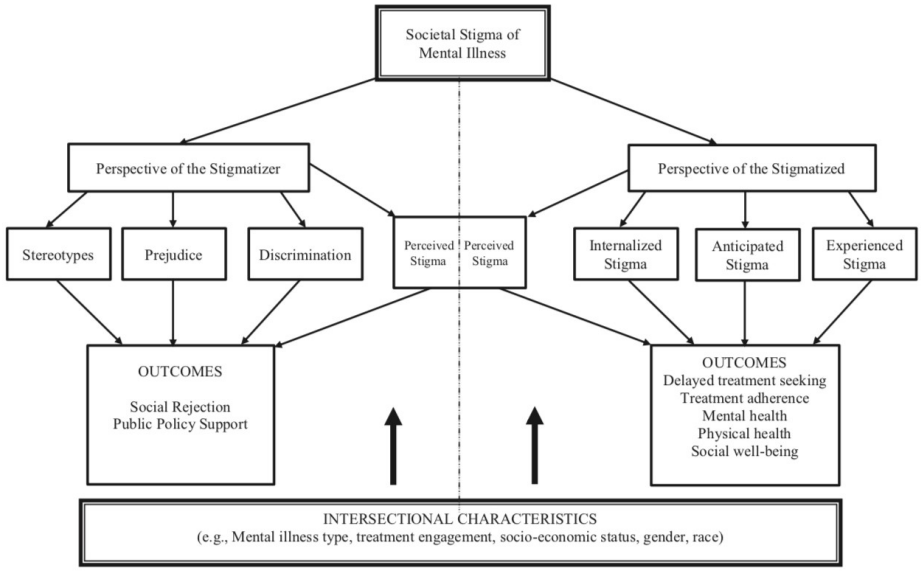


2.2. Interpersonal model of stigma

Since the sociological model focusses on the macro level process of stigma, we also wanted to be able to describe the processes of stigma at the personal and interpersonal level (i.e. the perspectives of patients with PSS, and healthcare professionals who manage people with PSS). Therefore, the second stigma model we rely on is known as the Mental Illness Stigma Framework (35). This explicitly acknowledges the role of different

perspectives of people, from those that are stigmatised, and those who are the stigmatisers. For clarity, we describe patients from the perspective of stigmatised, and healthcare professionals in their role as prospective stigmatisers. Though of course, these are not always exclusive roles (it is possible that healthcare professionals themselves are stigmatised for having PSS).

Figure 2: The mental illness stigma framework (reproduced from Fox et al., 2018) (35)



From the patient perspective, experienced stigma (sometimes called 'enacted stigma') is defined as experiences of stigmatisation from others (35, 39). This includes recurring, everyday experiences of stigma as well as acute, major experiences. There are many examples of this in PSS in clinical consultations, from having symptoms invalidated, to abusive behaviour in the consultation room (15). Anticipated stigma (sometimes called 'felt stigma') is the extent to which a stigmatised person expects to be the target of stigma in the future (35, 39). Because people are aware of negative stereotypes, they may worry about other people viewing them as less legitimate than other patients. It is possible to have anticipated stigma without experienced stigma. Examples of anticipated stigma in PSS include patients avoiding seeking treatment because they expect to have negative encounters with healthcare professionals (15). Internalised stigma

describes the extent to which people endorse negative stereotypes and emotions about the stigmatised themselves (35, 39).

From the healthcare professional perspective, the framework focusses on interacting cognitive mechanisms (stereotypes), affective mechanisms (prejudices) and behavioural mechanisms (discrimination):

- Stereotypes are "*beliefs, or 'cognitive schemas' about the characteristics and behaviours of groups of individuals*" *(35)*. These beliefs are characterised by their inaccuracy, negativity, and overgeneralisation of the stigmatised group of individuals. Stereotypes associated with PSS include a belief that patients have less severe symptoms than those with a clearer pathology (40). Other stereotypes include patients pressuring professionals for somatic testing (28), a belief that patients are fixated on somatic explanations of illness (41), and a belief that validation of symptoms will worsen the severity or prevalence of symptoms (42).

- Prejudices describe personal attitudes, defined as the "*emotional reaction or feelings that people have toward a group or member of a group*" *(35)*. Expressions of prejudice by healthcare professionals towards people with PSS include frustration that they are 'difficult' to work with (23, 43), a 'heartsink patient' (44), or resentment that patients can be manipulative (38). Prejudices can also manifest as anxiety, such as fear of saying the wrong thing (45), or defensiveness during communication (46).

- Discrimination is defined as "*unfair or unjust behaviours… existing on a spectrum from subtle to overt*," *(35)*(p132) in the "*differential and disadvantaged treatment of the stigmatised*" *(47)*(p93). Types of discrimination directed towards people with PSS include invalidation of symptoms (12), avoidance of providing PSS patients with a diagnosis or avoidance of PSS patients in general (27).

2.3. Characteristics of stigmatised conditions: stigma dimensions

The sociological framework and the interpersonal framework describe how stigma happens. But they don't account for the *forms* or the *characteristics* of the stigma that are present. For example, the type of stigma seen against people with PSS is different to discrimination based on racism,

or the stigma against people who have an alcohol addiction. Even within a seemingly similar category of health condition (i.e. an illness caused by acute infection), the stigma against people with acute COVID-19 infection is fundamentally different to that of acute HIV infection. Therefore, a third influential stigma framework used in this thesis is based on the concept of stigma dimensions.

To describe different stigmatised identities, Jones et al., (36) proposed six dimensions of stigma along which stigmatised attributes will vary. These dimensions have been confirmed in research evaluating 93 stigmatised conditions, among experts and the general public (48):

1. Concealability: how obvious or detectable a characteristic is to others
2. Course: whether the difference is life-long or reversible over time
3. Disruptiveness: the impact of the difference on interpersonal relationships
4. Aesthetics: whether the difference elicits a reaction of disgust or is perceived as unattractive
5. Origin: the causes of the difference, particularly whether the individual is perceived as responsible for this difference
6. Peril: the degree to which the difference induces feelings of threat or danger in others

Each stigmatised condition (for example, having PSS) is characterised by one or more of these dimensions. Some conditions share dimensions and are therefore clustered. (48) Though not specifically tested, PSS typically fit under the cluster labelled as '*innocuous persistent*' – that is, low levels of perceived peril, but high levels of perceived origin (perceived responsibility for origin of symptoms), course (symptoms are perceived as irreversible), and disruptiveness (awkwardness in communication). While these dimensions are common across different types of PSS, there will be also be differences between specific symptoms. For example, the concealability of persistent fatigue contrasts with the lack of concealability in functional seizures. These stigma dimensions have important explanatory power for the types of stigma that plays out.

2.4. Structural approaches to stigma

Stigma is not only based on interpersonal interactions. Rather, it has structural elements, occurring at different, intersecting scales (49). This includes the

level of nations and governments which decide healthcare policy, the amount of money available for healthcare, socioeconomic capital (for example in being able to purchase healthcare insurance), and the role of the media in shaping perceptions of people (49). While PSS are prevalent across many countries, different cultural expressions and interpretations of illness play an important role in how these are stigmatised. This thesis explicitly focuses on personal and interpersonal stigma in a clinical context, namely in Western Europe (Netherlands and the UK). However, it is only by addressing this and structural change (for example, improving knowledge and awareness about PSS through the role of the media) that stigma will be meaningfully addressed.

3. Contributing factors to stigmatisation in persistent somatic symptoms

There are several, often layering factors that contribute to PSS related stigmatisation and reduced quality of life**.** These affect people directly experiencing symptoms, friends, families and caregivers, but also healthcare professionals who often face challenges in managing people and providing appropriate treatments. This contributes to what is referred to as a "vicious cycle" - increasing distress from the patient perspective, and frustration from the perspective of healthcare professionals (50). These factors can be considered from the perspective of people with PSS, the perspective of healthcare professionals, and the perspective of the wider social context.

3.1. Perspective of people with PSS

1) *Persistence of somatic symptoms*: Persistence of symptoms can lead people to develop strategies to avoid the negative consequences of symptoms. These include withdrawal from social activities, delaying seeking care, or seeing different healthcare professionals to avoid being seen as demanding (51). Social circumstances can be further complicated by illness, including giving up employment, changes in relationships, and being unable to fulfill desired roles such as care-giving (52). Being affected by persistent symptoms is associated with higher levels of perceived stigma (53).

2) *Characteristics of presenting persistent somatic symptoms*: As well as through the persistence of symptoms, people are stigmatised on the basis of their symptoms according to their specific presenting characteristics.

For example, people with fatigue have reported that their symptoms are trivialised because it is perceived as common tiredness, as well as not outwardly looking sick (54). For symptoms related to irritable bowel syndrome (such as bloating, gas, diarrhea or constipation) there are often cultural taboos which discourage help-seeking (55). For a patient with chronic low back pain, negative test results from lumbar radiography or MRI might mean they are interpreted as malingering (56). These characteristics can be described as *dimensions* of stigma, which affect the type of stigma that plays out (see section 1.3.3).

3) *Intersectionality:* PSS related stigmatisation is highly influenced by *who* has symptoms. Intersectionality is a concept that explores how social categories interact with each other. These can create overlapping and interdependent systems of discrimination and disadvantage (57). For example, women presenting with common somatic symptoms in primary care were found to receive fewer physical examinations, less diagnostic imaging and fewer referrals to secondary care, while receiving more symptom diagnoses (rather than somatic disease diagnoses) than men (58). Women with non-specific, functional and somatoform syndromes perceived the relational conduct of their physician to be poorer when compared with men and with other healthcare conditions (59). The influence of sex and gender does not only affect women. Acting as a threat to gender identity and perceived masculinity, men have reported concealing persistent pain even at the risk of further distress (60). As well as sex and gender, racial and cultural aspects of PSS also play an important role, including differences in the perception of illness and disease concepts, and idioms of distress (61). For example, a review of pain care found that there were significant disparities across lifespan and treatment settings, with racial and ethnic minorities receiving lesser quality pain care (62).

3.2. Perspective of healthcare professionals

4) *Fragmented care across healthcare systems:* Responsibility for care of people with PSS often falls between traditional healthcare boundaries. Diagnostic and referral procedures for PSS were recently found to be inefficiently structured across European countries, with a lack of specialised treatment centres and lack of use of clinical guidelines (31). The structure of healthcare systems reinforces the notion that certain health conditions are more valued than others. Further, this fragmentation means that the care of

people with PSS often involves multiple and repeated consultations between different specialists. This puts patients at risk of unnecessary referrals, testing and treatment due to misdiagnosis (11).

5) *Challenges in communication:* PSS are not exclusively attributed to somatic diseases, nor are they exclusively symptoms of a mental disorder (4, 63). As a consequence, many healthcare professionals experience 'incongruence' or a 'mismatch' between the severity and persistence of symptoms and the absence of somatic abnormalities. This results in a struggle in communication and in providing explanations to patients with PSS (25, 64).

There is growing evidence that healthcare professionals communicate in a less person-centred way than in consultations with patients with clearer pathologies, often failing to explore patients' reason for encounter and their ideas and expectations about the symptoms (65). This is because there can be powerful discrepancies between perceptions of communication (expectations of how patients or healthcare professionals will communicate), and actual communication as seen in observational research. While there are no systematic variations in language use for people with PSS (66), healthcare professionals have been found to adjust their communication (fewer structuring techniques during the consultation (65) less positive framings (67), and more uncertainty markers (68)) in response to the perceived inexplicability of symptoms.

6) *Lack of knowledge*: There is a lack of formal training for healthcare professionals about PSS. For example, one study in the UK found that only 11% of medical schools offered formal training about PSS (69). A lack of knowledge leads to less competence in critical skills such as providing explanations. Meaningful explanations of symptoms to patients with PSS are important for several reasons. First the patient has sufficient explanation to reduce their need to find further reasons for their symptoms. Second, explanation can lead to a rationale for engaging with appropriate forms of treatment (70). Third, explanation can help with social integration, such as discussing illness with friends and family, or reintegration into work. A lack of adequate explanations provided about the nature of a patient's symptoms can cause further emotional distress, rumination about symptoms, and contribute to internalisation of stigma (56).

3.3. Perspective of the wider social context

7) *Mind-body dualism:* Underlying the problem of perceived inexplicability is a model of health and illness that divides symptoms to either biomedical or psychosocial factors. This 'mind-body dualism' places a particular focus of healthcare professionals on finding and treating disease. If no immediate underlying cause is found for a symptom, then symptoms are assumed to be psychological or less valid.

This tension between somatic and psychological factors form key stereotypes of patients with PSS by healthcare professionals. Patients are often described as having a 'somatic fixation', seeking only a somatic explanation for illness (41), whereas healthcare professionals themselves struggle to explore emotions and psychosocial factors during consultation (71). Similarly, healthcare professionals also reported feeling pressure from patients to conduct testing and make specialist referrals, even though in practice this further action was mostly suggested first by professionals (27, 28, 142).

Clinical and research practice is increasingly moving towards a biopsycho-social approach to health and illness, where biomedical, psychological, social and contextual factors are integrated in understanding the experience of symptoms (72). Despite being integrated into medical training, implementation of the biopsychosocial approach remains a challenge (73, 74). Supporting this is the importance of providing person-centred care, which means treating patients as individuals and as equal partners (75). This is valued by people with PSS, and enhances the feeling of being taken seriously (76).

4. Terminology and diagnostic categories in persistent somatic symptoms

4.1 Language matters

The language used to describe PSS acts as a *zeitgeist* - a mirror of current perceptions and attitudes. The wide array of terms used and the shifts over the years to describe PSS are a marker of its troubled history, but also point towards how we might reduce stigma in the future.

Historically people with PSS have been referred to as 'heartsink patients' (44), or 'hysterical' (77). In contrast to identifying disease in order to treat it, these terms instead became markers of illegitimate patients: *"These [names for] symptoms didn't just signify what a person had. They told you what a person was."* (78). As well as being clinically vague, these terms reinforced harmful stereotypes about the health-seeking behaviour of women that persist to this day (79).

Many different terms to describe symptoms like persistent somatic symptoms have emerged through the years. These posit their own ideas about the cause of symptoms, recommended treatments, and characterisations of patients themselves. The framing of these terms is often based on the perspective of the treating healthcare disciplines. Conversion, psychogenic, and somatisation described a patients' tendency to experience psychological distress in the form of somatic symptoms. 'Medically unexplained' symptoms emerged as a term in primary care that aimed not to blame patients for the experience of symptoms, but only emphasised an *absence* of explanation (e.c.i). In recent years this is considered as problematic because: 1) being labelled as 'unexplained' creates distress in patients; 2) assessing if there is a medical explanation or not is unreliable; 3) the concept reinforces that if an illness is not 'of the body' then it must be 'in the mind'; and 4) many patients did not approve of the term (3, 80).

Currently, diagnostic terms are used according to treating specialty. There are currently two major classification systems involving PSS: the World Health Organisation's International Classification of Diseases (ICD) and the American Psychiatric Association's Diagnostic and Statistical Manual (DSM). ICD includes all somatic and mental disorders, whereas the DSM focuses only on mental disorders. Specific symptoms or syndromes (clusters of

symptoms) are defined in the ICD, such as irritable bowel syndrome or fibromyalgia. Persistent somatic symptoms or persistent physical symptoms are terms mainly used in primary care, whereas specialised medical fields more commonly employ the term 'functional syndromes' or 'functional disorders' (81). Specific mental disorders such as Bodily Distress Disorder or Somatic Symptom Disorder are diagnosed when persistent somatic symptoms are combined with distressing psychological symptoms. We predominately use the term PSS in this thesis because: 1) it is the preferred umbrella term by people living with symptoms [3], 2) it was acknowledged as a preferred term from an international group of experts across Europe (31); 3) is particularly used and recognised in primary care settings. However, throughout the thesis, specific functional disorders are referred to as appropriate, such as Functional Neurological Disorder (FND). This is when there is a focus on particular symptoms or diagnostic categories.

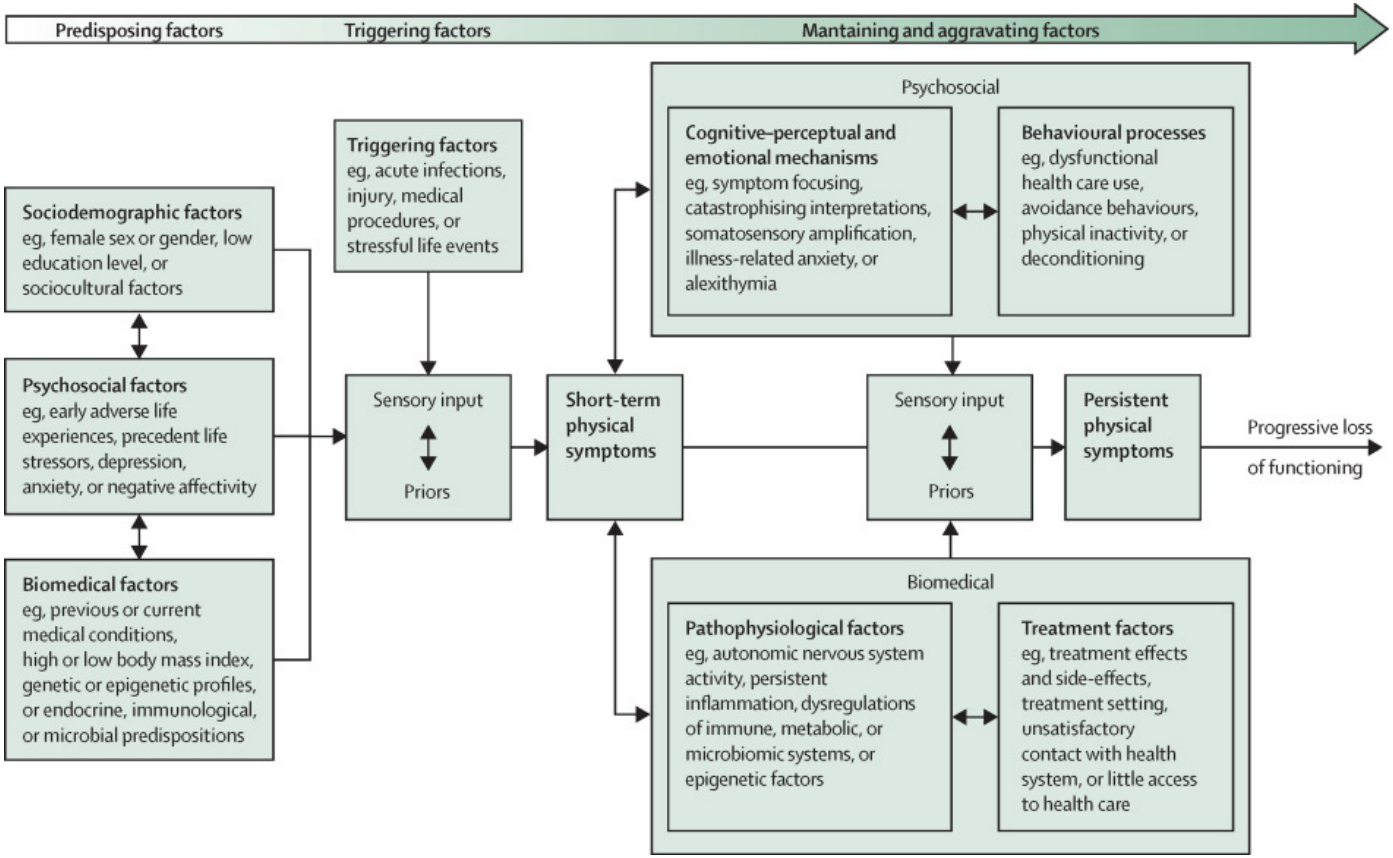
The language around PSS has evolved significantly in recent years, and will likely continue to do so. The trajectory of these shifts has not been consistent or linear, as seen in both clinical practice and research. This thesis cannot help but also reflect this transition and moment in time. With our research focus on stigma, what matters more than the specific terms deployed are two semantic shifts: a shift away from perceived inexplicability, and a shift away from a mind-body dualism.

4.2 Current understanding of aetiological mechanisms of PSS

While symptoms have historically been distinguished as those with a clear biomedical pathophysiology or not, there is increasing evidence that all persistent symptoms share neuropsychological mechanisms (82, 83). Understanding of aetiological mechanisms in PSS is growing, with current models linking predisposing factors, triggering factors, and maintaining factors **(Figure 3)** (84).

Regarding specific mechanisms, neuroscientific models suggest that PSS are due to dysfunction in symptom processing in the central nervous system. Symptom perceptions emerge through an interaction of sensory inputs, prior experience (leading to negative symptom expectations) and contextual cues such as affective state (85). Improved understanding of mechanisms is a key factor in developing explanations that are suitable and acceptable to patients (86, 87).

Figure 3: Conceptual working model of risk factors and mechanisms currently under discussion for the persistence of somatic symptoms (reproduced from Löwe et al., 2024 (84))



4.3. Towards positive diagnostic criteria

Increasingly, general practitioners try to make a biopsychosocial diagnosis, considering different contributing factors to presenting symptoms, and eliciting the patient's thoughts and emotions about the symptoms. This information can be used to construct an explanation for the symptoms in close collaboration with the patient. However, key to the stigmatisation process in PSS is that diagnoses for PSS are sometimes made solely through a diagnosis of exclusion. This means that a diagnosis is presented as what something *isn't*, rather than through its presenting characteristics.

Typically to confirm something as PSS, other conditions have been first ruled out, there is an absence of structural damage, or the symptom distress is disproportionate to currently understood pathology. Ruling out other conditions in the differential diagnosis is an important component of healthcare. Indeed, missing underlying causes for symptoms is a key concern for many healthcare professionals and often the reason for repeated diagnostic tests (30). However, *only* being presented with a diagnosis of exclusion effectively reinforces the stereotype that PSS is less legitimate than other health conditions.

- Example of diagnosis by exclusion: the diagnosis of myalgic encephalomyelitis /chronic fatigue syndrome (ME/CFS) is made on the basis of persistent fatigue that is not explained by another condition (88).

For some functional disorders such as functional neurological disorder, great progress has been made in identifying internally consistent diagnostic signs, communicating these clearly and making them publicly available (89). The challenge will be to find similar positive identification for other non-neurological symptoms that can be used to aid diagnosis and provide better explanations:

- Example of positive diagnostic criteria by internally consistent diagnostic signs: the Hoover test is commonly used to identify functional weakness, a symptom of functional neurological disorder (89). This is done by testing the weakness of voluntary hip extension with normal involuntary hip extension during contralateral hip flexion against resistance.

5. Measuring stigmatisation

Addressing stigma in PSS is increasingly being recognised as a priority in research and clinical practice (16, 90). Despite this, very little attention has been paid to the quality of stigma measurement in PSS. Extensive reviews of measures have been done in the field of mental health stigma (35, 91-93), but not in PSS. Measuring PSS related stigmatisation by healthcare professionals can establish how prevalent a problem it is, which groups of healthcare professionals should be targeted for more support and intervention, and whether interventions against stigma are effective.

Since stigma is a social construct, there is no 'gold standard' for measuring PSS related stigmatisation by healthcare professionals. However, there are ways to establish how valid, reliable and responsive different measurement instruments are (in this thesis we refer to these quality indicators as measurement properties). In recent years, a consistent taxonomy of measurement properties has been developed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group (94, 95). This provides a standardised and systematic way to compare the quality of instruments, as well as design studies (96).

5.1. Explicit measures

Stigma is most commonly assessed using self-reported questionnaires, which allows for testing of specific stigma concepts, comparison between different groups of healthcare professionals, and evaluation of stigma reduction interventions. Questionnaires can be developed to assess stigma relating to a specific PSS, assess stigma across PSS in general, or assess stigma across different health conditions. It is important that the measurement properties of instruments are sufficient. However, understanding detailed experiences of healthcare professional is also crucial, as seen in several qualitative studies with healthcare professionals (25, 38, 97).

This raises important limitations of self-reported instruments. Limitations include people not explicitly endorsing negative attitudes, or preferring not to reveal their attitudes through a social desirability bias (98). This is particularly important when there are social pressures against explicit stigmatising behaviour, such as the risk of professional liability. We use

multiple methods to try and understand the potential for social desirability bias throughout this thesis.

5.2. Implicit measures

Implicit measures of stigma aim to directly measure attitudes, thus reducing the risk of social desirability biases. These can typically be divided into response latency tests where differences in response times are measured to assess bias (99) or response priming studies where differences in vignettes are compared (40). The most commonly used method of measuring implicit attitudes in clinical settings is the Implicit Association Test, a type of response latency test (98).

6. Rationale for this thesis

In recent years there has been increased attention to how healthcare professionals work with people with PSS. This includes many examples of stigmatisation by healthcare professionals towards people with PSS, philosophical exploration of why this happens, and the development of educational interventions to improve knowledge and specific skills such as communication. However, little attention has been paid to how we *measure* stigma in this context. Measurement of stigma is important because you can target particular groups of healthcare professionals for stigma reduction interventions (such as increased support or education), and you can meaningfully design and evaluate stigma reduction interventions.

As described in section 1.4, there are multiple methods (explicit and implicit) methods to measure PSS related stigma. Multiple types of measurement are needed to understand dynamic and complex behaviour such as stigma. In our initial searches for the suitability of instruments, we found that many instruments existed, but most without validation or evidence of their development. This is particularly so for implicit measurement instruments such as the Implicit Association Test (IAT), where despite its regular use, doubts had been raised about the validity and reliability of these tests (100, 101). We also wanted to use a measure with higher feasibility among larger samples of healthcare professionals. Therefore, we made the decision to focus on explicit measures, in order to establish a foundation to which to compare to other measures in the future.

By establishing a valid and reliable measurement instrument, we would be able to provide a foundation for future stigma intervention development and evaluation.

7. Aims for this thesis

The aim of this thesis is to:

- develop a valid and reliable measurement questionnaire instrument to measure stigmatisation by healthcare professionals towards people with PSS
- develop increased understanding of how stigmatisation by healthcare professionals unfolds and impacts people with PSS
- develop specific theoretical understanding of stigmatisation by healthcare professionals towards people with PSS in order to aid future development of stigma reduction interventions

8. Outline of this thesis

In **Chapter 2,** we performed a systematic review of questionnaire measurement instruments for stigmatisation by healthcare professionals towards patients with PSS. We used COSMIN criteria to evaluate the methodological quality and measurement properties of available stigma instruments. Our conclusion from this review shifted our focus towards the development of a new instrument to measure PSS related stigmatisation.

In **Chapter 3**, we developed a new stigma scale to measure stigmatisation by healthcare professionals towards people with PSS: the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). We followed an iterative process of generating and reviewing items, as well as cognitive interviews with healthcare professionals to select and improve items important for measuring stigma.

In **Chapter 4**, we validated the newly developed stigma scale (the PSSS-HCP) in an observational study of healthcare professionals in the UK. We presented the final 13-item version of the PSSS-HCP, evaluation of the validity and reliability of the scale, and an exploratory analysis of the factor structure.

In **Chapter 5**, we validated a Dutch version of the PSSS-HCP in an observational study of healthcare professionals in the Netherlands. We confirmed the factor structure of the PSSS-HCP as well as further evaluating the validity and reliability of the scale.

In **Chapter 6**, we explored patient experiences of stigma in functional neurological disorder through a qualitative study. We used semi-structured interviews to explore how stigma unfolded from symptom onset, through diagnosis and afterwards in everyday encounters. We explored the experience of stigma from multiple perspectives, including from healthcare professionals, as well as internally (self) and the influence of friends of family.

In **Chapter 7**, we explored the quality of health information related to specific diagnostic categories related to functional disorders. We rated the stigmatising content, credibility, comprehensiveness, and usefulness of Wikipedia articles across 14 languages, making recommendations for future health information.

In **Chapter 8**, we present the main findings of the thesis, and draw conclusions bring together the different threads of stigma theory, research and clinical practice.

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Chapter 2

A review of stigma questionnaire measurement instruments

McGhie-Fraser B, Lucassen P, Ballering A, Abma I, Brouwers E, van Dulmen S, olde Hartman T. Persistent somatic symptom related stigmatisation by healthcare professionals: A systematic review of questionnaire measurement instruments. *Journal of Psychosomatic Research. 2023 Mar 1;166:111161.*

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Abstract

Objective: Patients with persistent somatic symptoms (PSS) experience stigmatising attitudes and behaviours by healthcare professionals. While previous research has focussed on individual manifestations of PSS related stigma, less is known about sound ways to measure stigmatisation by healthcare professionals towards patients with PSS. This review aims to assess the quality of questionnaire measurement instruments and make recommendations about their use.

Methods: A systematic review using six databases (PubMed, Embase, CINAHL, PsycINFO, Open Grey and EThOS). The search strategy combined three search strings related to healthcare professionals, PSS and stigma. Additional publications were identified by searching bibliographies. Three authors independently extracted the data. Data analysis and synthesis followed COSMIN methodology for reviews of outcome measurement instruments.

Results: We identified 90 publications that met the inclusion criteria using 62 questionnaire measurement instruments. Stereotypes were explored in 92% of instruments, prejudices in 52% of instruments, and discrimination in 19% of instruments. The development process of the instruments was not rated higher than doubtful. Construct validity, structural validity, internal consistency and reliability were the most commonly investigated measurement properties. Evidence around content validity was inconsistent or indeterminate.

Conclusion: No instrument provided acceptable evidence on all measurement properties. Many instruments were developed for use within a single publication, with little evidence of their development or establishment of content validity. This is problematic because stigma instruments should reflect the challenges that healthcare professionals face when working with patients with PSS. They should also reflect the experiences that patients with PSS have widely reported during clinical encounters.

1. Introduction

The term 'persistent somatic symptoms (PSS)' is used as an umbrella term to describe subjectively distressing somatic complaints, irrespective of their aetiology, that are present on most days for at least several months (1)(p2). These symptoms are common as these are present in over one third of primary care and secondary care consultations (2,3). Many patients with PSS experience stigmatising attitudes and behaviours from the general public, family and friends, as well as from healthcare professionals. Patients have reported that their symptoms are not taken seriously by others, their conditions are dismissed as emotional problems, and their truthfulness and accuracy in describing symptoms is questioned (4,5).

An important factor contributing to stigma by healthcare professionals is that PSS are not exclusively attributed to somatic diseases, nor are they exclusively symptoms of a mental disorder (6,7). As a consequence, many health professionals experience incongruence between the severity and persistence of symptoms and the absence of somatic abnormalities, resulting in a struggle to provide explanations to patients (8). Incongruences between healthcare professionals and patients can act as a barrier to successful communication and a strong therapeutic relationship (9). While the concept of medical inexplicability in PSS is increasingly seen as problematic (10), this appears to play a role in the stigmatising process with higher levels of perceived stigma compared to conditions with more established pathophysiology (11,12). While most recent research on PSS has focussed on medically unexplained symptoms in primary care, or functional somatic symptoms/ syndromes in other medical specialities, we do not consider PSS as just a new name for these. We argue that PSS both reflects advances in understanding of aetiological mechanisms (13) and a focus on the impact of symptoms on patients (14).

There is increasing evidence of stigmatisation towards patients with PSS by healthcare professionals. Negative emotions about these patients are well documented (15,16). These negative emotions may be developed through exposure to adverse attitudes of senior professionals during training, time and service pressures, and emotional reactions experienced when working with these patients (17). Stigmatisation of patients by healthcare professionals is known to reinforce feelings of rejection and act as a barrier to care seeking and treatment engagement (18). For these reasons,

challenging PSS related stigmatisation by healthcare professionals is increasingly a priority in research and clinical practice.

A first step in this process is to assess the levels of stigma towards patients with PSS by healthcare professionals. While there is strong evidence of individual manifestations of stigma, less is known about sound ways to measure stigma by healthcare professionals towards patients with PSS. Valid, reliable and responsive measurement of stigma is needed to understand its prevalence and severity, but also to design effective interventions against stigma and evaluate their effectiveness. This review aims to: 1) identify existing questionnaire measurement instruments of PSS related stigmatisation by healthcare professionals; 2) assess the measurement properties of these instruments; 3) make recommendations about further development and use of stigma measurement instruments.

1.1. Definition of stigma

From its original meaning of a physical marker or brand onto a person, stigma was described as the negative evaluation and discrediting of a person due to an undesired characteristic (19). An influential model by Link and Phelan (2001) describes stigma as a social process where elements of labelling of the stigmatised person, stereotyping, separating, status loss and discrimination occur in the context of power (20). This model usefully highlights the psychosocial processes of stigmatisation. However, this stigma model is less relevant when evaluating interventions against stigma because it focuses less on the perspective of stigmatisers. This study focusses on the stigma by healthcare professionals towards patients, considering healthcare professionals in the role as stigmatisers.

Among stigmatisers, stigma models focus on interacting cognitive mechanisms (stereotypes), affective mechanisms (prejudices) and behavioural mechanisms (discrimination) ((21), (22), (23)). Stereotypes are *"beliefs, or 'cognitive schemas' about the characteristics and behaviours of groups of individuals"* (22)(p132). These beliefs are characterised by their inaccuracy, negativity, and overgeneralisation of the stigmatised group of individuals (24,25). Stereotypes associated with PSS include patients having a personality problem (15), and a belief that patients have less severe symptoms than those with a clear pathology (26). Other stereotypes include patients pressuring professionals for somatic testing (27,28), a belief that patients are

fixated on somatic explanations of illness (29) and a belief that validation of symptoms will worsen the severity or prevalence of symptoms (30). Stereotyping has the effect of perceiving a group as fundamentally different from the perceived norm and linked with undesirable characteristics. Prejudices are the "*emotional reaction or feelings that people have toward a group or member of a group*" (22)(p132). Expressions of prejudice by healthcare professionals towards people with PSS include frustration that they are 'difficult' to work with (15,16), or resentment that patients can be manipulative (31). Prejudices can also manifest as anxiety, such as fear of saying the wrong thing (32), or defensiveness during communication (33). Discrimination is defined as "*unfair or unjust behaviours... existing on a spectrum from subtle to overt*," (22)(p132) in the "*differential and disadvantaged treatment of the stigmatised*" (34)(p93). Types of discrimination directed towards people with PSS include invalidation of symptoms (35), avoidance of providing PSS patients with a diagnosis or avoidance of PSS patients in general (36). More explicit forms of discrimination include jokes at the expense of patients (37,38).

1.2. Measuring stigmatisation by healthcare professionals

There is no 'gold standard' for assessing PSS related stigmatisation by healthcare professionals. Stigma is most commonly assessed using self-reported questionnaires, which allows for testing of specific stigma concepts, comparison between different groups of healthcare professionals, and evaluation of anti-stigma interventions. For these reasons, we have focused our review on questionnaire instruments. Questionnaires can be developed to assess stigma relating to a specific PSS, assess stigma across PSS in general, or assess stigma across different health conditions. It is important that the measurement properties of instruments are sufficient.

2. Method

This systematic review follows the protocol recommended by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (39). To assess the measurement properties of instruments, we have followed the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (40). This review was pre-registered using the PROSPERO International Prospective Register of Systematic Reviews (CRD42021287798). The current study is part of the innovative training

network ETUDE (Encompassing Training in fUnctional Disorders across Europe) (41).

2.1. Search strategy

To identify publications using or validating relevant questionnaire stigma measurement instruments, we searched in four databases that include published literature (PubMed, Embase, PsycINFO, and CINAHL) and two databases of unpublished literature (Open Grey and EThOS). Search strategies were developed by translating our research question to PICO criteria. The search strategy combined three search strings related to healthcare professionals, PSS and stigma mechanisms respectively (**Appendix 1, supplementary data**). The terms related to these three components in the search string were based on previous systematic reviews (42,43,8,44). Since pilot searches identified several publications without validation of instruments, we decided not use a specific search string for measurement properties. We manually searched for additional publications by screening the reference lists in the included publications, screening the reference lists in relevant systematic reviews, and other publications by included authors.

2.2. Eligibility criteria

To be included, the publication had to meet the following criteria:

1) Related to healthcare professionals working with patients with PSS.

2) Related to PSS. At the symptom level, we considered single PSS (e.g. chronic low back pain) or terms which refer to symptoms in general (e.g. medically unexplained symptoms). When there was no secondary disease provided, we included publications relating to chronic pain. This aligns with recent evidence referring to chronic pain as a health condition in its own right (45). At the syndrome level, we considered clusters of symptoms according to organ or physiological system (e.g. irritable bowel syndrome). At the diagnosis level, we considered conditions that fulfil the criteria of functional somatic disorders or psychiatric disorders (e.g. somatic symptom and related disorders in the DSM-5). We did not include publications that focussed solely on race or gender-related stigma among patients, even if these patients in some cases also had PSS.

3) Related to stigma. We considered a publication to be relating to stigma when at least one item in the questionnaire measure instrument assessed one or more stigma mechanism (stereotypes, prejudice, discrimination). Publications that focussed only on clinical knowledge of symptoms (e.g. knowledge about contributing factors to low-back pain) or appropriate treatment options were excluded.

4) Used at least one questionnaire measurement instrument. We included all relevant instruments regardless of the validation of the instruments. Systematic reviews, qualitative publications or publications not using questionnaire instruments were excluded.

5) The publication was published in a language accessible to the reviewers (English, Dutch, French or German).

There were no limits on year of publication (from start of database until the search date of 5 November 2021) or publication status.

2.3. Publication selection

Titles and abstracts were screened and assessed for inclusion by two independent male reviewers (BMF and PL) with a background in medical anthropology and sociology, and general practice respectively. Any disagreements were discussed and the articles were reviewed together against the inclusion and exclusion criteria until consensus was reached. We made decisions around what constituted a unique version of a measurement instrument based on what we considered to be a significant adaptation. This included item or sub-scale addition or reduction, or change in target population (for example, from patients to healthcare professionals).

2.4. Data extraction, analysis and synthesis

To systematically review the quality of measurement properties in included instruments, we used the taxonomy developed by the COSMIN group (46). This taxonomy comprises three domains of measurement properties: 1) reliability (the degree to which a measurement is free from measurement error); 2) validity (the degree to which an instrument measures the construct(s) it aims to measure) and; 3) responsiveness (the ability of an instrument to detect change over time in the construct to be measured).

Further definitions of measurement properties are provided in **Appendix 2 (supplementary data)**. Since there is no 'gold standard' for assessing PSS related stigma, it is not possible to assess criterion validity. In addition, we assessed reported information about the development of measurement instruments (a component of content validity), interpretability and feasibility of instruments. Analysis followed the COSMIN guidelines for the systematic reviews of outcome measurement instruments (40,47,48). While these guidelines are primarily designed for patients as target population, the process was simply adapted by applying the same criteria to healthcare professionals.

First, we recorded basic characteristics of each included publication, such as author information, content of the measurement instrument, target population, publication location, sample size and measured stigma mechanism. For each measurement instrument, we made a distinction between: 1) outcome only publications that used an existing instrument to measure an outcome, with no validation of the instrument; 2) development publications, that developed an instrument and; 3) validation publications, that explicitly evaluated the measurement properties of an existing instrument. We did not contact authors for additional information about instrument or measurement properties, relying on information included in the original manuscripts and supplementary data only.

Second, for instruments used in development and validation publications, we used the COSMIN risk of bias checklist to assess the methodological quality of publications (47). The checklist contains multiple questions to critically appraise the methods for each reported measurement property, using a four point scale: 'inadequate', 'doubtful', 'adequate' and 'very good'. The overall rating of the quality of each publication is determined by taking the lowest rating of any standard in the checklist.

Third, the result of each publication on a measurement property was rated against the most recent criteria for good measurement properties **(Appendix 3, supplementary data).** These criteria provide a benchmark to determine whether the measurement instruments' results of testing are acceptable and provide ratings of either sufficient (+), insufficient (–), or indeterminate (?)**.**

Lastly, the quality of the evidence for each measurement instrument was summarised and graded using a modified GRADE approach. Due to the high

heterogeneity and limited numbers of publications for each measurement instrument, results were qualitatively summarised rather than statistically pooled. The GRADE approach uses five factors to determine the quality of the evidence: risk of bias, inconsistency of the results of the publications, indirectness (evidence comes from different populations, interventions or outcomes than the ones of interest in the review), and imprecision (wide confidence intervals). The quality of the evidence is graded as high, moderate, low, or very low. All stages of data analysis were performed by three independent reviewers (BMF and PL/AB). Disagreements were resolved in discussion, with a further author of the article providing advice (IA).

3. Results

The database searches found 10,037 publications after removing duplicates. Based on the title and abstract, 224 appeared to meet the inclusion criteria. After assessing the full text, 86 publications were included. Four additional publications were identified by checking reference lists and using citation tracking resources. In total, we included 90 publications and 62 questionnaire measurement instruments **(Fig. 1, Table 1).**

Figure 1: Flow chart for included publications and measurement instruments

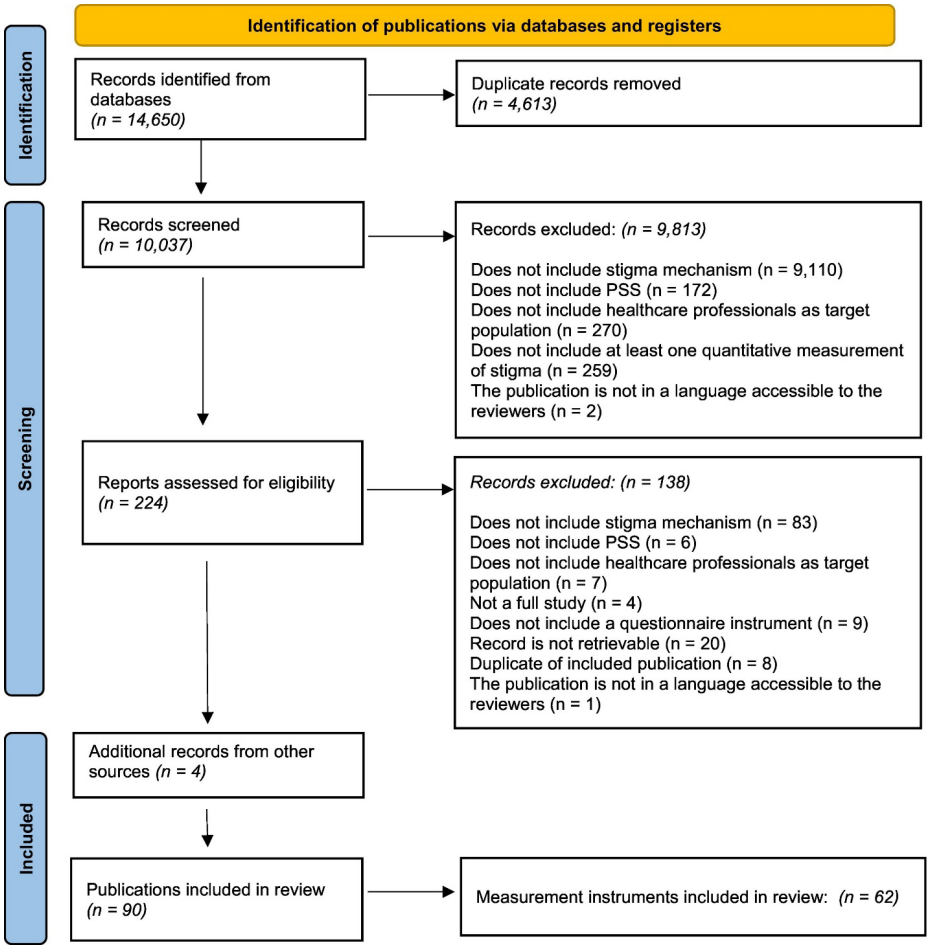


Table 1: General characteristics of measurement instruments

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
3*I-P-J	Homma et al., 2016 (49)	development	Japan
Attitudes, Experiences and Management Strategies of GPs Towards Medically Unexplained Symptoms†	Sirri et al., 2017 (50)	outcome only	Italy
Attitudes of Nurses toward Patients With Conversion/Functional Symptoms†	Ahern et al., 2009 (51)	development	UK
Attitudes to the Care of Children/ Adolescents and Adults with TMD†	Lindfors et al., 2016 (52)	outcome only	Sweden
Attitudes towards Cause and Management of Medically Unexplained Symptoms†	Reid et al., 2001 (15)	development	UK
	Nawal Hassan Gholome et al., 2008 (53)	outcome only	Kuwait
	Husain et al., 2011 (54)	outcome only	Pakistan
Attitudes toward Pain†	Wilson et al., 1992 (55)	outcome only	USA
Attitudes and Knowledge about Irritable Bowel Syndrome†	Longstreth and Burchette 2003 (56)	development	USA
Attitudes and Practices in the Evaluation and Treatment of Irritable Bowel Syndrome†	Lacy et al., 2006 (57)	development	USA
Attitudes of General Practitioners (GPs) towards Somatising Patients†	Garcia-Campayo et al., 1998 (58)	development	Spain
Attitudes to Patients with a Self Diagnosis of Myalgic Encephalomyelitis†	Scott et al., 1995 (59)	development	UK
Attitudes towards Somatising Patients†	Rosendal et al., 2005 (60)	development	Denmark
Back Pain Advice and Pharmacists' Experience of Back Pain†	Silcock et al., 2007 (61)	development	UK
Barriers for Primary Care Physicians in Diagnosis and Treatment of Persistent Somatic Symptoms†	Lehmann et al., 2021 (62)	development	Germany
Beliefs and Attitudes in GPs in the Management of Mechanical Low Back Pain†	Chaudhary et al., 2004 (63)	development	UK
Beliefs and Attitudes of General Pediatricians and Pediatric Gastroenterologists Regarding Functional Gastrointestinal Disorders†	Sood et al., 2011 (64)	development	USA; Canada

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
rheumatologists	Fibromyalgia	stereotype, prejudice, discrimination	invalidation
general practitioners	Medically unexplained symptoms	prejudice	attitudes, experiences, management
nurses	Functional neurological disorder (FND) symptoms / conversion symptoms	stereotype, prejudice, discrimination	attitudes
dentists	Temporomandibular disorders	prejudice	knowledge, attitudes and clinical experience
general practitioners	Medically unexplained symptoms	stereotype, discrimination	attitudes towards cause and management
general practitioners	Medically unexplained symptoms	stereotype, discrimination	attitudes towards cause and management
general practitioners	Medically unexplained symptoms	stereotype, discrimination	attitudes towards cause and management
medical students	Chronic pain	stereotype, prejudice	attitudes
primary care physicians	Irritable Bowel Syndrome	stereotype, prejudice	attitudes and knowledge
internal medicine physicians; family practice physicians; gastroenterology physicians	Irritable Bowel Syndrome	prejudice	attitudes and practices in evaluation and treatment
general practitioners	Somatising patients	prejudice	attitudes
general practitioners	Myalgic Encephalomyelitis	stereotype, prejudice, discrimination	attitudes
general practitioners	Somatising patients	stereotype, prejudice	attitudes
community pharmacists	Chronic low back pain	prejudice	attitudes, knowledge and reported practice
primary care physicians	Persistent somatic symptoms	stereotype, prejudice, discrimination	diagnostic and treatment barriers
general practitioners	Low back pain	stereotype, prejudice, discrimination	beliefs and attitudes
general pediatricians; pediatric gastroenterologists	Functional gastrointestinal disorder	stereotype	beliefs and attitudes

Table 1: Continued

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
B-IPQ (Brief Illness Perception Questionnaire)[1]	Aatti et al., 2016 (65)	outcome only	France
	Medina et al., 2021 (66)	outcome only	USA
B-IPQ-P-I†[2] (Brief Illness Perception Questionnaire – physicians point of view - Israel)	Aloush et al., 2021 (67)	development	Israel
BIPQ-P-J†[2] (Brief Illness Perception Questionnaire – physicians point of view - Japan)	Homma et al., 2016 (49)	development	Japan
CAT (Chronic Fatigue Syndrome Attitudes Test)	Shlaes et al., 1999 (68)	development	USA
CFSAT (Chronic Fatigue Syndrome Attitudes Test)	Friedberg et al., 2008 (69)	outcome only	USA
CFS-KAB (Chronic Fatigue Syndrome – Knowledge Attitudes and Behaviour)	Brimmer et al., 2010 (70)	development	USA
Challenges and Barriers in Diagnosing, Treating and Relating to Patients with Fibromyalgia†	Hayes et al., 2010 (71)	development	Canada
Chronic Pain and Distress among Elderly in the Community†	Hall-Lord et al., 1999 (72)	development	Sweden
Clinicians' perception towards FND†	Medina et al., 2021 (66)	development	USA
COBS-Adapted (City of Boston's Rehabilitation Professionals' Knowledge and Attitude Survey Regarding Pain – adapted)	Rochman et al., 2013 (73)	development	USA
CPMS-E (Chronic Pain Myth Scale – English)	Martorella et al., 2019a (74)	development	USA
	Martorella et al., 2019b (75)	outcome only	USA
CPSS-HCP (Chronic Pain Stigma Scale – Health Care Providers)	Betsch et al., 2017 (76)	development	Canada

[1] The Brief Illness Perception Questionnaire and Illness Perception Questionnaire Revised are questionnaire instruments designed with patients as target population. When there was no information about adaptation to a healthcare professional point of view and no validation of measurement properties, we decided to treat these as outcome only publications.

[2] In these publications the Brief Illness Perception Questionnaire were independently modified from the patient's to the physician's point of view. Given the different cultural contexts in which they were adapted and little information about how they were adapted, we have treated these as different instrument versions.

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
psychiatrists	Psychogenic nonepileptic seizures	stereotype	illness perceptions
psychiatrists; neurologists	Functional neurological disorders	stereotype	illness perceptions
rheumatologists	Fibromyalgia	stereotype	illness perceptions
rheumatologists	Fibromyalgia	stereotype	illness perceptions
psychology students	Chronic Fatigue Syndrome / Myalgic Encephalomyelitis	stereotype, prejudice, discrimination	attitudes
medical students	Chronic Fatigue Syndrome / Myalgic Encephalomyelitis	stereotype, prejudice, discrimination	attitudes
physicians; nurse practitioners; physician assistants; occupational therapists; nurses	Chronic Fatigue Syndrome	stereotype	knowledge, attitudes, and beliefs
general practitioners; specialists (anesthesiologists, neurologists, psychiatrists and rheumatologists)	Fibromyalgia	stereotype, prejudice	challenges and barriers in diagnosing, treating and relating to patients
nurses	Chronic pain	stereotype	assessment of pain and distress
psychiatrists; neurologists	Functional neurological disorders	stereotype, prejudice	perception
occupational therapists	Chronic pain	stereotype	knowledge
nurses	Chronic pain	stereotype	knowledge, beliefs, and attitudes
nurses	Chronic pain	stereotype	knowledge, beliefs, and attitudes
physiotherapists; pediatricians; psychologist	Fibromyalgia/ chronic widespread pain	stereotype, prejudice	pain stigma

Table 1: Continued

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
DDPRQ-10 (Difficult Doctor–Patient Relationship Questionnaire)	Aloush et al., 2021 (67)	outcome only	Israel
	Hahn 2001 (77)	outcome only	USA
GP Attitudes to CFS/ME†	Bowen et al., 2005 (78)	outcome only	UK
General Practitioners' Views on Reattribution for Patients with Medically Unexplained Symptoms†	Dowrick et al., 2008 (79)	development	UK
HC-PAIRS-15 (Health Care Providers' Pain and Impairment Relationship Scale)	Rainville et al., 1995 (80)	development	USA
	Rainville et al., 2000 (81)	validation	USA
	Macdonald et al., 2018 (82)	validation	UK
	Moran et al., 2017 (83)	validation	New Zealand
	Latimer et al., 2004 (84)	outcome only	Australia
	Louw et al., 2019 (85)	outcome only	USA
	Morris et al., 2012 (86)	outcome only	UK
	Rankin et al., 2018 (87)	outcome only	Sweden; Australia
	Ryan et al., 2010 (88)	outcome only	UK
	Springer et al., 2018 (89)	outcome only	Israel
	Burnett et al., 2009 (90)	validation	Australia; Taiwan; Singapore
HC-PAIRS-15 (Health Care Providers' Pain and Impairment Relationship Scale)	Domenech et al., 2013 (91)	validation	Spain
	Van Biesen and Alvarez 2020 (92)	outcome only	Spain
	Ferreira et al., 2004 (93)	validation	Australia; Brazil
	de Jesus-Moraleida et al., 2021 (94)	outcome only	Brazil
	Magalhães et al., 2011 (95)	validation	Brazil
	Magalhães et al., 2012 (96)	outcome only	Brazil

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
rheumatologists	Fibromyalgia	stereotype, prejudice	difficulty in doctor patient relationship
primary care physicians	Physical symptoms	stereotype, prejudice	difficulty in doctor patient relationship
general practitioners	Chronic Fatigue Syndrome / Myalgic Encephalomyelitis	stereotype, prejudice, discrimination	attitudes and knowledge
general practitioners; nurse prescribers	Medically unexplained symptoms	stereotype, prejudice	views on reattribution
physical therapists; occupational therapists; nurses; physicians; psychologists; counsellors; excercise therapists	Chronic low back pain	stereotype	attitudes and beliefs
family physicians; orthopedic surgeons	Chronic low back pain	stereotype	attitudes and beliefs
osteopaths	Chronic low back pain	stereotype	attitudes and beliefs
osteopaths; physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
nurses; physicians; psychologists; pharmacists; social workers; physical therapists; occupational therapist	Chronic pain	stereotype, prejudice	attitudes and beliefs
medical students	Chronic low back pain	stereotype	attitudes and beliefs
medical students	Chronic pain	stereotype	attitudes and beliefs
physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
nursing students; physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapy students; family physicians	Chronic low back pain	stereotype	attitudes and beliefs
osteopaths	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
medical students; physiotherapy students; nursing students; pharmacy students	Chronic low back pain	stereotype	attitudes and beliefs
physical therapists	Chronic low back pain	stereotype	attitudes and beliefs
physical therapists	Chronic low back pain	stereotype	attitudes and beliefs

Table 1: Continued

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
HC-PAIRS-13 (Health Care Providers' Pain and Impairment Relationship Scale)	Briggs et al., 2013 (97)	outcome only	Australia
	Chance-Larsen et al., 2020 (98)	outcome only	UK
	Colleary et al., 2017 (99)	outcome only	UK; Ireland
	Cross et al., 2014 (100)	outcome only	New Zealand
	Jacobs et al., 2013 (101)	outcome only	UK
	Rufa et al., 2021 (102)	outcome only	USA
	Houben et al., 2004 (103)	validation	Netherlands
	Epstein-Sher et al., 2017 (104)	outcome only	Israel
	Roitenberg 2019a (105)	validation	Israel
	Roitenberg 2019b (106)	outcome only	Israel
HC-PAIRS-12 (Health Care Providers' Pain and Impairment Relationship Scale)	Caner Aksoy et al., 2021 (107)	validation	Turkey
IPQ-R[2] (Illness Perception Questionnaire Revised)	Dickman et al., 2011 (108)	outcome only	Israel
	Levy et al., 2014 (109)	outcome only	Israel
IPQ-R-PNES[†] (Illness Perception Questionnaire Revised - psychogenic nonepileptic seizures)	Whitehead and Reuber 2012 (110)	development	UK
	Whitehead et al., 2013 (111)	validation	UK
Knowledge and Attitude Statements About Back Pain[†]	Buchbinder et al., 2001 (112)	development	Australia
	Buchbinder and Jolley 2007 (113)	outcome only	Australia
Knowledge Attitudes and Perceptions Regarding Irritable Bowel Syndrome and Treatment[†]	Purdy 2017 (114)	development	USA
Likeability of Headaches and Other Neurological Disorders[†]	Evans and Evans 2010 (115)	development	USA
MA-S (Medical Authoritarianism Scale)	Burgess et al., 2011 (116)	development	USA
MCRS (Medical Condition Regard Scale)	Christison et al., 2002 (117)	development	USA
	Hirsh et al., 2014 (118)	outcome only	USA

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
chiropractor students; medical students; occupational therapy students; pharmacy students; physiotherapy students	Low back pain	stereotype	attitudes and beliefs
physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapy students	Chronic low back pain	stereotype	attitudes and beliefs
occupational therapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
physical therapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapists; manual therapists; chiropractors; Cesar therapists)	Chronic low back pain	stereotype	attitudes and beliefs
primary care practitioners	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
physiotherapists	Chronic low back pain	stereotype	attitudes and beliefs
gastroenterologists; nurses	Irritable Bowel Syndrome	stereotype	illness perceptions
gastroenterologists, nurses	Irritable Bowel Syndrome	stereotype	illness perceptions
neurologist	Psychogenic nonepileptic seizures	stereotype	illness perceptions
neurologists; psychiatrists	Nonepileptic attack disorder	stereotype	illness perceptions
general practitioners	Back pain	stereotype, prejudice, discrimination	beliefs
general practitioners	Back pain	stereotype, prejudice, discrimination	beliefs
nurses	Irritable Bowel Syndrome	stereotype, prejudice	knowledge, perceptions, attitudes
neurologists	psychogenic (functional) neurological disorders	prejudice	likeability
general internal medicine physicians	Chronic pain	stereotype, prejudice, discrimination	medical authoritarianism
medical students	Somatoform conditions	stereotype, prejudice	medical condition regard
mix of medical specialists, medical students	Chronic pain	stereotype, prejudice	medical condition regard

Table 1: Continued

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
MUS-Q (Attitudes of Paediatric Staff to Caring for Children with Medically Unexplained Symptoms)	Glazebrook et al., 2009 (119)	development	UK
NKASRP-P (Nurses' Knowledge and Attitude Survey Regarding Pain – Paramedics)	Pocock 2013 (120)	development	UK
Nurses' Perceptions of Irritable Bowel Syndrome (IBS) and Sufferers of IBS†	Letson and Dancey 1996 (121)	development	UK
Nurses' Perceptions of Patients with IBS†	Chen and McCutcheon 2001 (122)	development	Taiwan
Nurses' Perception to Chronic Pain†	Desai and Chaturvedi 2012 (123)	development	India
OCPPS (Orientation to Chronic Pain Patients Scale)	Evans et al., 2011 (124)	outcome only	USA
PBBQ (Pharmacists' Back Beliefs Questionnaire)	Abdel Shaheed et al., 2015 (125)	outcome only	Australia
Perceived difficulty of various patients with headache†	van Wilgen et al., 2013 (126)	development	Netherlands
Persistent Pain Attitude Questionnaire†	Weiner et al., 2002 (127)	development	USA
Physician Attitudes and Beliefs related to Patients with Psychogenic Nonepileptic Spells†	Shneker and Elliott, 2008 (128)	development	USA
Physicians' Attitudes Regarding CFS/FM†	Lu et al., 2007 (129)	development	USA
Physicians' Opinions about Functional Seizures (Psychogenic Nonepileptic Seizures) †	Dastgheib et al., 2020 (130)	development	Iran
Practice Impact Statements†	Louw et al., 2019 (85)	outcome only	USA
Provider Perceptions of Patients with SSRDs†	Malas et al., 2018 (131)	development	USA
Self-Efficacy and Attitudes among Pediatricians in Managing Medically Unexplained Physical Symptoms†	Broekhuizen van Henten et al., 2015 (132)	development	Netherlands
Questionnaire for General Practitioners about Chronic Fatigue Syndrome†	Prins et al., 2000 (133)	development	Netherlands

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
mix of paediatrician professionals	Medically unexplained symptoms	stereotype, prejudice	attitudes
paramedics	Pain	stereotype, discrimination	attitudes
nurses	Irritable Bowel Syndrome	stereotype, prejudice	perceptions
nurses	Irritable Bowel Syndrome	stereotype, prejudice	knowledge, perceptions, beliefs and learning needs
nurses	Chronic pain	stereotype	perception
primary care physicians	Chronic pain	stereotype, prejudice	attitudes
pharmacists	Low back pain	stereotype, prejudice	knowledge, attitudes and beliefs
psychologists; physicians; physical therapists; psychology students	Somatic symptoms without medical explanation	prejudice	difficulty in doctor patient relationship
nurses; nursing assistants	Persistent pain	prejudice	attitudes
primary care physicians; emergency physicians; internal medicine physicians	Psychogenic nonepileptic spells	stereotype	attitudes and beliefs
medical students	Chronic Fatigue Syndrome / Myalgic Encephalomyelitis; Fibromyalgia	stereotype, prejudice	perceptions and attitudes
neurologists; psychiatrists	functional seizures (psychogenic nonepileptic seizures)	stereotype	opinions
nurses; physicians; psychologists; pharmacists; social workers; physical therapists; occupational therapists	Chronic pain	stereotype, prejudice	attitudes and beliefs
primary care pediatricians, family medicine physicians, primary care nurse practitioners, primary care physician assistants	Somatic Symptom and Related Disorders	stereotype, prejudice	perceptions
pediatricians	Medically unexplained physical symptoms	stereotype, prejudice	self-efficacy and attitudes
general practitioners	Chronic Fatigue Syndrome / Myalgic Encephalomyelitis	stereotype, prejudice	doctor-patient relationship

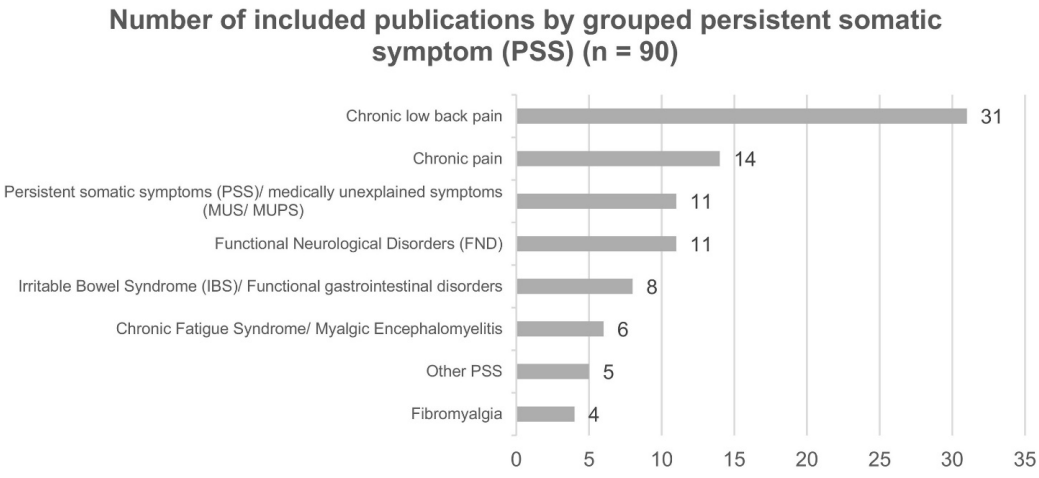
Table 1: Continued

Measurement instrument († = no name of instrument provided in publication)	**Authors**	**Type of publication**	**Country of publication**
Questionnaire on Beliefs and Experiences about the Treatment of Chronic Pain in Emergency Departments (EDs) †	Wilsey et al., 2008 (134)	development	USA
Understanding and Management of Conversion Disorder - Psychiatrists†	Dent et al., 2020 (135)	development	UK; Australia
Understanding and Management of Conversion Disorder - Neurologists†	Kanaan et al., 2011 (136)	development	UK
VASC (Visual Analogue Sympathy Scale)	Bestch et al., 2017 (76)	outcome only	Canada
Views about Functional Neurological Disorders†	Lehn et al., 2019 (137)	development	Australia
Willingness to Accept Patients with FM†	Homma et al., 2016 (49)	outcome only	Japan

The majority of included publications focussed on medical specialists (mostly internal medicine specialists, neurologists, psychiatrists and pediatricians), physical therapists and general practitioners **(Table 1)**. A wide range of PSS were studied, with the highest number of publications focusing on chronic low back pain **(Fig. 2)**. Stereotypes were the most studied stigma mechanism, with 92% of measurement instruments including at least one relevant item. Prejudices were explored in 52% of instruments, and discrimination in 19% of instruments. Most publications did not explicitly name stigma as their construct of interest, but rather related constructs such as attitudes, beliefs, perceptions towards patients, intended behaviours and management of patients.

Healthcare professional population	Persistent somatic symptom	Stigma mechanisms assessed	Construct(s)
emergency department physicians; nurses	Chronic pain	stereotype, prejudice, discrimination	attitudes and beliefs
psychiatrists	Conversion disorder	stereotype	understanding and management
neurologists	Conversion disorder	stereotype	understanding and management
physiotherapists; pediatricians; psychologists	Fibromyalgia	stereotype, prejudice	sympathy
neurologists; psychiatrists; psychologist; general practices; nurses; physiotherapists	Functional neurological disorders	stereotype, prejudice, discrimination	views
rheumatologists	Fibromyalgia	stereotype, prejudice, discrimination	willingness to accept patients

Figure 2: Number of included publications by grouped persistent somatic symptom



3.1. Methodological quality of publications (risk of bias)

The development of a measurement instrument was described in 47 publications. Construct validity, structural validity, internal consistency and reliability were the most commonly investigated measurement properties. Three publications evaluated measurement error, and only one publication measured responsiveness. There were no content validity publications.

The development of measurement instruments in these publications was rated as doubtful in 10 publications, and inadequate in 37 publications. New instruments were typically developed through a literature review and approved by an unclear number of reviewers before testing in the target population. Most instruments were not clearly named. While some publications described involvement of patients and healthcare professionals in the concept elicitation process, these were in insufficient detail to make a judgement of their appropriateness. Only one publication clearly described qualitative research methods during development (120). Some publications described a piloting process, but it was unclear if the comprehensiveness and comprehensibility of the instrument were assessed. Inadequate scores were assigned because of a lack of clarity about the construct being measured, lack of qualitative methods in the concept elicitation process, or lack of pilot testing. It was notable that most development publications (32 out of 47) sought to develop a measurement instrument and use it to assess PSS stigmatisation by healthcare professionals without evaluating any measurement properties.

The methodological quality of structural validity studies was rated as very good in five publications, adequate in seven publications, doubtful in one publication and inadequate in one publication. In some cases, exploratory factor analysis was used when confirmatory analysis would have been more appropriate. Internal consistency studies were rated as very good in 16 publications, doubtful in six publications, and inadequate in one publication. Publications received a score of doubtful when it was unclear whether the construct was unidimensional.

Assessment of construct validity through hypothesis testing was rated as very good in three publications, adequate in six publications and doubtful in five publications. Reasons for doubtful scores included a lack of explicit hypotheses, not making the characteristics of comparator groups clear and not displaying results clearly across the comparator groups.

The methodological quality of cross-cultural validity studies was rated as doubtful in two publications, due to relatively small sample sizes in each group, or the lack of explanation about how differences in samples might affect instrument scores. Assessments of other measurement properties that scored doubtful or inadequate either had a small study population or had other methodological limitations. A full table of the methodological quality of the development and validation publications is shown in Appendix 4 (supplementary data).

3.2. Application of good criteria for measurement properties and quality of evidence

The results for the quality of measurement properties and the quality of the total evidence are shown in **Table 2**.

3.2.1. Content validity

Content validity was inconsistent or indeterminate across all instruments, at low or very low quality level of evidence due to a lack of specific content validity studies. Only the CAT (68) and the MCRS (117) had both sufficient relevance and comprehensiveness based on face validity. Only eight instruments had sufficient relevance, defined by at least 85% of items being considered relevant to one of the stigma mechanisms based on face validity (66,68,74,(115), (116), (117),129,131). Only eight instruments had sufficient comprehensiveness at least one item focusing on each mechanism of stereotypes, prejudices and discrimination (51,59,62,68,112,117,134,137). Only one instrument had sufficient comprehensibility (120). Comprehensibility was mostly indeterminate due to lack of reported involvement of healthcare professionals during instrument development. Evidence around each component of content validity is described in more detail in Appendix 5 (supplementary data). Results for other measurement properties with a risk of bias with a score of 'adequate' or better are described below in more detail.

Table 2: Quality of evidence for measurement properties of measurement instruments

Measurement instrument († = no name of instrument provided in publication)	**Content validity**		**Structural validity**		**Internal consistency**	
	Overall rating	**Quality of evidence**	**Overall rating**	**Quality of evidence**	**Overall rating**	**Quality of evidence**
3*I-P-J	?	Very low	?	High	+	High
Attitudes of Nurses Toward Patients With Conversion/Functional Symptoms†	?	Very low			+	Low
Attitudes Towards Cause and Management of Medically Unexplained Symptoms†	±	Low				
Attitudes and Knowledge about Irritable Bowel Syndrome†	±	Very low				
Attitudes and Practices in the Evaluation and Treatment of Irritable Bowel Syndrome†	±	Very low				
Attitudes of General Practitioners (GPs) towards Somatising Patients†	±	Very low				
Attitudes to Patients with a Self Diagnosis of Myalgic Encephalomyelitis†	±	Very low				
Attitudes towards Somatising Patients†	±	Very low				
Back Pain Advice and Pharmacists' Experience of Back Pain†	±	Very low				
Barriers for Primary Care Physicians in Diagnosis and Treatment of Persistent Somatic Symptoms†	±	Low				
Beliefs and Attitudes in GPs in the Management of Mechanical Low Back Pain†	±	Very low				
Beliefs and Attitudes of General Pediatricians and Pediatric Gastroenterologists Regarding Functional Gastrointestinal Disorders†	±	Very low				
B-IPQ-P-I	±	Very low				
BIPQ-P-J	±	Very low	?	Moderate	?	Low
CAT	±	Very low	?	Moderate	?	High
CFS-KAB	±	Very low	?	Moderate	-	High

	Cross-cultural validity		Reliability		Measurement error		Construct validity		Responsiveness	
	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence
			?	Low			+	Low		

Table 2: Continued

Measurement instrument († = no name of instrument provided in publication)	**Content validity**		**Structural validity**		**Internal consistency**	
	Overall rating	**Quality of evidence**	**Overall rating**	**Quality of evidence**	**Overall rating**	**Quality of evidence**
Challenges and Barriers in Diagnosing, Treating and Relating to Patients with Fibromyalgia†	±	Very low				
Chronic Pain and Distress among Elderly in the Community†	?	Very low				
Clinicians' perception towards FND†	±	Very low				
COBS-Adapted†	?	Very low				
CPMS-E	±	Low	?	Moderate	-	High
CPSS-HCP	?	Very low			+	Very low
General Practitioners' Views on Reattribution for Patients with Medically Unexplained Symptoms†	±	Very low				
HC-PAIRS-15	±	Very low	?	Moderate	+	High
HC-PAIRS-13	±	Very low	+	High	+	High
HC-PAIRS-12	±	Very low	+	High	+	High
IPQ-R-PNES	±	Low			-	High
Knowledge and Attitude Statements About Back Pain†	±	Very low				
Knowledge Attitudes and Perceptions Regarding Irritable Bowel Syndrome and Treatment†	±	Very low			-	Moderate
Likeability of Headaches and Other Neurological Disorders†	±	Very low				
MA-S	±	Very low			+	Low
MCRS	±	Low	+	High	+	High
MUS-Q	±	Low	?	Very low	-	HIgh
NKASRP-P	±	Low				
Nurses' Perceptions of Irritable Bowel Syndrome (IBS) and Sufferers of IBS†	±	Very low				
Nurses' Perceptions of Patients with IBS†	?	Low				
Nurses' Perception to Chronic Pain†	?	Very low				

	Cross-cultural validity		Reliability		Measurement error		Construct validity		Responsiveness	
	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence
							+	Moderate		
							+	Very low		
	±	Low	+	Moderate	?	High	+	High	+	High
							+	High		
			+	High	?	High	+	High		
							+	Low		
			?	Moderate						

Table 2: Continued

Measurement instrument († = no name of instrument provided in publication)	**Content validity**		**Structural validity**		**Internal consistency**	
	Overall rating	**Quality of evidence**	**Overall rating**	**Quality of evidence**	**Overall rating**	**Quality of evidence**
Perceived difficulty of various patients with headache†	?	Very low	-	Low	-	Low
Persistent Pain Attitude Questionnaire†	?	Very low				
Physician Attitudes and Beliefs Related to Patients with Psychogenic Nonepileptic Spells†	±	Very low				
Physicians' Attitudes Regarding CFS/FM†	±	Very low				
Physicians' Opinions about Functional Seizures (Psychogenic Nonepileptic Seizures) †	±	Very low				
Provider Perceptions of Patients With SSRDs†	±	Low				
Self-Efficacy and Attitudes Among Pediatricians in Managing Medically Unexplained Physical Symptoms†	±	Very low				
Questionnaire for General Practitioners about Chronic Fatigue Syndrome†	±	Very low				
Questionnaire on Beliefs and Experiences about the Treatment of Chronic Pain in Emergency Departments (EDs) †	+	Very low				
Understanding and Management of Conversion Disorder - Psychiatrists†	±	Low				
Understanding and Management of Conversion Disorder - Neurologists†	±	Very low				
Views about functional neurological disorders†	±	Very low	?	Moderate		

+ = sufficient; - = insufficient; ? = indeterminate; ± = inconsistent

	Cross-cultural validity		Reliability		Measurement error		Construct validity		Responsiveness	
	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence	Overall rating	Quality of evidence
			?	Very low						
							+	Low		

3.2.2. Structural validity and internal consistency

For the HC-PAIRS-13 (103,105), HC-PAIRS-12 (107) and MCRS (117) there was a high level of evidence for sufficient structural validity. For the 3*I-P-J, there was indeterminate evidence for structural validity. Reasons for indeterminate evidence for structural validity included a lack of information provided about the analysis, or only exploratory factor analysis/ principal components analysis conducted. There was a high level of evidence for sufficient internal consistency for the 3*I-P-J (49), HC-PAIRS-15 (80,82,83,90,91,95), HC-PAIRS-13 (103,105) and HC-PAIRS-12 (107). For the MCRS, there was a low level of evidence for sufficient structural validity and internal consistency (117). For the CPMS-E (74), CFS-KAB (70) and IPQ-R-PNES (110,111) there was a high level of evidence for insufficient internal consistency. For the Knowledge Attitudes and Perceptions Regarding Irritable Bowel Syndrome and Treatment scale (114) there was a moderate level of evidence for insufficient internal consistency. The reason for insufficient internal consistency was that at least one sub-scale had a Cronbach's alpha of less than 0.7.

3.2.3. Reliability (test-retest)

There was a high level of evidence for sufficient reliability of the HC-PAIRS-12 (107), and a moderate level of evidence for sufficient reliability of the HC-PAIRS-15 (81,83,91,95). There was a low level of evidence for sufficient reliability of the MCRS (117). There was indeterminate evidence for reliability of the Persistent Pain Attitude Questionnaire (127).

3.2.4. Measurement error

The only instruments measuring measurement error were the HC-PAIRS-15 (83,95) and the HC-PAIRS-12 (107), for which there was indeterminate evidence. This is because we were not able to judge whether the Smallest Detectable Change (SDC) or Limits of Agreement (LoA) were smaller than the Minimal Important Change (MIC).

3.2.5. Hypothesis testing for construct validity

There was a moderate level of evidence for sufficient convergent validity of the HC-PAIRS-15 (83,90,91,95), HC-PAIRS-13 (103,105), HC-PAIRS-12 (107) and the CAT (68). There was a moderate level of evidence for sufficient known-groups validity of the CPMS-E (74).

3.2.6. Responsiveness

There was a low level of evidence for sufficient responsiveness of the HC-PAIRS-15 (91).

3.2.7. Interpretability and feasability

There was very little information reported about interpretability of measurement instruments. No floor or ceiling effects were reported for the HC-PAIRS-12 (107). For the other instruments, floor and ceiling effects, MIC, and subgroup scores were not reported. There was no information reported about the feasibility of measurement instruments.

4. Discussion

4.1. Main results

Although we found that 62 questionnaire measurement instruments have been used to assess PSS related stigmatisation by healthcare professionals, most of these instruments were developed for use within a single publication, with insufficient evidence of their development or establishment of their content validity. There is a general absence of assessment of measurement properties, and for no single instrument acceptable evidence on all measurement properties was found. Therefore, there is currently no questionnaire instrument that we can recommend for assessing PSS related stigmatisation by healthcare professionals.

We did not find a single instrument with sufficient evidence of content validity relating to stigma. This is not surprising considering that only one of these instruments explicitly named stigma as their construct of interest (76). Instead, we found that the instruments measured a variety of related constructs, which were rarely adequately defined. Further, there was little evidence that healthcare professionals and patients were involved in instrument development. This is problematic because stigma instruments should reflect the challenges that healthcare professionals face when working with patients with PSS. They should also reflect the experiences that patients with PSS have widely reported during clinical encounters. Further, for many instruments it has not been tested if the professionals interpret the items as intended (comprehensibility). The lack of content validity of existing instruments is particularly problematic given that many

publications in this review measured the effectiveness of an anti-stigma intervention (such as an educational programme). The use of unvalidated measurement instruments means that it cannot be certain their results are valid.

4.2. Comparison with the literature

Among identified instruments, the HC-PAIRS instruments (HC-PAIRS-15, HC-PAIRS-13, HC-PAIRS-12) were the most established, used among the largest number of healthcare professionals. These had sufficient evidence for several measurement properties. This is a similar finding to a previous review conducted about instruments exploring attitudes and beliefs about low back pain (138). While the HC-PAIRS instruments were designed to explore beliefs about low back pain, they largely explore beliefs about the ability of patients to function (continuing daily activities, managing pain) rather than stigmatising attitudes about these patients. Given a lack of content validity relating to stigma, we would not recommend this instrument for further stigma research.

It is commendable that there are so many publications exploring stigmatising attitudes and behaviours of healthcare professionals, across several decades and covering most major clinical professions. This demonstrates the enduring interest of stigma and commitment to tackling it. These publications provide practical examples of professional assumptions about patient behaviour which have been challenged. For example, while healthcare professionals frequently reported challenges in communication, analysis of linguistic markers found no systematic variations between patients with a clear pathology and those without (139). Rather, professionals appear to adapt their language to patients with PSS (140,141). Healthcare professionals also reported feeling pressure from patients to conduct testing and make specialist referrals, even though in practice this further action was mostly suggested first by professionals (27,28,142). While many of these publications developed instruments before COSMIN guidelines were available, this does not explain the lack of explanation of constructs being explored, lack of patient and healthcare professional involvement in developing these instruments, and lack of validation. A lack of connection between similar publications has resulted in a fragmented research landscape.

This inconsistency is not unique to PSS related stigma. A previous systematic review for mental health related stigma among healthcare professionals found that there were no instruments that presented strong levels of evidence for all measurement properties (42). In reviews of health stigma instruments, the authors concluded that more generic measures of stigma should be used because the outcomes of stigma are similar across different health conditions (143,144). This has been echoed in a recent review for functional neurological disorder related stigma (145). The main advantage of using a generic instrument is to more easily compare scores across health conditions. This would enable researchers to compare stigma across different PSS, for example where there is additional stigma associated with specific symptoms (146), or to compare PSS related stigma to comparable health conditions (11) or even to other types of health stigma such as mental health stigma (147).

4.3. Strengths and limitations

A major strength of our publication is that we performed a systematic review according to PRISMA guidelines with extensive database searches. We conducted independent selection and assessment of included publications with a diversity of male/ female reviewers and academic perspectives (medical sociology and anthropology, general practice, epidemiology and biomedical sciences). Furthermore, we followed established COSMIN taxonomy and guidance in describing and assessing the measurement properties of instruments. The COSMIN methodology provides a highly structured and systematic way to assess the quality of available stigma instruments.

We found that the inclusion criteria of a questionnaire measurement instrument were challenging to administer. Rather than a clear binary of self-reported questionnaires or behaviour-based instruments, measurement instruments existed on a spectrum between these (often facilitated by experimental methods in the research design). Although we reached consensus on these instruments through discussion between reviewers, a different operationalisation of questionnaire measurement instruments could have led to different publications included.

We are aware that there may be differences in opinion around the naming, version history and categorisation of included measurement instruments. This may have affected our results when summarising results for

measurement properties (for example, if there are few publications for a particular version of an instrument, this will result in a lower graded quality of evidence). However, we think the likely impact of this was negligible.

Lastly, despite developing a comprehensive search strategy and searching several databases, it is possible that not all relevant measurement instruments were identified. Similarly, contacting authors of all included publications may have resulted in additional information about measurement properties. However, we are confident that our strategy was sufficiently extensive to make valid conclusions about PSS related stigma questionnaire measurement instruments.

4.4. Consequences for further research

Considerable work is still needed in this field to be confident of a high quality stigma questionnaire measurement instrument for healthcare professionals. This could be done through validation of the instruments with both sufficient relevance and comprehensiveness relating to content validity (the CAT and the MCRS), validation of other health stigma instruments in a PSS context, or the development and validation of a new PSS related stigma questionnaire instrument. Our recommendation is the development of a generic PSS related stigma instrument using examples here for item generation and hypothesis testing for convergent validity. Establishing sufficient content validity should be the priority, with a clear stigma model defined from the outset.

A key assumption about content validity of self-reported measurement instruments is that it can be assessed by asking healthcare professionals about the relevance, comprehensiveness and comprehensibility of items. This is to say that that participants are epistemologically authoritative; they are the experts in their own experience and are able to reflect on that. There is increasing evidence to suggest that negative attitudes and behaviours can be expressed in more indirect and subtle ways. This is despite people regarding themselves as non-stigmatising, or in some cases actively eschewing explicit forms of discrimination (148). This raises the question as to whether as potential stigmatisers, healthcare professional involvement is enough on its own to inform content validity. Existing COSMIN resources give sufficient insight into how this can be achieved. For development of stigma instruments with sufficient content validity, a combination of meaningful involvement of healthcare professionals and also patients is

needed. Theoretical insights from stigma research should both shape initial development and supplement this involvement. We encourage researchers to follow COSMIN guidelines such as their study design checklist when designing their research (149). Further, we believe that a positive framing looking to understand the challenges of healthcare professionals would lessen the incentive for socially desirable answers, while also providing a more honest reflection of the aim of the research.

This raises important limitations of self-reported instruments. Limitations include people not explicitly endorsing negative attitudes, or preferring not to reveal their attitudes through a social desirability bias (150). This is particularly important when there are social pressures against explicit stigmatising behaviour, such as the risk of professional liability. Other types of stigma measurement instruments also exist, for example instruments which aim to reveal stigmatising behaviours. Examples of these behaviour-based instruments include response latency techniques (e.g. through a type of Implicit Association Test (151)), or analysis of stigmatising behaviours during clinical consultation (140). Therefore, particular focus is needed on how discrimination can be explored through suitable items, and how results from self-reported instruments can be compared to behaviour-based instruments.

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Supplementary material

Appendix 1: Search strategy

Search strategy: #1 healthcare professionals AND #2 persistent somatic symptoms AND #3 stigma measurement

PubMed (syntax adaptations for other databases available on online supplementary material)

(Health Personnel [Mesh] OR professional*[tw] OR doctor*[tw] OR personnel[tw] OR physician*[tw] OR clinician*[tw] OR practitioner*[tw] OR medical[tw] OR nurs*[tw] OR provider[tw] OR GP[tw] OR GPs'[tw]) AND (Somatoform disorders[Mesh] OR Hypochondriasis[Mesh] OR Neurasthenia[Mesh] OR Conversion disorder[Mesh] OR Psychophysiological disorder[Mesh] OR Psychosomatic medicine[Mesh] OR "Fibromyalgia"[Mesh] OR "Fatigue Syndrome, Chronic"[Mesh] OR "Irritable Bowel Syndrome"[Mesh] OR "Colonic diseases, Functional" [MESH] OR "Pelvic Pain"[Mesh] OR "Tension-Type Headache"[Mesh] OR "Cumulative Trauma Disorders"[Mesh] OR "Whiplash Injuries"[Mesh] OR "Back Pain"[Mesh] OR "Neck Pain"[Mesh] OR "Chronic Pain"[Mesh] OR "Multiple Chemical Sensitivity"[Mesh] OR "Tinnitus"[Mesh] OR "Burning Mouth Syndrome"[Mesh] OR "Premenstrual Syndrome"[Mesh] OR "Dizziness"[Mesh] OR "Temporomandibular Joint Disorders"[Mesh] OR somatization[tw] OR somatisation[tw] OR ((somatoform[tw] AND (disorder*[tw] OR pain[tw] OR symptom*[tw] OR syndrome*[tw] OR illness[tw]))) OR hypochondriasis[tw] OR neurasthen*[tw] OR conversion disorder*[tw] OR psychophysiological disorder*[tw] OR psychosomat*[tw] OR (((functional[tw] OR unexplained[tw]) AND somatic sympt*[tw])) OR functional somatic syndrom*[tw] OR functional syndrom*[tw] OR FSS[tw] OR functional neurological disorder[tw] OR FND[tw] OR unexplained sympt*[tw] OR medically unexplained[tw] OR unexplained medical sympt*[tw] OR unexplained physical symptom*[tw] OR MUS[tw] OR MUPS[tw] OR psychogen*[tw] OR non-organ*[tw] OR non-specific complain*[tw] OR non-specific sympt*[tw] OR fibromyalgia[tw] OR fibrositis[tw] OR "fatigue syndrome"[tw] OR effort syndrome[tw] OR "chronic epstein barr virus"[tw] OR myalgic encephalopathy*[tw] OR myalgic encephalomyelitis*[tw] OR post viral fatigue syndrome*[tw] OR post viral syndrome*[tw] OR postviral syndrome*[tw] OR post infectious fatigue*[tw] OR postinfectious fatigue[tw] OR "irritable bowel*"[tw] OR IBS[tw] OR ((irritable[tw] AND (bowel*[tw] OR colon[tw]))) OR "functional bowel disease*"[tw] OR "functional colonic

disease*"[tw] OR functional urinary disorder*[tw] OR urethral syndrome*[tw] OR micturition dysfunction*[tw] OR irritable bladder*[tw] OR "pelvic pain"[tw] OR "abdominal pain"[tw] OR "tension-type headache*"[tw] OR "tension headache*"[tw] OR "cumulative trauma disorder*"[tw] OR "carpal tunnel syndrome"[tw] OR "repetitive strain injuries"[tw] OR "repetitive strain injury"[tw] OR RSI[tw] OR whiplash*[tw] OR "back pain"[tw] OR "neck pain"[tw] OR backache[tw] OR "chronic pain"[tw] OR "chronic widespread pain"[tw] OR "multiple chemical sensitivity"[tw] OR (electromagnetic hypersensitivity[tw] OR electro-hypersensitivity[tw] OR electrosensitiv*[tw] OR IEI-EMF[tw] OR environmental illness*[tw]) OR "gulf syndrome"[tw] OR "idiopathic environmental intolerance*"[tw] OR "chemical intolerance"[tw] OR tinnitus[tw] OR ("burning mouth"[tw] AND (syndrome[tw] OR symptom[tw])) OR "burning tongue"[tw] OR (((premenstrual[tw] OR pre-menstrual[tw]) AND (syndrome[tw] OR tension[tw]))) OR dizziness[tw] OR "Temporomandibular Joint Disorder*"[tw] OR (((nonspecific[tw] OR non-specific[tw] OR "non cardiac"[tw] OR noncardiac[tw]) AND "chest pain"[tw])) OR ((nonepileptic[tw] OR "non epileptic"[tw] OR psychogenic[tw]) AND (seizure*[tw] OR attack*[tw]))) AND (Social Stigma [Mesh] OR Prejudice [Mesh] OR Attitude of Health Personnel [Mesh] OR Stereotyping [Mesh] OR stigma*[tw] OR prejudic* [tw] OR discriminat*[tw] OR OR bias*[tw] OR invalidat*[tw])

Appendix 2: Definitions of measurement properties according to the COSMIN group

Reproduced from COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures (PROMs), page 11-12.

For more information, please see:
Mokkink LB, Terwee CB, Patrick DL, Alonso J, Stratford PW, Knol DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. Journal of clinical epidemiology. 2010;63(7):737-45.

Domain	Measurement property	Aspect of a measurement property	Definition
Reliability			The degree to which the measurement is free from measurement error
Reliability (extended definition)			The extent to which scores for patients who have not changed are the same for repeated measurement under several conditions: e.g. using different sets of items from the same PROM (internal consistency); over time (test-retest); by different persons on the same occasion (inter- rater); or by the same persons (i.e. raters or responders) on different occasions (intra-rater)
	Internal consistency		The degree of the interrelatedness among the items
	Reliability		The proportion of the total variance in the measurements which is due to 'true'† differences between patients
	Measurement error		The systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured
Validity			The degree to which a PROM measures the construct(s) it purports to measure
	Content validity		The degree to which the content of a PROM is an adequate reflection of the construct to be measured

Domain	Measurement property	Aspect of a measurement property	Definition
		Face validity	The degree to which (the items of) a PROM indeed looks as though they are an adequate reflection of the construct to be measured
	Construct validity	Hypothesis testing for construct validity	The degree to which the scores of a PROM are consistent with hypotheses (for instance with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups) based on the assumption that the PROM validly measures the construct to be measured
		Structural validity	The degree to which the scores of a PROM are an adequate reflection of the dimensionality of the construct to be measured
		Cross-cultural validity	The degree to which the performance of the items on a translated or culturally adapted PROM are an adequate reflection of the performance of the items of the original version of the PROM
	Criterion validity		The degree to which the scores of a PROM are an adequate reflection of a 'gold standard'
Responsiveness			The ability of a PROM to detect change over time in the construct to be measured
	Responsiveness		Idem responsiveness
Interpretability*			Interpretability is the degree to which one can assign qualitative meaning - that is, clinical or commonly understood connotations – to a PROM's quantitative scores or change in scores.

† The word 'true' must be seen in the context of the CTT, which states that any observation is composed of two components – a true score and error associated with the observation. 'True' is the average score that would be obtained if the scale were given an infinite number of times. It refers only to the consistency of the score, and not to its accuracy (22)

* Interpretability is not considered a measurement property, but an important characteristic of a measurement instrument

Appendix 3: Updated criteria for good measurement properties (and additional guidance for structural validity)

Reproduced from COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures (PROMs), page 28-29.

Measurement property	Rating[a]	Criteria
Structural validity	+	**CTT: CFA:** CFI or TLI or comparable measure >0.95 OR RMSEA <0.08[b] **IRT/Rasch:** No violation of unidimensionality[c]: CFI or TLI or comparable measure >0.95 OR RMSEA < 0.20 OR Q3's < 0.37 AND no violation of monotonicity: adequate looking graphs OR item scalability >0.30 AND adequate model fit: IRT: $\chi 2$ >0.01 Rasch: infit and outfit mean squares ≥ 0.5 and ≤ 1.5 OR Z-standardized values > -2 and <2
	?	**CTT:** Not all information for '+' reported **IRT/Rasch:** Model fit not reported
	-	Criteria for '+' not met
Internal consistency	+	At least low evidence[d] for sufficient structural validity[e] AND Cronbach's alpha(s) ≥ 0.70 for each unidimensional scale or subscale[f]
	?	Criteria for "At least low evidence[d] for sufficient structural validity[e]" not met
	-	At least low evidence[d] for sufficient structural validity[e] AND Cronbach's alpha(s) < 0.70 for each unidimensional scale or subscale[f]
Reliability	+	ICC or weighted Kappa ≥ 0.70
	?	ICC or weighted Kappa not reported
	-	ICC or weighted Kappa < 0.70
Measurement error	+	SDC or LoA < MIC[e]
	?	MIC not defined
	-	SDC or LoA > MIC[e]
Hypotheses testing for construct validity	+	The result is in accordance with the hypothesis[g]
	?	No hypothesis defined (by the review team)
	-	The result is not in accordance with the hypothesis[g]

Measurement property	Rating[a]	Criteria
Cross-cultural validity\measurement invariance	+	No important differences found between group factors (such as age, gender, language) in multiple group factor analysis OR no important DIF for group factors (McFadden's $R^2 < 0.02$)
	?	No multiple group factor analysis OR DIF analysis performed
	-	Important differences between group factors OR DIF was found
Criterion validity	+	Correlation with gold standard ≥ 0.70 OR AUC ≥ 0.70
	?	Not all information for '+' reported
	-	Correlation with gold standard < 0.70 OR AUC < 0.70
Responsiveness	+	The result is in accordance with the hypothesis[h] OR AUC ≥ 0.70
	?	No hypothesis defined (by the review team)
	-	The result is not in accordance with the hypothesis[h] OR AUC < 0.70

Initials used:
AUC = area under the curve, CFA = confirmatory factor analysis, CFI = comparative fit index, CTT = classical test theory, DIF = differential item functioning, ICC = intraclass correlation coefficient, IRT = item response theory, LoA = limits of agreement, MIC = minimal important change, RMSEA: Root Mean Square Error of Approximation, SEM = Standard Error of Measurement, SDC = smallest detectable change, SRMR: Standardized Root Mean Residuals, TLI = Tucker-Lewis index

Notes:
a "+" = sufficient, " –" = insufficient, "?" = indeterminate
b To rate the quality of the summary score, the factor structures should be equal across studies
c Unidimensionality refers to a factor analysis per subscale, while structural validity refers to a factor analysis of a (multidimensional) patient-reported outcome measure
d As defined by grading the evidence according to the GRADE approach
e This evidence may come from different studies
f the criteria 'Cronbach alpha < 0.95' was deleted, as this is relevant in the development phase of a PROM and not when evaluating an existing PROM.
g The results of all studies should be taken together and it should then be decided if 75% of the results are in accordance with the hypotheses

Appendix 4: Quality of studies on measurement properties (risk of bias assessment)

Measurement instrument († = no name of instrument provided in study)	Reference	Instrument development	Structural validity	Internal consistency
3*I-P-J	Homma et al., 2016 (49)	Inadequate	Very good	Very good
Attitudes of Nurses towards Patients With Conversion/ Functional Symptoms†	Ahern et al., 2009 (51)	Inadequate		Doubtful
Attitudes towards Cause and Management of Medically Unexplained Symptoms†	Reid et al., 2001 (15)	Doubtful		
Attitudes and Knowledge about Irritable Bowel Syndrome†	Longstreth and Burchette 2003 (56)	Inadequate		
Attitudes and Practices in the Evaluation and Treatment of Irritable Bowel Syndrome†	Lacy et al., 2006 (57)	Inadequate		
Attitudes of General Practitioners (GPs) towards Somatising Patients†	Garcia-Campayo et al., 1998 (58)	Inadequate		
Attitudes to Patients with a Self Diagnosis of Myalgic Encephalomyelitis†	Scott et al., 1995 (59)	Inadequate		
Attitudes towards Somatising Patients†	Rosendal et al., 2005 (60)	Inadequate		
Back Pain Advice and Pharmacists' Experience of Back Pain†	Silcock et al., 2007 (61)	Inadequate		
Barriers for Primary Care Physicians in Diagnosis and Treatment of Persistent Somatic Symptoms†	Lehmann et al., 2021 (62)	Doubtful		
Beliefs and Attitudes in GPs in the Management of Mechanical Low Back Pain†	Chaudhary et al., 2004 (63)	Inadequate		
Beliefs and Attitudes of General Pediatricians and Pediatric Gastroenterologists Regarding Functional Gastrointestinal Disorders†	Sood et al., 2011 (64)	Inadequate		
B-IPQ-P-I†	Aloush et al., 2021 (67)	Inadequate		
BIPQ-P-J†	Homma et al., 2016 (49)	Inadequate	Adequate	Doubtful
CAT	Shlaes et al., 1999 (68)	Inadequate	Adequate	Very good
CFS-KAB	Brimmer et al., 2010 (70)	Inadequate	Adequate	Very good
Challenges and Barriers in Diagnosing, Treating and Relating to Patients with Fibromyalgia†	Hayes et al., 2010 (71)	Inadequate		

	Cross-cultural validity	**Reliability**	**Measurement error**	**Construct validity** (convergent validity)	**Construct validity** (known groups validity)	**Responsiveness**
		Doubtful		Adequate		

Appendix 4: Continued

Measurement instrument († = no name of instrument provided in study)	Reference	Instrument development	Structural validity	Internal consistency
Chronic Pain and Distress among Elderly in the Community†	Hall-Lord et al., 1999 (72)	Inadequate		
Clinicians' perception towards FND†	Medina et al., 2021 (66)	Inadequate		
COBS-Adapted	Rochman et al., 2013 (73)	Inadequate		
CPMS-E	Martorella et al., 2019a (74)	Doubtful	Adequate	Very good
CPSS-HCP	Betsch et al., 2017 (76)	Inadequate		Doubtful
General Practitioners' Views on Reattribution for Patients with Medically Unexplained Symptoms†	Dowrick et al., 2008 (79)	Inadequate		
HC-PAIRS-15	Rainville et al., 1995 (80)	Inadequate	Adequate	Inadequate
	Rainville et al., 2000 (81)			
	Macdonald et al., 2018 (82)			Very good
	Moran et al., 2017 (83)			Very good
	Burnett et al., 2009 (90)			Doubtful
	Domenech et al., 2013 (91)		Adequate	Very good
	Ferreira et al., 2004 (93)			
	Magalhães et al., 2011 (95)			Very good
HC-PAIRS-13	Houben et al., 2004 (103)		Very good	Very good
	Roitenberg 2019a (105)		Very good	Very good
HC-PAIRS-12	Caner Aksoy et al., 2021 (107)		Very good	Very good
IPQ-R-PNES	Whitehead and Reuber 2012 (110)	Doubtful		Very good
	Whitehead et al., 2013 (111)			Very good
Knowledge and Attitude Statements About Back Pain†	Buchbinder et al., 2001 (112)	Inadequate		
Knowledge Attitudes and Perceptions Regarding Irritable Bowel Syndrome and Treatment†	Purdy 2017 (114)	Inadequate		Very good
Likeability of Headaches and Other Neurological Disorders†	Evans and Evans 2010 (115)	Inadequate		
MA-S	Burgess et al., 2011 (116)	Inadequate		Doubtful
MCRS	Christison et al., 2002 (117)	Doubtful	Very good	Very good
MUS-Q	Glazebrook et al., 2009 (119)	Doubtful	Inadequate	Very good
NKASRP-P	Pocock 2013 (120)	Doubtful		
Nurses' Perceptions of Irritable Bowel Syndrome (IBS) and Sufferers of IBS†	Letson and Dancey 1996 (121)	Inadequate		

	Cross-cultural validity	**Reliability**	**Measurement error**	**Construct validity** (convergent validity)	**Construct validity** (known groups validity)	**Responsiveness**
					Adequate	
					Doubtful	
					Doubtful	
		Adequate				
		Adequate	Adequate	Very good		
	Doubtful	Adequate		Very good	Doubtful	
		Adequate		Adequate		Very good
	Doubtful					
		Adequate	Very good	Adequate		
				Adequate		
				Adequate		
		Very good	Very good	Very good		
					Doubtful	
		Adequate				

Appendix 4: Continued

Measurement instrument († = no name of instrument provided in study)	**Reference**	**Instrument development**	**Structural validity**	**Internal consistency**
Nurses' Perceptions of Patients with IBS†	Chen and McCutcheon 2001 (122)	Doubtful		
Nurses' Perception to Chronic Pain†	Desai and Chaturvedi 2012 (123)	Inadequate		
Perceived difficulty of various patients with headache†	van Wilgen et al., 2013 (126)	Inadequate	Doubtful	Doubtful
Persistent Pain Attitude Questionnaire†	Weiner and Rudy, 2002 (127)	Inadequate		
Physician Attitudes and Beliefs Related to Patients with Psychogenic Nonepileptic Spells†	Shneker and Elliott, 2008 (128)	Inadequate		
Physicians' Attitudes Regarding CFS/FM†	Lu et al., 2007 (129)	Inadequate		
Physicians' Opinions about Functional Seizures (Psychogenic Nonepileptic Seizures) †	Dastgheib et al., 2020 (130)	Inadequate		
Provider Perceptions of Patients With SSRDs†	Malas et al., 2018 (131)	Doubtful		
Self-Efficacy and Attitudes Among Pediatricians in Managing Medically Unexplained Physical Symptoms†	Broekhuijsen-van Henten et al., 2015 (132)	Inadequate		
Questionnaire for General Practitioners about Chronic Fatigue Syndrome†	Prins et al., 2000 (133)	Inadequate		
Questionnaire on Beliefs and Experiences about the Treatment of Chronic Pain in Emergency Departments (EDs) †	Wilsey et al., 2008 (134)	Inadequate		
Understanding and Management of Conversion Disorder - Psychiatrists†	Dent et al., 2020 (135)	Doubtful		
Understanding and Management of Conversion Disorder - Neurologists†	Kanaan et al., 2011 (136)	Inadequate		
Views about functional neurological disorders†	Lehn et al., 2019 (137)	Inadequate	Adequate	

	Cross-cultural validity	**Reliability**	**Measurement error**	**Construct validity** (convergent validity)	**Construct validity** (known groups validity)	**Responsiveness**
		Adequate				
					Doubtful	

Appendix 5: Evaluation of content validity of measurement instruments

Measurement instrument († = no name of instrument provided in study)	**Relevance**	**Comprehen-siveness**	**Comprehen-sibility**	**Content validity**	**Quality of evidence**
3*I-P-J†	?	?	?	?	Very low
Attitudes of Nurses towards Patients With Conversion/Functional Symptoms†	?	+	?	?	Very low
Attitudes towards Cause and Management of Medically Unexplained Symptoms†	±	-	?	±	Low
Attitudes and Knowledge about Irritable Bowel Syndrome†	±	-	?	±	Very low
Attitudes and Practices in the Evaluation and Treatment of Irritable Bowel Syndrome†	±	-	?	±	Very low
Attitudes of General Practitioners (GPs) towards Somatising Patients†	±	-	?	±	Very low
Attitudes to Patients with a Self Diagnosis of Myalgic Encephalomyelitis†	±	+	?	±	Very low
Attitudes towards Somatising Patients†	±	-	?	±	Very low
Back Pain Advice and Pharmacists' Experience of Back Pain†	±	-	?	±	Very low
Barriers for Primary Care Physicians in Diagnosis and Treatment of Persistent Somatic Symptoms†	±	+	?	±	Low
Beliefs and Attitudes in GPs in the Management of Mechanical Low Back Pain†	±	-	?	±	Very low
Beliefs and Attitudes of General Pediatricians and Pediatric Gastroenterologists Regarding Functional Gastrointestinal Disorders†	±	-	?	±	Very low
B-IPQ-P-I†	±	-	?	±	Very low
BIPQ-P-J†	±	-	?	±	Very low
CAT	+	+	?	±	Very low
CFS-KAB	±	-	?	±	Very low

Appendix 5: Continued

Measurement instrument († = no name of instrument provided in study)	**Relevance**	**Comprehen-siveness**	**Comprehen-sibility**	**Content validity**	**Quality of evidence**
Challenges and Barriers in Diagnosing, Treating and Relating to Patients with Fibromyalgia†	±	-	?	±	Very low
Chronic Pain and Distress among Elderly in the Community†	?	?	?	?	Very low
Clinicians' perception towards FND†	+	-	?	±	Very low
COBS-Adapted†	?	?	?	?	Very low
CPMS-E	+	-	?	±	Low
CPSS-HCP	?	?	?	?	Very low
General Practitioners' Views on Reattribution for Patients with Medically Unexplained Symptoms†	±	-	?	±	Very low
HC-PAIRS-15	±	-	?	±	Very low
HC-PAIRS-13	±	-	?	±	Very low
HC-PAIRS-12	±	-	?	±	Very low
IPQ-R-PNES†	±	-	?	±	Low
Knowledge and Attitude Statements About Back Pain†	±	+	?	±	Very low
Knowledge Attitudes and Perceptions Regarding Irritable Bowel Syndrome and Treatment†	±	-	?	±	Very low
Likeability of Headaches and Other Neurological Disorders†	+	-	?	±	Very low
MA-S	+	-	?	±	Very low
MCRS	+	+	?	±	Low
MUS-Q	±	-	?	±	Low
NKASRP-P	±	-	+	±	Low
Nurses' Perceptions of Irritable Bowel Syndrome (IBS) and Sufferers of IBS†	±	-	?	±	Very low
Nurses' Perceptions of Patients with IBS†	?	?	?	?	Low
Nurses' Perception to Chronic Pain†	±	-	?	?	Very low
Perceived difficulty of various patients with headache†	±	-	?	?	Very low
Persistent Pain Attitude Questionnaire†	?	?	?	?	Very low

Appendix 5: Continued

Measurement instrument († = no name of instrument provided in study)	**Relevance**	**Comprehen-siveness**	**Comprehen-sibility**	**Content validity**	**Quality of evidence**
Physician Attitudes and Beliefs Related to Patients with Psychogenic Nonepileptic Spells†	±	-	?	±	Very low
Physicians' Attitudes Regarding CFS/FM†	+	-	?	±	Very low
Physicians' Opinions about Functional Seizures (Psychogenic Nonepileptic Seizures) †	±	-	?	±	Very low
Provider Perceptions of Patients With SSRDs†	+	-	?	±	Low
Self-Efficacy and Attitudes Among Pediatricians in Managing Medically Unexplained Physical Symptoms†	±	-	?	±	Very low
Questionnaire for General Practitioners about Chronic Fatigue Syndrome†	±	-	?	±	Very low
Questionnaire on Beliefs and Experiences about the Treatment of Chronic Pain in Emergency Departments (EDs) †	±	+	?	+	Very low
Understanding and Management of Conversion Disorder - Psychiatrists†	±	-	?	±	Low
Understanding and Management of Conversion Disorder - Neurologists†	±	-	?	±	Very low
Views about functional neurological disorders†	±	+	?	±	Very low

Chapter 3

Development of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP)

McGhie-Fraser B, McLoughlin C, Lucassen P, Ballering A, van Dulmen S, Brouwers E, Stone J, olde Hartman T. Measuring persistent somatic symptom related stigmatisation: Development of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). *Journal of Psychosomatic Research. 2024 June 111689.*

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Abstract

Objective: Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months. These include single symptoms such as chronic pain, combinations of symptoms, or functional disorders such as fibromyalgia or irritable bowel syndrome. While stigmatisation by healthcare professionals is regularly reported, there are limited measurement instruments demonstrating content validity. This study develops a new instrument to measure stigmatisation by healthcare professionals, the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP).

Methods: Development was an iterative process consisting of research team review, item generation and cognitive interviewing. We generated a longlist of 60 items from previous reviews and qualitative research. We conducted 18 cognitive interviews with healthcare professionals in the United Kingdom (UK). We analysed the relevance, comprehensibility and comprehensiveness of items, including the potential for social desirability bias.

Results: After research team consensus and initial feedback, we retained 40 items for cognitive interviewing. After our first round of interviews (n = 11), we removed 20 items, added three items and amended five items. After our second round of interviews (n = 7), we removed four items and amended three items. No major problems with relevance, comprehensibility, comprehensiveness or social desirability were found in remaining items.

Conclusion: The provisional version of the PSSS-HCP contains 19 items across three domains (stereotypes, prejudice, discrimination), demonstrating sufficient content validity. Our next step will be to perform a validation study to finalise item selection and explore the structure of the PSSS-HCP.

1. Introduction

Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months (1). These include single symptoms such as chronic pain, combinations of symptoms, or syndromes meeting the criteria for functional disorders such as fibromyalgia or irritable bowel syndrome.

While symptoms have historically been distinguished as those with a clear biomedical pathophysiology or not, there is increasing evidence that all persistent symptoms share neuropsychological mechanisms (2). We use the term PSS here also because it is the preferred umbrella term by people living with symptoms (3), although we acknowledge that other terms are used including functional disorders (4). As well as causing distress and disruption to daily functioning (5), many people with PSS face stigmatisation which can delay help-seeking (6, 7). Perceived stigma by people with PSS is associated with decreased wellbeing (8), increased depression and anxiety (9), and treatment non-adherence (10).

Stigmatisation is a dynamic process where elements of labelling, stereotyping, separation, status loss, and discrimination occur in the context of power (11). During clinical consultations, people with PSS have reported being dismissed, accused of faking or exaggerating symptoms, and having truthfulness in reporting symptoms questioned ((12), (13), (14), (15), (16)). Healthcare professionals themselves report stigmatising attitudes towards people with PSS, questioning the legitimacy of symptoms and motivations for seeking treatment ((17), (18), (19)). An important contributing factor to stigmatisation by healthcare professionals is incongruence between the severity and persistence of symptoms against biomedical expectation, or an absence of structural abnormalities (20). This incongruence provides barriers to successful communication and explanations (21, 22), and feelings of helplessness in the professional role (23).

There is increasing evidence that across stigmatised statuses, stigma can be considered a mediator that fundamentally shapes health outcomes and equalities (24). In the context of PSS this link is correlational. A potential explanation for this is that when someone experiences a lack of validation about their symptoms, or an implication that their symptoms are illegitimate, this can cause psychological distress (6, 9). Further, stigmatised people

may conceal their condition to protect themselves further from stigmatising experiences. In doing so they are less likely to seek care or more likely to delay care (9). Through this connection to health outcomes and quality of life, reducing stigmatisation by healthcare professionals is a priority in both research and clinical practice.

Stigmatisation occurs at different, intersecting scales (25). An influential model by Link and Phelan describes stigma as starting when a person is labelled based on particular characteristics, such as having PSS. They are then linked to undesirable characteristics (stereotypes) and devalued or excluded through status loss or discrimination. Throughout this process, emotional reactions can occur (11). We wanted to focus more specifically on the role of the healthcare professional, where interventions can be focused and evaluated. Here, we conceptualise stigmatisation as a model of interacting stereotypes, prejudices and discrimination. Stereotypes are "*beliefs, or 'cognitive schemas' about the characteristics and behaviours of groups of individuals*", characterised by their inaccuracy and negativity (26)(p132). Prejudices are the negative "*emotional reaction or feelings that people have toward a group or member of a group*" (26)(p132). These can include emotions of anger and hostility, but also fear or discomfort. Discrimination describes behaviours in the "*differential and disadvantaged treatment of the stigmatised*" (27)(p93). These behaviours exist on a spectrum from subtle 'microaggressions', avoidance, maintaining social distance, to more explicit jokes, bullying or denial of opportunities.

While there are many reported examples of stigmatisation by healthcare professionals, there is a lack of validated measurement instruments. Methods used to measure PSS related stigmatisation include explicit measures (e.g. questionnaires, interviews) (17,(28), (29), (30), (31)), and implicit measures (e.g. response latency tests (32) or response priming studies (33)). While multiple methods are needed, we focused on developing a self-reported questionnaire that could primarily be used as an outcome measurement for stigma reduction interventions. In a recent systematic review (34), we found that there was no candidate instrument with sufficient content validity. Assessed constructs were rarely defined and development lacked involvement of healthcare professionals or pilot testing. Among identified instruments, the HC-PAIRS was the most established (35), used most frequently among our included studies. However, this largely explored beliefs about the ability of patients to

function rather than stigmatising attitudes about these patients. While a couple of instruments demonstrated sufficient relevance (>85% of items) based on face validity (the Chronic Fatigue Syndrome Attitudes Test (30) and the Medical Condition Regard Scale (29), we felt that some items were not relevant in this context. Further, we felt that these scales were not comprehensive relating to stigma (sufficiently exploring each component of stereotypes, prejudices and discrimination). Therefore, we decided to develop a new instrument to measure PSS related stigmatisation by healthcare professionals, the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP).

2. Methods

The development of the PSSS-HCP was an iterative process consisting of research team review, item generation and cognitive interviewing. This study was pre-registered on Open Science Framework (https://osf.io/7jtr4). The research was approved by the University of Edinburgh and the South Central - Oxford C Research Ethics Committee (22/SC/0473). The current study is part of the innovative training network ETUDE (Encompassing Training in fUnctional Disorders across Europe) (36).

2.1. Taxonomy of measurement properties

The taxonomy of measurement properties developed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group was used (37, 38). While COSMIN guidelines are primarily designed for outcome measurements in patients, the process can simply be adapted by applying the criteria to healthcare professionals.

2.2. Regular review by research team

Feedback was regularly reviewed by a research team which consisted of a medical anthropologist/ sociologist, consultation-liaison psychiatrist, general practitioners, psychologist, epidemiologist, neurologist and social scientist with expertise in stigma. During the item generation phase, we also received feedback on face validity from other healthcare professionals including two general practitioners, an internal medicine specialist, and two people with lived experience of PSS.

2.3. Item generation

We generated an initial longlist of 60 items from: 1) adapting the most relevant examples by face validity from a previous systematic review of PSS related stigma instruments (34); 2) adapting items from stigma instruments relating to other health conditions; and 3) adapting items from a recent scoping review on stigmatisation during clinical consultation (15). We grouped each item according to stereotypes, prejudices and discrimination. Response categories were created in a five-point Likert scale. A five-point scale was chosen due to its familiarity and ease of administration. From collective feedback, items were removed or amended resulting in a 40 item scale for cognitive interviewing. The rationale for 40 items at this stage was pragmatic, based on a proportion of reduction similar to other developed stigma instruments (39) while allowing for each remaining item to be suitably tested during interviewing.

2.4. Cognitive interviewing

Cognitive interviewing aimed to evaluate and improve the content validity of items. Cognitive interviewing is a qualitative technique that evaluates how a participant responds to a questionnaire (40, 41). Response processes include comprehension, the retrieval of necessary information from memory, evaluation of retrieved information, and the selection of a response (40, 42). Related to this is the measurement property of content validity: the "degree to which the content of an instrument is an adequate reflection of the construct to be measured" (38). Within content validity, there are components of relevance (items must be relevant to the construct, target population and context of use, including appropriate response categories), comprehensiveness (ensuring no key concepts are missing), and comprehensibility (ensuring that items and response options are understood as intended) (43).

We conducted cognitive interviews with healthcare professionals within the United Kingdom (UK) over two rounds, with recommendations made after each round. Participants were recruited using a purposive sampling method. We aimed to recruit from each of the following disciplines: general practitioners, internal medicine specialists, neurologists, psychologists, nurses, and physiotherapists. We also aimed to maximise variation in the sample by recruiting healthcare professionals with varying years

of experience of working with people with PSS and varying attitudes towards people with PSS. Inclusion criteria were as follows: 1) participant was willing and able to give informed consent; 2) over 18 years of age; 3) completion of basic healthcare professional training (but not necessarily specialisation); 4) some experience of interacting with people with PSS in their healthcare role (thought this contact did not have to be regular or recent); 5) they were fluent in English (language of interviewer). Healthcare professionals were initially recruited through two sources: NHS Lothian (Scotland, UK) and contacts of the research group based in the UK. These contacts were asked to share our advertisement of the research. Potential participants gave verbal consent to be contacted by the lead researcher (BMF), who waited at least 24 h before contacting the potential participant to discuss the study and arrange a meeting time for consent and interview. All participants were offered a £25 high street voucher to compensate them for their time and input.

Each participant was interviewed one time, lasting between 45 and 90 min. Cognitive interviews were conducted by an experienced qualitative researcher (BMF). We adopted a hybrid approach of the 'think aloud' method and verbal probing (40, 42). Within the think aloud method, participants think out aloud while answering, or recall thoughts after completing an item. Verbal probing occurs where the interviewer administers specific probe questions. These can be anticipated probes, designed to search for potential problems (for example comprehension of a particular phrase) or they may be reactive probes to the response of the participant (40, 42). We explored relevance and comprehensibility for each item, and included a specific prompt for comprehensiveness at the end of the interview. We also explored understanding of the umbrella term 'persistent somatic symptoms' and related terms. Finally, we discussed if participants felt pressured to respond to items in a particular way (social desirability). Our topic guide is shown in **Appendix 1, supplementary material**. Interviews were audio recorded following informed consent and transcribed verbatim.

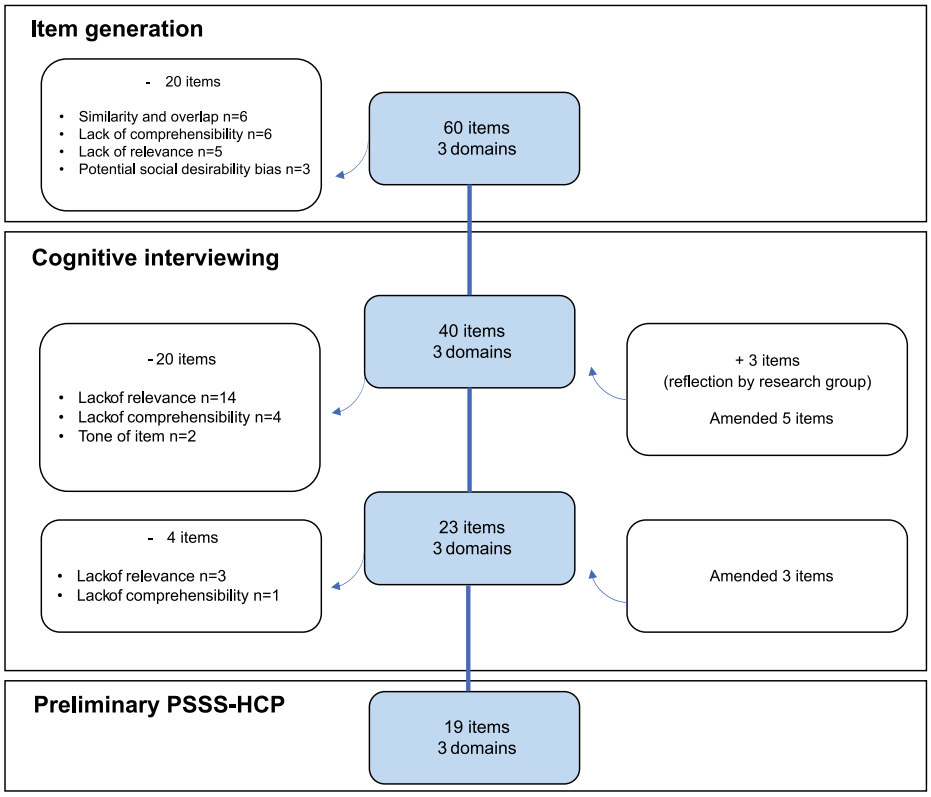
Interview transcripts were analysed using a codebook developed from examples in cognitive interviewing research (42, 44) and content validity components. Analysis was conducted using MaxQDA 2022. All interviews were coded by the interviewer (BMF). A sample of interviews (n = 4) was independently coded by a consultation-liaison psychiatrist (CM) and coding compared. Coding was further discussed with a general practitioner

experienced in qualitative research (PL). Where there were differences in coding, these were discussed until consensus was reached. We used these discussions to update the codebook. To assess comprehensiveness, saturation of item generation was evaluated across each round of interviews. Each component of content validity and social desirability was assessed, with recommendations made for each item. Removal or amendment of items were made following consensus of the research group. This process was repeated until only a few minor problems could be identified.

3. Results

The development process of the PSSS-HCP is summarised in **Fig. 1** A five-point Likert scale is used and ranges from 1 = Strongly disagree, to 5 = Strongly agree.

Figure 1: Development process of the PSSS-HCP



3.1. Item generation

We generated 60 initial items, grouped into domains of stereotypes, prejudices and discrimination as defined in the introduction. After research team consensus and initial feedback, this was reduced to a shortlist of 40 items for cognitive interviewing. Reasons for removal included similarity and overlap with other items (*n* = 6), lack of comprehensibility (n = 6), lack of relevance (*n* = 5), and potential social desirability bias (*n* = 3). Indicative examples of removed items are presented in **Table 1**. The full list of generated items including original sources is provided in **Appendix 2, supplementary material.**

Table 1: Indicative examples of removed items in item generation stage

Reason for removal	Example item
Similarity or overlap to other items	• If I had persistent somatic symptoms, I would never admit this to my family
Lack of relevance	• I feel powerless when working with people with persistent somatic symptoms
Lack of comprehensibility	• I tend to ignore people with persistent somatic symptoms
Potential influence of social desirability bias	• I am able to avoid stigmatising comments towards people with persistent somatic symptoms

3.2. Cognitive interviewing

We conducted 18 cognitive interviews with healthcare professionals, including four general practitioners, three nurses, two internal medicine specialists, two physiotherapists, two neurologists, a psychologist, a speech and language therapist, and a doctor in the NHS Foundation Programme (training for newly qualified doctors preceding specialisation). Participant characteristics were missing for two participants. Participant characteristics are summarised in **Table 2**. Interviews were done in two rounds (*n* = 11, *n* = 7), with amendments made after each round. Interviews were coded to identify potential problems with relevance, comprehensibility, comprehensiveness, social desirability and response options (our codebook is available in **Appendix 3, supplementary material)**. A summary of recommendations at each round is available in **Appendix 4, supplementary material).**

Table 2: Participant characteristics

Variable		n	% (from 16 participants)
Age (years)	18-25	1	6.25
	26-44	7	43.75
	45-64	7	43.75
	≥ 65	1	6.25
Gender	Woman	9	56.25
	Man	7	43.75
Type of healthcare professional	General practitioner	4	25
	Nurse	3	18.75
	Internal medicine specialist	2	12.5
	Physiotherapist	2	12.5
	Neurologist	2	12.5
	Psychologist	1	6.25
	Speech and language therapist	1	6.25
	NHS Foundation Programme	1	6.25
Years of experience working with people with PSS	0-1	1	6.25
	1-2	2	12.5
	2-5	3	18.75
	5-10	1	6.25
	≥ 10 years	9	56.25

After our first round of interviews, we removed 20 items (14 removed for lack of relevance, four removed for lack of comprehensibility and two removed for perceived social desirability). We added three items from suggestions of the research group (two items were variants of existing concepts and one item explored a new concept, diagnostic overshadowing). We amended five items that were perceived as relevant but comprehensibility was slightly inconsistent. The second version of the PSSS-HCP for pilot testing included 23 items. After our second round of interviews, we removed four items (three for lack of relevance and one for lack of comprehensibility) and amended three items. Remaining items are presented in **Table 3**.

Table 3: Preliminary version of the PSSS-HCP

#	Item (* = reverse score)	Domain	Sub-domain/ concept
1	Anybody could develop persistent somatic symptoms under the right circumstances.*	Stereotype	Beliefs about people with PSS
2	Most people with persistent somatic symptoms don't try hard enough to get better.	Stereotype	Beliefs about people with PSS
3	There is little I can do to help people with persistent somatic symptoms.	Stereotype	Beliefs about people with PSS
4	I find that people with persistent somatic symptoms often exaggerate their symptoms.	Stereotype	Beliefs about people with PSS
5	It is important to encourage hope for improved quality of life for people with persistent somatic symptoms.*	Stereotype	Responsibility of healthcare professional
6	I feel as comfortable talking to a person with persistent somatic symptoms as I do other people.*	Prejudice	Emotions about people with PSS
7	I find it satisfying to support people with persistent somatic symptoms.*	Prejudice	Emotions about people with PSS
8	People with persistent somatic symptoms evoke negative feelings in me, such as aversion, stress or impatience.	Prejudice	Emotions about people with PSS
9	I secretly hope that people with persistent somatic symptoms will not return.	Prejudice	Emotions about people with PSS
10	I often feel uncomfortable about providing care to people with persistent somatic symptoms.	Prejudice	Emotions about people with PSS
11	I struggle to feel compassion for people with persistent somatic symptoms.	Prejudice	Emotions about people with PSS
12	Sometimes I lose patience with people with persistent somatic symptoms.	Discrimination	Responsibility of healthcare professional
13	If I had a choice, I would rather not care for people with persistent somatic symptoms.	Discrimination	Responsibility of healthcare professional
14	When someone with persistent somatic symptoms presents a new symptom or complaint, I am less likely to pay attention to other diseases than in other people.	Discrimination	Diagnostic overshadowing
15	If a colleague told me they had persistent somatic symptoms, this would not change how willing I was to work with them.*	Discrimination	Social distance
16	Employers should hire a person with persistent somatic symptoms if they are the best person for the job.*	Discrimination	Social distance
17	If I developed persistent somatic symptoms, I would be reluctant to tell my friends.	Discrimination	Disclosure and help-seeking
18	If I were under treatment for persistent somatic symptoms, I would try not to disclose this to any of my colleagues.	Discrimination	Disclosure and help-seeking
19	I would be reluctant to seek help if I knew I had persistent somatic symptoms.	Discrimination	Disclosure and help-seeking

3.2.1 Relevance

3.2.1.1. Terminology

While the umbrella term PSS was not always spontanenously recognised, all participants were able to clearly distinguish people with these symptoms after being provided with a definition and examples. There was a split in familiar terminology according to treatment setting. Participants in primary care settings were more familiar with PSS, though the term PSS was sometimes used interchangeably with 'persistent physical symptoms' or 'medically unexplained symptoms' in education settings. Participants in secondary care settings were typically more familiar with specific PSS such as chronic pain, or functional disorders such as functional neurological disorder or irritable bowel syndrome. Most participants did not express a preference for terms, but some noted the need to highlight examples and relevant diagnostic terms. We adjusted our definition of PSS to include examples at the symptom, syndrome and disorder level in **Appendix 5, supplementary material.**

3.2.1.2. Relevance to stigma

Items focused on causal attribution (for example: "Persistent somatic symptoms are primarily a psychological disorder") were initially viewed as relevant, with participants noting that psychological attributions of symptoms were often perceived as more stigmatising. This fed into discussions about mind and body dualisms within medicine, with some preferring not to engage with a mutually exclusive concept. Some participants felt unable to answer causal attribution questions due to a perceived lack of knowledge. We reflected that causal attribution was less related to stigma than illness perception, so we removed these items.

Some items exploring personal prejudices were reflected as healthcare systems challenges rather than stigma. This included items such as people with PSS being 'difficult to manage', being 'demanding' or feeling 'frustrated' as a healthcare professional. Participants reflected that these emotions were more a reflection of fragmentation of healthcare, lack of guidance and challenging interactions:

> "*By the time they come to see me as a patient, they have been run around the houses quite a bit… it sometimes feels like hard work because you're having to unpick some of that.*" *(P12, physiotherapist).*

We amended two items to focus more on specific prejudices explored in interviews, including 'feeling uncomfortable'.

Items relating to social distance were generally not perceived as relevant, with participants noting that other factors would influence willingness to interact with people before PSS would play a role. Some participants noted that disruption to daily functioning from symptoms may limit possibilities for interaction, but this was distinct from willingness to engage. Items assessing social distance in the workplace and during clinical consultation were generally perceived as more relevant than items relating to friendship or more personal interaction. We removed items on prospective friendship, instead focusing on relationships in the work context.

3.2.1.3. Relevance to healthcare professionals

One item about treatment was considered not relevant to all healthcare professionals. Treatment was considered only applicable to clinicians, with some nursing participants suggesting that in their role they didn't personally treat people. We rephrased this item from 'treating' to 'supporting'.

3.2.1.4. Relevance of response categories

No problems emerged with the proposed response categories. Two participants expressed a desire for more response categories (such as a seven-point Likert scale), with more explicit 'slightly agree' or 'slightly disagree' options. Two participants expressed a desire for open questions, to provide further qualification or context for responses. In a recent analysis comparing different numbers of response categories (three, five and seven point Likert scales), the authors reported that using a five point response category provided no disadvantage compared to a seven point response, while offering more ease of responding (45). Therefore, we decided to retain the simpler five-point Likert scale and provide an optional open question in future testing.

3.2.1.5. Responsiveness to change

While examples of stigmatisation by other healthcare professionals were regularly discussed, we reflected that items exploring perceived stigmatisation by others were not relevant. Since we aimed to evaluate stigmatisation by individual healthcare professionals, the score on these items would not necessarily be responsive to change. We removed these items.

3.2.1.6. Recall

No problems with the recall and retrieval of relevant information from memory were found. Some participants reflected that their amount of experience affected their responses. One participant reflected that their answers would have been a lot more negative during an early stage of their career, without the years of experience, support, or successful communication strategies that they would later rely on.

3.2.2. Comprehensibility

For items focusing on control, participants questioned if they referred to voluntary control and motivation for secondary gain (stigmatising), or successful management of symptoms and improved functioning (not stigmatising):

> *"Control is a difficult one... it has connotations with almost choice... some people can modify some of their symptoms, but it's not straightforward or consistently effective... the other side of [control] is the notion that you can through processes of education and understanding... you can help people to live with their symptoms and potentially manage them more effectively." (P02, general practitioner).*

There was also doubt if control referred to initial onset of symptoms, or how a person could modify symptoms once presenting. Because of this inconsistency we removed this item. We adjusted an item about motivation to be more explicitly a stereotype: we re-phrased (people with PSS would) 'get better if they really wanted to be healthy' with 'don't try hard enough to get better'.

An item about recovery was also interpreted inconsistently. Some participants interpreted recovery in a biomedical model, referring to complete alleviation of symptoms or a return to baseline condition. For others, recovery could refer to improved functioning and achieving personal goals. We amended this to more explicitly ask about the role of the healthcare professional in encouraging a better quality of life. An item about potential health seeking was also seen as unclear, with 'If I thought I had PSS, I would seek help'. Participants noted that seeking clarity about symptoms is an important motivation for help-seeking. We amended this item to 'knew', where it was clearer that an explanation had previously been provided.

3.2.3. Comprehensiveness

Comprehensiveness was discussed in every interview, with item generation coded both to spontaneous suggestions and to a specific prompt at the end of the interview. Participants commented that they felt the breadth of the items was sufficient: "*it covers most aspects you know... everything comes to mind*" (P16, internal medicine specialist). When asked to generate new ideas: a typical response was "*[I] can't think of anything specific*" (P17, psychologist). In our first round of interviews, we coded four quotes relating to item generation. In our second round, we coded two quotes for idea generation, both of which had been considered in the item generation stage. Therefore we consider it likely that saturation was reached.

3.2.4. Influence of social desirability

When asked about the use of a confidential online survey to answer questions, participants raised no concerns about social desirability. A typical response was "*... if somebody's gonna commit to doing it, I think you'll get honest answers*" (P05, nurse). Some participants commented on the value of reflecting on experiences as a healthcare professional and acknowledging difficult feelings:

> "*I think that sort makes you think... how you portray a group of patients and how we feel about them and get that out a little bit... I think it's helpful. You know these people desperately need something and if our mission is here to help somehow, how can we help rather than just hoping they go away and not come back*" *(P08, nurse)*.

However, for two items we noted that there was hesitance to answer the question if they found the tone negative. For example, we tested an item about personality traits of people with PSS, which one participant described as "*not liking the concept*" (P02, general practitioner). Similarly, an item about manipulation was described as being a "*judgmental statement*" (P04, general practitioner). We removed these items and reflected that early items were predominately negative formulations. We amended the order to include reverse-scored items higher in the item order.

4. Discussion

Perceived stigmatisation is associated with poorer health outcomes and quality of life for people with PSS. While there is increasing need to evaluate the effectiveness of stigma reduction interventions, there are limited stigma measurement instruments that explain their development process or demonstrate content validity. In this study we developed a measurement instrument to assess PSS related stigmatisation by healthcare professionals, the PSSS-HCP. The preliminary version of the PSSS-HCP contains 19 items within three domains (stereotype, prejudice, discrimination). The total sum of scores can range from 19 to 95 and a lower score indicates less stigma. Items 1, 5, 6, 7, 15, 16 are reverse-scored. This has been developed in the context of healthcare professionals working in the UK, requiring further evaluation of validity and reliability before we can consider the scale complete.

We found that responses of healthcare professionals were influenced by perceived structural factors. The importance of structural factors such as policy, legislation and media are well reported in stigma literature (25), as well as in the context of PSS clinical consultations (15). Participants cited structural factors including lack of knowledge and guidelines (46), lack of support from senior healthcare professionals (47), and fragmented healthcare systems (48, 49). All of these factors are highly likely to influence stigmatisation, for example through factors such as perceived confidence and competence. However, we took care in the development process to remove items that were solely considered in the context of healthcare systems factors.

Many of our items exploring social distance were not perceived as relevant. The relevance of social distance might be attributable to dimensions of stigmatised conditions. These dimensions include concealability (the extent to which the visibility of the condition is controllable), aesthetics (how repellant or upsetting the condition is perceived as) or peril (how dangerous the condition is perceived as) (50, 51). Studies regularly using measures of social distance include conditions with perceived risks of infection such as HIV or leprosy, or conditions with stereotypes about danger, unpredictability or violence (such as mental health stigma) (52). It could be that desired social distance as operationalised in other stigma instruments are less of a contributing factor to stigmatisation in PSS, or that health

conditions with particular dimensions (such as higher perceived peril and lower perceived aesthetics) are better suited to explore questions of social distance than PSS. Therefore, we focused on social distance questions that were perceived as more relevant to participants.

In this study we found evidence that self-report questionnaires can offer reflection for professionals about their own experiences. In a review of outcome measurement instruments, it was similarly reported that self-reported questionnaires are not a neutral act of information retrieval. Rather, self-reporting can influence communication in a clinical setting (53). Questionnaires like the PSSS-HCP may similarly offer a starting point for self-reflection and improved clinical communication. Indeed, there are some specific PSS educational interventions that focus on self-reflection, for example through the use of group supervision (54). This supports the notion that both explicit and implicit measurement instruments are needed to explore complex social processes such as stigma, with triangulation made between them. While demand characteristics pose a threat to the validity of the instrument, these could also apply to implicit measurement instruments (55) or study designs when evaluating stigma reduction interventions (56). Therefore, we believe in the utility of self-reported instruments while also emphasising the importance of assessing the influence of demand characteristics where possible.

While we have used a well-recognised conceptualisation of stigma during this development, it is important to acknowledge that there are multiple possible conceptualisations. For example, recent interventions against mental health stigma have conceptualised stigma as a problem of knowledge, attitudes and behaviour (57). This was based on the work of Thornicroft et al., who described stigma as a problem of ignorance (knowledge), attitudes (prejudices), and behaviour (discrimination) (58). There is typically convergence between these conceptualisations, with items broadly covering cognitive, affective and behavioural components of stigma from the perspective of a potential stigmatiser. However, points of divergence might include the approach in which items are asked. For example, while we proposed to measure the extent of agreement with stereotypes, another conceptualisation of stigma may lead to items that test knowledge more prescriptively (with correct or incorrect responses). Similarly, while there is recently some critique about the role of emotional reaction as a necessary part of the stigma conceptualisation (59), this

critique is aimed at the perspective of someone who is stigmatised rather than the perspective of a potential stigmatiser. In the context of PSS, we know that the emotional reactions experienced by healthcare professionals are particularly relevant, so we developed items to specifically explore this. Therefore, while conceptualisation of stigma will undoubtedly affect the operationalisation of items, these approaches are not necessarily exclusive of each other.

The terminology used to describe persistent somatic symptoms continues to be debated (1, 4, 60, 61). We explored comprehensibility of PSS as part of this study and updated our definitions through testing. A broad spectrum of recognised terms was expected, reflecting both available training to healthcare professionals and familiarity with relevant diagnostic criteria. We found no consensus on preferred terminology, and no significant problems with understanding or coherence when definitions and examples were provided. So long as appropriate definitions are provided, we consider it likely that the term persistent somatic symptoms in the scale could be substituted for related terms such as functional disorders, or syndromes including irritable bowel syndrome or fibromyalgia in future versions of the scale without compromising content validity.

4.1. Strengths and limitations

The most important strength of this study is the use of robust methodology throughout development. We have followed COSMIN guidance throughout development and reporting, offering transparency about analysis and decision-making. We involved healthcare professionals throughout the process, evaluating the instrument and individual items for their content validity.

A further strength of this study is the consideration of the influence of social desirability bias. We discussed the potential influence of social desirability on individual item responses as well as overall responses by distribution format. We also explored factors that might influence the likelihood of giving socially desirable answers. This included removing items that were perceived to be too negative in tone.

This study also has limitations. While the PSSS-HCP is developed for healthcare professionals, our involvement of people with PSS is limited.

We gained feedback from people with lived experience of PSS in early stages of development through a convenience sample, but there is scope to include perspectives more formally. We mitigated the impact of this by adapting items from a recent scoping review exploring patient perspectives of stigmatisation in the clinical consultation (15) and an ongoing interview study exploring patient perspectives of stigmatisation during diagnosis of functional neurological disorder (16). Therefore, we do not believe that any important domains are missing. However, additional research may be warranted to expand the item pool.

Another limitation is that there may be a risk of bias in our sample of healthcare professionals. Participants were self-selecting and therefore more likely to have an interest or willing to discuss their experiences working with people with PSS. This is reflected in the high proportion of participants with extensive experience of working with people with PSS. Therefore, these participants may not represent the full spectrum of beliefs and attitudes of healthcare professionals in the UK. However, since the aim of the study was to assess the content validity of items rather than measure stigmatisation outright, we think the likely impact is negligible. The effect of recruitment bias was further limited with the use of a detailed topic guide.

Our next step will be to perform a validation study of healthcare professionals in the UK to finalise item selection, explore the structure of the PSSS-HCP, and assess the validity and reliability of the scale. Only through both studies can we consider the development complete. However, we believe that this initial study represents an important step in the development of the scale and in the measurement of PSS related stigma.

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Supplementary material

Appendix 1: Cognitive interview topic guide

WRITTEN INFORMED CONSENT:

(participants will have already looked at information sheet, agreed to be contacted, and agreed to a research appointment)

Thanks for coming in today, I'll explain to you a little more about what we'll be doing today.

- We are testing a new survey with the help of healthcare professionals such as yourself.
- I'll ask you questions and you answer them, just like a regular survey. There are no right or wrong answers. The aim for us to help develop the questions.
- I'll be taking some notes, and we will record the interview to make notes after the interview. Once the recording has been written, we will delete the recording. We'll do this for about 45 minutes, unless we have finished the questions before then.
- Do you have any questions before we start? If so, please sign the consent form.

INSTRUCTIONS

- I'll ask you questions and you answer them, just like a regular survey. There are no right or wrong answers.
- However, our goal is to get a better idea of how the questions are working. So I'd like you think to 'think aloud' as you answer the questions – just tell me everything you are thinking about as you go about answering them.
- At times I'll also stop and ask you more questions about the terms of phrases in the questions and what you think the question is asking about.
- Please keep in mind that I really want to hear all of your opinions and reactions. Don't hesitate if something seems unclear, is hard to answer, or doesn't seem to apply to you.

INTRODUCTION TO PSS

In this survey we are asking you questions about persistent somatic symptoms.

- Can you explain to me in your own words what you think the term "persistent somatic symptoms" means?
- Do your recognise the term? What other terms are you familiar with?
- (Provide definition): This is the definition that we will be using: is this a clear group for you?

PSSS-HCP COGNITIVE INTERVIEW:

To read out full question alongside response categories (Strongly Disagree, Disagree, Neither Disagree or Agree, Agree, Strongly Agree).

- Probes may include the anticipated probe for each question, conditional probes or spontaneous probes.
- Some probes focus on the social desirability of items (would they likely to answer this in the same away if they did the questionnaire alone? What influences how they answer these?)

Item #	Item (* = reverse scoring)	Anticipated probes
1	Most people with persistent somatic symptoms have control over their symptoms	In your own words, what is this question asking? What does the term "control" mean to you in this question?
2	Many people experience physical symptoms without being distressed by them.*	What do you think this question is asking?
3	People with persistent somatic symptoms are responsible for their symptoms	What does the term "responsible" mean to you in this question?
4	People with persistent somatic symptoms would get better if they really wanted to be healthy	In your own words, what is this question asking? Why do you say this?
5	Persistent somatic symptoms are primarily a psychological disorder	Was this hard or easy to answer?
6	People with persistent somatic symptoms have personality problems	What does "personality problems" mean to you in this question? Do you find this a relevant question to ask?
7	People with persistent somatic symptoms really want to get better*	What do you think this questions is asking?
8	People with persistent somatic symptoms communicate clearly*	What does clear communication mean to you here? How sure are you of this?
9	People with persistent somatic symptoms are manipulative.	What do you think "manipulative" means in this question? If you were answering this question anonymously, would you answer any differently? Why do you say this?
10	People with persistent somatic symptoms communicate less clearly than other people	What do you think "less clearly" means in this question?

Interview: Continued

Item #	Item (* = reverse scoring)	Anticipated probes
11	People with persistent somatic symptoms are truthful about their symptoms.*	What do you think "truthful" means in this question? If you were answering this question anonymously, would you answer any differently?
12	People with persistent somatic symptoms are unwilling to accept psychological reasons for their symptoms	Do you discuss psychological reasons for symptoms? What do you think "unwilling to accept" means in this question?
13	I find people with persistent somatic symptoms demanding	What do you think "demanding" means in this question?
14	People with persistent somatic symptoms are lifelong users of the health care system	What do you think this questions is asking? How sure are you of this?
15	If I had persistent somatic symptoms, I would not be able to live a satisfying life.	What do you think "satisfying life" means in this question?
16	People with persistent somatic symptoms are difficult to manage	What do you think "difficult" means in this question? What do you think "manage" means in this question?
17	The management of people with persistent somatic symptoms is time-consuming	Was this hard or easy to answer? What does "time consuming" mean to you?
18	I feel as comfortable talking to a person with persistent somatic symptoms as I do with other people*	What influences you feeling comfortable? If you were answering this question anonymously, would you answer any differently?
19	I find it satisfying to treat people with persistent somatic symptoms*	What does "satisfying" mean to you?
20	I find working with people with persistent somatic symptoms frustrating	What does "frustrating" mean to you?
21	There is little I can do to help people with persistent somatic symptoms	What help?
22	I enjoy working with people who have persistent somatic symptoms*	Was this hard or easy to answer?
23	I secretly hope that people with persistent somatic symptoms will not return	Was this hard or easy to answer? What does "not return" mean to you in this question? What does "secretly" mean to you in this question?
24	I often feel unsure of what to do with people with persistent somatic symptoms	What does this question mean to you? If you were answering this question anonymously, would you answer any differently?
25	I struggle to feel compassion for people with persistent somatic symptoms	Was this hard or easy to answer?

Interview: Continued

Item #	Item (* = reverse scoring)	Anticipated probes
26	People with persistent somatic symptoms are often treated unfairly	What does "treated unfairly" mean to you in this question?
27	People with persistent somatic symptoms are not taken seriously by healthcare professionals	What does "not taken seriously" mean to you in this question?
28	Employers should hire people who have persistent somatic symptoms if they are qualified for the job*	Is it easy or difficult to answer this?
29	Sometimes I lose patience with people with persistent somatic symptoms	What does "lose patience" mean to you in this question?
30	I enjoy giving extra time to people with persistent somatic symptoms*	Are you able to give extra time if needed? What influences giving extra time to people?
31	If my colleague told me they had persistent somatic symptoms, I would still want to work with them	What does this question mean to you? If you were answering this question anonymously, would you answer any differently?
32	If my colleague told me they were unable to work because of their persistent somatic symptoms, I would not hesitate to take on their responsibilities	What does the word "responsibilities" mean to you here? How often do you take on the responsibilities of colleagues through sickness?
33	I would make close friends with someone who had persistent somatic symptoms*	What does this question mean to you?
34	If I had a choice I would rather not care for people with persistent somatic symptoms	What does this question mean to you?
35	One role for the healthcare professional is to foster hope for recovery*	What does "foster hope" mean to you in this question? Are there other roles of healthcare professionals that you think are important?
36	I often struggle with the discussion of associated psychological problems with people with persistent somatic symptoms	How often do you discuss associate psychological problems with people?
37	If I had persistent somatic symptoms, I would never admit this to any of my friends.	What does this question mean to you? If you were answering this question anonymously, would you answer any differently?
38	Discussing persistent somatic symptoms only makes the symptoms worse	What does this question mean to you?
39	I would seek help if I thought I had persistent somatic symptoms.*	What does "seek help" mean to you in this question?
40	If I had persistent somatic symptoms, I would never tell this to my colleagues.	What does this question mean to you?

Interview: Continued

GENERAL QUESTIONS

Now that we have finished the survey, I have some short general questions:

- **Social desirability** - this survey can ask about personal attitudes and beliefs – this can be quite difficult to answer openly. Do you think that you would fill out the questionnaire differently: anonymously on an online survey? What would influence you filling in different answers to your true beliefs?
- **Comprehensiveness:** Are there any important beliefs or attitudes about PSS that you think have been missed here?

END RECORDING

Thank you so much for going through this survey with me. We will take the feedback from this to improve the questions.

END OF INTERVIEW

GIFT VOUCHER

Appendix 2: Initial item generation for PSSS-HCP (60 item longlist)

#	Item (* = reverse scoring)	Concept	Source	Reason for initial removal from item generation stage
	Stereotypes			
1	These people's symptoms are real.*	Beliefs about people with PSS	Ahern et al., 2009	Lack of comprehensibility
2	Most people with persistent somatic symptoms have control over their symptoms.	Beliefs about people with PSS	Adapted from Ahern et al., 2009, Brief Illness Perception Questionnaire	
3	Persistent somatic symptoms are a women's problem.	Beliefs about people with PSS	Adapted from 3*I	Lack of comprehensibility
4	Many people experience physical symptoms without being distressed by them.*	Beliefs about people with PSS	Continuum Beliefs Questionnaire	
5	Anybody could develop persistent somatic symptoms under the right circumstances.*	Beliefs about people with PSS	Adapted from Continuum Beliefs Questionnaire	Similar or overlap with other items
6	People with persistent somatic symptoms are to blame for getting sick.	Beliefs about people with PSS	CAT	Potential for social desirability bias
7	People with persistent somatic symptoms are responsible for their symptoms.	Beliefs about people with PSS	Ahern et al., 2009, Lehn et al., 2019	
8	If a person with persistent somatic symptoms is not getting better despite treatments, that means they are malingering.	Beliefs about people with PSS	Adapted from Desai, 2012.	Potential for social desirability bias
9	People with persistent somatic symptoms would get better if they really wanted to be healthy.	Causal attribution	Adapted from CAT	
10	Persistent somatic symptoms are primarily a psychological disorder.	Causal attribution	Adapted from Reid et al., 2001	
11	People with persistent somatic symptoms have personality problems.	Causal attribution	Adapted from Reid et al., 2001	
12	People with persistent somatic symptoms really want to get better*.	Beliefs about people with PSS	Adapted from CPMS-E	
13	People with persistent somatic symptoms try to obtain sick leave to stop working.	Beliefs about people with PSS	Adapted from CPMS-E	Lack of comprehensibility

Appendix 2: Continued

#	Item (* = reverse scoring)	Concept	Source	Reason for initial removal from item generation stage
14	People with persistent somatic symptoms are less likely to be compliant than other people with persistent somatic symptoms.	Beliefs about people with PSS	Adapted from Evans et al., 2011	Lack of relevance
15	People with persistent somatic symptoms communicate clearly*	Beliefs about people with PSS	Adapted from CAT	
16	People with persistent somatic symptoms are manipulative.	Beliefs about people with PSS	Adapted from Ahern et al., 2009	
17	People with persistent somatic symptoms are lazy.	Beliefs about people with PSS	Adapted from 3*I	Lack of relevance
18	People with persistent somatic symptoms are more likely to exaggerate their symptoms.	Beliefs about people with PSS	new (othering, adapted from Treufeldt & Burton, submitted)	Similar or overlap with other items
19	People with persistent somatic symptoms communicate less clearly than other people with persistent somatic symptoms.	Beliefs about people with PSS	new (othering, adapted from Treufeldt & Burton, submitted)	
20	People with persistent somatic symptoms are truthful about their symptoms.*	Beliefs about people with PSS	Ahern et al., 2009	
21	People with persistent somatic symptoms are not as trustworthy as the average person.	Beliefs about people with PSS	Adapted from Devaluation/ Discrimination scale	Lack of relevance
22	People with persistent somatic symptoms are unwilling to accept psychological reasons for their symptoms.	Beliefs about people with PSS	Adapted from Lehman et al., 2021	
23	I find people with persistent somatic symptoms demanding.	Beliefs about people with PSS	Adapted from Lehn et al., 2019	
24	People with persistent somatic symptoms are lifelong users of the health care system.	Recovery	Adapted from OMS-HC	
25	If I had persistent somatic symptoms, I would not be able to live a satisfying life.	Recovery	Adapted from OMS-HC	

Appendix 2: Continued

#	Item (* = reverse scoring)	Concept	Source	Reason for initial removal from item generation stage
	Prejudices			
26	People with persistent somatic symptoms are difficult to manage.	Emotions about people with PSS	Adapted from Reid et al., 2001	
27	The management of people with persistent somatic symptoms is time-consuming.	Emotions about people with PSS	Adapted from Garcia-Campayo et al 1998.	
28	I feel as comfortable talking to a person with persistent somatic symptoms as I do other people*.	Emotions about people with PSS	Adapted from MICA	
29	It is satisfying to treat people with persistent somatic symptoms*.	Emotions about people with PSS	Adapted from Glazebrook et al., 2001	
30	I find working with people with persistent somatic symptoms frustrating.	Emotions about people with PSS	Glazebrook et al., 2001	
31	I feel powerless when working with people with persistent somatic symptoms.	Emotions about people with PSS	Adapted from Sirri et al., 2017	Lack of relevance
32	I often feel uncomfortable about providing care to people with persistent somatic symptoms	Emotions about people with PSS	Adapted from Rosendal et al., 2005	Similar or overlap with other items
33	I enjoy working with people who have persistent somatic symptoms*.	Emotions about people with PSS	Adapted from Glazebrook et al., 2001	
34	I secretly hope that people with persistent somatic symptoms will not return.	Emotions about people with PSS	DDPRQ-10, Hahn et al., 2001	
35	I often feel unsure of what to do with people with persistent somatic symptoms.	Emotions about people with PSS	Adapted from Rosendal et al., 2005	
36	People with persistent somatic symptoms evoke negative feelings in me, such as aversion, stress or impatience.	Emotions about people with PSS	Lehman et al., 2021	Similar or overlap with other items
37	I struggle to feel compassion for people with persistent somatic symptoms.	Responsibility of professional	Adapted from OMS-HC	
38	There is little I can do to help people with persistent somatic symptoms.	Responsibility of professional	MCRS	

Appendix 2: Continued

#	Item (* = reverse scoring)	Concept	Source	Reason for initial removal from item generation stage
	Discrimination			
39	People with persistent somatic symptoms are often treated unfairly.	Perceived stigma of others	Adapted from OMS-HC	
40	People with persistent somatic symptoms are not taken seriously by healthcare professionals.	Perceived stigma of others	Adapted from Letson et al., 1996.	
41	Employers should hire people who have persistent somatic symptoms if they are qualified for the job*.	Recovery	Adapted from Devaluation/ Discrimination scale.	
42	I am likely to conduct medical tests because people with persistent somatic symptoms expect me to do so.	Responsibility of professional	Adapted from Buchbinder et al., 2001	Lack of comprehensibility
43	I am able to avoid stigmatising comments towards people with persistent somatic symptoms.*	Responsibility of professional	Lehman et al., 2021	Potential for social desirability bias
44	Sometimes I lose patience with people with persistent somatic symptoms.	Responsibility of professional	Lehman et al., 2021	
45	Discussing persistent somatic symptoms only makes the symptoms worse	Responsibility of professional	new (denial, adapted from Treufeldt & Burton, submitted)	
46	I enjoy giving extra time to people with persistent somatic symptoms*	Responsibility of professional	MCRS	
47	I try not to get too involved with people with persistent somatic symptoms.	Social Distance	Adapted from Reid et al., 2001	Lack of comprehensibility
48	If my colleague told me they had persistent somatic symptoms, I would still want to work with them.	Social Distance	Adapted from MICA	
49	I would make close friends with someone who had persistent somatic symptoms*.	Social Distance	Adapted from OAMI scale	
50	I would not mind if a person with persistent somatic symptoms lived next door to me.*	Social Distance	Adapted from OAMI scale	Similar or overlap with other items
51	If I had a choice I would rather not care for people with persistent somatic symptoms.	Social Distance	Adapted from Ahern et al., 2009	

Appendix 2: Continued

#	Item (* = reverse scoring)	Concept	Source	Reason for initial removal from item generation stage
52	One role for the healthcare professional is to foster hope for recovery*.	Responsibility of professional	Adapted from OMS-HC	
53	I tend to ignore people with people with persistent somatic symptoms.	Responsibility of professional	Adapted from Wilsey et al., 2008	Lack of comprehensibility
54	I often struggle with the discussion of associated psychiatric/psychological problems with these people.	Responsibility of professional	Lehn et al., 2019	
55	If my colleague told me they were unable to work because of their persistent somatic symptoms, I would not hesitate to take on their responsibilities.	Responsibility of professional	new (from research team)	
56	I treat some people with persistent somatic symptoms using placebos	Responsibility of professional	Lehman et., 2021	Lack of relevance
57	If I had persistent somatic symptoms, I would never admit this to any of my friends.	Disclosure	Adapted from MICA scale	
58	I would seek help if I thought I had persistent somatic symptoms.*	Disclosure	Adapted from OAMI scale	
59	If I had persistent somatic symptoms, I would never admit this to my colleagues.	Disclosure	Adapted from MICA	
60	If I had persistent somatic symptoms, I would never admit this to my family.	Disclosure	new (from research team)	Similar or overlap with other items

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Appendix 3: codebook for analysis of cognitive interviews

Code	Memo
Relevance	Refers to how relevant the questions are: to the construct of stigma, but also to the participants.
Relevance > General reflection	General reflections on relevance of questionnaire - postive or negative sentiment
Relevance > Relevance stigma	This includes examples where the assumption of the question relating to stigma is not as hypothesised. This might include a different interpretation of the question, or a factor not making the question relevant to stigma.
Relevance > Relevance participant	The question is not relevant to the participant (for example, the question does not apply in their role).
Comprehensibility	Comprehensibility refers to how easily understood the questions are to the participant. Are they understood in the same way as the researchers intended?
Comprehensibility > Unclear	If a question is unclear, or the participant does not know how to answer.
Comprehensibility > Too generalising	This includes if a question is too generalising (the participant cannot generalise enough for the whole population).
Comprehensibility > Technical term	Discussion over a particular term used in the question, and different semantics of the term (definitions, consequences of using words, alternatives). This is generally coded if there are multiple interpretations of a concept. Different to 'wording' - wording refers to general unclarity due to the wording (for example, the order of words).
Comprehensibility > Technical PSS	Discussion of terminology (meaning of PSS, discussion of alternative terms and diagnoses provided, preference of terms). Interviews typically started with the question: how familiar are you with the term PSS?
Comprehensibility > Wording	Refers to general clarity of the question (for example, word order, small changes to improve clarity).
Social desirability	Coded if the participant discusses any social pressure or influence to answer a question in a particular way. Sometimes this is openly discussed by the interviewer - sometimes it is introduced by the participant. Includes both: General social desirability: about how they would answer across all of the questions (in this questionnaire) Specific social desirability: about how they would anwer a particular question.

Appendix 3: Continued

Code	Memo
Social desirability > Influence on answering	When the participant comments that there are social factors that would influence how they would answer the questionnaire (either in general, or to a specific question)
Social desirability > Tone of questions	When the participant comments on the tone of the question (particularly, negative tone of question, such as re-inforcing a stereotype)
Comprehensiveness	Comprehensiveness refers to how comprehensive the questions are related to our construct of stigma. Have we covered all of the important questions and concepts relating to stigmatisation in healthcare professionals? We explored this mostly at the end of interviews, asking "have we missed anything", and "if you could ask a question yourself, what would you ask?"
Comprehensiveness > General reflection	General reflections on comprehensiveness of questionnaire - postive or negative sentiment
Comprehensiveness > Idea generation	Specific suggestions for new questions or concepts to test in questionnaire.
Response categories	Are the response categories (likert scale, strongly disagree to strongly agree) appropriate? Coded if the answer does not appear to fit the likert categories given, or if the participant expresses a desire to choose a different option.
Response categories > Desire for open questions	Participants express a desire for open questions (to explain decision behind a likert response)
Order and flow	Issues with the order of questions, or 'flow' of questions. Do questions have a logical order? Is there progression?
Retrieval	Coded if participant cannot reliably recall the answer (for example, if we are asking about a particular time period).
Responsiveness	Reflections on responsiveness - can you reduce stigma? Can you improve attitudes and beliefs?
Items	Item specific coding

Appendix 4: Item recommendations at each stage of cognitive interviewing

Available in online supplementary material

Appendix 5: Preliminary version of the PSSS-HCP and scoring instructions

Available in online supplementary material

Chapter 4

Validation of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP)

McGhie-Fraser B, Ballering A, Lucassen P, McLoughlin C, Brouwers E, Stone J, olde Hartman T, van Dulmen S. Validation of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). Journal of Clinical Epidemiology. 2024 Aug 111505.

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Abstract

Objective: Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months. These include single symptoms such as chronic pain, combinations of symptoms, or functional disorders such as fibromyalgia or irritable bowel syndrome. While many studies have explored stigmatisation by healthcare professionals towards people with PSS, there is a lack of validated measurement instruments. We recently developed a stigma scale, the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). The aim of this study is to evaluate the measurement properties (validity and reliability) and factor structure of the PSSS-HCP.

Study Design and Setting: The PSSS-HCP was tested with 121 healthcare professionals across the United Kingdom to evaluate its measurement properties. Analysis of the factor structure was conducted using principal component analysis. We calculated Cronbach's alpha to determine the internal consistency of each (sub)scale. Test re-test reliability was conducted with a sub-sample of participants with a two-week interval. We evaluated convergent validity by testing the association between the PSSS-HCP and the Medical Condition Regard Scale (MCRS) and the influence of social desirability using the short form of the Marlowe Crowne Social Desirability Scale (MCSDS).

Results: The PSSS-HCP showed sufficient internal consistency (Cronbach's alpha = 0.84) and sufficient test-retest reliability, intraclass correlation = 0.97 (95% CI 0.94 to 0.99, $p<0.001$). Convergent validity was sufficient between the PSSS-HCP and the MCRS, and no relationship was found between the PSSS-HCP and the MCSDS. A three factor structure was identified (othering, uneasiness in interaction, non-disclosure) which accounted for 60.5% of the variance using 13 of the 19 tested items.

Conclusion: The PSSS-HCP can be used to measure PSS stigmatisation by healthcare professionals. The PSSS-HCP has demonstrated sufficient internal consistency, test-retest reliability, convergent validity and minimal influence of social desirability bias. The PSSS-HCP has demonstrated potential to measure important aspects of stigma and provide a foundation for stigma reduction intervention evaluation.

1. Introduction

Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months (1). These include single symptoms, combinations of symptoms, or syndromes meeting the criteria for functional disorders such as fibromyalgia or irritable bowel syndrome. As well as causing suffering, distress, and disruption to daily functioning (2), many patients report stigmatising attitudes from healthcare professionals when seeking help. Patients with PSS have reported that their symptoms are not taken seriously, that their illness is not treated as legitimate, and that they are dismissed or accused of exaggerating symptoms (3-8). Stigmatisation is increasingly understood as a casual factor in shaping health outcomes, through poorer living conditions and socioeconomic status, decreased access to and quality of health services, and development of physiological and psychological symptoms due to managing stigma (9-12). In the context of PSS there is less data on the outcomes of stigma, but stigmatisation is associated with decreased wellbeing (13), increased depression and anxiety (14), treatment non-adherence (15) and increased burden for caregivers (16). Key explanations provided for these associations include the psychological distress of not having an illness validated by a healthcare professional (14, 17), and disruption to health-seeking behaviour when stigmatisation is anticipated (7, 14).

It is well reported that healthcare professionals find working with patients with PSS challenging, highlighting challenges in communication and finding acceptable explanations (18), barriers to diagnosis (19), fragmented healthcare systems (20), and lack of formal education (21, 22). These structural barriers are suggested to contribute to personal negative attitudes, including anxiety and desired avoidance of patients with PSS (21), and a sense of powerless in the professional role (23). In recent years, there are increasing educational resources and stigma reduction interventions being developed. Providing information (about a health condition, or the impact of the stigma on health outcomes), skills building activities, facilitated contact with the stigmatised group, and structural approaches (changing policy, or facility restructuring) are all strategies that have been used to reduce stigmatisation in healthcare settings (24). Examples of current PSS stigma reduction interventions for healthcare professionals include educational interventions about the biopsychosocial components of PSS (25-28), and

interventions to improve specific skills such as communication during clinical consultation (29-32). These interventions typically feature contact-based sessions, where healthcare professionals interact with people with experience of PSS who are managing their illness or who have recovered.

Key to determining the effectiveness of stigma reduction interventions is using a valid and reliable measurement instrument. While there have been many studies exploring the beliefs and attitudes of healthcare professionals towards people with PSS, there is a lack of validated measurement instruments to assess them (33). The characteristics of PSS as a stigmatised status are unique, since these symptoms fall outside traditional dualisms of illness as solely psychological or somatic in aetiology (34). We initially explored the feasibility of adapting both existing PSS related stigma scales and mental health stigma scales for this population. However, we found that a substantive proportion of items of these were not relevant to the context of PSS (for example, with items focusing on perceived peril, or assumed psychological treatment pathways). Therefore we focused on developing a specific stigma scale, using the most relevant items from existing examples and establishing their content validity.

In a previous study, we described the development of a stigma scale to measure stigmatisation by healthcare professionals towards people with PSS. This stigma scale is named the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP) (35). We used input from healthcare professionals and people with lived experience of PSS to select and improve items important for measuring stigma. We defined stigma as a dynamic process where elements of labelling, stereotyping, separation, status loss, and discrimination occur in the context of power (36). Within the healthcare professional context, we conceptualised stigma as interacting domains of stereotypes, prejudices and discrimination (37). The preliminary version of the PSSS-HCP contained 19 items within three domains (stereotypes, prejudices, discrimination). While this previous study demonstrated sufficient content validity of the PSSS-HCP, further evaluation is needed to ensure sufficient validity and reliability. The aim of this study is to evaluate the measurement properties (validity and reliability) and factor structure of the PSSS-HCP on a sample of healthcare professionals in the United Kingdom.

2. Methods

The study was pre-registered on Open Science Framework (https://osf.io/7jtr4) and approved by the University of Edinburgh and the South Central - Oxford C Research Ethics Committee (22/SC/0473). The study is part of the innovative training network ETUDE (Encompassing Training in fUnctional Disorders across Europe) (38). A STROBE checklist is included in **Appendix 1.**

2.1. Participants and design

We identified participants by searching for healthcare professional organisations across the UK. We asked these to distribute an online survey (through Qualtrics XLM) for members to complete. Criteria for eligibility included: 1) being a healthcare professional working in the UK; 2) completion of basic healthcare professional training and; 3) some experience of working with patients with PSS (though this did not have to be regular or recent contact). We considered healthcare professionals participating in the NHS Foundation Programme (training for newly qualified doctors, preceding specialist training) as eligible. Once a participant clicked on the link, they received information about the aim of the survey, followed by a mandatory consent form to be agreed. After providing consent to participate in the study, participants were eligible for the survey. The period of data collection was July 31 to October 20, 2023.

The online survey consisted of sociodemographic questions, a block on perceived adequacy of knowledge, training and resources about PSS, the preliminary version of the PSSS-HCP, the Medical Condition Regard Scale (MCRS) and the short form of the Marlowe–Crowne Social Desirability Scale (MCSDS). The MCRS explores the degree to which healthcare professionals find patients with a given medical condition to be enjoyable, treatable, and worthy of medical resources. It is a generic questionnaire that can be used across different medical conditions (39). Since the survey was exploring potentially stigmatising views, it is possible that individuals might present themselves in an overly positive way and downplay negative attributes during self-reporting. The short form of the MCSDS assesses individual level social desirability bias - whether or not participants are concerned with social approval (40). An overview of the survey is provided in **Appendix 2.**

2.2. Measurement instrument

The preliminary version of the PSSS-HCP is a self-report questionnaire, consisting of 19 items relating to stigmatisation by healthcare professionals towards people with PSS. A five-point Likert scale was used and response options were 1 = Strongly disagree, 2 = Disagree, 3 = Neither agree nor disagree, 4 = Agree and 5 = Strongly agree. Scores can range from 19 to 95 and a lower score indicates less stigma towards people with PSS. Items 1, 5, 6, 7, 15, and 16 are reverse-scored.

2.3. Taxonomy of measurement properties

We have followed the taxonomy of measurement properties of outcome measurement instruments developed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group (41, 42). We referred to the COSMIN study design checklist throughout study design (43).

2.4. Statistical analysis

All statistical analyses were conducted using IBM SPSS v27.

2.4.1. Item analysis and principal component analysis

An item analysis of the PSSS-HCP was conducted, with items with a corrected Pearson item-total correlation coefficient <0.2 being removed (44). Analysis of the structure of the PSSS-HCP was conducted using principal component analysis. The suitability of factor analysis was verified by calculating the Kaiser–Meyer–Olkin (KMO) coefficient and by performing Bartlett's test of sphericity. A KMO of ≥0.7 was considered adequate and significance of Bartlett's test of sphericity at a two-sided alpha-level of 0.05 indicated sufficient correlation between items to perform factor analyses. COSMIN guidance additionally specifies that for an appropriate sample size for factor analysis, the number of participants should be at least 5 times the number of items and ≥100 (43).

We performed the principal component analysis with oblimin rotation. We used oblimin rotation because we expected correlations between the different factors (45). Retention of factors was based on minimum eigenvalues >1.0 and inspection of the scree plot. Items not loading on a single factor with a loading coefficient of ≥0.5 were removed one at a time from the analysis, with

items loading ≥0.3 on more than one factor being considered for removal (46). We also required each factor to consist of at least three items (44).

2.4.2 Internal consistency

We calculated Cronbach's alpha to determine the internal consistency of the (sub)scales. Cronbach's alpha coefficients ≥ 0.70 for each (sub)scale were considered sufficient (47).

2.4.3. Test-retest reliability

Test-retest reliability of the scale was conducted with a sub-sample of healthcare professionals who agreed to be contacted two weeks after the first time they completed the survey. While there is no specific guidance on sufficient time intervals (44), we considered two weeks enough time that memory of completion would not influence the scores, but short enough that scores should be consistent within individuals (for example, not being impacted by a stigma reduction intervention). We calculated test-retest reliability of the total scores for each (sub)scale using the intraclass correlation (ICC) for consistency of agreement, comparing the total scores at the two time periods. An ICC ≥ 0.70 was considered sufficient (47).

2.4.4. Hypothesis testing for construct validity

We evaluated convergent validity by testing the association between the PSSS-HCP and the MCRS. In the context of healthcare professionals working with people with PSS, we previously reported that the MCRS demonstrated sufficient structural validity at a high quality of evidence, sufficient internal consistency at a high quality of evidence (Cronbach's alpha of 0.87), doubtful test-retest reliability at a moderate level of evidence (reliability test coefficient of 0.84) (33). Regarding content validity, we found that while the MCRS had many items relevant to stigma (particularly around prejudices and personal interactions), it was not sufficiently comprehensive for stigma. Therefore, we considered the MCRS's construct of 'regard' to be a similar and related construct to stigma. Using the hypotheses template suggested by the COSMIN group (47), our expectation was a moderate negative association of ≥ 0.5 between the PSSS-HCP and MCRS. We used Spearman's rank correlation coefficient to determine the relationship between the total scores of the PSSS-HCP and the MCRS.

We were also interested in the relationship between the PSSS-HCP and perceived adequate knowledge, training, resources and time during consultations with people with PSS. These perceptions were assessed through single item statements on a five-point Likert scale, ranging from 1 = Strongly disagree to 5 = Strongly agree. We did not formulate a pre-defined hypothesis for this relationship. We used Spearman's rank correlation coefficient to determine the relationship between the total scores of the PSSS-HCP and each statement about perceived adequate knowledge, training, resources and time during consultations with people with PSS.

2.4.5 Influence of social desirability bias

We also sought to determine the relationship between the PSSS-HCP and the influence of social desirability. The 13-item short form of the MCSDS has been previously shown to have sufficient structural validity and internal consistency (Kuder-Richardson 20 coefficient = 0.76) (40). We used the Spearman's rank correlation coefficient to determine the relationship between the total scores of the PSSS-HCP and the short form of the MCSDS. If items of the PSSS-HCP correlate highly with scores of social desirability, this suggests that the scale could be influenced by social desirability bias. Our expectation was a low association of ≤0.3 between the PSSS-HCP and MCSDS.

2.4.6 Floor and ceiling effects

Floor and ceiling effects were defined as the proportion of respondents scoring the minimum (floor) or maximum (ceiling) possible score across any given scale/subscale. Floor and ceiling effects were considered present if ≥15% of participants achieved minimum or maximum scores (44).

2.4.8 Missing data

Participants completing the online survey were not allowed to progress in the survey while leaving any items unanswered. However, participants completing some questions, but not completing the whole survey are included here as missing data (total across the whole survey: n=3 (2.5%). Where possible, we have analysed results using results of participants who have at least completed all relevant blocks (complete case analysis).

3. Results

3.1. Descriptive statistics

The online survey received 118 complete responses from healthcare professionals, all of which provided complete data. The PSSS-HCP instrument was completed by 121 participants, with 62.8% women, mean and SD age 44.4 years (11.9). The mean and SD total score for the 19-item scale was 45.4 (9.9)(95% C.I. 43.6 – 47.2; min-max: 22.0 – 78.0). Descriptive characteristics of participants completing the PSSS-HCP are shown in **Table 1**.

Table 1: Participant characteristics

Characteristics		**Frequency (%) (n=121)**
Age (years)	18-25	1 (0.8)
	26-44	63 (52.1)
	45-64	49 (40.5)
	≥ 65	8 (6.6)
Gender	Woman	76 (62.8)
	Man	44 (36.4)
	Prefer not to say	1 (0.8)
Type of healthcare professional	General practitioner	6 (5)
	Internal medicine specialist	28 (23.1)
	Neurologist	6 (5)
	Paediatrician	6 (5)
	Psychiatrist	32 (26.4)
	Surgeon	1 (0.8)
	Psychologist	6 (5)
	Nurse	7 (5.8)
	Physician associate	2 (1.7)
	Psychotherapist	1 (0.8)
	Physiotherapist	10 (12.5)
	Occupational therapist	1 (0.8)
	Speech and language therapist	6 (5)
	Medical graduate (e.g. NHS Foundation Programme)	2 (1.7)
	Other	7 (5.8)
Years of experience working with patients with PSS	0-1	6 (5)
	1-2	5 (4.1)
	2-5	19 (15.7)
	5-10	30 (24.8)
	10-20	22 (18.2)
	20-30	27 (22.3)
	≥ 30	12 (9.9)

3.2. Item analysis and principal component analysis

The Kaiser–Meyer–Olkin coefficient (KMO = 0.792) and Bartlett's test of sphericity ($p < 0.001$) of the 19-item scale verified suitability for the analysis. After item analysis, two items were shown to have item-total correlations below 0.2 and so were removed (items 15 and 16, see **Table 2**).

Table 2: Item-total correlations for 19-item version of PSSS-HCP

Item (* = item is reverse scored)		**Item total correlation**
1*	Anybody could develop persistent somatic symptoms under the right circumstances	0.349
2	Most people with persistent somatic symptoms don't try hard enough to get better.	0.412
3	There is little I can do to help people with persistent somatic symptoms.	0.436
4	I find that people with persistent somatic symptoms often exaggerate their symptoms.	0.468
5*	It is important to encourage hope for improved quality of life for people with persistent somatic symptoms	0.389
6*	I feel as comfortable talking to a person with persistent somatic symptoms as I do other people	0.552
7*	I find it satisfying to support people with persistent somatic symptoms	0.605
8	People with persistent somatic symptoms evoke negative feelings in me, such as aversion, stress or impatience.	0.624
9	I secretly hope that people with persistent somatic symptoms will not return.	0.630
10	I often feel uncomfortable about providing care to people with persistent somatic symptoms.	0.624
11	I struggle to feel compassion for people with persistent somatic symptoms.	0.554
12	Sometimes I lose patience with people with persistent somatic symptoms.	0.311
13	If I had a choice, I would rather not care for people with persistent somatic symptoms.	0.522
14	When someone with persistent somatic symptoms presents a new symptom or complaint, I am less likely to pay attention to other diseases than in other people.	0.416
15*	If a colleague told me they had persistent somatic symptoms, this would not change how willing I was to work with them	**0.149**
16*	Employers should hire a person with persistent somatic symptoms if they are the best person for the job	**0.188**
17	If I developed persistent somatic symptoms, I would be reluctant to tell my friends.	0.394
18	If I were under treatment for persistent somatic symptoms, I would try not to disclose this to any of my colleagues.	0.464
19	I would be reluctant to seek help if I knew I had persistent somatic symptoms.	0.439

Bold = removed from PSSS-HCP due to low item-total correlation.

Principal component analysis of the remaining items revealed a three-factor structure accounting for 60.5% of the total variance explained. Four items were removed because they did not have sufficient loading on any of the factors (item 10 in **Table 2**), or insufficient loading across one or more factors (item 5, 12 and 14 in **Table 2**). **Appendix 3** summarises the iterative stages of the analysis. The three factors were reflected to refer to 1) othering (four items), 2) uneasiness in interaction (six items) and non-disclosure (three items). **Table 3** shows the factor loadings after rotation. In order of explained total variance, the uneasiness in interaction subscale explained 35.4% of the total variance, the non-disclosure subscale explained 14.0% of the total variance, and the othering subscale explained 11.1% of the total variance. All items had factor loadings >0.6, with all items clearly loading onto a single factor.

Table 3: Factor loadings for the 13-item version of PSSS-HCP

	Rotated factor loadings		
Item (* = reverse scored)	**Factor 1: Othering**	**Factor 2: Uneasiness in interaction**	**Factor 3: Non-disclosure**
Anybody could develop persistent somatic symptoms under the right circumstances*	**0.613**[1]	-0.022	0.047
Most people with persistent somatic symptoms don't try hard enough to get better.	**0.763**[1]	-0.016	0.006
There is little I can do to help people with persistent somatic symptoms.	-0.098	**0.660**[2]	-0.009
I find that people with persistent somatic symptoms often exaggerate their symptoms.	**0.788**[1]	0.055	-0.013
I feel as comfortable talking to a person with persistent somatic symptoms as I do other people*	0.148	**0.679**[2]	-0.045
I find it satisfying to support people with persistent somatic symptoms*	0.055	**0.827**[2]	-0.025
People with persistent somatic symptoms evoke negative feelings in me, such as aversion, stress or impatience	0.107	**0.696**[2]	0.080
I secretly hope that people with persistent somatic symptoms will not return.	0.143	**0.663**[2]	0.081
I struggle to feel compassion for people with persistent somatic symptoms.	**0.638**[1]	0.189	0.000
If I had a choice, I would rather not care for people with persistent somatic symptoms.	-0.053	**0.801**[2]	-0.014
If I developed persistent somatic symptoms, I would be reluctant to tell my friends.	0.098	-0.148	**0.932**[3]

Table 3: Continued

Item (* = reverse scored)	Rotated factor loadings		
	Factor 1: Othering	**Factor 2: Uneasiness in interaction**	**Factor 3: Non-disclosure**
If I were under treatment for persistent somatic symptoms, I would try not to disclose this to any of my colleagues.	0.122	-0.036	**0.876**[3]
I would be reluctant to seek help if I knew I had persistent somatic symptoms.	-0.211	0.277	**0.735**[3]

1: Items included in factor 1 'othering'; 2: Items included in factor 2 'uneasiness in interaction'; 3: items included in factor 3 'Non-disclosure'

Means, standard deviations, and range of scores for each scale of the PSSS-HCP are shown in **Table 4**. The mean total score and SD for the 13-item scale was 32.2 (7.8) (95% C.I. 30.8 – 33.6). Scores ranged from 14.0 to 57.0. For the 13-item scale, the mean total score for the othering subscale was 8.2 (95% C.I. 7.7 to 8.6), the mean total score for the uneasiness in interaction subscale was 15.2 (95% C.I. 14.3 to 16.1) and the mean total score for the non-disclosure subscale was 8.9 (95% C.I. 8.4 to 9.3).

Table 4: Mean total score, standard deviation and range for each instrument

Instrument	n = completed block	Mean total score (SD)	Minimum recorded score	Maximum recorded score	Range recorded score
PSSS-HCP-19	121	45.4 (9.9)	22.00	78.0	56.0
PSSS-HCP-13[a]	121	32.2 (7.8)	14.0	57.0	43.0
Subscale 1: Othering[b]	123	8.2 (2.7)	4.0	16.0	12.0
Subscale 2: Uneasiness in interaction[b]	121	15.2 (4.8)	6.0	27.0	21.0
Subscale 3: Non-disclosure[b]	121	8.9 (2.7)	3.0	15.0	12.0
MCRS	121	42.9 (8.0)	20.0	60.0	40.0
MCSDS	118	20.7 (2.5)	14.0	26.0	12.0

a Scores for the PSSS-HCP-13 can range from 13 to 65.

b Scores for the othering subscale can range from 4 to 20, scores for the uneasiness in interaction subscale can range from 6 to 30, and scores for the non-disclosure scale can range from 3 to 15.

3.3. Internal consistency

The Cronbach's alpha coefficient of the 19-item version of the PSSS-HCP was α = 0.86. The Cronbach's alpha coefficient of the 13-item version of the PSSS-HCP was α = 0.84 (α = 0.71 for othering subscale, α = 0.84 for uneasiness in interaction subscale and α = 0.82 for non-disclosure subscale). The correlation between subscales ranged from 0.22 to 0.46 **(Appendix 4**).

3.4. Test-retest reliability

Of the total participants opting in for retesting in two weeks (n = 66), 21 participants completed the retest (response rate from opt-in = 31.8%). The test-retest reliability of the total score of both the 19 and the 13-item scale was high (ICC PSSS-HCP-19 = 0.98 (95% CI 0.96 to 0.99, p<0.001; ICC PSSS-HCP-13 = 0.97 (95% CI 0.94 to 0.99, p<0.001). The test-retest reliability of the total score for each subscale was also high: othering ICC = 0.97 (95% CI 0.93 to 0.99, p<0.001), uneasiness in interaction ICC = 0.97 (95% CI 0.93 to 0.99, p<0.001), non-disclosure ICC = 0.88 (95% CI 0.70 to 0.95, p<0.001).

3.5. Construct validity

Both the 19-item version and the 13-item version of the PSSS-HCP had a strong negative and significant correlation with the MCRS (PSSS-HCP-19 r_s = –0.80, p<0.001; PSSS-HCP-13 r_s = -0.83, p<0.001). This suggests that healthcare professionals scoring higher for stigma held less positive regard towards patients with PSS.

Table 5: Correlations of total PSSS-HCP scores and perceived adequate knowledge, training, tools and time in consultations

	Perceived adequate knowledge	**Perceived adequate training**	**Perceived adequate tools**	**Perceived adequate time in consultations**
PSSS-HCP-19	-0.47**	-0.47**	-0.47**	-0.48**
PSSS-HCP-13	-0.48**	-0.45**	-0.51**	-0.49**

**p<0.01

Mean scores and SD for perceived adequate knowledge were 3.7 (1.0), for perceived adequate training were 3.1 (1.1), for perceived adequate tools to manage and/or diagnose patients were 3.0 (1.1), for perceived adequate time

in consultations to discuss PSS were 2.9 (1.3). The total score of the PSSS-HCP was moderately negative and significantly correlated with perceived adequate knowledge, training, tools and time during consultations **(Table 5).**

3.6. Social desirability bias

Both the 19-item version and the 13-item version of the PSSS-HCP had an insignificant correlation with the MCSDS (PSSS-HCP-19 r_s = –0.15, *p*=0.15; PSSS-HCP-13 r_s = -0.07, *p*=0.46). This suggests that there was no significant social desirability bias on answering the PSSS-HCP.

3.7. Floor and ceiling effects

There were no ceiling or floor effects found for either the total scales of the 19 and 13-item versions of the PSSS-HCP, or the othering, uneasiness in interaction or non-disclosure subscales. Percentages of minimum and maximum scores reached for each scale and subscale are included in **Appendix 5.**

4. Discussion

4.1 Main findings

In this study we present the validated version of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals, the PSSS-HCP. The PSSS-HCP has demonstrated sufficient convergent validity, internal consistency, re-test reliability, and minimal influence of social desirability bias among a sample of healthcare professionals in the UK. The PSSS-HCP, guidance about use and scoring instructions are available in **Appendix 6**.

4.2 Comparison to literature

Three factors were found from the principal component analysis of the PSSS-HCP. These subscales, including othering (four items), uneasiness in interaction (six items), and non-disclosure (three items) all demonstrated sufficient internal consistency. While these factors broadly mapped to our conceptualisation of items during development (stereotypes, prejudice, discrimination), we felt that these names better reflected the specific stigma

elements in the context of PSS. Items in the 'othering' subscale explore the extent to which healthcare professionals view people with PSS as fundamentally different and less legitimate than other people. Items in the 'uneasiness in interaction' subscale focus on attitudes and comfort about personal interactions with people with PSS.

The items in the non-disclosure subscale ask healthcare professionals to consider themselves in the prospective role of a person with PSS. Perspectives on disclosure and help seeking may indicate whether a healthcare professional feels that a stigmatised condition is something needed to keep concealed or to be ashamed of (48). Further, disclosure and help seeking are important steps in reducing self-stigma, when people internalise negative beliefs and attitudes. Our principal component analysis placed these items in a separate subscale, which we found was correlated (albeit less strongly correlated) to the other subscales. Both the distinction of disclosure as a separate factor and the reduced correlation strength to the other factors is consistent with psychometric testing of mental health stigma scales among healthcare professionals (49-51). Therefore, the non-disclosure subscale contributes to overall PSS related stigmatisation while also being distinct. This is also relevant because many healthcare professionals also experience PSS including chronic pain (52, 53).

We found that items that we had conceptualised as relating to discrimination and behavioural intentions were removed from the final 13-item version. In some cases these were placed in other subscales (e.g. 'There is little I can do help people with persistent somatic symptoms' was found to relate more to uneasiness during interactions). Other items such as those relating to social distance were removed during item analysis. This finding was consistent with our previous development study, where we found that many traditional approaches to social distance were not perceived as relevant by participants (35). Reasons for this might include specific dimensions of the stigmatised condition (such as low perceived peril and low perceived dangerousness) (54, 55). It may well be that the behavioral component of PSS related stigmatisation is best addressed through other methods (e.g. analysing communication during consultation or treatment behaviour), while this stigma scale focusses particularly on beliefs and attitudes of healthcare professionals. Indeed, there is increasing focus on identifying specific processes of social distancing relevant to PSS in clinical consultations (56).

We found that the total PSSS-HCP score was moderately correlated with lower perceived adequate knowledge about PSS, lower perceived adequate training, lower perceived adequate resources and lower perceived adequate time during consultations. We know that insufficient knowledge is often associated with negative attitudes (9, 57). For example, healthcare professionals have reported a desire for further training, and desire to avoid patients with PSS due to a lack of confidence and competence (21). However, more knowledge about a healthcare condition as a healthcare professional might lead to increased pessimism to long term outcomes (58). So while the correlation between the PSSS-HCP score and perceived adequate knowledge, training, resources and consultation time are consistent with other PSS research (and therefore indicative of construct validity), we did not formally hypothesise a relationship between these aspects.

We found that the PSSS-HCP did not correlate significantly with the short form of the MCSDS. This suggests that the responses to the PSSS-HCP were not strongly related to social desirability. Limitations of self-reported stigma instruments are well-reported, such as participants not explicitly endorsing negative attitudes, or preferring not to reveal their attitudes through a social desirability bias (33, 59). This is particularly important in the context of healthcare professionals when there are social and professional pressures against explicit stigmatising behaviour. While social desirability remains a threat to the validity of stigma instruments in general, we have shown through development and validation that it was not strongly related to responses to the PSSS-HCP scale.

4.3 Strengths and limitations

The most important strength of our study was that we have followed COSMIN guidelines throughout the development and validation of the PSSS-HCP. While there has been enduring interest in stigma by healthcare professionals, validation of measurement instruments in this population is limited. We have previously demonstrated sufficient content validity of the PSSS-HCP (35), building a stigma model from well-recognised frameworks. We used the COSMIN study design checklist to assess sufficiency of each measurement property (43), and used a systematic approach to reach the final 13-item version.

An additional strength is that we have considered the potential impact of social desirability bias in the study design. While there is recent debate

about what social desirability scales essentially measure (60), we believe it is important to consider the potential role of response biases when evaluating self-reported measurement instruments. We have approached this in a mixed-methods design, combined with the qualitative assessment of social desirability in the developmental phase of the PSSS-HCP (35). Further research should continue to explore the influence of social desirability for example by testing the PSSS-HCP alongside implicit measurement instruments or more direct behaviour.

There are also limitations of this study. One limitation is the relatively limited number of participants, which is particularly evident for our cohort of participants opting in for test-retest reliability. Since the PSSS-HCP demonstrated sufficient internal consistency and the results for test-retest reliability were very high, we can be reasonably confident of sufficient reliability. However, these results for test-retest reliability should be considered as indicative and require further testing in future research. Further, we ensured suitability of the sample for principal component analysis.

Another limitation of this study is that there may be a recruitment bias in participating healthcare professionals. Since we contacted organisations and asked them to distribute, participants were self-selecting and likely more willing to share their beliefs and attitudes. Therefore, it is possible that this sample of healthcare professionals is not representative of the wider population of the healthcare professionals working in the UK. This was reflected in the high levels of years of experience working with people with PSS and moderate levels of perceived adequate knowledge and training. Further, while the study sample consisted of healthcare professionals of various professions, ages, and work experience, the majority of participants were internal medicine specialists or psychiatrists. While these are important groups of healthcare professionals working with patients with PSS in secondary care, we recognise that healthcare professionals such as general practitioners play a critical role as first point of contact and providing a gatekeeper function. There are two potential implications of this regarding generalisability of results. First, it could be hypothesised that this recruitment bias resulted in a study sample that had less stigmatising attitudes and beliefs towards people with PSS. However, since the aim of this study was to assess the measurement properties of the PSSS-HCP rather than measure stigmatisation of the population of healthcare professionals outright, we think the impact of this is limited. Second, this population sample could affect the

factor structure identified here. A larger sample is needed to further explore and confirm the factor structure identified here. This could also explore how often healthcare professionals interact with people with PSS, as well as the overall amount of experience.

4.4 Implications for future research and policy

Further research should evaluate the validity of the PSSS-HCP in different cultural and healthcare professional contexts. This research should confirm the structure of the PSSS-HCP and further evaluate retest reliability. Responsiveness to change should also be evaluated, including the minimally important change (the meaning of changes in PSSS-HCP score over time), which is an important aspect of intervention evaluation. Further, potential cut-off scores for severity banding of the PSSS-HCP could have policy implications, such as helping target stigma reduction interventions.

As we reported in the development of the PSSS-HCP, terminology used to describe persistent somatic symptoms continues to be debated (1, 61-63). We consider the PSSS-HCP suitable for testing stigmatisation in related conditions such as functional disorders, or syndromes including irritable bowel syndrome or fibromyalgia in future versions of the scale without compromising content validity (so long as appropriate definitions are provided). Future research should evaluate the measurement properties of the PSSS-HCP used in these specific contexts.

5. Conclusion

We present the validated version of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals, the PSSS-HCP. The 13-item PSSS-HCP can be used measure PSS stigmatisation by healthcare professionals. The PSSS-HCP has demonstrated sufficient convergent validity, internal consistency and test re-test reliability, and minimal influence of social desirability bias among healthcare professionals working in the UK. The PSSS-HCP has demonstrated potential to measure important aspects of stigma and provide a foundation for stigma reduction intervention evaluation.

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Supplementary material

Appendix 1: STROBE Statement

Available on online supplementary material

Appendix 2: Online survey printout (Qualtrics XM)

Available on online supplementary material

Appendix 3: Summary of iterations of principal component analysis

From initial inspection of eigenvalues (>1), as many as five factors could be meaningful. From scree plot inspection, a point of inflexion was not clear, but could be three and four factors. Analysis was run extracting each number of factors with oblimin rotation. A three factor rotated solution made most sense conceptually to stigma, linked broadly to cognitive processes (stereotypes), emotional/ affective processes (prejudices) and items linked to non-disclosure and help-seeking.

Round of principal components analysis (three factors extracted, oblimin rotation)	Number of items included in analysis	Interpretation of factors identified	Interpretation of item loadings	Recommendation
1	17	*Component 2 relates strongly to disclosure and help-seeking (n=3). *Component 3 relates to stereotypes (n=4). *Component 1 relates to prejudices (negative emotions or attitudes) (n=10)	One item loading poorly (<0.3) on only one factor (item 12): "Sometimes I lose patience with people with persistent somatic symptoms."	Remove item 12: "Sometimes I lose patience with people with persistent somatic symptoms."
2	16	*Component 2 relates strongly to disclosure and help-seeking (n=3). *Component 3 relates to stereotypes (n=4). *Component 1 relates to prejudices (negative emotions or attitudes) (n=8). *One item loading poorly on all factors (n=1)	One item loading poorly (<0.3) on all factors (Item 14): "When someone with persistent somatic symptoms presents a new symptom or complaint, I am less likely to pay attention to other diseases than in other people."	Remove item 14: "When someone with persistent somatic symptoms presents a new symptom or complaint, I am less likely to pay attention to other diseases than in other people."
3	15	*Component 2 relates strongly to disclosure and help-seeking (n=3). *Component 3 relates to stereotypes (n=4). *Component 1 relates to prejudices (negative emotions or attitudes) (n=7). *One item loading poorly across two factors (n=1)	*One item loading poorly (<0.3) across two factors (Item 10): "I often feel uncomfortable about providing care to people with persistent somatic symptoms."	Remove item 10: "I often feel uncomfortable about providing care to people with persistent somatic symptoms."

Appendix 3: Continued

Round of principal components analysis (three factors extracted, oblimin rotation)	Number of items included in analysis	Interpretation of factors identified	Interpretation of item loadings	Recommendation
4	14	*Component 2 relates strongly to disclosure and help-seeking (n=3). *Component 3 relates to stereotypes (n=4). *Component 1 relates to prejudices (negative emotions or attitudes) (n=6). *One item loading poorly across one factor (n=1)	One item loading poorly (<0.3) on one factor (item 5): "It is important to encourage hope for improved quality of life for people with persistent somatic symptoms"	Remove item 5: "It is important to encourage hope for improved quality of life for people with persistent somatic symptoms"
5	13	*Component 2 relates strongly to disclosure and help-seeking, labelled "non-disclosure" (n=3). *Component 3 relates to stereotypes, labelled "othering" (n=4). *Component 1 relates to prejudices (negative emotions or attitudes), labelled "uneasiness in interaction" (n=6).	All items load highly (>0.6) on one factor.	13-item solution across three factors, explaining 60.52% variance.

Appendix 4: Correlations between subscales (Spearman's rank correlation coefficient)

	Othering subscale	Uneasiness in interaction subscale	Non-disclosure subscale
Othering subscale	1.000	.459**	.217*
Uneasiness in interaction subscale	.459**	1.000	.307**
Non-disclosure subscale	.217*	.307**	1.000

**p<0.01 *p<0.05

Appendix 5: Percentages of possible minimum and maximum scores reached for each scale and subscale

Instrument	n = completed block	Mean total score (SD)	Minimum possible score	Minimum recorded score	Maximum possible score	Maximum recorded score	Percentage of participants with minimum score (floor)*	Percentage of participants reaching maximum score (ceiling)*
PSSS-HCP-19	121	45.4 (9.9)	19.0	22.00	95.0	78.0	0.0	0.0
PSSS-HCP-13	121	32.2 (7.8)	13.0	14.0	65.0	57.0	0.0	0.0
Othering subscale	123	8.2 (2.7)	4.0	4.0	20.0	16.0	7.3	0.0
Uneasiness in interaction subscale	121	15.2 (4.8)	6.0	6.0	30.0	27.0	1.7	0.0
Non-disclosure subscale	121	8.9 (2.7)	3.0	3.0	15.0	15.0	3.3	0.8

*Floor and ceiling effects were considered present if ≥15% of participants achieved minimum or maximum scores.

Appendix 6: Persistent Somatic Symptom Stigma scale for Healthcare Professionals, guidance about use and scoring instructions

The PSSS-HCP is a self-report questionnaire, consisting of 13 items relating to stigmatisation by healthcare professionals towards people with persistent somatic symptoms (PSS).

Instructions for use

A five-point Likert scale is used and response options are: 1 = Strongly disagree, 2 = Disagree, 3 = Neither agree nor disagree, 4 = Agree and 5 = Strongly agree. Scores can range from 13 to 65 and a lower score indicates less stigmatisation towards people with PSS. Items 1, 5, and 6 are reverse-scored. The instruction for participants was: "For the following questions, please select the answer that best corresponds to your views."

Items of the PSSS-HCP

	Item (* = reverse scored)
1*	Anybody could develop persistent somatic symptoms under the right circumstances
2	Most people with persistent somatic symptoms don't try hard enough to get better.
3	There is little I can do to help people with persistent somatic symptoms.
4	I find that people with persistent somatic symptoms often exaggerate their symptoms.
5*	I feel as comfortable talking to a person with persistent somatic symptoms as I do other people
6*	I find it satisfying to support people with persistent somatic symptoms
7	People with persistent somatic symptoms evoke negative feelings in me, such as aversion, stress or impatience
8	I secretly hope that people with persistent somatic symptoms will not return.
9	I struggle to feel compassion for people with persistent somatic symptoms.
10	If I had a choice, I would rather not care for people with persistent somatic symptoms.
11	If I developed persistent somatic symptoms, I would be reluctant to tell my friends.
12	If I were under treatment for persistent somatic symptoms, I would try not to disclose this to any of my colleagues.
13	I would be reluctant to seek help if I knew I had persistent somatic symptoms.

Explanation of subscales

The PSSS-HCP contains 13 items across three subscales:

- Items in the 'othering' subscale (items 1, 2, 4, 9) explore the extent to which healthcare professionals view people with PSS as fundamentally different and often less legitimate than other people.
- Items in the 'uneasiness in interaction' subscale (3, 5, 6, 7, 8, 10) focus on attitudes about personal interactions with people with PSS.
- Items in the non-disclosure subscale (items 11, 12, 13) ask healthcare professionals to consider themselves in the role of a person with PSS. Perspectives on disclosure and help seeking may indicate whether a healthcare professional feels that PSS is something needed to keep concealed or to be ashamed of.

Standardisation

We have developed and evaluated the PSSS-HCP for healthcare professionals who have completed basic training, and some experience of working with patients with persistent somatic symptoms. This experience does not have to be recent or regular.

The terminology around persistent somatic symptoms continues to be debated. During development we tested understanding of these definitions. We consider it likely that the term 'persistent somatic symptom' can be substituted for specific persistent somatic symptoms, syndromes or functional disorders without compromising content validity, so long as appropriate definitions are provided before administration. Further research should validate the PSSS-HCP in these specific contexts.

Feasibility information

There are no costs associated, or specialist equipment requirements for administering the PSSS-HCP.

Permission to use

This project has received funding from the European Union's Horizon 2020 research and innovation programme under the Marie Skłodowska-Curie grant agreement No 956673. The PSSS-HCP can be used freely without permission of authors so long as the original authors and this publication is properly cited.

Chapter 5

Validation of the Dutch version of the Persistent Somatic Symptoms Stigma Scale for Healthcare Professionals (PSSS-HCP)

McGhie-Fraser B, Lucassen P, Ballering A, van der Linden L, Brouwers E, olde Hartman T, van Dulmen S. Validation of the Dutch version of the Persistent Somatic Symptoms Stigma Scale for Healthcare Professionals (PSSS-HCP).

Submitted.

Abstract

Objectives: Reducing stigma by healthcare professionals towards people with persistent somatic symptoms (PSS) is a priority in research and clinical practice. We recently developed and validated an instrument to measure stigmatisation towards people with PSS by healthcare professionals: the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). This study developed a Dutch version of the PSSS-HCP and evaluated its measurement properties.

Methods: The PSSS-HCP was tested with 157 healthcare professionals across the Netherlands to evaluate its measurement properties. We conducted a confirmatory factor analysis to examine dimensionality and model-based reliability. Test re-test reliability was conducted with a sub-sample of 49 participants at a two-week interval. We established hypotheses for known groups validity, and convergent validity with the Medical Condition Regard Scale (MCRS). We tested for social desirability bias with the Marlowe Crowne Social Desirability Scale (MCSDS).

Results: Model fits were similar between a two-factor correlated model and a modified bifactor model, and bifactor analysis suggested that the PSSS-HCP was mostly influenced by a general factor. Reliability was strong for the total scale (ω = 0.87) and for the non-disclosure subscale (ωS = 0.80). Test-retest reliability was excellent for the total scale (ICC = 0.90) and non-disclosure subscale (ICC = 0.81). All prior hypotheses for construct validity were confirmed with no evidence of social desirability bias. Participants reported that people with PSS were taken less seriously (80.2%), treated as less of a priority (67.5%) and provided with poorer quality care (61.2%) when compared to people with a clearer pathology.

Discussion: Our results demonstrated sufficient construct validity, test-retest reliability, model-based reliability, and no evidence of social desirability bias. The PSSS-HCP can be used to measure stigmatisation by healthcare professionals towards people with PSS in the Dutch context. We recommended scoring the PSSS-HCP using a total score and an optional additional non-disclosure subscale score.

1. Introduction

People living with persistent somatic symptoms (PSS) do not only face distress and suffering caused by the symptoms themselves, reporting stigmatising attitudes and behaviours from healthcare professionals while seeking medical help (1-6). Specifically, people have reported that their symptoms are not taken seriously, dismissed as emotional problems, or outright fabrication, and their truthfulness and accuracy in describing symptoms is questioned (1-5). Stigmatisation is a dynamic process where elements of labelling, stereotyping, separation, status loss, and discrimination occur in the context of power (7). Stigma is associated with negative health outcomes and poor quality of life for people with PSS including increased depression and anxiety (8), decreased wellbeing (9), and non-adherence to treatment (10).

Persistent somatic symptoms describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months (11, 12). These include single symptoms, combinations of symptoms, or syndromes meeting the criteria for functional disorders such as fibromyalgia or irritable bowel syndrome. An important contributing factor for stigmatisation by healthcare professionals towards people with PSS is a lack of perceived explicability of symptoms. A tension between the severity and persistence of symptoms being presented against biomedical expectation, or an absence of structural abnormalities, creates challenges in communication during the clinical consultation (13). This creates a perception that people with PSS are less legitimate than people with health conditions with clearer pathologies and explanations of illness (4, 14). This difference is further reflected and potentially reinforced by fragmented healthcare systems for people with PSS (15), and a lack of personal competence and confidence in treating people with PSS (16). When a lack of security is provided to patients about the nature or reality of their symptoms, this can cause further distress and contribute to internalisation of stigma (17). Therefore, reducing stigmatisation by healthcare professionals is increasingly a priority in research and clinical practice.

Key to this agenda is the development of specific interventions to reduce stigma towards people with PSS, and the accompanying use of valid and reliable measurement instruments to assess their effectiveness. Despite increased clinical and research attention to stigma, very few measurement instruments have had their measurement properties evaluated for use

by healthcare professionals (18). We recently developed and validated a new stigma scale to measure stigmatisation towards people with PSS by healthcare professionals: the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP) (19). In a sample of healthcare professionals in the United Kingdom, we found that the 13-item version of the scale showed a three-factor solution, demonstrating sufficient construct validity, internal consistencies, and test-retest reliability (20).

We identified further need to evaluate the measurement properties of the PSSS-HCP. Given the limited availability and sufficient measurement properties of Dutch scales to measure stigmatisation, we aimed to develop a translated version of the PSSS-HCP that can be used in Dutch healthcare professional populations. Second, we aimed to assess the previously identified factor structure of the PSSS-HCP in the Dutch context, including the viability of using total scales and subscale scores. Third, we aimed to measure test-retest reliability in a larger sample of healthcare professionals. Fourth, we wanted to revisit the PSSS-HCP with a larger healthcare professional population, since participants for the original validation had high representation from people with extensive experience of working with people with PSS, and moderate self-perceived levels of knowledge, training, tools and time available in consultations with people with PSS. Therefore the aim of this study is to evaluate the measurement properties (validity and reliability) of the PSSS-HCP on a sample of healthcare professionals in the Netherlands.

2. Materials and Methods

This study was a mixed methods study in two parts: a qualitative part which developed translated items of the PSSS-HCP, and a quantitative part using an online survey to measure stigmatising attitudes of healthcare professionals towards people with PSS. The study was pre-registered on Open Science Framework (https://osf.io/j96yv). The Research Ethics Committee Arnhem/Nijmegen reviewed this project and supporting documentation, considering it not applicable for the Medical Research Involving Human Subjects Act (WMO) (registration number 2024-17028). The study is part of the innovative training network ETUDE (Encompassing Training in fUnctional Disorders across Europe) (21).

The Dutch version of the PSSS-HCP was developed following the taxonomy of measurement properties of outcome measurement instruments developed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group (22, 23). We referred to the COSMIN study design checklist throughout study design (24).

2.1 Translation of PSSS-HCP into Dutch

Translation of the PSS-HCP involved the following stages according to COSMIN study design recommendations (24):

1) Translation of the English version to Dutch language: The original PSSS-HCP was translated by two Dutch native speakers independently with proficiency in English (LvdL and PL), after which the versions were compared. Differences were resolved through consensus until a draft Dutch version was agreed.
2) Back-translation into English: The scale items were back-translated into English by a native English speaker with proficiency in Dutch (BMF).
3) Synthesis: Translations were compared by all three translators and differences were resolved in discussion until consensus was reached.
4) Research committee: The original English version, Dutch version and synthesis of translation differences were discussed by a research committee consisting of a medical sociologist/anthropologist, general practitioners, a medical student, a psychologist, an epidemiologist and an expert in mental health stigma.
5) Pilot testing through cognitive interviews: The pilot version of the PSSS-HCP was tested in two rounds on Dutch healthcare professionals through a convenience sample. Participants include five general practitioners, one psychologist and a psychiatrist in specialisation. Items were assessed through cognitive interviews (conducted by LvdL). We adopted a hybrid approach of the 'think aloud' method and verbal probing (25, 26). Items were assessed for content validity (relevance, comprehensibility, comprehensiveness), with items that were difficult to understand being highlighted for revision. A preliminary version of the Dutch PSSS-HCP was developed according to participant feedback.
6) The preliminary translation was presented to two expert groups for feedback, comprising both healthcare professionals and people with lived experience of PSS: 1) Stichting De Bagagedrager who develop anti-stigma tools and workshops including for healthcare professionals and; 2) the advisory group of the Dutch Network for PSS (NALK). Based on

this feedback, the final version was prepared. The Dutch translation is presented in **Appendix 1, supplementary material.**

2.2. Participants and design

Participants were gathered through both contacting multiple Dutch healthcare professional organisations, asking for their members to participate, and inquiring with already existing clinical and research contacts, all within the Netherlands. Participants were asked to complete an online survey (through LimeSurvey Cloud Version 6.6.1). The inclusion criteria of the study were: 1) being aged 18 years or above; 2); being a healthcare professional working in the Netherlands; 3) completion of basic healthcare professional training; and 4) some experience of working with patients with PSS (though this did not have to be regular or recent contact); 5) ability and willingness to consent to participate in the research. Once a participant clicked on the link, they received information about the aim of the survey, followed by a mandatory consent form to be agreed. After providing consent to participate in the study, participants were eligible for the survey. The period of data collection was March 26 to May 24, 2024. A STROBE checklist is included in **Appendix 2, supplementary material**.

2.3. Measurement instruments

The online survey consisted of several blocks, beginning with sociodemographic questions, and perceived adequacy of knowledge, training and resources about PSS. This was followed by the PSSS-HCP, a block on perceived stigma for people with PSS, the Medical Condition Regard Scale (MCRS) and a short form of the Marlowe–Crowne Social Desirability Scale (MCSDS).

2.3.1. *Perceived adequacy of knowledge, training and resources*

We asked participants to reflect on their own adequacy of knowledge, training, tools and time during consultations for people with PSS. These perceptions were assessed through four single item statements on a five-point Likert scale, ranging from 1 = Strongly disagree to 5 = Strongly agree.

2.3.2. *Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP)*

The PSSS-HCP is a self-report questionnaire, measuring stigmatising attitudes by healthcare professionals towards people with PSS. The subscales

previously identified in the validation of a sample of healthcare professionals in the UK included: 1) othering (the extent that people with PSS are different and often less legitimately ill than other people); 2) uneasiness in interactions (comfort about personal interaction with people with PSS); and 3) non-disclosure (reluctance to reveal prospective illness of PSS to other people). For the total scale, the internal consistency was $\alpha = 0.84$. For the subscales, the internal consistency was $\alpha = 0.71$ (othering), $\alpha = 0.84$ (uneasiness in interactions) and $\alpha = 0.82$ (non-disclosure) (20). Test-retest reliability was ICC=0.97 for the total scale, and ICC=0.88-0.97 for the subscales. A five-point Likert scale is used and response options are between 1 = Strongly disagree and 5 = Strongly agree. Total scores can range from 13 to 65 and a lower score indicates less stigma towards people with PSS. Three items are reverse-scored.

2.3.3. Perceived stigma

Perceived stigma was assessed by asking participants to reflect on how much they believed that people with PSS were stigmatised compared to people with conditions with a clearer pathology. These perceptions were assessed through single item statements on a five-point Likert scale, ranging from 1 = Definitely not to 5 = Definitely.

2.3.4. The Medical Condition Regard Scale (MCRS)

The MCRS explores the extent to which healthcare professionals find patients with a given medical condition to be enjoyable, treatable, and worthy of medical resources (27). A six-point Likert scale is used (1 = Strongly disagree, 6 = Strongly agree). In a previous systematic review of measurement instruments we found that the MCRS demonstrated sufficient structural validity at a high quality of evidence, sufficient internal consistency at a high quality of evidence (Cronbach's alpha of 0.87), test-retest reliability at a moderate level of evidence (reliability test coefficient of 0.84, with doubtful methodological quality) (18). The MCRS had previously been translated into Dutch through a forward-backward method (28).

2.3.5. Marlowe Crowne Social Desirability Scale Short Form (MCSDS)

Since the survey was exploring potentially stigmatising views, it is possible that individuals might present themselves in an overly positive way and downplay negative attributes during self-reporting. The Dutch short form of the Marlowe Crowne Social Desirability Scale (MCSDS) explores the extent to which participants are influenced by social desirability bias (29).

2.4. Statistical analysis

All statistical analyses were conducted using IBM SPSS v27. Descriptive data were presented as mean, minimum and maximum, standard deviation, and percentages. The SPSS AMOS module v29 was used for confirmatory factor analysis. Additional bifactor statistics were calculated using a specific 'Bifactor Indices Calculator' tool (30).

2.4.1. Confirmatory factor analysis

We conducted a confirmatory factor analysis (maximum likelihood estimation) to examine the dimensionality of the PSSS-HCP and the viability of calculating subscale scores. We compared the fit of three models: a unidimensional model, a correlated three factor model that followed from our initial validation of the PSSS-HCP in a sample of healthcare professionals in the UK (20), and a bifactor model.

We applied several restrictions to these models. In the unidimensional model, all items loaded on one general factor only. In the three-factor model, items were allowed to load on only one of the three specific factors. The factors were free to correlate with each other. In the bifactor model, each item was allowed to load only on its specific factor and the general factor, and factors were not allowed to correlate with each other. Model fit was examined using the chi-square statistic, the comparative fit index (CFI), the standardised root mean square residual (SRMR) and the root mean square error of approximation (RMSEA). Sufficient model fits were considered to be CFI ≥ 0.95, SRMR ≤ 0.08, and RMSEA ≤ 0.06 (31).

2.4.2. Model based reliability

We assessed the dimensionality and reliability of the PSSS-HCP using omega coefficient (ω), hierarchical omega coefficient (ωH), explained common variance (ECV), and percent uncontaminated correlations (PUC).

The omega coefficient is an index of model-based reliability. Values range between 0 (indicating no reliability) and 1 (indicating perfect reliability) (32). For comparison, we also calculated Cronbach's alpha coefficient to determine the internal consistency of the (sub)scales. Cronbach's alpha coefficients $\geq$ 0.70 for each (sub)scale were considered sufficient (31).

The hierarchical omega coefficient (the scale-level variance explained by a general factor) ECV (the item-level variance explained by a general factor), and the PUC (proportion of total number of item correlation coefficients affected only by a general factor) are all indicators of dimensionality (33). If the PUC values are higher than 0.80, the general ECV values are larger than 0.60, and the ωH values are higher than 0.70, the presence of some dimensionality is not severe enough to disqualify the interpretation of the instrument as 'essentially' unidimensional (33).

2.4.3. Test-retest reliability

Test-retest reliability of the scale was conducted with a sub-sample of healthcare professionals who agreed to be contacted two weeks after the first time they completed the survey. We considered that a two-week time interval was long enough that memory of completion would not influence the scores, but short enough that scores should be consistent within individuals (for example, not being impacted by a stigma reduction intervention). We calculated test-retest reliability of the total scores for the total scale/ subscales using the intraclass correlation (ICC) for consistency of agreement using an absolute, two-way mixed-effects model. We compared the total scores at the two time periods. We considered an ICC ≥ 0.70 to be sufficient (31).

2.4.4. Construct validity

We established hypotheses for construct validity based on known groups validity, convergent validity and influence of social desirability bias.

Known groups validity measures an instrument's ability to distinguish among distinct groups. Group differences were determined using the independent t-test. Three hypotheses were previously defined:

- There would be a significant difference between the mean PSSS-HCP scores of healthcare professionals with a special interest in PSS compared to those who did not have a special interest in PSS. To operationalise the special interest group, we sent a separate survey link to identify participants associated with the Dutch Network for PSS (NALK). Further, we included healthcare professionals who stated that their healthcare profession was in psychosomatic medicine or psychosomatic therapies. Our expectation was that healthcare professionals with a special interest in PSS would score lower for stigma.

- There would be a significant difference between the mean PSSS-HCP scores of healthcare professionals with a close friend or relative with persistent somatic symptoms and those who did not. Our expectation was that healthcare professionals a close friend or relative with PSS (and therefore more contact with people with PSS) would score lower for stigma.
- There would be a significant difference between the mean PSSS-HCP scores of healthcare professionals who had themselves previously been treated for persistent somatic symptoms and those who had not. Our expectation was that healthcare professionals with personal experience of treatment would score lower for stigma.

Convergent validity was assessed by testing hypotheses about the relationships between different instruments. We used the hypothesis template suggested by the COSMIN group, considering both the relation of the constructs and the expected direction of association (31).

- We considered the MCRS's construct of 'regard' to be a similar and related construct to stigma. Our expectation was a moderate negative association between the PSSS-HCP and MCRS. We used Spearman's rank correlation coefficient to determine the relationship between the mean scores of the PSSS-HCP and the MCRS.
- We considered perceived adequate knowledge, training, resources and time during consultations with people with PSS to be different but related to stigma. During validation in a sample of healthcare professionals in the UK, we found that there was a moderate negative association between these (20). Therefore, we would expect the relationship here to be similar in this context.

We also sought to determine the relationship between the PSSS-HCP and the influence of social desirability. We used the Spearman's rank correlation coefficient to determine the relationship between the total scores of the PSSS-HCP and the short form of the MCSDS. If items of the PSSS-HCP correlate highly with scores of social desirability, this suggests that the scale could be influenced by social desirability bias. Our expectation was a low or insignificant association between the total scores of the PSSS-HCP and MCSDS.

2.4.5. Floor and ceiling effects

We analysed responses to determine if any floor or ceiling effects were present for each (sub)scale. Floor and ceiling effects were considered present if ≥15% of participants achieved minimum or maximum scores (34).

2.4.6. Missing data

Participants completing the online survey were not allowed to progress in the survey while leaving any items unanswered, resulting in minimal missing data. However, participants completing some questions, but not completing the whole survey are included here as missing data. The total across the whole survey was n=3 (1.9%). Where possible, we have analysed results using results of participants who have at least completed all relevant blocks (complete case analysis).

3. Results

3.1. Participant information

The online survey received 154 complete responses from healthcare professionals. The PSSS-HCP was completed by 157 participants. Most participants were female (65.0%) with a mean age of 45.8 years (SD = 11.9, range 22-71). Most participants worked in psychology (n = 35, 22.3%), or as a general practitioner (n = 29, 18.5%). Most participants reported that they had a close friend or relative with PSS (58%), and 10.2% had personally been treated for PSS. Characteristics of participants are provided in **Table 1.**

Table 1: Sample characteristics (n=157)

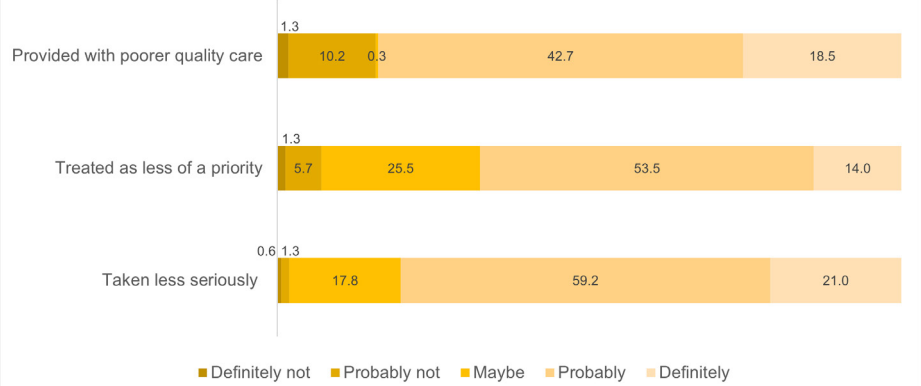
Characteristics		Frequency (n)	Percentage (%)
Age (years)	18-25	5	3.2
	26-44	71	45.2
	45-64	70	44.6
	≥ 65	11	7.0
Sex	Female	102	65.0
	Male	53	33.8
	Other	1	0.6
	Prefer not to say	1	0.6
Type of healthcare professional	Psychologist	35	22.3
	General practitioner	29	18.5
	Physiotherapist	21	13.4
	Neurologist	12	7.6
	Psychotherapist	10	6.4

Table 1: Continued

Characteristics		Frequency (n)	Percentage (%)
Type of healthcare professional	Psychiatrist	9	5.7
	Company doctor	8	5.1
	Nurse	3	1.9
	Surgeon	1	0.6
	Psychosomatic exercise therapist	3	1.9
	Paediatrician	2	1.3
	Social worker	2	1.3
	Basic doctor/ doctor not in specialisation	2	1.3
	Psychomotor therapist	2	1.3
	Insurance doctor	1	0.6
	Occupational therapist	1	0.6
	Other	16	10.2
Years of experience working with patients with PSS	0-1	2	1.3
	1-2	11	7.0
	2-5	24	15.3
	5-10	31	19.7
	10-20	44	28.0
	20-30	35	22.3
	≥ 30	10	6.4
Close friend or family member with experience of PSS	Yes	91	58.0
	No	63	40.1
	Unsure	3	1.9
Previous treatment for PSS	Yes	16	10.2
	No	140	89.2
	Unsure	1	0.6

When asked to compare how people with PSS were treated by healthcare professionals compared to people with symptoms with a clearer pathology, most participants said that people with PSS were taken less seriously (80.2%), treated as less of a priority (67.5%) and provided with poorer quality care (61.2%) (see **Figure 1**. A detailed table of responses is provided in **Appendix 3, supplementary material**).

Figure 1: Perceived stigmatisation of people with PSS by healthcare professionals compared to symptoms with a clearer pathology (percentage)



3.2. Confirmatory factor analysis and model-based reliability

Table 2 displays the fit indices for each tested factor model. Specific factor loadings for each tested model can be found in **Appendix 4, supplementary material**.

Table 2: Model fit indices for tested models

Model	χ2	Df	CFI	SRMR	RMSEA
Unidimensional	266.6	65	0.699	0.105	0.141
3-factor correlated	125.3	62	0.905	0.063	0.081
Bifactor	Failed to converge				
2-factor correlated	127.4	64	0.905	0.063	0.080
Modified bifactor	126.9	62	0.903	0.063	0.082

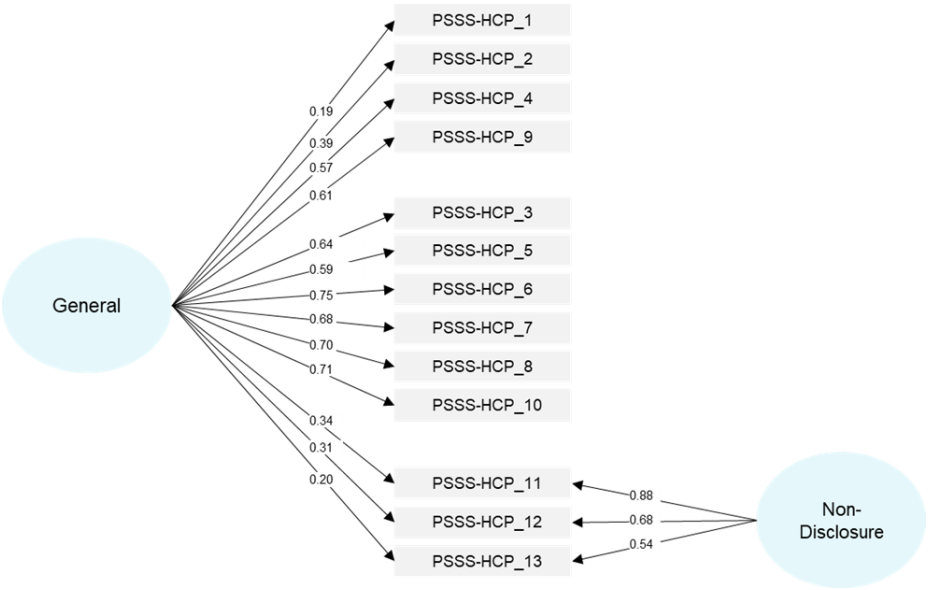
Note: χ2 = chi-square; Df = degrees of freedom; CFI = comparative fit index; SRMR = standardised root-mean-square residual; RMSEA = root-mean-square error of approximation. *Best fit of the data

The unidimensional model showed a poor model fit, suggesting that the PSSS-HCP is not strictly unidimensional. The CFI, the RMSEA and SRMR did not approach acceptable thresholds of model fit.

The correlated three-factor model showed a better model fit, with the CFI approaching acceptable fit, and the SRMR and RMSEA being borderline acceptable fit. Latent factor correlations were moderate to high: othering and uneasiness in interaction = 0.99, $p < 0.001$; othering and non-disclosure = 0.47, $p < 0.001$; uneasiness in interaction and non-disclosure: = 0.35, $p < 0.001$). These results suggest that the factors are all related (sharing a conceptual theme of stigma). While non-disclosure is measuring a distinct concept, the othering and uneasiness in interaction factors appear to be substantively measuring the same concept.

The bifactor model failed to converge due to negative residual variances on two items (PSSS-HCP_9 and PSSS-HCP_10). Through this and the analysis of the three-factor correlated model, we decided to test modified versions of the models. Specifically, we tested a two-factor correlated model (othering and uneasiness in interaction as a combined factor, and non-disclosure), and a modified bifactor model (with all items loading onto a general factor, and items PSSS-HCP_11, PSSS-HCP_12 and PSSS-HCP_13 loading onto a general factor and a specific non-disclosure factor). We used additional bifactor indices to calculate model-based reliability and to assess the dimensionality of the PSSS-HCP.

Figure 2: Factor loadings in the modified bifactor model. Rectangles represent observed item scores and ovals represent latent factors. Path coefficients are standardised coefficients.



The correlated two-factor model and the adjusted bifactor model showed similar model fits, with the CFI, RMSEA and SRMR approaching acceptable fit. In the bifactor model, factor loadings for the general factor were moderate across all items (0.19 – 0.71, **Figure 2**), which were in general only slightly lower than those for the unidimensional model. This suggests that the loadings for the unidimensional model were not substantially distorted by multidimensionality. However, after partialing out the general factor, all three items from the non-disclosure specific factor remained high (>0.5). One item loaded poorly onto the general factor (PSSS-HCP_1).

3.3. Model based reliability

Results from model-based reliability testing are presented in **Table 3**.

Table 3: Model based reliability

	ECV	ω/ ωS	ωH/ ωHS	PUC
General factor	0.722	0.866	0.789	0.962
Specific factor: Non-disclosure	0.278	0.804	0.690	

Note: ECV = explained common variance; ω = omega oefficient; ωS = omega coefficient subscale; ωH = coefficient omega hierarchical; ωHS = coefficient omega hierarchical subscale; PUC = percent of uncontaminated correlations.

The ECV of the general factor was 0.72, the omega hierarchical coefficient of the general factor was ωH = 0.79, and the PUC value was 0.96, suggesting that the PSSS-HCP is 'essentially' unidimensional. From these findings together, we concluded that the PSSS-HCP can be scored using a total scale score, and an optional additional non-disclosure specific subscale score. The mean total score and SD for the PSSS-HCP was 26.81 (6.41), with scores ranging from 13.0 to 44.0. The mean score and SD for non-disclosure subscale was 6.96 (2.42).

The omega coefficient of the PSSS-HCP was ω = 0.87, indicating high reliability of the total score. The omega coefficient for the specific non-disclosure factors was (ωS = 0.80), also indicating high reliability. For comparison, we also calculated Cronbach's alpha coefficient across the scales. The Cronbach's alpha coefficient of the total score was α = 0.83, and α = 0.79 for the non-disclosure subscale).

3.4. Test-retest reliability

A subsample of participants (n = 49) completed the survey a second time after a two week period (response rate from opt-in = 40.83%). **Table 4** demonstrates that the test-retest reliability between the two administrations of the PSSS-HCP is high for both the total scale and subscales. This indicates that the items of the PSSS-HCP are stable in time.

Table 4: Test re-test reliability measures

Scale/subscale	ICC	95% CI of ICC
PSSS-HCP total scale	0.90	0.83 – 0.94
Non-disclosure subscale	0.81	0.69 – 0.89

Note: ICC = Intra Class Correlation; CI = Confidence Interval

3.5. Construct validity

3.5.1. Known groups validity

The constructed special interest group included healthcare professionals associated with the Dutch Network for PSS (n = 50) and healthcare professionals not associated with the Dutch Network for PSS but working in psychosomatic medicine and therapies (n = 3). There was a significant difference between the mean PSSS-HCP scores of the participants who were not in the special interest group (n = 104, mean = 29.02, SD = 5.47) and those who were (n = 53, mean = 22.47, SD = 5.91), [t(155) = 6.903, p = <0.001], Cohen's *d* = 1.165. Therefore, we can confirm the hypothesis that healthcare professionals with a special interest in PSS scored lower for stigma.

There was a significant difference between the mean PSSS-HCP scores of the participants who did not have a close friend or relative with PSS (n = 63, mean = 28.4, SD = 6.2), and those who did (n = 91, mean = 25.7, SD = 6.3) [t(152) = 2.703, p = .009, Cohen's *d* = 0.43]. Therefore, we can confirm the hypothesis that healthcare professionals with a close friend or relative with PSS scored lower for stigma.

There was a significant difference between the mean PSSS-HCP scores of the participants who had not themselves been treated for PSS (n = 140, mean = 27.2, SD = 6.4), and those who had (n = 16, mean = 23.3, SD = 5.9),

[t(154) = 2.321, p = 0.022, Cohen's *d* = 0.61]. Therefore, we can confirm the hypothesis that healthcare professionals who had themselves been treated for PSS scored lower for stigma.

3.5.2. Convergent validity

The total score of the PSSS-HCP had a strong negative correlation with the total MCRS score (r_s = -0.78, p<0.001). Therefore, we can confirm the hypothesis that healthcare professionals scoring higher for stigma held less positive regard towards patients with PSS.

Mean scores and SD for perceived adequate knowledge were 3.91 (0.74), for perceived adequate training were 3.73 (0.87), for perceived adequate tools to manage and/or diagnose patients were 3.61 (0.88), for perceived adequate time in consultations to discuss PSS were 3.67 (1.08). The total score of the PSSS-HCP had a moderate negative correlation with perceived adequate knowledge (r_s = -0.40, p<0.001), training, (r_s = -0.46, p<0.001), tools (r_s = -0.37, p<0.001), and time during consultations (r_s = -0.43, p<0.001)**.** Therefore, we can confirm the hypothesis that healthcare professionals with less perceived adequate knowledge, training, tools and time during consultations scored higher stigma.

3.6. Social desirability bias

The PSSS-HCP had an insignificant correlation with the MCSDS (r_s = –0.03, p=0.75). This suggests that there was no significant association between social desirability bias and the PSSS-HCP.

3.7. Floor and ceiling effects

There were no ceiling or floor effects found for the total PSSS-HCP, or the specific factors. Percentages of minimum and maximum scores reached for each scale and subscale are included in **Appendix 5.**

4. Discussion

4.1. Main findings

We developed a Dutch version of the PSSS-HCP and evaluated its measurement properties among a sample of healthcare professionals in the

Netherlands. Our results demonstrated sufficient construct validity, test-retest reliability, model based reliability and no evidence of social desirability bias. This version of the PSSS-HCP can be used to measure stigmatising attitudes of healthcare professionals towards people with PSS in the Dutch context.

The results from the confirmatory factor analysis suggested similar model fits for a two-factor correlated model and a modified bifactor model (where all items load on a general underlying factor and three items load on a specific non-disclosure factor). The model-based reliability results suggest that the general factor explained a substantial proportion of the variance. Therefore, depending on the specific research question, we suggest that the PSSS-HCP can be interpreted both as a total stigma score, or an optional additional non-disclosure subscale score.

4.2. Comparison to literature

Most participants perceived there to be higher stigma (treated less seriously, with less priority and provided with less quality care) for people with PSS than with symptoms with a clearer pathology. This aligns with perspectives of healthcare professionals from qualitative studies, who have reflected on the unique challenges of communicating and providing acceptable treatment for people with PSS (13, 35, 36). This also resonates with perspectives from patients, who have described feeling less legitimate than other patients going through the healthcare system (6, 37, 38). Several studies directly comparing functional disorders to symptoms with a more clearly structural pathology also found higher levels of perceived stigma (39, 40). This finding underlines the perceived relevance of stigma in this field and the importance of developing and evaluating stigma reduction interventions.

There is increased evidence pointing to the increased use of bifactor models when measuring stigma, assessing both general and specific factors that may affect levels of stigma and health outcomes. A recent review of the Opening Minds Stigma Scale for Healthcare Professionals (examining mental health stigma among healthcare professional) examined the factor structure of the scale on a large sample of psychiatrists across 32 countries in Europe. They concluded that the bifactor model was the best approximation of the factor structure of the scale, but also that the general factor and the disclosure and help-seeking factor could be the best scoring choices when assessing stigma among healthcare professionals (41).

Similarly, bifactor solutions have been identified for self-stigma for people with psychiatric disabilities (42) and HIV-related stigma (43).

All of our hypotheses for construct validity (including known groups and convergent validity) were supported. Through differences in PSSS-HCP scores between groups (having a close friend or relative with PSS, being personally treated for PSS, or having a special interest in PSS), these results suggest that increased contact with people with PSS is an important component in reducing stigma. This highlights the importance of person-centred care for managing people with PSS, including paying attention to psychosocial cues, treating the concerns of someone seeking medical help seriously, and building a relationship as equal partners (44). Contact is regularly described in stigma research as one of the key types of stigma reduction intervention, which can be effective both through direct contact (e.g. in person), or indirect (e.g. online, pre-recorded contact or through media) (45). However, particular conditions are important for contact interventions, including equal status between participants, frequent contact with individuals who disprove stereotypes, and real-world opportunities to interact (46).

Further, these results suggest that lower perceived adequacy of knowledge and training is associated with higher levels of stigma. This aligned with our findings from the original validation of the PSSS-HCP in a sample of healthcare professionals in the United Kingdom (20). There is also qualitative evidence supporting this, where perceived lack of competence of healthcare professionals had an influence on discriminating actions such as avoiding patients (16). Therefore, targeting interventions towards healthcare professionals earlier in their training may also be worthwhile. This is particularly important as through a limited formal and intended curriculum, healthcare professionals learn through a 'hidden curriculum' of informal social processes such as role modeling and interactions with senior colleagues (47).

The test-retest reliability with a two-week retest period was strong, both in the general and specific factors. We also found that the PSSS-HCP demonstrated very strong reliability in previous validation among UK healthcare professionals (20). This suggests strong stability between test periods.

Throughout development and evaluation of the PSSS-HCP, we aimed to explore and mitigate the impact of a specific type of demand characteristic, social desirability. Though social desirability could be a threat to the validity

of the instrument, we have consistently found minimal influence of this during testing. While there are recent criticisms of the validity of social desirability scales (48), further research could include triangulation of the PSSS-HCP with other methods, such as implicit measures.

4.3. Strengths and limitations

An important strength of our study was that we followed COSMIN guidelines throughout design and reporting of results. We used guidelines from the COSMIN study design checklist to assess sufficiency of each measurement property (24). Further, we followed extensive guidance in the translation of the PSSS-HCP into Dutch (including forwards and backwards translations, multiple translators and rounds of feedback), and evaluation of measurement properties. We used feedback from multiple organisations with both healthcare professional experience and lived experience of PSS.

A further strength is that we followed recommendations during confirmatory factor analysis. While we tested additional models following initial analysis, we retained the original versions of these models without post hoc modifications such as correlating error terms or removing items. This reduces the tendency for researchers to optimise their model fit in a specific sample (49, 50). Further, it allows our model to be replicated and evaluated in further research.

The main limitation of this study is that there may be a recruitment bias in participating healthcare professionals. Since we contacted national organisations and asked them to distribute among their network, participants were self-selecting. Participants reported high levels of years of experience working with people with PSS and moderate to high levels of perceived adequate knowledge and training. This is further reflected in the high numbers of participants reporting personal treatment for PSS, or had a close friend or relative with PSS. Further research should study the measurement properties of the PSSS-HCP in less experienced healthcare professionals.

A second limitation is that we were not able to test the sensitivity of the PSSS-HCP to change during an intervention.

4.4. Implications for future research and policy

Further research with bifactor analyses could explore the structure of the PSSS-HCP in different healthcare professional and cultural contexts. This would assess to what extent our findings here are universal, or unique to the Dutch healthcare context.

Responsiveness to change should also be a research focus, including the minimally important change (the meaning of changes in PSSS-HCP score over time). This is an important aspect of intervention evaluation.

5. Conclusion

We have developed a Dutch version of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP) and evaluated its measurement properties.

The results from the confirmatory factor analysis suggested similar model fits for a two-factor correlated model and a modified bifactor model (where all items load on a general underlying factor and three items load on a specific non-disclosure factor). The model-based reliability results suggest that the general factor explained a substantial proportion of the variance. Therefore, depending on the specific research question, we suggest that the PSSS-HCP can be interpreted both as a total stigma score, and an optional additional non-disclosure subscale score.

Our results demonstrated sufficient construct validity, test-retest reliability, model-based reliability and no evidence of social desirability bias. This version of the PSSS-HCP can be used to measure stigmatisation by healthcare professionals towards people with PSS in the Dutch context. Future research should conduct further evaluation with an emphasis on responsiveness (sensitivity to change).

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Supporting information

Appendix 1: Dutch version of Persistent Somatic Symptom Stigma scale for Healthcare Professionals

In hoeverre bent u het eens met deze stellingen?
(1 = Helemaal mee oneens, 5 = Helemaal mee eens)

	Item (* = reverse scored)
1*	Iedereen kan aanhoudende lichamelijke klachten krijgen onder bepaalde omstandigheden.
2	De meeste mensen met aanhoudende lichamelijke klachten doen niet goed genoeg hun best om beter te worden.
3	Er is weinig dat ik kan doen om mensen met aanhoudende lichamelijke klachten te helpen.
4	Ik vind dat mensen met aanhoudende lichamelijke klachten vaak hun klachten overdrijven.
5*	Ik voel me net zo comfortabel om met iemand met aanhoudende lichamelijke klachten te praten als met mensen die dit niet hebben.
6*	Het geeft mij voldoening om mensen met aanhoudende lichamelijke klachten te ondersteunen.
7	Mensen met aanhoudende lichamelijke klachten roepen bij mij negatieve gevoelens op, zoals afkeer, stress of ongeduld.
8	Ik hoop stiekem dat mensen met aanhoudende lichamelijke klachten niet terug komen.
9	Ik vind het moeilijk om mee te leven met mensen met aanhoudende lichamelijke klachten.
10	Als ik de keuze had, zou ik liever geen zorg verlenen aan mensen met aanhoudende lichamelijke klachten.
11	Als ik aanhoudende lichamelijke klachten zou krijgen, zou ik het liever niet tegen mijn vrienden zeggen.
12	Als ik behandeling zou krijgen voor aanhoudende lichamelijke klachten, zou ik dit liever niet aan mijn collega's vertellen.
13	Als ik zou weten dat ik aanhoudende lichamelijke klachten heb, zou ik terughoudend zijn om hulp te zoeken.

Appendix 2: STROBE Statement

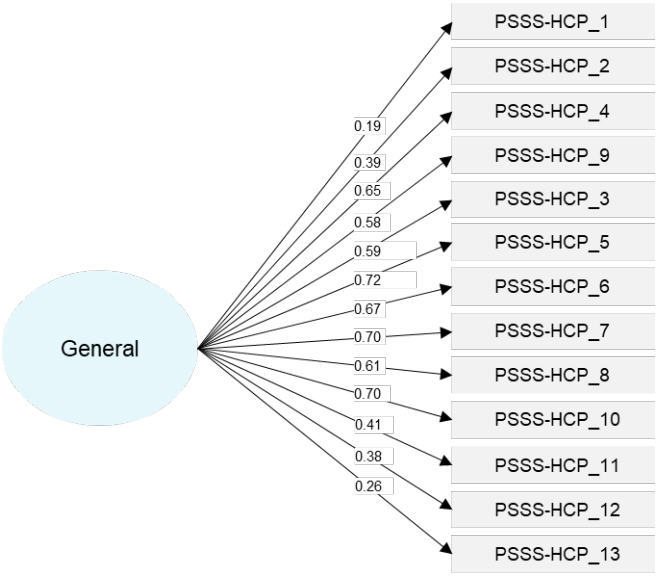
Available in online supplementary material

Appendix 3: Perceived stigmatisation of people with PSS by healthcare professionals, compared to symptoms with a clearer pathology

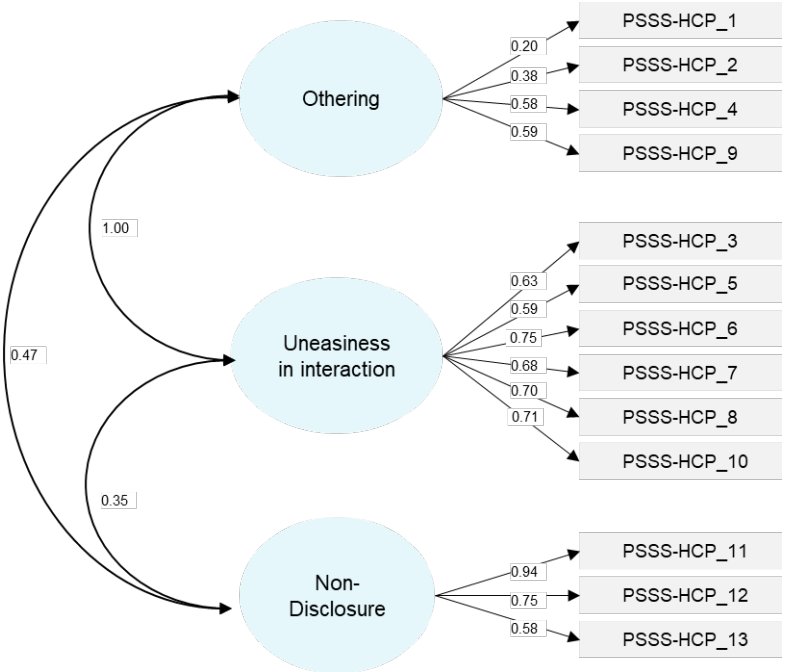
	Taken less seriously		Treated as less of a priority		Provided with poorer quality care	
Response options	**Frequency (n)**	**Percentage (%)**	**Frequency (n)**	**Percentage (%)**	**Frequency (n)**	**Percentage (%)**
Definitely	33	21.0	22	14.0	29	18.5
Probably	93	59.2	84	53.5	67	42.7
Maybe	28	17.8	40	25.5	43	27.4
Probably not	2	1.3	9	5.7	16	10.2
Definitely not	1	0.6	2	1.3	2	1.3
Aggregated scores						
Total agree	126	80.2	106	67.5	96	61.2
Total disagree	3	1.9	40	25.5	43	27.4
Total maybe	28	17.8	11	7.0	18	11.5

Appendix 4: Tested measurement models for the PSSS-HCP

Unidimensional model

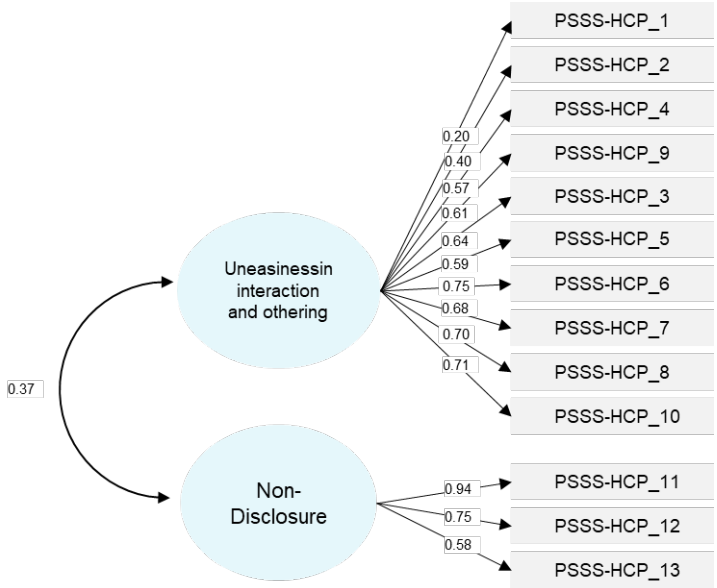


3-factor correlated model

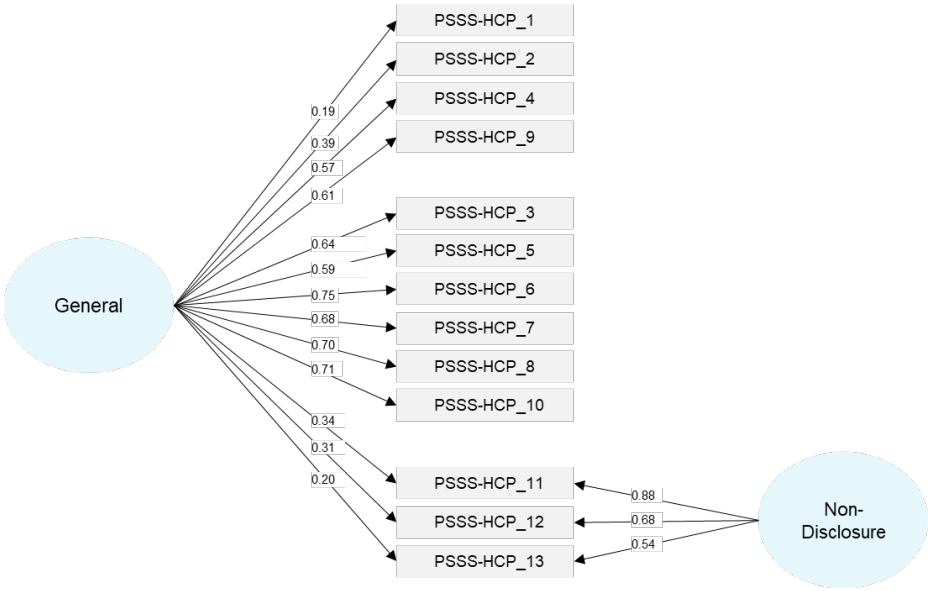


Bifactor model – failed to converge

2-factor correlated model



Modified bifactor model



Appendix 5: Percentages of possible minimum and maximum scores reached for each scale and subscale

	Item (* = reverse scored)
1*	Iedereen kan aanhoudende lichamelijke klachten krijgen onder bepaalde omstandigheden.
2	De meeste mensen met aanhoudende lichamelijke klachten doen niet goed genoeg hun best om beter te worden.
3	Er is weinig dat ik kan doen om mensen met aanhoudende lichamelijke klachten te helpen.
4	Ik vind dat mensen met aanhoudende lichamelijke klachten vaak hun klachten overdrijven.
5*	Ik voel me net zo comfortabel om met iemand met aanhoudende lichamelijke klachten te praten als met mensen die dit niet hebben.
6*	Het geeft mij voldoening om mensen met aanhoudende lichamelijke klachten te ondersteunen.
7	Mensen met aanhoudende lichamelijke klachten roepen bij mij negatieve gevoelens op, zoals afkeer, stress of ongeduld.
8	Ik hoop stiekem dat mensen met aanhoudende lichamelijke klachten niet terug komen.
9	Ik vind het moeilijk om mee te leven met mensen met aanhoudende lichamelijke klachten.
10	Als ik de keuze had, zou ik liever geen zorg verlenen aan mensen met aanhoudende lichamelijke klachten.
11	Als ik aanhoudende lichamelijke klachten zou krijgen, zou ik het liever niet tegen mijn vrienden zeggen.
12	Als ik behandeling zou krijgen voor aanhoudende lichamelijke klachten, zou ik dit liever niet aan mijn collega's vertellen.
13	Als ik zou weten dat ik aanhoudende lichamelijke klachten heb, zou ik terughoudend zijn om hulp te zoeken.

*Floor and ceiling effects were considered present if ≥15% of participants achieved minimum or maximum scores.

Chapter 6

How stigma unfolds for patients with Functional Neurological Disorder (FND)

McLoughlin C, McGhie-Fraser B, Carson A, olde Hartman T, Stone J. Journal of Psychosomatic Research. 2024 June 111667.

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Abstract

Objective: The aim of this study was to explore experiences of stigma in Functional Neurological Disorder (FND) from the perspective of the patient as it manifests from the onset of symptoms, up to diagnosis and subsequently.

Background: The aim of this study was to explore experiences of stigma in Functional Neurological Disorder (FND) from the perspective of the patient as it manifests from the onset of symptoms, up to diagnosis and subsequently.

Methods: We performed a qualitative interview study with patients who were diagnosed with FND, using data based on semi-structured interviews. Participants were recruited purposively via outpatient clinics. We analysed the data using a reflexive thematic analytic approach, through the lens of recognised stigma frameworks.

Results: 15 participants were included in the study, aged between 19 and 68 years, with varying presentations of FND. We identified six themes and 16 subthemes relevant to their stigma trajectory. We found that stigma unfolds through four main domains: 1) through their symptom experience; 2) through "othering" by the healthcare system; 3) through everyday interactions; and 4) from within the self. Across these four domains was a central theme of 5) stages of knowledge, which both fuelled and countered stigma. Lastly, 6) validation of the patient experience emerged as a theme that alleviated stigma.

Conclusion: Stigma did not unfold as a linear process, rather it came from multiple interacting sources. Interventions to target stigma could take the form of improved clinician training, communication, especially around point of diagnosis, and public interventions, co-produced with patients with FND.

1. Introduction

Functional Neurological Disorder (FND) is a common condition that can present in varying ways including, weakness, seizures, movement disorders and speech problems (1). Though recognition of FND as a valid and treatable disorder is growing, it remains a neglected condition, influenced by outdated misperceptions and attitudes ((2), (3), (4)). Training on the subject has been reported to be poor ((5), (6), (7)), and patients and clinicians report referrals to clinical services have been rejected based on the FND label ((8), (9), (10)).

A recent survey of 503 participants run by charity FND Hope, showed that 81.6% felt they had been treated poorly due to stigma (11). Stigma is a multi-factorial, social process and has been conceptualised in different ways ((12), (13), (14), (15)). Link and Phelan (2001) in their sociological model describe stigma as the co-occurrence of the following: labelling, stereotyping, separation, status loss, and discrimination, all occurring the context of power (12). Stigma has been further considered as an interpersonal process involving *prejudice, stereotyping* and *discrimination* ((13), (14), (15)). From the perspective of the person experiencing stigmatisation, stigma can be *experienced, anticipated* and/or *internalised* (self-stigma) (13,15). It has been described how a collective social rejection of a group influences policy and healthcare planning, perpetuating a damaging cycle that has been described in other functional syndromes (16,17).

While there is a lack of longitudinal data on the outcomes of stigma in FND, stigma has been associated with depression and poor treatment engagement in other conditions ((18), (19), (20), (21)). Quantitative studies show stigmatisation is around 40% more likely for patients with FND than epilepsy – the latter also a highly stigmatised condition (22,23). Stigma with functional seizures is associated with poorer quality of life and caregiver burden (22,24,25). Reviews on this topic show there are several qualitative studies examining patient experiences of FND from which stigma themes naturally emerged, but most studies did not aim to explore stigma specifically (2,3). Furthermore, the majority of studies in this sphere relate to functional seizures – not covering the fuller spectrum of FND symptoms.

Therefore, while it is clear that stigma exists in FND, it is less clear where stigma originates from, how it unfolds, and how it can be alleviated.

Increased knowledge about the development of stigma in FND could direct the formation of “anti-stigma” interventions, and potentially improve stigma-related outcomes for this group.

1.1. Aim

Therefore, the aim of this study is to explore experiences of stigma in FND from the perspective of the patient as it unfolds from symptom onset through diagnosis and thereafter.

2. Methods

2.1. Study design

We performed a qualitative interview study with patients diagnosed with FND using reflexive thematic analysis (RTA) (26,27). We used the COREQ guideline for the reporting of this study (28).

2.2. Study approval

The study was approved by the University of Edinburgh and South-Central Hampshire A Research Ethics Committee (reference 21/SC/0418). The current study is part of the innovative training network ETUDE (Encompassing Training in fUnctional Disorders across Europe) ultimately aiming to improve the understanding of mechanisms, diagnosis, treatment and stigmatisation of Functional Disorders (29).

2.3. Participants

Participants were recruited consecutively via neurology/neuropsychiatry clinics. Inclusion criteria were as follows: 1) participant was willing and able to give informed consent; 2) of any sex/gender; 3) over 18 years; 4) their diagnosis of FND was given by a neurologist/neuropsychiatrist; 5) they were fluent in English (language of interviewer). We wanted to get a range of opinions and experiences related to stigma, and therefore employed purposive sampling to ensure diversity in age, gender, symptom presentation, and diagnosing clinician.

2.4. Recruitment

Prior to recruitment into the study, participants had to be diagnosed with FND by a neurologist/neuropsychiatrist. The clinician had copies of the participant information sheet and sought verbal consent from potential participants to be contacted by the lead researcher (CM), who waited at least 24 hours before contacting the potential participant to discuss the study and arrange a meeting time for consent and interview. Given that we were interested in stigma from symptom onset, we recruited patients as close to their formal diagnosis as possible (aiming for within four months) to reduce possible influence of a long recall gap on answers.

2.5. Interview structure and procedure

All interviews were recorded using a secure encrypted Dictaphone following informed written consent. Each interview lasted 45 to 90 minutes. Questions were informed by the various components of stigma as it has been described in the literature (12–15). We chose open questions about patient experience as not to be leading, deliberately not mentioning stigma, giving space for both positive and negative experiences. See **Appendix 1, supplementary material** for the discussion guide. We checked the patient's medical record to verify the diagnosis and history (including time from symptom onset and diagnosis to interview).

2.6. Analysis

We analysed the data using a reflexive thematic analysis approach (26, 27, 30). To reduce the risk of bias, we strived to ensure the analysis remained grounded in the data, remaining cognisant and reflective about existing assumptions from our clinical and research experience. CM (liaison psychiatrist), JS (neurologist), AC (neuropsychiatrist) and TOH (general practitioner) are involved in the clinical care of patients with FND, and BMF (sociologist) has extensive experience in qualitative research. All researchers are involved in researching stigma as part of the ETUDE program (29).

The audio-recorded interviews were transcribed verbatim and analysed independently by two researchers (CM and BMF), using MaxQDA 2022 software. We followed six iterative steps, namely; familiarisation, coding, generating initial themes, reviewing and developing themes, refining, defining

and naming themes, and writing up (27,30). We regularly compared our analysis, clarifying differences and refining our codes, themes and subthemes. We wrote memos throughout to capture our ideas and reflections on codes and themes as they emerged. We further discussed our analysis with experts in the field (TOH, AC, JS). We discussed differences in coding until consensus was reached.

3. Results

Nine interviews were conducted face to face and six interviews via secure video platform. After 15 interviews, using the latest coding framework, no new categories were found, and we considered saturation was reached. We selected quotes which we considered depicted a theme/subtheme well.

3.1. Participant characteristics

There were 15 participants in total, 11 identified as women, one as non-binary and three as men. Participants were aged between 19 and 68 years. All diagnoses were confirmed by a consultant neurologist (five consultants in total), often after it had been raised as a possibility in the emergency/primary care setting. See **Table 1** for clinical and demographic characteristics of participants. Twelve patients were recruited within six months of diagnosis. In order to maximise sample variation, the remaining three were recruited nine months to five years post-diagnosis.

3.2. Main themes and subthemes

We found the patient experience of stigma did not unfold as a linear trajectory. Rather, six key themes dynamically interacted with each other. Stigma unfolded through four main domains: 1) their *symptom experience*, 2) *"othering" by the healthcare system*, 3) *everyday interactions* with friends, family, colleagues and online, and 4) from *within the self*. Across these four domains was a central theme of: 5) *stages of knowledge*; and lastly, 6) *validation of patient experience* emerged as a theme that countered or opposed stigma (**see Fig. 1**). Furthermore, we found 16 subthemes within these themes, see **Table 2 Case Boxes 1–3** are examples of stigma narratives depicting the trajectory of stigma themes. Note, these are not true cases, but adapted from individual cases to protect anonymity.

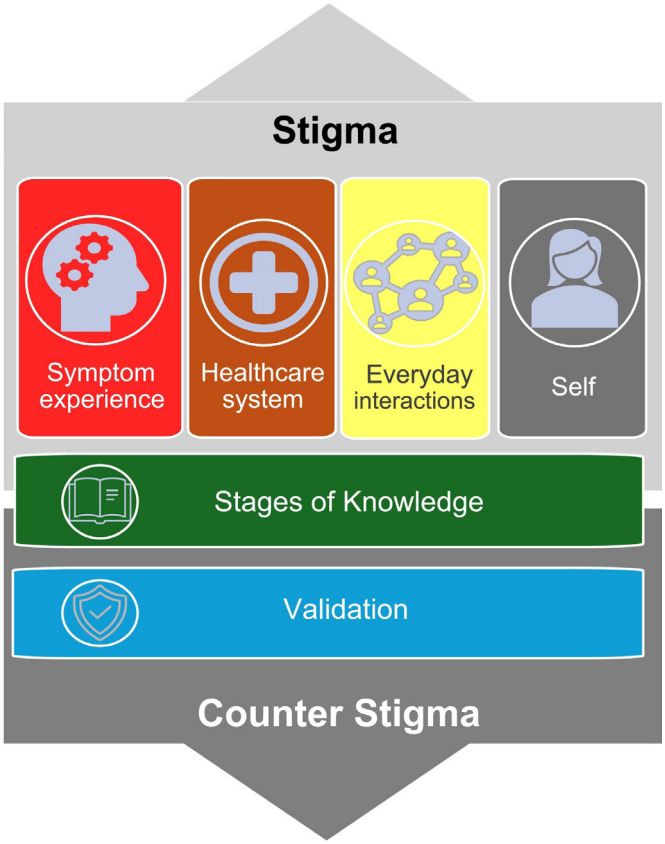
Table 1: Clinical and demographic characteristics of participants (pseudonyms)

Pseudonym	Gender	Age	Clinical presentation	Symptom duration prior to diagnosis	Time from diagnosis to interview
Charles	M	46	Functional sensory symptoms	2.5 years	4 weeks
Una	F	68	Functional gait disorder Functional visual disturbance	3 years	2 years
Orla	F	51	Functional tremor Functional gait disturbance Functional cognitive symptoms Right upper limb functional numbness	11 months	2 months
Sam	Non-binary	35	Dissociative seizures Functional cognitive symptoms	3 weeks	2 months
Grace	F	37	Bilateral functional leg weakness Functional achromatopsia Dissociative episodes Functional ankle dystonia	1 week	5 years
Ali	M	60	Functional left upper limb tremor	2 years	5 months
Brendan	M	48	Right sided functional weakness Functional speech disturbance	1 week	2 months
Rose	F	38	Functional tremor Functional left sided weakness Functional sensory disturbance Functional gait disorder	4 weeks	1 year
Poppy	F	34	Functional bilateral leg weakness 3 days post-partum	4 days	3 weeks
Hailey	F	19	Dissociative episodes Functional speech disturbance Functional weakness/paralysis	3 years	6 months
Maggie	F	29	Functional left sided lower limb weakness	2 years	5 weeks
Martha	F	53	Dissociative seizures	9 years	3 weeks
Laura	F	20	Dissociative episodes Functional sensory symptoms Functional tremor Functional speech problems	2 years	4 weeks
Norah	F	48	Functional left sided and generalised weakness Functional speech disturbance Dizziness	18 months	6 months

Table 1: Continued

Pseudonym	Gender	Age	Clinical presentation	Symptom duration prior to diagnosis	Time from diagnosis to interview
Bridget	F	39	Left sided functional weakness Left sided functional sensory disturbance Functional speech difficulties Functional cognitive difficulties	3 months	9 months

Figure 1: Core themes depicting how stigma unfolds for patients with FND.



Stigma unfolds through four main domains, impacted by stages of knowledge. Stigma as alleviated by increasing knowledge and validation of the patient experience.

Table 2: Main themes and subthemes with illustrative quotes

Theme	Subtheme with illustrative quote
1. FND symptom experience	**a) Visibility of symptoms** *"when you can hear other people talking over you and about you yeah … they couldn't believe they had seen something like this before…I'm literally kicking everything out; my arms, my legs, …I just feel really hurt"* – Martha **b) Invisible** *"I was out in the garden and there was a guy that I've no seen for absolutely years and he walked by and he made some sort of comment about there's nothing wrong with your legs you can walk fine there… I think people just find it difficult because like one day I can be in a wheelchair the next day my walking can be pretty decent"* – Grace **c) Voluntary control** *"There is a little bit of shame in losing control … you can't help but feel embarrassed because you have lost control…that is something that I spent days in the hospital trying to come to terms with…"*- Sam
2. "Othered" by health system	**a) Healthcare professional attitudes** *"the experience with the GPs about it hasn't been very…ehm and it just, it actually upset me quite a fair bit… he did say … could you not walk in and I'm like I cannae.. made me feel like he was saying it was all in my head and I was making it up and stuff and I was just sort of like why would anybody make this up…."* - Grace **b) Point of diagnosis** *"it was like a kick in the back because it wasn't even really a consultation, it was a case of … suddenly oh you've got FND, it's almost like just putting me in a bracket, you've got FND here's the website, go away"* -Charles **c) Functional is left out and "lesser"** *"I kinda got the general feeling that the way to treat FND is to ignore it and so do ignore it means to not talk about it so … what professional do I get to say this is happening, this is hurting"* -Bridget
3. Everyday interactions	**a) Friends and family** *"I feel like my mum is the sort of person that always belittled my health…um she… she would say… 'I don't know if I should believe you anymore because what if you're making it up'"* -Laura **b) Work** *"I haven't told um the people I work for and I don't know if they would let me continue to work there if they knew…part of me thinks that I would be written off … like they won't trust me to do things… because…its difficult though because there are times when it would be useful for them to know"* – Hailey **c) Online** *"I never heard of it and it wasn't until I started reading (online) oh my god, oh my god that's when I just lost all respect, all my self-respect… just reading things like you're nuts basically"* - Norah

Table 2: Continued

Theme	Subtheme with illustrative quote
4. Self	**a) Devaluation** *"I feel like a useless piece of … flesh that doesn't work properly"* – Una **b) Shame** *"it had a huge impact on my self-esteem because it made me feel my on my god you are a complete fruit loop, I mean you made this happen…you are making this happen to yourself and it made me, it was actually yeah, I ashamed of myself"* -Norah **c) Strength** *"I've learnt a lot through the process, I think probably ehm value myself more"* -Orla
5. Stages of knowledge	**a) Misperceptions/public awareness** *"I've been going back and forward to the doctor with but nobody knew what was wrong because I think there is not enough knowledge about it, it's a really like underrated…. because you say it to folk, I've got an FND, and they look at you like you have got three heads like what's that"* – Rose **b) Importance of explanations** *"and then leaving from there I felt a little bit more satisfied that somebody took it more seriously…and understands that there is a problem there, but we don't know what…I think the way to explain things saying it could be this, it could be that… he says right what will we do with you, we take a step at a time"* – Ali
6. Validation of patient experience	**a) Within professional context** *"She … kind of propped it up, you know…and didn't make me feel ashamed…I think because she was the first doctor that actually made me feel like that, I then felt a bit better about having it, does that make sense"* - Norah **b) Within wider context** *"I was really lucky I got quite a positive from my friendship group and from my family…other people have said your just lazy it's just a title, you….you've looked for this…a few like my closest friends have downloaded the app and they're like that how you are feeling today… is there anything we can do, can we make it better like"* - Rose

Case boxes 1-3: Describing typical stigma trajectories (fictional), adapted from individual cases

Case 1. Leo developed weakness in his leg and face and thought he was having stroke. He felt worried and felt self-conscious.	**Symptom experience**
After a few days in hospital, he was suddenly diagnosed with FND and promptly discharged. He did not understand what the diagnosis meant, as it all felt really rushed. He went to his GP after discharge to get more information, who told him he must accept the diagnosis or he would not get better.	**Othering through healthcare system**
Leo was confused by this so so looked FND up online. He found the online information scary and demoralising – where experts in the area were discredited and people saying FND was a pseudonym for faking.	**Stages of knowledge**
He was referred for regular physiotherapy where the therapist worked closely with him for over a year, educating him about his condition and helping him with exercises to get his power back.	**Validation**
He is able now to do the activities that are important for him, and feels the diagnosis of FND has made him stronger.	**Self**

Case 2. Mary dreaded the loss of control that happened whenever she had a functional seizure. She felt scared and also feared she appeared strange to others.	**Symptom experience**
Her friends called her weird, and she internalised this negative attribute.	**Everyday interactions**
The emergency staff discharged her whenever she came to the Emergency Department with no diagnosis, leaving Mary with no words to explain her condition to others.	**Othering through healthcare system**
By the time she got to see neurologist several months later, she had withdrawn a lot socially partly due to shame around losing control.	**Self**
Her neurologist was able to spend time giving her a clear explanation and diagnosis, with a care plan tailored to her individual needs. She stopped reattending ED, and having a clear words for her diagnosis with analagies, empowered her to explain what was happening when the symptoms occurred.	**Stages of knowledge**

Case 3. Farah worked in healthcare and started to experience dissociative events where she felt strange and unreal. Over the next few weeks, she noticed she was unable to remember things, felt dazed, and got lost on the way home.	**Symptom experience**
She was afraid to tell her healthcare colleagues as she suspected they held negative preconceptions of functional disorders.	**Everyday interactions**
She went to her GP where she felt dismissed, as they did not give the perception there was any urgency to her presentation, despite her being too afraid to work and drive.	**Othering through healthcare system**
She started to believe she was imagining her symptoms and blamed herself for being stressed.	**Self**
She had a helpful interaction with her neuropsychologist who acknowledged the seriousness of her condition and	**Validation**
explained it in terms that made sense to her, integrating the connection of mind and body.	**Stages of knowledge**

3.3. Main themes and subthemes

3.3.1. *Theme 1. FND symptom experience*

The experience of having FND symptoms gave rise to stigma – due to their conspicuous nature, variability which was not easy to explain, or conversely because they appeared "normal", with no disability. Comments from others led patients to become self-conscious or "noticed": *"for me it is just the normal … until somebody points out that it's weird"* (Maggie). Several patients expressed concern about how their symptoms might be perceived; *"I suddenly started taking tremors… …it was like does she think I've got the DTs (delirium tremens) or something"* (Orla). The variability inherent in FND led to patients feeling doubted around the legitimacy of their experience; *"he asked me to walk…its really hard for someone to diagnose something when you look normal and walk normal"* (Una).

Many symptoms felt vague and were hard to articulate. Being unable to satisfactorily communicate the symptom challenged the credibility of experience, which led to patients feeling they needed "proof". *"…at times I have thought if I had some sort of proof that my condition is like disabling I would feel more comfortable sitting on disabled chairs"* (Laura). Many patients had the impression that others thought they could control their symptoms. For some it was felt implicitly; *"they do make you feel like it's all in your head, you're dreaming this, you're making this happen"* (Norah). For some the loss

of control during episodes hampered social participation, forming a cycle of decreased confidence and exclusion: "*I may have had a few episodes of not being able to talk … having to kind of withdraw from the classroom….it does feel embarrassing that I don't have control over it*" (Hailey).

Theme 2. Self

Self-stigma emerged as a significant theme, where patients internalised negative beliefs and attitudes about FND. In some cases, this led to disturbed self-identity and devaluation; "*it's kind of that weird imposter syndrome….am I trying to make things more wrong with me… other people have it worse*" (Hailey). This self-judgement often abated when given the official diagnosis: "*it was a relief when I found out what it was because I thought ehm, I thought I was causing it*"" (Orla). Many described themselves in derogatory terms – as if their FND represented something intrinsically deficient; "*I am wrong*" (Una). Several patients internalised negative stereotypes such that people with FND were malingering or crazy. Some described feeling undeserving of care or healthy relationships, or judged themselves for perceived inabilities. "*I think it's stolen my life…who would want this because realistically nobody, nobody wants to have to deal with somebody that's like this*" (Bridget).

This feeling of shame affected how patients would choose to interact with others. Some patients became afraid to socialise, losing confidence and amplifying self-stigma; "*I've lost friends…because of it, so it's kinda …she's not got a lot to say*" (Bridget). One patient didn't want to go out in case anyone could "*identify a weakness*"" (Orla). Shame led to many not disclosing their diagnosis, linking to anticipated stigma around misperceptions of FND; "*I'm not ashamed to tell people that I have ME uhm but if it ever gets to that stage with FND I don't know… there is so many people out there insisting that it's Freud's conversion disorder*" (Norah).

Though many experienced self-devaluation, several patients also adapted in positive ways to stigma-related difficulties, becoming more assertive and assured of their worth. Many harnessed inner resilience, choosing to ignore negativity and focus on recovery. It was often through the process of accepting the diagnosis, allowing it to be integrated as a valid part of themselves that allowed strength and confidence to blossom; "*I think generally it's actually made me a stronger person..ehm and like I say I've, I'm a completely different person for what I was before FND…ehm and I know it's rough at times but it's my life has changed for the better*" (Grace).

Theme 3: "Othered" by the healthcare system

Through interactions with the healthcare system, many patients experienced a feeling of being different or less legitimate than patients with other medical conditions. This "othering" happened in both subtle and more explicit ways, and led patients to feel set apart and separated on the basis of having FND. This process of othering mainly occurred through negative professional interactions, though it was not the only route. For several patients, the route to diagnosis was protracted and difficult – for example they saw multiple specialists, had to seek care privately or attend the emergency department repeatedly for years before a diagnosis. Many perceived a sense of confusion about FND from professionals, in contrast to other conditions they sought help for.

While not arising for every patient, negative professional attitudes were quite formative. A common scenario involved an invalidating consultation where there was a discordance between patient experience and what professionals saw as "normal"; "*I feel like I was gaslit a lot by medical professionals…was essentially making things up… because everything came back as normal*" (Laura). This was more strongly described in primary care or emergency department (ED) settings. There was a general sense that patients were bothersome and unwanted, leading to feelings of rejection; "*you can almost feel them sighing*" (Norah). Some interactions represented more serious derision and ridicule: "(its)*as if you didn't exist, you're down …they're mocking around you… they said she is just an attention seeker*" (Martha).

FND was further set apart throughout the process of diagnosis. While some found it a positive moment, many felt confused and isolated: "*professionals can be…um put you in the deep end and see if you start swimming …. I think it's better to say that sounds like something is wrong…you're not completely crazy*" (Hailey). The diagnosis was often delivered in unusual ways – not mentioned in the work-up, rushed or sprung as a surprise. In some cases, a website appeared to entirely replace a satisfactory consultation. One patient was told that diagnosis was her choice; "*it was quite strange the whole process you don't have to be diagnosed with this…it's your choice*" (Hailey). Another had an opposing experience where diagnosis felt forced; "*…got to the point where I ended up crying like…I don't understand, she was demanding me just to confirm I believe that I had FND*" (Bridget).

FND was almost never mentioned in differential diagnosis. When it was, it felt vague and mysterious; "*Nobody mentioned FND… it's so…I don't want to say*

niche but …different, you know" (Una). There was an implication that despite frightening symptoms, FND wasn't serious; "*I didn't feel as though there was any urgency… and to me … I felt like my memory was leaving me*" (Sam). While many were open to psychological components in their formulation, simply attributing FND to mental illness felt invalidating, and an excuse for professional inaction; "*ehm it just felt like a dismissive kind of you've got FND eh…saying without saying it's all in your head …it's all because you were abused…it's very easy for people to kind of block you*" (Bridget). Once the diagnosis of FND was given, several were discharged with no perceived plan, a contrast to other conditions.

Theme 4. Everyday interactions

While family and friends were often supportive, several patients experienced stereotyping and dismissal, threatening the veracity of their condition; "*As much as my family have grown and become supportive there were moments when they said 'get up just get up, go to school you're just being dramatic', so I guess that did change when we talked to the psychologist*" (Hailey). Several patients anticipated stigma from colleagues, which led to patients not disclosing their diagnosis, especially those who worked in health and social care settings; "*I was worried that they would be judgemental…I somehow believed and still do believe, that they will take the seizure bit seriously but not the functional bit*" (Sam). One patient described feeling stereotyped by colleagues, which they linked to them not believing the functional impairment with FND; "*they were like nah you just want time off …you just want extra benefits…I've never had a benefit in my life*" (Rose). Some gave up work altogether impacting sense of identity which again, triggered a cycle of reduced self-esteem and social exclusion.

Several patients used the online space to interact about FND, and for several their stigma experience really ignited in this domain. Reading inaccurate posts drove self-doubt, which was exacerbated when professionals were discredited; "*do you know what was very unhelpful recently… …from a COVID group…eh and basically it was to do with FND research being led by (name removed) basically he is a fraud, they are all frauds uhm… that just set me back just all the way* (Norah). It also arose when they encountered individuals whose experience did not fit at all with their own, leading to confusion and fear; "*I was like …wait a minute here …but I'm nothing like these guys on this website so what are they talking about*" (Charles).

Theme 5. Stages of knowledge

A consistent issue in all interviews was the lack of awareness of FND; *"if you had something that people have heard of they would be more sympathetic perhaps? But I think when you have something that people haven't heard of, that brings its own challenges"* (Una). It was demoralising and "othering" for patients to try and explain FND difficulties that were already hard to verbalise, to people who had never heard of it. The knowledge that did exist often reflected inaccurate, outdated models. Several patients educated others about misperceptions, adding additional burden; *"when I first told my best mate it was like, 'this is fake'...but in the end he went away and read it and he was like ken (you know what) this actually does make sense"* (Brendan).

The importance of having an explanation that fit their experience was outlined in all interviews, helping them feel less "othered" and more confident in narrating their difficulties to others; *"but just seeing the professional for maybe forty minutes like changes everything, I was like oh wow ok, that was easy once I actually was given the proper care"* (Laura). It also helped them with any self-doubt/blame that FND was their fault. All patients were realistic about how a clinician might not have all the answers, expressing a desire for open communication. Several commented on the lack of knowledge in the medical profession; *"I think they could maybe do with like more training in GP surgeries...but that just I think the general medical world could do with more education"* (Rose). Online information had the potential to be overwhelming and stigmatising, though it was often useful. However, FND remained elusive and hard to grasp for many; *"I don't find it hard to explain to them, I find it hard that they don't understand what I'm telling them"* (Ali).

Theme 6. Validation of patient experience

The majority of patients commented they felt understood and supported by professionals at some point in their trajectory. These professionals comprised several disciplines, and usually had existing knowledge of FND. When this happened, they felt that they and their FND experience were seen as valid and worthy of attention, in contrast to what they had heard before, read online, or internalised. *"I think it was a relief...that I was taken seriously if I'm honest and it wasn't just all in my head again"* (Orla). Specifically, taking time, being appreciated as an individual and demonstrating visibility of clinical signs were helpful. Having a follow-up appointment and a clear treatment plan were important: *"it just meant that...something significant*

was happening despite the vagueness of the diagnosis…there was a clear way of way of getting help" (Sam).

Despite some stigmatising experiences, family and friends they were also frequently sources of recognition and understanding. Families particularly were key in supporting patients; "*when I am at home I have people around me that understand and recognise when I could be having a day when I am more likely to have one of these episodes*" (Hailey). Given the difficulties grasping FND, it was often a journey for family/friends to get to the stage where they could be understanding and supportive, usually influenced by desire to learn more about FND; "*my partner … he has got the app on his phone and he's… he's like I don't understand it…I but I will learn, if there is anything I can do to help…he's been absolutely…it's been positive*" (Rose).

4. Discussion

4.1. Main findings

Our findings show that stigmatisation experienced by patients with FND unfolds and interacts though four main domains – the symptom experience, self, healthcare system and everyday interactions. FND was often perceived as something different, "niche", and mysterious. Patients were often not given the diagnosis in a typical way, and had to educate others about their condition, leading to distress, "othering" and feelings of separation. A negative cycle often ensued, where patients internalised their difficulties, feeling unable to share with people who could potentially support them, leading to avoidance and exclusion. Knowledge was a key factor throughout the process – a lack of knowledge propelled stigma, but was also an effective tool in countering stigma. Validation – recognising and affirming the patient and their experience was helpful towards alleviating stigma. While clinicians were not the sole origin of stigmatising experiences; nonetheless in all these cases, they played a powerful role in helping allay them.

4.2. Comparison with previous literature

Our findings reflect the literature on stigma in FND (2,3,31,32), and provide some further insights. We found that patients with a range of FND symptoms, including speech disturbance, visual and cognitive symptoms experience

stigma, phenotypes which have not been explored much in this realm previously. We found that negative mental health connotations associated with FND, while present, did not emerge as prominently as represented in the existing literature (2,3,31,32) and patients in this study were open to psychological components to their formulation and treatment. Furthermore, while concepts related to the self and identity have been explored previously in relation to FND (3) the weight of self-stigma and its impact on patients with FND were pertinent findings in this study.

The finding that stigma arises from everyday interactions is elsewhere in the literature in relation to functional seizures (2), with patients describing experiences of being misunderstood and stereotyped, similar to this study. However, these findings are usually overshadowed by explicit negative healthcare interactions which are far more pervasive in the literature (2,3,32). While negative healthcare interactions were significant in our study, they presented as one of several manifestations of the broader culture of the "othering" of functional within the healthcare system. Furthermore, while there is some evidence that representation of FND online is derogatory and offensive towards patients (33), the burden of online stigma experienced by patients is an unexplored area in the literature, and a further important finding in our study.

4.3. Strengths and limitations

Our findings are limited to a small sample, however, despite this, this sample was diverse in terms of gender, age and clinical presentation. There may be differences between those who decided to participate and those who didn't. Though we wanted to recruit patients as close to possible as diagnosis to minimise potential recall bias, a minority (three) were recruited nine months to five years post-diagnosis. We felt this was acceptable – the criterion of duration between diagnosis and interview was not in our formal inclusion criteria, as we felt it was more important to maximise sample variation, and include rarer presentations such as functional visual symptoms. Nonetheless, this could be regarded as a potential limitation. Our findings were obtained from a predominantly white UK sample, meaning we did not obtain experiences from other patient subgroups. We used open-ended questions which were not leading, allowing for a range of stigma-related experiences to emerge. Our sample was limited to a small region, where some clinicians have a particular interest in researching and treating FND, though we did

recruit from a range of clinicians, whose expertise lay outside FND. However, because of this, it is possible that other sources of stigmatisation were able to emerge as well as healthcare professional interactions.

4.4. Implications of results

Our findings suggest there are numerous ways that stigma could potentially be reduced. Given how stigma occurs as a cyclical process, it is likely that addressing one key area might impact another. The crucial role of the clinician has been described, linking in with the aspect of power that is inherent in the stigma process (12). In addition to the frontline patient interaction, clinicians are in a position to tackle stigma on a structural level, such as advocating for research/service funding and designing training programs and health policy.

The findings of this study suggest that training for clinicians could be improved, so FND is not perceived as "lesser" or "other". FND should be placed within formal core curricula, at an early stage in training, for all relevant specialties involved in the care of FND, (for example nursing, paramedical training, physiotherapy, speech and language therapy, occupational therapy and medical social work). In addition, the "hidden curriculum" (34) is also worth serious consideration by educators. This more subtle method of learning, which includes processes such as role modelling and informal conversations, is an influential vehicle for perpetuating stereotypes, but can serve to impart helpful attitudes.

Regarding the area of clinical communication, patients would benefit if clinicians imparted the diagnosis in a compassionate and confident way. There have been some studies focussing specifically on this area with positive results ((35), (36), (37)). Mentioning FND as a potential in the differential in the same way as any other condition would be useful during work-up. It could be possible that clinicians are aware of the stigma and misperceptions surrounding FND, and are therefore reluctant to mention FND as they are worried about alienating the patient or engendering mistrust. It is possible too, that clinicians may fear missing a diagnosis such as multiple sclerosis or epilepsy, which may be perceived as a larger clinical error than missing FND. That said, in a systematic review of 1466 patients with FND, the proportion of misdiagnosis was less than 4% after an average of 5 years of follow-up (38). Even after lengthy follow-up, the diagnosis remains stable—a recent 14-year follow-up study described a diagnostic revision rate of 1% (39).

Indeed, misdiagnosis occurring in the opposite direction (diagnosing FND as epilepsy for example) has the potential to be harmful (40). Regarding other areas of communication, it is important to validate the patient experience and where possible, allow time for a follow up appointment. Regarding the use of unguided internet self-education, directing patients to a single website is an approach that needs to be used thoughtfully, as an adjunct to appropriate care. While patients find this type of self-education valuable, it is not a replacement for treatment (41).

Regarding the symptom experience and self-stigma, it is worth remembering that patients are unlikely to bring forward these concerns. Self-stigma has been discussed often as occurring in tandem with perceived stigma – an individual's recognition that the healthcare system and public hold prejudice and will discriminate against them because of their presentation and diagnostic label (42). Therefore, clinicians may have a role in propagating self-stigma for patients, further highlighting the pressing need to address negative professional attitudes and public misperceptions. Furthermore, it would be helpful to be cognisant of other potential origins of self-stigma and explore further if necessary, or explain more specific aspects of FND that might bring this about. For example, the variability and distractibility inherent in FND could be explained so people understand why the symptoms are not constant or always visible. It is important to balance a sensitive approach while also not “othering” the patient further.

Regarding stigma from other sources, maintaining an open dialogue where possible with work, family and school/university could be helpful. Family and friends could be included more actively in the process of diagnosis and treatment, educating them alongside the patient. Maintaining communication with work, schools or universities such as written advice on what do to during seizures, or outlining a patients' abilities or restrictions could be helpful, to increase inclusion and reduce the potential of feeling “othered” in these domains. Regarding online stigma, advising patients and their caregivers about the fallacies/outdated models in the public domain and emphasising the selective trustworthiness of sources could also be useful.

Improving public knowledge around FND is paramount, and much work led by patients and professionals has already been done in this regard (43,44). The online domain will continue to be used by patients to interact about their illness and though it can be harmful, there are beneficial aspects to the online

space. This is an emerging area of importance in functional disorders (45,46). Future studies could assess the accuracy of online information and perceptions of FND in the public domain, and direct interventions accordingly.

Going forward, all the above interventions should be co-developed/delivered by patients with FND. In the last few years, individuals and groups have successfully navigated the complexities around the historical dualism that surrounds FND, acting as "translators" between clinicians and patients (11,44). Their continued involvement will be critical in transforming the misperceptions throughout the FND landscape.

5. Conclusion

Stigma unfolds as a layered process, influenced by surrounding structures, relationships, what is held internally and what has gone on before. It is alleviated by increasing knowledge and validating the patient experience. Interventions to target stigma could take the form of improved clinician training, communication, especially around point of diagnosis, and public interventions, co-produced with patients with FND.

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Supplementary material

Appendix 1: Discussion guide for interviews

Introduction

- Confirmation of audio recording
- Introduction or researcher and research aims
- Anonymity, right to withdraw (or take breaks)
- Reminder that discussion guide is only a guide and free to speak as things come to mind

Living with FND symptoms

- Now we might start with how things were before you received a diagnosis.
- What symptoms or problems were you having that prompted you to seek help? *Prompt:* Can you describe/list them?
- How did/do your symptoms impact on your life? (*Prompts:* physical impact, emotional impact (e.g. worry, anxiety), work/ employment impact, social impact, financial impact)
- What has it been like living with these symptoms? *Prompts:* how long had you had them for? Have they changed over time? How did you manage these symptoms?
- How did you decide you needed help?
- What has been/is the worst part? What helps the symptoms/makes them worse?

Getting help

- Can you tell me about your experience of getting help for your FND symptoms from a healthcare professional? (*Prompts* - What kind of reaction did you expect? What kind of help did you ask for?) Ask to elaborate on positive and negative aspects
- How was it the first time you went to your doctor (GP)? *Prompts:* Did you feel they understood your concerns? What happened following the appointment? What kind of reaction did you expect?
- Did the visit help you understand what was happening? Did you feel the GP understood what was happening? Yes/no – why did you feel that way
- After you saw the GP, before you saw the specialist, did you have any hopes or expectations about what might happen next? What was it like waiting to see them? Did you have an idea what your diagnosis might be? What were your thoughts on this?

- How was it the first time you went to a specialist? (This could be a neurologist or neuropsychiatrist) *Prompts:* did it feel any different seeing a specialist? Did you feel they understood your concerns? Did they explain your problems/symptoms? What kind of reaction did you expect? How was your understanding of what was going on after you saw the specialist?

Other medical history and experience of getting help outside FND setting

- We might just take a brief pause from your experience of FND now and visit some of your other healthcare history – it can be interesting to see how it compares with your FND experience.....Can you tell me briefly about any other medical problems outside your FND symptoms? (*Prompt:* Is there anything else you attend the doctor for?)
- Was your experience of getting help for your other health problems any different to getting help for your FND problems? {Might need to pick one here (for example your asthma/epilepsy/ulcer or whatever they have listed} (*Prompt*: Were there any differences in getting the care you needed?
- Would you say there were any differences in the way other diagnosis(es) were communicated?
- Would you say there were any differences in the way you were u felt understood/cared for/supported?

Diagnosis

- Going back to your FND now...you were telling me about when you went to the specialist – did you get a diagnosis/name for the problems you were having at this point? Can you tell me about your experience of receiving a diagnosis here? *Prompts*; How did they explain it to you? Did you understand it/Was it helpful? How did you feel after you got a diagnosis?
- Ask to elaborate on positive and negative aspects
- Did you agree with your diagnosis? *Prompts:* Has it changed how you view your symptoms? If you don't agree can you tell me a bit about how you view your symptoms?
- How would you explain your diagnosis to a close friend or family member?
- Outside the healthcare setting, what do you think others think about your FND – prompt, partner, family, friends, work (symptoms/diagnosis)?
- Did/do you have any negative/positive experiences in this regard? (*Prompt* – have you felt you have been untreated unkindly/disrespectfully in any way with regards to your FND outside the healthcare setting - for example by friends, family or online)?

- Or the opposite, have you felt you have been treated kindly by others outside the healthcare setting, in relation to your FND?
- Has your diagnosis changed how you view yourself?(prompt; opinion of yourself, self esteem, confidence, other areas).

Information and resources

- Thank you for sharing this information with me, it's really helpful to get a sense of all the different aspects to your experience. Going to something a little different now
- Were you given any information, educational material or other resources about your FND symptoms? *Prompts:* how was it given to you? Was it helpful? Would you share this information with others?
- Have you looked up any information yourself (for example, websites, forums, apps)? *Prompts:* how did you find it? Was it helpful? Would you share this information with others?)
- Is there anything that is still unclear to you? *Prompt:* what information do you need?

Future expectations

- Do you expect anything to change for you now that you have a diagnosis? *Prompt:* what does the future look like for these symptoms?
- Do you have any advice for healthcare professionals when treating people with FND symptoms?
- Is there anything we haven't covered that you feel might be important in relation to all this?
- Any final comments?

Close of interview

Close of interview: thanks, information about next contact etc

Chapter 7

Stigmatising content in health information related to functional disorders

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Abstract

Objective: While the internet is an increasingly popular place for people to access health information, the quality of information varies significantly between sources and clinical topics. To our knowledge there is no evidence about the quality of online information relating to functional disorders. In this study we analysed the quality and stigmatising content of Wikipedia articles in multiple languages.

Methods: We analysed Wikipedia articles of diagnostic categories relating to functional disorders across 14 languages. We rated the quality of each article on a self-developed 11-point scale in the domains of stigma, credibility, comprehensibility and usefulness. We also conducted a thematic analysis of articles, with the focus on categorising the types of stigmatising content.

Results: We identified 24 articles across 14 languages. Articles were rated moderate to high for stigmatising content, moderate to low for credibility and comprehensibility and low for usefulness. In many articles, we found examples of stigmatising beliefs, attitudes and suggested behaviour towards patients.

Conclusion: This analysis highlights the need to improve the quality of online health information in functional disorders. We call for other scientific researchers, healthcare professionals and interested people to engage with how information about functional disorders is presented online.

1. Introduction

While the internet is an increasingly popular place for people to access health information, the quality of information varies significantly between sources and clinical topics (1,2). Wikipedia is a prominent resource for the public, patients and healthcare professionals. Articles can be created and edited by anonymous users in almost 300 languages, which can result in differences in information quality. Low quality or inaccurate health information has the potential to mislead patients, contributing to unrealistic expectations or poor decision making (3). This is clearly seen in functional disorders, where inaccurate beliefs and negative attitudes are widespread.

Functional disorders is an umbrella term for a group of recognisable medical conditions which are thought to be due to changes to the functioning of the systems of the body rather than due to a disease affecting the structure of the body. To our knowledge there is no evidence about the quality of online information relating to functional disorders. In this study we analysed the quality and stigmatising content of Wikipedia articles in multiple languages.

2. Methods

Diagnostic categories in functional disorders vary significantly according to symptoms, country and treatment setting (4). Given the wide range of diagnostic categories used, we targeted a selection of categories rather than specific syndromes or symptoms:

- Functional Disorder (classification suggested by EURONET-SOMA group) (5).
- Somatoform Disorder (DSM-IV and ICD-10. Since articles were typically accompanied by somatisation disorder articles, we analysed these separately).
- Somatic Symptom Disorder (DSM-V)
- Bodily Distress Disorder (ICD-11)

We analysed articles in the following languages, spoken by the authors: Arabic, English, Croatian, Danish, Dutch, French, German, Greek, Italian, Japanese, Polish, Portuguese, Spanish and Swedish. First, we rated the quality of each article on a self-developed 11-point scale in the domains of stigma, credibility, comprehensibility and usefulness (anchors included 0 = not at all,

10 = absolutely). Ratings were based on the following definitions: stigma (associated negative or undesirable characteristics); credibility (validity of presented information); comprehensibility (coherence and ease of used language); and usefulness (practicality of the information). More information about quality ratings are provided in **Appendix 1, supplementary material**.

Second, we conducted a thematic analysis of articles (6). This had the focus on categorising the types of stigmatising content. The current study is part of the innovative training network ETUDE (Encompassing Training in fUnctional Disorders across Europe; https://etude-itn.eu/), ultimately aiming to improve the understanding of mechanisms, diagnosis, treatment and stigmatisation of functional disorders (7).

3. Results

We identified 24 articles across 14 languages. We found the most articles for Somatoform Disorder and Somatisation Disorder, and no articles for Bodily Distress Disorder. Article view counts were typically very low, with many receiving less than 30 views. Quality ratings are shown in **Table 1**, with individual language ratings in **Appendix 1, supplementary material.**

Articles were rated moderate to high for stigmatising content, moderate to low for credibility and comprehensibility and low for usefulness.

Credibility was rated low in articles that displayed factually incorrect or outdated information. For example, using diagnostic criteria found in older versions of diagnostic manuals (DSM-II or III) to define new cases, and suggesting that development of symptoms was age-defined (rather than diagnostic criteria defining a time period for the onset of symptoms). Comprehensibility was rated low when language use was overly technical and unclear. Thematic analysis of content found a wide range of stigmatising beliefs, negative attitudes and encouraged behaviours **(Table 2).**

Table 1: Quality rating of articles by stigmatising content, credibility, comprehensibility, and usefulness (0 = not at all, 10 = absolutely)

	Number of articles across languages analysed (n=13)	Stigma mean* (SD)	Credibility mean (SD)	Comprehensibility mean (SD)	Usefulness mean (SD)
Functional Disorder	4	6.38 (2.06)	6.63 (3.40)	4.75 (2.02)	3.25 (1.55)
Somatoform Disorder	8	6.06 (1.94)	6.06 (2.09)	6.19 (1.71)	4.94 (1.99)
Somatisation Disorder	9	5.22 (2.32)	4.89 (2.22)	5.94 (2.39)	4.22 (1.94)
Somatic Symptom Disorder	3	5.67 (2.89)	3.83 (1.26)	3.83 (1.89)	3.17 (1.44)
Bodily Distress Disorder	0	.	.	.	.

**Higher stigma scores indicates lower quality*

Table 2: Summary of stigmatising content in articles

Theme	Sub-theme	Summary
Stereotypes (beliefs about illness or patients)	Legitimacy	Suggestions that functional disorders are less legitimate or 'real' than illnesses with clearer pathology.
	Countering	Statements that appear to counter stereotypes and suggest equivalence with other illnesses. These include: Direct counters: statements that directly address a stereotype. Counters by diagnosis definition: statements that address a stereotype by referring to the diagnostic criteria of different conditions.
	Gender	When discussing prevalence, articles state that functional disorders are unseen, or rarely seen in men.
	Benefits	Suggestion that symptoms are associated with benefits of illness (avoiding conflicts, claiming benefits).
	Somatic fixation	Suggestion that patients refuse to accept, or have difficulty accepting, psychological explanations of illness.
	Communication	Suggestion that symptoms are poorly or vaguely described by patients, and descriptions are inconsistent over time.
Prejudices (attitudes towards patients/ emotions)	Discomfort	Suggestion that healthcare professionals are uncomfortable working with these patients and find it stressful.
Discrimination (behaviour towards patients)	Invalidation	Encouraging healthcare professionals to avoiding validation of symptoms.
	Negative language	Encouraging healthcare professionals to use negative language during description of clinical results and explanations.

	Selected example from article:
	"Normal emotional responses do not cause general health concerns or significant distress or dysfunction when actually suffering" (Somatisation Disorder, Japanese, p2)
	"Patients with somatization disorder will usually visit many doctors trying to get the treatment they imagine they need." (Somatisation Disorder, Spanish, p1)
	Direct:
	"The patient experiences real physical complaints, which are not imaginary. The complaints are also not consciously or deliberately imitated..." (Somatoform Disorder, Dutch, p1)
	"Although ordinary people, and even doctors, may see them as symptoms that have no physical basis, and that they "exist only in the patient's imagination", they are real symptoms for patients who suffer from them." (Somatisation Disorder, Arabic, p2)
	Counter by diagnosis definition:
	"The pain is not intentionally produced or feigned (as in feigned disorder or simulation" (Somatoform Disorder, German, p5)
	"Very rare its occurrence in men, it affects predominantly women." (Somatisation Disorder, Italian, p2)
	"They usually appear suddenly in stressful situations, allowing a person to choose a certain activity or responsibility or to focus a strong desired attention" (Somatoform Disorder, Croatian, p1).
	"Symptoms should not stem from an attempt by the patient to get attention or gain anything else by pretending to be ill" (Somatisation Disorder, Greek, p1)
	"A significant proportion of sufferers, rejecting the idea that their ailments may have a psychological origin, avoid help in that direction and turn to many doctors and specialists in search of new clinical examinations and treatment that will satisfy them" (Somatisation Disorder, Italian, p2)
	"The symptoms are vague in nature, often ill-defined but very serious..." (Somatisation Disorder, Italian, p2)
	"These disorders are very uncomfortable and stressful for health professionals" (Somatoform Disorder, Portuguese, p4)
	"These diseases pose a double problem: for the doctor who finds nothing, and for the patient for whom nothing is found. This leads to a double suffering: for the doctor who is not recognized as a carer, and for the patient who is not recognized as a patient" (Functional disorders, French, p1)
	"Refusal of the doctor to take the disorder into account, telling the patient "You have nothing, I have seen nothing". Negativity can momentarily reassure the patient (especially in hypochondriacs focused on organ dysfunction)." (Functional disorders, French, p2)
	"...the presence or absence of physical illness should not be discussed, and explanations of relaxation such as "there is no abnormality in the body" have already been said in other departments" (Somatisation Disorder, Japanese, p2)

4. Discussion

We found that the quality of health information on functional disorders in Wikipedia articles was inconsistent across diagnostic categories and languages. This reflects the fragmented landscape of healthcare systems and diagnostic criteria used in functional disorders. The low number of article views suggests these diagnostic terms are not widely used or known about by people seeking information.

In many articles, we found examples of stigmatising beliefs, attitudes and suggested behaviour towards patients. While these are not new, we also found examples of attempts to challenge stigma. This was done in two ways: by directly countering a stereotype, or by reference to the diagnosis criteria of other conditions **(Table 2).** There is evidence to suggest that actively addressing a stereotype can activate them despite being intended to suppress them (8,9). Authors would be better advised to provide appropriate context and evidence that demonstrates their concept rather than simply stating it. For the example of illness legitimacy, this might include discussion of mechanisms and treatment options rather than a single sentence statement that symptoms are real.

This analysis highlights the need to improve the quality of online health information in functional disorders. This is important as knowledge about these disorders is low (10), and the demand for health information through the internet is increasing (11). To support this, the ETUDE group has built a prototype for a Wikipedia article building on current scientific understanding. This is currently available on the EURONET-SOMA website (12). We are translating this into several languages, and we encourage healthcare providers worldwide as well as EAPM members to edit articles and translate into other languages. Future research should focus on sources of health information most used by patients with functional disorders, including specific functional somatic symptoms and syndromes. Further, more systematic quality assessment could be done using scales such as DISCERN (13). We call for other scientific researchers, healthcare professionals and interested people to engage with how information about functional disorders is presented online.

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Supplementary material

Appendix 1: Article information and quality ratings

Functional Disorder

Article information						Quality ratings (0 = not at all, 10 = absolutely)			
LanguageID (ISO 639-1 Code)	Date accessed	Name of article	Reference count	Word. count	Page views	Stigma rating	Credibility rating	Comprehensibility rating	Usefulness rating
AR	.	.	.	.	.	.	.	.	.
DA	02.05.2022	Funktionelle lidelser	4	843	<30	6	8	6	4
DE	28.04.2022	Funktionelle Syndrome	73	14800	<30	9	10	4.5	3.5
EN	02.05.2022	Functional Disorder	1	205	42	4	2	2	1
EL	.	.	.	.	.	.	.	.	.
ES	.	.	.	.	.	.	.	.	.
FR	28.04.2022	Trouble fonctionnel	1	683	<30	6.5	6.5	6.5	4.5
IT	.	.	.	.	.	.	.	.	.
HR	.	.	.	.	.	.	.	.	.
JA	.	.	.	.	.	.	.	.	.
NL	.	.	.	.	.	.	.	.	.
PL	.	.	.	.	.	.	.	.	.
PT	.	.	.	.	.	.	.	.	.
SE	.	.	.	.	.	.	.	.	.

Somatic Symptom Disorder

	Article information					Quality ratings (0 = not at all, 10 = absolutely)			
LanguageID (ISO 639-1 Code)	**Date accessed**	**Name of article**	**Reference count**	**Word. count**	**Page views**	**Stigma rating**	**Credibility rating**	**Comprehensibility rating**	**Usefulness rating**
AR	28.04.2022	اضطراب العرض الجسدي	14	875	<30	9	2.5	2.5	1.5
DA	.	.	.	.	.	.	.	.	.
DE	.	.	.	.	.	.	.	.	.
EN	02.05.2022	Somatic Symptoms Disorder	21	1126	281	4	5	3	4
EL	.	.	.	.	.	.	.	.	.
ES	.	.	.	.	.	.	.	.	.
FR	.	.	.	.	.	.	.	.	.
IT	.	.	.	.	.	.	.	.	.
HR	.	.	.	.	.	.	.	.	.
JA	28.04.2022	身体表現性障害 (Somatic Symptom Disorder)	31	6079	<30	4	4	6	4
NL	.	.	.	.	.	.	.	.	.
PL	.	.	.	.	.	.	.	.	.
PT	.	.	.	.	.	.	.	.	.
SV	.	.	.	.	.	.	.	.	.

Somatoform Disorder

Article information						Quality ratings (0 = not at all, 10 = absolutely)			
LanguageID (ISO 639-1 Code)	Date accessed	Name of article	Reference count	Word. count	Page views	Stigma rating	Credibility rating	Comprehensibility rating	Usefulness rating
AR	.	.	.	.		.	.	.	.
DA	.	.	.	.		.	.	.	.
DE	28.04.2022	Somatoforme Störung	28	2800	42	5.5	9	8.5	7
EN	.	.	.	.		.	.	.	.
EL	.	.	.	.		.	.	.	.
ES	28.04.2022	Trastorno somatomorfo	13	1094	<30	6.5	6.5	5.5	7.5
FR	28.04.2022	Trouble somatoforme	6	790	<30	5.5	7.5	7	5.5
IT	28.04.2022	Disturbo somatoforme	1	319	<30	4.5	6.5	7	2.5
HR	28.04.2022	Somatoformni poremećaj	3	480	<30	10	3	5	4
JA	.	.	.	.		.	.	.	.
NL	28.04.2022	Somatoforme stoornis	2	412	<30	3.5	6	8	5
PL	02.05.2022	Zaburzenia somatoformiczne	4	274	137	6	3	3.5	2
PT	28.04.2022	Transtorno somatoforme	9	1063	<30	7	7	5	6
SV	02.05.2022	Somatoforma störningar	2	274	<30	8	3	3	3

Somatisation Disorder

Article information						Quality ratings (0 = not at all, 10 = absolutely)			
LanguageID (ISO 639-1 Code)	Date accessed	Name of article	Reference count	Word. count	Page views	Stigma rating	Credibility rating	Comprehensibility rating	Usefulness rating
AR	28.04.2022	اضطراب الجسدنة	8	1071	<30	9	2	1.5	2.5
DA	.	.	.	.		.	.	.	.
DE	.	.	.	.		.	.	.	.
EN	02.05.2022	Somatization Disorder	31	1258	160	3	4	5	5
EL	28.04.2022	Σωματοποιητική διαταραχή	2	587	<30	8	8	9	2
ES	28.04.2022	Trastorno de somatización	2	281	<30	6.5	4.5	5.5	4
FR	.	.	.	.	.	.	.	.	.
IT	28.04.2022	Disturbo da somatizzazione	17	1377	<30	6	7	7	5.5
HR	.	.	.	.	.	.	.	.	.
JA	28.04.2022	身体化障害 (Somatization Disorder)	31	6079	<30	4	5	6	5
NL	28.04.2022	Somatisatiestoornis	1	471	<30	4	5	8	4
PL	02.05.2022	Zaburzenie somatyzacyjne	1	275	<30	4.5	1.5	3.5	2
PT	28.04.2022	Transtorno de somatização	10	555	<30	2	7	8	8
SV	.	.	.	.	.	.	.	.	.

Appendix 2: Prototype Wikipedia articles (English language versions)

Available on online supplementary material

Chapter 8

General discussion

General discussion

The aim of this thesis was to gain deeper insights into the measurement and manifestations of stigmatisation towards people with persistent somatic symptoms (PSS) by healthcare professionals. While there has been increased attention to stigmatisation in this context in recent years, little attention has been paid to the quality of its measurement. This thesis has filled in a gap here, providing a foundation for future stigma reduction intervention and evaluation. This thesis has developed and validated a measurement instrument that can be used to measure stigmatisation, the PSSS-HCP, explored manifestations of stigma in a clinical context, and the experience of stigma for patients.

This final chapter draws some conclusions that try to bring together the different threads of stigma theory, research and clinical practice. First, the main findings of the research are presented. Second, reflections on the findings are offered and contextualised within recent research. Third, some methodological reflections are provided. Fourth, implications for stigma interventions and future research are discussed.

1. Main findings

Our initial review of stigma measurement instruments **(Chapter 2)** found that while there were many examples of publications and instruments exploring stigmatising attitudes and behaviours of healthcare professionals, little attention was paid to their development or evaluation of measurement properties. Many instruments were developed for a single study only, constructs of measurement were rarely named or defined, there was insufficient evidence of instrument development, and insufficient evidence of content validity. This was particularly since several instruments used unvalidated instruments to assess the effectiveness of anti-stigma interventions. Our recommendation was the development of a generic PSS related stigma instrument, with the priority of establishing content validity.

We used our learning from the review to develop a new scale to measure stigmatisation towards people with PSS, the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP) **(Chapter 3).** Development was an iterative process consisting of research team review, item generation and cognitive interviewing of healthcare professionals in the UK. We analysed the relevance, comprehensibility and comprehensiveness of potential items, including the potential for social desirability bias. The provisional version of the PSSS-HCP contained 19 items, grouped across three domains (stereotypes, prejudice, discrimination).

The PSSS-HCP was then tested with 121 healthcare professionals across the UK to evaluate its factor structure, validity and reliability **(Chapter 4).** A three factor structure was identified (we named these othering, uneasiness in interaction, and non-disclosure) which accounted for 60.5% of the variance using 13 of the 19 tested items. The PSSS-HCP showed sufficient internal consistency (Cronbach's alpha = 0.84 for the total scale, 0.71-0.84 for the subscales) and sufficient test-retest reliability, intraclass correlation = 0.97 (95% CI 0.94 to 0.99, $p<0.001$). Convergent validity was sufficient between the PSSS-HCP and the Medical Condition Regard Scale, and we found no evidence of social desirability bias. We concluded that the PSSS-HCP can be used to measure PSS stigmatisation by healthcare professionals, encouraging further validation.

In **Chapter 5** we developed a Dutch version of the PSSS-HCP and evaluated its measurement properties among a sample of healthcare professionals in

the Netherlands. Translation was an iterative process involving forwards and backwards translations, cognitive interviewing, and expert recommendations. Our study design mirrored that of the UK validation, assessing the factor structure, validity and reliability of the PSSS-HCP. Our results demonstrated sufficient construct validity, reliability (model-based reliability and test-retest reliability) and minimal influence of social desirability bias. Model fits were similar between a two-factor correlated model and a modified bifactor model, and bifactor analysis suggested that the PSSS-HCP was mostly influenced by a general factor. This study provided further evidence that the PSSS-HCP is an appropriate measurement of stigmatising attitudes of healthcare professionals towards people with PSS.

Concerning the manifestations of stigma, this study highlighted the perceived poor treatment of people with PSS. When asked to compare how people with PSS were treated by healthcare professionals compared to people with symptoms with a clearer pathology, most participants said that people with PSS were taken less seriously (80.2%), treated as less of a priority (67.5%) and provided with poorer quality care (61.2%). This finding underlines the perceived relevance of stigma in this field and the importance of developing and evaluating stigma reduction interventions.

We also explored experiences of patients with a specific type of persistent somatic symptom, Functional Neurological Disorder (FND) **(Chapter 6).** While existing literature demonstrated that stigma exists for many patients with FND, and is associated with poorer quality of life, it was less clear how stigma unfolded in this context. We interviewed patients with a recent diagnosis of FND in the UK to explore experiences of stigma as it unfolded from symptom onset, through diagnosis and afterwards. We found that stigma did not unfold as a linear process. Rather it came from multiple interacting sources: through presenting characteristics of FND symptoms, through everyday social interactions, through negative internalised beliefs (self-stigma), and being treated as less legitimate (othered) by the healthcare system. Across these sources was a central theme of knowledge, which both fueled and countered stigma. Validation of the patient experience emerged as a theme that alleviated stigma. Specific advice for healthcare professionals included mentioning FND as a potential in the differential diagnosis, building education of FND into the core curriculum for healthcare professionals, validating the patient experience, and being sensitive to the potential origins of self-stigma.

Lastly, we explored the quality and stigmatising content of health information in Wikipedia articles **(Chapter 7).** While the internet is an increasingly popular place for people to access health information, the quality of information varies significantly between sources and clinical domains. Low quality or inaccurate health information has the potential to mislead patients, contributing to unrealistic expectations or poor decision making. Through an analysis of different diagnostic categories across 14 languages, we found that the quality of articles was very inconsistent, containing many examples of stigmatising content. We also found examples of attempts to challenge stigma. This was done in two ways: by directly countering a stereotype, or by reference to the diagnosis criteria of other conditions. We gave practical recommendations for online health information and a prototype that can be used for future communication.

2. Reflections on findings

2.1. Stigma continues to affect the quality of care people with PSS receive

Despite years of clinical and research interest, stigmatisation by healthcare professionals continues to be prevalent and negatively affects the care people with PSS receive.

The underlying stereotype behind this stigma is that people with PSS are less legitimate than people with illnesses with clearer pathologies. Specific contributing factors are consistent: a lack of formal education on PSS, and lack of specific skills in communication. Despite this consensus, many people with PSS continue to receive poor quality care and experience negative health outcomes – not only through the burden of their symptoms but through the stigma that they face. This thesis solidifies this evidence base and points to the need for structural changes to how people with PSS are cared for.

2.2 Stigma is present across healthcare systems

While the care trajectory for people with PSS is complex due to variability in diagnoses and treatments (1), our thesis demonstrates that healthcare professional stigma is present across countries and healthcare systems. This points to the need for interventions that are structural as well as interpersonal.

Among the heterogeneity of measurement instruments identified in our systematic review **(Chapter 2)** were key commonalities: endorsement of negative stereotypes, negative attitudes and emotional reactions towards people with PSS, and lack of confidence. This was across 24 different countries of sampled healthcare professionals (Europe, North America, South America, Asia, and Australasia), highlighting the widespread phenomenon of stigma against people with PSS. This breadth of stigma has been further highlighted in reviews of stigma in PSS from the patient perspective (2) and in FND from both patient and healthcare professional perspectives (3).

It is worth considering characteristics which might shape stigma within specific healthcare systems. We tested the PSSS-HCP in the UK (**Chapter 4**) and the Netherlands (**Chapter 5**), finding evidence of stigmatising attitudes in both countries. While the total score of the PSSS-HCP was higher in the UK than in the Netherlands (suggesting higher stigma), definitive comparisons cannot be made due to differences in the composition of sample populations. However, we might explore some possibilities for future investigation. There are key similarities between the healthcare systems of the UK and the Netherlands, namely support for universal access, and the role of general practitioners making referrals to further specialists (the 'gatekeeper' function). There are however key differences, including the healthcare insurance system of provision in the Netherlands, and the recent system pressures that are affecting care delivery in the UK (4). Given that perceived lack of time and resources of healthcare professionals is a key barrier to involvement in research (5), systems pressure in the UK might also force an evaluation of 'priority' and 'less priority' patients.

There is increasing evidence describing the impact of healthcare systems on the provision of care for people with PSS. In a recent qualitative study across Germany, the Netherlands, Poland and Italy, it was found that the interaction of structural and interpersonal factors within the healthcare system influenced the course of PSS symptoms (6). Systemic barriers such as limited consultation times and issues with insurance coverage were prevalent in Germany and the Netherlands, while access and trust issues were more prominent in Italy and Poland. While there were some common presented solutions to improve care (adequate consultation times, included reimbursement and treatment eligibility for PSS, improving education about PSS and establishing collaborative care pathways), factors such as a lack of trust are likely to exacerbate negative interactions.

2.3. The personal impact of stigma

By understanding stigma as a fundamental cause of health inequalities, reducing it should become even more a priority in clinical and research practice.

Stigmatisation in general is increasingly understood as a casual factor in shaping health outcomes. In the context of PSS there is less quantitative data on the outcomes of stigma, but stigmatisation is associated with decreased wellbeing (7), increased depression and anxiety (8), treatment non-adherence (9) and increased burden for caregivers (10). Key explanations provided for these associations include the psychological distress of not having an illness validated by a healthcare professional (8, 11), and disruption to health-seeking behaviour when stigmatisation is anticipated (8, 12).

The findings from our qualitative study (**Chapter 6**) support these explanations. Participants in our interviews described several ways in which the stigma affected their everyday lives. Self-stigma, where feelings of shame and embarrassment were internalised, led to people being afraid to socialise and losing confidence. Through feeling stereotyped by colleagues in the workplace, some participants gave up their work, losing an important part of their identity and sense of contribution. Some people described not disclosing their FND with healthcare professionals for fear of receiving further stigma. These findings align with the substantive body of qualitative research in this field, namely the withdrawal from the social sphere and loss of employment and educational opportunities (13), the distress of invalidation (2), and decreased engagement with healthcare (14). While further quantitative research could further demonstrate the casual link, the influential role of the healthcare professional is clear.

2.4. The need for formal education opportunities

A lack of knowledge about PSS plays an important role in stigma. Throughout the validation studies of the PSSS-HCP 9 (**Chapter 4 and 5**), we found that the total PSSS-HCP score was moderately correlated with lower perceived adequate knowledge about PSS and lower perceived adequate training. PSS is not widely or consistently taught in medical curricula, despite there being recognition of the importance of education in this area (15, 16). The curriculum itself symbolises what is important to learn, and which healthcare conditions are worthy of attention.

A lack of formal education has been consistently highlighted in the area of PSS, by healthcare professionals early in their careers (16, 17), as experienced specialists (18, 19), and through reviews of curricula taught to medical students (15). In a survey of healthcare professionals across Europe, a lack of perceived knowledge and adequate training was also highlighted (20). This is linked with perceived confidence and competence when caring for people with PSS, which affects the quality of interactions (16). Through **Chapter 6**, we reflected that this lack of knowledge by healthcare professionals presented itself in several ways, such dismissal of symptoms as 'normal', or multiple referrals to protracted routes to specialists. Inconsistent diagnostic behaviour by healthcare professionals included a reluctance to give a diagnosis, presenting a diagnosis as mysterious, or presenting a diagnosis as a choice. When healthcare professionals lacked knowledge, this forced patients with FND themselves to explain their symptoms to others, when the symptoms were already difficult to verbalise.

Further, where formal education opportunities are lacking, healthcare professionals learn instead through informal means. This has been described as the 'hidden curriculum', where healthcare professionals learn through informal social processes such as role modeling and interactions with senior colleagues (21, 22). While social modelling processes in themselves are not problematic, negative beliefs and attitudes can develop without being challenged through formal education. Therefore, it is important that knowledge about PSS is presented formally in healthcare professional education. This includes components of education such as mandatory assessment which assert its importance.

Online health information is becoming increasingly influential and high in demand (23). In **Chapter 6** we described people relying on the internet when information was lacking from healthcare professionals. While some examples were overwhelming for patients who had just received a diagnosis, lacked relevancy and highlighted extreme examples of FND, some participants highlighted their potential usefulness. In **Chapter 7** we highlighted examples of outdated and factually incorrect diagnostic information, as well as many examples of stigmatising content. This is an area which deserves more focus, though examples are emerging of online information developed with interactive explanatory models and actionable health advice in mind (24). AI-based approaches are also gaining increased research attention with the

aim of matching information to the right people, improving health literacy and combating an 'infodemic' (an abundance of information) (25).

2.5. Addressing the stereotypes against people with PSS

Stigma works through the labelling of difference between people, associating these differences with negative characteristics, and providing separation between "us" and "them" (26). We described this process throughout this thesis as othering. We found examples of othering during cognitive interviewing (**Chapter 3**), identified this as subscale during psychometric testing of the PSSS-HCP (**Chapter 4 and 5**), and a major theme through the experiences of patients with FND as they move through the healthcare system (**Chapter 6**).

The experience of feeling 'othered' is a core component of several qualitative studies of people with PSS, who have described being taken less seriously and treated more dismissively than other patients (2, 12, 27). This is supported in several observational studies, where perceived stigma was higher in people with functional disorders than comparable health conditions with a more clearly structural pathology (11, 28). Healthcare professionals themselves agree with this perception of othering and differential treatment. In our Dutch validation of the PSSS-HCP we found that healthcare professionals agreed that people with PSS were taken less seriously (80.2%), treated as less of a priority (67.5%) and provided with poorer quality care (61.2%) **(Chapter 5).**

Many stereotypes about people with PSS have consistently shown to not be true in recent research. These apply to understanding of aetiology of symptoms, patient understanding of aetiology, patient expectations, patient communication, and the role of explanation. For example, while the concept of 'somatic fixation' (patients only accepting somatic explanations of illness) is a concern among healthcare professionals, there is evidence that it is healthcare professionals themselves who prompt further diagnostic testing (29, 30) and frequently miss psychosocial cues from patients (31). Likewise, recent research has suggested that there are increased numbers of contacts and management strategies provided for people with a disease diagnosis in primary care than people with a persistent symptom diagnosis (Chaabouni et al., 2024, submitted). The educational approach taken to address these is often done through a 'myth busting' – directly naming

these stereotypes and then showing the balance of evidence to disprove or dismantle them (32).

While describing and dismantling stereotypes is an important component of stigma reduction, this is not enough without sufficient accompanying action. We found evidence of this in **Chapter 7**, where health information tried to counter stereotypes, but inadvertently acted to reinforce a sense of illegitimacy:

> *"The patient experiences real physical complaints, which are not imaginary. The complaints are also not consciously or deliberately imitated..." (Article for Somatoform Disorder, Dutch, p1)*

There is evidence to suggest that actively addressing a stereotype can activate it despite being intended to suppress, known as stereotype rebound (33, 34). So while 'myth busting' sheets are often used in educational material, it may be necessary to add a third column: applying learning in practice. This includes addressing and negating a stereotype, but providing practical advice about how to demonstrate this learning. An example of how this could happen is provided in **Appendix 1.**

2.6. Validation through communication

There is a growing body of evidence describing what patients with PSS expect from their interactions with healthcare professionals: they want emotional support, for the healthcare professional to believe and show interest in their symptoms, to be treated as equal partners, and a tangible explanation that can act as a plan to manage those symptoms (35, 36). Communication is a crucially important skill that can achieve this.

Communication is increasingly understood in its role in recognising the humanity of a patient. That is, building a connection with a person that focusses on their needs, rather than as a transactional relationship (37). In comparison to the biomedical aspect of the healthcare professional role (finding, isolating and treating disease), the importance of the psychosocial role can be understated. In interviews with general practitioners, they typically described their relationship-work as 'just support' or 'just reassurance' (5). While recent evidence suggests that expectations of patients with PSS are no different to other patients, they may seek more emotional and moral support (29, 30). Our qualitative study in **Chapter 6** suggested this too,

where patients with FND highlighted that being given sufficient time and being appreciated as an individual were helpful and validating. Therefore, building a strong relationship should be valued as important and meaningful work that can make a tangible impact to people with PSS.

Providing a meaningful explanation for symptoms is another key component of proper communication. The process of explanation offers an opportunity to demonstrate that the healthcare professional is taking the person with PSS seriously, a chance to discuss how the symptoms arise, and emphasise the potential for improvement or reversibility (38). While it may be incomplete (for example focussing on symptom generation and exacerbating mechanisms rather than root causes), it can help someone with PSS make sense of their experience (39). A recent intervention suggests that healthcare professionals can be taught to create personalised explanations for PSS using a REAL framework (40). This intervention involved: 1) Recognition (validating the patient experience; 2) Explanation (providing an explanation of symptoms that integrates brain and body); 3) Action (negotiating self-management strategies); and 4) Learning (jointly reviewing and modifying the other components as necessary). This has been evaluated as encouraging biographical repair in people with PSS – increasing acceptance and agency, helping them to find a new normal, and coming to terms with a revised biography which incorporates elements of their illness without being defined by it (41). Therefore, communication not only provides an important bridge for validation, but can play an active role in the management of PSS.

3. Methodological considerations

This thesis combines multiple methods to explore the measurement and manifestations of stigma by healthcare professionals. These included a systematic review, semi-structured interviews with patients, cognitive interviewing with healthcare professionals, surveys, and secondary data analysis of available online health information. The combination of methods allowed us to conceptualise PSS related stigma and apply our theory in a range of contexts.

While specific strengths and weaknesses of studies have been discussed in each thesis chapter, there are some methodological considerations that cut across this thesis.

3.1. Reflexivity in stigma research

When trying to understand experiences of stigmatisation, there can be serious consequences of not exploring this in an ethical and sensitive way (42). We have tried to question our own position as researchers.

During interviews with healthcare professionals (**Chapter 3**), we wanted to understand the barriers and frustrations of professional work while making it clear that we did not endorse the statements in the tested items. This could be challenging when participants agreed with stigmatising statements. We used the opportunity to explore how and why they held that view, and what could be improved. In some cases healthcare professionals used the PSSS-HCP to reflect on their own beliefs and attitudes.

While interviewing patients with FND (**Chapter 6**), we were aware that many participants were sharing distressing experiences of stigmatisation. This was particularly so for people with PSS who have often experienced *epistemic injustice* – being challenged in their capability to reliably narrate their own experience (43). We felt it was important to validate these experiences, and encourage the reduction of stigma (44).

We felt that it was important to consider that research might inadvertently reproduce stigma (44). We managed this by conducting the research with an interdisciplinary team. The researchers consisted of a medical sociologist/anthropologist, general practitioners, psychologists, stigma experts in mental health and employment, epidemiologists, medicine students, a consultant liaison psychiatrist and a neurologist. We have also worked closely with *Stichting De Bagagedrager* throughout, who have experience in developing and running workshops to discuss stigma in a safe and even playful way. This integration of perspectives has been essential for researching stigma in a way that is embedded in theory and practice.

3.2. Measurement theories

Throughout the development and evaluation of the PSSS-HCP we adopted the measurement theory of Classical Test Theory (CTT). CTT assumes that an observed score of an observable construct (stigma) is a reflection of the 'true score' plus an associated measurement error. This theory has been widely adopted, with sufficiency of measurement properties increasingly

standardised through the COSMIN group (45). Key assumptions of CTT are that individual items contribute equally to the construct of stigma, and that in order to assess any change of score over time, the exact same test must be administered.

Another approach to measurement is item response theory (IRT). This theory posits that a person's individual performance on an individual test-item is shaped by the amount of the underlying trait (in our case, stigma), that that person has. Therefore, everyone falls on the continuum of that trait to a greater or lesser extent. The extra assumption for IRT models is that the items can be ordered according to their 'difficulty'. While these measurement theories offer different analysis approaches, they are not mutually exclusive. IRT could therefore be used in future research to investigate individual item characteristics of the PSSS-HCP, and the extent to which each item individually contributes to stigma.

In **Chapter 6** we used confirmatory factor analysis to assess different model fits of the PSSS-HCP. The adoption and evaluation of higher order models (such as the bifactor model) is a relatively recent development. While there is some guidance around interpretation of sufficient reliability coefficients (such as omega coefficients) and ancillary indicators of dimensionality (46, 47), it would benefit researchers to have specific guidance from the COSMIN group.

3.3. The use of generic stigma instruments

This thesis questions the use of generic stigma instruments without evaluation of content validity. Specifically, we found problems relating to the use of mental health instruments, and use of items measuring social distance.

First, we found that several items related to mental illness stigma to be inappropriate in the context of PSS stigma. These included items exploring perceived peril (e.g. how dangerous people were perceived to be, the protection of the public from people with mental illness, appropriateness of working with children), psychological treatment pathways (e.g. references to psychiatric treatment), and specific stigmatising lexicon ('mad, nutter, crazy'). The perceived interpretation of PSS as psychological (and the anticipated reaction to this) plays an important role in the stigmatising process. Even the name of a scale (e.g. 'mental Illness scale adapted for PSS') might give a false impression of the intentions of the researchers.

Second, we found that items relating to social distance were often not relevant or comprehensible. Social distance refers to the desired proximity between oneself and another in social situations (48). From the perspective of healthcare professionals, this could mean the desired proximity in a professional context, but also through personal social situations. We found that items based on social distance were interpreted as not relevant, or not clearly understood (**Chapter 3**). Similarly, when we tested social distance items quantitatively, these did not psychometrically fit through item-total correlations (**Chapter 4**). We suggested that this could be because social distance measures are commonly tested where the dimensions of the stigmatised condition are high in perceived peril.

These insights have consequences for generic questionnaires that continue to be used in the context of PSS, such as the Illness Perception Questionnaire and Social Distance Scale. Specific versions of these scales for PSS might need to be developed in the future, for example more nuanced items around contributing causal factors (49).

3.4 The need for triangulation of measurement

As we explained in our general introduction (**Chapter 1**), we focused on the development of an explicit self-reported instrument in this thesis. However, given the complexity of stigma as a social process, the comparison and triangulation of different measurement methods is needed.

Implicit measurement instruments are increasingly being used in the context of PSS, aiming to reveal stigmatising attitudes and behaviour through experimental study designs. Examples of these include judgements of pain in hypothetical patients (50) or response latencies (through a type of Implicit Association Test) (51). One recent study of healthcare professional directly compared explicit and implicit attitudes towards FND (52). They found that while participants reported strong explicit attitudes that FND was legitimate, they demonstrated an FND illegitimate bias through implicit testing. Attitudes about FND-illegitimacy were negatively associated with likelihood of referral to physical interventions such as physiotherapy and treatment optimism. This suggests that implicit bias is an important indicator of behaviour. Further, while this and other recent stigma research suggests that implicit and explicit measures are not necessarily related, they might be reflecting different dimensions of stigma (53).

Further types of measurement of stigma include assessment of real-world behaviour. Examples of this include communication during clinical consultations (54, 55), treatment recommendations (51) and comparing the management strategies towards patients with persistent symptoms versus those with somatic or psychiatric diagnoses (Chaabouni, 2024).

3.5. Addressing social desirability bias

We have considered the potential influence of social desirability throughout this thesis. While social desirability bias can never be eliminated from research, we aimed to directly address and mitigate this. Through the cognitive interviewing process we tested this (**Chapter 3**), both through individual items and when answering the instrument as a whole. Through our validation studies (**Chapter 4 and 5**), we used short forms of the Marlowe Crowne Social Desirability Scale to assess potential social desirability bias.

These attempts are not without their limitations. In recent years there has been a debate about what social desirability scales such as the Marlow Crowne Social Desirability Scale essentially measure - either a particular 'response style' or a personality 'substance' that drives responses. A recent review found that while researchers using social desirability scales interpreted higher associations as evidence of desirability bias, these interpretations were not always clear (56). Rather, their recommendation was that researchers should test specific traits of interest (such as self-control) instead of ambiguous social desirability scores.

A more pressing type of social desirability bias might present itself through sample bias. That is, to avoid expressing negative views, a participant would choose not to participate in the study itself. While we aimed to maximise variation in our validation studies by contacting a range of healthcare professional organisations, the sample was self-selecting and therefore more willing to share their views about people with PSS. This is a limitation, which can be addressed in future research by adopting more systematic sampling methods.

4. Implications for practice and research

The results from this thesis have several implications for stigma reduction interventions and future research. These are summarised as a series of statements in **Appendix 2**.

While there are important technical aspects of PSS that warrant specific skills and support for healthcare professionals (for example around communication), there is as much focus on returning to fundamental aspects of person-centred care. These are particularly important for managing people with PSS, including attention to a person as much as a patient, treating the concerns of someone seeking medical help seriously, discussing and treating PSS as an illness like any other, and building a relationship as equal partners (57).

4.1. Recommendations for stigma reduction interventions

Interventions to reduce stigma have previously been categorised as educational (providing information about the stigmatised group to help make more informed decisions), contact (meeting members of the stigmatised group), or protest-based (protesting situations where experiences of stigma occur) (58). There are several key components of interventions that are important for reducing stigma:

First, interventions should encourage increased understanding and application of the biopsychosocial model of medicine. Where there are perceived dualisms between the mind and body, it is important that interventions encourage healthcare professionals to consider all relevant biological, psychological, and social factors that might be influencing the persistence of symptoms. This includes actively listening and responding to cues that patients raise about relevant factors. Healthcare professionals should learn how to explore these factors in conversation with a patient, and how to provide explanations that clearly explain how the brain and body interact with each other. Collaborative care between healthcare professionals can further improve the integration of different disciplines and care provision (59).

Second, it is important that interventions build specific communication skills for healthcare professionals when working with people with PSS. These act as a demonstration of the legitimacy of PSS, helping alleviate difficult encounters and building confidence of healthcare professionals. There are

a number of promising communication interventions reporting higher self-efficacy among healthcare professionals (60) and patient outcomes (61).

Third, reflection is an important component of interventions that can help healthcare professionals understand their own attitudes and behaviours. This could be done by directly assessing participants' own level of stigma (for example by using the PSSS-HCP), which could comprise an educational outcome itself (62). Further, reflection is an important attribute of maintaining a professional identity (63), which can be strained through challenging interactions. Given that negative emotions play such an important role in the stigmatising process, reflection can provide a mechanism to talk through difficult encounters and learn from peers.

Fourth, interventions should involve meaningful contact with people with PSS. Examples of this could include facilitation of the intervention by people with PSS, role-playing of clinical consultations, or more indirectly through videos. However, there is evidence to suggest that more active forms of contact are more effective than passive forms (64). There are also conditions for effective contact. The intervention should be targeted to specific healthcare professionals, be local (for example, within the regional context), people with lived experience should be 'credible' (meaning, be in the process of management of PSS or in recovery), and continuous (requiring ongoing effort) (65).

Lastly, it is important that future stigma interventions are designed so that they can be appropriately evaluated for their impact. This includes a clear conceptualisation of stigma, which is linked to the healthcare professional role (66). The intervention should discuss the mechanisms in which the desired change should happen, and use a measurement instrument with suitable content validity for the target population. We have built the PSSS-HCP to be able to be used as such an outcome measurement instrument.

4.2. Recommendations for future research

There are five key areas of research that can continue from the research presented here.

First, there is room for further evaluation of measurement properties of the PSSS-HCP. The priority here is evaluation of the responsiveness to change. Detecting minimally important change is integral to the evaluation of stigma

reduction interventions. Throughout the development of the PSSS-HCP, we have been attentive to the range of terminology applied both for specific types of PSS, and as an umbrella term. Given a lack of consensus in our cognitive interviewing, we suggested that it was possible to substitute specific terms without compromising content validity. Further evaluation of the PSSS-HCP could test this assumption, including the viability of the PSSS-HCP for specific types of PSS, functional disorders, and PSS that are diagnosable as mental health disorders (such as Somatic Symptom Disorder).

Second, the PSSS-HCP could be used alongside and compared to other types of measurement of PSS related stigma. There are very few examples of research doing this currently, with a notable example being the comparison of implicit and explicit measurement instruments in the case of functional neurological disorder (FND) (52). Given that the PSSS-HCP can readily be administered online, it would be feasible to test this among other research methods.

Third, the PSSS-HCP could be used to predict which types of healthcare professionals, and what characteristics of healthcare professionals have the most stigma towards people with PSS. This would establish a priority for increased support and stigma reduction intervention.

Fourth, is the examination of intersectionality and PSS related stigma. Further research could be done to assess what types of patient characteristics (for example, sex-based and racial differences) influence stigmatising attitudes and behaviours.

Fifth, there is scope to build on existing qualitative evidence from the perspectives of healthcare professionals. We know that there is extensive research about the challenges healthcare professionals face in communication (67), their concerns about somatic testing (29, 30), the absence of formal education (16), and the ways in which negative attitudes develop and persist (22). However, we know less about what happens when stigmatising attitudes are identified. How do healthcare professionals recognise and respond to their own biases? Are healthcare professionals aware of multiple sources of stigma (such as self-stigma of patients), and how do they address this? Recent research exploring healthcare systems in PSS found that while negative attitudes could inspire some healthcare professionals to be active and improve their care, it could force others to

disengage (6). Where there is reflection on this, the results are candid. For example, junior doctors expressed their desire to avoid patients with medically unexplained symptoms (16). During the clinical consultation, there is qualitative research exploring how communication can be improved when patient and clinician review the consultation together (55). Further qualitative research could explore how negative attitudes are resolved in practice, and what support healthcare professionals seek.

5. Conclusion

While there has been increased attention to stigmatisation in the context of PSS in recent years, little attention has been paid to the quality of its measurement. This thesis has developed and validated a measurement instrument that can be used to measure stigmatisation by healthcare professionals, the PSSS-HCP. We have explored manifestations of stigma in a clinical context, the experience of stigma for patients, and assessed the quality of online health information.

There is great potential for interventions to improve the knowledge, attitudes and behaviour of healthcare professionals towards people with PSS. While not the only source of stigma for people with PSS, reducing stigmatisation by healthcare professionals can have a major and lasting impact on health outcomes. We hope that this thesis provides a foundation for future stigma reduction intervention and evaluation.

Appendix 1: A modified 'myth-busting' sheet for PSS

Myths	Current scientific understanding
Persistent somatic symptoms are psychological ('all in the head')	Multiple biological, psychological, and social risk factors contribute to the persistence of somatic symptoms (68)
Persistent somatic symptoms are always a diagnosis of exclusion	There is increasing progress on diagnosing PSS based on internally consistent presenting characteristics of symptoms (32, 69)
Patients with persistent somatic symptoms use more resources than people with other disease diagnoses	There is recent evidence suggesting that patients with persistent symptom diagnoses use less resources than people with disease diagnoses (even adjusted for number of visits) (Chaabouni et al., 2024)
Patients with persistent somatic symptoms are less willing to consider psychosocial contributors to illness ('somatic fixation')	Healthcare professionals themselves are more likely to avoid raising psychosocial contributors to illness (31)
Patients with persistent somatic symptoms are more likely to pressure healthcare professionals for somatic testing and treatment.	Patients with PSS are not more likely to pressure healthcare professionals for testing or treatment than other patients. However, they may seek more emotional and moral support. (29, 30).
Patients with persistent somatic symptoms communicate about their symptoms differently to patients with similar but explained conditions.	Evidence exploring patient language found no systematic variations between those with 'medically unexplained symptoms' than those with more 'clearly explained' symptoms (71).
Discussing symptoms will necessarily exacerbate them (somatisation theory) (72)	Interventions focussed on healthcare professional communication can lead to sustained improvement in single and multiple PSS (73).

	In practice
	Exploring relevant biological, psychological and social factors during presentation of PSS. Countering stereotypes when raised (by patients or healthcare professionals).
	Using positive diagnostic criteria where possible to rule in PSS, rather than a diagnosis of last resort. This includes raising the possibility of PSS earlier in the differential diagnosis. This explanation of PSS should be reconsidered when symptoms change.
	PSS should be considered using a stepped care approach, based on the severity of symptoms and level of impairment (70). A first step from a general practitioner should involve education about the symptoms, shared decision making towards a time contingent treatment plan, and regularly scheduled appointments. If PSS is more severe, general practitioners should focus on collaborating with or referring to other specialists as appropriate.
	Raising personally relevant contributors to illness when they are prompted by the patient, and excluding factors then they are not considered relevant to the patient.
	Providing validation of the distress of symptoms.
	Following on raised cues by patients. Mirroring relevant language that is introduced by the patient (for example, if patient refers to fatigue).
	Following the REAL model of clinical consultation (Recognition, Explanation, Action, Learning) (40).

Appendix 2: Some propositions for further research and discussion

- **Healthcare professional stigma directly affects health outcomes.** While research in the context of PSS points to associations between perceived stigma and poor health outcomes, wider stigma research emphasises causal effects. Discriminatory behaviour can cause iatrogenic harm, while negative interactions with healthcare professionals can themselves be traumatic. By understanding stigma as a fundamental cause of health inequalities, reducing it should become even more a priority in clinical and research practice.

- **We can improve knowledge and build specific skills to reduce stigma.** Examples of these include positive communication strategies, and guided group reflection about experiences of working with people with PSS. Contact with people with PSS should be a key component of these interventions. Given the high prevalence of PSS, these should be built into formal medical curricula and be seen as central and mandatory, rather than optional. Interventions should be thoroughly evaluated for their impact on stigma and health outcomes for people with PSS.

- **Stigma relies on grouping and labelling people as different, rather than seeing an individual.** Person-centred care is essential to understanding the factors that shape PSS (biological, social, psychological), providing explanations that are the most relevant and acceptable, and building a strong relationship.

- **Stating that a stereotype is not true is not enough on its own to reduce stigma.** For example, stating that an illness is 'real' as a healthcare professional does not necessarily provide comfort. Rather, it separates from conditions that are more 'obviously' or 'demonstrably' real. Demonstration of legitimacy comes through validation of patients' distress, using acceptable explanations of illness, and management strategies developed in partnership between healthcare professional and patient.

- **Treat PSS the same as any other health condition:** while we are learning more about the aetiological mechanisms of PSS, this does not prevent healthcare professionals from taking action now. This includes introducing PSS earlier in the differential diagnosis, making a confident diagnosis

based on identifying characteristics, and using explanations to make a plan of action together with the patient.

- **If we truly believe in a biopsychosocial approach to medicine, this must include all health conditions, not just those that are 'contested' or lacking positive diagnostic criteria.** When applied to PSS, this includes evaluating new treatments for PSS that do not just focus on psychological contributing mechanisms such as cognitive behavioural therapy. When psychological treatments are used, their multifactorial use should also be explained (for example, the role of antidepressants on the central nervous system).

- **The act of measurement is not neutral, and never 'complete'.** Rather, specific constructs are developed, and measurement is always done in a specific context. Similarly, measurement instruments are never completely validated – rather they are evaluated in specific contexts and continually tested.

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Summary

Introduction and rationale for thesis

Persistent somatic symptoms (PSS) describe recurrent or continuously occurring symptoms such as fatigue, dizziness, or pain that have persisted for at least several months. These include single symptoms such as chronic pain, combinations of symptoms, or syndromes meeting the criteria for functional disorders such as fibromyalgia or irritable bowel syndrome. These symptoms are known to have a high personal impact on daily functioning and quality of life, work participation, and healthcare costs. On top of the distress and suffering caused by the symptoms themselves, people with PSS have reported that their symptoms are not taken seriously, dismissed as emotional problems, or outright fabrication, and their truthfulness and accuracy in describing symptoms is questioned. Throughout this thesis, we refer to these negative beliefs, attitudes, and behaviours as stigma.

A key component to stigma in healthcare is the personal beliefs, attitudes and behaviours of healthcare professionals. It is well reported that many healthcare professionals find the care of people with PSS difficult, and that there are many barriers to providing healthcare to people with PSS. Healthcare professionals in particular report feeling insecure about communication. This includes providing adequate explanations for PSS, addressing psychosocial factors that can play a role in symptoms, or applying a more person-centred communication style. This lack of perceived confidence and competence has negative effects, for example limiting interactions with patients with PSS where possible, or compensating by ordering inappropriate diagnostic procedures and interventions.

In recent years there has been increased attention to how healthcare professionals work with people with PSS. Research has primarily focused on the many examples of stigmatisation by healthcare professionals towards people with PSS, qualitative exploration of why this happens, and the development of educational interventions. However, little attention has been paid to how we measure stigma in this context. By establishing a valid and reliable measurement instrument, we would be able to provide a foundation for future stigma intervention development and evaluation.

This thesis studies stigmatisation by healthcare professionals towards people with PSS. This includes the development of a valid and reliable measurement instrument that can be used to measure healthcare professionals'

stigmatisation. This aims to provide a foundation to develop and evaluate future stigma interventions, as well as providing a more specific understanding of stigma in the context of PSS. We explore manifestations of stigma in a clinical context, the experience of stigma for patients, and the quality of online health information.

Main findings

Our initial review of stigma measurement instruments **(Chapter 2)** found that while there were many examples of publications and instruments exploring stigmatising attitudes and behaviours of healthcare professionals, little attention was paid to their development or evaluation of measurement properties. Many instruments were developed for a single study only, constructs of measurement were rarely named or defined, there was insufficient evidence of instrument development, and insufficient evidence of content validity. While we found the enduring attention to measuring stigma (and related constructs) commendable, we found the lack of evaluation of measurement properties problematic. This was particularly since several instruments used unvalidated instruments to assess the effectiveness of anti-stigma interventions. Our recommendation was the development of a generic PSS related stigma instrument, with the priority of establishing content validity.

We used our learning from the review to develop a new scale to measure stigmatisation towards people with PSS, the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP) **(Chapter 3).** Development was an iterative process consisting of research team review, item generation and cognitive interviewing of healthcare professionals in the UK. We analysed the relevance, comprehensibility and comprehensiveness of potential items, including the potential for social desirability bias. The provisional version of the PSSS-HCP contained 19 items, grouped across three domains (stereotypes, prejudice, discrimination).

The PSSS-HCP was then tested with 121 healthcare professionals across the UK to evaluate its factor structure, validity and reliability **(Chapter 4).** A three factor structure was identified (we named these othering, uneasiness in interaction, and non-disclosure) which accounted for 60.5% of the variance using 13 of the 19 tested items. The PSSS-HCP showed sufficient

internal consistency (Cronbach's alpha = 0.84 for the total scale, 0.71-0.84 for the subscales) and sufficient test-retest reliability with an intraclass correlation = 0.97 (95% CI 0.94 to 0.99, p<0.001). Convergent validity was sufficient between the PSSS-HCP and the Medical Condition Regard Scale, and we found no evidence of social desirability bias. We concluded that the PSSS-HCP can be used to measure PSS stigmatisation by healthcare professionals, encouraging further validation.

In **Chapter 5** we developed a Dutch version of the PSSS-HCP and evaluated its measurement properties among a sample of healthcare professionals in the Netherlands. Our results demonstrated sufficient construct validity, reliability (model-based reliability and test-retest reliability) and minimal influence of social desirability bias. Model fits were similar between a two-factor correlated model and a modified bifactor model, and bifactor analysis suggested that the PSSS-HCP was mostly influenced by a general factor. This study provided further evidence that the PSSS-HCP is an appropriate measurement of stigmatising attitudes of healthcare professionals towards people with PSS.

An additional and important finding from this study was the perceived poor treatment of people with PSS. When asked to compare how people with PSS were treated by healthcare professionals compared to people with symptoms with a clearer pathology, most participants said that people with PSS were taken less seriously (80.2%), treated as less of a priority (67.5%) and provided with poorer quality care (61.2%). This finding underlines the perceived relevance of stigma in this field and the importance of developing and evaluating stigma reduction interventions.

We also aimed to explore in this thesis how stigma unfolds from the perspectives of patients. To do this, we explored experiences of patients with a specific type of persistent somatic symptom, Functional Neurological Disorder (FND) **(Chapter 6).** While existing literature demonstrated that stigma exists for many patients with FND, and is associated with poorer quality of life, it was less clear how stigma unfolded in this context. We interviewed patients with a recent diagnosis of FND in the UK to explore experiences of stigma as it unfolded from symptom onset, through diagnosis and afterwards. We found that stigma did not unfold as a linear process. Rather it came from multiple interacting sources: through the symptom experience, through "othering" by the healthcare system, through everyday

interactions and from within the self. Across these sources was a central theme of knowledge, which both fueled and countered stigma. Validation of the patient experience emerged as a theme that alleviated stigma. Specific advice for healthcare professionals included mentioning FND as a potential in the differential diagnosis, building education of FND into the core curriculum for healthcare professionals, validating the patient experience, and being sensitive to the potential origins of self-stigma.

Lastly, we explored the quality and stigmatising content of health information in Wikipedia articles **(Chapter 7).** While the internet is an increasingly popular place for people to access health information, the quality of information varies significantly between sources and clinical domains. Low quality or inaccurate health information has the potential to mislead patients, contributing to unrealistic expectations or poor decision making. Through an analysis of different diagnostic categories across 14 languages, we found that the quality of articles was very inconsistent, containing many examples of stigmatising content. We also found examples of attempts to challenge stigma. This was done in two ways: by directly countering a stereotype, or by reference to the diagnosis criteria of other conditions. We gave practical recommendations for online health information and a prototype that can be used for future communication.

Implications for practice and research

There are several key components of interventions that are important for reducing stigma:

- Interventions should encourage increased understanding and application of the biopsychosocial model of medicine. Where there are perceived dualisms between the mind and body, it is important that interventions encourage healthcare professionals to consider all relevant biological, psychological, and social factors that might be influencing the persistence of symptoms.
- Developing skills in personal communication is essential for healthcare professionals providing care to patients with PSS. the process of explanation offers an opportunity to demonstrate that the healthcare professional is taking the person with PSS seriously, a chance to discuss how the symptoms arise, and emphasise the potential for improvement or

reversibility. Building a strong relationship should be valued as important and meaningful work that can make a tangible impact to people with PSS.

- Reflection is an important component of interventions that can help healthcare professionals understand their own attitudes and behaviours. This could be done by directly assessing participants' own level of stigma, which could comprise an educational outcome itself. Given that negative emotions play such an important role in the stigmatising process, reflection can provide a mechanism to talk through difficult encounters and learn from peers.
- Interventions should involve meaningful contact with people with PSS. Examples of contact could include facilitation of the intervention by people with PSS, role-playing of clinical consultations, or more indirectly through videos. However, there is evidence to suggest that more active forms of contact are more effective than passive forms. There are also conditions for effective contact. The intervention should be targeted to specific healthcare professionals, be local (for example, within the regional context), people with lived experience should be 'credible' (meaning, be in the process of management of PSS or in recovery), and continuous (requiring ongoing effort).
- Future stigma interventions should be designed so that they can be appropriately evaluated for their impact. This includes a clear conceptualisation of stigma, which is linked to the healthcare professional role. The intervention should discuss the mechanisms in which the desired change should happen, and use a measurement instrument with suitable content validity for the target population. We have built the PSSS-HCP to be able to be able to be used as such an outcome measurement instrument.

The five key areas of research that can continue from the research presented here include: 1) further evaluation of the measurement properties of the PSSS-HCP; 2) comparison of different methods to measure stigmatisation by healthcare professionals; 3) prediction of which types of healthcare professionals, and what characteristics of healthcare professionals have the most stigma towards people with PSS; 4) examination of intersectionality and PSS related stigma; and 5) qualitative research exploring how healthcare professionals recognise and respond to their own biases.

Conclusion

While there has been increased attention to stigmatisation in the context of PSS in recent years, little attention has been paid to the quality of its measurement. This thesis has developed and validated a measurement instrument that can be used to measure stigmatisation by healthcare professionals, the PSSS-HCP. We have explored manifestations of stigma in a clinical context, the experience of stigma for patients, and the stigma in online health information.

There is great potential for interventions to improve the knowledge, attitudes and behaviour of healthcare professionals towards people with PSS. While not the only source of stigma for people with PSS, reducing stigmatisation by healthcare professionals can have a major and lasting impact on health outcomes. We hope that this thesis provides a foundation for future stigma reduction intervention and evaluation.

Samenvatting

Inleiding en rationale van het proefschrijft

Aanhoudende lichamelijke klachten (ALK; persistent somatic symptoms (PSS) in het Engels) beschrijven steeds terugkerende of persisterende symptomen zoals vermoeidheid, duizeligheid of pijn die al meerdere maanden bestaan. Hieronder vallen enkelvoudige symptomen zoals chronische pijn, combinaties van symptomen of syndromen die voldoen aan de criteria voor functionele stoornissen zoals fibromyalgie of prikkelbare darm syndroom. Van deze symptomen is bekend dat ze een grote persoonlijke impact hebben op het dagelijks functioneren en de kwaliteit van leven, de arbeidsparticipatie en de zorgkosten. Naast het leed veroorzaakt door de symptomen zelf, rapporteren veel mensen met ALK negatieve ervaringen met zorgverleners. Zo melden mensen met ALK dat hun symptomen niet serieus worden genomen door zorgverleners en soms worden afgedaan als emotionele problemen of verzinsels. Ook rapporteren zij dat hun waarheidsgetrouwheid in het beschrijven van de symptomen in twijfel wordt getrokken. In dit proefschrift verwijzen we naar deze negatieve overtuigingen, houdingen en gedragingen als stigma.

Een belangrijk onderdeel van stigmatisering in de gezondheidszorg zijn de *persoonlijke* overtuigingen, houdingen en gedragingen van zorgverleners. Het is algemeen bekend dat veel zorgverleners de zorg voor mensen met ALK moeilijk vinden en dat er veel barrières worden ervaren bij het verlenen van zorg aan mensen met ALK. Zorgverleners melden in het bijzonder dat ze zich onzeker voelen over communicatie met mensen met ALK. Deze communicatieomvat het geven van adequate uitleg over ALK, het aanpakken van psychosociale factoren die een rol kunnen spelen bij de symptomen, of het toepassen van een meer persoonsgerichte communicatiestijl. Dit ervaren gebrek aan vertrouwen en competentie heeft negatieve gevolgen, bijvoorbeeld het vermijden van interacties met patiënten waar mogelijk of het compenseren door niet-passende diagnostische procedures en interventies te bestellen.

De afgelopen jaren is er meer aandacht gekomen voor de manier waarop zorgverleners werken met mensen met ALK. Wetenschappelijk onderzoek heeft zich voornamelijk gericht opde vele vormen en voorbeelden van stigmatisering door zorgprofessionals ten opzichte van mensen met ALK, kwalitatief onderzoek naar waarom dit gebeurt en de ontwikkeling van educatieve interventies. Er is echter weinig aandacht besteed aan het meten van stigmatisering in de context van ALK. Door een valide en betrouwbaar

meetinstrument te ontwikkelen, kunnen we een basis leggen voor de ontwikkeling en evaluatie van toekomstige stigma-interventies.

Deze thesis onderzoekt stigmatisering door zorgprofessionals ten opzichte van mensen met ALK. Dit omvat de ontwikkeling van een valide en betrouwbaar meetinstrument dat gebruikt kan worden om stigmatisering door zorgprofessionals te meten. Het doel hiervan is om een basis te leggen voor het ontwikkelen en evalueren van toekomstige stigma-interventies en om een duidelijker begrip te krijgen van stigmatisering in de context van ALK. We onderzoeken uitingen van stigma in een klinische context, de ervaring van stigma van patiënten en de kwaliteit van online gezondheidsinformatie.

Resultaten

Uit onze eerste beoordeling van stigmametingsinstrumenten (**hoofdstuk 2**) bleek dat er weliswaar veel voorbeelden waren van publicaties en instrumenten die stigmatiserende houdingen en gedragingen van zorgverleners onderzochten, maar dat er weinig aandacht werd besteed aan de ontwikkeling ervan of aan de evaluatie van de meeteigenschappen.

Veel instrumenten waren ontwikkeld voor slechts één onderzoek, meetconstructen werden zelden benoemd of beschreven en er werd onvoldoende bewijs gerapporteerd voor zowel de ontwikkeling van het instrument als de inhoudsvaliditeit. Hoewel we de blijvende aandacht voor het meten van stigma (en verwante constructen) waardevol vonden, vonden we het gebrek aan evaluatie van de meeteigenschappen problematisch. Dit was vooral omdat verschillende studies niet-gevalideerde instrumenten gebruikten om de effectiviteit van antistigma-interventies te beoordelen. Daarom was onze aanbeveling de ontwikkeling van een generiek ALK-gerelateerd stigma-instrument, met als prioriteit het vaststellen van de inhoudsvaliditeit.

We gebruikten onze lessen uit dit reviewom een nieuw meetinstrument te ontwikkelen om stigmatisering van mensen met ALK te meten, de ‘Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP)’ (**hoofdstuk 3**). De ontwikkeling van dit instrument was een iteratief proces dat bestond uit een review door het onderzoeksteam, het creëren van meetitems en cognitieve interviews met professionals in de gezondheidszorg in het Verenigd Koninkrijk. We analyseerden de relevantie, begrijpelijkheid

en volledigheid van potentiële items, inclusief de kans op sociale wenselijkheidsbias. Deze eerste versie van de PSSS-HCP bevatte 19 items, gegroepeerd over drie domeinen (stereotypen, vooroordelen, discriminatie).

De PSSS-HCP werd vervolgens getest met 121 zorgverleners in het Verenigd Koninkrijk om de factorstructuur, validiteit en betrouwbaarheid te evalueren (**hoofdstuk 4**). Er werd een drie-factorstructuur geïdentificeerd (othering, uneasiness in interaction en non-disclosure) die verantwoordelijk was voor 60.5% van de variantie met behulp van 13 van de 19 geteste items. De PSSS-HCP vertoonde voldoende interne consistentie (Cronbach's alpha = 0.84 voor de totale schaal, 0.71-0.84 voor de subschalen) en voldoende test-retest betrouwbaarheid met een intraclass correlatie = 0.97 (95% CI 0.94 tot 0.99, $p<0.001$). De convergente validiteit tussen de PSSS-HCP en de Medical Condition Regard Scale was voldoende en we vonden geen bewijs van sociale wenselijkheid bias. We concludeerden dat de PSSS-HCP gebruikt kan worden om stigmatisering van ALK door zorgverleners te meten, maar dat verdere validatie van het instrument aanbevolen is.

In **hoofdstuk 5** ontwikkelden we een Nederlandse versie van de PSSS-HCP en evalueerden we de meeteigenschappen ervan onder een steekproef van zorgverleners in Nederland. Onze resultaten toonden voldoende constructvaliditeit, betrouwbaarheid (modelgebaseerde betrouwbaarheid en test-retest betrouwbaarheid) en een minimale invloed van sociale wenselijkheidsbias. Model fits waren vergelijkbaar tussen een twee-factor gecorreleerd model en een gemodificeerd bifactor model, en bifactor analyse suggereerde dat de PSSS-HCP vooral werd beïnvloed door een algemene factor. Deze studie leverde verder bewijs dat de PSSS-HCP een geschikte meting is van stigmatiserende houdingen van zorgverleners ten opzichte van mensen met ALK.

Deze studie benadrukte de slechte behandeling van mensen met ALK. De deelnemers werden gevraagd om te vergelijken hoe mensen met ALK door zorgverleners worden behandeld in vergelijking met mensen met symptomen met een duidelijkere pathologie. De meeste deelnemers antwoordden dat mensen met ALK minder serieus worden genomen (80.2%), minder prioriteit krijgen (67.5%) en een slechtere kwaliteit van zorg ontvangen (61.2%). Deze bevinding onderstreept het belang van stigmatisering op dit gebied en het belang van het ontwikkelen en evalueren van stigma reducerende interventies.

In dit proefschrift wilden we ook onderzoeken hoe stigmatisering zich ontvouwt vanuit het perspectief van patiënten. Om dit te doen, onderzochten we de ervaringen van patiënten met een specifiek type van persisterende somatische symptomen: Functionele Neurologische Stoornis (FNS, FND in het Engels) (**hoofdstuk 6**). Terwijl bestaande literatuur aantoont dat stigma bestaat voor veel patiënten met FNS en geassocieerd wordt met een slechtere levenskwaliteit, is het minder duidelijk hoe stigma zich in deze context ontwikkeld. Wij interviewden patiënten met een recente diagnose van FNS in het Verenigd Koninkrijk om ervaringen met stigma te onderzoeken vanaf het begin van de symptomen, tot aan de diagnose en daarna. Wij ontdekten dat stigma zich niet ontwikkelt als een lineair proces. In plaats daarvan vonden wij een proces met meerdere, op elkaar inwerkende factoren: door de symptoomervaring, door 'othering' door het gezondheidszorgsysteem, door alledaagse interacties en vanuit de *zelf*. Hierbinnen was een centraal thema van kennis te vinden, die het stigma zowel aanwakkerde als tegenwerkte. Het erkennen van de ervaring van de patiënt kwam naar voren als een thema dat het stigma verlichtte. Specifieke adviezen voor professionals in de gezondheidszorg waren onder andere: het vermelden van FNS als een mogelijkheid in de differentiaaldiagnose, het opnemen van onderwijs over FNS in het kerncurriculum voor professionals in de gezondheidszorg, het serieus nemen van de ervaring van de patiënt en het gevoelig zijn voor de mogelijke oorsprong van zelfstigma.

Tot slot onderzochten we de kwaliteit en de stigmatiserende inhoud van gezondheidsinformatie in Wikipedia artikelen (**hoofdstuk 7**). Hoewel het internet een steeds populairdere plek is voor mensen om toegang te krijgen tot gezondheidsinformatie, verschilt de kwaliteit van de informatie aanzienlijk tussen informatiebronnen en medische vakgebieden. Lage kwaliteit of onnauwkeurige gezondheidsinformatie kan patiënten misleiden en bijdragen aan onrealistische verwachtingen of slechte besluitvorming. Door middel van een analyse van verschillende diagnosecategorieën van ALK in 14 talen ontdekten we dat de kwaliteit van de artikelen zeer inconsistent was en veel voorbeelden van stigmatiserende inhoud bevatte. We vonden ook voorbeelden van pogingen om stigmatisering tegen te gaan. Dit gebeurde op twee manieren: door een stereotype direct te weerleggen of door te verwijzen naar de diagnosecriteria van andere aandoeningen. Op basis hiervan gaven wij praktische aanbevelingen voor het verminderen van stigmatiserende online gezondheidsinformatie en een prototype dat gebruikt kan worden voor toekomstige communicatie.

Implicaties voor praktijk en onderzoek

Op basis van deze thesis worden de volgende onderdelen geidentificeerd die essentieel zijn voor het verminderen van stigmatisering van mensen met ALK.

- Interventies moeten een beter begrip en toepassing van het biopsycho-sociale model van de geneeskunde aanmoedigen. Waar dualismen tussen lichaam en geest bestaan, is het belangrijk dat de interventies zorgverleners aanmoedigen om rekening te houden met alle relevante biologische, psychologische en sociale factoren die van invloed kunnen zijn op het voortduren van symptomen.
- Het ontwikkelen van vaardigheden in persoonlijke communicatie is essentieel voor zorgverleners van mensen met ALK. Door tijd te nemen voor communicatie en uitlegmaakt het mogelijk om te laten zien dat de zorgverlener de persoon met ALK serieus neemt, om te bespreken hoe de symptomen ontstaan en om de kans op verbetering of omkeerbaarheid te benadrukken. Het opbouwen van een sterke relatie moet gezien worden als belangrijk en zinvol werk dat een tastbare impact kan hebben op mensen met ALK.
- Reflectie is een belangrijk onderdeel van interventies die zorgverleners kunnen helpen inzicht te krijgen in hun eigen houding en gedrag. Dit kan worden gedaan door de mate van stigmatisering van deelnemers direct te beoordelen, wat op zichzelf een educatief resultaat zou kunnen zijn. Aangezien negatieve emoties zo'n belangrijke rol spelen in het stigmatiseringsproces, kan reflectie een mechanisme bieden om moeilijke ontmoetingen te bespreken en te leren van collega's.
- Interventies moeten zinvol contact met mensen met ALK bevatten. Voorbeelden van contact kunnen zijn: het faciliteren van de interventie door mensen met ALK, het deelnemen aan rollenspellen van klinische consulten, of - meer indirect – het bekijken van video's met ervaringsverhalen. Er zijn echter aanwijzingen dat actievere vormen van contact effectiever zijn dan passieve vormen. Er zijn ook andere voorwaarden voor effectief contact. De interventie moet gericht zijn op specifieke zorgverleners, lokaal zijn (bijvoorbeeld binnen de regionale context), mensen met ervaring moeten 'geloofwaardig' overkomen (dat wil zeggen, bezig zijn met het managen van ALK of in herstel zijn), en continu zijn.
- Het is belangrijk dat toekomstige stigma-interventies zo ontworpen worden dat hun effect op de juiste manier geëvalueerd kan worden. Dit omvat een duidelijke conceptualisatie van stigma, die gekoppeld is aan de rol van de

zorgprofessional. De interventie moet de mechanismen bespreken waarin de gewenste verandering moet plaatsvinden en een meetinstrument gebruiken met geschikte inhoudsvaliditeit voor de doelpopulatie. We hebben de PSSS-HCP zo ontwikkeld dat deze gebruikt kan worden als een dergelijk uitkomstmeetinstrument.

Er zijn vijf belangrijke onderzoeksgebieden die verder kunnen gaan op basis van het hier gepresenteerde onderzoek, waaronder: 1) verdere evaluatie van de meeteigenschappen van de PSSS-HCP; 2) vergelijking van verschillende methoden om stigmatisering door zorgprofessionals te meten; 3) voorspelling van welke typen zorgprofessionals en welke kenmerken van zorgprofessionals het meest stigmatiserend zijn tegenover mensen met ALK; 4) onderzoek naar intersectionaliteit en ALK-gerelateerd stigma; en 5) kwalitatief onderzoek naar hoe zorgprofessionals hun eigen vooroordelen herkennen en erop reageren.

Conclusie

Hoewel er de laatste jaren meer aandacht is voor stigmatisering in de context van ALK, is er weinig aandacht besteed aan de kwaliteit van de meting ervan. In dit proefschrift is een meetinstrument ontwikkeld en gevalideerd dat gebruikt kan worden om stigmatisering van mensen met ALK door zorgverleners te meten, de PSSS-HCP. We hebben uitingen van stigmatisering in een klinische context onderzocht, de ervaringen met stigmatisering van patiënten en de kwaliteit en stigmatiserende inhoud van online gezondheidsinformatie beoordeeld. Er is een groot potentieel voor interventies om de kennis, de houding en het gedrag van zorgverleners ten opzichte van mensen met ALK te verbeteren. Hoewel het niet de enige bron van stigmatisering is voor mensen met ALK, kan het verminderen van stigmatisering door zorgverleners een grote en blijvende invloed hebben op de gezondheidsresultaten. We hopen dat dit proefschrift een basis biedt voor toekomstige stigmareducerende interventies en evaluaties.

Appendices

Research data management
PhD Portfolio
Publications list
Acknowledgements / Dankwoord

Research data management

Ethics and privacy

This thesis is based on the results of medical-scientific research with human participants. The studies described in **Chapter 3, 4, 5 and 6** were conducted in accordance with the ICH-GCP guidelines (Good Clinical Practice). No studies were subject to the Medical Research Involving Human Subjects Act (WMO).

The studies in **Chapter 3 and 4** were approved by the University of Edinburgh and the South Central - Oxford C Research Ethics Committee (22/SC/0473). The study in **Chapter 5** was reviewed by the medical ethical review committee 'METC Oost-Nederland' and was viewed not to be subject to the Medical Research Involving Human Subjects Act (WMO) (registration number: 2024-17028). The study in **Chapter 6** was approved by the University of Edinburgh and South- Central Hampshire A Research Ethics Committee (reference 21/ SC/ 0418).

Informed consent was obtained from participants to collect and process their data for this research project. Consent was also obtained for sharing the (pseudonymized) data after research. Technical and organisational measures were followed to safeguard the availability, integrity and confidentiality of the data (these measures include the use of independent monitoring, pseudonymisation, access authorisation and secure data storage).

Data collection and storage

Data for **Chapter 3** were collected through interview recordings and transcribed verbatim. Interview transcripts were imported into MaxQDA (VERBI Software, Berlin, Germany). Data for **Chapter 4** were collected through an online survey using Qualtrics XLM. From Qualtrics XLM data were exported to SPSS (SPSS Inc., Chicago, Illinois, USA). Data for chapter for **Chapter 5** were collected through an online survey using LimeSurvey. From LimeSurvey data were exported to SPSS (SPSS Inc., Chicago, Illinois, USA). Data for **Chapter 6** were collected through interview recordings and transcribed verbatim. Interview transcripts were imported into MaxQDA (VERBI Software, Berlin, Germany).

Pseudonymised data were stored and analysed in workspace (dws-1736-PSSSHCP) in the Azure DRE (**DRE Portal**). The data were only accessible by project members.

Availability of data

All studies are published open access. Data were made reusable by adding sufficient documentation (research protocol, codebook), by using preferred and sustainable data formats and by publishing under the CC.BY.4.0 license. The datasets from Chapters **3**, **4**, **5** and **6** were published with restricted access. Requests for access will be checked against the conditions for sharing the data as described in the signed Informed Consent. The data will be archived for 15 years after termination of the study.

PhD Portfolio

Training	Year
RIHS Introduction course	2021
ETUDE training week 1: introduction to functional disorders and persistent somatic symptoms (online)	2021
Dutch language B1-B2, (Radboud In'to languages)	2021
Dutch language B2-C1 (Radboud In'to languages)	2022
Basic course for clinical investigators (BROK)	2022
Clinimetrics: Assessing Measurement Properties of Health Measurement Instruments (WV40)	2022
ETUDE training week 2: diagnosis	2022
ETUDE training week 3 mechanisms	2023
Facilitation and interactive workshop methods: Sessiemakers	2023
Statistics for PhD candidates using SPSS	2023
Project management for PhD candidates	2023
ETUDE training week 4: stigma	2023
Research integrity	2024
ETUDE training week 5: treatment	2024

Symposia and congresses	Year
Medical Education on Medically Unexplained Symptoms and Intercultural Communication, Netherlands: participant	2021
European Association of Psychosomatic Medicine conference, Austria: poster presentation	2022
European Association of Psychosomatic Medicine conference, Poland: oral and poster presentation	2023
Network for Persistent Somatic Symptoms (NALK), Netherlands: oral presentation	2023
ETUDE summerschool, Switzerland: oral presentation, workshop facilitator and organising committee	2024
European Association of Psychosomatic Medicine conference, Switzerland: oral presentation	2024

Lectures	Year
Invited lecture UKE Hamburg, Germany	2021
Invited lecture NHS Lothian, United Kingdom	2023
Invited lecture University of Hull, United Kingdom	2023

Secondments and internships	Year
UKE Hamburg, Germany	2021-2022
Stichting De Bagagedrager, Netherlands	2023
WMU Wroclaw, Poland	2023

Teaching activities	Year
Supervising medical students Radboudumc	2022-2024

Publications list

Kustra-Mulder A, McGhie-Fraser B, Petzke T, Fila-Pawłowska K, Rosmalen J, Cosci F, Löwe B, Weigel, A. Healthcare Professionals' Views on Healthcare-Related Factors Influencing Symptom Course in Persistent Somatic Symptoms: A Qualitative Study of Four European Countries. Submitted, 2024.

McGhie-Fraser B, Lucassen P, Ballering A, Abma I, Brouwers E, Van Dulmen S, olde Hartman T. Persistent somatic symptom related stigmatisation by healthcare professionals: A systematic review of questionnaire measurement instruments. Journal of Psychosomatic Research. 2023:111161.

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McGhie-Fraser B, McLoughlin C, Lucassen P, Ballering A, van Dulmen S, Brouwers E, Stone J, olde Hartman T. Measuring persistent somatic symptom related stigmatisation: Development of the Persistent Somatic Symptom Stigma scale for Healthcare Professionals (PSSS-HCP). Journal of Psychosomatic Research. 2024:111689.

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McGhie-Fraser B, Lucassen P, Ballering A, van der Linden L, Brouwers E, olde Hartman T, van Dulmen S. Validation of the Dutch version of the Persistent Somatic Symptoms Stigma Scale for Healthcare Professionals (PSSS-HCP). Submitted, 2024.

McLoughlin C, McGhie-Fraser B, Carson A, olde Hartman T, Stone J. How stigma unfolds for patients with Functional Neurological Disorder. Journal of Psychosomatic Research. 2024:111667.

Treufeldt H, Burton C, McGhie-Fraser B. Stigmatisation in clinical consultations for persistent physical symptoms/functional disorders: A best fit framework synthesis. Journal of Psychosomatic Research. 2024:111828.

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