



Bodily distress syndrome:

Concerns about scientific credibility in research and implementation

Diane O'Leary^{1, 2, *}

¹ Rotman Institute of Philosophy, Western University, London, Ontario, Canada

² Fondation Brocher, Hermance, Switzerland

The credibility of psychosomatic medicine has recently been called into question through challenges to the scientific integrity of the “PACE trial”, which claimed success for psychiatric treatment in managing myalgic encephalomyelitis/chronic fatigue syndrome. The newest product of research in psychosomatic medicine is “bodily distress syndrome” (BDS), a Danish diagnostic construct developed to replace “somatoform disorders”, “medically unexplained symptoms” and “functional somatic syndromes”. I set out in this paper to examine the science that supports the construct of BDS, both in design and in implementation. Following the Introduction, in §2 I clarify the details that define BDS and the problems the construct is designed to resolve. In §3 I explore three problems with the science behind BDS. In §4 I consider the World Health Organization (WHO)’s effort to implement BDS in the International Classification of Diseases (ICD), noting that while BDS criteria fail in both WHO studies, the workgroup nonetheless insists that the ICD should recommend them for global use. I conclude that BDS gives support to recent concern that scientific standards in psychosomatic medicine are inadequate, closing with a brief discussion of ethical problems that arise when global health policy decisions are not grounded in science.

Keywords: evidence-based medicine, International Classification of Diseases, myalgic encephalomyelitis/chronic fatigue syndrome, psychosomatic medicine, scientific integrity

1. INTRODUCTION: THE CREDIBILITY OF PSYCHOSOMATIC MEDICINE

In recent years, the field of psychosomatic medicine has begun to make its way out of the dark corners of medical practice and into the public light of day. As a small subdiscipline of psychiatry, the scientific credibility of the field was secured in recent years by a large-scale research study, “Pacing, graded activity, and cognitive behaviour therapy; a randomised evaluation” [1], commonly known as the “PACE trial”. A UK government-funded study, the PACE trial established in 2011 that professional debate over the biological reality of myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) could be put to rest. Having followed a large group of patients over five years, the study showed that the psychiatric approach succeeds in resolving physical symptoms for the quintessential contested condition.

Beginning in 2014, however, scientists [2], journalists [3, 4] and government health authorities [5, 6] joined the patient community in questioning the science behind PACE’s conclusions. Following the conclusion of the US Academy of Medicine in 2015 that ME/CFS is not psychosomatic [7], the US Agency for Healthcare Research and Quality published a report in 2016 offering an extensive list of scientific failings in the PACE trial’s

design, including three distinct forms of bias [8]. Several peer-reviewed articles have now examined the original PACE data [9, 10] and concluded that, although PACE dictated management of ME/CFS across the globe for many years, the study fails to meet basic standards of scientific methodology.

A challenge to the scientific credibility of the PACE trial can only be viewed as a challenge to the credibility of the field of psychosomatic medicine. No matter which side of the PACE debate we might prefer, there has been enough evidence to call into question the scientific standards of PACE researchers, those who supported the study both scientifically and in terms of funding, and those who allowed the trial to pass through the peer review process. Indeed, it is difficult to imagine how such a large-scale investigation could have developed, proceeded and passed through the review process unless its scientific failings were actually characteristic of its field. For these reasons, the PACE controversy suggests a need to evaluate the scientific credibility of psychosomatic medicine generally.

As a first step toward that larger goal, this paper considers the science behind “bodily distress syndrome” (BDS), the newest product of research in psychosomatic medicine [11]. BDS is a Danish diagnostic construct, one developed to replace “somatoform disorders” in a way that would also capture patients with “medically unexplained symptoms” and “functional somatic

* E-mail: doleary8@uwo.ca

syndromes” [12, 13]. The category of “somatoform disorders” has directed diagnosis and management of psychosomatic conditions across the globe since the 1980s, and BDS failed to replace this category in the DSM-5¹ in 2013. Studies suggest [14, 15], however, that because of its ability to unify all three diagnostic categories, BDS will have a substantial rôle to play in the new edition of the global diagnostic coding manual, the International Classification of Diseases, ICD-11, which is nearing publication at the present time.

Following this Introduction, in §2 I clarify the details that define BDS, and the problems the construct is designed to resolve. In §3 I shall explore three problems with the science behind BDS, suggesting that researchers misrepresent neuroscientific support for the construct, that research fails to establish the construct’s scientific success at the task for which it was designed and, most importantly, that research ignores the evidence-based requirement to successfully exclude patients with similar but distinct conditions; that is, patients who suffer from medical conditions that require medical care. In §4 I consider the efforts of the World Health Organization (WHO) to implement BDS in the International Classification of Diseases, noting that while BDS criteria fail in both WHO studies, the workgroup nonetheless insists that the ICD should recommend them as a tool for determining when physical symptoms can safely be diagnosed and managed in psychiatry. I conclude in §5 that BDS gives support to recent concern that scientific standards in psychosomatic medicine are inadequate, offering a brief discussion of ethical problems that arise when global health policy decisions are not grounded in science.

2. WHAT IS BODILY DISTRESS SYNDROME?

“Bodily distress syndrome” is a diagnostic construct that gathers together the disparate areas of medicine and psychiatry where bodily symptoms are understood to be caused by the mind (or perhaps by central neurological pathologies that masquerade as peripheral bodily pathologies) [16]. To understand the meaning and value of the construct as a diagnostic tool, it is important to take stock of the three core concepts it claims to unify: the very broad category of medically unexplained symptoms; the smaller category of “contested conditions” (known within psychiatry as “functional somatic syndromes”); and the overlapping category of mental health disorders known as “somatoform disorders” (Fig. 1). Professionals in psychosomatic medicine generally presently agree that all three categories are in need of substantial revision.

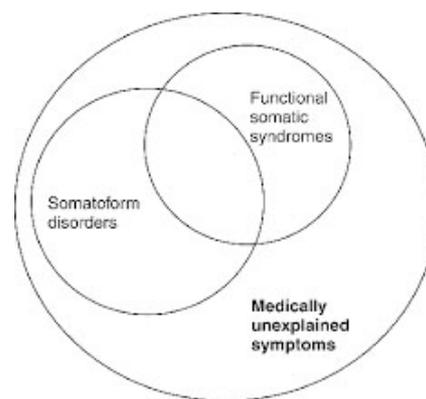


Figure 1. Medically unexplained symptoms [17].

2.1 Medically unexplained symptoms

Most of us have unthinkingly adopted a model of medicine where patients present their symptoms to physicians, who investigate and reach a diagnosis, which then directs the course of treatment. In reality, however, the diagnostic process very often fails. While prevalence estimates for medically unexplained symptoms (MUS) vary widely [18, 19], both the “Up to Date” review system in the US [20] and the Joint Commissioning Panel on Mental Health in the UK [21] settle in the middle range of estimates, suggesting that 52% of symptoms are unexplained in outpatient settings. According to the lead researcher for the BDS construct, Danish psychiatrist Per Fink, “it is ... more the exception than the rule that a physical symptom can be explained by a known physical disease” [22].

For reasons that are far too complex to examine here, “medically unexplained symptoms” are understood and classified as psychiatric conditions rather than medical conditions. That is to say, while most non-medical, non-psychiatric professionals would understand the phrase “medically unexplained symptoms” to label symptoms for which no diagnosis has been determined, in practice the term is understood as a diagnostic answer that shifts the orientation of care from medicine to psychiatry [23]. Research and practice directives for MUS often focus on primary care, because early diagnosis of MUS can protect patients from the iatrogenic harms of unnecessary testing and trial treatments [24, 25].

Three sorts of concerns have supported professional consensus during the last fifteen years that change is needed in the area of medicine that addresses MUS. First, the diagnostic construct of “medically unexplained symptoms” has substantial theoretical problems, ambiguous as it surely is [12]. Second, patients typically feel resentful when they are told they suffer from MUS; this triggers a

¹ The American Psychiatric Association’s *Diagnostic and statistical manual of mental disorders: DSM-5*TM (5th ed.).

very common, deeply intractable form of conflict between patients and doctors [26]. It is useful to note that this problem has persisted since Freud. While core ideas about mental causes for symptoms have not substantially changed over the last hundred and fifty years, names for those ideas have frequently shifted to avoid triggering resentment in patients who have become familiar with the current labels [27, 28]. Third, and perhaps most importantly, MUS consume an unusually large portion of health resources, hence national health systems, insurers and policymakers have given considerable attention to changes in this area that could substantially reduce expenditure [24, 29, 30].

2.2 Contested conditions or “functional somatic syndromes”

For the most part, the general public is aware that “chronic fatigue syndrome”, or ME/CFS, is a “contested condition”—that is, one that has stirred vigorous debate about biological versus psychiatric causes. Alongside ME/CFS, fibromyalgia is generally recognized as contested, as is the condition known as “multiple chemical sensitivity” (MCS). In the professional context of medicine and psychiatry, this group is also understood to include a number of conditions that the public typically does not recognize to be contested, such as tension headache, premenstrual syndrome (PMS), irritable bowel syndrome (IBS), and chronic pain in the absence of abnormality in diagnostic imaging [22, 30, 31, 35]. All of these conditions share a lack of consensus about biological aetiology. Though many contested conditions have been substantially explored in biological research that claims to support a biological disease model, research of this kind has been unconvincing to leading professionals in psychosomatic medicine.

Within psychiatry, conditions in this group are called “functional somatic syndromes” and they are generally understood as consistent, organized groupings of medically unexplained symptoms. In 1999, leading researchers in the field began to suggest that [31]

... the existence of specific somatic syndromes is largely an artefact of medical specialization. That is to say that the differentiation of specific functional syndromes reflects the tendency of specialists to focus on only those symptoms pertinent to their specialty, rather than any real differences between the patients.

What these researchers propose is that distinct contested conditions are actually “an artefact of medical specialization” [31]. This hypothesis has been attentively explored and defended in a substantial list of publications since 1999 based on similarities in symptoms, diagnostic

definitions, recommended treatments and patient characteristics, such as “difficult” relationships with doctors and a predominantly female population of patients receiving diagnoses of this kind. Some biological research has noted similarities in biological pathophysiology for some of these conditions. Finally, neurobiological research has often been construed to support common neuropathological underpinnings for this collection of conditions [16, 22] (a suggestion I shall challenge in §3).

With a foundational empirical study in 2010, Per Fink and his colleague Andreas Schröder showed that diagnostic criteria for BDS will effectively “capture” six functional somatic syndromes under a single diagnostic rubric in mental health: ME/CFS, fibromyalgia, irritable bowel syndrome, non-cardiac chest pain, hyperventilation syndrome, and chronic unexplained pain [13]. In doing so, they have shown that BDS can address the three core problems that have generally driven change in this area. First, BDS resolves a theoretical problem (the problem of disparate medical diagnoses for a single, unified mental health condition); and second, it addresses the conflict that so often arises when patients are told they suffer from “functional somatic syndromes”, a term now often recognized as a psychiatric diagnosis [14]; and third, perhaps most importantly, BDS criteria greatly reduce the costs of continued medical investigation and trial treatments for contested conditions [24]. This gives insurers and national health systems strong reasons to support implementation of the construct.

2.3 Somatoform disorders

When patients have medically unexplained symptoms that are sufficiently varied and sufficiently persistent, they meet criteria for diagnosis of a full-fledged mental health disorder. Since the 1980s that diagnosis has been, primarily, “somatization disorder”, which is central to the “somatoform disorders” category in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) and the International Classification of Diseases (ICD). “Somatoform disorders”, it is useful to note, did not substantially alter the original Freudian construct of “hysteria” when they were first included in the third edition of the DSM. Rather, it was renamed in an effort to avoid the cycle of conflict that arose when patients were told their symptoms were caused by hysteria [27, 28].

As with MUS and functional somatic syndromes, in the last fifteen years a professional consensus has gathered around the idea that somatization disorder, and the larger category of somatoform disorders, have three sorts of problems that can only be addressed with substantial revisions. First, somatization disorder has the irresolvable theoretical challenge of defining a psychiatric

disorder purely on the basis of physical symptoms that lack medical explanation [32]. Second, patients are nowadays largely familiar with the concept of somatization, recognizing it, resentfully, as a mental health diagnosis for what they understand to be medical problems [26]. For this reason, physicians have long been reluctant to make use of the category of somatoform disorders in everyday medical practice [33]. Third, this reluctance creates budgetary problems, as physicians choose instead to pursue continued medical investigation and trial treatments [24].

On these grounds it is generally agreed that the core construct of somatoform disorders is no longer tenable. In 2013 the DSM-5 replaced it with “somatic symptom disorder” (despite the strongly expressed objections of some psychiatric professionals). At present the World Health Organization is finalizing its decisions on the constructs that could replace somatoform disorders in the eleventh edition of the International Classification of Diseases (ICD-11). Some recent papers suggest that BDS will have a substantial rôle to play [14, 15].

2.4 How BDS criteria combine these three groupings

The core idea of BDS is that “one single diagnosis” can cover the whole range of medically unexplained symptoms, functional somatic syndromes and most of the somatoform disorders.² This is a difficult challenge given the highly disparate set of symptoms and conditions that fall under these headings. The construct accomplishes the task with diagnostic criteria that specify a very wide range of symptoms within four distinct “symptom clusters” [11, 13, 34]:

1. Cardio-pulmonary: palpitations/heart pounding, precordial discomfort, breathlessness without exertion, hyperventilation, hot or cold sweats, trembling or shaking, dry mouth, churning in stomach, flushing or blushing;

2. Gastro-intestinal: abdominal pains, frequent loose bowel movements, feeling bloated/full of gas/distended, regurgitations, constipation, diarrhoea, nausea, vomiting, burning sensation in chest;

3. Musculo-skeletal: pains in arms or legs, muscular aches or pains, pains in the joints, feelings of paresis or localized weakness, back ache, pain moving from one place to another, unpleasant numbness or tingling sensations;

4. General symptoms: concentration difficulties, impairment of memory, excessive fatigue, headache, dizziness.

For diagnosis of “modest, single-organ BDS” a patient must have three or more symptoms in any one of the

symptom groups, hence single-organ BDS is specified as one of the four subtypes. By contrast, “severe, multi-organ BDS” requires three or more symptoms from three or four of these symptom groups [11, 13, 34].

Fink and Schröder’s 2010 empirical study showed that BDS “succeeded in capturing 10 diagnostic categories of functional somatic syndromes and somatoform disorders”, leading to the conclusion that it can “replace numerous overlapping diagnostic labels and... reduce the diagnostic confusion that currently prevails in the field” [13]. In the context of primary care, researchers have often commented on the benefits of replacing “medically unexplained symptoms” with BDS [12, 17, 35], though given the ambiguity of that original construct, it is more difficult to establish the success of BDS in this context.

There is no question that if BDS did replace MUS, contested conditions and somatoform disorders, it would improve all three problems that have driven change in this area. First, BDS eliminates most of the theoretical challenges that have concerned psychiatric researchers when it comes to medically unexplained symptoms, functional somatic syndromes and somatoform disorders. Second, BDS is a new and unfamiliar diagnostic label that does seem likely to alleviate patient resentment and examination room conflict. The construct carries well-developed manuals and guidelines with a heavy focus on establishing trust with patients heading into psychiatric care [22]. Third, and perhaps most importantly, symptoms in these groups “form one of the most expensive categories of healthcare expenditure”, and many researchers in psychosomatic medicine have supported the idea of “shifting some of this expenditure away from numerous investigations for organic disease and toward effective treatment of bodily distress” [30]. It is clear that implementing BDS to replace these three categories would be a highly effective cost-saving measure for national health systems and insurers [24, 36, 37].

In §3, I will consider the success of the BDS construct more broadly, stepping away from the concerns of professionals in psychosomatic medicine towards universal standards for scientific accuracy, scientific method and evidence-based medicine.

3. THREE CHALLENGES FOR THE SCIENCE BEHIND BDS

3.1 Scientific accuracy: Misrepresentation of neuroscience

Within the context of psychosomatic medicine, BDS is strongly recommended by its ability to address the three

² It is important to note that BDS does not include the somatoform disorder of hypochondriasis. Instead, the BDS research team has reimagined hypochondriasis as “health anxiety”, which it understands to be distinct from the actual experience of symptoms in bodily distress syndrome [15].

core problems that have motivated change to diagnostic structures in this area over the last fifteen years. It is important to also acknowledge, however, that within its field the construct is additionally recommended by a longstanding sensibility that functional somatic syndromes should be understood as “central sensitivity syndromes” (CSS) [38–40].

With a presentation late in 2017, lead BDS researcher Per Fink offered a helpful overview of BDS, its very current definitions, goals and explanatory models [41]. During that presentation Fink presented a diagram that pictures the whole range of contested conditions circling a centre point labeled “central sensitivity”. He offered this explanation:

Central sensitization can be defined as an amplification of neural signaling within the CNS. When the response is prolonged, central sensitization becomes a pathological state characterized by a dysfunctional response to different and normally non-noxious stimuli that can manifest itself as pain hypersensitivity.

This kind of suggestion is very common in BDS research [22, 35], as it has been in psychosomatic medicine for decades. As with this very recent Fink presentation, and a similarly recent article by a colleague of Fink's, Marianne Rosendal [35], in the context of BDS, discussion of CSS typically implies a consensus within neuroscience about a single, unified central nervous system disorder where benign bodily sensations become amplified through a cycle involving psychological distress. Fink does acknowledge in the book *Functional Disorders and Medically Unexplained Symptoms* that “the results of neuroimaging ... and pathophysiological studies have been disparate and sometimes in direct conflict with each other” [22], but in reality neuroscience presents a substantial problem for the central sensitization model in psychiatry. On two different grounds, recent neuroscientific studies challenge the idea that MUS and functional somatic syndromes can be explained by a single “central sensitivity syndrome” where psychological distress leads to amplification of benign stimuli.

The first is that, while for some conditions the “amplification of benign stimuli” model seems to be accurate, some contested conditions have been discovered to involve stimuli that are not benign, or “non-noxious” [42]. Studies have suggested, for example, that patients with fibromyalgia do not amplify a benign pain experience. Rather, their pain experience, neurologically speaking, is identical to that of healthy patients whose pain stimuli have been purposefully amplified twofold [43, 44]. To the extent that this kind of model has been supported for other contested conditions [45–47], it is

inaccurate to characterize conditions of this kind as, in the words of Professor Fink, “a dysfunctional response to different and normally non-noxious stimuli” or as “pain hypersensitivity” [41]. As conditions of this kind involve a healthy psychological response to a genuine experience of pain or illness, it is difficult to see any sense in which these could accurately be characterized as mental health conditions.

The second and perhaps more important ground is that in their extensive 2016 review of neuroimaging of central sensitivity syndromes, Wallit et al. emphatically conclude that “a coherent picture of a ‘central sensitization’ mechanism that bridges across all of these syndromes does not emerge from the existing scientific evidence” [42]:

... the concept that “central sensitivity syndromes” are biologically-related entities is not strongly supported by the sum of the neuroimaging evidence. Some passing similarities are noted, but are far outweighed by heterogeneity and inconsistency when results are compared between disorders [42].

Before neuroscience developed to evaluate the idea, professionals in psychosomatic medicine theorized that MUS and contested conditions could ultimately be unified as a single neurobiological malady, a “central sensitivity syndrome” that involves benign bodily symptoms amplified by psychological distress. Now that neuroscience has been able to explore that idea, however, we find it to be unsupported. While the notion of CSS in psychosomatic medicine does strongly invite a diagnostic construct exactly like BDS, neurological imaging studies have failed to support the theory that contested conditions can be unified by a single pathology, a single “sensitivity syndrome”, in the central nervous system.

3.2 Scientific method: Failure to support the construct's success for its purpose

The scientific success of any hypothesis depends upon clarity about the question the hypothesis is proposed to answer. Similarly, the scientific success of any conceptual construct depends upon clarity about the purpose the construct is intended to serve, and proof that the construct does successfully accomplish that goal. On this very rudimentary scientific level, the construct of bodily distress syndrome is unsupported.

Whatever else we might say about the diagnostic categories of MUS and somatoform disorders, it is clear how practising medical providers understand these categories and put them to use:

In the front line of the health care system ... interpretation and categorisation of symptoms

are inherent parts of daily clinical practice. For the GP, a diagnostic category serves as a decision node or a working diagnosis on which treatment, further investigations and conclusions on the absence of serious disease are based [48].

As a replacement for MUS and somatoform disorders, physicians would primarily make use of BDS as a tool for distinguishing bodily symptoms best managed along a mental health track from those that require further biologically-oriented testing and treatment. It is in this sense that BDS would “serve as a decision node...on which conclusions on the absence of serious disease are based”. Does research provide scientific support for the success of BDS at this task? Is there sufficient evidence to show that criteria for BDS do safely and reliably distinguish symptoms of disease from those best classified and managed in mental health?

The most salient fact to note about BDS research is that it simply fails to answer this question. The central 2010 study begins with a paragraph that reminds us of the range of functional somatic syndromes, then it presents a list of references that explore similarities among those syndromes [13]. After that opening paragraph the study accepts that functional somatic syndromes are a single condition best diagnosed and managed in mental health, and the question is set aside. Similarly, in the book *Functional Disorders and Medically Unexplained Symptoms*, Fink and colleagues begin by listing a range of functional syndromes within thirteen medical specialties, and then they assert, still within the first page, that “the syndromes in Table 1.1 are best understood as an expression of bodily stress and not as classical physical diseases” [22]. Like the study, the book then proceeds to explore “subtypes” of bodily distress, tools for management, and ties with historical conceptions of psychosomatic conditions. At no point does the book offer its own research to support the conclusion that conditions in this group, individually or collectively, are correctly diagnosed and managed within mental health rather than medicine.

The categorization of BDS would lead physicians to conclude that a condition meeting its criteria can safely be diagnosed and managed within mental health rather than medicine – while BDS research can support only this far less useful conclusion: that the condition is correctly grouped as a contested condition; that is, that the condition belongs within the group often theorized to be unified by a single underlying mental health pathology. To determine whether those theories are correct for any given patient, a physician would need to undertake an assessment of the research that supports psychiatric

diagnosis for the patient’s condition and the research that disputes it. There is nothing in BDS research that should give physicians grounds for trusting that evaluations of this kind are unnecessary.

3.3 Evidence-based medicine: Failure to establish the construct’s success at excluding disease

Basic tenets of scientific method suggest problems with BDS in terms of its misrepresentation of neuroscience, and its failure to verify the success of the construct with respect to the purpose for which it was designed. By far the most important shortcoming of BDS research, however, has to do with the fundamental requirements of evidence-based medicine. To adequately support the basic BDS hypothesis that medically unexplained symptoms, functional somatic syndromes and somatoform disorders arise from a single condition best classified and managed in mental health, evidence-based practice requires two distinct forms of caution: It requires that criteria for diagnosing BDS successfully capture all of the patients who suffer from the condition—and it also requires that criteria successfully exclude patients who suffer from similar conditions that are actually distinct. No diagnostic construct can be said to be evidence-based if research fails to show not only that its criteria cast a sufficiently wide net, but also that they do not cast the net too widely, mistakenly capturing patients who suffer from similar conditions.

When it comes to replacing MUS in primary care, evidence-based support for BDS would have to show that when physicians apply BDS criteria, they safely and reliably distinguish symptoms of disease from symptoms best classified and managed in mental health. Studies of this kind would not be difficult to develop or implement in a way that does not induce unusual caution in physicians’ decision-making processes [49]. There is strong evidence, moreover, that physicians feel studies of this kind are necessary. In the WHO study that first examined the success of BDS diagnostic strategies in primary care outside of Denmark, physicians in three countries independently expressed concern that “the variety of symptoms was so extensive that almost any patient could be labeled as such”, that “a positive diagnosis ... might make it more likely that a significant organic pathology would be missed”, and that BDS criteria “might lead to missing underlying/more severe illnesses” [14]. No research since that time has addressed these three concerns.

When it comes to functional somatic syndromes, evidence-based support for BDS would have to establish not only that criteria capture the functional somatic syndromes, but also that functional somatic syndromes “are best understood as an expression of bodily stress

and not as classical physical diseases" [22]. To accomplish that, evidence-based support would have to (1) evaluate available evidence for biological medical diagnosis and management for every functional somatic syndrome, (2) compare that evidence to research in psychosomatic medicine supporting unified mental health diagnosis and management, and (3) offer convincing evidence to show that the mental health approach is better defended than the approach of biological medicine.

There are strong reasons to expect that BDS researchers—and indeed any researchers in psychiatry—would fail to be successful with research of this kind. The “Center for Functional GI and Motility Disorders” at the University of North Carolina, for example, straightforwardly asserts that “functional gastrointestinal disorders [such as IBS] are ... not psychiatric disorders [50]. Similarly, in its diagnostic criteria for the various forms of headache, the International Headache Society (IHS) states that “evidence supporting psychiatric causes of headache remains scarce”, so scarce that the IHS recommends psychiatric diagnosis for headache only with the far more stringent criteria that independent psychiatric disorders have been previously diagnosed, and proven to play an immediate causal rôle in headache development [51]. Similar claims are abundant in medical research into most of the functional somatic syndromes [52–54], and it is difficult to imagine how psychiatric specialists could ever succeed in disputing them if they did set out to do so. Conceptually speaking, psychiatrists lack the expertise to refute the conclusions of biological medical researchers, or those of biological medical specialists who evaluate research within their own specialties.

By far the most obvious challenge along these lines arises from recent US reversal of opinion on ME/CFS. BDS criteria have been specifically designed to ensure that they capture ME/CFS as a mental health condition, and the 2010 study by Fink and Schröder established that they do succeed at that task [13]. In 2014, however, the US National Institutes of Health concluded that “although psychological repercussions (i.e., depression) may accompany ME/CFS, it is not a primary psychological disease in etiology” [5]. Moreover, the US Department of Health and Human Services now recommends that guidance for managing ME/CFS should include “a clear indication that the disease is not a psychiatric or somatoform disorder” [6]. Finally, the US National Academy of Medicine did proceed through a three-step, evidence-based evaluation of the comparative merits of biological and psychiatric approaches to diagnosis and management of ME/CFS; in 2015 they unequivocally concluded that “ME/CFS is a serious, chronic, complex systemic disease” [7].

Because the US conclusion establishes a strong possibility that patients with ME/CFS suffer from a biological disease that requires biological medical care, it also establishes an unacceptable level of risk with any diagnostic construct that would systematically channel care for ME/CFS down the mental health track. In this way, ME/CFS is an important cautionary tale. Regardless of which side we might favour in the ME/CFS debate, the very existence of the debate establishes that BDS carries significant medical risk.

No diagnostic criteria can be said to meet the standards of evidence-based medicine if they cannot be shown to successfully exclude patients with similar conditions that are actually distinct. While research does establish that BDS casts its net widely enough, there exists no research that evaluates whether BDS criteria might be over-inclusive, capturing some conditions, or indeed many conditions, in error. Moreover, strong evidence exists that casts doubt on the safety of including many of the conditions that BDS captures. Based on the conclusions of medical specialist organizations about the biological nature of conditions like irritable bowel syndrome and tension headache, and the emphatic US conclusion that ME/CFS is a serious biological disease, evidence-based caution forces us to acknowledge very serious problems with the medical safety and reliability of the BDS construct.

4. BDS IN THE INTERNATIONAL CLASSIFICATION OF DISEASES

4.1 The rôle of the WHO in evaluating BDS

Problems with the science behind BDS are not difficult to discern. Indeed, as soon as we step outside the realm of psychosomatic medicine to consider the construct from a larger scientific and evidence-based perspective, we see that researchers misrepresent neuroscientific support for the construct, that research fails to establish the construct's success at the task for which it was designed and, most importantly, that research fails to show that the construct safely excludes patients with disease.

It is reasonable to imagine that in considering BDS for implementation in the ICD, the WHO, has, or at least should, play the kind of oversight rôle that would evaluate the construct from this broader perspective. The objective of the WHO, after all, is “the attainment by all peoples of the highest possible level of health” [55] and the ICD is key to meeting that goal. As the diagnostic and coding manual that unifies medical and psychiatric practice across the globe, the ICD determines how conditions in medicine and mental health are organized and, in many cases, how they are conceived. It is difficult

to see what rôle the WHO serves in overseeing the ICD if not to evaluate the scientific success, and certainly the medical safety, of diagnostic constructs proposed for global use.

It has been clear for many years that a replacement for somatoform disorders will be necessary for the 2018 edition of the International Classification of Diseases [32], and at the present time it seems certain that the construct of BDS has not been selected to serve in that rôle [56]. The ICD-11 “Beta Draft” has been available online for well over a year now, and this slot has been filled by “bodily distress disorder” (BDD)—a very close relation to “somatic symptom disorder”, which was implemented in DSM-5 in 2013. But that is not the end of the story.

For the last edition of the ICD, the WHO also published a version specifically for use in primary care (ICD-10-PHC), and it is within that manual that we find the psychiatric diagnosis of medically unexplained symptoms. Though the current primary care ICD is really just an abridged version of the general ICD, the WHO has been working to develop a more substantial version for the upcoming edition, including a new mental health manual that has been specifically designed for use in primary care [57]:

... it has been proposed that the identification and management of common mental disorders needs to be carried out in primary care settings. For this reason, in addition to a version of ICD-11 Mental and Behavioural Disorders for use by mental health specialists, it was considered important to develop a version for use by primary health care providers.

MUS are considered one of the two most common mental health disorders in primary care [14], and it has been clear for many years that the WHO intends to replace MUS in the primary care mental health manual with some version of BDS [12, 14, 15]. Unfortunately, it is also clear that the WHO has chosen not to offer the broad oversight that could verify the science behind BDS and establish its medical safety.

4.2 WHO studies on BDS as “bodily stress syndrome” in primary care

In 2013 the WHO published a focus group study of BDS criteria under the name of “bodily stress syndrome” (BSS), exploring “the opinions of primary care professionals” on BDS criteria as a replacement for the construct of MUS [14]. It was in this study that physicians expressed such considerable concern about medical safety, stating that “the variety of symptoms was so extensive that almost any patient could be labeled as

such”, that “a positive diagnosis ... might make it more likely that a significant organic pathology would be missed”, and that BDS criteria “might lead to missing underlying/more severe illnesses”.

The WHO working group acknowledged a “mixed reaction” to the BDS-based formulation of bodily stress syndrome in that 2013 study, and an intention to modify the BSS construct in ways that would address physicians’ central concerns. Then in 2016 the working group published an extensive field study on the use of BSS in primary care settings around the globe [15]. With this study the working group was careful to evaluate two distinct sets of criteria. The first is a set of very simple criteria adapted for use in primary care settings from the diagnosis of bodily distress disorder proposed for the main ICD-11. These require (1) three or more somatic symptoms not considered by the treating PCP to have a medical basis, and (2) excessive and unjustified anxiety about health. In addition, this study sets out to “examine the importance of the specific symptoms clusters emphasized by Fink and colleagues based on work in Denmark” [15].

The most important problem to note in the second WHO study is that at no point does it address, or even consider, substantial concern among physicians in the first study that BDS criteria will mistakenly capture patients in need of medical care. The second study makes no attempt to refine symptom cluster criteria in a way that will reduce error, and it makes no attempt to evaluate physicians’ views on the medical safety of the new construct. In addition to that very serious problem, the second study also determines that in the vast majority of countries physicians do not find BDS symptom clusters to be a viable replacement for MUS.

Given that at this point BDS criteria have failed both WHO studies, while the simpler criteria based on the construct in the general ICD have broadly succeeded, it seems most reasonable for the working group to conclude that BDS criteria should be abandoned in favour of the simpler formulation. That decision would have the great advantage of aligning the primary care ICD with the general ICD, rather than asking physicians to juggle two wholly different formulations in the two manuals. It is useful to note, moreover, that a 2015 study in Austria independently validated the conclusion that while the simple core criteria for BSS are successful, the addition of BDS symptom cluster criteria is not recommended by physicians outside of Denmark [58].

Surprisingly, however, while the second study acknowledges the failure of symptom cluster criteria in all but one country, the WHO working group immediately reaches this conclusion [15]:

Therefore, for the ICD-11-PHC description of BSS, the best course would seem to be to describe these common patterns but not to require that symptom presentations conform to them in assigning a diagnosis.

This response to the data is deeply puzzling. It is certainly clear that, given the study results, the working group had no choice but to abandon the idea of defining the new construct of BSS with BDS criteria. It is equally clear, however, that the study offers no basis at all for recommending them in the ICD as a useful tool. Based on this study, there is simply no sense in which BDS symptom patterns can be said to be “common”.

It is reasonable to look to the WHO for the kind of oversight that could take note of problems in research with scientific accuracy, scientific method and evidence-based medicine. While the details of neuroscience might be outside the scope of WHO expertise, it seems clear that WHO administrators and working groups are capable of discerning when research for a construct fails to establish success in the goal for which it was designed. Most importantly, it is clear that the WHO is equipped to evaluate whether research meets the standards of evidence-based medicine, and to do so it must demand not only that a construct effectively captures those who suffer from the condition, but also that it effectively excludes those who do not. Unfortunately, when it comes to BDS, the WHO has chosen not to serve as a scientific arbiter of this kind, limiting the scope of its evaluations to the concerns of psychosomatic medicine.

Though there exists no research that examines whether BDS criteria will effectively exclude patients who suffer from disease, though physician focus groups have expressed concern that BDS criteria are medically unsafe – and though both WHO studies have failed to support these criteria for global use – it seems the WHO still intends to recommend them to physicians as a tool for determining when patients’ bodily conditions can safely be diagnosed and managed in psychiatry.

5. CONCLUSIONS

Bodily distress syndrome comes highly recommended within the psychiatric subdiscipline of psychosomatic medicine, based on its ability to resolve theoretical problems with current diagnostic constructs, address conflict between patients and doctors in the examination room, and reduce the cost of MUS and contested conditions for national health systems and insurers. From a broader scientific perspective, however, BDS research misrepresents support for a unified “central sensitivity

syndrome” in neuroscience, and it fails to establish the construct’s scientific success in the task that defines its diagnostic category. Most importantly, as a matter of evidence-based medicine, research fails to establish that the construct succeeds in excluding patients with medical conditions that require medical care.

BDS presents itself to decision makers in the WHO in much the same way that the PACE trial conclusions presented themselves to reviewers and editors at the *Lancet* in 2011. As was the case with the PACE trial (at least with the benefit of hindsight), it seems difficult to imagine how any scientific or evidence-based review could lead decision makers to be convinced about BDS. The conclusion we are forced to draw is—again, as with the PACE trial—that the failings of the science are comfortable for the discipline. While research into BDS could not begin to pass scientific or evidence-based review outside of psychosomatic medicine, professionals within the field find it to be well defended. In short, BDS supports recent concern that scientific standards in psychosomatic medicine are inadequate.

This reality is concerning in a way that goes far beyond the integrity of science—because the decisions of the WHO are not actually like those of journal reviewers and editors. The decisions of the WHO on this matter will determine the health of tens of millions of suffering patients across the globe, so this is as much a matter of ethics as it is a matter of science. The WHO has an ethical obligation to proceed with medical caution; to withhold recommendations in the ICD that can lead to medical error. Above all, the WHO has an ethical obligation to protect access to medical care for every group of patients who might need it.

It is not unusual for researchers in psychosomatic medicine to overlook the ethical imperative to protect access to medical care for every patient who faces a significant possibility of medical need [23], and in a sense it is not surprising that they do so. They are, after all, psychiatric professionals. Though they are called upon to consider conditions that might well be medical, it is clear that calling upon them will lead to psychiatric recommendations. The WHO, however, is in a position to determine when to call upon psychiatric researchers and when to call upon medical researchers. In fact, it is hard to see any more important element of the WHO’s rôle in this area of medicine than to carefully consider when psychiatry is and is not qualified to make a determination about patient care. For the WHO to simply decline to serve in that capacity constitutes an ethical problem of the highest order.

REFERENCES

- White, P.D., Goldsmith, K.A., Johnson, A.L., Potts, L., Walwyn, R., DeCesare, J.C., Baber, H.L., Burgess, M., Clark, L.V., Cox, D.L., Bavinton, J., Angus, B.J., Murphy, G., Murphy, M., O’Dowd, H., Wilks, D., McCrone, P., Chalder, T. & Sharpe, M. PACE trial management group. Comparison of adaptive pacing therapy, cognitive behavior therapy, graded exercise therapy, and specialist medical care for chronic fatigue syndrome (PACE): a randomised trial. *Lancet* **377** (2011) 823–836.
- Ablashi, D., Baraniuk, J., Barcellos, L. et al. An Open Letter to Psychological Medicine about “Recovery” and the PACE Trial. In: *Virology Blog*, 13 March 2017 (<http://www.virology.ws/2017/03/13/an-open-letter-to-psychological-medicine-about-recovery-and-the-pace-trial/>).
- Tuller, D. Trial by Error: The Troubling Case of the PACE Chronic Fatigue Syndrome Study. In: *Virology Blog*, 21 October 2015 (<http://www.virology.ws/2015/10/21/trial-by-error-i/>).
- Rehymeyer, J. & Tuller, D. Getting it wrong with chronic fatigue syndrome. *New York Times Sunday Review* (18 March 2017).
- Green, C.R., Cowan, P., Elk, R., O’Neil, K.M. & Rasmussen, A.L. National institutes of health pathways to prevention workshop: advancing the research on myalgic encephalomyelitis/chronic fatigue syndrome. *Ann. Intern. Med.* **162** (2015) 860–865.
- Recommendations from the HHS CFSAC following publication of: Institute of Medicine *Beyond Myalgic Encephalomyelitis/Chronic Fatigue Syndrome* and National Institutes of Health *Pathways to Prevention Workshop: Advancing the Research on Myalgic Encephalomyelitis/Chronic Fatigue Syndrome*. Washington, DC: Chronic Fatigue Syndrome Advisory Committee (CFSAC) (August 2015).
- IOM (Institute of Medicine). *Beyond Myalgic Encephalomyelitis/Chronic Fatigue Syndrome: Redefining an Illness* (report brief). Washington, DC: National Academy Press (2015).
- Diagnosis and Treatment of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome* (AHRQ Publication No 15-E001-EF, 2014; Addendum 2016). Washington, DC: Agency for Healthcare and Research Quality (2016).
- Geraghty, K. ‘PACE-Gate’: when clinical trial evidence meets open data access. *J. Health Psychol.* **22** (2017) 1106–1112.
- Wilshire, C.E., Kindlon, T., Courtney, R., Matthews, A., Tuller, D., Geraghty, K., & Levin, B. Rethinking the treatment of chronic fatigue syndrome—a reanalysis and evaluation of findings from a recent major trial of graded exercise and CBT. *BMC Psychol.* **6** (2018) 6–17.
- Fink, P., Toft, T., Hansen, M.S., Ornbøl, E. & Oleson, F. Symptoms and syndromes of bodily distress: an exploratory study of 978 internal medical, neurological and primary care patients. *Psychosomatic Med.* **69** (2007) 30–39.
- Ivbijaro, G. & Goldberg, D. Bodily distress syndrome (BDS): the evolution from medically unexplained symptoms (MUS). *Mental Health Family Med.* **10** (2013) 63–64.
- Fink, P. & Schröder, A. One single diagnosis, bodily distress syndrome, succeeded to capture 10 diagnostic categories of functional somatic syndromes and somatoform disorders. *J. Psychosomatic Res.* **68** (2010) 415–426.
- Lam, T.P., Goldberg, D.P., Dowell, A.C., Fortes, S., Mbatia, J.K., Minhas, F.A. & Klinkman, M.S. Proposed new diagnoses of anxious depression and bodily stress syndrome in ICD-11-PHC: an international focus group study. *Family Practice* **30** (2013) 76–87.
- Goldberg, D.P., Reed, G.M., Robles, R., Bobes, J., Fortes, S., de Jesus Mari, J., Lam, T.P., Razzaque, B., Garcia, J.A., Rosendal, M., Dowell, C.A., Gask, L., Mbatia, J.K. & Saxena, S. Multiple somatic symptoms in primary care: a field study for ICD-11 PHC, WHO’s revised classification of mental disorders in primary care settings. *J. Psychosomatic Res.* **91** (2016) 48–54.
- Yunus, M.B. An update on central sensitivity syndromes and issues of nosology and psychobiology. *Current Rheumatol. Rev.* **11** (2015) 70–85.
- Creed, F., Barsky, A. & Leiknes, A.K. Epidemiology: Prevalence, causes and consequences. In: *Medically Unexplained Symptoms, Somatisation and Bodily Distress: Developing Better Clinical Services* (ed. F. Creed, P. Henningsen & P. Fink), pp. 1–42. Cambridge: University Press (2011).
- Kroenke, K. & Mangelsdorff, A.D. Common symptoms in ambulatory care: Incidence, evaluation, therapy, and outcome. *Am. J. Med.* **86** (1989) 262–266.
- Swanson, L.M., Hamilton, J.C. & Feldman, M.D. Physician-based estimates of medically unexplained symptoms: a comparison of four case definitions. *Family Practice* **27** (2010) 487–493.
- Greenberg, D.B. Somatization. In: *Up to Date* (eds J. Dimsdale & D. Solomon) (2018).
- Guidance for Commissioners of Services for People with Medically Unexplained Symptoms*. London: Joint Commissioning Panel for Mental Health (2017).
- Fink, P. & Rosendal, M. (eds). *Functional Disorders and Medically Unexplained Symptoms: Assessment and Treatment*. Aarhus: University Press (2015).
- O’Leary, D. Why bioethics should be concerned with medically unexplained symptoms. *Am. J. Bioethics* **18** (2018) 6–15.
- Rask, M.T., Ornbøl, E., Rosendal, M. & Fink, P. Long-term outcome of bodily distress syndrome in primary care: A follow-up study on health care costs, work disability, and self-rated health. *Psychosomatic Med.* **79** (2017) 345–357.
- Fink, P. Surgery and medical treatment in persistent somatizing patients. *J. Psychosomatic Res.* **36** (1992) 439–447.
- Stone, L. Managing the consultation with patients with medically unexplained symptoms: A grounded theory study of supervisors and registrars in general practice. *Family Practice* **15** (2014) 1–15.
- van Gijn J. In defence of Charcot, Curie, and Wittmann. *Lancet* **369** (2007) 462–480.
- Trimble, M. *Somatoform Disorders: A Medicolegal Guide*. Cambridge: University Press (2004).
- Medically Unexplained Symptoms/Functional Symptoms. Positive Practice Guide*. London: Department of Health (2014).

30. Creed, F. Henningsen, P. & Fink, P. Preface. In: *Medically Unexplained Symptoms, Somatisation, and Bodily Distress: Developing Better Clinical Services* (eds F. Creed, P. Henningsen & P. Fink), pp. vi–viii. Cambridge: University Press (2011).
31. Wessely, S., Nimnuan, C. & Sharpe, M. Functional somatic syndromes: one or many? *Lancet* **354** (1999) 936–939.
32. Creed, F. & Gureje, O. Emerging themes in the revision of the classification of somatoform disorders. *Intl Rev. Psychiatry* **24** (2010) 556–567.
33. Fink, P., Rosendal, M. & Olesen, F. Classification of somatization and functional somatic symptoms in primary care. *Australian–New Zealand J. Psychiatry* **39** (2005) 772–781.
34. Budtz-Lilly, A., Fink, P., Ornbol, E., Vestergaard, M., Moth, G., Christensen, K.S. & Rosendal, M. A new questionnaire to identify bodily distress in primary care: the 'BDS checklist'. *J. Psychosomatic Res.* **78** (2015) 536–545.
35. Rosendal, M., Hartman, T.C.O., Aamland, A., Van der Horst, H., Lucassen, P., Budta-Lilly, A. & Burton, C. "Medically unexplained" symptoms and symptom disorders in primary care: prognosis-based recognition and classification. *BMC Family Practice* **18** (2017) 18–24.
36. Schröder, A., Ornbol, E., Jensen, J.S., Sharpe, M. & Fink, P. Long-term economic evaluation of cognitive-behavioural group treatment versus enhanced usual care for functional somatic syndromes. *J. Psychosomatic Res.* **94** (2017) 73–81.
37. Schröder, A., Rehfeld, E., Ornbol, E., Sharpe, M., Licht, R.M. & Fink, P. Cognitive-behavioural group treatment for a range of functional somatic syndromes: randomized trial. *Br. J. Psychiatry* **200** (2012) 499–507.
38. Barsky, A.J., Goodson, J.D., Lane, R.S. & Cleary, P.D. The amplification of somatic symptoms. *Psychosomatic Med.* **50** (1988) 510–519.
39. Rief, W. & Barsky, A.J. Psychobiological perspectives on somatoform disorders. *Psychoneuroendocrinology* **30** (2005) 996–1002.
40. Wallace, D.J. & Clauw, D.J. (eds). *Fibromyalgia and Other Pain Syndromes*. Philadelphia: Lippincott, Williams and Wilkins (2005).
41. Fink, P. Syndromes of bodily distress or functional somatic syndromes: where are we heading? (lecture on the occasion of receiving the Alison Creed Award (30 June 2017 in Barcelona).
42. Wallit, N., Ceko, M., Gracely, J.L. & Gracely, R.H. Neuroimaging of central sensitivity syndromes: key insights from the scientific literature. *Current Rheumatol. Rev.* **12** (2016) 55–87.
43. Gracely, R.H., Petzke, F., Wolf, J.M. & Clauw, D.J. Functional magnetic resonance imaging evidence of augmented pain processing in fibromyalgia. *Arthritis Rheumatol.* **46** (2002) 1333–1343.
44. Pujol, J., López-Solà, M., Ortiz, H., Vilanova, J.C., Harrison, B.J., Yücel, M., Soriano-Mas, C., Cardoner, N. & Deus, J. Mapping brain response to pain in fibromyalgia patients using temporal analysis of fMRI. *PLoS ONE* **4** (2009) e5224.
45. Giesecke, T., Gracely, R.H., Grant, M.A., Nachemson, A., Petzke, F., Williams, D.A. & Clauw, D.J. Evidence of augmented central pain processing in idiopathic chronic low back pain. *Arthritis Rheumatol.* **50** (2004) 613–623.
46. Hampson, J.P., Reed, B.D., Clauw, D.J., Bhavsar, R., Gracely, R.H., Haefner, H.K. & Harris, R.E. Augmented central pain processing in vulvodynia. *J. Pain* **14** (2013) 579–589.
47. Kawakubo, N., Miyamoto, J.J., Katsuyama, N., Ono, T., Honda, E., Kurabayashi, T., Taira, M. & Moriyama, K. Effects of cortical activations on enhancement of handgrip force during teeth clenching: an fMRI study. *Neurosci. Res.* **79** (2014) 67–75.
48. Rask, T.M., Anderson, R.S., Bro, F., Fink, P. & Rosendal, M. Towards a clinically useful diagnosis for mild-to-moderate conditions of medically unexplained symptoms in general practice: a mixed methods study. *BMC Family Practice* **15** (2014) 118–125.
49. Nimnuan, C., Hotopf, M. & Wessely, S.C. Medically unexplained symptoms: How often and why are they missed? *Q. J. Med.* **93** (2000) 21–28.
50. University of North Carolina Center for Functional GI and Motility Disorders. What are FGIDs? Patient resource (<https://www.med.unc.edu/ibs/patient-education/what-are-fgimds/>).
51. International Headache Society. International Headache Society Classification ICHD-3, 12: Headache attributed to psychiatric disorder (<https://www.ichd-3.org/12-headache-attributed-to-psychiatric-disorder/>).
52. Pustilnik, A. Painful disparities, painful realities. *SSRN Electronic Journal* (2014); doi: 10.2139/ssrn.2407265.
53. Chiara, D.L., Raskovic, D., Pacifico, V., Chung Sheun Thai, J. & Korkina, L. The search for reliable biomarkers of disease in multiple chemical sensitivity and other environmental intolerances. *Intl J. Environ. Res. Public Health* **8** (2011) 2770–2797.
54. Bontle, G.M., Meyer, H., Mason, S., Engelke, U.F.H., Wevers, R.A., Reenen, M.V. & Reineke, C.J. A diagnostic biomarker profile for fibromyalgia syndrome based on an NMR metabolomics study of selected patients and controls. *BMC Neurology* **17** (2017) 88.
55. *Constitution*. Geneva: World Health Organization (1946).
56. Gureje, O. & Reed, G. Bodily distress disorder in ICD-11: problems and prospects. *World Psychiatry* **15** (2016) 291–292.
57. Robles García, R. & Reed, G.M. WHO's ICD-11 for primary health care. *Salud Mental* **40** (2017) 45–46.
58. Spiegel, W., Goldberg, D., Princz, D. & Fellingner, P. New concepts for ICD-classification of common mental disorders from the perspective of general medical practice. *Wiener Medizinische Wochenschrift* **165** (2015) 310–314.