

BRAIN COMMUNICATIONS

Briquet syndrome revisited: implications for functional neurological disorder

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With the creation of the Somatic Symptom and Related Disorders category of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition in 2013, the functional neurological (symptom) disorder diagnostic criteria underwent transformative changes. These included an emphasis on ‘rule-in’ physical examination signs/semiological features guiding diagnosis and the removal of a required proximal psychological stressor to be linked to symptoms. In addition, the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition somatization disorder, somatoform pain disorder and undifferentiated somatoform disorder conditions were eliminated and collapsed into the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition somatic symptom disorder diagnosis. With somatic symptom disorder, emphasis was placed on a cognitive-behavioural (psychological) formulation as the basis for diagnosis in individuals reporting distressing bodily symptoms such as pain and/or fatigue; the need for bodily symptoms to be ‘medically unexplained’ was removed, and the overall utility of this diagnostic criteria remains debated. A consequence of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition restructuring is that the diagnosis of somatization disorder that encompassed individuals with functional neurological (sensorimotor) symptoms *and* prominent other bodily symptoms, including pain, was eliminated. This change negatively impacts clinical and research efforts because many patients with functional neurological disorder experience pain, supporting that the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition would benefit from an integrated diagnosis at this intersection. We seek to revisit this with modifications, particularly since pain (and a Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition somatization disorder comorbidity, more specifically) is associated with poor clinical prognosis in functional neurological disorder. As a first step, we systematically reviewed the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition somatization disorder literature to detail epidemiologic, healthcare utilization, demographic, diagnostic, medical and psychiatric comorbidity, psychosocial, neurobiological and treatment data. Thereafter, we propose a preliminary revision to the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition allowing for the specifier functional neurological disorder ‘with prominent pain’. To meet this criterion, core functional neurological symptoms (e.g. limb weakness, gait difficulties, seizures, non-dermatomal sensory loss and/or blindness) would have ‘rule-in’ signs *and* pain (>6 months) impairing social and/or occupational functioning would also be present. Two optional secondary specifiers assist in characterizing individuals with cognitive-behavioural (psychological) features recognized to amplify or perpetuate pain and documenting if there is a pain-related comorbidity. The specifier of ‘with prominent pain’ is etiologically neutral, while secondary specifiers provide additional clarification. We advocate for a similar approach to contextualize fatigue and mixed somatic symptoms in functional neurological disorder. While this preliminary proposal requires prospective data and additional discussion, these revisions offer the potential benefit to readily identify important functional neurological disorder subgroups—resulting in diagnostic, treatment and pathophysiology implications.

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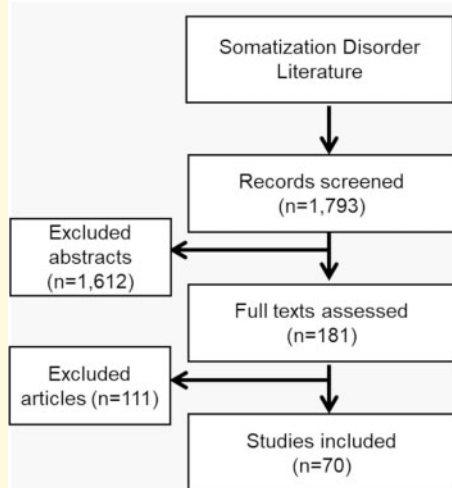
Abbreviations: CBT = Cognitive behavioural therapy; DSM-5 = Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition; DSM-IV = Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition; DSM-IV-SD = Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition Somatization disorder; FND = functional neurological disorder; HC = healthy control; IBS = irritable bowel syndrome; ICD-10 = International Classification of Diseases, Tenth Edition; MDD = major depressive disorder; PCI = psychiatric consultation intervention; PTSD = post-traumatic stress disorder; SD = somatization disorder; SSD = somatic symptom disorder.

Graphical Abstract

Revisiting the Intersection of Sensorimotor Functional Neurological Disorder and Other Distressing Somatic Symptoms: Towards an Integrated Diagnosis

	Briquet Syndrome	Somatization Disorder	Functional Neurological Disorder	Proposed Functional Neurological Disorder Preliminary Revision
	1859	1994	2013	2020
Pain	✓	✓	✗	✓
Neurological symptoms	✓	✓	✓	✓

Systematic Review using PRISMA Guidelines



Proposed Preliminary Revision

Diagnostic and Statistical Manual of Mental Disorders – 5th Edition Functional Neurological (Symptom) Disorder Criteria

Specify if:

- With prominent pain
- With prominent fatigue
- With prominent mixed somatic symptoms

Optional secondary specifiers:

- With symptom-related cognitive-behavioral (psychological) features
- With a contributing comorbidity associated with the somatic symptom(s) of concern

Introduction

Pierre Briquet, the French physician and psychologist, published his *Treatise on Hysteria* in 1859 on 430 patients that provided the basis for the modern-day somatization disorder (SD) (Briquet syndrome) diagnosis (see Fig. 1) (Briquet, 1859; Mai and Merskey, 1980,

1981). Briquet wrote that hysteria was a ‘neurosis of the brain in which the observed phenomena consist chiefly of a perturbation of vital activities, which serve as the manifestation of affective feeling’. While sensorimotor functional neurological symptoms were part of the original symptom complex, pain was a core symptom. Briquet wrote, ‘there is not a single woman with this neurosis

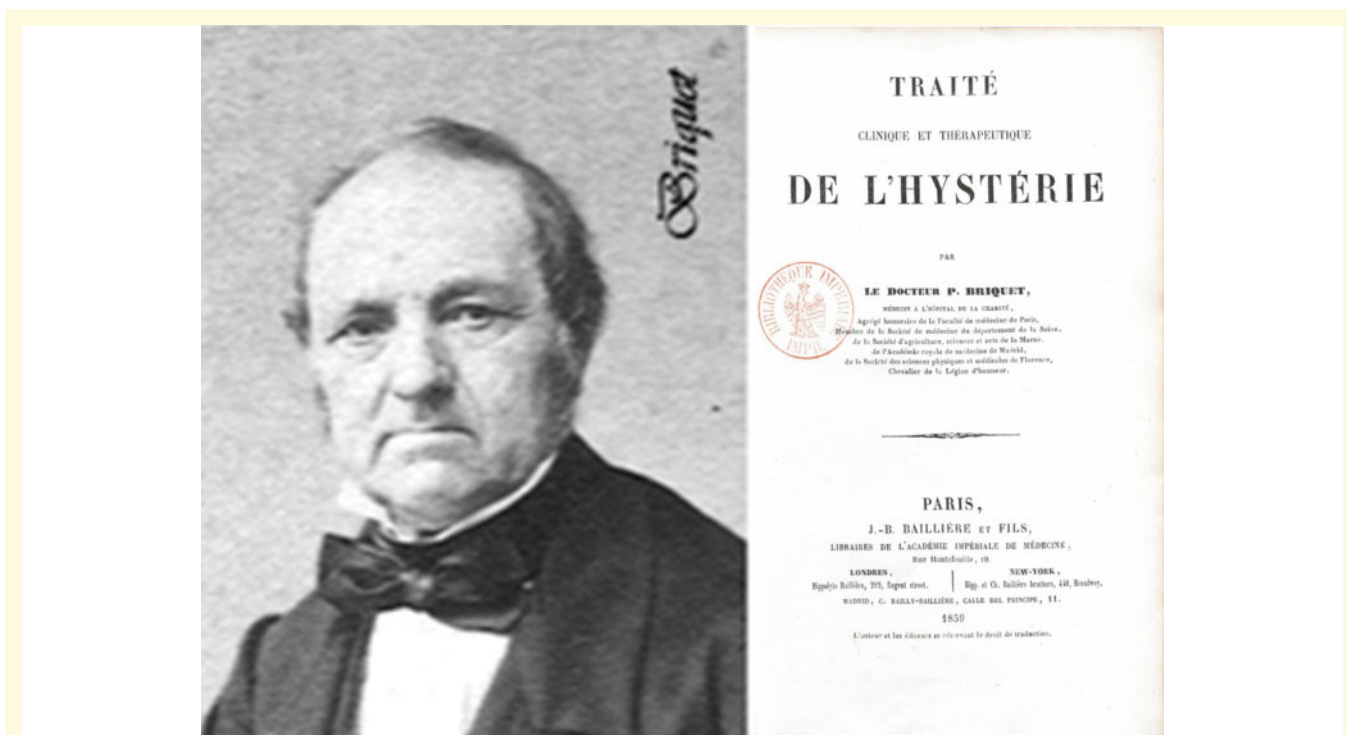


Figure 1 Depicts Pierre Briquet (1796–1881; left panel) alongside his *Treatise on Hysteria* book published in 1859 (right panel). Left panel image reproduced with permission from Fontoura P. The ‘Ajuda Paralytes’: history of a neuropsychiatric debate in mid-19th-century Portugal. *Brain* 2010; 133: 3141–52. Right panel image is in the public domain. Source: Bibliothèque nationale de France, département Sciences et techniques.

who does not have some muscle pain during the course of the illness’ (Briquet, 1859). In the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) Somatization disorder (DSM-IV-SD) category, the presence of at least one functional neurological symptom occurring at some point during the illness course was required—along with four pain symptoms, two gastrointestinal symptoms and one sexual symptom (American Psychiatric Association, 2000). While the DSM-IV-SD criteria were criticized for its somewhat arbitrary symptom domain requirements (Mayou *et al.*, 2005; Rief *et al.*, 2011), a strength was having one diagnosis encompass the frequently encountered intersection of functional neurological disorder (FND) with prominent pain.

With Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), the ‘Somatic Symptom and Related Disorders’ section underwent major changes (American Psychiatric Association, 2013; Dimsdale *et al.*, 2013). The DSM-IV-SD, somatoform pain disorder and undifferentiated somatoform disorder diagnoses were re-conceptualized into one condition—somatic symptom disorder (SSD). The SSD diagnostic criteria removed the need for physical symptoms to be ‘medically unexplained’, and instead emphasized a cognitive-behavioural (psychological) formulation whereby individuals were deemed to engage with bodily symptoms (>6 months duration) using

unhelpful thought patterns, behavioural strategies and/or emotional responses. Hypochondriasis was reframed as illness anxiety disorder, and conversion disorder was re-conceptualized as FND. Major changes to the FND diagnostic criteria included an emphasis on positive neurological examination signs ‘ruling-in’ diagnosis, as well as the removal of the need to relate a proximal stressor to symptom onset; the requirement to exclude feigning, a diagnostic challenge relevant only to a minority of cases, was also eliminated (Stone *et al.*, 2010a). Notably, the DSM-5 FND diagnostic category focuses on motor symptoms (e.g. limb weakness, abnormal movements, seizures) and sensory deficits (e.g. non-dermatomal sensory loss, blindness). As such, a single diagnosis encompassing patients with FND *and* prominent pain is now no longer present in the DSM-5 framework, requiring clinicians to consider dual FND and SSD diagnoses. This is problematic given that the psychological diagnostic criteria for SSD and its potential application to individuals with known symptom-related medical problems have been met with mixed reviews (van der Feltz-Cornelis *et al.*, 2018; Lehmann *et al.*, 2019; Burton *et al.*, 2020; Scamvougeras and Howard, 2020), resulting in variable use of the SSD diagnosis amongst FND experts (Aybek *et al.*, 2020).

In FND, pain is common and clinically relevant (Glass *et al.*, 2018). For example, in a large cohort ($n=107$) of patients with functional limb weakness, pain beyond the

affected limb (64%), headache (40%) and back pain (38%) were frequently present and differentiated FND from neurological controls (Stone *et al.*, 2010b). In 160 functional movement disorder patients, one-fourth reported pain with functional motor symptom onset (Gelauff *et al.*, 2020). Robust associations have also been described between functional dystonia and complex regional pain syndrome (Popkirov *et al.*, 2019). Notably, the presence of chronic pain differentiates individuals with functional (psychogenic non-epileptic/dissociative) seizures from those with epilepsy (Gazzola *et al.*, 2012). In pediatric FND, pain is especially common, with one study in 194 children reporting that 56% had concurrent pain (Kozłowska *et al.*, 2007). Reduced quality of life and poor clinical outcomes have been linked to pain in FND (Ibrahim *et al.*, 2009; Myers *et al.*, 2012). In addition, FND patients with predominantly pain-related medical disability have been excluded from physiotherapy clinical trials, highlighting that this subgroup is being identified indirectly as a distinct entity (Nielsen *et al.*, 2017). Relatedly, pathophysiological models of FND emphasizing altered predictive processing, multimodal integration and emotion processing fit well with a close intersection between sensorimotor FND and pain (Edwards *et al.*, 2012; Diez *et al.*, 2019; Pick *et al.*, 2019), further supporting the need to better characterize the FND—pain intersection.

In this article, we first revisit the explicit intersection of FND and pain in the DSM-IV-SD diagnosis by performing a systematic review of the DSM-IV-SD literature to detail relevant epidemiologic, healthcare utilization, demographic, diagnostic, medical and psychiatric comorbidity, psychosocial, neurobiological and treatment data. Thereafter, we subsequently propose a preliminary revision to the DSM-5 FND diagnostic criteria that allows for the etiologically neutral specifier of FND ‘with prominent pain’ (akin to a FND plus syndrome). This important distinction will greatly aid cohort characterization across diagnostic, treatment and pathophysiology studies, including providing increased clarity regarding the types of patients enrolled in clinical and translational research studies.

Materials and methods

Search strategy

This study was registered in PROSPERO (CRD42020159179). We searched PubMed, PsycINFO and Embase with the terms ‘somatization disorder’ OR ‘Briquet’ from inception to 30 June 2019 in accord with PRISMA guidelines. Reference lists from identified original research articles and reviews were also scrutinized to identify articles meeting eligibility criteria.

Eligibility criteria

Original research studies with patients meeting DSM-IV criteria for SD were included. Only articles in which the DSM-IV-SD diagnosis was obtained by interview (structured or unstructured), as opposed to symptom checklist or self-report questionnaire, were included (Carson *et al.*, 2015). We identified within and between-group studies in the following content areas: epidemiology, healthcare utilization, diagnosis, medical and psychiatric/psychological comorbidities, predisposing vulnerabilities, neural mechanisms, treatment and prognosis. Case reports or series ($n < 10$), review articles, those not written in English, and content published only in abstract or dissertation form were excluded. Articles not using DSM-IV-SD diagnostic criteria [e.g. abridged SD, Diagnostic and Statistical Manual of Mental Disorders, Third Edition, International Classification of Diseases, Tenth Edition or Perley & Guze criteria (Perley and Guze, 1962)] were also excluded (see Table 1). The rationale for this exclusion criterion is that only DSM-IV-SD diagnostic criteria explicitly required at least one functional neurological symptom. Structural neuroimaging studies of DSM-IV-SD were also omitted given that they were previously reviewed elsewhere (Begue *et al.*, 2019).

Data extraction

EndNote was used to compile abstracts from search results of all three databases. After removing duplicates, J.M. and P.R.A. independently applied inclusion/exclusion criteria to determine articles to be read. Discrepancies between the two reviewers were independently resolved by D.L.P. From the list of articles selected, J.M. and P.R.A. narrowed down studies based on inclusion/exclusion criteria. Included articles were evaluated for quality using the National Institutes of Health Study Quality Assessment Tools guidelines (National Institutes of Health: National Heart Lung and Blood Institute) (see Supplementary Table 1). See Fig. 2 for a PRISMA flow diagram of the systematic review (Moher *et al.*, 2009).

Results

Epidemiology

The prevalence of DSM-IV-SD was recorded in eight studies (Escobar *et al.*, 1998; Lynch *et al.*, 1999; Simon and Gureje, 1999; Fink *et al.*, 2004, 2005; Smith *et al.*, 2005; Prerana *et al.*, 2017; Chander *et al.*, 2019). The largest study ($n = 5447$) performed in 14 countries, determined that DSM-IV-SD had a prevalence of 1.4% among primary care outpatients (Simon and Gureje, 1999). Two studies performed in psychiatry clinics, found that 2.6–6.7% met criteria for DSM-IV-SD (Prerana *et al.*, 2017; Chander *et al.*, 2019). Among first time neurology

Table 1 The historical evolution of Briquet syndrome diagnostic criteria

Diagnostic criteria	Name(s)	Date first described	Number of symptoms required	Number of symptom groups required	Neurologic symptoms	Age of onset requirement
Briquet	Briquet Syndrome or Hysteria	1859	25	9 of 10 ^a	Not required	Onset of symptoms by 30
Robins and O'Neal	Hysteria	1953	18	7 of 10 ^a	Not required	More than one major operation or two hospitalizations by 21
Perley and Guze	Hysteria	1962	15	9 of 10 ^a	Not required	Significant medical history by 35
Feighner	Hysteria	1967	Definite diagnosis: 25 Probable diagnosis: 20–24	9 of 10 ^a	Not required	Chronic or recurrent illness by 30
ICD-9	Hysteria unspecified	1977	Not specified	Not specified	Not specified	Not specified
DSM-III	SD	1980	Women: 14 Men: 12	Not required	Not required	Onset of symptoms by 30
DSM-III-R	SD	1987	13	Not required	Not required	Onset of symptoms by 30
Abridged Escobar or Somatic Symptom Index	SD	1987	Women: 6 Men: 4	Not required	Not required	Not specified
ICD-10	SD	1990	Not specified	Not specified	Not required	2-year duration of unexplained somatic symptoms needed
DSM-IV	SD	1994	8	4 ^b	Required	Onset of symptoms by 30
DSM-5	SSD	2013	1	Not specified	Not required	>6 months of symptoms duration

^aGroup 1: Feeling sickly for most of life, or headache; Group 2: blindness, paralysis, anaesthesia, aphonia, fits or convulsions, unconsciousness, amnesia, deafness, hallucinations or urinary retention; Group 3: fatigue, lump in the throat, fainting spells, visual blurring, weakness or dysuria; Group 4: breathing difficulty, palpitation, anxiety attacks, chest pain or dizziness; Group 5: anorexia, weight loss, marked fluctuations in weight, nausea, abdominal bloating, food intolerances, diarrhea or constipation; Group 6: abdominal pain or vomiting; Group 7: dysmenorrhea, menstrual irregularity, including amenorrhea for at least 2 months, or excessive menstrual bleeding; Group 8: sexual indifference, sexual frigidity, dyspareunia, other sexual difficulties or vomiting for all 9 months of pregnancy; Group 9: back pain, joint pain, extremity pain, burning pains of the sexual organs, mouth or rectum or other bodily pains; Group 10: nervousness, fears, depressed feelings, need to quit working or inability to carry on regular duties because of feeling sick, crying easily, feeling life was hopeless, thinking a good deal about dying, wanting to die, thinking of suicide or suicide attempts.

^bGroup 1: pain symptoms; Group 2: gastrointestinal symptoms; Group 3: functional neurological symptoms; Group 4: sexual symptoms.

referrals ($n = 120$), 7% in one cohort were diagnosed with DSM-IV-SD (Fink *et al.*, 2005).

Healthcare utilization

Five studies investigated healthcare utilization in DSM-IV-SD patients, although none had an isolated DSM-IV-SD cohort (Lynch *et al.*, 1999; Hiller *et al.*, 2003; Smith *et al.*, 2005; Frosthholm *et al.*, 2014; Weiss *et al.*, 2017), and two studies had very limited inclusion of DSM-IV-SD patients and will not be further discussed (Lynch *et al.*, 1999; Smith *et al.*, 2005). Weiss *et al.* examined 254 patients (55 with DSM-IV-SD) across 7 outpatient psychotherapy clinics, identifying that those with DSM-IV-SD had a higher number of outpatient doctor visits (an average of 36.5 outpatient doctor visits in the prior year) compared to those with undifferentiated somatoform disorder, somatoform pain disorder and severe DSM-5 SSD ($x = 25.9, 24.7$ and 32.5 outpatient doctor

visits, respectively) (Weiss *et al.*, 2017). The somatoform disorder cohort (including those with DSM-IV-SD) showed two times higher outpatient healthcare utilization compared to the general German population. Negative illness perceptions in 144 patients with somatoform disorders (26 with DSM-IV-SD) correlated with greater healthcare expenditures (Frosthholm *et al.*, 2014). In 172 subjects (54 with DSM-IV-SD) enrolled in an inpatient cognitive behavioural therapy (CBT) treatment programme for somatic symptoms, individuals with DSM-IV-SD had higher outpatient costs than those with abridged SD, and the entire cohort had 2.5% higher outpatient costs compared to average German healthcare system costs (Hiller *et al.*, 2003).

Demographics

Seven studies investigated demographic characteristics in DSM-IV-SD cohorts ($n > 50$) (Guz *et al.*, 2004; Allen

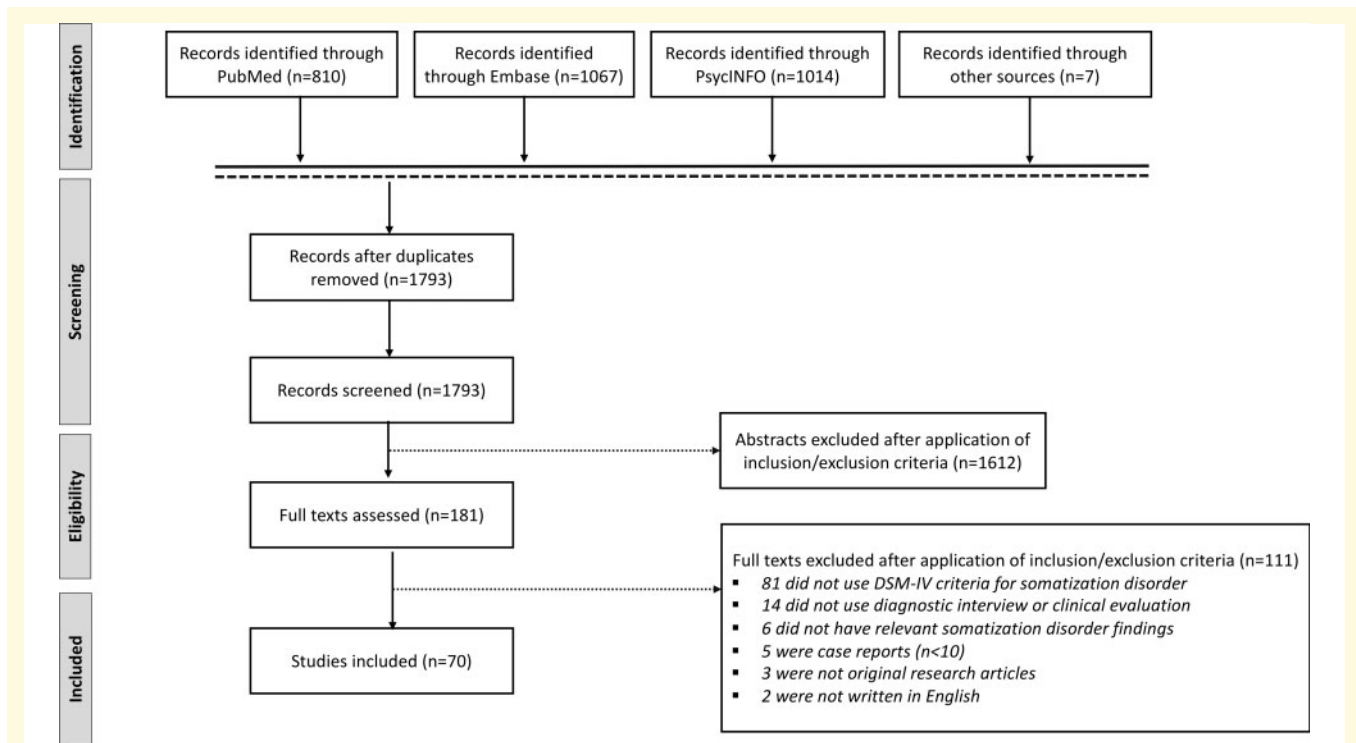


Figure 2 PRISMA flow diagram of the systematic review of somatization disorder, as defined using DSM-IV diagnostic criteria.

et al., 2006; Garcia-Campayo *et al.*, 2007; Kuwabara *et al.*, 2007; Manchikanti *et al.*, 2007; Fjorback *et al.*, 2013; Prerana *et al.*, 2017). DSM-IV-SD had a female predominance (75–89%), with an age range typically between 20 and 50 years old (Guz *et al.*, 2004; Allen *et al.*, 2006; Garcia-Campayo *et al.*, 2007; Kuwabara *et al.*, 2007; Manchikanti *et al.*, 2007; Fjorback *et al.*, 2013; Prerana *et al.*, 2017). Educational background reflected regional differences; 22 ± 8.4 years of schooling were reported in a Japanese cohort (Kuwabara *et al.*, 2007), while only 14% of participants in a Turkish DSM-IV-SD study completed college (Guz *et al.*, 2004). Rates of unemployment were reported in 4 of 7 studies and ranged from 5% to 45% (Guz *et al.*, 2004; Allen *et al.*, 2006; Fjorback *et al.*, 2013; Prerana *et al.*, 2017). Patients receiving disability ranged from 19% to 53% (Allen *et al.*, 2006; Garcia-Campayo *et al.*, 2007; Fjorback *et al.*, 2013). The majority of patients were married (Guz *et al.*, 2004; Allen *et al.*, 2006; Garcia-Campayo *et al.*, 2007; Fjorback *et al.*, 2013; Prerana *et al.*, 2017).

Diagnostic criteria

Four studies evaluated the stability or specificity of the DSM-IV-SD diagnostic criteria (Lynch *et al.*, 1999; Simon and Gureje, 1999; Fink *et al.*, 2005; Smith *et al.*, 2005). A random sample of 3196 primary care patients from the World Health Organization Psychological Problems in General Health Care study conducted in 15 sites across 14 countries were assessed regarding the stability of a

lifetime DSM-IV-SD diagnosis (Simon and Gureje, 1999). Of the 74 (2.3% of total sample) patients initially meeting lifetime DSM-IV-SD criteria, only 21 (28%) of those same individuals in a subsequent interview 1 year later again endorsed a history of DSM-IV-SD. This suggests that the DSM-IV-SD diagnosis was subject to recall bias (an issue not necessarily specific to the DSM-IV-SD diagnosis). Compared to other multiple somatic symptom classification systems, the DSM-IV-SD diagnostic criteria were the most narrowly defined and restrictive (Lynch *et al.*, 1999; Fink *et al.*, 2005; Smith *et al.*, 2005). For example, in 119 patients attending a primary care centre, only 1 (0.84%) met DSM-IV-SD criteria; 10 (8%) met SD when only one gastrointestinal symptom was required and sexual/reproductive symptoms were excluded (Lynch *et al.*, 1999). Similarly, the DSM-IV-SD prevalence was 1% versus 7% by International Classification of Diseases, Tenth Edition in a sample of 198 patients with medically unexplained symptoms (Fink *et al.*, 2005); in 206 primary care patients with medically unexplained symptoms, only 3 (1.5%) met DSM-IV-SD criteria while 39 (19%) met the abridged Escobar criteria (Smith *et al.*, 2005).

Medical comorbidities—functional somatic disorders

Eight studies evaluated the intersection of DSM-IV-SD and medical comorbidities (Hiller *et al.*, 2000; Miller *et al.*, 2001; North *et al.*, 2004; Schrag *et al.*, 2004;

Manchikanti *et al.*, 2007, 2008; Padhy *et al.*, 2016; Chander *et al.*, 2019). The intersection of DSM-IV-SD and functional somatic disorders were primarily investigated in two distinct groups, patients with irritable bowel syndrome (IBS) and individuals with chronic pain disorders (Hiller *et al.*, 2000; Miller *et al.*, 2001; North *et al.*, 2004; Manchikanti *et al.*, 2007, 2008; Padhy *et al.*, 2016). Three studies were conducted in IBS cohorts ($n=24-56$), two from outpatient gastroenterology (Miller *et al.*, 2001; North *et al.*, 2004). The prevalence of DSM-IV-SD in those with IBS ranged from 16% to 25% (Miller *et al.*, 2001; North *et al.*, 2004; Padhy *et al.*, 2016). In these studies, patients with IBS with comorbid DSM-IV-SD showed increased abnormal illness behaviours, greater psychiatric comorbidities and higher rates of other functional somatic disorders compared to individuals with IBS alone (Miller *et al.*, 2001; North *et al.*, 2004; Padhy *et al.*, 2016). Three studies looked at the intersection of chronic pain and DSM-IV-SD (Hiller *et al.*, 2000; North *et al.*, 2004; Manchikanti *et al.*, 2007). Ten of 60 individuals attending an inpatient chronic pain programme had DSM-IV-SD and were grouped together with 31 total patients framed as ‘other functional somatic disorders’ (Hiller *et al.*, 2000). The chronic pain group with a comorbid other functional somatic disorder displayed a greater number of bodily symptoms and pain symptoms, but similar psychological pain distress profiles, depression and anxiety as the chronic pain only group. In a separate outpatient cohort of 500 patients taking opioids for chronic pain, 30% had comorbid DSM-IV-SD (Manchikanti *et al.*, 2007). Higher rates of current illicit drug use were also observed in men with SD versus without (22% versus 9%) (Manchikanti *et al.*, 2007). In 438 patients with chronic spinal pain, a DSM-IV-SD diagnosis ($n=162$) did not significantly influence false positive (placebo) rates following a single anesthetic administration (Manchikanti *et al.*, 2008). While not well studied, in addition to functional somatic disorders, patients with DSM-IV-SD were also reported to have cardiovascular and endocrine conditions, and peripheral injuries (including fractures) as common comorbid medical conditions (Schrag *et al.*, 2004; Chander *et al.*, 2019).

Psychiatric comorbidities

Eleven studies characterized psychiatric comorbidities in DSM-IV-SD (Battaglia *et al.*, 1998; Rief *et al.*, 2001; Carey *et al.*, 2003; Mohlman *et al.*, 2004; Brown *et al.*, 2005; Garcia-Campayo *et al.*, 2007; Ozturk and Sar, 2008; Sertoz *et al.*, 2009; Spitzer *et al.*, 2009; Taycan *et al.*, 2014; Chander *et al.*, 2019). Many studies reported high rates of mood disorders (10–90%) (Brown *et al.*, 2005; Sertoz *et al.*, 2009; Taycan *et al.*, 2014; Chander *et al.*, 2019); panic attacks (41%) (Brown *et al.*, 2005); lifetime post-traumatic stress disorder (PTSD) or complex PTSD (14–55%) (Brown *et al.*, 2005; Spitzer

et al., 2009; Taycan *et al.*, 2014); generalized anxiety disorder (45%) (Brown *et al.*, 2005); any anxiety disorder (15%) (Chander *et al.*, 2019); lifetime dissociative disorder (28–50%) (Brown *et al.*, 2005; Taycan *et al.*, 2014) and borderline personality disorder (15%) (Taycan *et al.*, 2014) within DSM-IV-SD cohorts. Similarly, DSM-IV-SD was found to be highly comorbid within PTSD (35%), panic disorder (11%) and dissociative disorder (33%) populations (Carey *et al.*, 2003; Mohlman *et al.*, 2004; Ozturk and Sar, 2008).

The largest sample of DSM-IV-SD patients ($n=70$) reported that 39% of patients did not have another DSM-IV Axis I comorbidity (Garcia-Campayo *et al.*, 2007). The remaining DSM-IV-SD population had comorbid mood disorders [major depressive disorder (MDD) 13%; dysthymia 11%], anxiety disorders (panic disorder 13%; generalized anxiety disorder 10%; agoraphobia 4%), or another mood/anxiety disorder (6%). Sixty-four percent of individuals with DSM-IV-SD also met criteria for one or more personality disorders (Garcia-Campayo *et al.*, 2007).

Several studies also examined relationships between psychiatric comorbidities and dimensional psychological characteristics in DSM-IV-SD. One study used the Brief Symptom Inventory to assess differences in psychopathology between DSM-IV-SD with complex PTSD ($n=10$), DSM-IV-SD without complex PTSD ($n=18$) and MDD without complex PTSD ($n=27$) (Spitzer *et al.*, 2009). Here, patients with DSM-IV-SD and complex PTSD exhibited higher psychoticism and interpersonal problems than both other groups, and higher obsessive compulsive and anger–hostility tendencies than the DSM-IV-SD without complex PTSD group. Both DSM-IV-SD groups showed greater obsessionality, anxiety, anger–hostility and global severity of psychopathology compared to the MDD group (Spitzer *et al.*, 2009). Furthermore, another study reported that the co-occurrence of major depression and somatization (37.5% with full-criteria DSM-IV-SD) was linked to increased psychopathology on the Symptom Checklist-90 Revised (Rief *et al.*, 2001). In another study, 18 DSM-IV-SD patients with comorbid panic disorder showed higher novelty seeking scores compared to 41 individuals with panic disorder and 22 healthy controls (HCs), and novelty seeking positively correlated with number of somatic symptoms (Battaglia *et al.*, 1998).

DSM-IV-SD versus Other DSM-IV somatoform disorders

Five studies compared DSM-IV-SD to other somatoform disorders (Escobar *et al.*, 1998; Sanyal *et al.*, 1998; Guz *et al.*, 2004; Espirito-Santo and Pio-Abreu, 2009; Kirpinar *et al.*, 2016), including two examining similarities and differences between DSM-IV-SD and hypochondriasis (Escobar *et al.*, 1998; Kirpinar *et al.*, 2016) and three comparing DSM-IV-SD and DSM-IV conversion

disorder (Sanyal *et al.*, 1998; Guz *et al.*, 2004; Espirito-Santo and Pio-Abreu, 2009). In a primary care setting of 1456 outpatients, 49 patients with hypochondriasis and 20 with DSM-IV-SD were identified (Escobar *et al.*, 1998). One in five patients with DSM-IV-SD also had hypochondriasis; a higher rate of DSM-IV-SD amongst hypochondriasis patients was also observed with a prevalence of 9% versus 1% in those without hypochondriasis. Kirpinar *et al.* characterized a consecutive sample of 73 outpatients in a somatoform disorders unit ([DSM-IV-SD ($n=51$) and hypochondriasis ($n=22$)], identifying a higher female prevalence in the DSM-IV-SD group (71% versus 41%) and higher health anxiety in the hypochondriasis group (Kirpinar *et al.*, 2016). The two cohorts showed similar Beck Anxiety Inventory, Hamilton Depression Rating Scale, Somatosensory Amplification Scale, Somatoform Dissociation Questionnaire, Dissociative Experiences Scale and State-Trait Anxiety Inventory scores.

DSM-IV-SD ($n=40$), conversion disorder ($n=26$) and dissociative disorders ($n=38$) were compared in one study (Espirito-Santo and Pio-Abreu, 2009), with more commonalities found amongst the dissociative and conversion disorder groups (e.g. greater dissociation) than between the DSM-IV-SD and conversion disorder groups. More depression and paranoia symptoms were seen in those with dissociative disorders, while more somatic and obsessive symptoms were appreciated in the DSM-IV-SD group. In a separate study, 71 DSM-IV-SD patients had lower paranoia and psychotic personality traits compared to 87 individuals with DSM-IV conversion disorder (Guz *et al.*, 2004). Individuals with DSM-IV-SD also exhibited lower self-appraisal compared to both patients with conversion disorder and HCs (Sanyal *et al.*, 1998).

Comorbid DSM-IV-SD in FND

Six studies reported on the co-occurrence of DSM-IV-SD and FND (Interian *et al.*, 2004; Schrag *et al.*, 2004; Marchetti *et al.*, 2008; Stone *et al.*, 2010b; Epstein *et al.*, 2016; Gelauff *et al.*, 2019). In one study, 8 (12.5%) of 36 patients with functional movement disorder met criteria for comorbid SD (Epstein *et al.*, 2016); 12 (32%) of 38 patients with probable or diagnosed functional dystonia had comorbid DSM-IV-SD in another study (Schrag *et al.*, 2004). Likewise, 19% of patients experiencing functional (non-epileptic/dissociative) seizures ($n=27$) also had DSM-IV-SD (Marchetti *et al.*, 2008). In a large sample of 107 patients with functional limb weakness, 27% had a comorbid DSM-IV-SD (Stone *et al.*, 2010b). However, the presence of functional neurological symptoms in 120 patients with medically unexplained symptoms was not predictive of a DSM-IV-SD diagnosis (Interian *et al.*, 2004). Importantly, the dual diagnosis of DSM-IV-SD and FND correlated with poor clinical outcomes in a 14-year follow-up study of 76 patients with functional limb weakness (Gelauff *et al.*, 2019).

Dimensional psychopathology

Eleven studies measured dimensional psychological characteristics using self-report or clinician-rated scales in DSM-IV-SD samples (Sanyal *et al.*, 1998; Brown *et al.*, 2005; Ozturk and Sar, 2008; Sertoz *et al.*, 2009; Spitzer *et al.*, 2009; Landa *et al.*, 2012; Taycan *et al.*, 2014; Kirpinar *et al.*, 2016; Prerana *et al.*, 2017; Davoodi *et al.*, 2018, 2019). Patients with DSM-IV-SD reported elevated depression scores compared to HCs in one study (Taycan *et al.*, 2014). Another study compared DSM-IV-SD patients to those with MDD, finding more anxiety in the DSM-IV-SD cohort (Spitzer *et al.*, 2009). The number of bodily complaints positively correlated with depression and anxiety scores in an DSM-IV-SD sample (Prerana *et al.*, 2017). Higher rates of suicide attempts and self-mutilation were also identified in DSM-IV-SD compared to HCs (Ozturk and Sar, 2008; Taycan *et al.*, 2014). Dissociation in DSM-IV-SD has also been characterized in multiple studies (Brown *et al.*, 2005; Taycan *et al.*, 2014; Kirpinar *et al.*, 2016). Individuals with DSM-IV-SD exhibited increased dissociation and more trance-like states compared to HCs (Taycan *et al.*, 2014), as well as higher dissociative amnesia rates compared to neurological controls (i.e. dystonia) (Brown *et al.*, 2005).

Studies have characterized an external locus of control in DSM-IV-SD compared to HCs (Sanyal *et al.*, 1998). Increased alexithymia, higher level of mistrust and an unmet need for interpersonal closeness were also identified in a mixed DSM-IV somatoform disorder cohort ($n=20$) compared to HCs ($n=20$) (Landa *et al.*, 2012). Furthermore, worse body image and self-esteem were identified in a mixed somatoform disorders sample (38% with DSM-IV-SD) compared to both healthy and medical controls (patients with breast cancer status-post total mastectomy) (Sertoz *et al.*, 2009). By contrast, when compared to patients with MDD, individuals with DSM-IV-SD exhibited less severe maladaptive schemas (Davoodi *et al.*, 2018) and use of more adaptive emotional regulation strategies (Davoodi *et al.*, 2019).

Traumatic life events

Three studies investigated childhood trauma burden in DSM-IV-SD cohorts (Brown *et al.*, 2005; Spitzer *et al.*, 2008; Taycan *et al.*, 2014). Forty women with DSM-IV-SD reported a greater number of childhood trauma types and were more likely to have experienced physical abuse (20%), emotional abuse (25%) and emotional neglect (30%) compared to 40 healthy women (Taycan *et al.*, 2014). In another study, 28 individuals with DSM-IV-SD reported more childhood sexual (43%) and physical abuse (54%) compared to 28 patients with MDD (Spitzer *et al.*, 2008). Furthermore, the severity of sexual and physical abuse distinguished the DSM-IV-SD group from the MDD cohort (Spitzer *et al.*, 2008). Similarly, more severe early-life maltreatment distinguished DSM-IV-SD

from neurological populations, with one study showing that 22 DSM-IV-SD patients reported more severe childhood physical abuse, increased emotional abuse exposure, greater number of emotional abuse perpetrators, and a longer duration of emotional abuse compared to 19 dystonia controls (Brown *et al.*, 2005). Those with DSM-IV-SD also reported more family conflict during childhood (Brown *et al.*, 2005). In a qualitative, interview-based study, several patients with DSM-IV-SD articulated links between early-life maltreatment, somatic symptoms and healthcare use (Morse *et al.*, 1997).

Regarding lifetime trauma, 40 women with DSM-IV-SD reported more traumatic events in adulthood (mean = 2.6) than 40 healthy women (mean = 1.2), and a higher number of adulthood traumatic experiences predicted the DSM-IV-SD diagnosis (Taycan *et al.*, 2014). Three studies assessed lifetime trauma in mixed cohorts (Carey *et al.*, 2003; Landa *et al.*, 2012; Brown *et al.*, 2014). Twenty individuals in a somatoform disorder cohort comprised of DSM-IV-SD, somatoform pain and undifferentiated somatoform disorders experienced more lifetime trauma than 20 HCs, and 76% of the traumatic experiences by the patient group were interpersonal in nature (e.g. assault, divorce and separation) (Landa *et al.*, 2012). In primary care, 36 individuals with DSM-IV-SD experienced more traumatic lifetime events than 165 medical patients without DSM-IV-SD (Carey *et al.*, 2003). In 898 twins discordant for lifetime trauma exposure, adverse experiences correlated with increased risk of developing DSM-IV-SD (Brown *et al.*, 2014); this highlights the potential etiological relevance of adversity in DSM-IV-SD.

Pathophysiology

Ten inter-related neuroimaging articles reported resting-state functional MRI (fMRI) findings in a single cohort of 25 drug-naive patients with DSM-IV-SD compared to 28 HCs (Su *et al.*, 2014; Song *et al.*, 2015; Su *et al.*, 2015, 2016; Wang *et al.*, 2016; Wei *et al.*, 2016; Guo *et al.*, 2017; Li *et al.*, 2018; Ou *et al.*, 2018; Pan *et al.*, 2019) (see Supplementary Table 2). Among the findings identified, patients with DSM-IV-SD showed increased functional connectivity within the right inferior temporal gyrus compared to HCs (Su *et al.*, 2015). Measuring local and distant functional connectivity, patients with DSM-IV-SD exhibited increased short-range functional connectivity in the right superior frontal gyrus and left pallidum, and increased long-range functional connectivity in the left middle frontal gyrus and right inferior temporal gyrus (Guo *et al.*, 2017).

In a single photon emission computed tomography study performed in 11 DSM-IV-SD patients, the most common findings were a normal single photon emission computed tomography image ($n=4$) and right cerebellum hypoperfusion ($n=4$) (Garcia-Campayo *et al.*, 2001). Another study used magnetic resonance spectroscopy to measure brain metabolites in patients with DSM-IV-SD,

fibromyalgia and HCs (10 subjects per group). The combined DSM-IV-SD and fibromyalgia cohort showed increased posterior cingulate glutamate levels, but this did not remain statistically significant when only comparing DSM-IV-SD patients and HCs (Fayed *et al.*, 2012).

One electrophysiology study measured attention and working memory mechanisms in 25 DSM-IV-SD patients and HCs using auditory-evoked potentials (Garcia Campayo *et al.*, 2007). Patients with DSM-IV-SD had greater latency in the time needed to perceive, identify and classify new information (Garcia Campayo *et al.*, 2007). In an autonomic study, 10 DSM-IV-SD patients showed higher baseline electrodermal activity (a marker of increased sympathetic tone) compared to individuals with DSM-IV conversion disorder and HCs (Sanyal *et al.*, 1998). Three articles investigated inflammatory markers in DSM-IV-SD, although no consistent pattern emerged across studies (Rief *et al.*, 2001; Hossain *et al.*, 2007a, b).

Treatment

Three treatment studies were specifically performed in DSM-IV-SD cohorts (Allen *et al.*, 2001, 2006; Fjorback *et al.*, 2013). In a small within-group study, 11 individuals assigned to 10 weekly outpatient CBT sessions reported less physical discomfort and improved physical functioning at the end of treatment (Allen *et al.*, 2001). In a trial comparing outpatient CBT plus a psychiatric consultation intervention (CBT+PCI; $n=43$) to a PCI alone ($n=41$), the CBT+PCI treatment arm showed greater improvement in somatic symptoms and physical functioning (Allen *et al.*, 2006). Fifty-nine subjects assigned to eight sessions (plus one follow-up) of mindfulness therapy showed improved general health, health anxiety, physical symptoms, anxiety and depression as compared to 60 patients receiving PCI (Fjorback *et al.*, 2013).

In addition, several mixed-cohort treatment studies included individuals with DSM-IV-SD. In a bodily distress syndrome cohort ($n=120$; 45% meeting DSM-IV-SD criteria), subjects who received nine sessions (over 4 months) of outpatient CBT-based treatment plus PCI showed greater improvement in physical functioning, bodily pain and vitality than those who received a PCI alone (Schröder *et al.*, 2012). In an inpatient CBT study comparing a mixed-somatoform disorders cohort ($n=172$, 31% meeting DSM-IV-SD criteria) to a mixed psychiatric cohort ($n=123$, primarily mood and anxiety disorders), both groups comparably improved in physical and mental health measures (Hiller *et al.*, 2003). In a somatization syndrome cohort (28% meeting DSM-IV-SD criteria), 107 subjects receiving eight sessions of inpatient CBT plus symptom management training and 84 subjects who underwent eight sessions of inpatient CBT plus relaxation training similarly improved in somatic symptoms, mental health and life satisfaction (Bleichhardt *et al.*, 2004).

Additional mixed-cohort studies with DSM-IV-SD making up <20% of the cohort have shown efficacy for intensive inpatient treatment (involving CBT) (Rief and Hiller, 2003), CBT-based outpatient treatment (Martin *et al.*, 2007; Zonneveld *et al.*, 2012; Schröder *et al.*, 2013), outpatient mindfulness therapy (van Ravesteijn *et al.*, 2013) and progressive muscle relaxation (Schröder *et al.*, 2013). Other studies have shown efficacy using antidepressants (Voon and Lang, 2005), and multidisciplinary treatment (Schrag *et al.*, 2004). See Supplementary Table 3 for additional treatment trial details in DSM-IV-SD.

Discussion

This systematic review of DSM-IV-SD identified a number of observations that parallel themes emerging in the DSM-5 FND literature (Ludwig *et al.*, 2018; Baslet *et al.*, 2020; Perez *et al.*, 2020). Patients with DSM-IV-SD were predominantly (although not exclusively) female with increased medical and psychiatric comorbidities, including functional somatic disorders (e.g. IBS) and elevated rates of mood, anxiety, trauma-related and personality disorders. Dimensionally, individuals with DSM-IV-SD reported increased alexithymia, dissociation, an external locus of control and health anxiety. Of etiological relevance, patients with DSM-IV-SD reported increased childhood and lifetime adverse life experiences, including higher rates compared to healthy and neuropsychiatric populations. A large twin study discordant for lifetime trauma exposure showed positive associations between adverse life experiences and increased risk for DSM-IV-SD (Brown *et al.*, 2014). Treatment studies also identified that skills-based psychotherapy was effective in reducing somatic symptoms in patients with DSM-IV-SD. Interestingly, neural mechanisms were particularly understudied in DSM-IV-SD, which may relate in part to the low 1-3% prevalence rates driven in the context of overly stringent diagnostic criteria.

The above overlapping characteristics between DSM-IV-SD and FND have relevant clinical and research implications in FND based on three additional observations: (i) pain is a common comorbid symptom in FND [including particularly high rates in pediatric FND (Kozłowska *et al.*, 2007)] that can limit treatment engagement and incur a poor prognosis (Glass *et al.*, 2018); (ii) a subset of FND patients meet criteria for comorbid DSM-IV-SD (Interian *et al.*, 2004; Schrag *et al.*, 2004; Marchetti *et al.*, 2008; Stone *et al.*, 2010b; Epstein *et al.*, 2016); (iii) comorbid DSM-IV-SD in FND is associated with a poor prognosis (Gelauff *et al.*, 2019). These interrelated themes demonstrate the importance of explicitly considering pain in the assessment, management and research of patients with FND, and underscore the need to better identify the co-occurrence of pain in FND populations. Furthermore, based on neurological examination alone, the assessment of pain requires a more nuanced approach

that is not as straight forward as a ‘rule-in’ diagnosis of motor FND based on clinical features such as Hoover sign or tremor entrainment with high diagnostic specificity (Daum *et al.*, 2014). Given concerns regarding SSD diagnostic criteria (based in part on its psychological framework) (van der Feltz-Cornelis *et al.*, 2018; Burton *et al.*, 2020), there is also evidence of heterogeneity in the willingness of FND clinical experts to diagnose SSD in patients with FND (Aybek *et al.*, 2020); an alternative would be to view pain as a commonly present, yet non-specific symptom of FND. The concern with the latter approach is that pragmatically, the presence or absence of pain has high clinical and research relevance, including but not limited to that predominant pain symptoms have been an exclusion criteria for physiotherapy trials in FND (Nielsen *et al.*, 2017). In a randomized feasibility study of physiotherapy for motor FND conducted by Nielsen and colleagues, 27% of the 210 patients screened were excluded based on predominant pain; notably, 47% of the 60 enrolled nonetheless reported ‘severe to extreme’ pain ratings (Nielsen *et al.*, 2017). Regarding neural mechanisms, the central pain matrix implicated in chronic pain disorders overlaps with salience (cingulo-insular) network areas implicated in the pathophysiology of FND (Denk *et al.*, 2014; Begue *et al.*, 2019), underscoring that pain has neurobiological consequences that need to be considered in FND pathophysiology research. Thus, to further advance clinical and research efforts in FND, there is a critical need to provide a practical approach to identify patients that also have prominent pain.

As a preliminary proposal that we hope will catalyse considerable discussion in the field [including amongst the FND Society (www.fndsociety.org) leadership], we argue that a revision to the DSM-5 should be considered to explicitly identify the subset of patients with FND that also have prominent pain. In Fig. 3, we detail the proposed diagnostic criteria allowing for the diagnosis of FND ‘with prominent pain’ (a specifier). Here, patients would meet rule-in criteria for sensorimotor FND and concurrently endorse pain symptoms for at least 6 months that are also impairing to social and/or occupational functioning. In an effort to remain etiologically neutral while acknowledging the biopsychosocial complexity of predisposing and perpetuating factors, we suggest an optional secondary specifier of ‘with cognitive-behavioral (psychological) features’. We propose this optional secondary specifier for the following reasons: (i) an exploration of psychological factors early in the diagnostic assessment for patients with physical symptoms can be challenging for both patients and clinicians alike (and psychological factors related to pain may not be present in all patients); (ii) identification of psychological constructs either amplifying or perpetuating pain have translatable clinical utility (CBT treatment targets), suggesting that there is merit in identifying this subgroup; (iii) the use of ‘rule-in’ psychological criteria as proposed in SSD and related diagnostic formulations remain debated, with

Proposed DSM-5 Functional Neurological (Symptom) Disorder Diagnostic Criteria Revision

- A) One or more symptoms of altered voluntary motor or sensory function.
- B) Clinical findings provide evidence of incompatibility between the symptom and recognized neurological or medical conditions.
- C) The symptom or deficit is not better explained by another medical or mental disorder.
- D) The symptom or deficit causes clinically significant distress or impairment in social, occupational, or other important areas of functioning or warrants medical evaluation.

Specify symptom type:

With weakness or paralysis

With abnormal movement (e.g., tremor, dystonic movement, myoclonus, gait disorder)

With swallowing symptoms

With speech symptom (e.g., dysphonia, slurred speech)

With attacks or seizures

With anesthesia or sensory loss

With special sensory symptom (e.g., visual, olfactory, or hearing disturbance)

With mixed symptoms

Specify if:

Acute episode: Symptoms present for less than 6 months

Persistent: Symptoms occurring for 6 months or more

Specify if:

With psychological stressor (specify stressor)

Without psychological stressor

Specify if:

With prominent pain

With prominent fatigue

With prominent mixed somatic symptoms

Optional secondary specifiers:

With symptom-related cognitive-behavioral (psychological) features

With a contributing comorbidity associated with the somatic symptom(s) of concern

Figure 3 Preliminary proposal for a revision to the DSM-5 for FND. We suggest the addition of three new specifiers: ‘with prominent pain’; ‘with prominent fatigue’ and ‘with prominent mixed somatic symptoms’. Patients must first meet complete criteria for FND (criteria A–D). In addition, pain, fatigue and/or mixed somatic symptoms should themselves be impairing to social and/or occupational functioning and present for at least 6 months. The above three specifiers are etiologically neutral, which acknowledges the biopsychosocial heterogeneity present in the development and maintenance of these somatic symptoms. To provide additional clarification, we also propose two optional secondary specifiers: (i) with symptom-related cognitive-behavioural (psychological) features; and (ii) with a contributing comorbidity associated with the somatic symptom(s) of concern. The former optional specifier allows the identification of individuals displaying psychological constructs either amplifying or perpetuating pain that can have clinical utility (e.g. CBT treatment targets). The latter optional specifier encourages an appropriate (not necessarily exhaustive) medical workup for the identified somatic symptoms, as well as aids the characterization of relevant medical and neurological comorbidities (including functional somatic disorders) that has been a shortcoming of FND research to date. If a relevant comorbidity is present, this should be noted when using this optional specifier. Regardless of whether or not this latter optional specifier is used, clinicians should be mindful to evaluate pain, fatigue and other somatic symptoms without ‘rule-in’ physical examination features in FND patients as they would in other populations (to prevent premature diagnostic anchoring).

a recent European workgroup for functional somatic disorders taking a similar etiologically neutral stance to that outlined above (Burton *et al.*, 2020).

An additional consideration is whether an individual’s chronic pain symptoms are driven by another recognized medical/neurological/functional somatic disorder known to cause pain (e.g. small fibre neuropathy, fibromyalgia, severe lumbar stenosis) or is part of the intrinsic disease processes occurring in FND itself. Here, we recommend another optional secondary descriptive specifier: ‘with a contributing comorbidity associated with the somatic symptom(s) of concern’. If such a condition is present, this can be recorded. The rationale for this specifier is 2-fold: (i) encourages an appropriate (not necessarily exhaustive) medical workup for the individual’s pain

symptoms; and (ii) makes transparent if an identified pain-related comorbidity is present, which can have both treatment and research implications. As a cautionary note, regardless of whether or not this optional specifier is used, clinicians should evaluate pain, fatigue and other somatic symptoms without ‘rule-in’ physical examination features in patients with FND as they would in other populations (to prevent premature diagnostic anchoring). While prospective studies are needed to test the reliability and utility of this suggested DSM-5 revision, we speculate that this diagnostic approach will identify the vast majority of patients with FND that are also endorsing prominent pain symptoms.

Operationalizing an FND with prominent pain subgroup has clear clinical and research advantages. By

aiding the identification of this important (and likely prevalent) FND subgroup, this approach allows clinicians to consider guiding individuals towards interdisciplinary mind-body pain programmes that may be potentially more suitable for initial management in comparison to emerging motor FND care models (Jimenez *et al.*, 2019). It also allows for the prioritization of treatment targets using a stepwise treatment approach in which a patient's most prominent physical symptoms are addressed first. In our opinion, this could optimize the likelihood of success with physiotherapy. With an improved ability to operationalize inclusion criteria, our proposal will also catalyse clinical trial research in this potentially more treatment refractory and costly population. Furthermore, with the boom in using brain imaging approaches to elucidate the neurocircuitry of FND, and to identify prognostic biomarkers, there is growing need to more precisely characterize patients that may help explain the high-degree of variability found in FND studies to date (Begue *et al.*, 2019).

A related question not yet addressed is how to also contextualize other prominent physical symptoms in patients with FND, most notably but not limited to fatigue (Aybek *et al.*, 2020; Gelauff *et al.*, 2020). Like pain, fatigue is a common symptom in FND that is also linked to reduced quality of life and reduced treatment engagement (Věchetová *et al.*, 2018). We suggest that the same approach taken for pain can also be used to categorize FND patients with prominent fatigue symptoms. This would include using the specifier 'with prominent fatigue' for those individuals that endorse fatigue (>6 months) is limiting their occupational and/or social functioning. Optional secondary specifiers can denote the presence or absence of cognitive-behavioural (psychological) features and the co-occurrence with a contributing comorbidity known to be associated with fatigue (e.g. multiple sclerosis, chronic fatigue syndrome). Lastly, for patients with prominent pain *and* fatigue, or those exhibiting a combination of two or more non-sensorimotor symptoms (e.g. widespread body pain and gastrointestinal distress), the diagnostic specifier FND 'with prominent mixed somatic symptoms' can be recorded (see Fig. 3).

Our systematic review and proposed revised DSM-5 diagnostic criteria for FND have several limitations. Alternative approaches such as systematically reviewing the frequency and relevance of pain, fatigue and other somatic symptoms in FND more broadly were not performed. Such efforts in the future could further inform the preliminary DSM-5 revisions proposed here (e.g. 6-month duration was chosen to be consistent with the symptom duration requirement for SSD, however, other durations could be considered). It is also important to note that many of the observations identified in DSM-IV-SD, such as increased rates of mood and anxiety disorders, alexithymia, dissociation and adverse life events, are non-specific features found in a range of other

neuropsychiatric disorders. However, our proposed DSM-5 FND diagnostic criteria revisions will help catalyse new research in FND with prominent pain compared to a range of healthy, medical and neuropsychiatric populations, which will allow for rigorous investigations of the relevance and specificity of clinical and neurobiological associations in this subgroup. In addition, we want to explicitly note that we are not advocating for a return of the term 'SD', which is fraught with limitations including a dualistic mind-brain framing and an arbitrary selection of symptom clusters—most precarious of which is the sexual symptoms cluster. Another question is the potential benefit, or lack thereof, of revising the diagnostic criteria for FND within the DSM-5 framework. While we recognize that psychological factors may not universally play important predisposing and/or perpetuating roles in all patients, for many patients cognitive-behavioural (psychological) factors are relevant—particularly for developing patient-centred treatment plans. As such, it is important to continue to actively engage psychiatrists, psychologists and allied mental health professionals as equal partners alongside neurologists and allied rehabilitation specialists in FND-related clinical and research activities. It will be important, however, for the FND field to achieve consensus across neurologic and psychiatric perspectives, and we hope our etiologically neutral specifier 'with prominent pain' assists in these efforts. We also welcome our proposal being considered in future diagnostic criteria considerations by the FND Society. Lastly, we acknowledge that this article does not address the question of how to contextualize patients without sensorimotor FND that experience other distressing physical symptoms (with normal neurological examinations and negative medical evaluations). While there are a range of potential diagnostic classification systems being actively debated (each with their own strengths and weaknesses) (Burton *et al.*, 2020), it will be important to bring together leaders (and patients) across the FND and functional somatic disorder fields to obtain consensus across the various stakeholders.

In conclusion, the intersection of FND with other prominent somatic symptoms, most notably pain, is clinically and prognostically relevant. The changes made from DSM-IV to DSM-5 eliminated DSM-IV-SD that encompassed individuals with functional neurological symptoms *and* other prominent somatic symptoms. We propose a preliminary revision to the DSM-5 diagnostic criteria for FND that adds an etiologically neutral specifier noting the presence of other prominent non-sensorimotor physical symptoms. While prospective research studies are needed to validate our proposal, we hope that this article catalyses discussion on how to optimally contextualize pain, fatigue and other physical symptoms in patients with FND across diagnostic, treatment and pathophysiology studies.

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Supplementary material

Supplementary material is available at *Brain Communications* online.

Competing interests

D.L.P. has received honoraria for continuing medication education lectures in functional neurological disorder. All other authors do not report any disclosures or conflicts of interest.

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