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EMOTION PROCESSING IN FUNCTIONAL NEUROLOGICAL DISORDER

Section A: A review of psychosocial associates of motor and sensory functional neurological symptoms

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A thesis submitted in partial fulfilment of the requirements of
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Doctor of Clinical Psychology

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SALOMONS
CANTERBURY CHRIST CHURCH UNIVERSITY

DECLARATION FOR MAJOR RESEARCH PROJECT

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Finally, thank you to all of the participants who very generously gave their time to take part in this study.

Summary of the Major Research Project

Section A is a narrative review, based on a systematic search of the literature, of case-control studies exploring psychosocial associates of functional motor and sensory symptoms. Findings in the areas of depressive symptoms, anxiety symptoms, personality traits and pathology, precipitating life events, and childhood trauma are synthesized, and the methodological quality of the studies is evaluated. The findings are discussed with reference to theoretical models and other relevant research, and recommendations for future research and clinical practice are made.

Section B describes an empirical study investigating alexithymia and mentalization in functional neurological disorder (FND). The differences between participants with FND and healthy control participants on a range of self-report measures were examined. Significant between-group differences in alexithymia and hypomentalization were found. Higher scores on measures of alexithymia and mentalization were also predictive of more somatic and neurological symptoms of a generalized nature, across all study participants. Section B concludes with a consideration of limitations of the study, and a discussion of the research and clinical implications of the findings.

Contents

Acknowledgements.....	iii
Summary of the Major Research Project.....	iv
List of Tables and Figures.....	vii
Appendices.....	viii

Section A

Introduction.....	3
Theoretical perspectives.....	4
Aim of this review	9
Methods.....	9
Search strategy	9
Assessment of quality	12
Results.....	12
Depressive symptoms	24
Anxiety symptoms	26
Personality traits and pathology.....	28
Precipitating life events/stressors.....	30
Trauma history	32
Assessment of quality	36
Discussion.....	44
Clinical and research implications	48
Conclusion	50
References.....	51

Section B

Abstract.....	68
Introduction.....	69
Definition and theoretical model	69
Revisiting childhood emotional adversity	73
Aims and hypotheses	76
Methods.....	77
Design	77
Participants.....	78

Materials	79
Procedure	82
Analysis.....	83
Results.....	85
Demographic and clinical characteristics	85
Hypothesis i	87
Hypothesis ii	88
Hypothesis iii	89
Hypothesis iv	89
Hypothesis v.....	93
Discussion	98
Limitations	101
Clinical implications	103
Conclusion	104
References.....	105

Section C

Appendices of supporting materials	118
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List of Tables and Figures

Section A

<i>Table 1.</i> Database search query	10
<i>Table 2.</i> Description and main findings of reviewed studies	13
<i>Table 3.</i> Summary of findings regarding childhood traumas	35
<i>Table 4.</i> Evaluation of methodological quality of the reviewed studies	39
<i>Figure 1.</i> PRISMA flow chart for literature search process	11

Section B

<i>Table 1.</i> Demographic and clinical characteristics of study participants	86
<i>Table 2.</i> Group mean scores on the TAS-20 and TAS-20 subscales, RFQ Certainty about mental states subscale, RFQ Uncertainty about mental states subscale, PHQ-15 checklist, and Neuro checklist	88
<i>Tables 3-10.</i> Logistic regression analyses for predictors of interest predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)	90
<i>Figure 1.</i> Scatter-plots of TAS-20 score as predictor of PHQ15 checklist and Neuro checklist scores	94
<i>Figure 2.</i> Scatter-plots of RFQ Certainty about mental states score as predictor of PHQ15 checklist and Neuro checklist scores	96
<i>Figure 3.</i> Scatter-plots of RFQ Uncertainty about mental states score as predictor of PHQ15 checklist and Neuro checklist scores	97

Appendices

Appendix 1 - Downs and Black (1998) checklist	119
Appendix 2 - Research Ethics Committee (REC) letter of favourable opinion	124
Appendix 3 - Health Research Authority (HRA) letter of approval	129
Appendix 4 - REC letter of favourable opinion for study amendment	138
Appendix 5 - Email confirming NHS Trust R&D approval (1)	141
Appendix 6 - Email confirming NHS Trust R&D approval (2)	142
Appendix 7 - Study questionnaires	144
Appendix 8 - FND group Participant Information Sheet	151
Appendix 9 - FND group consent form	156
Appendix 10 - Control group Participant Information Sheet	158
Appendix 11 - Control group consent form	163
Appendix 12 - Binary logistic regression SPSS outputs	165
Appendix 13 - Author submission guidelines for the Journal of Psychosomatic Research .	172
Appendix 14 - Declaration of end of study	186
Appendix 15 - Final study report for REC and study participants	188
Appendix 16 - Consent procedure for the study	190

Section A: A review of psychosocial associates of motor and sensory functional neurological symptoms

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Abstract

Theoretical models of functional neurological symptoms (FNS) have traditionally assumed that all of the symptoms have the same etiology, as they all resemble symptoms of neurological disorders. Recent research has questioned whether symptoms that are acute and intermittent (e.g. functional seizures) may be different from those that are chronic and continuous (e.g. motor and sensory FNS). This narrative review explored common psychosocial associates of motor and sensory FNS, based on a systematic literature search of case-control studies. The Ovid MEDLINE and PsycINFO databases were searched in November 2017, along with reference lists from relevant papers, and 26 studies were retrieved. Study quality was evaluated using the Downs and Black checklist for case-control studies. Depressive symptoms, alexithymia, and precipitating life events/stressors were the most consistently reported associates of motor/sensory FNS, while the findings regarding anxiety, personality pathology, and childhood trauma were more mixed. However, the sample sizes of reviewed studies were small, and FNS samples were predominantly drawn from specialist services, so the findings may not be generalizable. The results offer some support for a theoretical model differentiating chronic and acute symptom presentations, and also raise questions about the usefulness of categorizing psychological phenomena using medical classification models.

Keywords: Functional neurological symptoms, motor, sensory, psychosocial associates

A review of psychosocial associates of motor and sensory functional neurological symptoms

Introduction

Functional neurological symptoms (FNS) are ‘somatic symptoms which superficially resemble those of organic disorders of the nervous system, but for which no physical explanation can be found’ (Howlett, Grunewald, Khan, & Reuber, 2007, p. 354). Common functional neurological symptoms include functional seizures (also known as psychogenic non-epileptic seizures or dissociative non-epileptic convulsions), sensory symptoms (such as numbness or loss of sensation) and motor symptoms (such as limb weakness or paralysis, gait disturbances, tremors, spasms or tics).

The absence of identifiable organic pathology in FNS has led to greater attention being paid to possible psychological causes of the condition, resulting in FNS occupying a unique position at the intersection of several different clinical specialties, including neurology, neuropsychiatry, epileptology, occupational therapy, and psychology (McKee et al., 2018). There remains, however, a lack of consensus regarding the mechanisms that underpin FNS (Carson et al., 2012). Even the terms used to describe FNS have been the subject of debate and disagreement, with *hysterical*, *dissociative*, *psychogenic*, *psychosomatic*, *somatoform*, *conversion* and *medically unexplained* all used to describe the symptoms (and their putative origins) in different eras and contexts (Edwards, Stone, & Lang, 2014). More recently, *functional* (indicating that the symptoms represent a change with the functioning, rather than the structure, of the brain) has become the preferred term, as it is relatively agnostic about the etiology of symptoms (Edwards, Stone and Lang, 2014), and it is the preferred term of service users and service user advocacy organisations (Stone et al., 2002; Rommelfanger et al., 2017). This term, or FNS, is used throughout this paper.

The lack of consensus regarding the nomenclature and classification of FNS may partly account for the variability in prevalence estimates of FNS, which range from

3.9/100,000 for functional weakness (Stone, Warlow & Sharpe, 2010), to 5/100,000 for functional motor symptoms (Binzer, Anderson, & Kullgren, 1997), and 1.4 – 33/100,000 for functional (non-epileptic) seizures (Reuber, 2008). When all types of ‘conversion disorders’ are considered, prevalence rates of 50/100,000 cases known to health services at any one time, and twice that number over a one-year period, have been reported (Akagi & House, 2002). Individuals with FNS have levels of distress, disability and healthcare usage that are equal to, and often greater than, those of individuals with equivalent organic motor disorders (Carson et al., 2011), and the prognosis is often poor. Reviewing prognostic studies with a mean weighted follow-up duration of 7.4 years, Gelauff, Stone, Edwards, and Carson (2014) found that more than one-third of those with functional motor symptoms had the same or worse symptoms at follow-up. Durrant, Rickards, and Cavanna (2011) reported that fewer than 40% of those diagnosed with functional seizures as adults achieve seizure remission five years after diagnosis, compared to 60-80% of those diagnosed with epileptic seizures.

Theoretical perspectives

Historical and current theoretical models of FNS have typically encompassed all of the FNS symptom presentations, on the assumption that they are etiologically similar, as they all involve pseudoneurological symptoms inconsistent with their neuropathological equivalents (Kanaan, Duncan, Goldstein, Jankovic, & Cavanna, 2017). Early dissociation theories (e.g. Janet, 1889, 1907) proposed that functional symptoms arise from disturbances of the cognitive attentional system, caused by traumatic experiences. Some symptoms (such as a loss of motor function or sensory anesthesia) develop when traumatic experiences lead to heightened attentional focus on some sensory channels, and other channels consequently being neglected. This results in information from the neglected channels being processed outside of conscious awareness (i.e. in a dissociated fashion). Other symptoms (such as

sensory distortions or functional seizures) occur when memories linked to traumatic experiences are not integrated in to the main autobiographical memory store, leading to a loss of conscious control over when they are activated or triggered by external/internal reminders of the memories (Roelofs & Spinhoven, 2007).

Although Janet (1907) viewed dissociation as an abnormal response to traumatic experiences, seen in people with constitutional weakness ('hysterics'), later neodissociation theories (e.g. Hilgard, 1977; Kihlstrom, 1992; Oakley, 1999) demonstrated that dissociation is a normal coping response to trauma, that becomes maladaptive when it is overgeneralized (Brown, 2004). Other subsequent research on the impact of trauma on memory formation (e.g. van der Kolk, 1998) has offered further support for dissociation models. However, the centrality of trauma in dissociation models is problematic, as many people with FNS do not report traumatic experiences prior to their symptom onset (e.g. Roelofs & Pasman, 2017)

The conversion model of FNS (e.g. Breuer & Freud, 1893-1895/1982) had its origins in psychodynamic theory, and proposed that functional symptoms arise from the active repression by the ego of undesirable or emotionally painful thoughts, memories or emotions (Nemiah, 1989). While this repression initially produces a 'primary gain' of protecting a person from painful experiences, preventing these memories from being 'abreacted' (discharged) in a normal way eventually leads to them being 'converted' into somatic symptoms, which were either present at the time of the trauma, or were symbolic representations of it (Roelofs & Spinhoven, 2007). Freud originally believed that childhood sexual abuse was a key risk factor for the subsequent development of conversion as a defence mechanism, though he modified this view in later revisions of the theory, emphasising instead the role of unconscious psychosexual conflict (Halberstadt-Freud, 1996). Conversion models have been criticised due to the emphasis on unconscious processes making them unfalsifiable (Brown, 2004); however, the interest they generated in childhood abuse as a key

precipitant of FNS has been one of their enduring legacies (e.g. Roelofs, Keijsers, Hoogdumin, Näring, & Moene, 2002).

Conversion models of FNS also introduced the concept that there may be ‘secondary gains’ to FNS, such as eliciting positive care and attention from others, or enabling unpleasant tasks (such as work or caring responsibilities) to be avoided (Roelofs & Spinhoven, 2007). Secondary gains were a key area of interest in later social constructionist theories of FNS (Kozłowska, 2005) - Ruesch (1957) suggested that FNS may be an expression of emotions via ‘somatic language’, developmentally the earliest of three forms of human communication (succeeded by action and verbal language). In this model, the function of a symptom, rather than its method of expression, is key to understanding its etiology – for some people, FNS may be a way of communicating distress, used as action or verbal language abilities are underdeveloped; for others, they may be an indirect, symbolic expression of conflict (interpersonal or situational) that a person cannot engage with directly; and for others, they may be an expression of physiological arousal in situations where fear or anxiety has overwhelmed the more sophisticated verbal language system.

Interest in functional symptoms as expressions of emotional distress, rather than of repressed or converted trauma, also led Peter Sifneos (1973) to coin the term ‘alexithymia’ (from the Greek *alexi*, meaning ‘rebuff’, and *thymos*, meaning ‘emotions’) to describe a biological or developmental trait, characterized by difficulties in identifying and describing feelings, externally oriented thinking, and limited imaginal capacity (Taylor, 2000). In the context of FNS, alexithymia has been proposed as a neurobiological mediator between emotional distress and physical symptoms, as well as an explanation as to why some people apparently remain oblivious to their emotional distress. Although some early research findings regarding alexithymia and FNS appeared promising (e.g. Shipko, 1982), subsequent studies of groups of people with specific functional symptoms (e.g. functional seizures) have

suggested that rates of alexithymia are broadly comparable to those of people with other medical conditions (Brown & Reuber, 2016).

The most recent theoretical models of FNS have focused on synthesizing elements of previous theories, using strengths of some models to compensate for shortcomings of others (Brown, 2004). Richard Brown's (2004) integrative conceptual model of medically unexplained symptoms, echoing dissociation theories, is underpinned by a cognitive attentional framework. Drawing on Norman and Shallice's (1986) dual attention model, it suggests that attention is controlled via two cognitive mechanisms: A primary attentional system (PAS), which controls routine actions in a way that feels intuitive and not requiring of conscious effort, as it utilises previously learned mental models (schemata); and a secondary attentional system (SAS), which is responsible for novel or complex actions, for which no mental model is already available, and as a result, feels more effortful and demanding of conscious control (Brown, 2004). Functional symptoms characterized by alterations in experience (positive symptoms, such as pain, nausea, or pseudohallucinations) occur when inappropriate or inconsistent perceptual information is selected during the creation of schemata by the PAS; symptoms characterized by an inability to control perception, cognition, or action, arise when inappropriate cognitive mental models are automatically triggered during the process of schemata selection by the PAS.

Drawing on conversion theories, Brown's model includes a consideration of the impact of early life trauma, by explaining how increased body focus can act as a defense against the overwhelming affect or cognitions associated with trauma. This accounts for why trauma is a common (though not a necessary) predisposing factor in FNS. Elements of social constructionist theories are also incorporated, with the nature of the functional symptoms (e.g. seizures vs motor symptoms) explained as being partly influenced by a person's exposure to similar symptoms in family/friends, or via sociocultural transmission.

Importantly, Brown's model also proposes a mechanism to explain why symptoms may present in an acute or a chronic manner: for intermittent or time-limited phenomena (such as functional seizures), the automatic activation of a schema as an acute response to cues from the internal or external environment (e.g. fear or anxiety) is implicated; whereas for chronic motor or sensory symptoms, a chronic activation and selection of perceptual or behavioural schemata, is responsible (Brown, 2002).

This element of Brown's model, and the experiences of clinicians working with people with FNS, have led to increased questioning of the assumption of etiological equivalence of all functional symptoms, particularly in relation to functional seizures (e.g. Guz et al., 2003; Stone, Sharpe, & Binzer, 2004; Driver-Dunckley, Stonnington, Locke, & Noe, 2011). Functional seizures tend to be intermittent and dramatic, accompanied by alterations of awareness, and often prompt medical intervention, including attempted resuscitation (Hopp, Anderson, Krumholz, Gruber-Baldini, & Shulman, 2012). Motor and sensory symptoms, on the other hand, tend to be less dramatic, but are more likely to involve chronic and persistent symptoms, affecting everyday tasks (Ganos et al., 2014). Based on a review of studies comparing characteristics of people with functional seizures to those of people with functional motor/sensory symptoms, Kanaan et al. (2017) proposed that different psychological mechanisms may be responsible for different functional symptom presentations: Acute symptoms (e.g. functional seizures) may be indicative of a developmental disorder, characterized by people dissociating in response to anxiety in situations which evoke the recall of earlier traumas; Chronic symptoms (e.g. functional motor/sensory symptoms), by contrast, may be an idiosyncratic yet adaptive coping response to recent stressful life events, as their consequence is often to draw in support from others.

Aim of this review

Recent reviews of psychosocial risk factors of FNS (e.g. Brown & Reuber, 2016; Roelofs & Pasman, 2017) have focused primarily on studies of people with functional seizures. However, if the model proposed by Kanaan et al. (2017) is valid, it is unclear if their findings would also apply to people with functional motor or sensory symptoms. The aim of this review was therefore to explore common psychosocial associates of motor/sensory functional symptoms, via a systematic search for published case-control studies. Based on the model proposed by Kanaan et al. (2017), it was hypothesized that functional motor/sensory symptoms would be associated with depressive symptoms and with stressful recent life events, as these functional symptoms reflect an impaired coping response to recent or current stressors. Furthermore, functional motor/sensory symptoms would not show the same strength of association found in studies of people with functional seizures (e.g. Brown & Reuber, 2016) with anxiety, childhood trauma, and personality pathology, as these associates are more indicative of a dissociative response to anxiety, with its origins in developmental trauma.

Methods**Search strategy**

Studies were first identified by searching the Ovid MEDLINE (1946 to present) and PsycINFO (1806 to present) electronic databases on 17 November, 2017, using search terms (listed in Table 1) related to functional motor and sensory symptoms, and to the associates of interest. The search was limited to English language papers only. After removing duplicates, the titles and/or abstracts of the retrieved studies were screened to exclude studies that did not meet inclusion criteria for the review (such as book chapters, review articles or studies of other functional symptoms). The full text of the remaining articles was read to determine whether the inclusion/exclusion criteria were met.

The inclusion criteria were: (i) empirical primary research studies; (ii) studies with a clinical sample of people with functional motor symptoms and/or functional sensory symptoms; (iii) studies with a control group(s) of healthy participants, participants with a psychiatric diagnosis, and/ or participants with an organic neurological disorder; (iv) studies that assessed relevant psychosocial associates by means of a validated self-report measure or a clinical interview (as a primary or secondary measure); and (v) studies of adults. As this was intended as a review of functional motor and sensory symptoms, studies where more than 10% of the functional symptoms sample had a primary diagnosis of functional seizures were excluded. Studies that did not include a description of the symptom profile of the FNS sample, and studies of children/adolescents, were also excluded.

Following the database search, additional studies were identified from the reference lists of included articles, with the same inclusion/exclusion criteria applied. A PRISMA flow diagram (Liberati et al., 2009) of the search process is presented in Figure 1.

Table 1.

Database search query

Search query

- | | |
|----|--|
| #1 | "functional neurologic*" or FND or FNSD or "conversion disorder" or "psychogenic neurologic*" or "non-organic neurologic* disorder" or "unexplained neurologic*" or "somatoform neurologic*" or "functional weakness" or "functional tremor" or "functional sensory" or "functional movement" or "functional dystonia" or "functional gait disturbance" or "functional speech" or "functional swallowing" or "psychogenic weakness" or "psychogenic tremor" or "psychogenic sensory" or "psychogenic movement" or "psychogenic dystonia" or "psychogenic gait disturbance" or "psychogenic speech" or "psychogenic swallowing" |
| #2 | "clinical characteristics" or "personality" or "psychopathology" or "depression" or "anxiety" or "panic" or "mental health" or "psychiatric" or "psychological" or trauma or stress or "life events" or "childhood adversity" or abuse or neglect |
| #3 | #1 AND #2 |

Limit to English language.

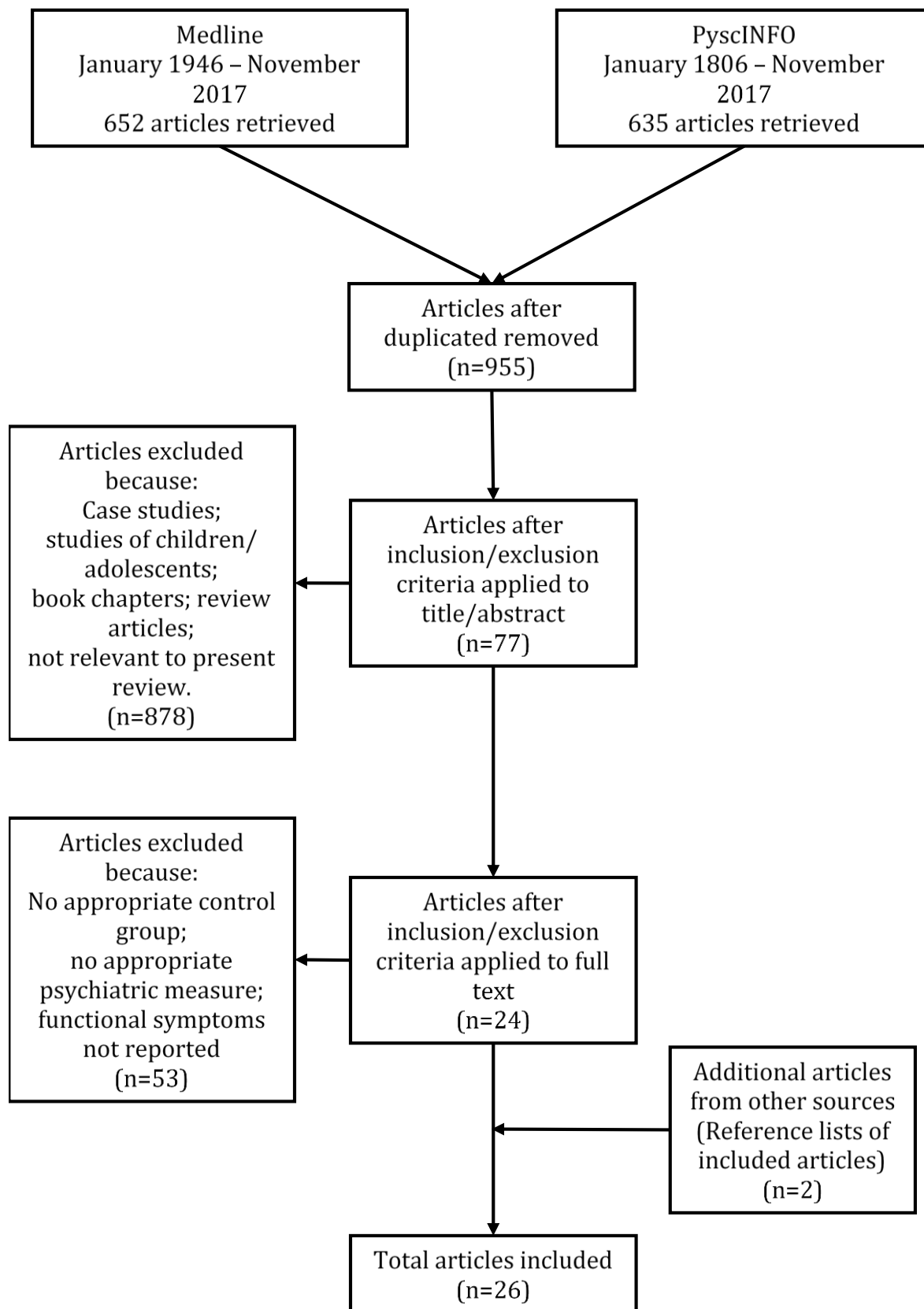


Figure 1. PRISMA flow chart for literature search process

Assessment of quality

The reviewed studies were evaluated using the 17 items relevant to case-control studies (Sanderson, Tatt & Higgins, 2007) of the Downs and Black (1998) checklist for assessing the methodological quality of randomised and non-randomised studies (Appendix 1). Checklist item 27 was simplified (as per MacLehose et al., 2000) to the question, “Was a power calculation reported for the primary outcome?”. Checklist items were scored as 0 (item criteria not fulfilled) or 1 (item criteria fulfilled), with the exception of item five (regarding confounding variables), which was scored as 0 (not fulfilled), 1 (partially fulfilled) or 2 (completely fulfilled). Quality scores therefore ranged from 0-18. As checklist items are not weighted by their relative risk to study quality (i.e. a lower score may not necessarily indicate lower study quality), the methodological quality of studies was also reviewed narratively.

Results

The search generated 24 papers, and two additional papers (Demartini et al., 2014; Fiess, Rockstroh, Schmidt, and Steffen, 2015) were identified from the references lists of included studies. The studies’ locations, participants, measures and key findings are described in Table 2. Several studies contained more than one control group, and the results of individual between-group comparisons are reported separately in the review. The main results in each of the five areas of interest – depressive symptoms, anxiety symptoms, personality traits and pathology, precipitating life events/stressors, and childhood trauma – are presented first. As many of the same methodological quality issues applied across the studies (and across the findings within them), these are then discussed in a separate section, with reference to specific findings, where applicable.

Table 2.

Description and main findings of reviewed studies

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Binzer, Andersen & Kullgren (1997)	30 participants with recent onset conversion disorder with motor symptoms	30 participants with recent onset organic motor symptoms	Umea and Kalmar, Sweden	Structured Clinical Interview for DSM-IV Axis I disorders (SCID-I): Major depression[†] Hamilton Rating Scale for Depression (HRSD) (mean)***	Not assessed.	Structured Clinical Interview for DSM-IV Axis II disorders (SCID-II): Any personality disorder[†]	5-item Life Events Inventory: Life events in 3 months* before symptom onset (mean); Life events in 12 months* before symptom onset (mean).	Not assessed	11

Measures where a significant difference between the clinical sample and the control sample was reported are highlighted in **bold**.

* Mean/median scores of clinical sample significantly higher than control sample, $p \leq 0.05$; ** Mean/median scores of clinical sample significantly higher than control sample, $p \leq 0.01$; *** Mean/median scores of clinical sample significantly higher than control sample, $p \leq 0.001$; [†] Diagnosis reported in significantly higher proportion of clinical sample than control sample, $p \leq 0.05$; ^ Other significant difference(s) between clinical sample and control sample - see narrative review for further information.

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Binzer & Eisemann (1998)	30 participants with recent onset conversion disorder with motor symptoms	30 participants with recent onset organic motor symptoms	Umea and Kalmar, Sweden	Not assessed	Not assessed	SCID-II: Any personality disorder † N of interview criteria fulfilled for Cluster A* disorders, Cluster B* disorders, Cluster C disorders, and Global Index* (mean)	Not assessed	Egna Minnen Beträffande Uppfostran (EBMU) (Own memories of childrearing experiences) ^ Clinical interview for self-reported childhood sexual abuse	9
Willinger & Aschauer (2005)	61 participants with functional dysphonia	61 healthy control participants	Vienna, Austria	Beck Depression Inventory (BDI), German version (mean)*	State-Trait Anxiety Inventory (STAI), German translation: STAI-State (mean) ns; STAI-Trait (mean)* . Interaction Anxiety Questionnaire: 'Illness anxiety' factor (mean)* ; 'Anxiety of social situations' factor (mean) ns.	Freiburg Personality Inventory: 'Extraversion versus introversion' subscale (mean)*	Not assessed	Not assessed	12

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Seignourel et al. (2007)	12 participants with psychogenic movement disorder (PMD)	12 healthy control participants	Florida, USA	BDI (mean)**	STAI - State (mean)** STAI - Trait (mean)**	Not assessed	Not assessed	Not assessed	12
Anderson et al. (2007)	41 participants with psychogenic movement disorder (PMD)	499 participants with Parkinson's disease (PD)	Maryland, USA	Brief Symptom Inventory (BSI-18), depression scale (mean)***	Brief Symptom Inventory (BSI-18), anxiety scale (mean)***	Not assessed	Not assessed	Not assessed	11
Espirito-Santo & Pio-Abreu (2009)	26 participants with DSM-IV conversion disorder (primarily motor and sensorial);	39 participants with DSM-IV dissociative disorders; 40 participants with DSM-IV somatization disorder; 46 participants with other DSM-IV psychiatric disorders.	Unspecified, Portugal	BSI depression scale (mean)	BSI anxiety, phobic anxiety and obsessive compulsive scales (mean)	Not assessed	Not assessed	Not assessed	14

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Voon et al. (2010)	16 participants with motor conversion disorder	16 healthy control participants	Bethesda, MD, USA (clinical sample); Unspecified USA (control sample)	BDI (mean)***	Beck Anxiety Inventory (BAI) (mean)***	Not assessed	Not assessed	Not assessed	11
Stone, Warlow & Sharpe (2010)	107 participants with functional weakness	46 participants with weakness attributable to neurological disease	South-East Scotland, UK	SCID-I[^] Hospital Anxiety and Depression Scale (HADS), Depression scale ¹ : Scoring above clinical cut-off†; Group median scores ns.	SCID-I[^] HADS anxiety scale ¹ : Scoring above clinical cut-off ns; Group median scores ns.	Not assessed	Not assessed	Not assessed	15

¹ Measures completed by 96/107 of the clinical sample and 40/46 of the control sample

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Kranick et al. (2011)	64 participants with psychogenic movement disorder (PMD) cases	39 participants with focal hand dystonia (FHD) controls; 39 healthy control participants	Bethesda, MD, USA (clinical and FHD samples); unspecified USA (healthy control sample)	SCID-I (PMD and FHD groups only): Major depression (lifetime) ns. BDI (mean) ***	SCID-I (PMD and FHD groups only): GAD, phobia and panic disorder ns. BAI (mean) ***	Revised Neuroticism-Extroversion-Openness Personality Inventory (NEO PI-R) (PMD & HC groups only): Conscientiousness domain (mean)* ; All other domains (means) ns.	Social Readjustment Scale (PMD & FHD groups only)	Childhood Trauma Questionnaire (CTQ)^ Traumatic Life Events Questionnaire (TLEQ)^ Parental Bonding Instrument (PBI)^	12
Parees et al. (2012)	18 participants with functional movement disorder (FMD)	18 healthy control participants	London, UK	HADS depression scale (mean) ***	HADS anxiety Scale (mean)***	Not assessed	Not assessed	Not assessed	11
Voon et al. (2013)	30 participants with psychogenic movement disorder and conversion disorder	30 healthy control participants	Bethesda, MD, USA (clinical sample); Unspecified USA (control sample)	BDI (mean)***	BAI (mean)***	Not assessed	Not assessed	Not assessed	12

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
DeMartini et al. (2014)	55 participants with functional motor symptoms (FMS)	33 participants with organic movement disorder (OMD) 35 healthy controls	London, UK	Montgomery-Asberg Depression Rating Scale (MADRS) (mean)**	Not assessed	SCID-II: OCPD †, all other PDs ns. Toronto Alexithymia Scale (TAS-20) (mean)**	Not assessed	Not assessed	11
Brown, Nicholson, Aybek, Kanaan, & David (2014)	21 participants with motor conversion disorder (inpatient and outpatient)	36 healthy control participants	South-East England, UK	HADS depression scale (mean)*	HADS anxiety scale (mean)*	Not assessed	Not assessed	Not assessed	11
Aybek et al. (2014)	12 participants with DSM-IV sensory / motor conversion disorder	13 healthy control participants	South-East London, UK	HADS depression scale (mean)*	HADS anxiety scale (mean)	Not assessed	Not assessed	Not assessed	12

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
van der Hoeven et al. (2015)	55 participants with functional movement disorder (FMD)	34 participants with organic movement disorder (OMD) 52 healthy control participants	Groningen, The Netherlands	Not assessed	Not assessed	Personality Diagnostic Questionnaire (PDQ-4)	Not assessed	Not assessed	13
Fiess, Rockstroh, Schmidt, & Steffen (2015)	20 participants with ICD-10 cases with ICD-10 dissociative disorders (motor, sensory, or mixed excluding functional seizures;)	20 healthy control participants	Konstanz, Germany	Not assessed	Not assessed	Toronto Alexithymia Scale, German version (TAS-26) (median)***	Not assessed	Not assessed	12

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Steffen, Fiess, Schmidt & Rockstroh (2015)	45 participants with motor and/or sensitivity dissociative disorders	45 healthy control participants	Konstanz, Germany	Not assessed.	Not assessed.	TAS-26 (mean)***	Life Events Questionnaire (LEQ): Negative life events in last 12 months (mean)***; Positive life events in last 12 months (mean) ns.	Early Trauma Inventory, German version (ETI) (all median scores): General trauma**; Emotional trauma***; Physical trauma ns; Sexual abuse ns.	12
Aybek et al. (2015)	12 participants with DSM-IV motor conversion disorder	14 healthy control participants	South-East London, UK	HADS depression scale: Scoring above clinical cut-off†; Group mean scores ns.	HADS anxiety scale: Scoring above clinical cut-off ns; Group mean scores ns.	Not assessed	Not assessed	Clinical interview for self-reported history of sexual abuse	12
Maurer et al. (2016a)	35 participants with functional movement disorder (FMD)	35 healthy control participants	Bethesda, MD, USA (clinical sample); Unspecified USA (control sample)	HRSD (mean)*** BDI (mean)***	Hamilton Rating Scale for Anxiety (HAM-A) (mean)***	Not assessed	Not assessed	CTQ (all scales)	11

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Maurer et al. (2016b)	35 participants with functional movement disorder (FMD)	38 healthy control participants	Bethesda, MD, USA (clinical sample); Unspecified USA (control sample)	HRSD (mean)*** BDI (mean)**	HAM-A (mean)***	Not assessed	Not assessed	Not assessed	11
Fiess, Rockstroh, Schmidt, Wienbruch, & Steffen (2016)	21 participants with ICD-10 dissociative disorders (motor, sensory, or mixed excluding functional seizures;)	21 healthy control participants	Konstanz, Germany	Symptom Check List Revised, German version (SCL-R-90): Depression scale (median)***	Not assessed	Not assessed	Not assessed	ETI, German version (median)***	12
Nicholson et al. (2016)	43 participants with motor conversion disorder	28 participants with psychiatric diagnosis (depression) 28 healthy control participants	London, UK	(Cases vs HC) HADS depression scale (mean)*	(Cases vs HC) HADS anxiety scale (mean)	Not assessed	Life Events and Difficulties Scale (LEDS)^	Clinical interview^	10

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Tomic et al. (2017)	39 participants with functional dystonia (DysF)	30 participants with idiopathic (primary) dystonia (DysP)	Belgrade, Serbia	HRSD (mean) Psychiatric interview [^]	HAM-A (mean) Psychiatric interview	NEO PI-R (all mean scores): Extraversion* ; Openness to experience* ; All other domains ns. Psychiatric interview	Psychiatric interview: presence of stressor before onset of symptoms [†]	Not assessed	10
Apazoglou, Mazzola, Wegrzyk, Polara & Aybek (2017)	16 participants with motor functional neurological disorder (motor FND)	15 healthy control participants	Geneva, Switzerland	BDI (mean)*	STAI - State scale (mean) STAI - Trait scale (mean)	Not assessed	Amiel-Lebigre questionnaire: N of sig. life events (last 5 yrs) (mean) Impact of sig. life events (last 5 yrs) (mean)	CTQ (mean scale scores): Sexual abuse* ; all other domains ns.	10

<u>Study</u>	<u>Clinical sample</u>	<u>Control sample</u>	<u>Location</u>	<u>Depression measure and findings</u>	<u>Anxiety measure and findings</u>	<u>Personality measure and findings</u>	<u>Life events measure and findings</u>	<u>Trauma measure and findings</u>	<u>Quality score</u>
Kienle et al. (2017)	60 participants with ICD-10 dissociative disorders (motor, sensory, or mixed excluding functional seizures)	39 participants with diagnosis of PTSD 40 healthy control participants	Konstanz & Gailingen, Germany (clinical sample); Mannheim, Germany (PTSD control sample); unspecified Germany (healthy control sample)	Not assessed	Not assessed	TAS-26 (median)***	Not assessed	Post-traumatic Stress Diagnostic Scale (PDS)^ German version of Maltreatment and Abuse Chronology of Exposure (KERF)^	12
Ekanayake et al. (2017)	59 participants with psychogenic movement disorder	26 healthy control participants (43 control participants with psychogenic non-epileptic seizures)	Maryland, USA (clinical sample); unspecified USA (control sample)	BDI-II (mean)**	Not assessed.	NEO PI-R: All five domains ns. Neuroticism facet of 'Depression'*	Not assessed	CTQ (mean scale scores): Emotional abuse* ; all other domains ns. TLEQ: Age when experienced most traumatic event.	12

Depressive symptoms

Of the 26 reviewed studies, 21 assessed depressive symptomology. Sixteen studies compared groups of participants with functional motor/sensory symptoms to healthy control participants, using validated self-report measures, including the Beck Depression Inventory (BDI; Beck & Steer, 1987; Beck, Steer, & Brown, 1996), the depression scale of the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983), the Hamilton Rating Scale for Depression (HRSD; Hamilton, 1960), the depression subscale of the Brief Symptom Inventory (BSI; Zabora et al., 2001), the Montgomery-Asberg Depression Rating Scale (MADRS; Davidson, Turnbull, Strickland, Miller, & Graves, 1986), and the Symptom Check List Revised (SCL-R-90; Derogitas & Unger, 2010). Fifteen (94%) of these studies found that the mean or median depression scores of the functional symptoms sample were significantly higher than those of healthy controls. One study (Aybek et al., 2015) did not find a significant difference in mean/median scores, but it did find that a significantly greater proportion of the functional symptoms sample scored above the established measure threshold for a diagnosis of ‘clinical depression’.

Six studies made comparisons to control samples of people with organic neurological disorders, and four (67%) reported significantly higher scores in the functional symptoms sample on self-report measures of depressive symptoms. A fifth study (Tomic et al., 2017) also reported higher mean scores, but the difference was just short of statistical significance ($p = 0.072$). The sixth study (Stone, Warlow, & Sharp, 2010) did not find a significant difference in median scores, but did find that a significantly higher proportion of the functional symptoms sample scored above the clinical threshold for ‘caseness’.

Four of the six studies with organic disorder control samples also assessed on categorical depression diagnoses. Three studies used the clinician-rated Structured Clinical Interview for DSM-IV Axis I disorders (SCID-I; First, Spitzer, Gibbon, & Williams, 1995).

Binzer et al. (1997) found that a significantly higher proportion of their functional symptoms sample met criteria for a current major depressive episode (corresponding with the findings from their self-report measure). Stone et al. (2010) also found that a significantly higher proportion of their functional weakness sample met the diagnostic criteria for a current major depressive disorder (corresponding with their findings regarding ‘caseness’, but not median scores, on a self-report measure). Kranick et al. (2011) found no significant between-group differences in categorical diagnoses of depression in their sample (contrasting with the finding of their self-report measure). The fourth study, Tomic et al. (2017) used a psychiatric interview to assess categorical diagnoses – they found that a significantly higher proportion of the functional symptoms sample had experienced a major depressive episode prior to developing functional symptoms, and also that a higher proportion of the functional symptoms sample met criteria for a current diagnosis of recurrent major depressive disorder, though this difference was marginally below statistical significance (19.4% vs 3.3%; $p=0.063$). There were no significant between-group differences on prior dysthymia, current major depressive episode, or current dysthymia, diagnoses.

Espirito-Santo and Pio-Abreu (2009) was the only reviewed study to evaluate depressive symptomology with control groups of people with other psychiatric diagnoses. They found no significant between-group differences in mean scores on the self-report BSI depression scale.

In summary, of the 21 studies that included a measure of depression, 16 (76%) found evidence of significantly higher depressive symptomology in the functional symptoms sample than in matched control groups (13/14 studies with healthy control samples, 2/4 studies with organic disorder control samples, and 1/2 studies with healthy and organic disorder control samples). Four (19%) of the reviewed studies (1/14 studies with healthy controls samples, 2/4 studies with organic disorders control samples, and 1/2 studies with

healthy and organic disorder control samples) reported a divergence in findings between multiple measures, with a least one measure finding significant differences, and one measure finding no significant differences. The study with a control group of participants with psychiatric disorder diagnoses was the only reviewed study that did not find any evidence of significantly higher depressive symptomology in the functional symptoms sample.

Anxiety symptoms

Seventeen of the reviewed studies assessed anxiety symptoms. Thirteen studies compared aspects of anxiety between functional symptoms samples and healthy control samples. Ten of these studies used a global self-report measure of anxiety, such as the anxiety subscale of the HADS (Zigmond & Snaith, 1983), the Hamilton Anxiety Rating Scale (HARS; Hamilton, 1959) or the Beck Anxiety Inventory (BAI; Beck, Epstein, Brown, & Steer, 1988). Seven (70%) of the ten studies found significantly higher mean or median anxiety scores in the functional symptoms samples, with the remaining three studies finding no significant differences.

The other three studies with healthy control samples used the State-Trait Anxiety Inventory (STAI; Spielberger, Gorsuch, & Lushene, 1970), which assesses anxiety as a reactive state (state anxiety), and as a stable personality characteristic (trait anxiety). These studies reported conflicting findings: Apazoglou, Mazzola, Wegrzyk, Polara and Aybek (2017) found no significant between-group differences on state or trait anxiety; Seignourel et al. (2007) found the functional symptoms sample had significantly higher mean scores on both the state and the trait scales; while Willinger and Aschauer (2005) found the functional symptoms sample had significantly higher mean scores on the trait scale, but not on the state scale. Willinger and Aschauer (2005) also found that their functional dysphonia sample had

significantly higher scores on the illness anxiety scale, but not on the social anxiety scale, of the Interaction-Anxiety Questionnaire (Becker, 1997).

Four studies made comparisons to control samples of people organic disorders using self-report measures – two of these studies found significantly higher mean/median anxiety scores in the functional symptoms samples, and two studies found no significant differences. Three of these studies also used clinician-rated categorical measure of anxiety, again reporting mixed results. Using the SCID-I, Stone et al. (2010) found that a significantly higher proportion of their functional weakness sample met the diagnostic criteria for panic disorder, somatization disorder and generalized anxiety disorder, but no significant between-group differences on agoraphobia, hypochondriasis, OCD or social phobia. Using the same measure, Kranick et al. (2011) found no significant between-group differences on diagnoses of generalized anxiety disorder, phobias, or panic disorder. Tomic et al. (2017) used a psychiatric interview to evaluate categorical anxiety diagnoses, and also found no significant between-group differences.

Espirito-Santo & Pio-Abreu (2009) was the only study to explore anxiety in control samples with psychiatric disorder diagnoses. They found no significant between-groups differences on the anxiety scale of the BSI.

In summary, of the 17 studies that assessed anxiety, eight studies (47%) found evidence of significantly higher anxiety in groups of people with functional symptoms than in control groups (7/12 studies with healthy control groups, and 1/3 studies with organic disorders control groups); six studies (35%) found no evidence of significantly higher anxiety in functional symptoms samples compared to control groups (4/12 studies with healthy control groups, 1/3 studies with organic disorders control groups, and the one study with control groups of people with psychiatric diagnoses); and the remaining three studies (18%) reported different findings according to different measures (1/12 studies with healthy control

groups, 1/3 studies with organic disorders control groups, and the one study with healthy control and organic disorders control groups).

Personality traits and pathology

Eleven of the reviewed studies assessed aspects of personality. Four studies assessed personality disorder diagnoses. As assessed by the clinician-rated Structured Clinical Interview for the DSM-IV Axis II disorders (SCID-II; First, Spitzer, Gibbons, Williams, & Benjamin, 1996), Binzer et al. (1997) found that a significantly higher proportion of the functional symptoms sample met the criteria for a personality disorder diagnosis, though there were no significant group differences on any specific personality disorder diagnoses. In subsequent analyses of the same data, Binzer and Eisemann (1998) reported that the functional symptoms sample met significantly more of the interview criteria for Cluster A (odd, bizarre or eccentric) and Cluster B (dramatic, erratic) disorders, but not Cluster C (anxious, fearful) disorders, than the organic disorders sample.

These findings regarding personality psychopathology were not replicated in other studies, however. DeMartini et al. (2014), using the SCID-II with samples with functional symptoms, organic motor disorders, and healthy controls, found no significant between-group differences on any personality disorder, with the exception of a significantly higher proportion of the functional symptoms sample meeting criteria for obsessive-compulsive personality disorder (OCPD), a Cluster C disorder. van der Hoeven et al. (2015) used a self-report measure, the Personality Diagnostic Questionnaire (PDQ-4; Hyler, Skodol, Kellman, Oldham, & Rosnick, 1990), and found no significant differences between the proportions of the samples with functional symptoms, organic disorders, and healthy control participants, scoring above the clinical cut-off for personality pathology. Tomic et al. (2017) also found no significant differences between their samples with functional dystonia and organic

dystonia, on diagnoses of histrionic, borderline, or undifferentiated personality disorders (as assessed by psychiatric interview).

Three of the reviewed studies assessed personality using the Neuroticism-Extroversion-Openness Personality Inventory Revised (NEO PI-R; Costa & McCrae, 1985), a self-report measure of five dimensional domains (Neuroticism, Conscientiousness, Extraversion, Agreeableness and Openness). There were no consistent findings across these studies: Kranick et al. (2011) found that a functional symptoms sample had significantly lower mean scores than a healthy controls sample on the Conscientiousness domain, but no significant differences on the other four domains; Ekanyake et al. (2017) found no significant differences between a functional symptoms sample and a healthy controls sample on any of the five domains; and Tomic et al. (2017) found that a functional symptoms sample had significantly lower scores than an organic disorders control sample on the 'Extroversion' and 'Openness' domains, but not on the other domains. Willinger and Aschauer (2005) also assessed extroversion as a dimensional aspect of personality, using "extraversion versus introversion" scale of the Freiburg Personality Inventory (Fahrenberg, Hampel, & Selg, 1994). They found that a functional symptoms sample had significantly lower levels of extroversion, than a healthy controls sample.

Four of the reviewed studies assessed alexithymia using the Toronto Alexithymia Scale (TAS-20; Bagby, Taylor, & Parker, 1994) or the German version of the scale, the TAS-26 (Kupfer, Brosig, & Brähler, 2001). Each of the three studies that compared samples with functional symptoms to healthy control samples found that the functional symptoms samples had significantly higher mean/median alexithymia scores, as did the study that made comparisons to an organic disorders control sample. Kienle et al. (2017) also compared the TAS-26 scores of a functional symptoms sample to a sample of people diagnosed with post-

traumatic stress disorder (PTSD) – they found that the functional symptoms sample had significantly lower median alexithymia scores.

In summary, one of the reviewed studies found significantly higher rates of personality disorder diagnoses in a functional symptoms sample, when compared to an organic disorders control sample, but two studies (one with an organic disorders control sample, and one with healthy and organic disorders control samples) found no significant differences. A fourth study, also with healthy control and organic disorders control samples, found significantly higher rates of obsessive-compulsive personality disorder diagnoses in the functional symptoms sample, but not other personality disorder diagnoses. There did not appear to be any consistent findings across the four reviewed studies that used dimensional measures of personality. On the other hand, all of the reviewed studies that assessed alexithymia found evidence of significantly higher alexithymic traits in people with functional symptoms (three studies with healthy control groups, and one study with an organic disorders control group), with the exception of one study that made comparisons to a control group of participants with PTSD.

Precipitating life events/stressors

Six of the reviewed studies included a measure of significant recent life events / stressors. Three of these studies made comparisons to samples of healthy controls participants. Steffen et al. (2015) found that the functional symptoms sample had experienced significantly more negative life events, but not positive life events, in the preceding 12 months, as assessed by the Life Events Questionnaire (LEQ; Norbeck, 1984). Negative life events were also a significant predictor of the severity of functional symptoms in a multiple logistic regression model. Nicholson et al. (2016) found that people with functional symptoms were significantly more likely to have experienced at least one severe life event in

the month before symptom onset, and/or at least one stressor where the development of functional symptoms would have secondary gains (i.e. would enable 'escape' from a stressor), as assessed by clinician-rated Life Events and Difficulties Schedule (LEDS; Brown and Harris, 1989). The mean number of severe events, and those linked to potential secondary gains, in the month preceding symptom onset, was also significantly higher in the functional symptoms sample. Apazoglu et al. (2017) found no significant between-group differences in the number of significant life events, and the subjective impact of those life events, in the preceding five years, as assessed by the Amiel-Lebigre questionnaire (Cottraux, Bouvard, & Légeron, 1985).

Three studies explored precipitating life events with control samples of people with organic disorders. Binzer et al. (1997), using an unvalidated measure (Perris, 1984) of significant life events in five areas (work, family life, economy, illness or death amongst family/friends, and personal illness), found that the functional symptoms sample reported a significantly higher mean number of significant life events in the 4-month and 12-month periods prior to symptom onset. As assessed by psychiatric interview, Tomic et al. (2017) found that a functional symptoms sample had significantly more frequent stressors preceding the onset of their symptoms. Experiencing stress before symptom onset also independently predicted having functional (versus organic) symptoms, in a binary logistic model. Kranick et al. (2011), however, found no significant between-group differences in number of stressors (and their impact) in the year prior to symptom onset, as assessed by the Social Readjustment Scale (Holmes & Rahe, 1967).

Finally, Nicholson et al. (2016) explored precipitating life events in a functional symptoms sample, and a control sample of people with a diagnosis of depression, using the LEDS. They found that people with functional symptoms were significantly more likely than people with a diagnosis of depression to have experienced at least one severe life event in the

month before symptom onset, and/or at least one stressor where the development of functional symptoms would have secondary gains. They also found that the mean number of severe events, and of events linked to potential secondary gains, in this month was significantly higher in the functional symptoms sample.

In summary, five of the seven studies (2/3 studies with healthy control samples, 2/3 studies with organic disorders control samples, and the one study with a control group of people with depression) found evidence of higher rates of significant life events / stressors preceding symptom onset in people with functional symptoms, than in control group participants.

Trauma history

Ten of the reviewed studies (with nine healthy control samples, two organic disorders control samples, and two psychiatric disorders control samples) assessed trauma history (childhood and/or lifetime), using clinician interview or validated self-report measures, such as the Childhood Trauma Questionnaire (CTQ; Bernstein & Fink, 1998), the Early Trauma Inventory (ETI; Wingenfeld et al., 2011), the Maltreatment and Abuse Chronology of Exposure scale (KERF; Isele et al., 2014), or the Traumatic Life Events Questionnaire (TLEQ; Kubany et al., 2000). As many of these measures assess trauma across similar domains (sexual abuse, physical abuse/neglect, and emotional abuse/neglect), the key findings are summarised in Table 3.

There were also some other findings of note regarding trauma. Although no significant between-group differences in sexual abuse were found by Steffen et al. (2015), they did find that individuals with multiple functional symptoms significantly more frequently reported sexual abuse, compared to those with single-domain functional symptoms.

Several studies also included measures of trauma across the lifespan. In addition to their findings regarding childhood trauma, Kienle et al. (2017) found that their functional symptoms sample had experienced more traumatic events across their lifetime than healthy control participants, as assessed by the PDS. Two studies used elements of the Traumatic Life Events Questionnaire (TLEQ; Kubany et al., 2000): Kranick et al. (2011) found that a functional symptoms sample reported a significantly more lifetime traumatic episodes, and significantly greater fear associated with traumatic events, than control groups of healthy participants or participants with organic disorders. The functional symptoms sample also reported a significantly greater number of traumatic events than people in the control groups (though this difference was no longer significant after correcting for multiple comparisons). Ekanyake et al. (2017) used the item from the TLEQ regarding the timing of the most traumatic event occurring – they found no significant differences between a functional symptoms sample and a healthy controls sample on the mean age at which people experienced their most traumatic event.

Finally, two studies looked at memories of parenting experiences in their samples. Binzer and Eisemann (1998) found that participants with functional symptoms recalled significantly less emotional warmth, and significantly more rejection, from both parents, than control participants with organic disorders, as assessed by the self-report Own Memories of Childrearing Experiences questionnaire (Perris, Jacobsson, Linndström, Knorrning, & Perris, 1980). Kranick et al. (2011), using the Parental Bonding Inventory (Parker, Tupling, & Brown, 1979), also found that a functional symptoms sample reported significantly lower perceived paternal and maternal parental care than control groups of healthy participants and participants with organic disorders (though the differences no longer met the level of statistical significance after correcting for multiple comparisons). Neither of these studies found significant between-group differences on experiences of parental overprotection.

In summary, eight (80%) of the 10 studies that assessed childhood trauma found some evidence of higher rates of childhood trauma in functional symptoms samples than in control samples (4/6 studies with healthy control groups, both of the studies with healthy and psychiatric diagnosis control groups, the one study with healthy and organic disorders control groups, and the one study with a control group of people with organic disorders). However, there was little consensus amongst the studies on which types of childhood trauma were most common, though emotional abuse was more commonly reported than sexual or physical abuse.

Table 3.

Summary of findings regarding childhood traumas

			Significantly higher rates reported in functional symptoms sample?					Overall childhood adversity
	Control group participants	Measure	Sexual abuse	Physical abuse	Physical neglect	Emotional abuse	Emotional neglect	
Kranick et al. (2011)	Healthy	CTQ	No	Yes ¹	Yes	Yes	No	N/A
Aybek et al. (2015)	Healthy	Clinical interview	No	N/A	N/A	N/A	N/A	N/A
Steffen et al. (2015)	Healthy	ETI	No		Yes		Yes	Yes
Fiess et al. (2016)	Healthy	ETI	N/A	N/A	N/A	N/A	N/A	Yes
Maurer et al. (2016)	Healthy	CTQ	No	No	No	No	No	N/A
Nicholson et al. (2016)	Healthy	Clinical interview	Yes	No	N/A	N/A	N/A	N/A
Apazoglou et al. (2017)	Healthy	CTQ	Yes	No	No	No	No	N/A
Ekanayake et al. (2017)	Healthy	CTQ	No	No	No	Yes	No	N/A
Kienle et al. (2017)	Healthy	KERF	No		Yes		Yes	N/A
Binzer & Eisemann (1998)	Organic disorders	Clinical interview	No	N/A	N/A	N/A	N/A	N/A
Kranick et al. (2011)	Organic disorders	CTQ	No	Yes ¹	Yes	Yes	No	N/A
Nicholson et al. (2016)	Psychiatric disorders (depression) ²	Clinical interview	No	No	N/A	N/A	N/A	N/A
Kienle et al. (2017)	Psychiatric disorders (PTSD)	KERF	No	No	No	No	No	No
Total number of significant findings			2/12	4/10	4/10	5/8	2/8	2/3

¹ Difference no longer statistically significant after controlling for multiple comparisons² Female participants only.

Assessment of quality

The quality of the reviewed studies was moderate, with study quality scores on the Downs and Black (1998) checklist ranging from 7-15 (mean 11.4). Further details of the quality assessment scoring are provided in Table 4.

A number of general and specific methodological issues warrant consideration. Firstly, none of the studies fully met the evaluation criteria regarding the representativeness of the samples, as all of the functional symptoms samples were recruited from specialist services (such as inpatient or outpatient neurology services, neurological rehabilitation services, or neuropsychiatry services). Many individuals with functional symptoms will be referred on to specialist services, but people with acute and/or transient symptoms, more likely to be seen in primary care, by community mental health teams, or not at all (Akagi and House, 2002), may be underrepresented in this research.

Secondly, sample sizes of the groups of participants with functional symptoms tended to be small ($M = 37$, $Mdn = 32.5$). Based on Cohen's (1992) widely-used criteria, none of the studies had sufficient power to detect small or medium effects, 17 (65%) studies had adequate power to detect large effects only, and the remaining nine studies were only powered to detect very large effects. Although small sample sizes are not unexpected due to the relatively low incidence of functional symptoms, there is an increased risk of Type II errors, due to studies being underpowered. Many of the studies also did not control for making multiple comparisons (increasing the risk of Type I errors). When such corrections were made, some significant findings were no longer statistically significant.

Thirdly, several of the reviewed studies included in the review also took place at the same research centres (see 'Location' column in Table 2). Findings do not appear to have been reported repeatedly across multiple studies, but the possible use of the same samples in multiple studies is a potential confounder of the findings of this review.

With regards to the findings regarding depressive symptoms, many of the reviewed studies excluded participants with a diagnosis of depression from their healthy control groups. As a result, depressive symptoms may have been less common in these control groups than would normally be found in the general population. However, the findings from these studies were broadly consistent with studies that did not exclude control group participants on this basis. Furthermore, Voon et al. (2013) excluded participants with current major depression of moderate severity from the functional symptoms sample, and still found that the functional symptoms sample had significantly higher mean BDI scores than the healthy controls sample.

Regarding the findings on personality disorders, the only study (Binzer and Eisemann, 1998) to find significantly higher rates of personality disorder diagnoses in a functional symptoms sample was also the only study to use exclusively in-patient participants. In order to reach a threshold for inpatient admission, these participants may have had more severe difficulties than average, and none of the other studies that measured personality psychopathology reported a prevalence rate even near the 50% reported in this sample. The functional symptoms sample also had significantly lower educational attainment than control participants, which may have been another significant confounding factor, as there is evidence that lower educational attainment is a predictor of personality disorder diagnoses (e.g. Huang et al., 2009).

A number of other methodological issues are also worth noting. The use of unvalidated measures, particularly clinical interviews, may have affected the reliability of some of the reported study results. There have also been queries raised regarding the validity and reliability of retrospective self-reports of trauma histories (Hardt & Rutter, 2004). However, the use of the same measures with the clinical and control groups negates the impact of these limitations somewhat. In addition, although studies that assessed depressive

and anxiety symptoms utilized the same measures with the clinical and control groups, there is a risk that between-group differences may have arisen from spurious associations between measure items assessing physical symptoms of depression/anxiety, and the physical manifestations of the functional motor/sensory symptoms themselves. Additional factor analyses of the types of depressive/anxiety symptoms being reported may be valuable in exploring this issue further.

Table 4.

Evaluation of methodological quality of the reviewed studies, based on Downs and Black (1998) checklist

Study	Downs and Black Checklist Item																											Total (max. 18)	Notes
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27*		
Binzer, Andersen & Kullgren (1997)	1	1	1		2	1	1			0	0	0			0	0		1		0	1	1			0		0	10	* Unvalidated Life Events Inventory used * Hypotheses not stated in introduction * Cases were all inpatients
Binzer & Eisemann (1998)	0	1	1		2	1	1			0	0	0			0	0		1		0	0	0			0		0	7	* Source (place/time) of control group not described. * Analyses by gender, not mentioned in original hypotheses, undertaken * Cases were all inpatients - more severe? * Only (unvalidated) clinical interview used to assess childhood sexual abuse
Willinger & Aschauer (2005)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			1		0	12	* Dysphonia cases were recruited consecutively - info not provided re controls * Cases recruited from specialist service; controls recruited from schools and acquaintances of students
Anderson et al. (2007)	1	1	0		2	0	1			1	0	0			0	1		1		1	1	1			0		0	11	* Reported that 80% of those eligible took part, but no actual Ns provided for majority of results (except PMD group for BSI). * No controlling for sig diffs in age, sex and educational achievement between groups. * BSI scores may have been confounded by QoL scores, and vice versa.

Study	Downs and Black Checklist Item																										Total (max. 18)	Notes		
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26			27*	
Seignourel et al. (2007)	1	1	1		2	1	1			0	0	0			0	1		1		1	1	0			1		0	12	* Actual p-values not reported * No information regarding timing of recruitment	
Espirito-Santo & Pio-Abreu (2009)	1	1	1		2	1	1			1	0	0			0	1		1		1	1	1			1		0	14	* Consecutive sampling used - however, not clear how participants were approached, if any declined etc.	
Stone, Warlow & Sharpe (2010)	1	1	1		2	1	1			0	0	1			0	1		1		1	1	1			1		1	15	* Cases not recruited consecutively, and recruited from specialist services (limitation acknowledged by authors) * Commonest diagnosis in controls (MS) associated with depression * No power calculation relevant as study of incidence	
Voon et al. (2010)		1	1	1		1	1	1			1	0	0			0	1		1		1	0	0			1		0	11	* No measure of IQ /education /employment status reported?
Kranick et al. (2011)		1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			1		0	12	* Cases recruited from tertiary care - may be more severe than normal? * Not specified when controls were recruited?
Parees et al. (2012)		1	1	0		2	1	1			1	0	0			0	1		1		1	0	0			1		0	11	* No info re voluntary / consecutive recruitment of cases. No info re timing. No info re inclusion/exclusion for controls.
Voon et al. (2013)		1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			1		0	12	* Rater blinding not stated * Cases recruited from specialist clinic

Study	Downs and Black Checklist Item																										Total (max. 18)	Notes	
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26			27*
DeMartini et al. (2014)	1	1	0		1	1	1			1	0	0			0	1		1		1	0	0			1		0	10	* No info re source of healthy controls reported * No measure of IQ/education used
Brown, Nicholson, Aybek, Kanaan, & David (2014)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			0		0	11	* Study was unable to recruit controls from same geographical area, so had to open up to opportunity sampling * Confounding effects of group diffs on IQ on anxiety/depression not investigated (as secondary measures)
Aybek et al. (2014)	1	1	0		2	1	1			1	0	0			1	1		1		1	0	0			1		0	12	* Reported that some participants excluded for "pragmatic reasons", but not reported what these were
Steffen, Fiess, Schmidt & Rockstroh (2015)	1	1	1		2	1	1			0	0	0			0	1		1		1	1	0			0		1	12	* Exact p-values not all provided * Info regarding recruitment (consecutive vs voluntary) not provided * Sig diff in years of education between cases and controls not controlled for
van der Hoeven et al. (2015)	1	1	1		2	1	1			1	0	0			0	1		1		1	1	0			1		0	13	* Recruitment of FMD and MD patients not consecutive * Healthy controls were spouses of MD patients - confounder?
Aybek et al. (2015)	1	1	1		2	1	1			1	0	0			0	1		1		0	1	0			1		0	12	* Participants recruited from specialist neuropsychiatry clinic, therefore more severe cases than average (?) * Unclear if raters were blind to case/control status * (Unvalidated) clinical interview used to assess sexual abuse history?

Study	Downs and Black Checklist Item																										Total (max. 18)	Notes		
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26			27*	
Fiess, Rockstroh, Schmidt, & Steffen (2015)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0				1		0	12	* Timing of recruitment for cases and controls not reported
Nicholson et al. (2016)	1	0	1		2	1	1			1	0	1			0	0		1		0	0	0				0		1	10	* FMD cases drawn from specialist secondary and tertiary services -may not be representative * IQ scores not available for psychiatric controls * Healthy controls had sig. higher IQ than FMD group, not corrected for in analysis
Fiess, Rockstroh, Schmidt, Wienbruch & Steffen (2016)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0				1		0	12	* Timing of recruitment for cases and controls not reported
Maurer et al. (2016a)	1	1	1		1	1	1			1	0	0			0	1		1		1	0	0				1		0	11	* No measure of IQ /education /employment status reported? * Comment in discussion regarding FMD participants not necessarily being representative of all FMD patients * Unclear when controls were recruited?
Maurer et al. (2016b)	1	1	1		1	1	1			1	0	0			0	1		1		1	0	0				1		0	11	* No measure of IQ /education /employment status reported? * Unclear when controls were recruited?

Study	Downs and Black Checklist Item																											Total (max. 18)	Notes
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27*		
Apazoglou, Mazzola, Wegrzyk, Polara & Aybek (2017)	1	1	1		1	1	1			0	0	0			0	1		1		1	0	0			1		0	10	* "Current psychiatric condition such as [...] depression with acute suicidality" excluded; however "comorbidities such as anxiety or depression without suicidality" were included - effect on psychiatric outcomes? * No measure of IQ/education reported?
Ekanayake et al. (2017)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			1		0	12	* PMD cases drawn from clinics, controls drawn from national database. * Not reported if PMD cases were consecutive or volunteered to participate
Kienle et al. (2017)	1	1	1		2	1	1			1	0	0			0	1		1		1	0	0			1		0	12	* Not reported where controls were drawn from * Not stated when recruitment took place
Tomic et al. (2017)	1	1	0		2	1	1			1	0	0			0	1		1		0	0	0			1		0	10	* Detailed inclusion/ exclusion criteria not provided * Query re the accuracy/validity of unstructured psychiatric interview?

Discussion

The aim of this review was to explore psychosocial associates of motor and sensory functional symptoms, via a review of relevant case-control studies. Several of the findings may offer support to the theoretical model proposed by Kanaan et al. (2017), which suggests that functional motor/sensory symptoms have a different etiology to functional seizures.

Of the associates reviewed, the strongest evidence appeared to be for depressive symptoms, with all of the studies with healthy or organic disorder control samples finding evidence of elevated depressive symptomology in the functional symptoms samples. By contrast, the findings on anxiety symptomology were mixed, with two-thirds of studies finding elevated rates of anxiety, and one-third finding no significant between-group differences.

The association between depressive symptoms and FNS has been noted as far back as Janet's (1907) original studies of functional symptoms, who referred to '*general sentiments of dissatisfaction [and] incompleteness*', '*innumerable lapses of the mental junctions, [including] incapacity of attention [and] memory*' and '*seeming emotional [but] in reality feeling nothing vividly*', experiences that would correspond to symptoms of depression in modern classification systems. Recent theoretical models of FNS (e.g. Brown, 2004) have not emphasized depressive symptoms as predisposing or precipitating factors for FNS, which may be because they are more strongly associated with some symptoms (e.g. motor/sensory symptoms) than others (e.g. seizures). Although case-control studies can only establish that depressive symptoms are common correlates of functional motor/sensory symptoms, and not that they are causal, there was evidence in the reviewed studies that depressive symptoms may be a precipitant of functional motor/sensory symptoms, with both studies of people with recent onset symptoms (Binzer et al., 1997; Stone et al., 2010) finding the association with depressive symptoms to already be present at this early stage. Including measures of FNS in

future prospective studies of people with depressive symptoms would be valuable in further exploring this hypothesis.

Several explanations could be proposed for the more mixed findings on anxiety symptoms. As previously mentioned, the small sample sizes of many of the reviewed studies may have meant that they were underpowered. However, this limitation would also apply to the findings regarding depressive symptoms, making it less likely that this was the sole cause. An alternative explanation, as proposed in Brown and Reuber's (2016) review of associates of functional seizures, is that people with functional symptoms either under-report, or lack awareness of, their anxiety. This hypothesis is supported by a number of findings in the reviewed studies: Apazoglou et al. (2017) found a divergence between subjective and biological measures of stress in response to an experimental social stress test in their functional symptoms sample, but not in their control sample, indicating an altered awareness of stress reactivity in those with functional symptoms; and Stone et al. (2010) found that a significantly higher proportion of their functional symptoms sample had significant anxiety symptoms when assessed by a clinician-rated measure, but not when assessed by a self-report measure. However, another study (Tomic et al., 2017) did not find this same pattern. In addition, if the findings regarding anxiety were predominantly due to people with functional symptoms under-reporting or lacking awareness of their anxiety, one might also have expected them to under-report/lack awareness of other affective symptoms. The findings regarding elevated depressive symptomology suggest that this was not the case.

A third explanation for the mixed findings regarding anxiety, consistent with the theory of Kanaan et al. (2017), is that the association with functional motor/sensory symptoms may genuinely be less robust than the association between functional seizures and depressive symptoms. Symptoms presenting as acute seizures may be more likely to be acute symptoms of anxiety that are not recognized as such (also referred to as 'non-fearful panic')

[Chen, Tsuchiya, Kawakami, & Furukawa, 2009]), a coping response to stress/anxiety that may have emerged as a developmental response to unescapable early-life trauma(s). On the other hand, symptoms presenting as more chronic motor/sensory symptoms may represent an 'escape' response to life stressors, arising from deficits in social problem-solving skills, which are also a common feature of depression (Nezu, 2004). These differences may point towards an important area of differentiation between functional motor/sensory symptoms, and functional seizures.

With regards to personality pathology, the initial findings from Binzer et al. (1997, 1998) of high rates of personality disorder diagnoses in a sample of people with functional motor symptoms do not appear to have been supported by the (albeit limited) subsequent research. This may be another area of difference between functional motor/sensory symptoms and functional seizures, as studies of functional seizure/PNES samples have found high rates of association (ranging from 10 to 86%) between functional seizures and personality disorders, especially borderline and other Cluster B disorders (Lacey, Cook, & Salzberg, 2007). As there is strong evidence implicating childhood maltreatment and trauma in the subsequent development of personality disorders (e.g. Johnson, Cohen, Brown, Smailes, & Bernstein, 1999; Ball & Links, 2009), higher prevalence of personality disorders in people with functional seizures would also be supportive of Kanaan et al.'s (2017) theory that functional seizures are symptoms of a disorder of developmental origin, whereas functional motor/sensory symptoms are not.

For functional motor/sensory symptoms, other features of personality may be more relevant than personality disorder diagnoses. All of the reviewed studies that assessed alexithymic traits found that people with functional motor/sensory symptoms had significantly higher scores than those of healthy or organic neurological disorders control participants. By contrast, studies of people with functional seizures have been much less

consistent in this area (Brown & Reuber, 2016). Alexithymia has a significant impact on engagement with medical and psychological therapies (e.g. Lumley, Neely, & Burger, 2007), so further research into its role in FNS may be beneficial for refining theoretical models, as well as informing clinical practice.

The findings of the reviewed studies with regards to childhood trauma were mixed, with stronger (though not unequivocal) evidence for emotional maltreatment, and weaker evidence for sexual or physical abuse. By contrast, a meta-analysis by Sharpe and Faye (2006) estimated that the odds of having a history of childhood sexual abuse were almost three times greater for people with functional seizures, than for the general population. A review of studies assessing prevalence of physical abuse in people with functional seizures (Fizman, Alves-Leon, Nunes, D'Andrea, & Figueira, 2004) reported lifetime rates of 50-77%, far greater than the prevalence rates reported in any of the studies in this review.

There was, however, some consensus amongst the reviewed studies regarding significant life events/stressors precipitating the onset of functional symptoms, with six of the seven studies assessing recent events (i.e. in the year prior to symptom onset) finding significantly higher rates of relevant events (and/or subjective distress associated with them) in functional symptoms samples. Tellingly, Nicholson et al. (2016) reported that a key event and/or a compelling psychological formulation could be generated for 91% of their participants with functional symptoms when a comprehensive, semi-structured assessment of life events was used, even though for 88% of this group, this key event had not been documented in their clinical notes. On the basis of research findings that precipitating psychological stressors could not always be elicited in people with functional symptoms (e.g. Roelofs, Spinhoven, Sandijck, Moene, & Hoogduin, 2005), identifying psychosocial factors associated with symptom onset was downgraded from an essential diagnostic criteria to an 'associated feature supporting diagnosis' in DSM-5 (APA, 2013) – this finding suggests that

such events may be more readily discoverable for those with functional motor/sensory symptoms, if appropriate methods are employed.

This pattern of findings on childhood trauma and life stressors would be consistent with Kanaan et al.'s (2017) theory that functional motor/sensory symptoms are more likely to be an acute response to immediate stress, whereas functional seizures are the result of a developmental trauma. Ekanayake et al.'s (2017) finding that the mean age at which people in a PNES sample experienced their most distressing traumatic event was significantly younger than those of people in functional movement disorder and healthy control samples (mean age 18.2 in PNES sample, vs 32.1 and 29.2 in FMD and HC samples, respectively), also points towards a different developmental trajectory for people with functional seizures, than for people with functional motor/sensory symptoms.

Clinical and research implications

The findings of this review highlight the diversity of psychosocial risk factors associated with functional motor/sensory symptoms, and the challenges of devising treatment approaches that apply to such a heterogeneous group. Comorbidity of symptoms, both pseudoneurological and psychiatric, is widely acknowledged to be the norm, rather than the exception, in populations with functional symptoms (Şar, Akyüz, Kundakçi, Kiziltan, & Doğan, 2004). Heterogeneity of symptoms and comorbidities underlines the importance of developing an individualized 'neuro-bio-psycho-social' formulation and treatment plan for those with FNS (McKee et al., 2018), which includes an assessment not only of the person's symptoms and experiences, but also their subjective interpretation of them. This review suggests that routine assessment of depressive symptoms, alexithymia, and recent life events may support clinicians to achieve this.

In advocating for a model of understanding distress that includes a broader consideration of the impact of psychosocial factors, rather than a model based on functional psychiatric diagnoses, Johnstone and Boyle (2018) point out that functional neurological disorders are one of the few diagnoses in the current psychiatric classification systems where the symptoms are explicitly acknowledged as responses to adverse psychosocial events or environments. The findings of this review offer support for clinical approaches that consider the function of functional symptoms, and their relationship to significant events in a person's life, rather than focusing on which neurological disorder they resemble.

The findings of this review, regarding a possible association between functional motor/sensory symptoms, and childhood emotional abuse/neglect, low parental emotional warmth, and high parental rejection, may also suggest a novel direction for future research. Caregiver emotional under-involvement with children is thought to be a significant risk factor for subsequent impairments in reflective functioning (also known as mentalization) (Fonagy & Bateman, 2007). Mentalization, defined as the ability to think about ourselves and others as having a mind (comprised of feelings, thoughts and intentions) (Subic-Wrana, 2011), is hypothesised to play a key role in the development of the ability to regulate one's own emotions, as well as to distinguish the sensations of affective states from those of physical states. Typically, these abilities are developed by a caregiver helping a child to make the implicit processing of emotions explicit, through the use of mirroring and the marking of affects, thus enabling them to differentiate their own inner states, and distinguish their own minds from those of others (Gergely & Watson, 1996). However, impairments in the ability to mentalize have been found to emerge when a child's affective expressions are consistently ignored by the caregiver, either due to emotional neglect, or because the caregiver themselves has difficulty distinguishing expressions of affective states (e.g. separation anxiety) from those of physical symptoms (Fonagy, Gergely, Jurist, & Target, 2002). Difficulties with

recognising emotional states, and with distinguishing them from physical states and symptoms, have typically been ascribed to alexithymia in people with FNS. For some people, however, these difficulties could plausibly be more usefully understood as a feature of an impairment in the ability to mentalize. Mentalization, and the ways in which it overlaps with, or is separate from, alexithymia, has not previously been explored in people with FNS, and further research in this area may contribute towards an improved understanding of the etiology of FNS.

Conclusion

The findings of this review of studies from nine different countries, spanning over 20 years of research, highlight the diversity of psychosocial associates of functional motor and sensory symptoms. While many of these are undoubtedly also common to functional seizures, there appear to be areas of subtle difference, particularly regarding depressive symptoms, personality pathology, precipitating life events, and childhood adversity. These findings offer tentative support to a perspective that functional neurological symptoms may be more helpfully conceptualised by their manner of presentation (acute vs chronic) and their relationship to stress and trauma (recent vs developmental), than by their equivalence to neurological conditions. Greater focus on the function of the functional neurological symptom(s) may enable better understanding of the symptom, and of its role for the person experiencing it, and lead to the development of treatment approaches that are more flexible, engaging, and helpful.

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Section B: Alexithymia, mentalization and symptom complexity in functional neurological
disorder (FND)

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Abstract

Objective: Alexithymia and hypomentalization, two traits associated with childhood emotional abuse/neglect, have not previously been studied in people with mixed-symptom functional neurological disorder (FND). This study examined whether these traits were more prominent in people with FND than in healthy control participants, and explored the relationships between alexithymia, hypomentalization, and somatic/neurological symptoms of a generalized nature. **Method:** Twenty-nine participants with FND and 41 healthy control participants completed a battery of self-report measures. Between-group differences in alexithymia and hypomentalization were investigated using parametric tests, and binary logistic regression analyses examined whether alexithymia and hypomentalization were predictive of FND (vs control) group status, after controlling for depressive symptoms, anxiety symptoms and education attainment. Linear regression analyses examined whether alexithymia and hypomentalization were associated with physical and neurological symptoms across the entire sample of FND group and control group participants. **Results:** Participants with FND had significantly higher scores on measures of alexithymia, hypomentalization, somatic symptoms and neurological symptoms, than healthy control participants. Between-group differences in alexithymia and neurological symptoms remained significant after controlling for covariates. High scores on the alexithymia and mentalization measures were also predictive of high scores on the measures of somatic and neurological symptoms across the entire sample. **Conclusion:** Alexithymia and hypomentalization appear to be significant issues for people with FND, and may contribute to the tendency to express distress via physical symptoms. Exploring these traits with individual service users may contribute to a more comprehensive conceptualisation of their difficulties, and inform the development of future treatment approaches.

Key words: functional neurological disorder (FND), mentalization, alexithymia,

Alexithymia, mentalization and symptom complexity in functional neurological disorder (FND)

Introduction

Definition and theoretical model

Functional neurological disorder (FND) is a condition characterised by symptoms of altered motor or sensory function, which are incompatible with recognized neurological or medical conditions, and which cause clinically significant distress and/or impairment in functioning (American Psychiatric Association, 2013). Common symptoms of FND include abnormal motor movements, weakness or paralysis; disturbances of speech or swallowing; sensory loss or anaesthesia; and dissociative attacks or seizures (also commonly known as psychogenic non-epileptic seizures or PNES).

As the symptoms of FND commonly resemble those of neurological disorders (such as Parkinson's disease, stroke or epilepsy), individuals with FND are often first seen in neurology clinics - Stone et al. (2009) reported that 30% of 3,781 service users seen at Scottish neurology clinics between December 2002 and February 2004 had neurological symptoms that were "Not at all" or only "Somewhat" explained by organic disease. If all symptoms (not just the presenting one) are considered, up to 61% of neurology service users have been reported to have functional symptoms (Fink, Hansen & Søndergaard, 2005). Estimates of the prevalence of FND range from 4-50 per 100,000 population per year, depending on the definition used (Carson et al., 2012). Due to the heterogeneity of symptoms, and their apparent overlap between the neurological and the psychological, FND was traditionally an "orphan disorder", with no single discipline keen to assert ownership of it (Baslet, Dworetzky, Perez, & Oser, 2015). There is now a growing consensus that an interdisciplinary, 'neuro-bio-psycho-social' approach to the assessment, formulation and treatment of FND is essential (Carson et al., 2012; Brown & Reuber, 2016; McKee et al., 2018).

In recent years, Richard Brown's (2004) integrative conceptual model has become a highly influential theoretical model of FND (Carson et al., 2012; Reuber & Brown, 2017). The model explicitly synthesises useful concepts from previous dissociation, conversion, and somatization models of FND, as well as integrating ideas and research from cognitive, social constructionist and neurobiological approaches.

Drawing on dissociation models (e.g. Janet, 1889, 1907; Oakley, 1999), the integrative conceptual model theorises that FND is the product of disturbances of the cognitive attentional system. It hypothesises that human actions are controlled via two mechanisms. Routine behaviours are controlled by a primary attentional system (PAS), which chooses the required attentional, cognitive and motor processes from previously learned mental models (schemata). This process requires little conscious effort, and is experienced as intuitive and automatic. For novel or complex behaviours, however, where there is no existing schemata, a secondary attentional system (SAS) is required, which draws on information from existing schemata that appears relevant, and from sensory input(s). Processing controlled by the SAS feels more effortful and demanding of conscious control – it is these processes that we are more aware of controlling. The integrative conceptual model suggests that functional symptoms arise when the PAS and SAS are activated inappropriately. It suggests that some symptoms (e.g. functional motor tremors), where a behaviour should be under the control of the SAS, become controlled by the PAS, leading to dissociation between the action and the experience of subjective volition; other symptoms (e.g. functional motor paralyses) develop when existing schemata (controlled by the PAS) are more compelling than the information being provided by the environment or the senses, leading to symptoms that are inconsistent with the objective sensory world, but are experienced by the person as subjectively 'real' (Brown, 2002).

Brown's model suggests that these compelling schemata ('rogue representations') may have their origins in exposure to certain physical states in the self (e.g. distressing and memorable physical symptoms of anxiety) or in others (e.g. illness or disability in family/friends). They may also develop via sociocultural transmission of illness representations: Reuber and Elger (2003), for example, report that a high proportion of functional seizures involve asynchronous limb movements, side-to-side head shaking, and closed eyelids, symptoms which are relatively rare in epileptic seizures, but are what people 'expect' to happen in a seizure.

Drawing on somatization theories, Brown's model suggests that the likelihood of developing FND is increased by factors such as heightened body-focussed attention, misattribution of the cause of symptoms, illness worry and rumination, and illness behaviour (the so-called "secondary gains" of FND, such as increased care or attention from others, or avoidance of unwanted tasks/responsibilities).

Finally, echoing conversion theories (e.g. Breuer & Freud, 1893-1895/1982), the model also integrates findings that childhood trauma is common in people with FND (e.g. Fiszman, Alves-Leon, Nunes, D'Andrea, & Figueira, 2004; Section A of this thesis). Increased body focus is hypothesised as acting as a defence against the overwhelming negative affect or cognition associated with trauma, which can be adaptive in the short term, but become maladaptive in the long term. This element of the model has been further expanded recently, with the suggestion that acute functional symptoms (e.g. functional seizures, paroxysmal movement disorders) may be the result of a developmental disorder with its origins in childhood trauma, while chronic symptoms are an idiosyncratic coping response to acute life stressors (Kanaan et al., 2017).

Treatments and prognosis

Although Brown's model has advanced understanding of the psychological mechanisms underpinning FND, there remains limited consensus as to how to translate this understanding into therapeutic practice (LaFrance, Rusch, & Machan, 2008). A Cochrane review of psychosocial interventions for FND (Ruddy & House, 2005) found only three relevant treatment studies, and these were assessed as offering only slight evidence in favour of help rather than harm, in terms of engagement, mental state, and physical functioning. Since this review, randomised controlled treatment trials of cognitive behaviour therapy-informed psychotherapy (Goldstein et al., 2010; LaFrance et al., 2014), cognitive behaviour therapy with guided self-help (Sharpe et al., 2011), and integrated inpatient physiotherapy and psychotherapy (Jordbru, Smedstad, Klungsøyr, & Martinsen, 2014; Nielsen et al., 2015) have been published. These, and other published case-series and uncontrolled treatment studies, have generally had small sample sizes and methodological limitations, and so are still considered recommendations for further research at this time (Perez & LaFrance, 2016).

An additional obstacle to treatment is that people with FND are often reluctant to engage with psychological interventions. Several studies have reported that more than half of service users with FND referred for psychological therapies do not engage with them (Howlett, Grunewald, Khan, & Reuber, 2007; Baslet & Prenskey, 2013). One potential reason for this is that many people with FND continue to attribute their symptoms to an undiscovered physical cause after the diagnosis of FND is confirmed, even though misdiagnosis of functional symptoms is actually very rare (Stone et al., 2005; Stone et al., 2009). Paradoxically, people with FND are also often less likely than those with organic disorders to attribute their symptoms to psychological factors, including stress (Stone, Binzer and Sharpe, 2004; Stone, Warlow, & Sharpe, 2010; Ludwig, Whitehead, Sharpe, Reuber, & Stone, 2015).

Currently, the prognosis for people with FND remains poor (Carson et al., 2012). Gelauff, Stone, Edwards, and Carson (2014) reported that more than one-third of people with FND had the same or worse symptoms when re-assessed (on average) 7.4 years later, while Durrant, Rickards, and Cavanna (2011) found that 40% or less of those diagnosed with functional seizures as adults achieved seizure remission five years after diagnosis, compared to 60-80% of those with epileptic seizures. Individuals with FND have been found to have levels of distress, disability and healthcare usage equal to, or even greater than, those of individuals with equivalent organic neurological disorders (Anderson et al., 2007; Carson et al., 2011). Research has also found that over 70% of people with functional seizures were treated with inappropriate and potentially harmful anticonvulsant medications prior to diagnosis (Reuber, Fernández, Bauer, Helmstaedter, & Elger, 2002; de Timary et al., 2002), highlighting that FND can have a significant impact on physical, as well as psychological, well-being.

Revisiting childhood emotional adversity

The combination of unexplained symptoms, reluctance to engage with psychological models, and poor prognostic outcomes, suggests that the dominant conceptualisations of FND remain incomplete. One consistent research finding, that may have important implications for the understanding and treatment of FND, is that people with FND have high rates of childhood emotional/psychological adversity. Brown and Reuber's (2016) review of psychological associates of functional seizures found that people with functional seizures reported higher rates of childhood emotional abuse/neglect than control participants in each of the six case-control studies that had explored this area. One of these studies (Salmon, Al-Marzooqi, Baker, & Reilly, 2003) also found childhood emotional abuse was a unique predictor of functional seizure (versus epilepsy) status, but physical or sexual abuse were not

(although it was no longer a significant predictor after controlling for other family characteristics). A review of published case-control studies of functional motor and sensory symptoms (Section A of this thesis) also found that these studies frequently reported higher rates of childhood emotional abuse/neglect in FND samples, while the findings regarding childhood sexual and physical abuse were more variable.

A number of consequences of childhood emotional abuse/neglect may be of relevance to the subsequent development of functional symptoms, the rejection of psychological causes for the symptoms, and the limited efficacy of psychological treatments. One outcome of childhood emotional abuse/neglect that has been suggested is alexithymia in later life (Zlotnick, Mattia, & Zimmerman, 2001; Aust, Härtwig, Heuser, & Bajbouj, 2013). Alexithymia describes a cluster of cognitive and affective features, characterised by difficulty identifying and describing feelings, difficulty distinguishing between emotions and bodily sensations of emotional arousal, restricted imagination and fantasy, and externally-oriented thinking (Taylor, 2000). It is generally considered to be a personality trait, with a high degree of stability over time (Aust et al., 2013), and it is of particular relevance to FND, as it was first described in the 1970s among people with classic psychosomatic symptoms (Sifneos, 1973).

Two mechanisms for the influence of alexithymia on FND have been suggested by Brown and Reuber (2016). Motivation not to recognise emotional states (because strong affect is seen as unacceptable or unmanageable), coupled with a tendency to suppress emotions, may lead to a heightened focus on the physical aspects of emotions. Alternatively, alexithymia may reflect intrinsic deficits in recognising, describing and thinking about emotions, potentially with neurodevelopmental origins. Functional symptoms may therefore be symptoms of affective arousal incorrectly interpreted as symptoms of a physical disorder, or they may act as a release mechanism for unrecognised emotional tensions. Alexithymia

has previously been studied in populations of people with functional seizures (e.g. Kaplan et al., 2013) and functional motor symptoms (e.g. Demartini et al., 2014); however, these studies typically excluded people with mixed functional symptoms, so it has not yet been established whether these findings would generalize to the heterogeneous FND population typically seen by NHS neuropsychiatry services.

A potential alternative, or additional, outcome of childhood emotional abuse/neglect is impairment of reflective functioning, or mentalization. Mentalization is the ability to think about ourselves and others as having a mind (comprised of feelings, thoughts and intentions), which enables us to be aware of our emotions, and to regulate them by thinking about them (Subic-Wrana, 2011). The ability to mentalize is thought to develop in early childhood, when a caregiver helps a child to make the implicit processing of emotions explicit, through the use of mirroring and the marking of affects. This enables a child to learn to distinguish between his/her own inner states, and those of others (Gergely & Watson, 1996). However, if the affective expressions of a child are consistently undetected by the caregiver, either due to emotional neglect, or because the caregiver themselves cannot distinguish the expressions of affective states (e.g. separation anxiety) from those of physical symptoms, then impairments may develop in the ability to distinguish between one's own mental states and those of others, and in the ability to develop mental representations of the physical sensations that belong to particular affective states (Fonagy, Gergely, Jurist, & Target, 2002). While deficits in mentalization are currently most commonly associated with the symptoms of borderline (emotionally unstable) personality disorder diagnoses (Fonagy & Luyten, 2009), this last element of mentalization theory, regarding the influence of mentalization on the ability to differentiate between emotional distress and physical distress, may also be highly relevant to the understanding FND.

Assessment of mentalization has previously only been possible by using the Reflective Functioning Scale (Fonagy, Target, Steele, & Steele, 1998) to rate the Adult Attachment Interview (George, Kaplan & Main, 1996), which has limited research value due to issues with the practicality, cost and duration of the measure (Badoud et al., 2015). However, a self-report measure, the Reflective Functioning Questionnaire (RFQ; Fonagy et al., 2016), has recently been developed as a brief screening measure of reflective functioning. The measure looks at two elements of mentalizing (certainty about mental states, and uncertainty about mental states), enabling evaluation of the tendency to hypermentalize (i.e. to develop models of the mind that go far beyond the available evidence) and to hypomentalize (i.e. to be unwilling or unable to develop models of the mind that are realistically nuanced and opaque) (Badoud et al., 2015). This study is the first to assess reflective functioning, and to do so using the RFQ, in a typical UK FND population.

Aims and hypotheses

Based on theory, and on the research findings described above, the aim of this study was to explore whether alexithymia, and/or difficulties with mentalizing, are more common in people with FND, than in people without FND. Exploring these areas is important as this may lead to the refinement theoretical models of FND, and may also have implications for treatment approaches for FND, particularly those reliant on emotional awareness and insight, as many psychological therapies are.

Alexithymia has previously been found to be associated with lower educational attainment (Lane, Sechrest, Riedel, 1998; Salminen, Saarijärvi, Äärelä, Toikka, & Kauhanen, 1999), and with symptoms of depression (Honkalampi, Hintikka, Tanskanen, Lehtonen, & Viinamäki, 2000) and anxiety (Berthoz, Consoli, Perez-Diaz, & Jouvent, 1999). As a result, measures of these variables were included in the study, so they could be examined as

covariates. To expand the exploration beyond categorical diagnoses of FND, measures of common somatic and neurological symptoms were also included, in order to examine whether alexithymia and mentalization would be predictive of people experiencing more physical symptoms, regardless of whether or not they had FND.

The study therefore aimed to test the following hypotheses:

- i. Individuals with FND will, on average, have greater impairments in their ability to understand and describe emotions (i.e. alexithymia), than healthy control participants.
- ii. Individuals with FND will have a greater tendency to hypomentalize (an impairment of reflective functioning characterized by low certainty, and high uncertainty, about mental states in one's self and others), than healthy control participants.
- iii. Individuals with FND will describe, on average, more somatic and neurological symptoms of a generalized nature, than healthy control participants (as they ascribe symptoms of affective arousal to physical defects).
- iv. Higher levels of alexithymia and hypomentalizing will predict FND (versus control) group membership in a logistic regression, when educational attainment, depressive symptoms, and anxiety symptoms are controlled for.
- v. Higher levels of alexithymia and hypomentalizing will predict higher levels of somatic/neurological problems of a generalized nature, in linear regression analyses of the FND and control groups combined.

Methods

Design

A cross-sectional, between-groups design was employed, with two groups of participants – one group of service users with a diagnosis of FND (the FND group), and one group of individuals with no history of FND (the healthy control group).

Participants

A target sample size of 26 participants per group was chosen, based on Cohen's (1992, p. 158) calculation to detect large effect sizes in independent two-sample t-tests, with conventional levels of significance (.05) and statistical power (80%). For the logistic regression analyses, this sample size also allowed for up to eight predictors to be included in the models, with sufficient power to detect a large effect size. For the linear regression analyses, a sample size of 52 allowed for a medium-large effect size to be detected.

Recruitment of service users with a diagnosis of FND took place at two tertiary neuropsychiatry services in England between October 2017 and March 2018. Recruitment continued until the target of 26 participants was reached. Eligibility for the study was confirmed by an experienced neuropsychologist or neuropsychiatrist, on the basis of a review of electronic medical records.

Recruitment of the healthy control group took place between January and March 2018, via an email invitation sent to university staff and students, and an advertisement on social media. Additional participants were recruited for the healthy control group, in the hope that the groups could be matched on educational attainment, but as this did not prove possible, instead all participants were included in the control group and educational attainment was controlled for as a covariate. (The implications of this are discussed further in the Limitations section.)

Inclusion criteria for both groups were being aged 18 or over, and being able to read English or access support to do so (in order to complete the questionnaires). Having a diagnosis of FND was an inclusion criterion for the FND group. Individuals with a primary diagnosis of chronic pain were excluded from the FND group, as this condition commonly has an organic etiology, such as arthritis/osteoarthritis, traumatic injury or surgery (Breivik, Collett, Ventafridda, Cohen, & Gallacher, 2006). Individuals with a primary diagnosis of

ME/chronic fatigue syndrome were also excluded, as these conditions are thought to often be associated with abnormalities of the central nervous system, immune system or neuroendocrine regulation system (Afari & Buchwald, 2003). Control group participants were excluded if they self-reported a current diagnosis of a neurological, psychiatric or serious physical health condition. Full demographic details of the two groups of participants are presented in the Results section.

Written or electronic informed consent was gained from all participants. The study was approved by the UK National Research Ethics Service, the Health Research Authority, and the relevant NHS Research and Development departments (Appendices 2-6).

Materials

A battery of self-report questionnaire measures was used to assess the variables of interest. For reference, all of the measures are presented in full in Appendix 7.

Alexithymia was measured via the *Toronto Alexithymia Scale* (TAS-20; Bagby, Taylor, & Parker, 1994a, 1994b), a 20-item questionnaire which generates scores for alexithymia sub-domains of Difficulty Identifying Feelings (TAS_DIF), Difficulty Describing Feelings (TAS_DDF), and Externally-Oriented Thinking (TAS_EOT), as well as an overall score for alexithymia (TAS_20_TOTAL). Participants were asked to rate their agreement with statements such as “*I am often confused about what emotion I am feelings*” on a five-point Likert-type response scale ranging from “strongly disagree” to “strongly agree”. Higher agreement generated higher scores on 15 of the measure items, and five items (e.g. “*Being in touch with emotions is essential*”) were reverse scored. Overall scores for alexithymia range from 20-100, with scores of ≥ 61 indicative of alexithymia (Taylor, Bagby, & Parker, 1997). The total and subdomain scores have been shown to have acceptable internal reliability (TAS_20_TOTAL, $\alpha = 0.86$; TAS_DIF, $\alpha = 0.80$; TAS_DDF, $\alpha = 0.76$;

TAS_EOT, $\alpha = 0.71$) (Parker, Taylor, & Bagby, 2003). The internal reliabilities of the TAS_20_TOTAL ($\alpha = 0.92$), TAS_DIF ($\alpha = 0.93$) and TAS_DDF ($\alpha = 0.84$) scales in this study were broadly equivalent to that reported in previous studies; however, as reported in other studies (e.g. Kooiman, Spinhoven, & Trijsburg, 2002) the internal consistency of the TAS_EOT scale was notably lower ($\alpha = 0.66$), and the results regarding this measure should be interpreted with this in mind.

Mentalizing was measured via the *Reflective Functioning Questionnaire* (RFQ; Fonagy et al., 2016). The RFQ is comprised of two subscales: the Certainty about Mental States subscale (RFQ_C) and the Uncertainty about Mental States subscale (RFQ_U). Both subscales consist of six items, which participants were asked to rate on a seven-point Likert scale ranging from “completely disagree” to “completely agree”. For the RFQ_C subscale, low levels of agreement with statements such as “*People’s thoughts are a mystery to me*” lead to high scores (indicating hypermentalizing), and lower scores signify normal levels of certainty about mental states. For the RFQ_U subscale, high levels of agreement with statements such as “*I don’t always know why I do what I do*” lead to high scores (indicating hypomentalizing), and lower scores signify normal levels of uncertainty about mental states. The total score on each subscale is calculated as the mean of the six responses, and ranges from 0-3. Initial analyses (Badoud et al., 2015) of the six-item scales reported acceptable internal consistency for the subscales (RFQ_C: $\alpha = 0.72$; RFQ_U: $\alpha = 0.64$). The internal consistency of the subscales in this sample was also acceptable (RFQ_C: $\alpha = 0.73$; RFQ_U: $\alpha = 0.78$).

Depressive symptoms were assessed using the *Patient Health Questionnaire depression scale* (PHQ-8; Kroenke & Spitzer, 2002), an 8-item measure used widely in research (e.g. Strine et al., 2008) and clinical practice (Kroenke et al., 2009). The PHQ-8 assesses the frequency with which common symptoms of depression (e.g. “*Feeling down,*

depressed or hopeless”) have been experienced in the previous two weeks, on a four-point response scale (“Not at all” = 0; “Some days” = 1; “More than half the days” = 2; “Nearly every day” = 3). Total scores range from 0-24, with a cut-off score of ≥ 11 suggested as having the best trade-off between sensitivity and specificity for indicating a major depressive disorder diagnosis (Manea, Gilbody, & McMillan, 2012). The measure has previously been reported to have good internal consistency ($\alpha = 0.86 - 0.89$) (Smarr & Keefer, 2011), and internal consistency in this study was also good ($\alpha = 0.94$).

Anxiety symptoms were assessed using the *Generalized Anxiety Disorder scale* (GAD-7; Spitzer, Kroenke, Williams, & Löwe, 2006). This seven-item scale assesses common symptoms of anxiety (such as “*Feeling nervous, anxious or on edge*”) in the previous two weeks, using the same four-point Likert-type response scale as the PHQ-8. Scores range from 0-21, with a cut-off score of ≥ 10 suggested as having the best sensitivity and specificity for clinically significant anxiety (Löwe et al., 2008). The measure has been reported to have acceptable internal consistency ($\alpha = 0.89$) (Löwe et al., 2008), which was also found in this study ($\alpha = 0.93$).

Comorbid physical health symptoms of a generalized nature were assessed via a checklist of 14 symptoms, based on the *Patient Health Questionnaire Somatic Symptom Severity Scale* (PHQ-15; Kroenke, Spitzer, & Williams, 2002) (referred to hereafter as the *PHQ-15 checklist*). The measure assesses the presence of fourteen common somatic symptoms (such as back pain, headaches and digestive difficulties) over the previous four weeks. One item (regarding pain related to menstruation) is gender specific, and was excluded to make the data consistent across the sample, as per Witthöft, Hiller, Loch and Jasper (2013). On the PHQ-15, participants are asked to rate whether they have been “Not bothered at all” (scoring 0), “Bothered a little” (1), or “Bothered a lot” (2) by the symptoms over the previous four weeks. This scale is reported as having acceptable internal

consistency ($\alpha = 0.80$) and test-retest reliability (intraclass correlation = 0.65) (Han et al., 2009). Due to a coding error, only six participants completed the measure using a three-point response scale, with the remaining participants instead rating whether the symptoms were absent (scoring 0) or present (1) over the previous four weeks. To explore the reliability of this scoring system, the data from the six participants who completed the full measure was recoded to the same present/absent scale (“Not bothered at all” = 0; “Bothered a little” or “Bothered a lot” = 1). The scores of the two rating scales correlated significantly ($R = 0.81$, $p = 0.049$), and the internal consistency of the measure with a present/absent rating scale was also adequate ($\alpha = 0.85$), so a decision was made to retain this measure. Potential scores on this measure therefore ranged from 0-14.

Finally, neurological symptoms were measured via a common neurological symptoms checklist, developed by Carson et al. (2014) (referred to hereafter as the *Neuro checklist*). Participants were asked to indicate whether or not they had experienced 10 common neurological symptoms (such as paralysis or weakness, loss of sensation, or seizures/fits) in the past month. Each endorsed item was allocated a score of one point, leading to a range of total scores from 0-10. The internal consistency of this measure in the current study was acceptable ($\alpha = 0.86$).

Procedure

Service users with FND, who were identified as being eligible for the study, were approached about participation in the study via a letter or a verbal invitation from a clinician at their service. After reading the Participant Information Sheet (Appendix 8), interested FND group participants were asked to either complete the consent form (Appendix 9) and questionnaires measures on paper forms, and to return these in the post, or alternatively, to visit an online survey website (www.qualtrics.com) and complete the consent form and study

measures electronically. Control group participants were invited (via email and advertisements on social media) to visit the same online survey website, where a Participant Information Sheet (Appendix 10) was available. Interested participants could then complete an online consent form (Appendix 11) and the study measures on the survey website.

In order to safeguard participants from harm, the Participant Information Sheets advised all participants of sources of emotional support that they could access if they found taking part in the study distressing. Participants with FND were also advised that they could contact the collaborating clinicians at their neuropsychiatry service if they wished to discuss any aspects of the study further.

In total, of the 65 service users with FND asked to participate, 29 (44.6%) agreed to take part, along with 42 healthy control participants. All 71 participants who started the measures completed them in full.

Analysis

Data from the study measures were analysed using SPSS (version 24). Means and SDs were calculated for continuous variables, and frequencies and percentages for categorical variables. Across all participants and measures, less than 0.1% of questionnaire responses were missing, with no more than one item missing on any one measure. Where no formal rules were available for managing missing data, missing items were replaced by the mean of the remaining items on the scale.

To explore between-group differences on measures of interest (*Hypotheses i-iii*), a series of t-tests were undertaken. Exploratory analyses of the data were conducted first, to establish whether the necessary assumptions for parametric tests were met. Kolmogorov-Smirnov tests for each group of participants revealed that data from a number of the measures (TAS_DIF, RFQ_C, RFQ_U, PHQ-15 checklist, PHQ-8, GAD-7, and Neuro Checklist) did

not meet the assumption of normal distribution. Levene's tests for homogeneity of variance were significant ($p < .05$) for the RFQ_U, PHQ-15 checklist, PHQ-8, GAD-7 and Neuro checklist, indicating that the assumption of equal variance was not met for these variables. As recommended by Wright, London, and Field (2011), bootstrap estimation was used on all of the parametric analyses as a more conservative measure. Significance values and confidence intervals are reported from the bootstrap analyses, as well as the test statistics and significance values from the parametric tests.

For *Hypothesis iv*, a series of hierarchical logistic regression analyses were undertaken, with group status (FND or control group) entered as the dependent variable; the PHQ8, GAD7 and educational attainment measures entered in the first block (as confounding variables); and the predictor of interest entered in the second block. Eight separate regressions were conducted, one for each of the eight predictors of interest (TAS_20_TOTAL, TAS_DIF, TAS_DDF, TAS_EOT, RFQ_C, RFQ_U, PHQ-15 checklist, and Neuro checklist). Prior to these analyses, the assumptions for logistic regression analyses were tested, as per Field (2013). The assumption of linearity of the logit was met, as the interactions between each of the predictors and the log of itself were not significant (all $p > .05$). Examination of the collinearity between variables indicated that all of the variables' Variance Inflation Factor indices were below 10, and the tolerance values were above 0.1, indicating that the assumption of no collinearity was met, with the exception of the PHQ-8. Bivariate correlations indicated that this measure correlated highly with one of the other covariates (the GAD-7), but also with one of the predictors of interest (the PHQ-15). As a result, the results of this particular regression should be interpreted with caution.

For *Hypothesis v*, six separate linear regressions were undertaken, with TAS_20_TOTAL, RFQ_C, and RFQ_U entered in to models as predictor variables, and the PHQ-15 checklist and Neuro checklist scores entered as dependent variables for each

predictor. Prior to the analyses, the scatterplots of standardised predicted values versus standardised residuals were examined, to establish if the data met the assumptions of homogeneity of variance and linearity, and if the residuals were approximately normally distributed. Some mild variations were noted, therefore bootstrap estimation of the linear regression analyses was used as a more conservative method (Wright et al., 2011).

Results

Demographic and clinical characteristics

The demographic and clinical characteristics of the total sample, and of the FND and control groups, are presented in Table 1. There were no significant between-group differences in gender (Fisher's exact test, $p = 1.00$) or mean age ($t[42.71] = -.948$, $p = .349$). There was a significant between-group difference on ethnicity, with a higher proportion of participants of White ethnic background in the control group (Fisher's exact test, $p = .018$). In order to compare the educational attainment of the groups, attainment was ranked on a scale from one to eight, in accordance with UK Regulated Qualification Framework (Office of Qualification and Examination Regulations, 2015). There was a significant between-group difference (FND group $Mdn = 3.50$, Control group $Mdn = 7.00$; $U = 136.00$, $z = -5.09$, $p < .001$), with educational attainment, on average, higher in the control group.

On the measure of depressive symptoms (PHQ-8), FND group participants, on average, scored higher than control group participants and the difference, -10.51 , BCa 95% CI $[-13.08, -7.69]$, was significant, $t(47.10) = -7.38$, $p < .001$, representing an effect size of $d = 2.23$. A significantly higher proportion of the FND group also scored above the clinical cut-off of 11 on the depression measure (69% vs 11.9%, Fisher's exact test, $p < .001$).

FND group participants, on average, scored higher than control group participants on the measure of anxiety (GAD-7), and the difference, -7.02 , BCa 95% CI $[-9.59, -4.34]$, was significant, $t(46.26) = -5.19$, $p < .001$, representing an effect size of $d = 1.60$. A significantly

Table 1.

Demographic and clinical characteristics of study participants

	Total sample (N=71) n (%)	FND group (n=29) n (%)	Control group (n=42) n (%)
Gender			
- Female	58 (81.7)	24 (82.8)	34 (81.0)
- Male	13 (18.3)	5 (17.2)	8 (19.0)
Ethnicity			
- White	64 (90.1)	22 (75.9)	42 (100)
- Other	4 (5.6)	4 (13.8)	0
- Not stated/Prefer not to say	3 (4.2)	3 (10.3)	0
Educational attainment			
- GCSE Grades D-G (1)	1 (1.4)	1 (3.4)	0
- GCSE Grades A*-C (2)	9 (12.7)	8 (27.6)	1 (2.4)
- A Level (3)	4 (5.6)	3 (10.3)	1 (2.4)
- Certificate of Higher Education (4)	2 (2.8)	1 (3.4)	1 (2.4)
- Diploma of Higher Education (5)	5 (7.0)	3 (10.3)	2 (4.8)
- Bachelor's Degree (6)	16 (22.5)	6 (20.7)	10 (23.8)
- Master's Degree / Postgraduate Certificate/Diploma (7)	25 (35.2)	2 (6.9)	23 (54.8)
- Doctorate (8)	4 (5.6)	0	4 (9.5)
- Not stated/Prefer not to say	5 (7.0)	5 (17.2)	0
	<i>Mean (SD)</i>	<i>Mean (SD)</i>	<i>Mean (SD)</i>
Age	36.23 (12.77)	38.10 (16.04)	34.93 (9.92)
Clinical comorbidity			
- PHQ8		14.91 (6.60)	4.40 (4.70)
- GAD7		12.31 (6.31)	5.29 (4.39)
		<i>n (%)</i>	<i>n (%)</i>
Scoring above clinical cut-off for depression (≥ 11 on PHQ8)		20 (69.0)	5 (11.9)
Scoring above clinical cut-off for anxiety (≥ 10 on GAD7)		20 (69.0)	7 (16.7)

higher proportion of the FND group also scored above the clinical cut-off of 10 on the anxiety measure (69% vs 16.7%, Fisher's exact test, $p < .001$).

Hypothesis i

The mean scores of the TAS-20, the TAS-20 subscales, RFQ_C, RFQ_U, PHQ-15 checklist, and Neuro checklist are presented in Table 2.

The first hypothesis was that participants with FND would have significantly greater alexithymic traits than control participants. Consistent with the hypothesis, on average, FND group participants had higher scores on the TAS-20, and the difference, -21.90, BCa 95% CI [-28.55, -15.78], was significant $t(69) = -7.43$, $p < .001$, representing an effect size of $d = 2.06$. A significantly higher proportion of the FND group also scored above the clinical cut-off (≥ 61) on the measure (55.2% vs 2.4%, Fisher's exact test, $p < .001$).

In addition, on average, FND group participants had higher scores on the of the subscales of the TAS-20. On the Describing Feelings subscale (TAS_DDF), the difference, -5.76, BCa 95% CI [-8.11, -3.41], was significant, $t(69) = -5.18$, $p < .001$, and represented an effect size of $d = 1.30$; on the Difficulty Identifying Feelings subscale (TAS_DIF), the difference, -10.85, BCa 95% CI [-14.42, -7.28], was significant, $t(69) = -6.88$, $p < .001$, and represented an effect size of $d = 1.86$; and on the Externally-Oriented Thinking subscale (TAS_EOT), the difference, -5.28, BCa 95% CI [-7.29, -3.30], was also significant, $t(69) = -5.19$, $p < .001$ and represented an effect size of $d = 1.32$.

In summary, Hypothesis i was fully supported, with the FND group participants having, on average, significantly higher scores on the TAS-20 and TAS-20 subscales, than control group participants, with large effect sizes in all cases.

Table 2.

Group mean scores on the TAS-20 and TAS-20 subscales, RFQ Certainty about mental states subscale, RFQ Uncertainty about mental states subscale, PHQ-15 checklist, and Neuro checklist

	FND group (n=29) <i>Mean (SD)</i>	Control group (n=42) <i>Mean (SD)</i>
TAS-20 Total	62.68 (14.24)	40.79 (10.61)
TAS-20 Difficulty Describing Feelings (DDF) subscale	16.93 (4.84)	11.17 (4.45)
TAS-20 Difficulty Identifying Feelings (DIF) subscale	24.68 (7.45)	13.83 (5.82)
TAS-20 Externally Oriented Thinking (EOT) subscale	21.07 (4.57)	15.79 (3.99)
RFQ Certainty about Mental States (RFQ_C) subscale	0.63 (0.72)	1.03 (0.62)
RFQ Uncertainty about Mental States (RFQ_U) subscale	1.26 (0.82)	0.50 (0.43)
PHQ-15 checklist	9.07 (3.41)	4.38 (2.78)
Neuro checklist	4.62 (2.38)	0.48 (1.02)

Hypothesis ii

The second hypothesis was that FND group participants would be more prone to hypomentalizing than control participants, an impairment of reflective functioning characterised by low certainty about mental states (indicated by significantly lower scores on the RFQ_C subscale), and high uncertainty about mental states (indicated by significantly higher scores on the RFQ_U states subscale).

The data supported both elements of this hypothesis. On average, FND group participants had lower scores on the RFQ_C subscale, and the difference, 0.40, BCa 95% CI [0.05, 0.77], was significant, $t(69) = 2.50$, $p = .015$, representing an effect size of $d = -0.65$. FND group participants also, on average, had higher scores on RFQ_U subscale (indicating higher uncertainty about mental states), and this difference, -0.77, BCa 95% CI [-1.10, -0.48], was significant, $t(38.90) = -4.64$, $p < .001$, representing an effect size of $d = 1.78$. This

pattern, of low certainty and high uncertainty about mental states, indicates significantly greater hypomentalyzing in the FND group participants.

Hypothesis iii

The third hypothesis was that FND group participants would have more somatic and neurological symptoms of a generalized nature, than control group participants, as assessed by the PHQ-15 checklist and Neuro checklist.

Consistent with the hypothesis, on average, FND group participants had higher scores on the PHQ15 checklist, and the difference, -4.69 , BCa 95% CI $[-6.06, -3.20]$, was significant, $t(52.03) = -6.13$, $p < .001$, representing an effect size of $d = 1.69$. FND group participants also, on average, had higher scores on the neurological symptoms checklist, and this difference, -4.14 , BCa 95% CI $[-5.03, -3.17]$, was also significant, $t(35.13) = -8.83$, $p < .001$, and represented an effect size of $d = 4.07$.

Hypothesis iv

The fourth hypothesis was that higher levels of alexithymia, and greater use of hypomentalyzing, would predict FND (vs. control) group membership in a logistic regression, when depressive symptoms, anxiety symptoms, and educational attainment were controlled for. Eight separate regression analyses were conducted, with PHQ-8, GAD-7, and educational attainment scores entered in step one of each model, and each of the eight predictors of interest (scores on the TAS_20_TOTAL, TAS_DDF, TAS_DIF, TAS_EOT, RFQ_C, RFQ_U, PHQ-15 checklist, and Neuro checklist) entered in step two.

The results of the omnibus test of model coefficients were examined for each regression, with a significant improvement in model fit after the addition of the predictor variable of interest indicating that this variable was a significant contributor to the model,

above and beyond the effects of the covariates (Open University, 2009). The full details of the regression analyses are presented in Appendix 12, with the key findings in Tables 3-10.

When TAS-20 total score was added as a predictor to a logistic regression model that had previously just included the covariates as predictors, the model fit significantly improved ($\chi^2(1) = 4.74, p = .03$), indicating that alexithymia predicted group (FND vs. control) membership, over and above any effects of the covariates. The Neuro checklist was also predictive of group status after controlling for covariates ($\chi^2(1) = 5.396, p = .02$). However, the model fit was not significantly improved when the TAS_DDF, TAS_DIF, TAS_EOT, RFQ_C, RFQ_U or the PHQ-15 checklist scores were added to the logistic regression as the predictor of interest (all $p > .05$), indicating that these variables were not significant predictors, after controlling for confounding variables.

Table 3. Logistic regression analysis for TAS-20 total score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.203	.143	2.028	1	.154
GAD-7	-.075	.140	.288	1	.592
Educational attainment	-.703	.252	7.808	1	.005
TAS_20_TOTAL	.095	.048	3.967	1	.046
Constant	-2.824				

Table 4. Logistic regression analysis for TAS_DDF score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.301	.142	4.516	1	.034
GAD-7	-.118	.145	.662	1	.416
Educational attainment	-.758	.261	8.454	1	.004
TAS_DDF	.177	.109	2.612	1	.106
Constant	-.627				

Table 5. Logistic regression analysis for TAS_DIF score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.205	.159	1.656	1	.198
GAD-7	-.063	.149	.180	1	.671
Educational attainment	-.744	.254	8.589	1	.003
TAS_DIF	.107	.080	1.778	1	.182
Constant	.144				

Table 6. Logistic regression analysis for TAS_EOT score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.288	.138	4.345	1	.037
GAD-7	-.077	.136	.324	1	.569
Educational attainment	-.622	.251	6.169	1	.013
TAS_EOT	.194	.108	3.242	1	.072
Constant	-2.786				

Table 7. Logistic regression analysis for RFQ_C score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.352	.152	5.340	1	.021
GAD-7	-.087	.145	.357	1	.550
Educational attainment	-.782	.261	8.960	1	.003
RFQ_C	.630	.694	.824	1	.364
Constant	.664				

Table 8. Logistic regression analysis for RFQ_U score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.305	.150	4.160	1	.041
GAD-7	-.095	.144	.436	1	.509
Educational attainment	-.738	.262	7.961	1	.005
RFQ_U	.166	.832	.040	1	.842
Constant	1.328				

Table 9. Logistic regression analysis for PHQ-15 checklist score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.277	.173	2.567	1	.109
GAD-7	-.083	.146	.322	1	.570
Educational attainment	-.756	.258	8.607	1	.003
PHQ-15 checklist	.071	.188	.144	1	.705
Constant	1.257				

Table 10. Logistic regression analysis for Neuro. checklist score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

	B	S.E.	Wald	df	Sig.
PHQ-8	.151	.157	.920	1	.337
GAD-7	-.110	.153	.516	1	.473
Educational attainment	-.558	.271	4.224	1	.040
Neuro checklist	.764	.470	2.642	1	.104
Constant	.503				

Hypothesis v

In order to explore whether alexithymia and hypomentalyzing would predict somatic and neurological symptoms of a more generalized nature, a series of linear regression analyses were undertaken on the whole sample (FND group and healthy control group participants), with TAS-20 total, RFQ_C, and RFQ_U scores as the independent variables, and the PHQ15 checklist and Neuro checklist scores as the dependent variables.

The scatter-plots showed strong *positive* linear relationships between TAS-20 score and PHQ-15 checklist score, and between TAS-20 score and Neuro checklist score (Figure 1). This was confirmed with Pearson's correlation coefficients of .66 and .67, respectively. The regression showed a significant relationship between TAS-20 score and PHQ-15 checklist score ($p < .001$), with a slope coefficient for the TAS-20 of .16. The R^2 value was .440, indicating 44% of the variation in PHQ-15 checklist score can be explained by the model containing only TAS-20 score. The regression also showed a significant relationship between TAS-20 score and Neuro checklist score ($p < .001$), with a slope coefficient for the TAS-20 of .10. The R^2 value was .443, indicating 44.3% of the variation in PHQ-15 checklist score can be explained by the model containing only the TAS-20 score.

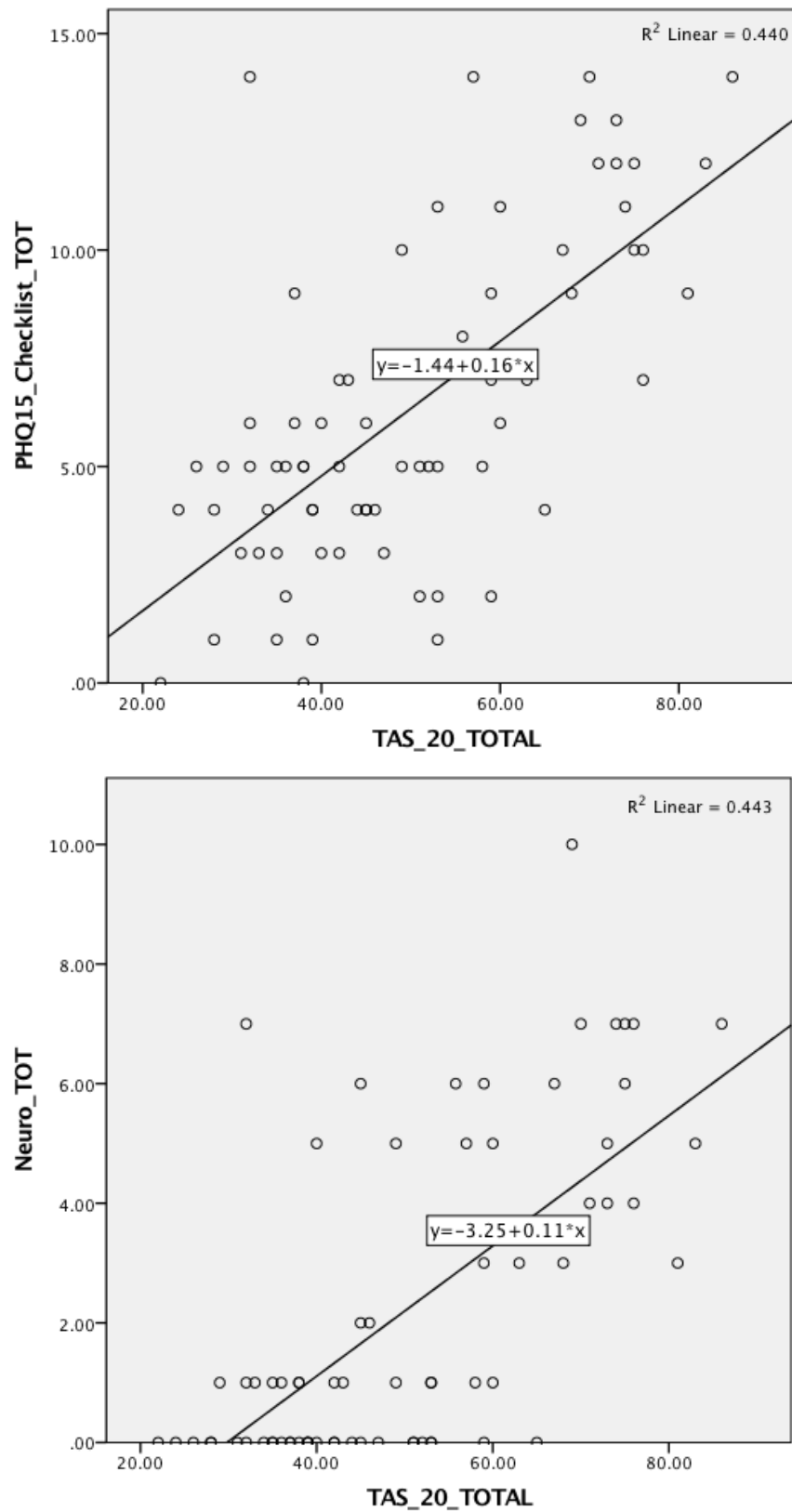


Figure 1. TAS-20 score (TAS_20_TOTAL) as predictor of PHQ15 checklist (PHQ15_Checklist_TOT) and Neuro checklist (Neuro_TOT) scores.

The scatter-plots showed strong *negative* linear relationships between RFQ_C score and PHQ-15 checklist score, and between RFQ_C score and Neuro checklist score (Figure 2). This was confirmed with Pearson's correlation coefficients of $-.48$ and $-.43$, respectively. The regression showed a significant relationship between RFQ_C score and PHQ-15 checklist score ($p < .001$), with a slope coefficient for the RFQ_C of -2.69 . The R^2 value was $.234$, indicating 23.4% of the variation in PHQ-15 checklist score can be explained by the model containing only RFQ_C score. The regression also showed a significant relationship between RFQ_C score and PHQ-15 checklist score ($p < .001$), with a slope coefficient for the RFQ_C of -1.68 . The R^2 value was $.187$, indicating 18.7% of the variation in Neuro checklist score can be explained by the model containing only the RFQ_C score.

As hypothesised, in contrast to the RFQ_C, strong *positive* linear relationships between RFQ_U score and PHQ-15 checklist score, and between RFQ_U score and Neuro checklist score, were shown by this scatterplots (Figure 3). This was confirmed with Pearson's correlation coefficients of $.58$ and $.61$, respectively. The regression showed a significant relationship between RFQ_U score and PHQ-15 checklist score ($p < .001$), with a slope coefficient for the RFQ_U of 3.07 . The R^2 value was $.336$, indicating 33.6% of the variation in PHQ-15 checklist score can be explained by the model containing only RFQ_U score. The regression also showed a significant relationship between RFQ_U score and Neuro checklist score ($p < .001$), with a slope coefficient for the TAS-20 of 2.26 . The R^2 value was $.373$, indicating 37.3% of the variation in Neuro checklist score can be explained by the model containing only the RFQ_U score.

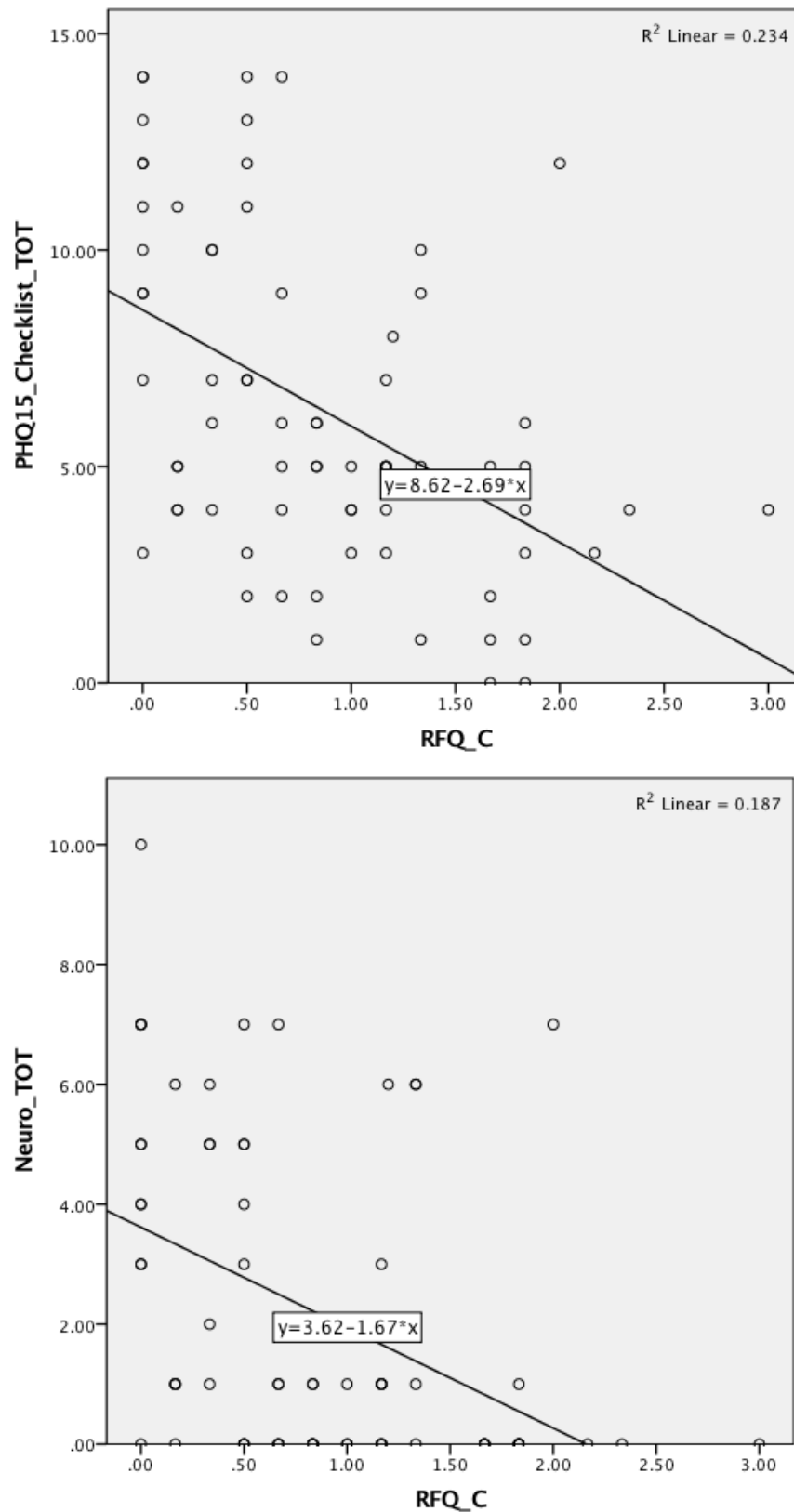


Figure 2. RFQ Certainty about mental states score (RFQ_C) as predictor of PHQ15 checklist (PHQ15_Checklist_TOT) and Neuro checklist (Neuro_TOT) scores.

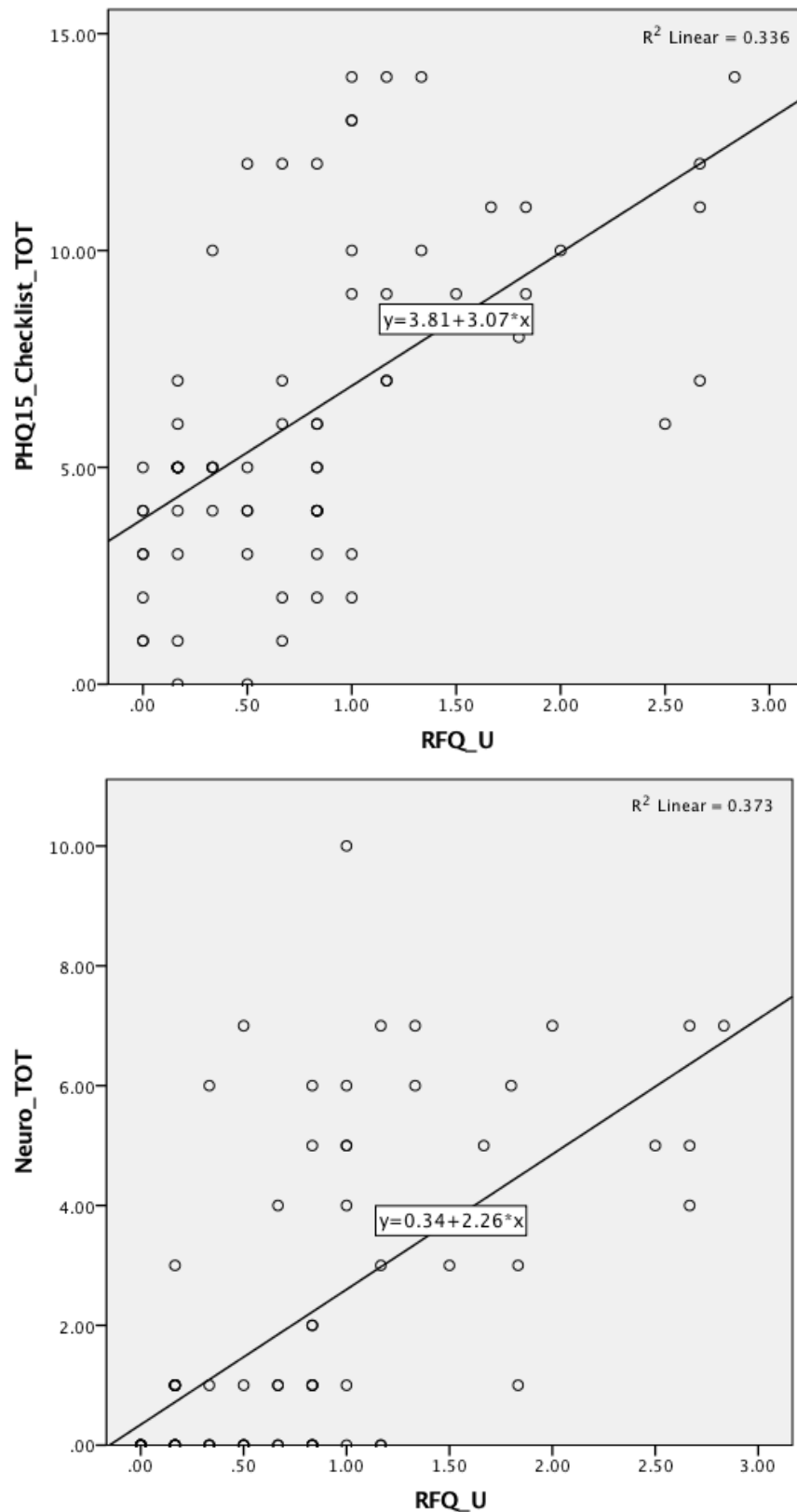


Figure 3. RFQ Uncertainty about mental states score (RFQ_U) as predictor of PHQ15 checklist (PHQ15_Checklist_TOT) and Neuro checklist (Neuro_TOT) scores.

In summary, Hypothesis v was fully supported by the data – higher scores on the PHQ-15 checklist and Neuro checklist (indicating somatic and neurological symptoms of a generalized nature) were associated with higher scores on the TAS-20 (indicating higher alexithymic traits), lower scores on the RFQ_C, and higher scores on the RFQ_U (indicating greater tendency to hypomentalize).

Discussion

The aim of this study was to investigate if alexithymia and impairments in mentalization were more common in people with FND than in control participants, and furthermore, to investigate whether such difficulties would also be predictive of somatic and neurological symptoms of a more generalized nature.

The mean total and subscales scores of the TAS-20 were significantly higher in the FND group than in the control group, suggesting that difficulties with identifying and describing feelings, and externally-oriented thinking, are significant issues for people with FND. These findings are consistent with previous studies of alexithymic traits in people with functional seizures (Kaplan et al., 2013; Urbanek, Harvey, McGowan, & Agrawal, 2014; Novakova, Howlett, Baker, & Reuber, 2015) and with functional motor/sensory symptoms (Demartini et al., 2014; Steffen, Fiess, Schmidt & Rockstroh, 2015; Kienle et al., 2017). This, however, was the first study to explore alexithymia in this type of FND sample (with mixed symptoms), typically seen in UK neuropsychiatry/neuropsychology services. The proportion of the FND group scoring above the clinical cut-off for alexithymia in this study (55.2%) was higher than the proportions reported in recent studies of groups of people with functional motor symptoms (34.5%: Demartini et al., 2014), and in some of the studies of functional seizures (40.4%: Kaplan et al., 2013; 31%: Wolf et al., 2015), though not all (63%; Urbanek et al., 2014). This finding, along with the finding that the mean score of the entire

FND group on the TAS-20 was above the recommended clinical cut-off for the measure (≥ 61), highlights that these findings have clinical, as well as statistical, significance.

Total alexithymia score was also predictive of FND (versus control group) status after controlling for group differences in depressive and anxiety symptoms, and educational attainment, suggesting that group differences in alexithymia were not attributable to these covariates alone. Although the subscales were no longer predictive of group status after controlling for covariates, the p values of the Difficulty Describing Feelings ($p = .093$) and Externally Oriented Thinking ($p = .057$) subscales were approaching significance, possibly indicating a Type II error due to insufficient power.

These results, along with the more clear-cut absence of significant between-group differences on the Difficulty Identifying Feelings subscale ($p = .182$) may also be informative, however. A lack of difficulty with identifying feelings, coupled with impairment in describing feelings, and greater use of externally oriented thinking, may suggest that people with FND are more likely to have Type II alexithymia. Type II alexithymia has been defined by Larsen, Brand, Bermond, and Hijman (2003) as being characterised by selective deficits in emotional cognition, but with a sparing of emotional experience (i.e. people feel the emotions, but have difficulty cognitively understanding them). Higher prevalence of Type II alexithymia has previously been reported to be predictive of somatization in a general population study (Bailey & Henry, 2007). The findings of this study suggest that this may be an area that warrants further research in people with FND too.

Regarding reflective functioning, there were significant between-group differences on both subscales of the RFQ, suggesting that people with FND do experience the absence of normal certainty about mental states that is characteristic of hypomentalizing. It has been hypothesised that the absence of an adaptive level of certainty about one's own and others' mental states may characterize people who are more prone to using affective coping

strategies, rather than cognitive coping strategies, to regulate the emotional arousal induced by everyday events (Badoud et al., 2015). A plausible mechanism for the development of functional symptoms may therefore be that people with FND are more likely to be reliant on affective coping strategies to manage arousal, but these affective strategies are also being influenced by alexithymic traits, with a consequence of them being ‘converted’ in to physical symptoms. The measures of alexithymia and hypomentalization in this study were highly correlated, so this particular hypothesis was not explored further, but it may be worth investigating in future research.

The results from the RFQ should also be interpreted with the caveat that neither subscale was predictive of FND status after controlling for covariates. This may indicate that hypomentalizing is more associated with comorbid affective symptoms and/or lower educational attainment. Alternatively, the study was only powered to find differences with a large effect size, and the use of a brief screening measure of reflective functioning may not have been sufficient to detect group differences. A more comprehensive measure of reflective functioning may have more sensitivity to detect differences in hypomentalizing, and the findings of this study offer support for further research in this area.

The nature of the symptoms (i.e. seizures, motor symptoms, or sensory symptoms) experienced by participants with FND in this study were not evaluated. This was partly because the clinical records of the recruiting services did not consistently record this information – diagnosis was typically recorded as FND when the American Psychiatric Association diagnostic criteria (APA, 2013) were used, or dissociative disorder when the World Health Organisation diagnostic criteria (WHO, 1990) were preferred. It was also partly because comorbidity with other functional and organic symptoms was so common among service users of the recruiting teams. As a result, the findings of this study cannot directly inform Kanaan et al.’s (2017) recent theory (described in the Introduction section),

that there may be etiological differences between functional symptoms that are acute (e.g. seizures) and those that are continuous or chronic (e.g. motor paralyses). According to the measures used in this study, many individual participants within the FND group had significant difficulties with alexithymia and/or hypomentalyzing – an interesting area for future research may be to explore whether particular patterns of impairment (e.g. high alexithymia and high hypomentalyzing vs high alexithymia and low hypomentalyzing) are more strongly associated with particular functional symptom profiles (acute vs chronic), particular types of adverse childhood experiences (e.g. emotional neglect/abuse vs sexual abuse), or another unrelated variable (or variables).

Finally, the results of the linear regression analyses support a hypothesis that alexithymia and hypomentalyzing increase vulnerability to experiencing somatic symptoms, and suggest that this mechanism may not be unique to people with FND. While links between alexithymia and other somatization disorders have previously been described (De Gucht & Heiser, 2003), this study is the first to show that hypomentalyzing is predictive of experiencing more somatic and neurological symptoms. However, these linear regressions were bivariate, and did not control for symptoms of anxiety and depression, or for educational attainment. These confounding variables could, therefore, potentially be more significant predictors of the higher reported somatic and neurological symptoms, than alexithymia or hypomentalyzing. Further research, where these variables are held constant in the analyses (and preferably with larger sample sizes to increase power) would be necessary before definite conclusions on this hypothesis could be drawn.

Limitations

A number of other methodological issues should also be kept in mind in the interpretation of these findings.

Firstly, participants in both groups took part on a voluntary basis. This raises the potential for self-selection bias, particularly as people who find this challenging or uncomfortable to think about emotions or their own/others' mental states may have been less likely to volunteer to take part in the study. As a result, scores on the measures of alexithymia and mentalization may not be an accurate representation of the true population values.

Secondly, FND group participants were all active service users of NHS neuropsychiatry services. The study did not assess where in their treatment participants were (e.g. waiting for a formal assessment vs being monitored following successful treatment), and it is plausible that alexithymic traits or hypomentalyzing may be improved by engagement in psychological treatment (Lumley, Neely, & Burger, 2007).

Thirdly, the FND group participants were highly heterogeneous, with a variety of symptom presentations. Participants with a comorbid neurological problem ('functional overlay') were also included. Previous research on FND has tended to focus on specific symptoms profiles, and some might argue that this FND sample was too heterogeneous varied to make useful findings. However, heterogeneity and complexity of symptoms is widely acknowledged to be the norm rather than the exception in FND (Şar, Akyüz, Kundakçi, Kiziltan, & Doğan, 2004), and the service users invited to participate in this study were representative of typical NHS neuropsychiatry service users, increasing the external validity of the findings.

Fourthly, there were significant differences between the FND and control groups on measures of educational attainment and ethnicity. Educational attainment was controlled for statistically in the analyses, but this was not possible with ethnicity (due to the small number of participants in categories other than White), and this may have been an additional confounding variable.

Finally, this study did not include a measure of childhood abuse/neglect, and therefore does not explicitly implicate such experiences in the subsequent development of alexithymia and/or hypomentalization. An alternative explanation may be that alexithymia/hypomentalization are indicative of a neurodevelopmental condition, such as autism (e.g. a case study reported by Miyawaki et al., 2016). This is an area of research in FND that appears to have been largely overlooked, and may warrant further exploration.

Clinical implications

This study illustrates that alexithymic traits appear to be the norm, rather than the exception, for people with FND. Using a relatively quick measure of alexithymia such as the TAS-20 may provide helpful information in the assessment and treatment planning stages, and guidance as to whether a person may engage better with therapies focusing on insight and emotional awareness, more structured cognitive-behavioural approaches (Lumley et al., 2006), or treatments with a primary focus on physical rehabilitation (McKee et al., 2018). The findings regarding alexithymia and reflective functioning also suggest that treatment approaches that specifically include a focus on affect recognition and management may be helpful for people with FND, such as mentalization-based cognitive therapy or dialectical-behaviour therapy (e.g. the DBT-informed treatment for functional seizures, recently described by Bullock, Mirza, Forte, & Trockel, 2015).

Equally, however, the findings of this study are based on mean group scores, and several of the FND group participants scored well within the normal range on the measures of alexithymia and reflective functioning. A “one size fits all” approach to FND seems highly unlikely given the heterogeneity and complexity of symptoms, and the retention and expansion of multi-disciplinary, individualised, formulation-based approaches (such as those

proposed in the Joint Commissioning Panel for Mental Health guidance, 2017) appears essential.

Conclusion

This study has highlighted that difficulties with identifying and describing feelings, and with uncertainty about their own and others' minds, appear to be common experiences for people with FND. These difficulties may contribute to a tendency to express distress via neurological or physical symptoms, and make understanding and accepting this idiosyncratic coping mechanism challenging. The findings of this study suggest that further exploration of these factors may lead to the development of a more comprehensive theoretical model of FND in future, and more importantly, may help professionals and others working with people with FND to better understand their difficulties, and to offer support that is more engaging and helpful.

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Section C: Appendix of supporting material

Appendix 1

Downs and Black (1998) checklist – *items not relevant to case-control studies italicised*

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Research Ethics Committee (REC) Letter of favourable opinion

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Appendix 3

Letter of approval from Health Research Authority (HRA)

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Appendix 4

REC letter of favourable opinion for study amendment

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Appendix 5

Email confirming Trust Research and Development (R&D) department approval from first
NHS trust

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Appendix 6

Email confirming Trust Research and Development (R&D) department approval from second NHS trust

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Appendix 7

Study questionnaires

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Appendix 8

FND group Participant Information Sheet

**Participant Information Sheet**

Hello. My name is Garret Coy and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study, which I am undertaking as part of the requirement of the Doctoral Programme in Clinical Psychology.

Before you decide, it is important that you understand why the research is being done and what it would involve for you. You can talk to your family, friends, psychologist or doctors about this study if you want to.

Part 1 of this information sheet tells you the purpose of this study and what will happen to you if you take part.

Part 2 gives you more detailed information about the conduct of the study.

Part 1

Formal study details:

Title of Project: Emotional awareness, mentalization, and symptom complexity in functional neurological symptom disorder (FND)

Name of Researcher: Garret Coy

IRAS Project ID: 211216

What is the purpose of the research study?

The broad aim of this study is to explore how people diagnosed with functional neurological disorder (FND) think about their emotions and problems with the physical health. Functional Neurological Disorder (FND) encompasses a diverse range of symptoms, including functional limb weakness and movement disorders, functional and dissociative attacks (non epileptic), sensory problems, cognitive problems, visual and speech symptoms. The symptoms may appear similar to neurological diseases, including those of multiple sclerosis, Parkinson's and epilepsy, and can be just as debilitating, but are not caused by structural disease of the nervous system, and rather appear to arise from a problem with the "functioning" of the nervous system. Further information about FND is available at www.fndaction.org.uk.

It is hoped that this study will lead to greater knowledge and understanding of the condition, and potentially to improved treatments for people with FND.

Why have I been invited to take part?

You have been invited to take part because you have a diagnosis of functional neurological disorder (FND), and are a service user of the Neuropsychiatry Service at

[REDACTED]

In total, 52 participants will take part in this study.

Do I have to take part?

It is your choice whether to take part in the study or not, and your access to treatment will not be affected in any way if you do not wish to take part in the study. If you decide to participate, you may withdraw your consent to further involvement in the research at any time without giving a reason.

What will happen if I decide to take part?

If you decide to take part, you will be asked to complete the consent form and six multiple-choice questionnaires, with questions about your basic details, and questions looking at the different ways that people experience and manage emotions and physical health problems. There are also some optional questions about your experience of taking part in the study, and your response to these questions will be shared with the service user group, the Salomon's Advisory Group of Experts by Experience (SAGE). It should take approximately 30-60 minutes to complete all of the questionnaires. The questionnaires are included with this letter, and can be completed and returned in the post to the address below or in person to your SLAM/KMPT Neuropsychiatry clinic, or can be completed in the clinic during your next appointment. The questionnaires can also be completed online at www.qualtrics.com/websitelinkTBC.

What are the possible disadvantages of taking part?

Some people might find that answering questions regarding their experiences of some emotions, or of ways of managing them, may cause some distress or discomfort. If you experience any distress, you are welcome to discuss this with your clinician at the [REDACTED]. There is also support available to anyone experiencing distress at any time from The Samaritans, who can be contacted at any time by calling 116 123, or visiting their website at www.samaritans.org. If you find it too difficult and upsetting to complete the questionnaires, you can stop at any time.

What are the benefits of taking part?

Your participation will help us to better understand functional neurological symptoms, and the processes that could be contributing to these symptoms. The findings might lead to further research in the area and even the development of better treatments for the problem. There is an option at the end of the questionnaires to leave your contact details if you would like to receive a summary of the findings of the study after it is completed.

To thank you for your time, you will also receive a £10 Amazon gift voucher, if you wish.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my participation in this study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

Part 2

Will my participation in this study be kept confidential?

All data and personal information will be stored securely within NHS and Canterbury Christ Church University premises, in accordance with the Data Protection Act 1998 and the University's own data protection requirements. All information collected about you during the course of the research will be kept strictly confidential, and data will be stored securely on a CD in a locked cabinet. Your answers will not be linked directly to your name. Data can only be accessed by the principal researcher and the supervisors, listed in the next section. The collected data will be used for statistical analysis, the results of which might be published in the future. You will, however, not be identified in any publication. The study documents and data will be disposed of securely after five years, and any documents or electronic files containing personal identifiable information (i.e. your email address) will be destroyed immediately after completion of the study. If you are interested in a summary of findings of this research, please let me know by ticking the box at the end of the consent form, and providing an email address to send the findings to.

Will anyone else be told I am participating in the study?

Your consultant at the [REDACTED] Neuropsychiatry service will be informed that you have participated in the study, for their records. They will only be informed that you have participated, and none of your responses will be shared with them.

Who is organising and funding the research?

This research project is being undertaken as part of the requirement of a Doctoral Programme in Clinical Psychology at Canterbury Christ Church University, with the following people involved:

- **Principal Investigator:** Garret Coy, trainee clinical psychologist, Canterbury Christ Church University
- **Lead research supervisor:** [REDACTED] (Canterbury Christ Church University)
- **Clinical supervisor:** [REDACTED] ([REDACTED])
- **Research consultant:** [REDACTED] ([REDACTED]).

This project is funded by Canterbury Christ Church University.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your safety and well-being. This study has been reviewed and given favourable opinion by the NHS Ethics Committee.

Who do I contact if I need more information?

If there is anything that is not clear, or if you would like more information regarding this study, please feel free to contact me by leaving a message for me on a 24-hour voicemail phone line at 0333 011 7070. Please say that the message is for me [Garret Coy] and leave a contact number so that I can get back to you. You can also email me at g.coy262@canterbury.ac.uk.




What will happen if I don't want to carry on with the study?


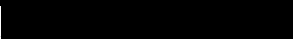
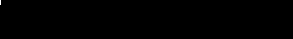
If you wish to withdraw from the study at any time, please contact me by leaving a message on a 24-hour voicemail phone line at 0333 011 7070. Please say that the message is for me [Garret Coy] and leave a contact number so that I can get back to you. You can also email me at g.coy262@canterbury.ac.uk.

What if there is a problem?

If you have a concern about any aspect of this study, please contact me and I will do my best to answer your questions (g.coy262@canterbury.ac.uk or 0333 011 7070). If you remain unhappy and wish to complain formally, you can do this by contacting Professor Paul Camic, Research Director on 0333 011 7114.

You can also contact the Patient Advice and Liaison Service (PALS), who offer confidential advice, support and information on health-related matters. They provide a point of contact for patients, their families and their carers. They can be contacted on:


 **Service Users** - Email: 
 Phone: 

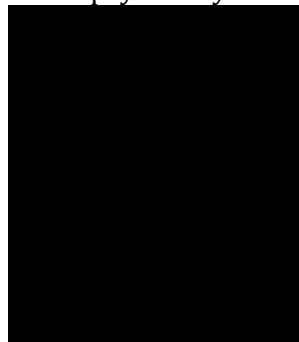
 **Service Users** - Email: 
 Phone: 

Further information about taking part in NHS research is also available on the NHS Choices website, at <http://www.nhs.uk/Conditions/Clinical-trials/Pages/Introduction.aspx>.

What if I want to take part?

If you have now decided that you want to take part in the study, thank you! I hope that you will find it to be an interesting experience.

To take part, please read and sign the enclosed consent form, and then complete the questionnaires and bring them to your next appointment at the  Neuropsychiatry clinic, or return them by post to:



You can also complete the questionnaires online by visiting www.tinyurl.com/FNDStudy17GC and using the following unique ID code when prompted:

.....

Don't forget – If you wish to receive the £10 Amazon gift voucher, please tick the relevant box on the consent form and provide an email address where the voucher can be sent.

Thank you for considering your participation in this study.

Appendix 9

FND group consent form



Office use only:
 Date received:
 Participant ID:

CONSENT FORM

Title of Project: Emotional awareness, mentalization, and symptom complexity in functional neurological symptom disorder (FND)
Name of Researcher: Garret Coy
IRAS Project ID: 211216

Please tick box

MANDATORY Questions (Required for participation in the study)

- 1. I confirm that I have read and understand the Participant Information Sheet, dated 21/7/17 (Version 5), for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
- 2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
- 3. I understand that relevant sections of data collected during the study may be looked at by the lead supervisors, [REDACTED] [REDACTED] and [REDACTED]. I give permission for these individuals to have access to my data.
- 4. I agree to my consultant in the [REDACTED] [REDACTED] service being informed of my participation in the study.
- 5. I agree to take part in the above study.

Name of Participant _____ Date _____

Signature _____

Name of Person taking consent (if applicable) _____ Date _____

Signature _____

Please tick box

OPTIONAL Questions (Not required for participation in the study)

1. I wish to provide feedback about my experience of taking part in the study, and agree for direct quotes from my feedback to be shared anonymously with the service user group, the Salomon's Group of Experts by Experience (SAGE).
2. I wish to receive a £10 Amazon e-voucher to thank me for the time I spent taking part in the study.
3. I wish to receive a summary of the results of the study, once it has been completed.

If you have ticked the box corresponding to OPTIONAL Questions 2 or 3 above, please provide an email address where you can be contacted with your e-voucher and/or the summary of the results of the study*:

Name of Participant _____ Date _____

Signature _____

**In accordance with the Data Protection Act, the email address provided will be used only for the purposes for which consent has been provided, and will not be stored after completion of the study.*

Appendix 10

Control group Participant Information Sheet

**Participant Information Sheet**

Hello. My name is Garret Coy and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study, which I am undertaking as part of the requirement of the Doctoral Programme in Clinical Psychology.

Before you decide, it is important that you understand why the research is being done and what it would involve for you. You can talk to your family, friends, psychologist or doctors about this study if you want to.

Part 1 of this information sheet tells you the purpose of this study and what will happen to you if you take part.

Part 2 gives you more detailed information about the conduct of the study.

Part 1

Formal study details:

Title of Project: Emotional awareness, mentalization, and symptom complexity in functional neurological symptom disorder (FND)

Name of Researcher: Garret Coy

IRAS Project ID: 211216

What is the purpose of the research study?

The broad aim of this study is to explore how people diagnosed with functional neurological disorder (FND) think about their emotions and problems with the physical health. Functional Neurological Disorder (FND) encompasses a diverse range of symptoms, including functional limb weakness and movement disorders, functional and dissociative attacks (non epileptic), sensory problems, cognitive problems, visual and speech symptoms. The symptoms may appear similar to neurological diseases, including those of multiple sclerosis, Parkinson's and epilepsy, and can be just as debilitating, but are not caused by structural disease of the nervous system, and rather appear to arise from a problem with the "functioning" of the nervous system. Further information about FND is available at www.fndaction.org.uk.

It is hoped that this study will lead to greater knowledge and understanding of the condition, and potentially to improved treatments for people with FND.

Why have I been invited to take part?

You have been chosen to take part to form part of the control group, against whom we will compare the results of the group of people with FND. In total, 52 participants will take part in this study.

Do I have to take part?

It is your choice whether to take part in the study or not. If you decide to participate, you may withdraw your consent to further involvement in the research at any time without giving a reason.

What will happen if I decide to take part?

If you decide to take part, you will be asked to complete the consent form and six multiple-choice questionnaires, with questions about your basic details, and questions looking at the different ways that people experience and manage emotions and physical health problems. There are also some optional questions about your experience of taking part in the study, and your response to these questions will be shared with the service user group, the Salomon's Advisory Group of Experts by Experience (SAGE). It should take approximately 30-60 minutes to complete all of the questionnaires.

The questionnaires can all be completed online by following this link:

www.qualtrics.com/FNDstudyGC18

What are the possible disadvantages of taking part?

Some people might find that answering questions regarding their experiences of some emotions, or of ways of managing them, may cause some distress or discomfort. There is support available to anyone experiencing distress at any time from The Samaritans, who can be contacted at any time by calling 116 123, or visiting their website at www.samaritans.org. If you find it too difficult and upsetting to complete the questionnaires, you can stop at any time.

What are the benefits of taking part?

Your participation will help us to better understand functional neurological symptoms, and the processes that could be contributing to these symptoms. The findings might lead to further research in the area and even the development of better treatments for the problem. There is an option at the end of the questionnaires to leave your contact details if you would like to receive a summary of the findings of the study after it is completed.

To thank you for your time, you can also be entered in to a prize draw to win one of two Amazon gift vouchers, worth £25 or £10, if you wish.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my participation in this study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

Part 2

Will my participation in this study be kept confidential?

All data and personal information will be stored securely within NHS and Canterbury Christ Church University premises, in accordance with the Data Protection Act 1998 and the University's own data protection requirements. All information collected about you during the course of the research will be kept strictly confidential, and data will be stored securely on a CD in a locked cabinet. Your answers will not be linked directly to your name. Data can only be accessed by the principal researcher and the supervisors, listed in the next section. The collected data will be used for statistical analysis, the results of which might be published in the future. You will, however, not be identified in any publication. The study documents and data will be disposed of securely after five years, and any documents or electronic files containing personal identifiable information (i.e. your email address) will be destroyed immediately after completion of the study. If you are interested in a summary of findings of this research, please let me know by ticking the box at the end of the consent form, and providing an email address to send the findings to.

Who is organising and funding the research?

This research project is being undertaken as part of the requirement of a Doctoral Programme in Clinical Psychology at Canterbury Christ Church University, with the following people involved:

- **Principal Investigator:** Garret Coy, trainee clinical psychologist, Canterbury Christ Church University
- **Lead research supervisor:** [REDACTED] (Canterbury Christ Church University)
- **Clinical supervisor:** [REDACTED] ([REDACTED])
- **Research consultant:** [REDACTED] ([REDACTED]).

This project is funded by Canterbury Christ Church University.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your safety and well-being. This study has been reviewed and given favourable opinion by the NHS Ethics Committee.

Who do I contact if I need more information?

If there is anything that is not clear, or if you would like more information regarding this study, please feel free to contact me by leaving a message for me on a 24-hour voicemail phone line at 0333 011 7070. Please say that the message is for me [Garret Coy] and leave a contact number so that I can get back to you. You can also email me at g.coy262@canterbury.ac.uk.

What will happen if I don't want to carry on with the study?

If you wish to withdraw from the study at any time, please contact me by leaving a message on a 24-hour voicemail phone line at 0333 011 7070. Please say that the

message is for me [Garret Coy] and leave a contact number so that I can get back to you. You can also email me at g.coy262@canterbury.ac.uk.

What if there is a problem?

If you have a concern about any aspect of this study, please contact me and I will do my best to answer your questions (g.coy262@canterbury.ac.uk or 0333 011 7070). If you remain unhappy and wish to complain formally, you can do this by contacting Professor Paul Camic, Research Director on 0333 011 7114.

You can also contact the Patient Advice and Liaison Service (PALS), who offer confidential advice, support and information on health-related matters. They provide a point of contact for patients, their families and their carers. They can be contacted on:

[REDACTED]

Further information about taking part in NHS research is also available on the NHS Choices website, at <http://www.nhs.uk/Conditions/Clinical-trials/Pages/Introduction.aspx>.

What if I want to take part?

If you have now decided that you want to take part in the study, thank you! I hope that you will find it to be an interesting experience.

To take part, please read and agree to the electronic consent form, and then complete the online questionnaires, by visiting www.qualtrics.com/FNDstudyGC18

Don't forget - If you wish to enter the prize draw to win an Amazon voucher, please tick the relevant box on the consent form and provide an email address where you can be contacted about the outcome of the prize draw.

Thank you for considering your participation in this study.

Appendix 11

Control group consent form



Office use only:
Date received:
Participant ID:

CONSENT FORM

NOTE: Consent for Control Group participants will be obtained via the online questionnaire, so this form is intended only to be representative of the questions and information that will comprise that form.

Title of Project: Emotional awareness, mentalization, and symptom complexity in functional neurological symptom disorder (FND)

Name of Researcher: Garret Coy

IRAS Project ID: 211216

Please tick box

MANDATORY Questions (Required for participation in the study)

- 1. I confirm that I have read and understand the Participant Information Sheet, dated 21/7/17 (Version 5), for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
- 2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
- 3. I understand that relevant sections of data collected during the study may be looked at by the lead supervisors, [REDACTED] and [REDACTED] I give permission for these individuals to have access to my data.
- 4. I agree to take part in the above study.

Name of Participant _____ Date _____

Signature _____

Name of Person taking consent (if applicable) _____ Date _____

Signature _____

Please tick box

OPTIONAL Questions (Not required for participation in the study)

1. I wish to provide feedback about my experience of taking part in the study, and agree for direct quotes from my feedback to be shared anonymously with the service user group, the Salomon's Group of Experts by Experience (SAGE).

2. I wish to enter the prize draw to win a £25 or £10 Amazon e-voucher to thank me for the time I spent taking part in the study.

3. I wish to receive a summary of the results of the study, once it has been completed.

If you have ticked the box corresponding to OPTIONAL Questions 2 or 3 above, please provide an email address where you can be contacted with the results of the prize draw for the Amazon e-voucher, and/or the summary of the results of the study*:

Name of Participant _____ Date _____

Signature _____

**In accordance with the Data Protection Act, the email address provided will be used only for the purposes for which consent has been provided, and will not be stored after completion of the study.*

Appendix 12

Binary logistic regression SPSS outputs

Block 0: Beginning Block**Variables not in the Equation**

			Score	df	Sig.
Step 0	Variable	PHQ8_TOT	32.884	1	.000
	s	GAD7_TOT	22.491	1	.000
		Highest educational qualification	25.950	1	.000
Overall Statistics			39.664	3	.000

Block 1: Method = Enter**Omnibus Tests of Model Coefficients**

		Chi-square	df	Sig.
Step 1	Step	48.991	3	.000
	Block	48.991	3	.000
	Model	48.991	3	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step 1 ^a	PHQ8_TOT	.316	.141	5.046	1	.025	1.371	1.041	1.807
	GAD7_TOT	-.094	.143	.429	1	.512	.910	.688	1.205
	Highest educational qualification	-.747	.257	8.421	1	.004	.474	.286	.785
	Constant	1.401	1.476	.901	1	.343	4.059		

a. Variable(s) entered on step 1: PHQ8_TOT, GAD7_TOT, Highest educational qualification.

Logistic regression analysis for TAS-20 total score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	4.735	1	.030
	Block	4.735	1	.030
	Model	53.727	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step 1 ^a	PHQ8_TOT	.203	.143	2.028	1	.154	1.225	.926	1.621
	GAD7_TOT	-.075	.140	.288	1	.592	.927	.704	1.221
	Highest educational qualification	-.703	.252	7.808	1	.005	.495	.302	.811
	TAS_20_TOTAL	.095	.048	3.967	1	.046	1.100	1.002	1.208
	Constant	-2.824	2.428	1.353	1	.245	.059		

a. Variable(s) entered on step 1: TAS_20_TOTAL.

Logistic regression analysis for TAS_DDF score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	2.823	1	.093
	Block	2.823	1	.093
	Model	51.814	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step	PHQ8_TOT	.301	.142	4.516	1	.034	1.351	1.024	1.784
1 ^a	GAD7_TOT	-.118	.145	.662	1	.416	.889	.670	1.180
	Highest educational qualification	-.758	.261	8.454	1	.004	.468	.281	.781
	TAS_DDF	.177	.109	2.612	1	.106	1.193	.963	1.478
	Constant	-.627	1.852	.115	1	.735	.534		

a. Variable(s) entered on step 1: TAS_DDF.

Logistic regression analysis for TAS_DIF score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	1.813	1	.178
	Block	1.813	1	.178
	Model	50.804	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step	PHQ8_TOT	.205	.159	1.656	1	.198	1.227	.898	1.677
1 ^a	GAD7_TOT	-.063	.149	.180	1	.671	.939	.701	1.257
	Highest educational qualification	-.744	.254	8.589	1	.003	.475	.289	.782
	TAS_DIF	.107	.080	1.778	1	.182	1.113	.951	1.303
	Constant	.144	1.696	.007	1	.932	1.155		

a. Variable(s) entered on step 1: TAS_DIF.

Logistic regression analysis for TAS_EOT score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	3.637	1	.057
	Block	3.637	1	.057
	Model	52.628	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step 1 ^a	PHQ8_TOT	.288	.138	4.345	1	.037	1.333	1.017	1.747
	GAD7_TOT	-.077	.136	.324	1	.569	.926	.709	1.208
	Highest educational qualification	-.622	.251	6.169	1	.013	.537	.328	.877
	TAS_EOT	.194	.108	3.242	1	.072	1.214	.983	1.499
	Constant	-2.786	2.566	1.179	1	.278	.062		

a. Variable(s) entered on step 1: TAS_EOT.

Logistic regression analysis for RFQ_C score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	.811	1	.368
	Block	.811	1	.368
	Model	49.803	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step	PHQ8_TOT	.352	.152	5.340	1	.021	1.422	1.055	1.916
1 ^a	GAD7_TOT	-.087	.145	.357	1	.550	.917	.690	1.218
	Highest educational qualification	-.782	.261	8.960	1	.003	.458	.274	.764
	RFQ_C	.630	.694	.824	1	.364	1.877	.482	7.312
	Constant	.664	1.649	.162	1	.687	1.943		

a. Variable(s) entered on step 1: RFQ_C.

Logistic regression analysis for RFQ_U score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	.040	1	.841
	Block	.040	1	.841
	Model	49.031	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step	PHQ8_TOT	.305	.150	4.160	1	.041	1.357	1.012	1.819
1 ^a	GAD7_TOT	-.095	.144	.436	1	.509	.909	.685	1.206
	Highest educational qualification	-.738	.262	7.961	1	.005	.478	.286	.798
	RFQ_U	.166	.832	.040	1	.842	1.180	.231	6.031
	Constant	1.328	1.529	.754	1	.385	3.772		

a. Variable(s) entered on step 1: RFQ_U.

Logistic regression analysis for PHQ15 Checklist score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	.145	1	.704
	Block	.145	1	.704
	Model	49.136	4	.000

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
Step 1 ^a	PHQ8_TOT	.277	.173	2.567	1	.109	1.319	.940	1.850
	GAD7_TOT	-.083	.146	.322	1	.570	.920	.691	1.226
	Highest educational qualification	-.756	.258	8.607	1	.003	.470	.284	.778
	PHQ15_Checklist_TOT	.071	.188	.144	1	.705	1.074	.743	1.552
	Constant	1.257	1.511	.692	1	.406	3.514		

a. Variable(s) entered on step 1: PHQ15_Checklist_TOT.

Logistic regression analysis for Neuro Checklist score predicting FND group status (FND diagnosis present or absent), controlling for covariates (depressive symptoms, anxiety symptoms, and educational attainment)

Block 2: Method = Enter

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	5.396	1	.020
	Block	5.396	1	.020
	Model	54.387	4	.000

Variables in the Equation

Step		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I. for EXP(B)	
								Lower	Upper
1 ^a	PHQ8_TOT	.151	.157	.920	1	.337	1.163	.854	1.582
	GAD7_TOT	-.110	.153	.516	1	.473	.896	.664	1.209
	Highest educational qualification	-.558	.271	4.224	1	.040	.572	.336	.974
	Neuro_TOT	.764	.470	2.642	1	.104	2.148	.854	5.398
	Constant	.503	1.653	.093	1	.761	1.654		

a. Variable(s) entered on step 1: Neuro_TOT.

Appendix 13

Author submission guidelines for the Journal of Psychosomatic Research

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Appendix 14

Declaration of end of study

DECLARATION OF THE END OF A STUDY

(For all studies except clinical trials of investigational medicinal products)

To be completed in typescript by the Chief Investigator and submitted to the Research Ethics Committee (REC) that gave a favourable opinion of the research within 90 days of the conclusion of the study or within 15 days of early termination. For questions with Yes/No options please indicate answer in bold type.

1. Details of Chief Investigator

Name:	Garret Coy
Address:	██
Telephone:	██
Email:	██
Fax:	n/a

2. Details of study

Full title of study:	Emotional awareness, mentalization and symptom complexity in functional neurological symptom disorder (FND)
Research sponsor:	Prof. Paul Camic, Canterbury Christ Church University
Name of REC:	East of England REC
REC reference number:	17/EE/0191

3. Study duration

Date study commenced:	06/08/2017
Date study ended:	25/07/2018
Did this study terminate prematurely?	No <i>If yes, please complete sections 4, 5, 6, & 7. If no, please go direct to section 8.</i>

4. Recruitment

Number of participants recruited	
----------------------------------	--

Proposed number of participants to be recruited at the start of the study	
If different, please state the reason or this	

5. Circumstances of early termination

What is the justification for this early termination?	
---	--

6. Temporary halt

Is this a temporary halt to the study?	Yes / No
If yes, what is the justification for temporarily halting the study? When do you expect the study to re-start?	<i>e.g. Safety, difficulties recruiting participants, trial has not commenced, other reasons.</i>

7. Potential implications for research participants

Are there any potential implications for research participants as a result of terminating/halting the study prematurely? Please describe the steps taken to address them.	
--	--

8. Final report on the research

Is a summary of the final report on the research enclosed with this form?	Yes <i>If no, please forward within 12 months of the end of the study.</i>
---	--

9. Declaration

Signature of Chief Investigator:	X
Print name:	Garret Coy
Date of submission:	27/08/2018

Appendix 15

Final study report for REC and study participants

**Research study final report**

Study title: **Emotional awareness, mentalization and symptom complexity in functional neurological symptom disorder (FND)**

What was being studied?

Previous research with people with functional neurological disorder (FND) has suggested that they may have difficulty with identifying and describing emotions (a concept known as 'alexithymia'). Other research has suggested that people who have difficulty understanding their own and others' mental states (a concept known as hypomentalization) may also have difficulty with working out which bodily sensations belong to which emotions. This study aimed to investigate if alexithymia and hypomentalization were difficulties for people with FND in the UK.

How was this studied?

A group of service users with FND, and a group of people who did not have FND (referred to as control group participants) completed a series of questionnaires. These questionnaires asked participants about how they thought about their emotions, and how they thought about their own minds and the minds of others. There were also questions about current symptoms of low mood and anxiety, and about symptoms of different neurological and physical health conditions. In total, 29 people with FND and 41 control participants took part. The questionnaire scores of the two groups were then compared, to explore whether there were significant differences between the groups.

What were the results?

Participants with FND did, on average, report more difficulties with identifying and describing emotions (alexithymia), and with understanding their own and others' mental states (hypomentalization), than control group participants. The participants with FND in

this study also, on average, had significantly more symptoms of low mood and anxiety than control group participants, as well as significantly fewer years spent in formal education. After these differences were factored in to the analyses, people with FND still, on average, had significantly higher scores on the alexithymia questionnaire, but the groups were no longer significantly different on the hypomentalization questionnaire.

When the scores of everyone that had taken part in the study were explored further, it was found that higher levels of alexithymia and hypomentalization were associated with higher numbers of neurological and physical health complaints. This suggests that alexithymia and hypomentalization may contribute to anyone experiencing problems with their neurological or physical health, and that this process is not unique to people with FND.

What does this mean for people with FND?

Difficulties with identifying and describing emotions, and with understanding their own and others' mental states, appear to be common problems for people with FND. As a result, it may be helpful for clinicians working with people with FND to explore these issues with service users. Questionnaires such as those used in this study could help with that exploration. This may help clinicians to think about how best to explain FND to people, and how to make sure that they are offering support that the person will find meaningful and helpful.

The findings of this study may also help other researchers and people working with people with FND to better understand why people develop FND, and other conditions with symptoms like it. A relatively small number of people took part in this particular study, and the participants with FND, on average, differed from the participants without FND in some other ways too (such as their current symptoms of low mood and anxiety, and their educational histories). As a result, it is not possible to say that the findings of this study apply to all people with FND. The results do suggest that exploration of the roles of alexithymia and hypomentalization in the thinking styles of people with FND may be a helpful area to research further in the future.

Thank you for taking part in the study. Your participation has helped us to understand FND a little better. I hope that you found taking part in the study interesting too.

Appendix 16

Consent procedure for the study

As per the guidance provided in the Canterbury Christ Church University Doctorate in Clinical Psychology Guidelines for the preparation of the Major Research Project (Assessment Handbook, Appendix 21), and the study protocol that was approved by the East of England Research Ethics Committee (REC Reference no. 17/EE/0191), the informed consent forms completed by study participants have been stored as follows:

- 1) Written consent forms, completed by 23 participants from the functional neurological disorders group, were submitted to the University Research Administrator in a sealed envelope for storage by the University, on 27th April, 2018.
- 2) Consent forms for the remaining 6 participants from the functional neurological disorders group, and from all 42 participants of the healthy control group, were obtained electronically via the University-approved Qualtrics online survey website. The University Research Director, Prof Paul Camic, was emailed on 26th April 2018, as per the University guidelines, to confirm that this was the reason why written consent forms were not being submitted for 48 study participants.