Is there a relationship between maladaptive schemas and functional neurological disorders?

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Table of Contents

Executive Summary

The Systematic Review

'Functional Neurological Disorders' (FND) is an umbrella term which describes the presence of neurological symptoms in the absence of any demonstrable neurological disease or injury. The symptoms of FND are varied, and can include non-epileptic seizures and movement difficulties, such as limb weakness or gait problems. Functional movement difficulties are referred to using the collective term functional motor symptoms. As well as non-epileptic seizures and functional motor symptoms, FND can also encompass disorders of voice and swallowing, memory problems, visual difficulties, and sensory problems. People with FND may present with one or several of these symptoms (Stone, 2013).

FND are a common presentation in neurology clinics (Stone, Warlow & Sharpe, 2010). Research suggest that people with FND have higher levels of distress and disability compared to populations with non-functional neurological diseases (Carson *et al*., 2011). In spite of the psychosocial impact of experiencing functional neurological symptoms and their potential cost to healthcare systems, FND are often described as poorly understood in the research literature. Similarly, the evidence base for psychological interventions in FND is limited (Martlew, Pulman & Marson, 2014).

There are a variety of psychological models of FND - including dissociative, psychodynamic, and cognitive-behavioural accounts – but a recent comprehensive systematic review by Brown and Reuber (2016) concluded that, regarding non-epileptic seizures at least, the evidence for all models is limited and inconsistent. Other reviews concerning FND have

examined a variety of issues, such as neuropsychological dysfunction, comorbidities with post-traumatic stress disorder and personality disorder, and the prevalence of traumatic experiences in those with the condition.

The present review focuses on a group of studies which were not covered by Brown and Reuber's (2016) review. It examines studies which compare groups with specific types of FND (such as participants with non-epileptic seizures only), based on their psychological characteristics. Brown and Reuber assert that this type of study has made an important contribution to understanding the psychology of FND and they are in need of systematic scrutiny. They hypothesise that, in comparison to individuals with only functional motor symptoms, people with non-epileptic seizures alone are younger, more likely to experience alterations in consciousness, and more likely to have experienced childhood abuse or stressful life events.

Searches of PsycINFO and PubMed electronic databases were carried out on the 9th March 2019. No publication date restrictions were applied. Only empirical quantitative studies of participants over 18 years old reported in English were included. Small sample ($N < 15$) designs were excluded.

Seven studies were included in the review, and all studies compared participants with non-epileptic seizures only and participants with functional motor symptoms only. All studies used clinical samples, and in three of the studies control groups of various types were also used. The total number of participants was 793.

The studies examined a variety of variables and had relatively few measures in common. Due to the heterogeneity of designs in the identified studies, a narrative synthesis approach was adopted. This involved tabulating the studies, appraising their research quality, exploring and synthesising relationships between their results, considering issues around the variability of findings, and critically appraising the synthesis itself.

The research quality of six of studies was rated as 'moderate', with one study rated as 'poor'. The main research issues which were present in the studies deemed 'moderate' in quality were the following: lack of power calculations; lack of information about the source of the sample (e.g. inpatient or outpatient settings); a focus on self-report measures; and lack of control groups.

The results of the review were that, taken as whole, patients presenting with these two types of FND tend to be female, have poorer self-reported mental and physical health, and elevated levels of depression, anxiety and alexithymia. Regarding the differences between the two types of FND, participants with non-epileptic seizures tended to be younger, and more likely to report traumatic or adverse experiences (particularly childhood trauma, sexual abuse and stressful life events). There was also some limited evidence regarding differences in personality traits and tendencies towards dissociative experience. This largely confirmed Brown and Reuber's (2016) predictions, thought there was little evidence concerning alterations to consciousness.

There were several limitations associated with the conclusions of the systematic review. The number of studies identified was small and they had relatively few measures in common. No study was rated 'strong' in quality due to methodological failings. The review was also unable to identify any studies comparing any combination of FND other than non-epileptic seizures and functional motor symptoms. This means it is unable to draw conclusions about the psychological characteristics of other types of FND (e.g. functional voice disorders). A methodological failing of the review itself was the failure to incorporate a second reviewer, which may have heightened the risk of bias at various stages of the review process. Nonetheless, the review was developed in accordance with best practice guidelines and existing systematic reviews, which arguably provided mitigation against the risk of bias.

The review recommends that future studies should seek to improve research quality in this area. Future research would benefit from following best practice research guidelines in regard to using and reporting power calculations. Furthermore, improvement is needed in terms of better characterising samples with FND to increase knowledge about the factors affecting variability in FND populations, such as the severity and complexity of functional symptoms manifesting within a sample. Further studies of this type also could use gender-balanced or exclusively male samples, to explore the extent to which findings are replicated in these groups.

Future research of this kind could also include comparisons of other types of functional neurological presentations, and incorporate groups with mixed functional neurological presentations. Comparative studies like these appear not to have yet been attempted.

Moreover, there may be a need for more comparative FND studies focusing on issues relevant to the cognitive-behavioural model of the disorder, given that this is the most likely kind of psychological intervention to be offered to those with the condition (Stone, 2010).

The Empirical Project

FND have been associated with a range of predisposing psychosocial factors. These include elevated rates of adverse experiences, emotional dysregulation and alexithymia. In addition, studies have also linked FND with elevated rates of personality disorders. The most consistently replicated findings associate FND with Borderline Personality Disorder (BPD) and Obsessive-Compulsive Personality Disorder (OCPD).

This project explores the utility of the theories associated with schema therapy (hereafter 'the schema model') as an explanatory model for understanding these findings. Schema therapy was developed for clients with long-standing, complex psychological difficulties, such as personality disorders. Schema therapy defines maladaptive schemas as a range of deep-rooted, self-perpetuating, cognitive-emotional processes which people use to cope with their emotions or relationships (Young, Klosko & Weishaar, 2003).

Based on the existing associations found between FND, BPD and OCPD, it was predicted that certain clusters of schemas, known as the Disconnection and Rejection and Excessive Responsibility schema domains, would be implicated in functional neurological difficulties. Consequently, it was hypothesised that scores for these schema domains would be negatively correlated with scores on a self-report measure of current health status in a

clinical sample of participants with FND. A secondary hypothesis, based on the schema model, was that the extent to which certain participants endorsed experiencing forms of unhelpful parenting in childhood would also be negatively correlated with their current health status. Lastly, it was predicted that scores for the identified schema domains in a clinical sample with FND would be significantly elevated when compared to large-scale data from a previous study (Bach, Lockwood & Young, 2018). This earlier study measured the schema profiles of two groups: a student sample and a sample of psychiatric outpatients described as having personality disorder traits.

The Empirical Study adopted a quantitative, cross-sectional design. In total, 25 participants with FND were recruited from inpatient, outpatient and day-patient neuropsychiatry services in South London and Maudsley NHS Foundation Trust. A power calculation based on finding large effect sizes for the first two hypotheses had suggested that 28 participants were needed. This meant that the study was likely to be somewhat underpowered. Participants completed a single set of standardised self-report questionnaires. Initial questionnaires measured current health status and levels of anxiety and depression. Participants then completed a screening questionnaire for personality disorder, followed by questionnaires measuring maladaptive schemas and perceived parenting experiences during childhood.

The first two hypotheses were analysed using correlations. As the health status data was found to be non-normally distributed, non-parametric correlations were used. There were small negative correlations between the identified schema domains and current health

status, but these correlations were not statistically significant. The correlations between the identified perceived parenting styles and current health status were also not statistically significant.

However, regarding the last hypothesis, the identified schema domain scores were found to be elevated in comparison to data from Bach *et al*.'s (2018) study. Specifically, the Disconnection and Rejection scores were significantly higher than those of the student sample, whereas the Excessive Responsibility scores were significantly higher than the student sample and not significantly different from those of the clinical sample. Visual analysis of individual schema subscales was then pursued to understand the ratings driving these results.

Elevated scores comparable with the clinical sample were noted for the following individual schemas: Pessimism, Failure to Achieve, Vulnerability to Harm, Self-Sacrifice and Self-Punitiveness. Two other schema subscales (Entitlement and Approval-Seeking) appeared to be lower than scores in the student sample. Overall, this was thought to suggest the following themes about the schema profile of the sample: inflexibility concerning internalised rules; anticipation of negative consequences; an exaggerated drive towards achievement, and a perceived need to sacrifice one's own needs in favour of those of other people. These findings were consistent with those from the personality disorder screening measure. This data suggested the sample contained elevated rates of Anankastic Personality Disorder; the criteria for this condition resemble these themes.

There were a number of limitations which applied to any conclusions which can be drawn from this project. Firstly, the fact that the study was underpowered to detect even large effect sizes might have heightened the risk of type II errors with regard to the first two hypotheses. Secondly, the measure of current health status used may have been confounded by the influence of other factors on a participant's health, thus limiting the precision with which it could measure the hypothesised effects. Thirdly, the large groups which the data was compared with to evaluate the third hypothesis were poorly defined, so it is not entirely clear what similarity or dissimilarity compared to these groups might mean. Lastly, it had been intended that formal service-user involvement would be used to inform the project. Unfortunately, due to time constraints this was not possible during data collection.

Overall the results of the Empirical Study suggested that there may be elevations in certain maladaptive schemas in people with FND. More research is warranted to further explore this result in larger samples. Given the findings of the systematic review, future research could also investigate comparisons of schema profiles in different types of FND, in order to gauge if differences are present.

Integration, Impact and Dissemination

The Systematic Review's conclusions helped to inform the development of the Empirical Project. The review highlighted a range of findings regarding the increased prevalence of adverse experiences and maladaptive emotional-relational coping strategies in FND. It also suggested that these findings appear to have some variability related to other factors, such

as the type of FND studied. It is argued, that, taken as a whole, these findings suggest that the schema model may have some application to understanding FND.

There were a number of challenges in conducting these two projects. One was arriving at an appropriate aim for the systematic review which was achievable within the time available, while not replicating an existing systematic review. Another was gaining ethical approval, which required liaison with several regulatory bodies and careful consideration of complex ethical issues. This created further challenges, such as limiting the time that was available for recruitment and service-user involvement. On reflection, aspects of the empirical project (such as being based on an original idea which took time to develop, and use of a clinical sample) involved greater challenges than might have been encountered otherwise. However, overall it was felt that a higher degree of learning and development was achieved through finding solutions to these problems.

These findings have potential impact in a number of areas. If further research substantiated a role of maladaptive schemas in FND, then this has the potential to inform future interventions. For instance, cognitive-behavioural interventions could be supplemented by schema therapy approaches and techniques, particularly because of the common-ground shared by the two approaches. The findings also highlight the potential clinical complexity of FND patients, which may have implications for the services that care for them. A multi-disciplinary team approach, reflective practice forums, and the opportunity to consult with psychological professionals may help staff teams cope with this complexity.

It is intended that the Systematic Review and Empirical Project will be disseminated in two separate publications. In order to improve the likelihood of publication for the project, an application for an extension to the ethical approval may be requested, so a sample equal to or greater than the power calculation can be collected. The project was a matter of interest to staff in services in which data was collected, and these teams have all requested presentations on the results. It is intended that these will be completed this year. It is also hoped that the project will be presented at relevant conferences.

A Systematic Review of Studies Comparing the Psychological Characteristics of Different Types of Functional Neurological Disorder

Abstract

The evidence concerning theoretical models for functional neurological disorders (FND) is limited and inconsistent. Previous systematic reviews have improved understanding of the available evidence. However, no existing systematic review has examined studies which compare specific types of FND based on their psychological characteristics. This area of research is in need of further examination. The present review examined quantitative studies which compared groups of participants with a specific type of functional neurological disorder (e.g. non-epileptic seizures) based on psychological characteristics. Searches were made of PubMed and PsycINFO databases. No date restriction was applied. Due to the heterogeneity of the study designs, a narrative synthesis approach was adopted. Seven studies were found to be eligible with a total of 793 participants. All studies used clinical samples, and all were comparisons of participants with non-epileptic seizures or functional motor symptoms. The majority of studies were evaluated to have moderate research quality. Overall, the findings indicated that patients presenting with these two types of FND tend to be female, have poor self-reported mental and physical health, and have elevated levels of depression, anxiety and alexithymia. Moreover, those with non-epileptic seizures tended to be younger and more likely to report childhood trauma. They were also more likely to report sexual abuse or stressful life events. These findings confirm the hypothesis of a previous review that relative to patients with functional motor symptoms, those with non-epileptic seizures tend to be younger and more likely to report

histories of stress and trauma. This review provides evidence that associations between FND and psychosocial characteristics may vary by the type of FND.

Introduction

'Functional Neurological Disorders' (FND) is an umbrella term which describes the presence of neurological symptoms in the absence of any demonstrable neurological disease or injury. Symptoms can include non-epileptic seizures and movement difficulties, such as limb weakness, balance problems and gait difficulties. Functional movement difficulties are referred to collectively as functional motor symptoms. Other symptoms of FND can include voice and swallowing difficulties, as well as alterations to cognition and awareness, such as memory problems, visual difficulties, or changes in bodily sensation. People with FND may have one or several of these symptoms (Stone, 2013).

The word 'functional' in this context indicates that what is observable in the condition is the abnormal function of a bodily system in the absence of pathology. It is intended to be an alternative to terms like 'psychogenic', which imply more assumptions about the causes of symptoms, and which tend to be poorly understood by service-users (Edwards & Bhatia, 2012; Stone, 2013).

The diagnostic categories applied to FND are currently in a state of change due to a recognition of uncertainty about their. FND have historically been referred using the diagnosis Conversion Disorder. This reflected the influence of psychodynamic models which held that these difficulties involved the 'conversion' of repressed emotional content into physical symptoms (Breuer & Freud, 1955). Recent neurological and psychiatric practice has moved towards a functional conceptualisation of the disorder, which makes fewer assumptions about its aetiology. A functional model emphasises the detection of positive

clinical signs of a functional problem through physical examination (Stone, Carson & Sharpe, 2005). It also involves a move away from diagnosing FND on the basis of associated psychosocial stress, or diagnosis by exclusion of other disorders (Ludwig *et al.*, 2018). Reflecting these changes, the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5) has changed the name of the diagnosis previously known as 'Conversion Disorder' to 'Conversion (Functional Neurological Symptom) Disorder', and removed a criterion that required the presence of psychological stress or conflict (American Psychiatric Association, 2013). The draft *International Classification of Diseases, 11th Revision* (ICD-11) has also removed references to conversion and uses the label 'Dissociative Neurological Symptom Disorder' (World Health Organisation, 2018).

FND are a common presentation in neurology clinics, making up around 16% of new referrals (Stone *et al.*, 2010). Research suggests that people with FND have poorer health outcomes than those with non-functional neurological symptoms. In a study of 3781 neurology outpatients, Carson *et al*. (2011) compared patients whose symptoms neurologists had rated as being 'not at all' or 'somewhat' unexplained by disease with patients whose symptoms were rated as 'largely' or 'completely' explained. Compared to the other patients, the less medically explained groups had significantly higher levels of distress and rated both their physical and mental health as poorer. They were also more likely to be unemployed due to ill health, and more likely to be in receipt of disability-related benefits.

In spite of the physical and psychosocial impact of FND, their aetiology is often described as

poorly understood in the literature (e.g. Edwards & Bhatia, 2012). Historically, FND have been under-researched, although since the millennium research into FND has increased (Carson *et al.*, 2012). Various interventions are recommended for FND, such as cognitive-behavioural therapy (CBT) for non-epileptic seizures (Stone, 2016), or physiotherapy for functional motor symptoms (Nielsen *et al.*, 2015). It is noteworthy that there is currently no National Institute for Clinical Excellence (NICE) guideline whose main concern is with FND. A draft guideline on suspected neurological conditions has been released (NICE, 2017), though this has been criticised for only superficially referring to FND as exclusion criteria for referral to neurology (FND Hope UK, 2017). Though psychological interventions in FND are often recommended, their evidence base is limited (Martlew, Pulman & Marson, 2014).

A wide variety of theoretical models for FND exist, though previous systematic reviews have found the evidence for these models is somewhat limited (Brown & Reuber, 2016). The earliest models in the modern era were dissociative (e.g. Janet, 1907). Dissociative models hold that FND arise from a breakdown in an individual's voluntary control in the face of intense emotional stress, resulting in the fragmentation of psychological systems. In this model, pre-existing tendencies regarding suggestibility and dissociation are also thought to be implicated, and relatedly hypnotic states are thought to be analogous with functional neurological symptoms (Oakley, 1999). Some contemporary accounts of FND continue to incorporate dissociative ideas (Bowman, 2006; Kuyk, Van Dyck & Spinhoven, 1996; Reuber, 2009). As discussed, later psychodynamic models instead conceptualised FND as a bodily manifestation of distressing, inexpressible mental content, which has been defended against through repression to the unconscious (Breuer & Freud, 1955; Howlett & Reuber,

2009).

Many recent theoretical models have adopted a broadly cognitive-behavioural orientation (Brown, 2004; Reuber & Brown, 2017; Roelofs & Spinoven 2007; Williams *et al*., 2011). Generally, these hold that FND depends on a person having a cognitive representation of an illness state. For example, in the case of non-epileptic seizures, this would be the sense of what it is like to experience a seizure, which could have been formed either through personal experience of seizures, or through other sources of information, such as perhaps witnessing seizures in a family member. In the context of distorted attentional processes (such as hypervigilance of bodily states or health worry) and heightened distress, this representation can become unintentionally activated, leading to changes in bodily function. In addition, cycles of behavioural avoidance of stimuli that are related to the illness representation are thought to support the development and maintenance of the condition.

Previous reviews have attempted to cast light on uncertainty about the aetiology of FND, through examining research into predisposing psychosocial factors. However, these have been limited either by not being systematic in nature, or having a narrow focus that excluded analysis of a broad range of factors. For instance, narrative reviews have examined the association between FND and particular psychological characteristics, such as neuropsychological dysfunction (Cragar, Berry, Fakhoury, Cibula & Schmit, 2002), post-traumatic stress disorder (Fiszman, Alves-Leon, Nunes, D'Andrea & Figuera, 2004) and personality disorder (Lacey, Cook & Salzberg, 2007). Several reviews have examined the large body of research concerning the prevalence of traumatic experiences in FND (e.g.

Fiszman *et* al., 2004; Sharpe & Faye, 2006). The most recent of these was a systematic review and meta-analysis of case control studies by Ludwig *et al.* (2018). It found, in common with other reviews, that FND were associated with higher rates of childhood abuse and stressful life events prior to onset in comparison to controls. However, it also found that in a substantial proportion of cases no history of abuse or stressful life events is disclosed. The fact that traumatic experiences may or may not be present in populations with FND has undermined psychodynamic and dissociative accounts of the condition (Reuber, 2018). However, as theoretical models of FND do not only differ in terms of assumptions about the role of trauma, it is important to consider other areas of evidence.

A systematic review by Brown and Reuber (2016) was the only systematic review of a broad range of psychosocial factors in FND that could be found by this author. Because of its scope, it allows for a thorough assessment of the evidence for different theoretical models of FND. The authors concluded that in general, the quality of most evidence identified was low to moderate, and that the support for all theories of FND was limited and inconsistent. However, a limitation of this review was that it only examined studies with participants who had a diagnosis of non-epileptic seizures, which restricts the conclusions that can be drawn about FND as a whole.

The review also excluded a class of study which compare the psychological characteristics of different types of FND, such as comparisons between groups with only non-epileptic seizures and those with only functional motor symptoms. These studies have the potential to be important in understanding FND, as they may highlight psychological differences

between types, or unifying features of the condition as whole. Brown and Reuber (2016) note that these studies have made valuable contributions to the field and would benefit from further independent systematic examination. They suggest that, at least anecdotally, such studies appear to show that compared to individuals with only functional motor symptoms, those with only non-epileptic seizures are generally younger, more likely to experience alterations in consciousness, and more likely to have experienced childhood abuse or stressful life events. It has also been the experience of this author that clinicians working with FND tend to assume there are differences in the psychosocial profiles of different functional neurological presentations, and this informs their clinical decision-making. This highlighted that this area of research would benefit from systematic examination to inform clinical practice.

Objectives

The aim of the review was to evaluate the evidence from empirical comparative studies of different types of FND. As outlined above, this is thought to be important as: a) no known previous review has examined this class of study; b) it may highlight differences in the psychological characteristics between types of FND; c) it may shed light on the aetiology of FND. This review was registered on the Prospero International Prospective Register of Systematic Reviews database (Prospero ID CRD42019123555; Newson & Fowke, 2019).

Methods

The Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) statement (Moher, Liberati, Tetzlaff & Altman, 2009) was used to inform the review methodology.

Eligibility Criteria

The review examined studies which compared the psychological or psychosocial characteristics of participants with different major types of FND (such as only non-epileptic seizures versus only functional motor symptoms). No publication date restrictions were imposed. The inclusion criteria were that studies: a) used empirical quantitative methods; b) used acceptable methods of statistical analysis; c) were published in English; d) were published in peer-reviewed academic journals; e) used participants who were aged over 18 years. Studies were excluded if they met the following criteria: a) they used small sample ($N < 15$) designs; b) they were qualitative studies; c) they were reviews, meta-analyses, case reports, theoretical articles, expert opinion, or book chapters.

Information Sources

Studies were identified via searches of the PsycINFO and PubMed electronic databases. Both searches were run on the 9th March 2019. Two additional studies were added to this list: these were referenced in the discussion of Brown and Reuber's (2016) review, though they were not formally included in that earlier systematic review. These two studies met the eligibility criteria but were not captured by the database searches.

Search

In order to identify relevant studies, a search strategy was used with two major terms, connected by an 'AND' operator. The first term included a wide variety of terms relating to psychological characteristics, such as 'psychological factor*', 'history of', 'personality' and 'risk factor'. The second term included a wide variety of names for FND, such as 'functional neurological', 'conversion disorder', 'psychogenic seizure' and 'hysteroepilepsy'. The full search strategy is given in Appendix 1. Development of the search strategy was informed by Brown and Reuber's (2016) methodology. Search terms were required to appear in the title or the abstract. Search limitations were set to conform to the review eligibility criteria.

Study Selection

The eligibility assessment was carried out by the author of this thesis. Duplicates were removed using reference management software. The remaining studies were then screened for eligibility from their titles and abstracts. Studies which did not appear to meet the eligibility criteria were subsequently excluded. Those which appeared to meet the criteria, or where it was unclear whether they met the criteria were escalated to the next stage of selection. The full texts of the remaining studies were reviewed, and any studies found not to meet the eligibility criteria were also excluded. The remaining studies were included in the narrative synthesis. The study selection process is summarised in a PRISMA flow diagram (Figure 1).

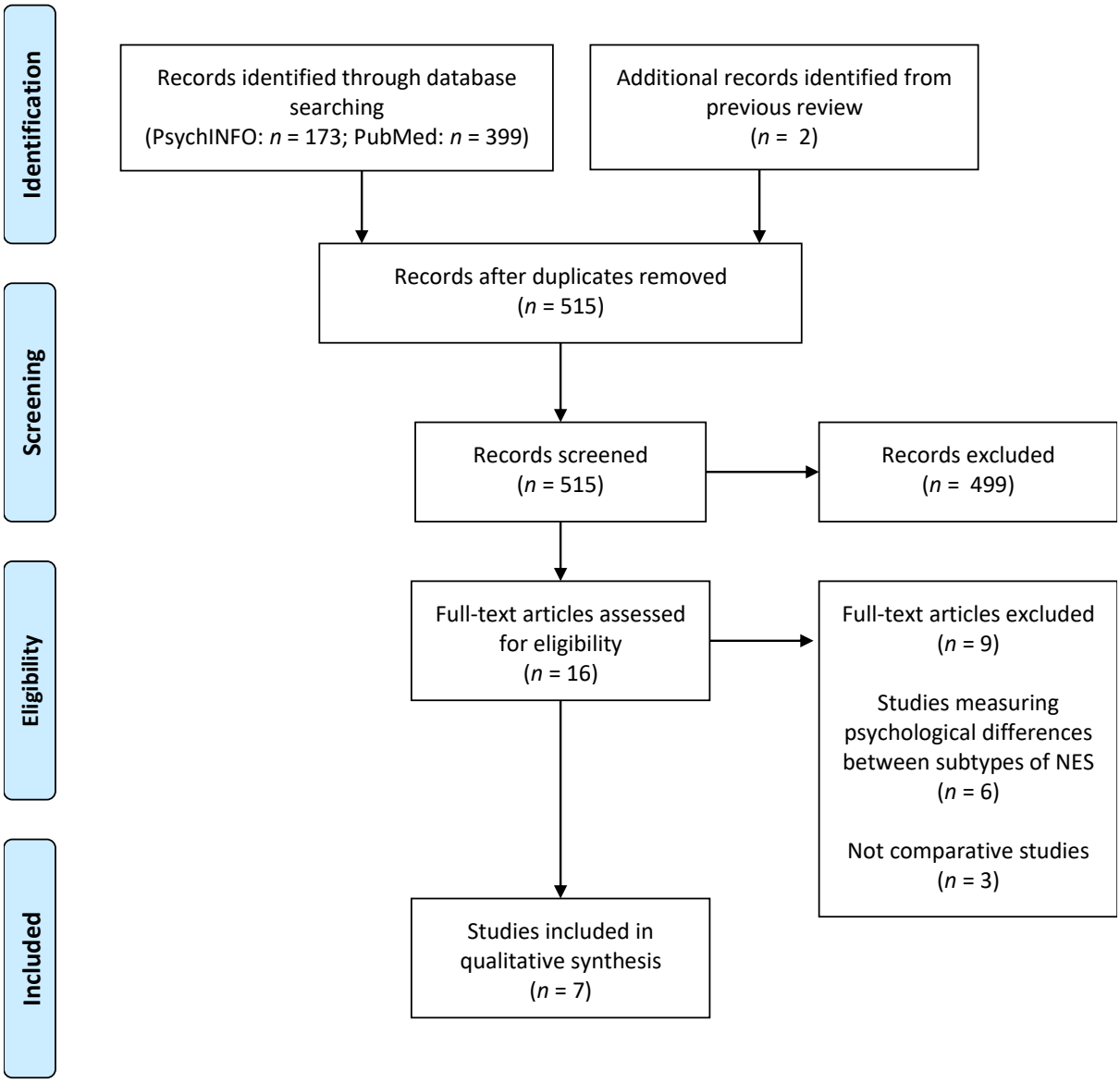

Figure 1. PRISMA flowchart detailing study selection

Data Collection Process and Data Items

A data extraction sheet was developed, and the following data was extracted from each study: author; location; year of publication; study design; participant characteristics; groups used; variables measured; and key findings. Table 2 shows the collected information extracted.

Methodological Quality Assessment

The methodological quality of the study was assessed by one reviewer (the author of this thesis), using the Critical Skills Appraisal Programme (CASP) Case Control Study Checklist (CASP, 2018). It was felt necessary to supplement the use of this quality appraisal framework with additional quality criteria specific to research into FND. These were derived primarily from Brown and Reuber's (2016) review and ensure well-controlled samples. These criteria included the stipulation that participants with non-epileptic seizures should be diagnosed using video electroencephalography (V-EEG) to ensure the validity of their diagnosis. Another was that participants with functional motor symptoms should be diagnosed using standardised criteria, such as those proposed by Fahn and Williams' (1988), as recommended by Edwards and Bhatia (2012). Table 3 gives the full list of specific quality criteria used

Planned Methods of Analysis

Analysis was based on the narrative synthesis approach set out by Popay *et al.* (2006). This approach involves the following: identifying and tabulating studies; rating their quality; exploring relationships between studies; considering variability in results between studies;

and evaluating the robustness of the synthesis. Analysis was also informed by the PRISMA statement (Moher *et al*., 2009).

Results

Study Selection

A total of seven studies were identified for final inclusion in the review (see Figure 1). Searches of PsycINFO and PubMed initially identified a total of 572 records. An additional two records (Driver-Dunckley, Stonnington, Locke & Noe, 2011; Hopp, Anderson, Krumholz, Gruber-Baldini & Shulman, 2012) were identified through mention in the earlier review by Reuber and Brown (2016). After duplicates were removed, 515 remained. 499 records were excluded as they did not meet the inclusion criteria based on screening by title and abstract. The full texts of the 16 remaining records were retrieved and examined in full.

Nine of these studies were excluded. Three (Ahsan *et al*., 2010; Akyüz, Gökalp, Erdiman, Oflaz & Karşidağ, 2017; Kuloglu, Atmaca, Tezcan, Gecici & Bulut, 2003) were excluded because they were not comparative studies: though they examined psychological variables in a sample with FND, they did not compare groups with different types of FND.

A further six studies were excluded as they were found to be comparative studies of types of non-epileptic seizure. These studies and the systems by which they subdivided types of non-epileptic seizure are given in Table 1. They were excluded for two reasons. Firstly, they used several different methods of categorising types of non-epileptic seizure, making it difficult to compare them to each other. Secondly, it was also felt that this type of study involved subclassification of functional neurological symptoms at too fine a level of detail to be comparable with the other types of study identified by the review. The ICD-11 was used to judge what counted as too fine a level of subclassification (World Health Organisation,

2018). Provided the criteria used for defining samples in a study broadly corresponded with at least two different ICD-11 diagnostic codes in the cluster 6B60 (Dissociative Neurological Symptom Disorder), this was considered an acceptable study for the review. If, however, the study compared groups which all met the criteria for just one code, then these were excluded. All the studies excluded examined groups covered exclusively by code 60B60.4 (Dissociative Neurological Symptom Disorder, with non-epileptic seizures), hence they were excluded.

Table 1

Studies excluded as they compared subtypes of non-epileptic seizure

Citation	Method of classifying non-epileptic seizures
An, Wu, Yan, Mu & Zhou, 2010	Minor vs. major vs. unresponsive
Brown *et al.*, 2013	Self-devised cluster analysis based on psychopathology
Griffith, Smith, Schefft, Szaflarski & Privitera, 2008	Minor vs. major vs. catatonic
Zeng, Myers & Lancman, 2018	Non-epileptic seizures with Post Traumatic Stress Disorder vs. non-epileptic seizures without Post Traumatic Stress Disorder
Dhiman *et al*., 2013	Abnormal hypermotor response vs. abnormal partial motor response vs. affective or emotional behaviour vs. dialeptic vs. aura vs. mixed type
Asadi-Pooya, Tinker & Fletman, 2016	Generalized motor vs. akinetic vs. focal motor vs. subjective symptoms only

Study Characteristics

Descriptive information on the seven selected studies is summarised in Table 2. All studies were published between 2004 and 2017. They were all either conducted in the United States or in Western European countries. All studies were found to be comparisons of a group with only non-epileptic seizures and a group with only functional motor symptoms.

Three studies also incorporated control groups of various types. Studies used retrospective, cross-sectional and prospective designs.

Table 2

Study Descriptive Information

Citation, country of origin	Design	Participants	Demographics Age in years (mean (*SD*), range), Gender (% female) Ethnicity (%)
1. Demartini *et al.* (2016), Italy	Cross-sectional comparative study	Total: *N* = 60 Group 1: Outpatients with NES (*n* = 20) (matched for age and gender) Group 2: Outpatients with FMS (*n* = 20) (matched for age and gender) Group 3: Healthy controls (*n* = 20) (matched for age and gender)	Total: Age: 44.9 (15.9) Gender: 80% Ethnicity: not given Group 1: Age: 45.9 (14.8), range not given Gender: 75% Ethnicity: not given Group 2: Age: 45.7 (15.8), range not given Gender: 85% Ethnicity: not given Group 3: Age: 43.1 (17.0), range not given Gender: 80% Ethnicity: not given
2. Driver-Dunkley *et al.* (2011), United States	Retrospective chart review	Total: *N* = 172 Group 1: Patients with NES (*n* = 116) Group 2: Patients with FMS (*n* = 56)	Total: Age: 46 (*SD* not given), 18-77 Gender: 82% Ethnicity: not given Group 1: Age: 41 (*SD* not given), 18-82 Gender: 82% Ethnicity: not given

			Group 2: Age: 45 (*SD* not given), 14-77 Gender: 82% Ethnicity: not given
3. Ekanayake *et al.* (2017), United States	Cross-sectional comparative study	Total: *N* = 128 Group 1: Patients with NES (*n* = 43) Group 2: Patients with FMS (*n* = 59) Group 3: Healthy controls (*n* = 26)	Total: Age: 44.4 (12.5) Gender: 75% Ethnicity: not given Group 1: Age: 40.5 (11.6), range not given Gender: 86% Ethnicity: 95.3% Caucasian Group 2: Age: 46.7 (12.7), range not given Gender: 73% Ethnicity: 93.9% Caucasian Group 3: Age: 45.9 (13.5) Gender: 65% Ethnicity: not given
4. Grimaldi, Dubuc, Kahane, Bougerol & Vercuil (2010), France	Prospective comparative study	Total: *n* = 17 Group 1: Inpatients or outpatients with NES (*n* = 8) Group 2: Inpatients or outpatients with FMS (*n* = 9)	Total: Age: 38.2 (15.0), 18-79 Gender: 76.5% Ethnicity: not given Group 1: Age: 36.2 (9.6), 23-53 Gender: 75.0% Ethnicity: not given

			Group 2: Age: 40.0 (19.8), 18-79 Gender: 77.8% Ethnicity: not given
5. Hopp *et al.* (2012), United States	Cross-sectional comparative study	Total: *N* = 139 Group 1: Outpatients and inpatients with NES (*n* = 35) Group 2: Outpatients with FMS (*n* = 104)	Total: Age: 45.8 (13.1), range not given Gender: 71.9% Group 1: Age: 41.8 (14.8), range not given Gender: 85.7% Ethnicity: not given Group 2: Age: 47.1 (12.5), range not given Gender: 67.3% Ethnicity: not given
6. Ludwig, Whitehead, Sharpe Reuber & Stone (2015), Scotland and England	Cross-sectional case-control study	Total: *N* = 227 Group 1: Patients with NES (*n* = 40) Group 2: Patients with functional weakness of a limb (*n* = 107) Group 3: Patients with epilepsy (*n* = 34) Group 4: Patients with neurological disease causing motor weakness (*n* = 46)	Total: Age: 37.8 (*SD* not given), 17-67 Gender: 62.5% Ethnicity: not given Group 1: Age: 37.0 (*SD* not given) 18-66 Gender: 62.5% Ethnicity: not given Group 2: Age: 39.1 (*SD* not given) 17-67 Gender: 79.4% Ethnicity: not given

			Group 3: Age: 33.2 (*SD* not given) 17-64 Gender: 79.4% Ethnicity: not given Group 4: Age: 39.3 (*SD* not given) 18-63 Gender: 82.6% Ethnicity: not given
7. Stone, Sharpe & Binzer (2004), Scotland	Cross-sectional comparative study	Total: *N* = 50 Group 1: Inpatients with NES (*n* = 20) Group: Inpatients with functional paresis or paralysis only (*n* = 30)	Total: Age: 34 (*SD* not given) 18-74 Gender: 66% Ethnicity: not given Group 1: Age: 27 (*SD* not given) 18-54 Gender: 75% Ethnicity: not given Group 2: Age: 39 (*SD* not given) 18-74 Gender: 60% Ethnicity: not given

Table 2

Study Descriptive Information (continued)

Citation	Main Variables Studied (Measure - Citation)	Summary of Findings (Effect sizes reported as Cohen's *d* or as odds ratios)
1. Demartini *et al.* (2016)	Psychoform dissociation (Cambridge Depersonalization Scale - Sierra & Berrios, 1998; Dissociative Experiences Scale - Carlson & Putnam, 1993) Somatoform Dissociation (Somatoform Dissociation Questionnaire - Nijenhuis, Spinhoven, Van Dyck, Van Der Hart & Vanderlinden, 1996) Alexithymia (Toronto Alexithymia Scale - Bagby, Taylor & Parker, 1994) Interoception (Heartbeat Detection Task - Schandry, 1981) Psychopathology (Hamilton Rating Scale for Depression - Hamilton, 1960; Hamilton Rating Scale for Anxiety - Maier, Buller, Philip & Heuser 1988)	Generally, participants with FND had higher levels of dissociation than healthy controls. The NES group had higher levels of psychoform dissociation than the FMS group (p = .007, d = .94) and healthy controls (p = .02, d = .76); no difference was found between the FMS group and healthy controls (p = 1). The FMS group had higher rates of somatoform dissociation than the NES group (p = .04, d = 2.36) and healthy controls (p = .03, d = 2.68); no difference was found between participants with NES and healthy controls (p = 1). Healthy controls had lower levels of alexithymia than the NES group (p = .04, d = .85) and the FMS group (p = .04, d = .82), but the two groups did not differ in alexithymia (p = 1). No differences found between groups regarding heartbeat detection (p = .8). Healthy controls had lower levels of depression and anxiety than the NES group (p = .002, d = 1.34; p = .004, d = 1.26) and the FMS group (p = .087, d = .85; p = .01, d = .97) The FND groups did not differ in depression and anxiety scores (p = .5; p = 1).
2. Driver-Dunkley *et al.* (2011)	Characteristics of clinical notes (no standardised measure used)	Participants with NES were found to be younger (p = .005, effect size not calculable) and younger at onset of functional symptoms than those with FMS (p = .038, effect size not calculable). Participants with NES were more likely to have histories of childhood abuse (p = .003, odds ratio = 2.87), sexual abuse (p = .03, odds ratio = 2.35) and emotional abuse (p = .006, odds ratio = 3.02), though not physical abuse (p = .06). They were also found to have higher rates of stressful life events preceding onset (p = .023, odds ratio = 6.22). Rates of chronic pain conditions were high in both groups (70% overall).

3. Ekanayake *et al.* (2017)	Personality Traits (The NEO Personality Inventory Revised - Costa & McRae, 1992) Traumatic Experiences (Childhood Trauma Questionnaire - Bernstein *et al.* (1994) Alexithymia (Toronto Alexithymia Scale - Bagby, Parker & Taylor, 1994) Dissociation (Dissociative Experiences Scale - Carlson & Putnam, 1993) Psychopathology (Symptom Checklist 90R - Derogatis, Lipman & Covi, 1973; Beck Depression Inventory – Beck, Steer & Brown, 1996; Beck Anxiety Inventory - Beck, Epstein, Brown & Steer, 1988) Healthy controls did not complete measures of alexithymia, dissociation or anxiety.	No difference in gender (p = .1) or ethnicity (p = .6) between groups with FND. The NES group was younger than the FMS group (p = .04, d = .5). The NES group had higher levels of neuroticism and lower levels of conscientiousness the FMS group (p = .002, d = .69; p = .006, d = .53) or healthy controls (p = .02, d = .90; p = .02, d = .65). The FMS group's personality profile appeared similar to that of healthy controls. Regarding childhood sexual abuse, the NES group had higher scores than the FMS group (p = .019, d = .53) and healthy controls (p < .001, d = 1.02), while FMS groups did not differ from controls (p > .01). Regarding childhood emotional abuse, NES and FMD groups scored higher than controls (p < .01 and p < .01 respectively; d = .87 and d = 1.13 respectively) but the two FND groups did not differ (p > .02). Regarding childhood physical abuse, the NES group scored higher than healthy controls (p = .042, d = .65), but the FMS group did not differ from healthy controls (p > .01). Regarding childhood physical neglect, the NES group scored higher than healthy controls (p = .007, d = .84), but the FMS group did not differ from healthy controls (p > .01). The NES group reported being a younger age when the most distressing event of their life occurred compared to the FMS group (p < .001, d = .95). The NES group also reported being younger when this event occurred compared to healthy controls (p = .003, d = .76). The NES group had higher levels of alexithymia than the FMS group (p = .0002, d = 1.20). The NES group had higher general levels of dissociative experience (p < .001, d = 1.10), though subscales for types of dissociation were not reported. The NES group were more anxious (p = .002, d = 0.70) and depressed (p < .02, d = 0.98) than the FMS group or healthy controls, and they had a greater number of mental health

		symptoms ($p < .012$, $d = .82$).
4. Grimaldi, Dubuc, Kahane, Bougerol & Vercuil (2010)	Clinical Characteristics (no standardised measure used) Diagnostic Interview by psychiatrist according to *Diagnostic and Statistical Manual for Mental Disorders, Fifth Edition* (American Psychiatric Association, 2013) Psychopathology (Beck Depression Inventory - Beck, 1996; Spielberger State Trait Anxiety Questionnaire - Gaudry, Vagg & Spielberger, 1975)	The article reports significance tests were used, but oddly no statistics related to these are reported. It also reports that the groups did not appear to significantly differ for any major demographic or psychopathological characteristics, except that the NES group tended to have higher scores for anxiety than the FMS group, and the NES group were also more likely to have had a family history of epilepsy or a movement disorder.
5. Hopp *et al.* (2012)	Health-Related Quality of Life (SF-12 Health Status Survey - Ware, Kosinski & Keller, 1996) Psychopathology (Brief Symptom Inventory 18 - Derogatis, 2000)	The NES group were more likely to be female ($p = .034$, odds ratio = 3.0) and younger ($p = .04$, $d = .39$) than the FMS group. The groups were not significantly different in terms of overall health-related quality of life, overall psychopathology, or subscales related to somatisation or depression ($p > .05$).
6. Ludwig, Whitehead, Sharpe Reuber & Stone (2015)	Illness perceptions (Illness Perception Questionnaire - Revised - Weinman, Petrie Moss-Morris & Horne, 1996)	No differences in age and gender were found between groups, though it was unclear from the publication whether demographic matching was used. Given the design and the lack of discussion of this finding in the article it was thought likely that demographic matching had been used. Both FND groups tended to report low levels of personal control and understanding of their condition compared to disease controls. Participants with NES rejected psychological explanations of their condition more strongly than the FMS group ($p < .01$, $d = .49$). The NES group reported that their condition had a greater impact on their lives and their families lives than the FMS group ($p < .01$, $d = .44$)

7. Stone, Sharpe & Binzer (2004)	Psychopathology (Structured Clinical Interviews (I & II) for the *Diagnostic and Statistical Manual for Mental Disorders, 4th Edition* - American Psychiatric Association, 2000) Perceived Parenting Childhood (Egna Minnen Beträffande Uppfostran (My Memories of Upbringing) Self Rating Inventory - Perris, Jacobsson, Linnströ, von Knorring & Perris, 1980) Stressful life events (Life Events Inventory, Cochrane & Robertson, 1973)	Participants with NES tended to be younger ($p < .005$, not enough information to calculate effect size), but there was no difference in gender ($p > .05$). There were high rates of any personality disorder (50-65%). Participant with NES were more likely to have Borderline Personality Disorder (35% vs. 5%, $p < .05$, odds ratio = 7.5), though rates of any personality disorder were similar ($p > .05$). They were also more likely to have experienced incest ($p < .01$, odds ratio = 12.42) and more likely to have experience parental divorce in childhood ($p < .05$, odds ratio = 4.89). The NES group had lower levels of perceived parental warmth in childhood ($p < .05$, $d = .54$ (fathers), $d = .86$ (mothers)), though findings regarding other unhelpful parenting styles were mixed. The NES group had experienced a greater number of stressful life events in the preceding 12 months than the FMS group ($p < .0001$, $d = 1.21$), although not in the preceding 3 months ($p < .05$).

Note. Abbreviations: NES, non-epileptic seizures; FMS, functional motor symptoms. Where statistics were absent, if possible, they have been estimated from others that were available according to the equations given by Field (2009). Effect sizes were very rarely reported so in almost all cases these have been calculated using methods given by Field (2009).

Participant Characteristics

The studies included a total of 793 participants. The number of participants per study ranged between 17 and 227. Clinical participants were generally recruited from both inpatient and outpatient services in hospitals, though two studies recruited exclusively inpatients and outpatients respectively (Demartini *et al.*, 2016; Stone *et al.*, 2004). One study also recruited two control groups with epilepsy and organic neurological motor difficulties (Ludwig *et al.*, 2015). Two other studies used healthy control groups (Demartini *et al.*, 2016; Ekanayake *et al.*, 2017).

The mean age across studies ranged between 34 and 46 years. The majority of total participants were female, as they were in all individual samples. Details regarding ethnicity were generally not recorded, but where available participants were described as predominantly caucasian.

Variables and Measures

The studies examined a range of psychological characteristics. They shared constructs of interest, though had almost no validated measures in common. Five studies used previously validated measures, while in one study, researchers only rated clinical notes for the presence of characteristics (Driver-Dunkley *et al.*, 2011). Most validated measures were self-report questionnaires. Two studies supplemented self-report questionnaires with other standardised measures, namely a semi-structured clinical interview (Stone *et al.*, 2004) and a psychophysiological task (Demartini *et al.*, 2016).

Psychopathology and Health-Related Quality of Life

Four studies used previously validated general measures of psychopathology: the Symptom Checklist 90R (Derogatis *et al.*, 1973) and the Brief Symptom Inventory 18 (Derogatis, 2000). Disorder specific measures of depression and anxiety were also used: the Hamilton Rating Scales for Depression and Anxiety (Hamilton, 1960; Maier *et al.*, 1988), the Beck Depression and Anxiety Inventory (Beck, 1996; Beck *et al.*, 1988), and the Spielberger State-Trait Anxiety Questionnaire (Gaudry *et al.*, 1975). One study (Stone *et al.,* 2004) study used the Structured Clinical Interviews (I & II) for the Diagnostic and Statistical Manual for Mental Disorders, 4th Edition (American Psychiatric Association, 1994). Two studies recorded large numbers of simple descriptive statistics regarding unstandardized ratings of clinical characteristics (Grimaldi *et al.*, 2010; Stone *et al.*, 2004). One study (Hopp *et al.*, 2012) measured Health-Related Quality of Life using the SF-12 Health Status Survey (Ware *et al.*, 1996).

Personality

One study measured personality traits using a self-report questionnaire (Ekanayake *et al.*, 2017). This study used The NEO Personality Inventory Revised (Costa & McRae, 1992) to examine a dimensional model of personality.

Traumatic Experiences

One study (Ekanayake *et al.*, 2017) used the Childhood Trauma Questionnaire (Bernstein *et al.* 1994). The perceived parenting style that participants had experienced in childhood was measured in one study (Stone *et al.*, 2004) using the My Memories of Upbringing Self Rating Inventory (Perris *et al.*, 1980). This study also measured stressful life events prior to onset of

functional neurological symptoms using the Life Events Scale (Cochrane & Robertson, 1973).

Dissociation

Two of the studies used self-report questionnaires which measured types of dissociative experience (Demartini *et al.*, 2016; Ekanayake *et al.*, 2017). The first study used both the Cambridge Dissociation Scale (Sierra & Berrios, 1998) and Somatoform Dissociation Scale (Nijenhuis, *et al.*, 1996) in order to make comparisons between psychoform and somatoform dissociation in participants. The second study used the Dissociative Experiences Scale (Carlson & Putnam, 1993) to compare levels of dissociation in the two groups, though subscale scores for types of dissociative experiences were unfortunately not reported.

Alexithymia and Interoception

The same two studies (Demartini *et al.*, 2016; Ekanayake *et al.*, 2017) also measured alexithymia in their samples using the Toronto Alexithymia Scale (Bagby *et al.*, 1994). These were the only studies found to share a measure in common. One study (Demartini *et al.*, 2016) used the Heartbeat Detection Task (Schandry, 1981) as a measure of interoception.

Illness Perceptions

One study measured illness perceptions related to functional neurological symptoms (Ludwig *et al.*, 2015). These were measured using the Illness Perception Questionnaire-Revised (Weinman *et al.*, 1996)

Methodological Quality

As discussed in greater depth above, the CASP Case Control Study Checklist (CASP, 2018)

was used to assess the quality of the studies. This framework was supplemented by specific quality standards in this research field as defined by previous papers (e.g. Brown & Reuber, 2016). Table 3 summarises these specific standards and their application to the studies. In most cases, studies were rated based on whether the design explicitly mentioned addressing a quality item.

Overall, the quality of the studies, with the exception of one, was judged to be moderate. One study was judged to be of poor quality (Grimaldi *et al.*, 2010). The key flaws identified in this study were that it used small samples (it compared groups of eight and nine participants); it had no power calculation; and did not provide any statistics regarding the outcomes of significance tests, though it reported most findings were not statistically significant. The reason no study was awarded a strong quality rating related either to the failure to use demographically matched disease control groups or the failure to report power calculations.

Concerning the other studies rated as moderate in quality, they showed both methodological strengths and weaknesses. For instance, regarding the risk of selection bias, generally these studies appeared to have taken reasonable steps to ensure adequately unbiased sampling. For instance, all cases of non-epileptic seizures were reported as having been diagnosed using V-EEG. Furthermore, in five out of seven studies, functional motor symptoms were explicitly reported as having been diagnosed using Fahn and Williams' (1988) criteria. No other alternative criteria were reported as having been used (e.g. Gupta & Lang, 2009).

It was however problematic that only one study (Hopp *et al.*, 2012) used a power calculation. This may have heightened the risk of type II error in these studies with regard to findings which were statistically non-significant. It was also problematic that only two studies (Demartini *et al.*, 2016; Stone *et al.*, 2004) were completely transparent about the settings from which their clinical samples were drawn. This may have increased the variability in the samples, as the severity of the FND involved is not clear.

Regarding the use of control groups, three of the studies used control groups in addition to comparing groups with FND (Demartini *et al.*, 2016; Ekanayake *et al.*, 2017; Ludwig *et al.*, 2015). However, only two of these control groups appeared to have been demographically matched to groups with participants who had FND. These studies used either healthy controls (Demartini *et al.*, 2016) or disease controls (Ludwig *et al.*, 2015).

Lastly, all but one of the studies (Driver-Dunkley *et al.*, 2011) used at least some measures that had previously been standardised, though these were almost entirely self-report questionnaires. Two studies (Demartini *et al.*, 2016; Stone *et al.*, 2004) included information on the blinding of interviewers or raters. However, in both cases this was regarding researchers being blinded to the study hypothesis, rather than the diagnosis of the participant.

Table 3

Methodological Quality Assessment Table

Authors	Demartini *et al.* (2016)	Driver-Dunkley *et al.* (2011)	Ekanayake *et al.* (2017)	Grimaldi *et al.* (2010)	Hopp *et al.* (2012)	Ludwig *et al.* (2015)	Stone *et al.* (2004)
Recruitment							
Sample Size	60	172	128	17	139	227	50
Consecutive Sampling	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Patient type clear (inpatient/outpatient)	Yes	No	No	No	No	No	Yes
NES diagnosed using V-EEG	Yes	Yes	Yes	Yes	Yes	Yes	Yes
FMS diagnosed standardised criteria	Yes	Yes	Yes	Yes	Yes	No	No
FND diagnoses confirmed by agreement of two neurologists	Yes	No	Yes	No	No	No	Yes
Mixed epilepsy and NES excluded	Yes	No	Yes	No	Yes	Yes	Yes
Mixed organic movement disorder and FMS excluded	Yes	No	Yes	No	Yes	Yes	Yes
Mixed clearly NES/FMS excluded	Yes	Yes	Yes	Yes	No	Yes	No
Procedures to exclude panic disorder	Yes	No	Yes	No	No	No	Yes
Power calculation documented	No	No	No	No	Yes (Met)	No	Yes
Controls Groups							
Any control group used	Yes	No	Yes	No	No	Yes	No
Healthy controls	Yes	No	No	No	No	No	No
Disease controls	No	No	No	No	No	Yes	No
Demographically matched controls	Yes	No	No	No	No	Yes	No
FND explicitly excluded from controls	Yes	No	No	No	No	No	No
Other methodological issues							
Standardised Measures Used	Yes	No	Yes	Yes	Yes	Yes	Yes
Evidence of blinding where appropriate	Yes	No	No	No	No	No	Yes
Statistical controls used (e.g. covariates)	Yes	No	Yes	No	No	No	No
Overall Quality Assessment (Poor/Moderate/Strong)	Moderate	Moderate	Moderate	Poor	Moderate	Moderate	Moderate

Note. Abbreviations: V-EEG, video-electroencephalography; NES, non-epileptic seizures; FMS, functional motor symptoms

Synthesis of Results

The results are grouped into seven categories below. In general, there were few studies with variables in common. This, combined with the fact that no study was rated as strong in quality, means that, the results should be understood as tentative. The single study rated as poor in quality was discounted from the synthesis as it was thought methodological issues had undermined its findings.

Demographics

Although demographic variables were not the primary focus of this review, it was thought important to consider these because of the bearing they may have on other findings. Regarding gender, the majority of all samples were female, and in clinical groups the female majority varied between 60% and 86%. One study found that participants with non-epileptic seizures were more likely to be female than those with functional motor symptoms (Hopp *et al.*, 2012). However, three other studies found no difference in gender between groups (Driver-Dunkley *et al.*, 2011; Ekanayake *et al.*, 2017; Stone *et al.*, 2004). In the remaining two studies, the use of demographic-matching between groups meant their findings were not relevant.

Regarding the age of participants, the studies included participants with a range of ages within adulthood. Setting aside those studies where demographic-matching was used, all remaining studies found that participants with non-epileptic seizures tended to be younger that those with functional motor symptoms (Driver-Dunkley *et al.*, 2011; Ekanayake *et al.*, 2017; Hopp *et al.*, 2012; Stone *et al.*, 2004). Where effect sizes could be calculated, these tended to be in the medium range according to Cohen (1988). Mean ages tended to differ

by around five years. Interestingly, one study also found that participants with non-epileptic seizures tended to report a younger age of onset for their functional symptoms (Driver-Dunkley *et al.*, 2011).

Ethnicity was rarely recorded, with only one study doing so (Ekanayake *et al.*, 2017). This study found no statistically significant difference in ethnicity between the clinical groups. However, it is noteworthy that all the studies involved were conducted with western countries within Europe or North America.

Psychopathology and Quality of Life

In studies using general measures of psychopathology, it was found that patients with FND had higher levels of mental health symptoms than normative values (Hopp *et al.*, 2012) and healthy controls (Ekanayake *et al.*, 2017). In terms of group differences, findings were mixed: one study found no group differences on overall scores or subscales (Hopp *et al.*, 2012), whereas the other found that participants with non-epileptic seizures had a greater number of mental health symptoms (Ekanayake *et al.*, 2017). Hopp *et al.*, 2012 also examined a health-related quality of life measure which produced mental health and physical health subscales. Both groups scored lower than normative values, but the study found no statistically significant differences between the two groups.

The findings regarding specific measures of depression and anxiety were very similar to those for general psychopathology. Two studies found that levels were above that of normative values (Ekanayake *et al.*, 2017) and controls (Demartini *et al.*, 2016). However, regarding between group differences, one study found no statistically significant differences

between group scores (Demartini *et al.*, 2016), while another found that participants with non-epileptic seizures had higher levels of depression and anxiety (Ekanayake *et al.*, 2017).

Personality

Only one study examined a dimensional model of personality in FND (Ekanayake *et al.*, 2017). It found that participants with non-epileptic seizures had higher levels of neuroticism and lower levels of conscientiousness than those with functional motor symptoms or healthy controls. In contrast, the personality profile of those with functional motor symptoms appeared to be similar to that of healthy controls. Another study (Stone *et al.*, 2004) recorded rates of personality disorder. It found elevated rates of any personality disorder in both groups (50% and 65%), though no statistically significant differences in the rates between groups. However, rates of Borderline Personality Disorder (BPD) were significantly higher in those with non-epileptic seizures (35% vs. 5%). Rates for other personality disorders were not reported.

Adverse Experiences

All studies investigating the prevalence of adverse experience generally found it was more likely to be reported by participants with non-epileptic seizures than those with functional motor symptoms, though for some specific abuse subscale differences were not statistically significant. For instance, Driver-Dunkley *et al.* (2011) found participants with non-epileptic seizures were more likely to have histories of childhood abuse, sexual abuse, and emotional abuse, though not physical abuse. Ekanayake *et al.* (2017) also found that on a measure of childhood trauma, scores for sexual abuse, physical abuse and neglect were significantly higher in participants with non-epileptic seizures, whereas in participants with functional

motor symptoms, scores were similar to healthy controls. The one exception was childhood emotional abuse, where both FND groups had scores which were significantly higher than controls and not significantly different from each other. Ekanayake *et al.* (2017) also found that participants with non-epileptic seizures reported they were at a younger age at the time of their most traumatic memory, when compared to participants with functional motor symptoms and healthy controls. Lastly, Stone *et al.* (2004) also found that participants with non-epileptic seizures were more likely to have experienced incest, which presumably is largely accounted for by sexual abuse.

Stressful life Events

Both studies which examined stressful life events generally found that that participants with non-epileptic seizures had higher overall rates than those with functional motor symptoms. Driver-Dunkley *et al.* (2011) found participants with non-epileptic seizures had a greater number of stressful life events preceding onset of functional symptoms. Stone *et al.* (2004) replicated this finding, though not when only the 3 months prior to onset of functional symptoms were considered, in which case the differences were not statistically significant. Stone *et al.* (2004) also found that that participants with non-epileptic seizures were more likely to have experienced parental divorce during childhood, as well more likely to report reduced parental warmth during childhood.

Alexithymia

Findings regarding alexithymia were mixed. One study found that levels of alexithymia in both groups were higher than in healthy controls, but not significantly different between groups (Demartini *et al.*, 2016). Another study also found that levels of alexithymia in both

groups were higher than in controls, but that levels in those with non-epileptic seizures were higher than in those with functional motor symptoms (Ekanayake *et al.*, 2017).

Dissociative Experience

Demartini *et al.* (2016) found qualitative differences in the type of dissociative experiences reported between groups. Participants with non-epileptic seizures were found to have higher levels of psychoform or detachment dissociation than participants with functional motor symptoms or healthy controls. Participants with functional motor symptoms were found to have higher levels of somatoform or compartmentalisation dissociation than participants with non-epileptic seizures or healthy controls. Using a different measure, Ekanayake *et al.* (2017) found participants with non-epileptic seizures had higher levels of overall dissociative experience, but unfortunately did not report comparisons using subscales for types of dissociation.

Illness-Perception and Self-Perceptions

Lastly, one study found that FND groups tended to report lower levels of personal control and understanding of their condition than disease controls (Ludwig *et al.*, 2015). Participants with non-epileptic seizures rejected psychological explanations of their condition more strongly than those with functional motor symptoms.

Critical Analysis

As has been acknowledged, this review draws on a limited number of studies with relatively few variables in common, meaning that its conclusions must be tentative. However, several other points are worthy of comment. Firstly, there was a lack of recording of measures of

ethnicity or culture, so it is unclear if ethnic or cultural background has any relation to these findings. Secondly, most studies relied on self-report questionnaires. None of these appeared to be validated in FND populations, which may call into question these results. Given that some of the findings suggest that people with FND may be distressed and have difficulties processing emotions, this could disrupt their answers on self-report measures. However, two studies of the moderate quality studies (Driver-Dunkley *et al.*, 2011; Stone *et al.*, 2004) also used ratings of clinical notes or standardised psychiatric interviews to assess psychological characteristics, and their results were comparable to other studies, suggesting there is some validity in the results which were reliant on self-report.

Thirdly, in most non-demographic findings, there appeared to be a high degree of variability in findings concerning differences between groups: some studies found no differences between groups, while others tended to find greater psychological dysfunction in non-epileptic seizure groups. It was striking that Driver-Dunkley *et al.* (2011), Ekanayake *et al.* (2017), Ludwig *et al.* (2015) and Stone *et al.* (2004) tended to find more differences between these groups, whereas Demartini *et al.* (2016) and Hopp *et al.* (2012) tended to find few differences between groups. Based on the quality appraisal of these studies, the reasons for this variability were not immediately obvious. However, on closer examination, both of these studies had comparatively small samples (20 and 35) of participants with non-epileptic seizures. This, combined with the absence of a power calculation in Demartini *et al.* (2016), may have led to a lack of power to detect medium to smaller effect sizes between groups.

Discussion

This review aimed to examine studies which compared types of functional neurological disorder based on their psychological characteristics. Seven relevant studies were identified, and in all cases, these were comparisons of participants with non-epileptic seizures and functional motor symptoms. It is important to reiterate that only a small number of studies were identified by the review, and generally the quality of research was found to be moderate. As such, any conclusions must be interpreted with caution. The key findings of this review are summarised and discussed below. Though it was not the primary intention of the review to concentrate on demographic findings, the first part of the discussion focuses on these issues, as they are thought to be important contextual factors for understanding the other findings.

Gender

The review identified that the majority of all FND samples were female. This replicates the findings of other reviews (Brown & Reuber, 2016; Ludwig *et al.*, 2018) and indicates that the majority of patients presenting clinically with FND appear to be female.

Age

Another demographic finding which was consistent across all studies was that participants with non-epileptic seizures tended to be younger than those with functional motor symptoms. Furthermore, one study also found that the mean reported age at onset of non-epileptic seizures was also lower (Ekanayake *et al.*, 2017), whilst another found that time the taken to diagnose a functional condition was not significantly different between the two groups (Driver-Dunkley *et al.*, 2011). This suggests that the age of those presenting with FND

is not simply an artefact of structural effects within health services. Instead, the finding suggests that the development of non-epileptic seizures is more closely linked to processes earlier in the lifespan.

Adverse Experiences

Another consistent finding was a tendency towards higher levels of self-reported adverse experiences amongst participants with non-epileptic seizures over those with functional motor symptoms. These effects appeared to be most consistent regarding childhood abuse and neglect, sexual abuse, and stressful life events. This suggests that the role of traumatic or stressful experiences may be more implicated in the development of non-epileptic seizures than in functional motor symptoms.

Continuing a theme of youth, Ekanayake *et al.* (2017) also found that participants with non-epileptic seizures tended to report being at a younger age when the most traumatic event of their life occurred compared to other groups. The average age for this event was 18 years in participants with non-epileptic seizures, as opposed to 32 years in participants with functional motor symptoms, and 29 years in healthy controls. These were both large effects. Taken together, these findings suggest that traumatic events during childhood, adolescence and early adulthood may be more involved in the development of non-epileptic seizures compared to functional motor symptoms. These findings are consistent with research highlighting elevated rates of trauma exposure and stressful life events in those with FND (Ludwig *et al.*, 2018). They are also consistent with elevated rates of trauma exposure and post-traumatic stress disorder (PTSD) in those with non-epileptic seizures in particular (Fizman *et al.*, 2004).

However, again it is important to qualify these findings with reference to gender bias in the samples. For instance, the life-time prevalence of PTSD in women is thought to be around twice that in men, and women are much more likely to experience sexual violence in their lifetimes than men (Cortina & Kubiak, 2006). Childhood sexual abuse may also be more common in female children (Walker, Carey, Mohr, Stein & Seedat, 2004). Given that patterns of trauma exposure vary by gender, it is unclear whether these findings would be true of men with FND.

Personality

There was limited evidence concerning comparisons of types of FND based on models of personality, with only one study examining the issue (Ekanayake *et al.*, 2017). Regarding personality traits, non-epileptic seizures were found to be associated with higher levels of neuroticism and lower levels of conscientiousness, whereas functional motor symptoms were not associated with statistically significant differences from healthy controls. Neuroticism is a persistent tendency to experience events negatively, or 'distress-proneness' (Deary, Chalder & Sharpe, 2007). Conscientiousness is a drive towards achievement, organisation and self-discipline (Digman & Takemoto-Chock, 1981), and is generally associated with increased well-being and productivity (Bogg & Roberts, 2012). These findings may represent underlying traits which predispose individuals to non-epileptic seizures. For instance, high levels of neuroticism may predispose individuals to be sensitive to the effects of adverse experiences, whereas reduced conscientiousness could limit their ability to cope productively with adversity.

Findings from another study were similar, in which FND were compared based on rates of personality disorder diagnoses (Stone *et al.*, 2004). Here it was found that rates of BPD were significantly higher in those who experienced non-epileptic seizures (35% vs. 5%). Saulsman and Page's (2004) meta-analysis has linked BPD to a dimensional personality profile of elevated neuroticism and diminished conscientiousness, so this finding is coherent with Ekanayake *et al.*'s (2017) findings. This is also consistent with existing research suggesting comorbidities between non-epileptic seizures and BPD. For instance, Howorka, Nezadal, Herman, Nemcova and Bajacek (2007) found that 31% of a sample of patients with non-epileptic seizures met criteria for BPD. However, Stone *et al.* (2004) also found overall rates of any personality disorder were similar in both groups (50-65%), suggesting a degree of different personality dysfunction was present in the functional motor symptom group also. Unfortunately this study did not report rates of any other personality disorders in their sample, meaning it is unclear what the nature of this dysfunction might be.

Research into BPD has also demonstrated high rates of reported trauma in the condition, particularly experience of childhood abuse (Ball & Links, 2009). More recent models of the condition, such as those derived from schema therapy or cognitive analytic therapy, tend to see the internalisation of trauma as central to the condition (Ryle, Leighton & Pollock, 1997; Young, Klosko & Weishaar, 2003). This suggests these findings are also largely consistent with those concerning adverse experiences.

Dissociative Experience

Similarly, there was limited evidence concerning dissociative experience, with only one comparatively small study examining it as a primary focus (Demartini *et al.*, 2016). Here it

was found that there were differences in dissociative experience between the two groups, whereby those with non-epileptic seizures showed higher levels of detachment or psychoform dissociation, while those with functional motor symptoms showed higher levels of compartmentalisation or somatoform dissociation. In Holmes *et al.*'s (2005) model of dissociative experience, these are distinct subtypes of dissociation that occur in different contexts. Detachment refers to feelings of estrangement or alienation from one's self, experience, or the world. Detachment is demonstrated in the phenomena of derealisation (the experience of feeling somehow distant from the world) and depersonalisation (the feeling of distance from or fragmentation of the self). Compartmentalisation dissociation instead refers to incomplete information transfer within psychological systems such as memory, self-awareness, or bodily awareness.

This model holds that both of these occur separately in a range of different psychological conditions. For instance, Holmes *et al.* (2005) identify that experiences of detachment are common in BPD, depression, Anorexia and Depersonalisation Disorder, whereas compartmentalisation is commonly present in conditions like Dissociative Fugue or Dissociative Identity Disorder. In some conditions both are thought to co-occur in different aspects of the disorder. This is most notable in PTSD, where for instance often experiences of derealisation or depersonalisation might occur alongside compartmental memory disturbances. In the original model, both non-epileptic seizures and functional motor symptoms were classified as types of compartmentalisation, so Demartini *et al*.'s (2016) findings do no support this prediction.

Demartini *et al.* (2016) suggest these results might best be understood as suggesting that

the two conditions have similar, but separate underlying dissociative mechanisms. They propose that non-epileptic seizures might be understood as an extreme form of detachment related to traumatic experience or stress. In contrast, they suggest that functional motor symptoms are a compartmentalisation of bodily systems, more likely related to a psychological concern about a physical stressor, such as an injury. As this was the only study of its kind identified, it is difficult to draw definite conclusions about this theory. However, it does continue an underlying theme in the studies linking FND to mental health conditions which may involve trauma, and the suggestion that are different underlying psychological processes in these two types of FND.

Other findings

Regarding studies of self-reported general psychopathology, health, depression, anxiety, and alexithymia, the studies identified by the review tended to find that levels of these constructs in the two groups were elevated above that of controls and normative values. However, whether differences were found between the two groups varied across studies. It is thought that this may in part relate to the quality of some studies, with some relying on small samples of participants with non-epileptic seizures, and only one study reporting a power calculation. This may have led to type II errors in these studies.

It is also noteworthy that few studies found those with functional motor symptoms scored higher than those with non-epileptic seizures on a measure where a higher score would be expected to indicate greater psychopathology. Generally, the pattern of findings was that no difference between groups was found, or participants with non-epileptic seizures had scores indicating significantly higher psychopathology. Again, this could be indicative of type II

errors. However, another factor could explain the variability of findings: it might be that there is greater variability in the psychology of people with non-epileptic seizures than those with functional motor symptoms. As a result, certain samples of people with non-epileptic seizures might contain higher levels of psychological dysfunction than others. In the studies identified here, this effect may have been masked by a low quality of information reported about the settings from which groups were recruited.

The idea that there is variability in levels of psychopathology within non-epileptic seizure populations has some support in recent research. For instance, in a study excluded from this review, of 156 patients with non-epileptic seizures, it was found that where patients also met criteria for PTSD, this was associated with higher levels of alexithymia and personality dysfunction (Zeng *et al*., 2018). This suggests there may be sub-groups within those presenting with non-epileptic seizures which have higher levels of trauma exposure and difficulties coping with emotion.

Aside from this issue, these results do highlight that participants with these two FND appear to score higher than controls on measures of psychopathology, ill-health, depression, anxiety, and alexithymia. This reiterates the levels of distress and impact on function associated with the condition (Carson *et al*., 2011). The elevation in levels of alexithymia also suggests a background of difficulties coping with emotional challenges, as did the findings around personality traits.

Summary

Overall, this review indicates that patients presenting with these two types of FND (non-

epileptic seizures and functional motor symptoms) tend to be female, have poor self-reported mental and physical health, and elevated levels of depression, anxiety and alexithymia. In regard to differences between the two types of FND reviewed, the most consistent findings were that those with non-epileptic seizures were younger, and more likely to report traumatic or aversive experiences (particularly childhood trauma, sexual abuse and stressful life events). There was also some limited evidence for personality dysfunction in both groups, differential patterns of dissociative experience between groups, and elevated rates of BPD in participants with non-epileptic seizures.

This largely confirms Brown and Reuber's (2016) hypothesis, in that participants with non-epileptic seizures appeared to be younger and to be more likely to report traumatic experiences, although there was little evidence concerning the prediction that they would be more likely to experience alterations to consciousness. The evidence concerning dissociative experience would in fact suggest that both groups are prone to different kinds of dissociative alterations to consciousness.

These conclusions are consistent with systematic reviews exploring the role of adverse experiences in FND, which have found associations with elevated rates of trauma and stressful life events (e.g. Ludwig *et al*., 2018). As discussed, these have led to criticism and a move away from older psychodynamic models which emphasised the role of trauma in FND (Reuber, 2018). However, this review suggests the picture is more complex, in that there is some evidence that the role of trauma may be more implicated in the development of non-epileptic seizures compared to functional motor symptoms. Any successful model of FND would need to recognise this. It was also striking to find that very few studies appeared

particularly relevant to the cognitive-behavioural model of FND. Instead, hypotheses tended to relate more to psychodynamic and dissociative models. This is despite the fact that a cognitive-behavioural approach currently appears to be the most common method of psychological treatment for people with FND. However, this may also highlight a need to incorporate more longitudinal factors into cognitive-behavioural treatment models, such as the role of personality traits or traumatic experiences.

Limitations

This review's conclusions are limited, in that only a small number of studies were identified which met the eligibility criteria and the majority of these were appraised as having only moderate methodological quality. In addition, many of the variables examined were only represented in one or two studies. This lack of replication reduces the confidence that can be placed in their conclusions. Most of these studies were also reliant on self-report questionnaires that did not appear to have been validated in target populations. As discussed, this may undermine the validity of the results of some studies. Given these limitations, and the fact that some of the studies excluded at the last stage of study selection actually proved useful in interpreting the findings (e.g. Zeng *et al*., 2018), it may be that the reviews' exclusion criteria were too strict. The analysis may have benefited from including studies which examined comparisons of subtypes of non-epileptic seizure.

Another methodological limitation is that there was no second reviewer at any point during the review process. This may have introduced bias into the processes of selection, extraction, appraisal or synthesis of the studies involved (Boland, Cherry & Dickson, 2017). However, the methodology of the review drew on the best practice guidance for systematic

reviews (Moher *et al*., 2009), existing published reviews (e.g. Brown & Reuber, 2016), best practice guidelines for research in the field (e.g. Edwards & Bhatia, 2012), and established research quality appraisal tools. Arguably, these measures will have provided mitigation against the risk of bias significantly affecting the review's conclusions.

A further limitation of the review was that no studies could be identified which examined any other type of FND other than non-epileptic seizures or functional motor symptoms. Whilst there is evidence to suggest that around 64% of all functional neurological symptoms will fall into these two categories (Ahmad & Ahmad, 2016), these are not the only manifestations of FND. Studies examining comparisons involving other types of FND, such as functional sensory alterations or functional voice difficulties could not be identified, meaning that the review is unable to comment on the psychological profiles of these disorders. Studies of these disorders have been reported examining similar variables (e.g. Kosztyła-Hojna *et al.*, 2018), so there is potential for future comparative studies.

Future Directions

More research is warranted in this area to further develop these preliminary findings. However, this review also highlights a number of quality issues which future research may need to improve on. For instance, very few studies included a power calculation, and there was some reason to believe that this may have led to type II errors in some analyses, so best practice around power calculations in studies should be adhered to. Future studies should also aim to use some measures that are not reliant on self-report, such as psychiatric interviews, observation schedules, or ratings of clinical notes.

One hypothesis for the variability of some findings was that this may have been caused by variability in levels of psychopathology between samples with non-epileptic seizures. It was noteworthy that very few of the studies identified were entirely clear about the referral source of their samples (such as an inpatient unit), which is important information for characterising the complexity or severity of a functional presentation. Indeed, few studies made any attempt to characterise the severity of the functional symptoms present within the samples used. It may be that greater psychopathology in some samples is the result of more severe functional symptoms, and further attention is needed in future research to consider and control for this possibility. A standardised clinician-rated measure known as the Functional Neurological Disorders Rating Scale (unpublished) is currently in development. This allows the severity of functional neurological presentations to be characterised numerically and may support improvements in future research designs.

It is also recommended that further research of this type should be conducted with other types of functional neurological presentation. Existing research appears to have concentrated exclusively on comparisons of participants with non-epileptic seizures and functional motor symptoms. Future research could, for instance examine comparisons involving isolated functional sensory symptoms, functional voice disorders or cognitive difficulties. There is also a need for the place of mixed functional neurological presentations to be considered. Research by Ahmad & Ahmad (2015) suggests around 25% of patients diagnosed with FND will sufferer from more than one class of functional neurological symptom. Given what has been discussed above, it might be hypothesised that the greater the number of functional neurological symptoms a patient presents with, the greater the level of psychopathology which might be evident.

Lastly, there is also appears to be a need for future studies of this kind to examine factors relevant to cognitive-behavioural models of the disorder, given that this is the most common form psychological intervention patients are likely to encounter. Research of this kind might examine the comparative prevalence of events predisposing those with FND to cognitive representations matching their symptoms, such as experience of prior epileptic seizures or organic motor difficulties.

Empirical Project: Is there a relationship between maladaptive schemas and functional neurological disorders?

Abstract

FND have been associated with a range of predisposing psychological factors, which include elevated rates of adverse experiences, disordered adult attachment style, and alexithymia. Studies have linked FND to increased rates of personality disorders, most consistently Borderline Personality Disorder and Obsessive-Compulsive Personality Disorder. It can be argued that these findings suggest that maladaptive schemas, a concept derived from schema therapy, may be useful in understanding FND. The present study examined the role of maladaptive schemas in participants with FND (*N* = 25) recruited from inpatient, outpatient and day-patient services. Based on existing findings and the schema model, it was hypothesised that scores for Disconnection and Rejection and Excessive Responsibility schema domains, as well as related perceived parenting scores, would be negatively correlated with participants' self-ratings of current health. Secondly, it was hypothesised that schema scores would appear elevated above normative data based on findings from a similar large-scale study. Participants completed a single set of self-report questionnaires measuring anxiety, depression, personality disorder prevalence, maladaptive schemas and perceived parenting in childhood. There were small negative correlations between the identified schema domains and the current health status. These correlations were not statistically significant, though this may have been because the study was somewhat underpowered. The correlations between the hypothesised perceived parenting styles and current health status were also not statistically significant. However, the schema domain scores were significantly higher than the large-scale student sample, and in some cases were comparable with those of a clinical sample. This is thought to be the first study of its

kind to examine the role of maladaptive schemas in FND. It provides preliminary evidence that a range of maladaptive schemas may be more prevalent in FND populations.

Introduction

'Functional Neurological Disorders' (FND) is an umbrella term which describes the presence of neurological symptoms in the absence of any demonstrable neurological disease or injury. Symptoms can include non-epileptic seizures and movement difficulties, such as limb weakness or gait problems. The latter are referred to collectively as functional motor symptoms. Other symptoms of FND can include voice and swallowing difficulties, as well as alterations to cognition and awareness, such as memory problems, visual difficulties, or changes in bodily sensation. People with FND may present with one or several of these symptoms (Stone, 2013).

The word 'functional' in this context indicates that what is observable in the condition is the abnormal function of a bodily system in the absence of pathology. It is intended to be an alternative to terms like 'psychogenic', which imply more assumptions about the causes of symptoms, and which tend to be poorly understood by service-users (Edwards & Bhatia, 2012; Stone, 2013). Accurate estimates of the prevalence of FND have proved difficult to calculate (Edwards & Bhatia, 2012). However, they are a common presentation in neurology clinics, where they are thought to make up around 16% of new referrals (Stone *et al.*, 2010).

Psychosocial Impact

FND have a significant impact on the lives of sufferers and have the potential to cause significant costs to health and social care systems. In a study of 3781 neurology outpatients, Carson *et al*. (2011) compared patients whose symptoms neurologists had rated as being 'not at all' or 'somewhat' unexplained by disease with those whose symptoms which were rated as 'largely' or 'completely' explained. Compared to controls, the medically

unexplained symptom group had significantly higher levels of distress, and rated both their physical and mental health as poorer. Participants in Carson *et al*.'s (2011) research were also more likely to be unemployed due to ill health and more likely to be in receipt of disability-related benefits. In 2009, it was estimated that medically unexplained illnesses like FND cost the National Health Service (NHS) around £18 billion a year; slightly more than the yearly cost of dementia at all ages (Bermingham, Hague, Cohen & Parsonage, 2010).

Despite this level of impact, the aetiology of FND is often described in the literature as poorly understood (Edwards & Bhatia, 2012). Historically, FND has been under-researched, though since the millennium, the number of studies examining FND has increased (Carson *et al.*, 2012). Various psychological models for the condition exist, though reviews have found the evidence for these to be limited and inconsistent (Brown & Reuber, 2016). The various psychological models of FND are discussed in greater depth in the introduction to the associated systematic review attached to this report.

Psychotherapy is often recommended as an intervention for FND, typically cognitive-behavioural therapy (CBT; Stone, 2016), though at present the evidence-base for psychological interventions in FND is also limited (Carson & Perry, 2017; Martlew, Pulman & Marson, 2014). Relatedly, there is no National Institute of Clinical Excellence (NICE) guideline concerning evidence-based treatments for FND. A draft guideline on suspected neurological conditions has been released (NICE, 2017), though this has been criticised for only superficially referring to FND as exclusion criteria for referral to neurology (FND Hope UK, 2017). Evidently, further research into FND is a priority, both to improve understanding of their origins, and to support effective treatment planning and management of the

condition.

Predisposing Psychological Factors in FND

In recent years, studies have concentrated on expanding knowledge concerning the predisposing psychological risk factors for FND. This introduction will survey several recent areas of research before drawing them together into a hypothesis concerning the role of maladaptive schemas in the condition.

Many studies have examined the role of past adverse experiences in FND. As a result of the influence of psychodynamic models, it was once held that traumatic experiences were central to the development FND (Reuber, 2018). However, this has only been partially supported by evidence. While rates of traumatic experiences are elevated in people with FND, a substantial proportion have no detectable traumatic experiences. A recent meta-analysis of case control studies indicated that rates of childhood emotional neglect, physical abuse and sexual abuse tend to be elevated in those with FND against controls, though the difference was most pronounced in emotional neglect (49% of FND cases vs. 20% controls). FND populations also show evidence of increased rates of adverse experiences in adulthood; the same meta-analyses found that rates of significant life events in the year before onset of FND were consistently higher than in controls (Ludwig *et al*., 2018).

The accumulation of findings like these have led to criticism of previous psychodynamic, trauma-focused models of FND, and a move towards a more present-focused approach to the disorder (Reuber, 2018). However, it is important to note that there appears to be some evidence that the prevalence of traumatic experiences may vary depending on the type of

FND. For instance, people with non-epileptic seizures alone appear to be more likely to report experience of childhood abuse and neglect, sexual abuse and stressful life events compared to people with only functional motor symptoms (see attached systematic review for further details).

Recent research has also suggested a general association between FND and various unhelpful psychological traits, which may be important in understanding the precise role of trauma in FND. For instance, Holman, Kirkby, Duncan and Brown (2008) examined the role of disordered attachment in FND. They found that 17 participants with non-epileptic seizures had significantly higher levels of fearful adult attachment style than controls with epilepsy, as well as significantly higher scores for experiences of abuse and neglect. This suggests that adverse early-life experiences and later resultant difficulties in coping with emotional and interpersonal situations may be implicated in the development of FND.

Other studies have found associations between FND and elevated rates of personality disorders. It is important to note again, however, that the specifics findings have varied by study. In a study of 56 patients with non-epileptic seizures, Howorka, Nezadal, Herman, Nemcova and Bajacek (2007) found 45% of patients with non-epileptic seizures met criteria for personality disorder, predominantly BPD (32%). Reuber, Pukrop, Bauer, Derfuss and Elger (2004) studied 85 patients with non-epileptic seizures using a dimensional measure of personality pathology. The main clusters they found within their sample were that 51% of the sample had a profile that resembled BPD, and 44% had a personality profile which was characterised by over-control, resembling the criteria for Obsessive Compulsive Personality Disorder (OCPD).

Similarly, in a study of participants with functional motor symptoms, Feinstein, Steriopolous, Fine and Lang (2001) found 45% of a sample of 88 patients met the criteria for a personality disorder, particularly antisocial personality disorder, BPD and dependant personality disorder. More recently, Demartini *et al.* (2014) compared a sample of 55 patients with functional motor symptoms to healthy controls, and found that the only significantly elevated rate of any personality disorder was for OCPD, with 25% of the sample meeting these criteria. In a similar comparative study, Stone, Sharpe and Binzer (2004) found that rates of any kind of personality disorder were elevated to similar levels in patients with functional motor symptoms and non-epileptic seizures (50 vs. 65%). However, when BPD alone was considered, patients with non-epileptic seizures had significantly higher rates (35% vs. 5%) than those with functional motor symptoms.

These studies suggest rates of personality disorder appear to be elevated in people with FND. This would suggest that sustained difficulties in emotional and interpersonal coping are present for a substantial proportion of those diagnosed with FND. The specific personality disorder diagnoses that have been over-represented in samples has varied between studies, although BPD and OCPD appear to be the most consistent associations. Difficulties in emotional coping have been highlighted by other studies on FND. A number of studies have found that people with FND show higher rates of alexithymia than controls (Demartini *et al*., 2016; Ekanayake *et al*., 2016; Demartini *et al*., 2014). Alexithymia refers to a difficulty in identifying, understanding and communicating one's emotional state, and is thought to be a factor in a range of mental health presentations. (Sifneos, 1973).

Taken as a whole, the studies discussed above suggest that FND are associated with adverse

experiences as well as difficulties coping with emotions and interpersonal situations. However, it also seems that the exact nature of these findings has varied across studies. As discussed in the systematic review attached to this study, these variations may be the result of a number of factors, including the type of functional neurological symptoms being studied, a high degree of variability between different samples of patients with FND, or in some cases the quality of research designs.

Maladaptive Schemas

The present study explores the utility of the theories associated with schema therapy (hereafter 'the schema model') as an explanatory model for understanding these findings concerning FND, with particular focus on the concept of a maladaptive schema. Schema therapy developed out of CBT in the 1990s, aiming to provide more appropriate therapeutic interventions for clients who had long-standing, complex psychological difficulties, such as personality disorders. While retaining many features of CBT, schema therapy "expands on traditional cognitive-behavioural therapy by placing much greater emphasis on exploring the childhood and adolescent origins of psychological problems, on emotive techniques, on the therapist-patient relationship, and on maladaptive coping styles" (p. 5, Young, Klosko & Weishaar, 2003). There is emerging evidence which indicates that schema therapy is a safe and effective treatment for BPD (Masley, Gillanders, Simpson & Taylor, 2012), as recognised by NICE clinical guideline 78 (2009).

The idea of a maladaptive schema has its roots in the work of Beck (1967), who defined a cognitive schema as: "a structure for screening, coding and evaluating the stimuli that impinge on the organised. It is the mode by which the environment is broken down and

organised into its many psychologically relevant facets. On the basis of schemas, the individual is able to … categorise and interpret his experiences in a meaningful way (p. 283)". Young (1999) developed this concept further by observing that a range of specific maladaptive schemas seemed to be common obstacles in traditional cognitive therapy. Young hypothesised that maladaptive schemas tended to have their origins in toxic childhood experiences, and developed a systematic account of various types of maladaptive schemas, as well as methods to assess and treat them.

Schema therapy defines maladaptive schemas as a range of self-perpetuating, cognitive-emotional patterns, which related to emotions or relationships with others, and which interfere with an individual's life to a significant degree. Drawing on psychodynamic, attachment and cognitive-behavioural theories, the schema model argues that the development of maladaptive schemas depends on a child's individual variability in emotional and relational needs, and the extent to which these are met by their environment. It argues that where childhood emotional needs are seriously and consistently unmet, this can lead to the internalisation of unhelpful messages about the self, which impair well-being and function in adulthood (Young *et al.*, 2003).

The schema model has identified lists of potential maladaptive schemas which have been revised over time, in part through factor analysis research. The present study will use the system of classification developed through a large-scale factor analysis by Bach, Lockwood and Young (2018). This defines 18 different schemas, grouped into four categories or schema domains: Disconnection and Rejection; Impaired Autonomy; Excessive Responsibility; and Impaired Limits. A schema domain refers to a general emotional and

interpersonal area with which an individual might struggle, defined by a set of individual schemas which factor analyses have clustered together. Table 1 gives a fuller account of this structure. It should be noted that aside from Table 1, for the sake of brevity I have opted to use shortened names for some schemas and schema domains in this report (e.g. 'Impaired Autonomy', 'Excessive Responsibility', 'Alienation',). Further details about the psychometric development of this system of classifying schemas can be found in below (Measures: The Young's Schema Questionnaire 3, Short Form).

Table 1

Descriptions of Maladaptive Schema Domains

Schema Domain	Schemas	Description
Disconnection & Rejection	Emotional Deprivation Social Isolation/Alienation Emotional Inhibition Defectiveness/Shame Mistrust/Abused Pessimism	"the expectation that one's needs for security, safety, stability, nurturance, empathy sharing of feelings, acceptance, and respect will [not] be met in a predictable manner" (Young *et al.*, 2003, p. 14)
Impaired Autonomy & Performance	Dependence Failure Subjugation Abandonment Enmeshment Vulnerability to Harm	"Expectations about oneself and the environment that interfere with one's perceived ability to separate, survive function independently, or perform successfully" (Young *et al.*, 2003, p. 14)
Excessive Responsibility & Standards	Self-Sacrifice Unrelenting Standards Self-Punitiveness	"Excessive emphasis on … meeting rigid internalized rules and expectations about performance and ethical behaviour, often at the expense of happiness, self-expression, relaxation, close relationships or health." (Young *et al.*, 2003, p. 16-17)
Impaired Limits	Entitlement Approval/Admiration Seeking Insufficient Self-Control	"Deficiency in internal limits, responsibility to others, or long-term goal orientation. Leads to difficulty respecting the rights of others, cooperating with others, making commitments, or setting and meeting realistic personal goals" (Young *et al.*, 2003, p. 15)

Note. In the rest of this report, for the sake of brevity I have opted to use shortened names for some schemas and schema domains (e.g. 'Impaired Autonomy', 'Excessive Responsibility', 'Alienation').

Schema therapy offers an evidence-based model for how adverse life experiences may inform adult strategies for coping with emotion and relationships, and the way in which this may influence various psychological disorders. Previous research has shown that maladaptive schemas and perceived parenting experiences can offer an explanatory model of psychological difficulties later in life. For instance, Sheffield, Waller, Emanuelli, Murray and Meyer (2009) found that in 124 participants with eating disorders, schema processes mediated the relationship between specific perceptions of parenting and specific forms of eating pathology. Similarly, in a study of 173 adolescents, Muris (2006) found that maladaptive schemas were predictive of a range psychological difficulties, including the symptoms of anxiety disorders and depression, disruptive behaviour, eating problems and substance misuse. In addition, a study of 145 psychiatric outpatients by Thimm (2011) found that levels of maladaptive schemas predicted the variation in personality disorder symptoms over and above five-factor dimensional personality traits.

By viewing FND through the lens of the schema model, it might be hypothesised that the increased rates of adverse experience in FND are implicated in an increased risk of developing maladaptive schemas. These may then manifest as personality dysfunction and other maladaptive traits later on in life. The schema model may also help to explain some of the heterogeneous findings in relation to different types of functional neurological presentation. It may be that different types of FND are associated with different patterns of maladaptive schemas. The schema model also has the benefit of defining psychological coping strategies to a fine degree of detail, offering the potential to characterise clinical samples in depth.

Objectives

The present empirical project was a cross-sectional study of a clinical sample of participants with FND. It aimed to explore the explanatory role of maladaptive schemas in this patient-group. No previous quantitative study could be found which had examined the role of maladaptive schemas in FND, though one case series of three patients was found (Flores, 2016), as well as one registration for an ongoing study (Levy & Guilhem, 2018) were found.

In order to design the study, it was first necessary to identify what predictions the schema model might make about FND. Though research has linked FND to a range of personality disorders, there appeared to be slightly firmer associations between FND, BPD and OCPD. Schema therapy broadly associates BPD and OCPD with the Disconnection and Rejection and Excessive Responsibility domains (Young *et al.*, 2003). It was predicted that the presence of schemas of this kind would be involved in the current functional neurological difficulties participants were experiencing. It was decided to operationalise participants' current difficulties as a self-report measure of health status. Hence, it was predicted that in a sample of participants with FND, greater endorsement of schemas in these domains would be associated with poorer self-reported health status.

The schema model also predicts that particular schemas are associated with the experience of particular parenting styles in childhood. Using existing research into the schema model (Bach *et al.*, 2018), a number of perceived parenting styles which have previously been correlated with the identified schema domains were also selected; these are listed below. As a result, it was predicted that endorsement of these perceived parenting styles on self-report measures would also be negatively correlated with current self-reported health.

Lastly, it was also predicted that scores for the identified schema domains would be elevated in comparison to normative data from a previous study (Bach *et al.*, 2018).

To summarise, the hypotheses were that:

1) Scores for the Disconnection and Rejection and Excessive Responsibility domains would be negatively correlated with scores on a self-report measure of current health.

2) Emotionally depriving, belittling, perfectionistic and controlling parenting subscales on a self-report measure of perceived parenting in childhood would negatively correlate with self-reported current health.

3) Scores for the Disconnection and Rejection and Excessive Responsibility domains would be elevated in comparison to normative data from a study by Bach *et al*. (2018).

As discussed, no previous study could be found which had examined the role of maladaptive schemas in FND quantitatively. The study was thought to have the potential to make an original contribution to an area of clinical research in need of further investigation. Schema therapy shares common ground with CBT, which is often recommended as part of treatment of FND (Stone, 2013). As a result, the research was also thought to be valuable as the findings might have the potential to inform future developments regarding psychological treatment for FND.

Methods

Participants

25 participants were recruited consecutively from neuropsychiatry services in South London and Maudsley NHS Foundation Trust over an eight-week period. Participants included inpatients, outpatients and day-patients. All of the services involved in the recruitment process operate as national specialist centres within the NHS. This means they accept referrals from anywhere in the UK for assessment and treatment. As such, it was thought that they are likely to represent a moderate to severe FND population, as their patients are likely not to have benefited sufficiently from any available local services. Demographic information on the participants is presented in Table 2.

Table 2
Demographic Statistics for the sample

Characteristics	Total (%) (*N* = 25)
Gender	
Female	17 (68%)
Male	7 (28%)
Prefer not to say	1 (4%)
Age (years)	
Mean	42
Range	20-64
Standard Deviation	13.65
Current Employment	
Employed	7 (28%)
Unemployed	14 (56%)
Student	1 (4%)
Retired	2 (8%)
Disability benefits	1 (4%)
Employment before FND	
Employed	20 (80%)
Unemployed	1 (4%)
Student	2 (8%)
Retired	0 (0%)
Disability benefits	0 (0%)
Marital Status	
Married/Civil Partnership	7 (28%)
Unmarried relationship	4 (16%)
Single	13 (52%)
Prefer not to say	1 (4%)
Pregnancy	0 (0%)

Parenthood	
Children	13 (52%)
No children	11 (44%)
Prefer not to say	1 (4%)
Sexual Orientation	
Heterosexual	22 (88%)
Bisexual	2 (8%)
Prefer not to say	1 (4%)
Perceived Disability	
Disabled	18 (72%)
Non-disabled	5 (20%)
Prefer not to say	2 (8%)
Current Patient Type	
Inpatient	7 (28%)
Outpatient	9 (36%)
Day-patient	9 (36%)
Stage of Current Treatment	
Start	7 (28%)
Middle	8 (32%)
End	10 (40%)
FND Symptoms	
Non-epileptic seizures	12 (48%)
Motor	18 (72%)
Sensory	4 (16%)
Speech/Swallowing	3 (12%)
Cognitive	2 (8%)
Visual	1 (4%)
Perceived Current Mental Health Problem	
Yes	9 (36%)
No	14 (56%)
Prefer not to say	2 (8%)
Previous Treatment for Mental Health Problems Reported	
Yes	13 (52%)
No	10 (40%)
Prefer not to say	2 (8%)

Table 3 summarises the functional motor symptoms experienced by sample. Dystonia refers to muscle contractions, while myoclonus refers to spasmodic jerky movements.

Table 3

Functional Motor Symptoms in the sample

Motor Symptoms	Total (%) (n = 17)
Tremor	4 (24%)
Gait Difficulties	3 (18%)
Balance Problems	4 (24%)
Dystonia	2 (12%)
Myoclonus	4 (24%)
Weakness	10 (59%)

Patients were excluded from the study for the following reasons: a) they were judged by their care team to lack capacity to give informed consent to participate; b) they were diagnosed with a functional disorder which seriously impaired their memory or cognition; c) they were diagnosed with a major mental health problem, such as Schizophrenia, Bipolar Affective Disorder, active Post-Traumatic Stress Disorder, or an active substance misuse problem; d) they were diagnosed with a degenerative brain disorder or brain injury; they had a diagnosis of epilepsy; e) they were diagnosed with a Learning Disability; f) they had a diagnosis of Autistic Spectrum Condition; g) their functional neurological disorder diagnosis was in doubt; h) their communication needs meant they could not understand or complete the required study materials; i) their care team felt that their participation might destabilise their mental health or lead to a risk of harm.

Diagnosis of a personality disorder was not included in the exclusion criteria. No participant was reported to have a diagnosis of a personality disorder by a referrer, though this was not formally recorded. This was because this was not explicitly reported by any participant in the study, or by the referring clinicians. The number of eligible participants who explicitly declined to participate was recorded where possible. In total, 11 (31%) people were recorded as declined to participate in the study when were approached by their clinician, not including those who did not respond to communications. The number of participants or were screened but not approached because they met one of the exclusion criteria was not recorded.

Ethical approval for the present study was granted by the Surrey–London NHS Research

Ethics Committee (see Appendix 2), the Health Research Authority (see Appendix 3), and the Joint R&D Office of South London and Maudsley NHS Foundation Trust and Institute of Psychiatry, Psychology & Neuroscience.

Power

In order to calculate the necessary sample size, Cohen's (1992) tables were used. The first two hypotheses would be analysed through correlations. The study explored a previously un-researched area and recruitment of a large sample was not feasible in the time available. As such, it was deemed justifiable to power the study only for large effect sizes. However, consideration of previous similar studies provided some justification for expecting medium to large effect sizes. For instance, Sheffield *et al*. (2009) examined the associations between schema measures of perceived parenting styles and eating disorder symptom measures in 353 healthy students and 124 patients with eating disorders. They found statistically significant correlations ranging between .23 and .35, which were almost all medium effect sizes according to thresholds given by Cohen (1988). In another similar study, Sheffield, Waller, Emanuelli and Murray (2006) examined whether subscales of schema measures of perceived parenting styles to distinguish between impulsive and non-impulsive patients with eating disorders (N = 124). In significance tests, Cohen's d ranged from 0.77 to 2.04. Almost all effect sizes they calculated were above the threshold considered indicative of large effects.

According to Cohen (1992), a large predicted effect size in a correlational study design necessitates a minimum sample size of 28 to achieve a power of .80. Due to the eight-week time period available, only 25 participants could be recruited. The reasons for this are

discussed in the Integration, Impact and Dissemination section of this thesis. This sample size means that the present study is likely to be slightly underpowered to detect even large associations in the sample.

Measures

All six measures used were self-report questionnaires. Measures are presented in the order they were delivered in. The first measure was a non-standardised demographics questionnaire devised for the study. This asked for participants the following: their age; ethnic group; relationship status; sexual orientation; whether they had children; pregnancy if applicable; their perceived disability status; their employment status before and after becoming unwell, and whether they had experienced mental health problems in the past (see Appendix 4 for demographics form). Participants' functional neurological diagnoses were also obtained from the clinicians who had referred them to the study, provided participants consented to this. Consent was given in all cases. To minimise the amount of confidential information held by the study, no other clinical information was obtained from referring clinicians for analysis.

The EQ-5D-5L (Herdman et al., 2011)

In order to characterise the current perceived health of participants, the EQ-5D-5L was used (see Appendix 5). The EQ-5D-5L, which is the unabbreviated name of the questionnaire, is a six-item measure assessing health status. Participants endorse one of five statements (e.g. "I am unable to walk") with regards to five dimensions of health - mobility, self-care, usual activities, pain/discomfort, anxiety/depression - for the day on which they complete the questionnaire. The sixth item is a visual analogue scale (VAS) which asks participants to rate

their current health in general by choosing a score between 0 and 100, for the day the questionnaire is completed. Using an algorithm, answers to the first five questions can be converted into a weighted EQ Index Score, which represents the value attached to this health state according to a given population. This means EQ Index Scores are standardised according to large datasets from various countries (Reenen & Jansen, 2015). In the present study EQ index scores were calculated using the supplied dataset for the United Kingdom. When this dataset is used, EQ Index Score can range between 1 and -.59.

Though the measure has not been validated in the target population, it has been validated for a range of chronic disorders associated with disability. For instance, Janssen *et al.* (2013) evaluated the EQ-5D-5L across six countries and eight patient groups, including cardiovascular disease, respiratory disease, depression, diabetes, liver disease, personality disorders, arthritis, and stroke, as well as a student cohort (*N* = 3,919). It also showed good convergent validity with the World Health Organisation-Five Well-being Index (*n* = 1001, UK only sample; World Health Organisation, 1998). In addition, it showed known-groups validity, improved discrimination and improved ceiling effects in comparison to previous versions. Research by Conner-Spady *et al*. (2014) has found that the EQ-5D-5L had good test-retest reliability in patients with osteoarthritis (*N* = 176), with interclass correlation coefficients between 0.61 and 0.77.

The Patient Health Questionnaire 9 (PHQ-9; Kroenke, Spitzer & Williams, 2001) and the Generalised Anxiety Disorder Questionnaire 7 (GAD-7; Spitzer, Kroenke, Williams & Löwe 2006)

In order to evaluate the general mental health of participants, PHQ-9 and GAD-7 were also

used. These are established self-report measures of depression and anxiety respectively. Previous research has established their reliability and validity in large samples (Kroenke, Spitzer & Williams, 2001; Löwe *et al.*, 2008; Spitzer *et al.*, 2006).

The International Personality Disorder Examination Screening Questionnaire (IPDE; (Loranger, Janca & Sartorius, 1997)

Given the background to the present study, it was thought necessary to include a measure from which to estimate the prevalence of personality disorders in the sample. The IPDE was selected for this purpose (see Appendix 6). The IPDE is a brief screening tool for personality disorders developed by the WHO as part of a wider interview schedule for assessing personality disorders. It uses 59 items which participants rate as true or false of themselves. Participants are asked to answer the items based on their experience of the past five years. The measure has nine subscales which relate to various personality disorders. According to the original directions for use, if a participant scores over a threshold of three items on any subscale, this indicates that they show signs of having a personality disorder and should be interviewed by a clinician.

However, the tool has also been used as a standalone measure to estimate the prevalence of personality disorders in large samples. Research has found that it has acceptable psychometric properties for this role (Lewin, Slade, Andrews, Carr & Hornabrook, 2005). Use of a higher threshold of four items improves its psychometric properties where it is used in populations known to have an elevated prevalence of personality disorders (Alvaro-Brun *et al.*, 2008). This is the threshold that has been used in the analyses below.

The Young's Schema Questionnaire 3, Short Form (YSQ3-SF; Young, 2016)

In order to measure maladaptive schemas within the sample, the YSQ3-SF was used (see Appendix 7). It includes a total of 90 items and 18 subscales for different maladaptive schemas. Items (e.g. "I don't belong; I'm a loner') are rated on a six-point scale as to how true the participant believes they are of themselves. The subscale scores are calculated as the means of five items, meaning scores they also between one and six. Participants are asked to answer the items based on their experience of the past year. The YSQ3-SF was developed from an already validated 205-item version (Young, 1994). This is the most recent short form version published by the Schema Institute (Young, 2016).

Unfortunately, much of the available evidence concerning the psychometric properties of the YSQ3-SF relates to older versions of the questionnaire, such as the Young Schema Questionnaire 2, Short Form (YSQ2-SF). For instance, Welburn, Coristine, Dagg, Potefract and Jordan (2002) administered the YSQ2-SF to 196 psychiatric outpatients. They found Cronbach's alpha coefficients for the schema subscales ranged from .76-.93, indicating moderate to good internal consistency. There were also statistically significant correlations with subscales of the Brief Symptom Inventory (Derogatis, 2000), indicating convergent validity. Lucia, Thorne, Waters and Preston (2001) also evidenced the parallel forms reliability of the YSQ2-SF with its longer version. In the YSQ3-SF three more subscales (approval-seeking, pessimism and self-punitiveness) and 15 more items have been added. These additions were made for theoretical reasons, but the evidence for them at present is limited (Young, 2016).

Recent research into the YSQ3-SF has suggested a two-layer, hierarchical factor solution.

Bach *et al.* (2018) analysed the measure in a sample of 1049 students and psychiatric outpatients. They found evidence for four 'schema domain' factors (larger groupings of individual schema subscales), as well as evidence for the hypothesised 18-factor solution. Schema domain subscales are also calculated as means of items, meaning they can range between one and six. As discussed in the introduction, this is the schema system used in the present study.

The Young's Parenting Inventory, Revised (YPI-R; Sheffield, Waller, Emanuelli, Murray & Meyer, 2006)

In order to measure participant's perception of the parenting they received during childhood, the YPI-R was used (see Appendix 8). It uses 37 items. Participants rate how true each item is of their memories of their mother and father in childhood and early adolescence (e.g. "When I was growing up as a child and young teenager [my mother/father] made me feel ashamed of myself in important respects"). Each item is rated twice on a six-point scale: one rating for their mother and one for their father. Nine subscales identify different kinds of unhelpful parenting styles: emotionally depriving; overprotective; belittling; perfectionistic; pessimistic; controlling; emotionally inhibited; punitive and conditional parenting. Subscale scores are again calculated as means of items, so they can range between one and six. The instructions indicate that if a participant feels a parent was absent to the degree that they cannot answer the questions (e.g. through bereavement), they should leave these items unanswered. Respondents are also encouraged to answer both sets of items using other important figures who may have taken the role of a parent if their family structure did not consist of a mother and/or father.

The YPI-R was developed by the authors through confirmatory factor analysis of an original 72-item measure (Young, 1994). In a student sample (*N* = 422) only those measures with a Cronbach's alpha greater than .65 were selected. Test-retest reliability was found to be adequate across all subscales. Relevant scores on the YPI-R were correlated with scores on the YSQ3-SF. There were also associations between some subscales, lending partial support for construct validity of the YPI-R. Sheffield *et al.* (2006) subsequently demonstrated the criterion validity of the YPI-R with subscales of the Brief Symptom Inventory (Derogatis, 2000) in their research examining eating disorders in a sample of women (*N* = 124). Furthermore, Bach *et al.* (2018) demonstrated that there is evidence that subscales of the YPI-R correlate with schema domain scores in a way predicted by the schema theory. For instance, the authors found that participant's scores for how emotionally depriving they rated their mothers as, had a .32 correlation with their scores for the Disconnection and Rejection schema domain subscale.

Procedure

Participation involved completing a single set of measures at one time-point. Participants completed the measures in a confidential space at the clinical service which they had been recruited from. Only the principal researcher was present. Measures were completed using pen and paper. Participants were given the Participant Information Sheet (see Appendix 9) to read at least 24 hours before participation, which included information about the aims of the research and the confidentiality procedures used. The appointment to complete the study questionnaires began with the principal researcher discussing the information sheet with the participant. Any questions the participant had about the study were answered. Participants were reminded that they were under no obligation to participate and could

withdraw without any consequence at any time. They were also reminded that some of the questionnaires contained content they might find distressing, such as items about negative self-image or memories of perceived parenting in childhood. At the suggestion of the NHS Research Ethics Committee, participants were also advised that knowledge about the causes of FND is limited, and that they should not assume that the content of the questionnaires was necessarily important in the development of FND in their particular case.

After recording informed consent (see Appendix 10 for Consent Form), participants completed the demographics form. They then completed the measures in the order they are presented above. After they had completed the questionnaires, participants were given the opportunity to briefly discuss any questions or feedback they had on the study with the principal researcher, and finally thanked for their participation in the study. This appointment typically took around 45 minutes to complete.

Plan for Statistical Analysis

Most statistical analyses were completed using SPSS Version 21. It was planned that the first and second hypotheses would be evaluated through bivariate correlations tested for statistical significance.

In order to evaluate the third hypothesis, unpublished data from a study by Bach *et al*. (2018) was requested and obtained from the lead author. This study was a cross-sectional study of 391 students and 658 psychiatric outpatients. The latter were specifically described as “592 non-psychotic psychiatric outpatients and 66 rehabilitants in treatment for alcohol/drug abuse, and all had predominant features of personality pathology (primarily

Cluster B and C personality disorders" (Bach *et al.*, 2018, p. 6). Hereafter these samples are referred to as the 'Student' and 'Clinical' sample. The measures administered in this study included the YSQ3-SF and YPI-R. The specific data requested from the authors were the means and standard deviations for all subscales of the YSQ3-SF and YPI-R for both samples. Means and standard deviations for the schema domain scores were then calculated. It was planned that the third hypothesis would be evaluated by conducting *t* tests of schema domain scores and visual analysis of graphs. As SPSS is unable to calculate *t* tests and tests of homogeneity of variance from summary data alone, two online calculators were used to perform these calculations. Unpaired *t* tests and Bartlett's test of unequal variance were completed using the StatTools online package offered by the Chinese University of Hong Kong (Chang & Sahota, 2017). Where Welch's *t* tests were used, these were completed using the online calculator provided by GraphPad software company (GraphPad, n.d.).

Service User Involvement

It was intended that the results of the study would be presented to an appropriate service-user group, in order to gather feedback to inform the interpretation of the results. Unfortunately, due to delays in beginning recruitment, it was not possible to secure time with a service-user group. However, notes were taken regarding participants' feedback on the study after they had completed the measures, and this has been used to inform the interpretation of the results. Issues connected with service-user involvement are discussed in greater depth in the Integration, Impact and Dissemination section of this thesis.

Results

Demographics statistics regarding the sample are presented in Table 2. Descriptive statistics for the main measures are presented in Table 4. Data for participants with non-epileptic seizures only or functional motor symptoms only is also shown. Significance testing between these two groups was not pursued as sample sizes were too small. However, the data is presented here for comparison as the systematic review highlighted that these groups may score differently on psychological measures.

Table 4

Descriptive Statistics for the sample

Scale	Total Mean (*SD*)	NES only Mean (*SD*)	FMS only Mean (*SD*)
GAD-7	(*N* = 25)	(*n* = 7)	(*n* = 13)
Total Score	10.60 (6.28)	13.14 (5.70)	10.31 (6.06)
PHQ-9	(N = 25)	(*n* = 7)	(*n* = 13)
Total Score	11.76 (7.53)	14.79 (7.69)	10.92 (7.55)
EQ-5D-5L	(*N* = 25)	(*n* = 7)	(*n* = 13)
EQ Index	.445 (.410)	.400 (0.492)	.438 (0.371)
VAS Score	61.80 (24.87)	53.86 (36.23)	61.77 (21.44)
YSQ3-SF			
Schema Domain Scores:	(*N* = 25)	(*n* = 7)	(*n* = 13)
Disconnection & Rejection	2.72 (1.08)	3.07 (1.19)	2.68 (1.16)
Impaired Autonomy	2.77 (0.93)	2.45 (0.60)	2.90 (1.08)
Excessive Responsibility	3.54 (1.03)	3.78 (0.98)	3.62 (1.07)
Impaired Limits	2.37 (0.81)	2.37 (0.73)	2.43 (0.96)
Individual Schema Scores:	(*N* = 25)	(*n* = 7)	(*n* = 13)
Emotional Deprivation	2.33 (1.32)	2.83 (1.79)	2.28 (1.16)
Alienation	2.96 (1.44)	3.29 (1.32)	2.85 (1.60)
Emotional Inhibition	2.66 (1.10)	3.37 (1.15)	2.52 (1.09)
Defectiveness	2.56 (1.50)	2.94 (1.77)	2.57 (1.47)
Mistrust	2.51 (1.33)	2.89 (1.65)	2.45 (1.29)
Pessimism	3.30 (1.30)	3.09 (1.19)	3.45 (1.46)
Dependence	2.70 (1.18)	2.31 (1.24)	2.77 (1.08)
Failure to Achieve	3.50 (1.25)	3.26 (1.28)	3.55 (1.31)
Subjugation	2.73 (1.26)	2.57 (0.88)	2.98 (1.55)
Abandonment	2.65 (1.50)	2.23 (1.43)	2.60 (1.71)
Enmeshment	2.25 (1.45)	1.66 (0.70)	2.60 (1.71)
Vulnerability to Harm	2.83 (1.33)	2.66 (0.79)	2.88 (1.34)
Self-Sacrifice	4.11 (1.21)	4.31 (0.89)	4.31 (1.26)
Unrelenting Standards	3.66 (1.41)	4.09 (1.43)	3.48 (1.48)
Self-punitiveness	2.84 (1.42)	2.94 (1.29)	3.08 (1.64)
Entitlement	2.07 (0.84)	2.06 (0.50)	2.17 (1.05)
Approval Seeking	2.36 (0.97)	2.43 (0.88)	2.43 (1.21)
Insufficient Self-control	2.66 (1.14)	2.63 (1.25)	2.68 (1.25)

YPI-R			
Maternal Scores:	(N = 25)	(*n* = 7)	(*n* = 13)
Emotionally Depriving	2.72 (1.82)	2.86 (2.03)	2.69 (1.71)
Over-protective	3.11 (1.51)	2.79 (1.57)	3.47 (1.53)
Belittling	2.29 (1.70)	1.98 (1.06)	2.68 (2.06)
Perfectionistic	3.29 (1.35)	3.05 (1.41)	3.79 (1.25)
Pessimistic	2.75 (1.47)	2.25 (1.20)	3.29 (1.55)
Controlling	2.77 (1.79)	2.52 (1.40)	3.38 (2.01)
Emotionally Inhibited	3.04 (1.67)	3.67 (1.83)	2.90 (1.59)
Punitive	3.32 (1.66)	3.14 (1.75)	3.97 (1.57)
Conditional	2.55 (1.41)	2.82 (1.80)	2.90 (1.19)
Paternal Scores:	(*n* = 24) [a]	(*n* = 6)	(*n* = 13)
Emotionally Depriving	3.10 (1.95)	3.21 (1.91)	3.25 (1.99)
Over-protective	2.30 (1.01)	2.11 (1.11)	2.37 (1.08)
Belittling	2.24 (1.80)	2.29 (1.83)	2.42 (1.98)
Perfectionistic	3.43 (1.55)	3.94 (1.95)	3.54 (1.35)
Pessimistic	2.50 (1.42)	2.33 (1.11)	2.56 (1.43)
Controlling	2.08 (1.44)	2.28 (1.58)	2.23 (1.54)
Emotionally Inhibited	3.75 (1.73)	4.11 (2.07)	3.85 (1.53)
Punitive	3.21 (1.86)	3.56 (2.13)	3.28 (1.79)
Conditional	2.58 (1.52)	2.88 (1.76)	2.69 (1.26)

Note. Abbreviations: NES, non-epileptic seizures; FMS, functional motor symptoms; VAS, Visual Analogue Scale GAD-7, Generalised Anxiety Disorder 7 Questionnaire; PHQ-9, Patient Health Questionnaire 9; YSQ3-SF, Young Schema Questionnaire 3 – Short Form; YPI-R, Young Parenting Inventory – Revised. [a] One participant only gave scores for one parent.

Table 5 shows the proportion of participants falling into clinical categories indicated by the GAD-7, PHQ-9 and IPDE. Again, data for those with isolated types of functional neurological symptoms are also shown for comparison.

Table 5

Descriptive statistics for the sample regarding the clinical categories of various measures

Scale / Categorisation	Total (%)	NES only (%)	FMS only (%)
GAD-7	(N = 25)	(*n* = 7)	(*n* = 13)
None (0-5)	28%	0%	31%
Mild (6-10)	28%	57%	23%
Moderate (11-15)	20%	14%	23%
Severe (16-21)	24%	29%	23%
PHQ-9	(N = 25)	(*n* = 7)	(*n* = 13)
None (0-4)	28%	14%	31%
Mild (5-9)	8%	0%	15%
Moderate (10-14)	24%	43%	23%
Moderately Severe (15-19)	24%	14%	15%
Severe (20-27)	16%	29%	15%
IPDE (4-item threshold)	(N = 25)	(*n* = 7)	(*n* = 13)
Paranoid	32%	43%	38%
Schizoid	36%	43%	23%
Dissocial	0%	0%	0%
Impulsive	16%	43%	8%
Borderline	12%	14%	15%
Histrionic	20%	14%	23%
Anankastic	60%	86%	54%
Anxious	40%	57%	38%
Dependant	40%	43%	31%

Note. Abbreviations: NES, non-epileptic seizures; FMS, functional motor symptoms; GAD-7, Generalised Anxiety Disorder 7 Questionnaire; PHQ-9, Patient Health Questionnaire 9; IPDE, International Personality Disorder Examination Screening Questionnaire.

Hypotheses One and Two

To recapitulate, the first hypothesis was that scores for the Disconnection and Rejection and Excessive Responsibility domains would be negatively correlated with current self-reported health status (i.e. EQ Index Scores). The second hypothesis was that scores for emotionally depriving, belittling, perfectionistic and controlling parenting subscales on the YPI-R would also negatively correlate with current health status.

All variables were assessed for the presence of outliers through boxplots as described in Field (2009). None were found as no cases were found to lie further than 1.5 times the interquartile range from the upper and lower quartiles.

EQ Index scores and scores from the YPI-R were mostly found to be non-normally

distributed through graphical analysis and significance tests. In contrast, schema domain scores were assessed to be normally distributed. Appendix 11 reports the results of Shapiro-Wilk tests of normality. Because of these findings, non-parametric correlations were used to evaluate the first two hypotheses, as shown in Table 6. Kendall's correlation coefficient was calculated as it is thought to be more accurate than Spearman's in small samples (Field, 2009). Following the argument of Grayson (2004), transformations were avoided as it was thought the transformed EQ index score was likely to be unintelligible in relation to the original hypothesis, given that it is already a composite score produced by an algorithm.

Table 6

Correlation Coefficients and significance tests for hypotheses one and two

Variable	Kendall's Correlation Coefficient in comparison with EQ Index Scores τ	*p* value (one-tailed)
YSQ3-SF		
Disconnection & Rejection	-.196	.087
Excessive Responsibility	-.091	.264
YPI-R		
Maternal Scores:		
Emotionally Depriving	-.090	.275
Belittling	-.035	.406
Perfectionistic	.260	.039
Controlling	.065	.333
Paternal scores:		
Emotionally Depriving	-.149	.163
Belittling	-.053	.368
Perfectionistic	.142	.171
Controlling	-.058	.357

Note. Abbreviations: YSQ3-SF, Young Schema Questionnaire 3 – Short Form; YPI-R, Young Parenting Inventory – Revised.

To summarise Table 6 EQ Index Scores were negatively correlated with Disconnection and Rejection domain scores, although this correlation was not statistically significant (τ = -.196, *p* = .087).The EQ Index Scores were also negatively correlated with Excessive Responsibility domain scores, however again this correlation was not statistically significant (τ = -.091, *p* = .224). None of the correlations between predicted perceived parenting styles and EQ index

scores were statistically significant; correlation coefficients ranged between -.149 and .260.

Hypothesis Three

Figure 1 shows all schema domain scores for the sample in comparison with data from the two large Danish samples collected by Bach *et al*. (2018; see Plan for Statistical Analysis for further details).

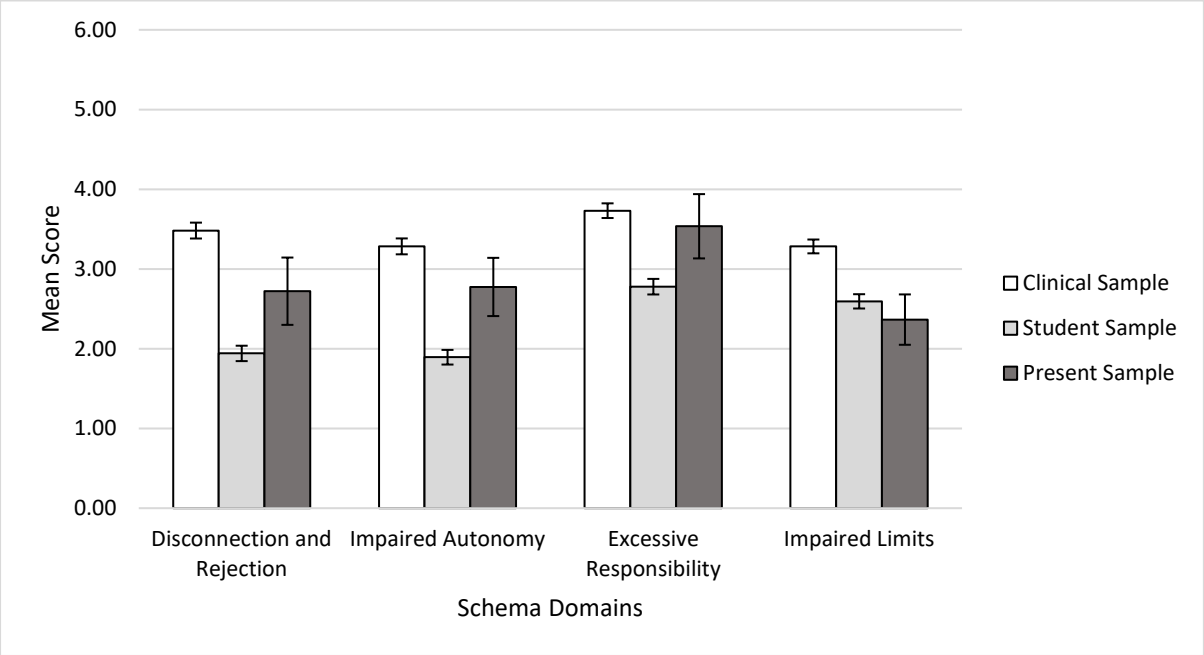

Figure 1. Graph showing schema domains scores in the present sample, and in Bach *et al.*'s (2018) two samples

Note. Error bars represent 95% confidence intervals.

It was found through statistical tests and graphical analysis that schema domain scores for the present sample were normally distributed (See Appendix 11 for further details). As the Bach *et al.* (2018) is drawn from two large samples, it was assumed that this data was normally distributed (Field, 2009).

Unpaired *t* tests between schema domain scores in the present samples and the two larger samples were conducted. These are reported in Table 7. Barlett's test for unequal variances was used as Levene's test for homogeneity of variance is impossible using summary statistics. Where Bartlett's test is statistically significant, Welch's unpaired *t* tests are reported. A Bonferroni correction for multiple corrections was applied to these *t* tests. Regarding comparisons with the student sample, *t* tests were one-tailed as it was hypothesised that the present sample's schemas scores would be elevated in comparison. Regarding comparisons with the clinical sample, *t* tests were two-tailed as there was no hypothesis as to whether the present sample's scores would be higher or lower in comparison.

Table 6
Significance Tests for Hypothesis Three

Sample Comparison	Bartlett's Test of Unequal Variances χ^2 (*df*)	*p* value	Unpaired *t* test *t (df)*	*p* value	Effect Size Cohen's *d*
Disconnection & Rejection					
Present vs Student	.55 (1)	.458	3.87 (414)	< .0001 [b d]	.76
Present vs Clinical	1.41 (1)	.236	-2.88 (681)	.004 [c d]	
Impaired Autonomy					
Present vs Student	.08 (1)	.078	4.16 (414)	< .0001 [b d]	.95
Present vs Clinical	4.20 (1)	.040	2.64 (27) [a]	.013 [c]	
Excessive Responsibility					
Present vs Student	.11 (1)	.736	3.75 (414)	< .0001 [b d]	.76
Present vs Clinical	.97 (1)	.324	-.78 (681)	.435 [c]	
Impaired Limits					
Present vs Student	.47 (1)	.495	-1.19 (414)	.117 [b]	
Present vs Clinical	4.36 (1)	.036	5.48 (27) [a]	< .0001 [c d]	.92

Note. [a] Welch's unpaired *t* test used due to significantly different variances [b] One-tailed [c] Two-tailed [d] Significant at $\alpha > 0.05$ with a Bonferroni correction applied, equivalent to $p < .006$

To summarise Table 7, regarding the Disconnection and Rejection schema domain, the present samples' scores were significantly elevated above those of the student sample, and were significantly lower than those of the clinical sample. In the Impaired Autonomy domain, the present sample's scores were significantly elevated above those of the student

sample, but not significantly different to those of the clinical sample. In the Excessive Responsibility domain, again the present sample's scores were significantly elevated above those of the student sample, and not significantly different to those of the clinical sample. Lastly, in the Impaired Limits domain, the present sample's scores were not significantly different to those of the student sample, and significantly below those of the clinical sample. All statistically significant effects were of medium to large size according to the thresholds given by Cohen (1988).

In order to investigate the reasons for these differences further, individual schema scores for the three samples were compared graphically. Figure 2 shows the individual schema scores for the present sample and the two samples collected by Bach *et al*. (2018).

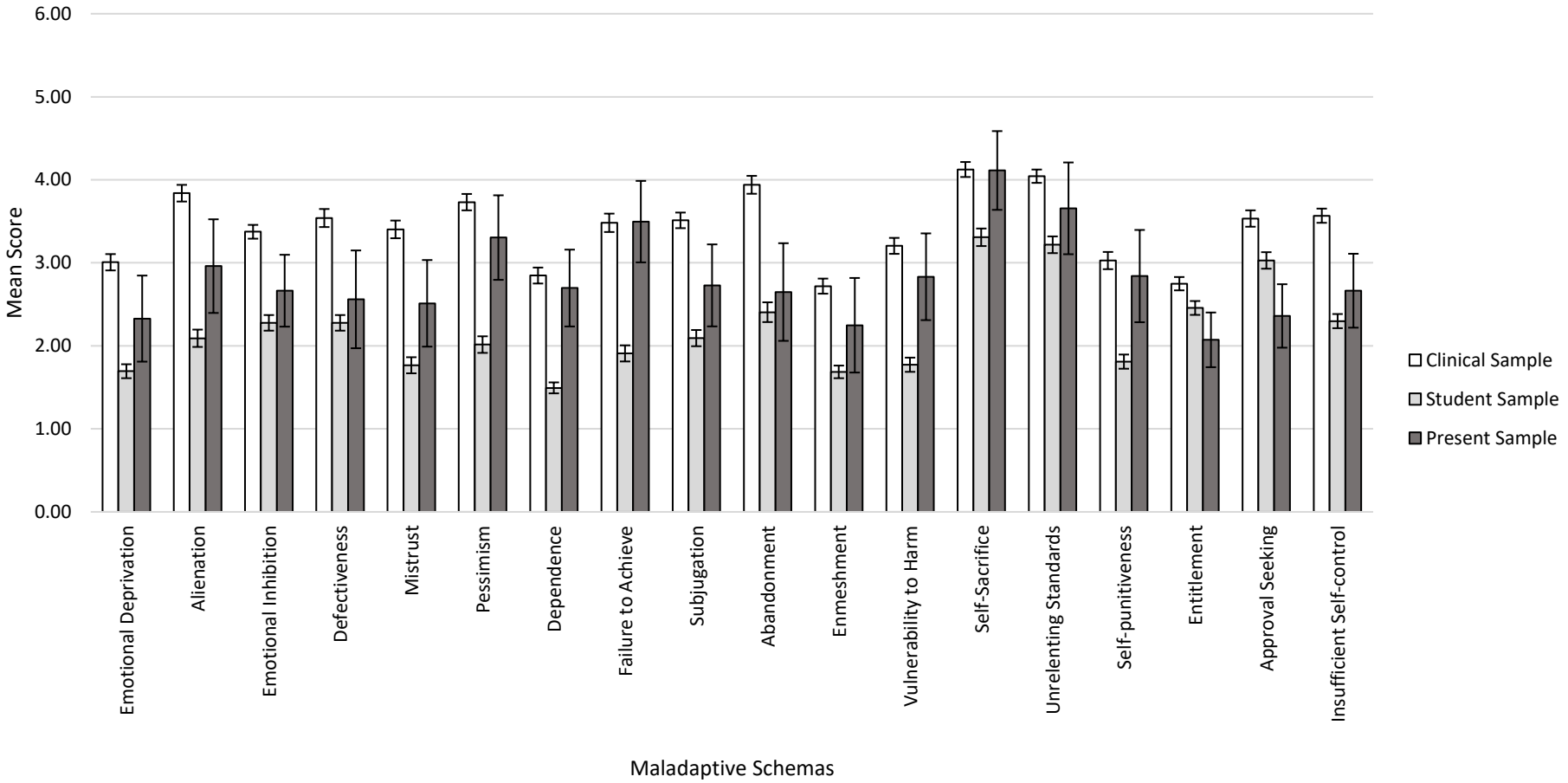

Figure 2. Graph showing individual schema scores in the present sample and in Bach *et al.*'s (2018) two samples
Note. Error bars represent 95% confidence intervals.

Visual inspection of the intersections of confidence intervals in Figure 2 suggests that the present samples' scores for individual schemas varied in their similarity to those of the other two samples. In the Disconnection and Rejection domain, most schema scores appeared to be between those of the student and clinical samples, or similar to those of the student sample. The Pessimism Schema scores were an exception, which appeared to be similar to those of the clinical sample. In the Impaired Autonomy domain, schema scores varied in whether they were similar to those of either sample. The Failure to Achieve and Vulnerability to Harm schemas in particular appeared to be similar to those of the clinical sample. In the Excessive Responsibility domain, the Self-Sacrifice and Self-Punitiveness schema scores of the present sample appeared to be similar to those of the clinical sample. In the Impaired Limits domain, all scores appeared to be either similar to or lower than the those of the student sample. The Entitlement and Approval-Seeking schema scores appeared to be lower than the student sample's scores.

Discussion

This study aimed to explore the role of maladaptive schemas in a clinical sample of participants with FND. Overall, the findings show that the participants' schema domain scores and perceived parenting style scores were not significantly correlated with self-reported scores for current health status. This means the first and second hypotheses were not confirmed. However, the results do show that hypothesised schema domain scores were elevated in the sample, in some cases to a level comparable with another clinical sample (Bach *et al.*, 2018). Further investigation of these results highlighted that these elevations appeared to be driven by scores for a number of individual schemas, which may provide some insight into the psychology of FND.

Sample Characteristics

The sample was predominantly (68%) female, which is consistent with existing studies using clinical samples of FND (see attached Systematic Review). It was also noteworthy that the proportion of participants who reported being in some form of employment prior to the development of FND (88%) appeared to have decreased substantially compared to the proportion who currently reported being employed (32%). Rates of self-reported disability also appeared to be high (72%). This is consistent with research demonstrating high levels of disability and unemployment in populations of FND (Carson *et al.*, 2011). Participants were drawn in fairly even proportions from inpatient (28%), outpatient (36%) and day-patient settings (36%). They were also broadly in even proportions at the beginning (28%), middle (32%) and end (40%) of their current form of treatment. There was a relatively high rate of refusal from potential participants (31%), which might relate to recruitment occurring in specialist services where patients are likely to be more unwell and have more complex

difficulties.

36% of the sample considered themselves to have a current mental health problem, and 52% disclosed that they had previously received some form of mental health treatment in the past. Using the recommended thresholds of the GAD-7, 44% of the samples' scores were indicative of moderate to severe anxiety. Using the four-item recommended thresholds for the PHQ-9, 64% of the samples' scores indicated moderate to severe depression. Inspection of the results of the IPDE appeared to indicate elevated levels of personality disorder. Using the four-item threshold for estimating personality disorder prevalence (discussed in the Measures section), the highest estimated rate was for Anankastic Personality Disorder, for which 60% of the sample scored above the threshold, and 86% in those with non-epileptic seizures. Anankastic personality disorder is characterised by "by pervasive perfectionism, conscientiousness, and insistent thoughts of a lesser severity than those in Obsessive–Compulsive Disorder" (Slade & Forrester, 2013, p. 209). Hence results appear to be consistent with studies which have linked FND to high rates of OCPD (Demartini *et al.,* 2014; Reuber *et al.*, 2004). In contrast to what was expected, the estimated rate of BPD was only 12%. Other personality disorder prevalence estimates also appeared relatively high, such as those for Dependant Personality disorder (40%). Overall these results appear to support the finding of the attached systematic review which found that patients with FND tend to have elevated mental health needs, though again this may have been influenced by the specialist services in which recruitment occurred. They also appear to again highlight the issue that FND groups may exhibit variability in their precise psychological characteristics.

Hypothesis One

It was predicted that participants' self-reported health status (EQ Index Scores) would be negatively correlated with schema scores for the Disconnection and Rejection and Excessive Responsibility domains. This was based on existing findings which show higher rates of certain personality disorders in FND groups. Though negative correlations were found between these variables, they were not statistically significant, so this hypothesis was not confirmed.

There are a number of reasons which could explain this result. The study recruited slightly less than its power calculation indicated, meaning that the study is likely to be underpowered to detect even large effect sizes. As such, these non-significant effects may indicate the absence of a relationship between schema scores and current self-reported health, or they may indicate that the strength of the relationship is smaller than the study was powered to detect.

Another factor which may have influenced these findings is that the participants varied a great deal in the amount of treatment that they had already received for their FND, meaning that they may not have been a very homogenous sample. In order to maximise recruitment, participants were drawn from inpatient, outpatient and day-patient services, where they were receiving a mixture of medical, psychological, physiotherapeutic and other interventions. The participants' stage of treatment also varied considerably. The duration and nature of previous interventions that participants had already received for their FND was not recorded. In feedback, some participants at later stages of treatment expressed the view that they were aware of the impact on their life of certain issues raised by the schema

questionnaires, but felt that through treatment they now felt more able to cope with them. This may mean that the study's sensitivity to detect a relationship between current health in FND and maladaptive schemas was confounded by variation in participants' levels of previous treatment, which might mitigate the impact of schemas on health. Because of this, it might be that if the study was repeated with participants at a controlled early stage of treatment, a stronger effect would be detected. One means to control for this might have been to re-analyse the hypotheses within subgroups of the sample only at a set stage of treatment. However, this was not thought to be feasible given the size of these subgroups.

Another confounding factor could have been whether a self-report questionnaire of general health status was in fact the most appropriate measure for the study. As the EQ-5D-5L is a generalised measure of mental and physical health, it does not measure a participant's health solely in regard to the impact of functional neurological symptoms. A myriad of other factors on the health of participants might also affect these scores, such as unrelated physical health conditions. This reduced the sensitivity of the measure to detect the hypothesised effects.

One corrective design feature for this problem might have been to gather data on participants' current physical health conditions. However, the need for this was overlooked when the study was planned. Another related solution might have been to exclude participants with any physical health problem which might produce similar effects to a functional neurological problem. Although the study did exclude any participant with a diagnosis of epilepsy, it did not exclude participants with other comorbid physical health conditions, such as those causing movement difficulties. This is common practice in many

studies examining participants with functional motor symptoms (e.g. Demartini *et al.*, 2016; Ekanayake *et al.*, 2017). However, this may have compounded difficulties recruiting adequate numbers of participants.

Thinking more broadly about the measure of health used in this study, the EQ-5D-5L was selected as no specific measure for the impact or severity of functional neurological symptoms could be found. The Somatoform Dissociation Questionnaire (Nijenhuis, Spinhoven, Van Dyck, Van Der Hart & Vanderlinden, 1996) might come close to this, but the construct it measures is not entirely the same. The lack of a validated, specific measure for the magnitude of all functional neurological symptoms presents an obstacle to future research. However, a clinician-rated measure known as the Functional Neurological Disorders Rating Scale (unpublished) is currently in development, coincidentally at South London and Maudsley NHS Foundation Trust. A measure of this kind has the benefit of allowing for more precise measurement of the level of functional symptoms within a sample. If this study were repeated, this might provide the most effective solution to the problems identified above.

Hypothesis Two

It was also hypothesised that participants' self-reported health (EQ Index Scores) would negatively correlate with perceived parenting styles experienced during childhood as measured by the YPI-R. These parenting styles – emotionally depriving, belittling, perfectionistic and controlling – were selected based on previous research by Bach *et al.* (2018), which found they were associated with the schema domains identified in Hypothesis One. Correlation coefficients between the perceived parenting styles and self-reported

health scores ranged between -.15 and .26, none of which were statistically significant. As such, the findings do not support this hypothesis.

The issues of lack of power, possible heterogeneity in the sample, and potential confounds in the measurement of health status that are discussed above are also potential explanations for this finding. A study discussed above by Sheffield *et al*. (2009), which examined correlations between parenting subscales of the YPI-R and current symptoms in a group of patients with eating disorders, tended to find medium effect sizes between these variables. This highlights the fact that again, the study may well have been underpowered to detect the hypothesized effects.

Some service-user feedback made similar points. Participants raised the issue that, although they could see the relevance of asking about perceived parenting, the study could have measured other factors which might have had an influence on the development of maladaptive schemas or FND. For instance, some mentioned the fact that they felt their functional neurological symptoms had been particularly influenced by difficulties within their family in adulthood. It may be that perceived parenting in childhood has a fairly distal influence, with only a small effect on current difficulties in FND, hence the study lacking to detect this effect.

The strongest associations found seemed to be between emotionally depriving parenting and current health status, with correlations ranging between -.15 and -.09. This is consistent with research by Bach *et al.* (2018), who found this to be one of the perceived parenting styles most strongly correlated with Disconnection and Rejection schema domain scores.

Unexpectedly, scores for participants' experience of perfectionistic parenting were positively correlated with scores for current health status, on both paternal and maternal subscales. These correlations ranged between .26 and .14, meaning that the greater participants' endorsement of experience of perfectionistic parenting during childhood, the higher they rated their health status on the day of participation. This maternal effect was strong enough that it would have provided a statistically significant *p* value had the hypothesis been in the opposite direction. It is difficult to find an obvious explanation for this finding. The items for this subscale include [the parent] "had high expectations for him/herself" and "expected me to do my best at all times". One possible explanation could be that it the experience of perfectionistic parenting in childhood is linked to having stronger beliefs around positive self-presentation, and a need to appear to be coping well with difficulties.

Hypothesis Three

Lastly, it was hypothesised that scores for the two schema domains would appear elevated in comparison to normative data. The findings indicated that scores for the Disconnection and Rejection domain were significantly above those of a student sample, but not as high as those of a clinical sample. In the Excessive Responsibility domain, scores were found to be above the level of the student sample and comparable with the clinical sample. Hence, these findings confirmed the hypothesis. In the Impaired Autonomy domain scores were also found to be above those of the student sample and comparable to those of the clinical sample. In contrast, in the Impaired Limits domain, scores were found to be below those of the clinical sample, and comparable to those of the student sample.

Visual analysis of the individual schema scores indicated which scores were driving these findings. A number of individual schema scores in the Disconnection and Rejection, Impaired Autonomy, and Excessive Responsibility domains appeared to be elevated to the level of the clinical sample. Scores for Pessimism, Failure to Achieve, Vulnerability to Harm, Self-Sacrifice and Self-Punitiveness schemas were the most elevated. Some scores in the Impaired Limits domain appeared lower than those of the student sample, namely those for Entitlement and Approval-Seeking schemas.

These findings provide a richer characterisation of the schema profile of the sample. To aid explanation, Table 8 provides definitions of these key maladaptive schemas.

Table 8
Descriptions of Selected Maladaptive Schemas

Schema	Definition
Pessimism	"a pervasive, lifelong focus on the negative aspects of life" combined with an exaggerated expectation that things will eventually go seriously wrong" (Young *et al.*, 2003, p. 17)
Failure to Achieve	"the belief that one has failed, will inevitably fail or is fundamentally inadequate relative to one's peers in areas of achievement" (Young *et al.*, 2003, p. 15)
Vulnerability to Harm	"exaggerated fear that imminent catastrophe will strike at any time and that one will be unable to prevent it" (Young *et al.*, 2003, p. 15)
Self-Sacrifice	"excessive focus on voluntarily meeting the needs of others in daily situations at the expense of one's own gratification" (Young *et al.*, 2003, p. 16)
Self-Punitiveness	a deep-rooted belief that one should be punished for making mistakes and a "difficulty in forgiving oneself" (Young *et al.*, 2003, p. 17)
Entitlement	"The belief that one is superior to other people; entitled to special rights and privileges; or not bound by the reciprocity that guides normal social interaction" (Young *et al.*, 2003, p. 15)
Approval-Seeking	Excessive emphasis on gaining approval, recognition, or attention from other people, or on fitting in at the expense of developing a secure and true sense of self." (Young *et al.*, 2003, p. 16)

Taken as a whole, the scores for these schema subscales seem to suggest a fairly coherent profile of maladaptive traits. The elevated schemas suggest a theme of anticipating negative consequences; an exaggerated, unrealistic drive towards achievement and adhering to internalised rules; and a perceived need to sacrifice one's own needs in favour of other people. Inflexibility around internalised standards also seems to be a common theme, for instance in the endorsement of self-sacrifice and self-punitiveness. The results regarding the two schemas that appeared to have lower scores than the student sample are both associated by schema theory with narcissism (Young *et al.*, 2003), and this is coherent with these themes. A diminished sense of entitlement agrees with a deep-rooted belief in the importance of self-sacrifice and a tendency to believe that one is a failure. The diminished

need to seek approval from others again suggests that a drive towards achievement and unrealistic standards is motivated by strict internal standards rather than the approval of others. It could be that these maladaptive schema patterns are factors in the development of functional neurological symptoms. As has been noted previously, prior studies in other conditions have found that scores for maladaptive schemas are significantly associated with greater psychological symptoms (e.g. Muris, 2006).

These findings also agree with the data from the IPDE, in that they appear to be consistent with an anankastic profile or that of OCPD, which are both characterised by perfectionism, inflexibility, and an over-controlled approach to emotional and interpersonal situations. The fact that schema scores for the Disconnection and Rejection domain were not as elevated as might have been expected may be explained by the fact that the present sample did not appear to have a high prevalence of BPD according to the IPDE scores.

Limitations

The conclusions of this study are limited by a number of factors. As has been discussed, it is likely to be underpowered to detect even large effect sizes in the main hypotheses. This may have raised the risk of type II errors regarding non-significant findings. Another limitation is that the first two hypotheses were tested using correlations, meaning causality could not have been inferred even from statistically significant findings. It is also important to acknowledge that this sample was recruited from national specialist services within the NHS, meaning that the participants are likely to have more complex difficulties and be more unwell than those seen elsewhere. As a result, it is unclear the extent to which these findings may generalise to wider functional neurological populations.

The analysis of the third hypothesis used large-scale data from another study, providing some of the benefits of using a control group. It was possible to compare the scores of the sample with those of student and clinical samples. However, this may have been problematic for a number of reasons. For instance, the use of student samples in psychological studies as a proxy for the performance of the general population has not always been supported by research (Hanel & Vione, 2016). Secondly, the exact nature of Bach *et al.*'s (2018) clinical sample was poorly characterised. It appears to have been a fairly heterogeneous sample of patients with substance misuse difficulties and personality disorder traits. As a result, the precise meaning of scores being found to be similar in this sample and the present sample is unclear.

As has been touched upon, another limitation is the possible inadequacy of some of the measures used in the study. In the first place, the study is reliant on self-report questionnaires that have not been validated in the target population. Evidence suggests that some participants with FND may struggle to regulate and process emotions, and this may distort their answers on self-report measures. This may present a confound in the design and mean that the measures used may not accurately track hypothesised constructs like maladaptive schemas.

A more specific problem with the self-report measures used, concerns the EQ-5D-5L. This questionnaire may have had a confounding influence on the findings, as scores its scores can easily be influenced by factors other than FND, such as physical health conditions. In addition, given previous research in this area, it may have been informative to include additional measures in the design. As discussed in the attached systematic review, there

appears to be some variability in the characteristics of samples with FND, so improved measurement of other factors relevant to the condition might have been important, such as the prevalence of traumatic experiences or stressful life events.

Lastly, another limitation was the absence of service-user involvement in the project. As discussed, it was intended that service-user involvement would be used to inform the interpretation of the study. Due to time constraints, this was not possible. Feedback from participants was drawn upon to gain an element of service-user perspective. However, had it been possible to gain formal input from a service-user group, this might have improved upon the quality of the interpretation of the findings. This is discussed in greater depth in the Integration, Impact and Dissemination section of this thesis.

Further Research

Further research would clearly be needed to demonstrate evidence that maladaptive schemas play a role in FND. The most obvious area for a new study to pursue would be a replication of a study like this one with a large enough sample to detect the hypothesised effects. Given the findings of the attached systematic review, which highlights psychological differences in types of FND, another area of future research might be to compare the schema profiles of groups with different types of functional neurological symptoms, in order to see if there are different schema patterns present within different types of FND.

Future research would also benefit from using measures which assess more precisely the complexity or severity of functional presentations, such as the Functional Neurological Disorders Rating Scale (unpublished). This would allow for more carefully controlled studies

in this area. It might also contribute to better understanding of the reasons for variability of psychological findings between studies of people with FND.

Integration, Impact and Dissemination

Integration

The overall purpose of this thesis was to explore the potential role of maladaptive schemas in understanding functional neurological disorders (FND). It argues that the findings of existing studies, such as those concerning the prevalence of adverse experiences, personality disorder and maladaptive coping strategies suggest that the theoretical models of schema therapy ('the schema model'; Young, Klosko & Weishaar, 2003) might be helpful in understanding functional neurological presentations. The Empirical Project is thought to be one of the first of its kind to explore the schema model within a clinical sample of people with FND.

The Systematic Review included in this thesis analyses studies which have compared clinical samples with two types of FND – non-epileptic seizures and functional motor symptoms – based on their psychological characteristics. These studies indicate that there appear to be a number of similarities between the two. The review found that both groups tended to be female and show elevated levels of depression, anxiety, psychiatric symptoms and alexithymia. However, it also highlighted differences between the two: patients with non-epileptic seizures tended to be younger and were more likely to report traumatic experiences and stressful life events. There was also some evidence for differences in personality and tendencies toward dissociative experience.

The Systematic Review can be summarised as showing that while there are common features in both of these functional neurological presentations, there also appear to be some psychological characteristics in which they differ. The review also highlights that

adverse experiences and maladaptive coping strategies appear to be associated with FND. It is argued that this combination of themes – adverse experiences and later maladaptive coping strategies, but variability in their precise nature – suggests that the schema model may be an appropriate explanatory framework for understanding the psychology of FND.

In combination with other literature, the results of the review informed the interpretation of the Empirical Project. The primary hypothesis concerned negative associations between maladaptive schemas and self-reported health in a clinical sample of participants with FND. The study found that, although there were inverse associations between scores for certain clusters of schema domain scores, these were not statistically significant. This may have been due to the small sample size, which meant that the study was somewhat underpowered. A second hypothesis regarding recalled experience of certain unhelpful parenting styles based on the schema model was also not confirmed. However, the study did find that various schema scores appeared to be significantly elevated, in some cases to clinical levels, in comparison to data from another larger study (Bach, Lockwood & Young, 2018).

Overall, the profile of schemas in the sample suggested themes of inflexibility, strongly internalised rules, anticipation of negative consequences, an exaggerated drive toward achievement, and a perceived need to sacrifice one's own needs in favour of those of other people. This was consistent with data from other measures used in the study which indicated elevated rates of Anankastic Personality Disorder.

Further research is warranted to further explore the role of maladaptive schemas in FND.

For firmer conclusions to be drawn, the results of this study would require replication in a larger, better controlled sample. Given the results of the systematic review, another avenue of future research would be to explore the maladaptive schema profiles of participants with different types of FND.

Challenges

In order to better characterise the relationship between the Systematic Review and the Empirical Project, this section outlines a number of challenges that were faced in carrying both of them out. This also section also helps to contextualise the process of carrying out the project as a whole.

Challenges in Carrying out the Systematic Review

The main challenge in carrying out the systematic review was selecting a suitable aim and scope for the review. Originally, it was intended that the review would examine a broad range of evidence concerning predisposing psychological factors in FND, in order to better understand the evidence which might support an argument for the empirical study. However, this plan was ultimately rejected, for two reasons. Firstly, it was decided that this would involve reviewing too large a number of studies than was feasible for the project. Secondly, it was felt that this was too similar to existing reviews which had recently been published (e.g. Brown & Reuber, 2016; Ludwig et al., 2018).

Instead, it was decided to focus the review on a more specific issue: studies of psychological characteristics comparing groups of participants with different types of FND. Though this topic might seem only indirectly related to the empirical project, it proved helpful in

explored issues in the argument for the hypothesis that schemas might be associated with FND. Moreover, the review process help me to become familiar with this area of research literature, through exposure to a large number of studies on FND, even if most of these were ultimately excluded from the review.

The systematic review topic was also selected because it had been highlighted for further exploration by an already published systematic review (Brown & Reuber, 2016). Furthermore, my experiences with clinicians working in the field had informed me that they were often of the view that there were certain psychological differences between types of functional neurological presentations, in a way that resembled the predictions of Brown and Reuber (2016). However, it appeared that these clinical opinions were not grounded in a systematic analysis of the available evidence.

Gaining Ethical Approval

There were a number of challenges in conducting the empirical project, the first of which was gaining ethical approval. In order to conduct the empirical project, it was necessary to gain approval from five different organisations, including the Health Research Authority, an NHS Research Ethics Committee, and the research and information governance departments of the recruitment sites. In discussions with these bodies, a number of important issues had to be carefully considered. For instance, it was recognised that schema-orientated measures ask about personal and potentially distressing content (such as experience of emotionally abusive parenting), so the way in which information was presented to potential participants had to be designed to support informed consent and mitigate the risk of distress. Confidentiality also had to be carefully considered, and the

extent to which personal information that might be disclosed by participants would be passed on to health professionals connected with their case. Specialised risks in the population also had to be considered, such as what procedures would be followed if a participant had a non-epileptic seizure while they were with the principal researcher.

Developing a comprehensive plan concerning all ethical issues surrounding the study, communicating this to the relevant organisations, and receiving their approval took up a large amount of the time involved in conducting the empirical project. It is estimated that the time taken to gain ethical approval from organisations not including Royal Holloway University of London departments was around 10 months. Though this process produced an ethically robust plan for conducting the study, it was challenging to come up with solutions to complex ethical issues, and to negotiate the relevant regulatory frameworks. The time taken to do this also created further challenges, limiting the time that could be spent recruiting participants to the study.

Recruitment

The total time spent on site recruiting participants was 29 days. Due to the time pressures around recruitment, these were compressed into a narrow window of time. Because of this, it was a challenge to recruit an adequate number of participants to the study. The power calculation for the empirical project suggested that a minimum of 28 participants was needed to ensure an acceptable level of power in the main hypotheses. Only 25 participants were actually recruited in the time available.

One factor in this may have been the relatively high rates of the potential participants

declining to participate; 31% of those approached are thought to have declined. This may relate to the content of schema-orientated measures, which as discussed above includes content which some people might have may find distressing. The fact that the power calculation was not met may have led to a small reduction in the ability of the study to detect statistically significant effects. However, given time pressures, the number recruited was thought to be a relatively good outcome.

On reflection, a number of strategies assisted me in maximising the number of participants it was possible to recruit in the short period of time available. Drawing on multi-disciplinary team working skills developed through clinical placements, I built productive relationships with a range of relevant staff across a number of different teams. Attending service meetings, seeking information about sources of appropriate patients within services, and proactively approaching relevant teams and professionals all proved useful strategies for enhancing the potential for recruitment. Later on in the recruitment window, focusing attention on the services which had demonstrated themselves to be more reliable sources of participants was also an important strategy.

Service User Involvement

Time pressures also limited the level of service-user involvement in the project. It had been intended that the results of the empirical project would be presented to a service-user group. It was hoped that their feedback would be used to inform the interpretation of the empirical project. Though requests were made to meet with a number of service-user groups, none were available in the time window between the end of recruitment and the deadline for submission. As a result, this aspect of the project had to be abandoned.

However, as part of the study procedure, feedback on the project separate to data collection was sought from participants, and details of these comments were recorded in note form as the project continued. Participants gave insightful comments on a range of topics, including their thoughts on the rationale for the study, the content and structure of the questionnaires used, and additional variables they thought it might have been helpful to have measured. As discussed in the report of the empirical project, these comments did inform interpretation of the results of the study. This process provided a degree of service-user perspective in the empirical project, which mitigated the loss of the planned level of involvement.

Reflections

This project was devised from an original idea by the author, in collaboration with internal and external supervisors. This in itself also added to the initial time needed to develop the project before recruitment could begin. However, in retrospect, the fact that the project developed from my own ideas about the literature on FND meant that I learnt more about the process of developing an empirical research project than I might have done from conducting a project which had been conceived by a supervisor. Similarly, although the fact that the project used a clinical sample led to challenges that I might not have encountered otherwise, I felt that I developed skills needed to conduct research in the NHS through adapting to these challenges. The fact that the study used a clinical sample also meant that I spent a large amount of time working with people with FND and experiencing NHS services designed to care for them. From this I learnt great deal about the realities of what it is like to live with functional neurological difficulties. This was a highly valuable experience to me,

both as a future researcher and as a future clinician.

Impact

FND have the potential to cause significant distress and disability to those they affect (Carson et al., 2012), as well as significant costs to health services. In 2009, it was estimated that medically unexplained illnesses like FND cost the NHS around £18 billion a year, slightly more than the yearly cost of dementia at all ages (Bermingham, Hague, Cohen & Parsonage, 2010). As such, improving knowledge about FND and its effective treatment is a priority. This thesis has the potential to contribute to better understanding of and improved interventions for functional neurological symptoms. However, it is important to reiterate that the findings presented here are limited by various factors and are contingent on future research. The sections below discuss potential aspects of the future impact of this research.

Therapeutic Interventions for FND

Cognitive-behavioural therapy (CBT) is the most likely psychological intervention patients with FND are likely to be offered in the NHS (Stone, 2016). As discussed in the introduction to the Empirical Study, schema therapy and CBT share common ground, in particular the concept of a maladaptive schema. While schema therapy incorporates aspects of other approaches, it retains many of the aspects of CBT, such as a structured approach to therapy, the use of psychometric measures, and the application of behaviour change strategies tailored to a psychological formulation (Young *et al.*, 2003). This means that if further research substantiated the idea that maladaptive schemas were important factors in the development or maintenance of FND, schema therapy approaches or techniques could be integrated into cognitive behavioural interventions for FND with relative ease (e.g. Padesky,

1994; Waller, Kennerly & Ohanian, 2007). This might contribute to improving the outcomes of therapeutic interventions for FND.

The findings might also have implications for how best to structure the care of those with FND. Schema therapy emphasises the importance of the therapeutic relationship more so than traditional CBT, as it suggests the presence of maladaptive schemas may impair a person's ability to engage in productive helping relationships. It also suggests that longer psychological interventions tend to be needed, in order to build up an effective therapeutic relationship and challenge deep-rooted beliefs. Other kinds of healthcare for FND, such as physiotherapy or neurology consultations, could benefit from incorporating some aspects of these ideas into their practice.

Psychological Complexity in FND Patients

Both the findings of the Systematic Review and the Empirical Study highlight the potential for patients with FND to be clinically complex and present with a range of mental health needs. The findings of the review also suggest that patients with non-epileptic seizures may have the potential to be more clinically complex than other FND patients, which again may be important information for clinicians working in the field to be aware of.

The potential clinical complexity of functional neurological presentations also suggests a need to think carefully about the way services which aim to meet their needs are structured. For instance, a multi-disciplinary approach to this patient-group may be needed to ensure that a patient's needs are considered from a range of perspectives (Demartini *et al.*, 2014a). This also suggests that indirect psychological interventions may be of use to other

professionals working with patients with FND, such as physiotherapists or neurologists. Having the opportunity to consult with psychologists may help other professionals to identify and address mental health needs in this population and understand unhelpful relationship dynamics which may arise in their work with patients. Reflective practice interventions may also help professionals to process the demands of working in this area and develop solutions to issues related to clinical complexity.

Dissemination

It is intended that the Systematic Review and Empirical Project will be disseminated in two separate publications. In order to improve the likelihood of publication for the Empirical Project, an application for an extension to the ethical approval is currently being considered. The intention would be to recruit further participants so that the sample exceeds the power calculation. Similarly, it may be necessary to integrate a second reviewer into a revised form of the Systematic Review for it to reach a publishable standard.

The Empirical Project was of interest to members of staff in the three services from which participants were recruited. All of these teams independently requested feedback on the results of the study. As a result, it is intended that over the summer presentations will be arranged with these local sites to communicate the results of the project. It is also being investigated whether there are upcoming relevant conferences at which the Empirical Project could be presented.

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Appendices

Appendix 1: Systematic Review Search Strategy

A title and abstract search of using the following logic:

“Psychological factor*” OR “Psychosocial factor*” OR “psycho-social factor*” OR “predisposing factor*” OR “risk factor*” OR “history of” OR personality OR alexithymia OR “core belief*” OR attribution* OR neuroticism OR perfectionism OR schema OR schemas OR coping OR “psychological adjustment” OR “emotional adjustment”

And

"functional neurological" or fnd OR "functional movement" or “functional motor” or “dissociative movement disorder” or “functional paralysis” OR “functional dystonia*” OR “functional tremor*” OR “functional weakness” OR “functional limb weakness” OR “functional myoclonus” or “gait disorder*” OR “functional voice” or “functional dysphonia*” or "non-epileptic" or nonepileptic OR "psychogenic seizure*" OR pseudoseizure* OR “pseudo-seizure*” OR “pseudo-epilepsy” OR pseudoepilepsy OR “functional seizure*” OR “hysterical seizure*” or “conversion seizure*” or “dissociative seizure*” or “dissociative convulsion*” or “hysterical convulsion*” OR hysteroepilepsy or “conversion disorder*” or “functional stroke*”

Inclusion Criteria

- Published empirical quantitative research reports studies which compare groups of participants with types of functional neurological disorders (e.g. non-epileptic seizures, functional motor symptoms) according to their psychological characteristics.
- English language studies published in peer-reviewed journals.
- Participants are older than 18 years.

Exclusion Criteria

- Qualitative studies
- Studies which only examine the role of simple demographic factors (e.g. age, gender)
- Do not use appropriate, replicable, quantitative measures to assess psychological characteristics
- Case reports or small n designs (n < 15)
- Interventions studies

- Expert opinion or guidance, commentaries, journalistic writing
- Literature or systematic reviews
- Republications of existing data

Databases

- PsycInfo
- PubMed

Appendix 2: Letter of Ethical Approval from NHS Research Ethics Committee

(This has been included a separate document)

Appendix 3: Letter of Ethical Approval from the Health Research Authority

(This has been included a separate document)

Appendix 4: Demographic Questionnaire

Participant Demographics Form

Participant Number	
What is your age?	
What is your ethnic group?	☐ White ☐ Asian / British Asian ☐ Arabic ☐ Black / African / Caribbean / Black British ☐ Multiple or mixed ethnic origins ☐ Prefer not to say ☐ I prefer to describe my ethnic origin as: .....
What is your sex?	☐ Male ☐ Female ☐ Intersex ☐ Transgender ☐ Prefer not to say ☐ I prefer another description: .....
What is your relationship status?	☐ Single ☐ In a relationship ☐ Married/Civil Partnership ☐ Divorced ☐ Widow/Widower ☐ Prefer not to say
What is your sexual orientation?	☐ Bisexual ☐ Gay / lesbian ☐ Heterosexual ☐ Prefer not to say ☐ I prefer another description: .....
Do you have children?	☐ Yes ☐ No ☐ Prefer not to say
If applicable, are you pregnant?	☐ Yes ☐ No ☐ Prefer not to say
Do you consider yourself to have a disability?	☐ Yes ☐ No ☐ Prefer not to say

What is your current employment status?	☐ Employed ☐ Self-employed ☐ Unemployed ☐ Retired ☐ Prefer not to say ☐ I prefer another description: .....
What was your employment status before you became unwell?	☐ Employed ☐ Self-employed ☐ Unemployed ☐ Retired ☐ Prefer not to say ☐ I prefer another description: .....
Apart from functional neurological symptoms, would you consider yourself to have any other kind of mental health difficulty?	☐ Yes ☐ No ☐ Prefer not to say
Have you even been treated for any other kind of mental health problems in the past?	☐ Yes ☐ No ☐ Prefer not to say

For Office Use FNS:

Appendix 5: The EQ-5D-5L

(Due to copyright restrictions, this is included as a separate document)

Appendix 6: The International Personality Disorder Examination Screening Questionnaire

(Due to copyright restrictions, this is included as a separate document)

Appendix 7: The Young Schema Questionnaire 3 – Short Form

(Due to copyright restrictions, this is included as a separate document)

Appendix 8: The Young Parenting Inventory - Revised

YPI-R

Name	Date

Instructions

Listed below are statements that you might use to describe your parents. Please read each statement and decide how well it describes your parents. Choose the **highest rating from 1 to 6** that describes your mother, then your father, **when you were a child or young teenager** and write the numbers in the boxes before each statement. If someone substituted as your mother or father, please rate the scale for that person. If you did not know, or have any contact with, your mother or a father when you were a child, leave the appropriate column blank.

Rating Scale

1 Completely untrue	3 Slightly more true than untrue	5 Mostly true
2 Mostly untrue	4 Moderately true	6 Describes him/her perfectly

	MOTHER	FATHER	When I was growing up as a child and young teenager...
1			Loved me, treated me as someone special.
2			Spent time with and paid attention to me.
3			Gave me helpful guidance and direction.
4			Listened to me, understood me, shared feelings with me.
5			Was warm and physically affectionate.
6			Worried excessively that I would get hurt.
7			Worried excessively that I would get sick.
8			Was a fearful or phobic person.
9			Overprotected me.
10			Made me feel I couldn't rely on my decisions or judgment.
11			Did too many things for me instead of letting me do things on my own.
12			Treated me as if I were younger than I really was.
13			Made me feel unloved or rejected.

	MOTHER	FATHER	When I was growing up as a child and young teenager...
14			Treated me as if there was something wrong with me.
15			Made me feel ashamed of myself in important respects.
16			Treated me as if I was stupid or untalented.
17			Didn't really want me to succeed.
18			Expected me to be a failure in life.
19			Treated me as if my opinions or desires didn't count.
20			Controlled my life so that I had little freedom of choice.
21			Had very high expectations for him/herself.
22			Expected me to do my best at all times.
23			Was a perfectionist in many areas; things had to be "just so".
24			I felt that I didn't have enough individuality or sense of self separate from him/her.
25			I felt that I didn't have my own sense of direction while I was growing up because he/she was such a strong person.
26			Worried a lot about the family's financial problems.
27			Had a pessimistic outlook; often expected the worst outcome.

	MOTHER	FATHER	When I was growing up as a child and young teenager...
28			Focused on the negative aspects of life or things going wrong.
29			Was uncomfortable expressing affection or vulnerability.
30			Was private; rarely discussed his/her feelings.
31			Would become angry or harshly critical when I did something wrong.
32			Would punish me when I did something wrong.
33			Would call me names (like "stupid" or "idiot") when I made mistakes.
34			Was concerned with social status and appearance.
35			Placed strong emphasis on success and competition.
36			Was concerned with how my behaviour would reflect on him/her in the eyes of others.
37			Seemed to love me more or pay more attention to me when I excelled.

Appendix 9: Empirical Study Participant Information Sheet

Participant Information Sheet

Title of Project: Is there a relationship between early maladaptive schemas and functional neurological symptoms?

You are invited to take part in a research study looking at the possible role of schemas in functional neurological symptoms (FNS).

Schemas are thought to be mental templates or 'blueprints' for how we think, feel and relate to others. They are formed as we grow up and affect how we see the world. It is not clear whether schemas are important or not in FNS, so this study is trying to gather more information about this. FNS may come about because of range of different causes in combination, so schemas may not be important in your case.

Before you decide to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to consider the following information. Discuss it with other people if you wish. Ask the lead researcher (Toby Newson) if anything is not clear or if you would like more information.

Your decision to take part will not affect your medical care.

What is the purpose of the study?
The study aims to gather more evidence about schemas in people with FNS. This is because some evidence suggests that schemas might play a role in the development of FNS.

Why have I been invited?
You have been invited because your care team at the Lishman Unit are aware that you have FNS.

Do I have to take part?
No. It is completely your choice whether you decide to take part. Your decision will not affect your medical care. Even if you agree to take part, you are free to leave the study at any time without giving a reason.

What will happen if I agree to take part?
You will be asked to complete a set of questionnaires. These will be on paper in a private space at the hospital. It should take about 45 minutes. A researcher will be with you while you do this, but they will not ask you the questions directly. There will be no further sessions after this.

The questionnaires will ask you about your thoughts, emotions and relationships. Many of the questions will ask if you have had negative thoughts or past negative experiences. Some of the questions will be about memories of your parents or caregivers during childhood.

None of the questions will ask for long answers. You will not need to give detailed information about your experiences. For nearly all questions you will only need to answer by ticking a box or writing a number.

We will ask your permission for your care team to tell us your diagnoses relating to FNS. We will also ask your permission for your care team to share your scores on three short questionnaires that you may have filled out for them already. These questionnaires were about FNS and dissociative states. Dissociative states are those times when we feel separated from a part of ourselves. Your care team

will not share any other information without your permission. We will not ask for any other information from your medical records.

What are the possible advantages of taking part?
We cannot promise the study will help you personally. However, by taking part you might contribute to better understanding and treatment of FNS in the future.

What are the possible disadvantages of taking part?
The questionnaires will ask you about sensitive personal information. They may remind you of upsetting experiences. You can discuss any concerns you have with the researcher if you feel negatively affected by filling out the questionnaires. Your care team can provide you with emotional support afterwards if you need it.

What if something goes wrong?
If you have a concern about any aspect of this study, you can speak to the lead researcher who will do their best to answer your questions. You can do this in person, or by phoning 01784 414 012. If you remain unhappy and wish to complain formally, you can do this by contacting the South London and Maudsley NHS Foundation Trust Complaints Department (Complaints Department, Maudsley Hospital, London, SE5 8AZ; Telephone: 020 3228 2444; email: complaints@slam.nhs.uk).

In the event that something goes wrong and you feel you have been harmed by the research and this is due to a researcher's negligence, then you may have grounds for a legal action for compensation against Royal Holloway University of London, but you may have to pay your legal costs. The researcher has professional indemnity insurance paid for by Royal Holloway University of London. The normal complaints procedure described above would also still be available to you.

Who is funding and organising this study?
The Department of Clinical Psychology at Royal Holloway University of London is organising and funding this study.

Will my information be confidential?
Yes. Your information will be kept confidential and secure, in line with the General Data Protection Regulation and the Data Protection Act (2018).

The information you give us about yourself on the questionnaires will be anonymised and stored securely. Any personal information you provide (such as your name or email address) will be stored separately from the questionnaires you fill out.

The information you provide will not be shared with anyone outside the research team. The only reasons why the research team would break this confidentiality are if they were concerned about danger to yourself or someone else, or if you appeared upset when completing the questionnaires. If either of these things happened, the researcher would share their concerns with your care team at the Lishman Unit. The researcher would always try to talk to you first before they did this.

General Data Protection Regulation Statement
Royal Holloway University of London is the sponsor for this study based in the United Kingdom. We will be using information from you and your medical records to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. We will keep identifiable information about you for 5 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways for the research to be reliable and accurate. If you withdraw from or can no-longer participate in the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

Royal Holloway University of London will collect information from you for this research study in accordance with our instructions. Royal Holloway University of London will use your name and contact details to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Individuals from The Lishman Unit and regulatory organisations may look at your medical and research records to check the accuracy of the research study. The Lishman Unit will pass these details to Royal Holloway University of London along with the information collected from you. The only people in Royal Holloway University of London who will have access to information that identifies you will be people who need to audit the data collection process. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details. Royal Holloway University of London will keep identifiable information about you from this study for 5 years after the study has finished.

Royal Holloway University of London will also collect further information about you for this research study from the Lishman Unit. We will ask The Lishman Unit for your functional neurological diagnoses and scores on two questionnaires, as specified above. This is classed as health information, which is regarded as a special category of information. We will use your health information to help us to analyse the questionnaire data you provide.

The study is likely to be written up as a university thesis and as an academic article. These would never include your name or any kind of information which could identify you. You can give your email address to the researcher if you would like to be provided with a summary of the results.

You can find out more about how we use your information by contacting the lead researcher, Toby Newson.

Ethical Approval
This study has been reviewed by the London Surrey NHS Research Ethics Committee.

Appendix 10: Empirical Study Consent Form

IRAS ID: 245770

Participant Identification Number:

CONSENT FORM

Title of Project: Is there a relationship between early maladaptive schemas and functional neurological symptoms?

Name of Researcher: Toby Newson

1. I confirm that I have read the information sheet (dated 07/01/2019, version 2) 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 consent to my care team disclosing my functional neurological disorder diagnoses from my medical records.

4. I consent to my care team at the Lishman Unit disclosing my scores on the Cambridge Depersonalisation Scale, the Dissociative Experiences Questionnaire and the Functional Neurological Disorders Scale, if I completed them on admission to the Lishman Unit.

5. I understand that the researchers will treat any information I give them confidentiality. I also understand that they may break confidentiality to speak to my care team if they are concerned about any risk of harm to myself or others, or if I appear distressed by participating.

6. I would like to be emailed a copy of the results of the study when they are completed. If so, please provide your email address: _____

7. I agree to take part in the above study.

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Person taking consent	Date	Signature

Appendix 11: Tests of Normality in the Empirical Project

Table 9

Significance test of normality for selected variables in the empirical project

Variable	Shapiro-Wilk Statistic *W*	*p* value
EQ Index Scores	.829	.001
Schema Domain Scores		
Disconnection & Rejection	.939	.154
Impaired Autonomy	.948	.242
Excessive Responsibility	.971	.681
Impaired Limits	.890	.014
Maternal YPI-R Scores		
Emotionally Depriving	.822	.001
Belittling	.743	< .001
Perfectionistic	.967	.591
Controlling	.843	.002
Paternal YPI-R Scores		
Emotionally Depriving	.849	.002
Belittling	.699	< .001
Perfectionistic	.951	.281
Controlling	.772	< .001