INVESTIGATING THE SUBJECTIVE EXPERIENCE OF LIVING WITH EPILEPSY OR PSYCHOGENIC NONEPILEPTIC SEIZURES

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**Abstract**

Psychogenic nonepileptic seizures (PNES) are paroxysmal events superficially resembling epileptic seizures. Although exploring the subjective experience is key in the management and care of patients with epilepsy or PNES, there is a lack of research into the subjective symptomologies. The primary aim of this thesis was to investigate the phenomenology and clinical implications (diagnostic, prognostic and therapeutic) of the lived experience of epilepsy or PNES. Firstly, a systematic synthesis of 21 qualitative studies investigating personal experiences of PNES was conducted. Five key themes emerged revealing experiences of treatment, impact to daily life, and emotional events, as well as differences between the seizure accounts of those with epilepsy. The first empirical study in this thesis explored a series of self-reported measures demonstrating that, subjective experiences could contribute to the diagnostic process as symptoms of panic associated with episodes of transient loss of consciousness could be used to distinguish between PNES and epilepsy or syncope. In study two, a series of multiple-regression analyses revealed that in both epilepsy and PNES, illness perception was a stronger predictor of health-related quality of life (HRQoL) when compared to demographic and condition-related factors. In study three, it was found that writing about experiences of living with a seizure disorder was associated with qualitative and quantitative benefits, including improved HRQoL one-month later. In the final series of studies, it was demonstrated that analysing and comparing individuals’ written accounts of PNES or epilepsy could help to improve our understanding towards some of the problems that individuals experience, as well as highlight clinical implications. In conclusion, research into the subjective experience of seizure disorders is a valuable area of research. More specifically, in-depth investigations are needed into the impact, prevalence and management of specific experiences, which include the voices of individuals that so far have been under-represented.

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# Glossary of Abbreviations

|  |  |
| --- | --- |
| **ACT** | Acceptance and Commitment Therapy |
| **ANOVA** | Analysis of variance |
| **APA** | American Psychiatric Association |
| **B-IPQ** | Brief Illness Perception Questionnaire |
| **CBT** | Cognitive Behavioural Therapy |
| **CT** | Computerised Tomography |
| **CIDI** | Centre for International Disaster Information |
| **DSM-V** | Diagnostic and Statistical Manual of Mental Disorders |
| **EEG** | Electroencephalography |
| **FEW** | Focused Expressive Writing |
| **GAD-7** | Generalised Anxiety Disorder 7-Item Scale |
| **GP** | General Practitioner |
| **HCP** | Healthcare Professionals |
| **HIV** | Human Immunodeficiency Virus |
| **HRQoL** | Health-Related Quality of Life |
| **ICD-10** | International Classification of Diseases |
| **LSSS-3** | Liverpool Seizure Severity Scale Version 3 |
| **MRC** | Medical Research Council |
| **NDDIE** | Neurological Disorders Depression Inventory for Epilepsy |
| **NEWQOL-6D** | Quality of Life in Newly Diagnosed Epilepsy – 6 Dimensions |
| **NICE** | The National Institute for Health and Care Excellence |
| **PNES** | Psychogenic Nonepileptic Seizures |
| **PRO** | Patient Reported Outcomes |
| **SPSS** | Statistical Package for the Social Sciences |
| **TLoC** | Transient Loss of Consciousness |
| **UK** | United Kingdom |
| **US** | Unites States |
| **WHO** | World Health Organisation |

**Chapter One**

# Epilepsy and psychogenic nonepileptic seizures: Background and clinical relevance

This chapter is original work written for this thesis, however, it does contain elements of work published previously in:

Reuber, M., & Rawlings, G. H. (2016). Subjective experience of psychogenic nonepileptic seizures. In M. Hallett, J. Stone, & A. Carson (v.139), Handbook of Clinical Neurology (p.283-296). Elsevier Science Inc.

I have confirmed with the publisher, Elsevier, that I have permission to include part of the article in this thesis.

## **1. Introduction**

## **1.1 Epilepsy**

Seizures have been reported in much of human history and across many cultures. The earliest known description of epilepsy is in a Babylonian diagnostic manual called the Sakikku (1067 - 1046 B.C.). In this, epilepsy is referred to as antašubba, which has been translated as “the falling disease” (Wilson and Reynolds, 1990). Seizures have often been surrounded by misconceptions, discrimination and myths. For example, they have been considered (and still are in many parts of the world) contagious, a sign of spirit possession, or a bad omen (Jacoby and Austin, 2007; Wilson and Reynolds, 1990). Despite great advances in the scientific understanding of epilepsy and significant investment in public educational campaigns, poor attitudes towards epilepsy and understanding of the disorder still persist (Jacoby et al., 2004; Baulac et al., 2012; Fernandes et al., 2011; Baker et al., 2000).

Epilepsy is one of the most common neurological conditions. In the United Kingdom (UK) it affects between 5 - 10 per 1,000 people (NICE, 2015). The International League Against Epilepsy has defined it as a disease of the brain, which is diagnosed on the basis of a single unprovoked seizure, plus the probability of seizure reoccurrence of at least approximately 60% or after two unprovoked seizures. While epilepsy is a long-term condition for many people affected, it can “resolve”, for example, in cases where individuals have been seizure-free for at least ten years, with a minimum of five years off anti-epileptic medication (Fisher et al., 2014).

Epilepsy is not a diagnosis, but is one manifestation of a problem with the brain. A diagnostic “scheme” outlining five different axes have been proposed to facilitate health professionals with the clinical care and diagnosis of individuals with (or suspected of having) the condition (Engel, 2001; Engel, 2006).

The first axis involves exploring the patients’ (and seizure witnesses’) subjective experience of before, during and after the episode. Depending on factors, such as the doctors’ questioning style and the patients’ level of recall, this can be a detailed or short account. This stage helps to differentiate between the myriad of potential diagnoses in patients presenting with paroxysmal symptoms which could be related to epilepsy (Krishnamoorthy, 2006).

In axis two, the epileptic seizure type experienced by the patient is identified from a pre-defined list of accepted seizure form. Epileptic seizures are characterised by sudden and involuntary events associated with a range of motor, sensory, and mental manifestations. However, to a great extent, ictal symptomatologies are determined by the location, spread and duration of epileptic activity in the brain, and clinical manifestations are heterogeneous in nature. For example, generalised seizures involve epileptic activity in both hemispheres of the brain, whereas focal seizures are localised to one area, which may or may not spread (Altrup et al., 2005). In epilepsy, there is great inter-individual variability, but the intra-individual variability is more limited. Although patients can, for instance, experience a range of different seizure types, it would be typical of epilepsy for each seizure type to be quite stereotyped in a particular patient. The stereotypical nature of attacks in one individual is a diagnostic indicator of epilepsy.

Axis three involves the diagnosis of a specific epilepsy syndrome. The final diagnosis should be made on the basis of all available evidence including subjective experience, clinical information and characteristics of the disorder.

The aetiology is defined in axis four. Epilepsy can be caused by a range of different problems with the brain such as, physical trauma, developmental problems, metabolic disorders, stroke, inflammation or tumors. It can also be the manifestation of genetic disorders. For some individuals however, despite best efforts, the cause of their epilepsy is not always known. Advances in the field of neuroimaging (investigating function and structure) can help to determine a range of underlying aetiological problems. This includes computerised tomography (CT) and magnetic resonance imaging scans. Epileptic seizures are characterised by hypersynchronous and excessive electrical discharges of cortical neurons. As such, electroencephalography (EEG) tests are often routine in the examination of individuals suspected of experiencing an epileptic seizure (Oguni, 2004).

In the final axis, the individual can undergo an optional evaluation to assess the impairment associated with epilepsy on their functioning, disability and health. Epilepsy can be debilitating and disquieting having profound consequences on a range of daily activities. As a result, individuals with epilepsy tend to report a lower health-related quality of life (HRQoL) (Taylor et al., 2011). Seizure events themselves can be a distressing and upsetting experience, the related-anxiety of which can be perpetuated by the uncertainty or unpredictability of when the next seizure will occur. Epilepsy can have a detrimental impact on psychosocial factors; for instance, individuals can experience stigma-related to having a seizure disorder (Jacoby and Austin, 2007). Those with epilepsy are also at an increased risk of experiencing emotional and behavioural disorders, such as anxiety, depression and somatic conditions, which is linked to both the development of the condition and/or sequelae (Tellez-Zenteno et al., 2007).

Approximately, two-thirds of patients can successfully control their epileptic seizures using anti-epileptic drugs, either by receiving monotherapy or polytherapy. Some individuals however experience refractory epilepsy. Other treatments are available (often in addition to anti-epileptic medication) including surgery, vagus nerve stimulation, special diets and psychological intervention (NICE, 2015).

## **1.2 Psychogenic nonepileptic seizures**

Psychogenic nonepileptic seizures (PNES) are episodes of impaired self-control. PNES superficially resemble syncope (recurrent fainting) or epileptic seizures, but are not associated with the pathophysiological changes characteristic of syncope or with epileptic neuronal discharges in the brain typical of epilepsy. Instead, studies using a wide range of different methodologies indicate that most PNES can be best understood as an experiential and behavioural response to internal or external stimuli (Reuber, 2009; Roberts and Reuber, 2014). The key symptoms of PNES, such as alterations in consciousness and the partial or complete loss of normal integration between memory, awareness of identity, and control of bodily movements, have increasingly been conceptualised as paroxysmal dissociative responses (Bowman, 2006; Brown and Reuber, 2016).

PNES are not rare. Together with epilepsy and syncope, PNES explain over 90% of presentations to neurologists with transient loss of consciousness (TLoC). Approximately, one in five patients referred to seizure clinics will have PNES (Angus-Leppan, 2008). The diagnosis of PNES is often an iterative process relying on the integration of data from multiple sources, including the patient’s account of their seizure experience, their previous history, witness reports and results of investigations, such as recordings of a typical attack with video-EEG (Malmgren et al., 2012; Lafrance et al., 2013b)

Given the clinical profile and multifaceted nature of the condition, the medical treatment of patients with PNES usually involves contact with many different healthcare professionals (Baslet et al., 2015). The current treatment of choice is psychological therapy (Gaynor et al., 2009), however, to date there is a lack of fully powered clinical trials and there is no clear agreement as to which type of therapy works best for whom (Lafrance et al., 2013b). In view of the heterogeneous nature of the condition, some authors suggest that treatment has to be flexible and individualised (LaFrance, 2007; Howlett and Reuber, 2009).

The prognosis of PNES in adults is variable, but many patients do not do well (Reuber et al., 2003a). A systematic review of 18 studies involving 1,024 patients reported that fewer than four in ten patients are seizure-free five years after their initial diagnosis (Durrant et al., 2011). Having said that, symptoms of psychopathology may continue following a reduction in the frequency of PNES so seizure remission alone should not be the only outcome of treatment (Reuber and House, 2002; Reuber et al., 2005).

Although different labels (including major hysteria and hystero-epilepsy) have been used to describe it, the clinical entity of what is today categorised as PNES has been recognised for a long time. However, especially since the introduction of synchronous video-EEG monitoring into routine care in the 1970’s and 1980’s, phenomenological research about PNES has focused almost entirely on demonstrating the absence of the physiological changes associated with epileptic seizures and on externally observable differences between PNES and epilepsy (Reuber, 2008). In contrast, the subjective symptomatology of PNES has largely been neglected, despite the current medical nosologies - International Statistical Classification of Diseases and Related-Health Problems (ICD-10), and The Diagnostic and Statistical Manual of Mental Disorders, Firth Edition (DSM-5) - categorising PNES as a mental disorder (World Health Organization, 1992; American Psychiatric Association, 2013).

Given that PNES may be a dissociative phenomenon; the reasons for this neglect may not be limited to the fact that visible PNES manifestations are easier to capture, objectify, analyse and report than subjective experiences, they may also include that PNES usually involve subjective states or experiences that are particularly difficult for patients to share.

**1.3 Subjective experience**

It is widely debated within and across academic disciplines what is meant by the term “subjective experience” (Paley, 2014). In his now famous paper, “What is it like to be a bat?” Nagel (1974; 2012) explains that the subjective character of experience consists of all subjective phenomena that are associated with a single point of view. Furthermore, if someone (or something) has an experience, then there is something that it is like to be that person. Due to the egocentric nature of how we experience an event, the subjective experience of one individual is uniquely different from that of another. For the purpose of this thesis, the term “subjective experience” is used to describe the subjective stream of consciousness encompassing phenomena including, but not limited to, thoughts, attitudes, emotions, perceptions, memories, beliefs and views – this also includes the subjective experience associated with the interaction of others, for example, the perception of how healthcare professionals treat individuals as a result of their condition. Ultimately, “subjective experience” encompasses any aspect of the individuals’ mental life or inner world specifically from their point of view.

Nagel (1974; 2012) goes onto to say that, the knowledge gained from an experience is obtained through direct and first-hand exposure. While we can attempt to understand what another person has experienced by making inferences, general observations, extrapolation or imagination, we are limited by the resources and structure of our own minds. These resources are inadequate to perform the task successfully as we will interpret another person’s experience from our own point of view - from the outside looking in. Nagel also makes a similar point in that, it is difficult to understand what an individual experiences through the means of objective measures and whilst such an approach may yield a more accurate or accessible view of the event, we would actually be taking a step away from truly understanding the experience, as less significance will be placed on the individuals’ viewpoint (pg. 445). As such, the best evidence that would be available to allow us to understand what it is really like to be that person would come from the experience of him or her (Nagel, 2012; Nagel, 1974). Unlike in his analogy of trying to comprehend what it is like to be a bat (or a wasp, or a flounder), i.e. a being unable to share their experiences with us, people are able to communicate through a range of mediums the different aspects of their experiences helping us to understand and appreciate what it is like to be them. In the current work, patients’ experiences will be investigated using an array of methodological approaches exploring such self-reported subjective accounts in order to explore the relevance of subjective experience in epilepsy or PNES.

**1.4 Bio-medical model of illness**

Western medicine has it origins in two distinct philosophical notions, which are in turn, responsible for driving the bio-medical model of illness: 1) the mind-body dualism, which states that the mind (psychology, emotion) and body (physical) are separate and can be dichotomised into different entities, these are detached from each other and therefore have little influence on one another, and 2) bio-medical reductionism that suggests pathogenic agents and biological mechanisms can explain all symptoms entirely independent of social or cultural factors, and any changes in health can be explained on the basis of bio-chemical or neurophysiological processes.

The bio-medical model of illness has had a profound impact on different aspects of healthcare:

1. It has meant that medical practice tends to discounts the significance of mental processes suggesting that doctors should not necessarily take into consideration psychosocial issues, but instead, focus their attention on physicalsymptoms making them their priority (Gendle, 2016).
2. It has placed a greater emphasis on objective evidence than on the subjective experiences of patients. Even today, it has been noted that it seems as if there is a gap between the patient and medical research or clinical care, whereby one narrative, typically the bio-medical one, is superior and therefore silences all others – especially the voices of those that it failed to understand (Bowman, 2016). This tendency risks de-humanising or depersonalising medicine, and replacing, changing or discounting patients’ accounts of their illness to fit in with the bio-medical model of illness. Although there has been significant progress in healthcare as a result of objective evidence-based medicine, there is clearly an audience and need for complimentary narrative-based practices (Kalitzkus and Matthiessen, 2009; Greenhalgh, 1999). For example, in patients presenting to emergency departments with upper respiratory infections, patient satisfaction was not related to whether they were given antibiotics, but instead, to their level of understanding of the illness following the consultation (Ong et al., 2007).
3. A greater weight is given to visible problems or manifestations. This is not only true from the perspective of health professionals or clinical guidelines, but also that of the patient. It has been demonstrated that, adults presenting with acute abdominal pain were more confident with their medical evaluation when laboratory testing and CT imaging were included in addition to history and physical examination (Baumann et al., 2011).
4. How illnesses are conceptualised determines the boundaries or responsibilities of healthcare professionals. For example, in the case of PNES, it is unclear what the most effective treatment pathway is and who should be responsible for treating these individuals. Sahaya et al. asked 115 healthcare professionals which specialty do they think is best for managing PNES (the sample consisted of those in primary care, neurologists and in-patient nurses). Psychiatry was the most preferred specialty, followed by neurology, and primary care was the least preferred. Given that responses were not unanimous, this would suggest that professionals are not working from the same model of the condition (Sahaya et al., 2012). Dworetzky asked epilepsy experts in the United States a range of questions about their practice around PNES. Following the diagnosis, n=93/134 experts reported offering all patients at least one follow-up appointment, n=35/134 provided follow-up until seizures were controlled. However, over 20% believed that only mental health follow-up is needed. The majority of professionals offered no follow-up for patients who doubted their diagnosis or who had not contacted a mental health provider. The author suggest this is sending a clear message that, “*we do not really care what happens after the diagnosis is made and that we are completely finished with the patient*” (pg. 356) (Dworetzky, 2015).
5. It has had an influence on healthcare professionals’ attitudes and behaviours towards patients with the condition, and vice versa. In the study above by Dworetzky, out of the 134 epilepsy experts interviewed, only 10% reported that they “always” discuss the possibility of PNES before patients undergo an EEG investigation. Overall, 43% reported only “occasionally” introducing the possibility of PNES. The author makes the point that in other specialties, it is unlikely that a new diagnosis would be “*sprung upon*” the patient. What is more, neglecting to introduce the potential diagnosis before the test may negatively impact the patient-doctor alliance (Dworetzky, 2015). In a study comparing neurologists’ perceptions of PNES to patients with the condition, Whitehead et al. reports that compared to patients (n=40), neurologists (n=45) were more likely to support psychological causes of PNES and less likely to report non-psychological causes. Neurologists viewed the most important cause of the condition as some form of “abuse”. Only one patient with PNES endorsed “abuse”, whereas nearly half of the patients involved in the study gave a physical reason for their condition. In other words, neurologists perceived PNES as a mostly or entirely psychological cause, whereas patients were more likely to endorse a wholly physical cause (Whitehead et al., 2013).
6. The bio-medical model of illness has been accused of failing to explain psychological conditions, presenting this group of problems as a result of purely biological factors (e.g. dysregulation of neurotransmitters, abnormalities in brain structure or function), which can be treated pharmaceutically (Deacon, 2013). PNES is conceptualised as a mental disorder, and even then its classification is debated within the medical nosologies. In the DSM-5, it is classified as a conversion disorder –neurological symptoms that cannot be explained by a neurological problem, whereas in the ICD-10, it is a dissociative disorder. A purely biological account of PNES for instance, does not take into account the aetiological complexities of the condition. Likewise in epilepsy, this model does not give importance to symptoms of psychopathology that have manifested as a result of the epilepsy or that may indeed have been a contributing factor to the development of the condition.

**1.5 Bio-psychosocial model of illness**

Within medicine, there is an emerging interest in examining patients’ subjective experience and perception of illness. The use of such findings can help to improve our understanding of disorders as well as the quality of healthcare provisions (Bury, 2001). Research focusing on patients’ perception demonstrates that what patients say (and how they say it) provides an important complementary voice to the bio-medical one. This can offer an unmatched window into the subjective experience, giving voice to distress that is distinct from the bio-medical approach (Hydén, 1997; Ochberg, 1988).

In response to the limitations of the bio-medical stance, the late George Engel proposed the bio-psychosocial model, in which biological, psychological and social factors interact to determine health (Figure 1.1). This model provides a holistic approach, taking into account patients’ experiences as well as the biological agents of illness, and their interactions (Engel, 1977). The model advocates that no patient or illness can be reduced to a single aspect, but instead, all factors are predominately relevant in all cases, at all times (Ghaemi, 2009). The bio-psychosocial model provides a framework to assess illness rather than to explain it. It invites researchers to collate qualitative and quantitative data in psychological, social and cultural domains, and incorporate them into the model. The purpose is not to devalue or replace medical knowledge, but instead, it seeks to include patients’ experiences into clinical practice allowing care to be more personalised and responsive to the patients’ needs (Atkinson and Rubinelli, 2012). Integrating the subjective experience (as well as acknowledging biological factors) into the diagnostic and therapeutic processes can help us to better understand the illness.

Biological

Health

Psychological

Social

Figure 1.1 Factors constituting the bio-psychosocial model.

In support of this model, Engel outlines several points suggesting how the inclusion of psychosocial factors could result in better patient care, a number of which have potential relevance to epilepsy and PNES:

1. He argues that, biological abnormalities are used as the diagnostic criterion for disease. However, a positive test only indicates the disease potential, which is inadequate to determine whether the patient is ill or not – “*it defines a necessary but not a sufficient condition for the occurrence of the human experience of the disease, the illness*” (pg. 131). Having said that, in PNES, the gold standard approach to making a diagnosis depends on the demonstration on the absence of objective changes – i.e. the recording of typical attacks simultaneously on video and EEG revealing an absence of ictal EEG changes before, during or after the event (Lafrance et al., 2013a). Although visible clinical manifestations can be used to achieve a diagnosis, how patients experience or report their symptoms can give further context, meaning, and help others to understand the nature of their symptoms. In the case of epilepsy and PNES, the differential diagnosis is crucial because the most effective treatment of these conditions is very different. Many patients with PNES receive an initial misdiagnosis of epilepsy and are therefore erroneously treated with anti-epileptic drugs (Reuber et al., 2002; Benbadis et al., 2004). The delayed recognition of PNES and the inappropriate treatment of presumed epilepsy may worsen the prognosis (potentially allowing symptoms to become established and more chronic) (Reuber et al., 2003a), cause iatrogenic harm from drug toxicity (Mayor et al., 2012) and emergency intervention (Gunatilake et al., 1997). Death due to the mistreatment of PNES as epilepsy has been reported (Reuber et al., 2004). Such outcomes constitute a considerable cost to health and social services, as well as the patients’ (and their family members’) emotional and physical wellbeing. A better understanding of the typical interictal and ictal symptom profiles of patients with epilepsy or PNES should help to improve the diagnostic process. In line with the dogma of the bio-medical model, the differentiation of epilepsy and PNES based on visible or objective charactierstics has been the subject of a considerable number of studies, for instance, using surface electromyography, ictal eye closure, ictal motor movements, ictal weeping, ictal stuttering, and seizure duration (Chung et al., 2006; Beniczky et al., 2015; Gates et al., 1985; Walczak and Bogolioubov, 1996; Vossler et al., 2004; Avbersek and Sisodiya, 2010). However, batteries of self-report questionnaires, which have included questions about interictal and ictal symptoms, have also been shown to distinguish accurately between PNES, epilepsy and syncope (Syed et al., 2009; Reuber et al., 2016). As no single measure or characteristic can or should be used to differentiate between conditions associated with TLoC, it follows that the final diagnosis should be based on all available clinical information including a range of subjective and objective data.
2. The model helps to establish a link between biological factors and clinical presentation - how the patient experiences the expression of their illness. The bio-medical model places greater importance on bio-chemical processes bypassing or disempowering patients’ accounts, when in fact, in those who have experienced an episode of TLoC, the act of taking and interpreting the patients’ narrative of their subjective experience is, arguably, still the most important diagnostic tool (Plug and Reuber, 2009; Malmgren et al., 2012).
3. Research that collects and analyses how patients experience and express their illness can help to gain a deeper meaning of their illness narrative in psychological and social terms. This information can facilitate the therapeutic process between the doctor and patient and provide a basis for psychotherapeutic intervention. For example, a better understanding of subjective perceptions associated with epilepsy or PNES is likely to have implications for attrition and adherence to interventions. The engagement of patients with PNES in psychotherapy is often difficult because patients perceive their disorder to be a “physical” rather than a “psychological” problem (Whitehead et al., 2013). Several studies have demonstrated that patients who accept the diagnosis of PNES are more likely to have better treatment outcomes (Ettinger et al., 1999a; Duncan et al., 2014). Furthermore, it may well be easier for clinicians to convince patients (and caregivers) that they experience PNES if the patient is aware of unpleasant warning symptoms, or at least of interictal mental health problems, and can accept that the seizures could be manifestations of arousal or distress (Thompson et al., 2009; Monzoni et al., 2011a; Monzoni et al., 2011b).
4. Biological factors are not always sufficient to explain the development of certain illnesses. Psychosocial factors on the other hand, have the propensity to influence the trajectory, onset and recovery of illness, and exposure to life events can play a pivotal role in the course of the disease. Reuber (2009) has applied the bio-psychosocial model in an attempt to understand the aetiology of PNES. He explains that, no single factor or mechanisms can be used to explain PNES in all patients. Instead, a range of interacting factors may contribute to the development of the condition including: *predisposing factors*, such as genetics, early abuse or neglect, and physical factors (structural or functional brain abnormalities); *precipitating factors*,occurring just prior to the onset of the seizure disorder (e.g. adult life events, psychopathologies, triggers); and *perpetuating factors* that make it difficult to take control of seizures (i.e. maladaptive coping tendencies, isolation, illness perceptions).
5. While bio-chemical factors may provide an indication of whether the patient has an illness or not, they are not sufficient to determine whether the patient perceives themselves as sick, and if they themselves accept the sick role. Notwithstanding the fact that the outcome of epilepsy and PNES is heterogeneous, individuals’ experience of and attitudes towards ictal and interictal states may provide insights into circumstances that are important in determining prognosis. While most studies into these conditions have investigated the association or risk of outcome with a range of clinical and demographic factors such as, seizure-related symptoms, aetiology, psychiatric comorbidities, age, education, employment status, socio-economic factors, seizure manifestations, and treatment (Reuber et al., 2003b; Mohanraj and Brodie, 2013; Durrant et al., 2011; Ettinger et al., 1999b; Arain et al., 2007; McKenzie et al., 2016), there is evidence to suggest that subjective variables are also important predictors. In PNES, patients who held an internal locus of control (the belief that they themselves can control events and outcomes, as opposed to holding an external locus of control) were more likely to be seizure free six-months following a psychological intervention (Duncan et al., 2016). Another study in PNES reported that, following diagnosis, those who continued to experience seizure events were more likely to have described feeling angry or confused in response to the diagnosis (Carton et al., 2003). In summary, in epilepsy and PNES, subjective factors including attitudes and self-reported symptoms appear to be important factors of outcome. Such characteristics could help to inform clinical interventions highlighting potential targets for therapeutic change.
6. Modifying the patients’ behaviour and thoughts has an influential effect on specific outcomes, “*for better or for worse*” (pg. 132). Engel explains that the responsibility of a healthcare professional includes educating the patient and providing psychological care. The disclosure and self-exploration of subjective symptoms associated with chronic disorders (such as the phenomenology of illness, or the predisposing, perpetuating or precipitating factors related to the development of symptoms) can, in itself, be therapeutic. In epilepsy, psychological interventions have been used, and indeed are recommended, in the management of epilepsies (NICE, 2015). For example, Cognitive Behavioural Therapy (CBT) involves the expression and formulation of an individual’s behavioural, emotional, cognitive and physiological reactions to a stimulus. In epilepsy, although findings vary between individual studies, this approach to treatment has been associated with a reduction in anxiety (Mula, 2013), depression (Gandy et al., 2013) and seizure frequency (McLaughlin and McFarland, 2011). In PNES, following CBT, patients have demonstrated a reduction in seizure remission and healthcare utilisation (Goldstein et al., 2010; Carlson and Perry, 2017).

## **1.6 Thesis outline and aims**

The overall aim of this thesis was to further our understanding of what it is like to live with epilepsy or PNES. This will be achieved by using the bio-psychosocial model as a framework looking to see if aspects of patients’ subjective accounts are of clinical relevance, specifically to the diagnosis, prognosis and treatment of these conditions.

While aspects of this work will be based on accounts of living with seizures in general (without differentiation between epilepsy and PNES), several chapters will compare and contrast the subjective accounts of people living with epilepsy with those of individuals living with PNES. This is in line with many studies in the literature. For example, most studies examining PNES symptoms have used self-report questionnaires comparing the mean responses of different groups of participants, mainly patients with PNES and epilepsy, less commonly healthy controls or other patient groups such as those with syncope. Indeed previous studies comparing and contrasting qualitative findings have provided useful insights into these disorders and have been especially valuable in aiding the differentiation of PNES and epilepsy: A series of qualitative studies have compared how individuals talk about living with epilepsy or PNES exploring different linguistic features (Schwabe et al., 2007; Schwabe et al., 2008; Plug et al., 2009; Plug et al., 2010; Monzoni et al., 2011b; Plug et al., 2011; Cornaggia et al., 2012; Robson et al., 2012). It has been demonstrated that these observations can facilitate the diagnostic process in routine clinic interactions between patients and doctors (Jenkins et al., 2016). The use of complementary approaches is to be particularly welcome in this area of research because different perspectives are likely to produce the best insights. A compare-and-contrast approach also helps us to test the specificity of our findings between two disorders that are superficially associated with the same visible manifestations - seizures. I am aware that the application of external categories (such as medical diagnoses) is considered problematic in many qualitative research approaches and that there is a clear preference to work closely with the data and base findings on the data itself. However, the previous literature makes it likely that there are important differences in life experiences of patients with epilepsy, PNES and other disorders causing paroxysmal neurological symptoms including loss of awareness, and the application of external categories in several parts of this thesis makes it easier for readers to relate the qualitative findings reported to previous and future studies demonstrating quantitative differences between these diagnostic groups.

This thesis has been written in accordance with the guidelines (including pagination) for the Alternative Format Thesis by The University of Sheffield. As such, this thesis is structured as follows:

*1.6.1 Chapter Two:* A systematic review has already been conducted investigating qualitative research into the impact of epilepsy on children and adults (Kerr et al., 2011), however, no such review existed for PNES. As such, the primary aim of this chapter was to conduct a systematic synthesis of published qualitative research investigating the subjective experience of living with PNES in adults. Reviews are seen as having a vital role in the decision making process in medical practice. Systematic reviews can identify, evaluate and summarise large banks of empirical literature investigating a similar problem, and thus making it easier for decision makers (who may not have the time to read the individual articles) to access and digest (Gopalakrishnan and Ganeshkumar, 2013). In this case, patients’ accounts of living with PNES yielded important insights into the psychological and social aspects associated with their condition, as well as highlighting opportunities to improve healthcare provisions. This review has been published (Rawlings and Reuber, 2016). Contributions of authors are as follows:

Rawlings, G: Design and formulation of research aims; collected, analysed and synthesised the data; and drafted the manuscript (first author).

Reuber, M: Design and formulation of research aims, reviewed the draft manuscript.

*1.6.2 Chapter Three:* In this chapter, the diagnostic implications of subjective experiences were explored. The overall aim of this study was to investigate the frequency and diagnostic value of panic symptoms just before, during and after episodes of TLoC in PNES, epilepsy and syncope. Exploring patients’ accounts of a specific manifestations associated with their condition (i.e. ictus-related panic) can demonstrate the heterogeneity of how symptoms are experienced and expressed. This allows us to further conceptualise the nature and cause of their illness. By comparing the subjective accounts between different conditions that are associated with similar visible manifestations (episodes of TLoC) we are able to gain a better understanding of how and to what extent subjective experiences can help clinicians to differentiate between the different problems. More specifically this study adds to previous literature about the disputed relationship between PNES and panic. The study has been published (Rawlings et al., 2017d). This study was an analysis of a dataset collected under the project title: *Development of a self-report and seizure witness observation tool – the Paroxysmal Experience Profile and Paroxysmal Event Observation Questionnaires.* The data was collected from 1st December 2005 – 30th November 2006. Professor Markus Reuber was the Principle Investigator. Contributions of authors are as follows

Rawlings, G: Design and formulation of the research aims specific to this study, statistical analysis and interpretation, and drafted the manuscript (first author)

Jamnadas-Khoda, J: Obtaining regulatory approval, identifying participants, data collection, data entry, and reviewed the draft manuscript.

Broadhurst, M: Development of Paroxysmal Event Profile Questionnaire, obtaining regulatory approval, and reviewed the draft manuscript.

Grünewald, RA: Development of Paroxysmal Event Profile Questionnaire, identifying participants, confirming participants’ diagnoses, and reviewed the draft manuscript.

Howell, SJ: Development of Paroxysmal Event Profile Questionnaire, identifying participants, confirming participants’ diagnoses, and reviewed the draft manuscript.

Koepp, M: Identifying participants, confirming participants’ diagnoses, and reviewed the draft manuscript.

Parry, S: Development of Paroxysmal Event Profile Questionnaire, obtaining regulatory approval, identifying participants, confirming participants’ diagnoses, data collection, and reviewed the draft manuscript.

Sisodiya, S: Identifying participants, confirming participants’ diagnoses, data collection, and reviewed the draft manuscript.

Walker, M: Identifying participants, confirming participants’ diagnoses, data collection, and reviewed the draft manuscript.

Reuber, M: Design and formulation of the research aims specific to this study, development of Paroxysmal Event Profile Questionnaire, obtaining regulatory approval, identifying participants, confirming participants’ diagnoses, data collection, and reviewed the draft manuscript.

*1.6.3 Chapter Four:* This chapter investigated the prognostic implications of illness perceptions in individuals with epilepsy or PNES. More specifically, this study examined the demographic (gender, age, education), condition-related (duration of the condition, and frequency and severity of seizures) and psychological (depression, anxiety, illness perception) profile of individuals with epilepsy or PNES, and investigated the relationship and predictive power of the aforementioned variables on HRQoL. The rationale of this study was to gain a greater understanding of the potential variables that are responsible for determining the impact of the condition on an individuals’ quality of life. The findings from this study also support that notion that multiple factors (condition, psychological, personal) are accountable in predicting health. This helps to inform treatment pathways and therapeutic targets. This study has been published (Rawlings et al., 2017a). Contributions of authors are as follows:

Rawlings, G: Obtaining regulatory approval; methodological design and formulation of the research aims; identifying, consenting and confirming the diagnoses of suitable participants; data collection, statistical analysis and interpretation; and drafted the manuscript (first author).

Brown, I: Reviewed the draft manuscript.

Reuber, M: Obtained regulatory approval, methodological design and formulation of the research aims, and reviewed the draft manuscript.

*1.6.4 Chapter Five:* In this chapter, the treatment relevance of subjective experiences was examined. The aim of this study was to develop and conduct a pilot randomised controlled trial of a home-based writing intervention for individuals with epilepsy or PNES assessing the feasibility, acceptability and preliminary effectiveness of such an approach. The purpose of this study was to demonstrate that, patients’ state of health could be improved by *treating* psychological facets associated with the condition. This paper has been written with the intent to publish in a scientific journal.

1.*6.5 Chapter Six:* This chapter aimed to add to our understanding of what it is like to live with epilepsy by thematically analysing individuals’ written accounts of their condition. The dataset was collected in the context of a randomised controlled trial investigating a home-based writing intervention for individuals with epilepsy or PNES. This study has been published (Rawlings et al., 2017b). Contributions of authors are as follows:

Rawlings, G: Obtaining regulatory approval; methodological design and formulation of the research aims; identifying, consenting and confirming the diagnoses of suitable participants; data collection, data analysis and interpretation; drafted the manuscript (first author).

Brown, I: Data analysis and reviewed the draft manuscript.

Stone, B: Data analysis and reviewed the draft manuscript.

Reuber, M: Obtaining regulatory approval, methodological design and formulation of the research aims, data analysis and reviewed the draft manuscript.

*1.6.6 Chapter Seven:* In this chapter, the same research design and methodology as chapter six was used, however the written accounts of those with PNES was thematically analysed. This study has been published (Rawlings et al., 2017c).

Rawlings, G: Obtaining regulatory approval; methodological design and formulation of the research aims; identifying, consenting and confirming the diagnoses of suitable participants; data collection, data analysis and interpretation; drafted the manuscript (first author).

Brown, I: Data analysis and reviewed the draft manuscript.

Stone, B: Data analysis and reviewed the draft manuscript.

Reuber, M: Obtaining regulatory approval, methodological design and formulation of the research aims, data analysis and reviewed the draft manuscript.

*1.6.7 Chapter Eight:* This chapter is a thematic comparison contrasting the written accounts of those with epilepsy and PNES. At the time of writing, this chapter has been submitted to a journal for publication. The reason behind the three qualitative chapters was to explore and compare individuals’ accounts of their conditions. The emergent findings have the potential to contribute to the bio-psychological model of PNES or epilepsy identifying factors that are associated with cause, differentiation, impact to health, symptoms, and treatment.

*1.6.8 Chapter Nine:* This thesis concludes with a summary of the major findings, recommendations for future research and clinical implications. It will discuss such outcomes using the bio-psychosocial model as a framework.

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**Chapter Two**

**What patients say about living with psychogenic nonepileptic seizures: A systematic synthesis of qualitative studies**

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**Candidate’s individual contributions:**

G.H. Rawlings was involved in the design and formulation of the research aims specific to this study; data collection, analysis and synthesis; drafted, submitted and corrected the published manuscript. The candidate is the first author.

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**Supplement**

Table 2.1 Summary of excluded studies

|  |  |  |
| --- | --- | --- |
| Primary author | Aim | Reason for exclusion |
| Arnold (1996) | To investigate psychopathology and trauma in patients with seizures | Used clinical interviews |
| Bautista (2008) | To investigate whether patients with PNES have observed epileptic seizures | Used close ended questions |
| Blank (2014) | To explore patients perceptions of referrals to epilepsy clinics | Only used patients with epilepsy |
| Ettinger (1999c) | To investigate symptoms following a seizure | Did not describe their qualitative approach/methodology |
| Ettinger (1999a) | To develop a clinical, psychiatric and psychosocial profile | Used a structured questionnaire |
| Ettinger (1999b) | To assess prognosis in patients with PNES | Used close ended questions |
| Griffith (1998) | To determine what relationship may exist between unspeakable dilemmas and PNES | Did not describe their qualitative approach/methodology |
| Hall- Patch (2010) | To evaluate the effectiveness and acceptability of a strategy for communicating the diagnosis of PNES | Whilst the authors did use semi-structured telephone interviews, if patients responses did not consist of the expected answer patients were then cued and may be biased by leading questions |
| Hendrickson (2014) | To explore the differences in panic in PNES and epilepsy | Used clinical interviews |
| Jenkins (2015) | To investigate doctors clinical encounters before and after a training intervention | Focused on doctors and not the patients experiences |
| Kozlowska (2011) | To examine emotional processing in patients with conversion disorders | Grouped patients with PNES with patients who have other disorders – PNES accounted for 17 of 76 patients |
| McMillan (2014) | To investigate healthcare professionals’ views on treatment of PNES | Focused on healthcare professionals’ views |
| Monzoni (2009) | To investigate if conversational displays can offer insight into coping behaviours in patients with epilepsy | Only used patients with epilepsy |
| Monzoni (2011a) | To investigate how neurologists discuss functional symptoms | Focused on doctors and not patients behaviours |
| Morrison (2011) | To investigate healthcare professionals’ experiences of driving regulation with PNES | Focused on healthcare professionals’ views |
| Plug (2009a) | To investigate conversational difference between PNES and epilepsy | Case study |
| Quinn (2012) | To examine factors that contributed to the successful treatment of PNES in men | Used a theory building case study approach |
| Reuber (2009a) | To investigate if conversational analysis can distinguish between PNES and epilepsy | Used qualitative findings only to make a differential diagnosis |
| Stone (2003) | To examine the meanings of labels for patients | Used close ended questions |
| Stone (2013) | To investigate experiences of prodromal symptoms | Did not describe their qualitative approach/methodology |
| Thimm (2011) | To explore the psychosocial effects of PNES on patients | Not published in a peer-reviewed journal |
| Toerien (2010) | To investigate how epilepsy specialists offered treatment and investigational options for patients with seizures | Focused on the doctors behaviours |
| Watson (2002) | To study differences in subjective experience of being in an earthquake between patients with epilepsy and patients with PNES | Did not describe their qualitative approach/methodology |
| Witgert (2005) | To investigate panic symptoms in patients with PNES | Used clinical interviews |
| Wo (2015) | To explore the factors affecting the employability in patients with uncontrolled seizures | Only used patients with epilepsy |

PNES = Psychogenic Nonepileptic Seizures

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**Chapter Three**

**Panic symptoms in transient loss of consciousness: Frequency and diagnostic value in psychogenic nonepileptic seizures, epilepsy and syncope.**

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**Chapter Four**

**Predictors of health-related quality of life in patients with epilepsy and psychogenic nonepileptic seizures**

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**Chapter Five**

**A pilot randomised controlled trial of a home-based writing intervention for individuals with epilepsy or psychogenic nonepileptic seizures**

**5.1 Introduction**

Processing the emotion associated with a distressing life event has profound consequences on psychological and physical health (Silver et al., 1983; Gross, 1989; Cole et al., 1996). This has long been recognised in psychotherapy (Breuer and Freud, 1875/1955) and indeed, forms the basis of many modern psychological treatments, including Cognitive Behavioural Therapy and Acceptance and Commitment Therapy.

Clinical research has applied the process of emotional expression to the practice of writing. The best examined variant of therapeutic writing is based on Pennebaker and Beall’s (1986) focused expressive writing (FEW). In a standard FEW intervention, participants are asked to write about a distressing experience for 15-20 minutes at a time, on three to four separate days. The effectiveness of FEW has been examined by a large number of randomised controlled trials, in which participants are either assigned to a FEW or a control writing group - the latter typically involving writing about emotionally devoid topics

Individual studies have demonstrated the benefits of FEW in a range of long-term conditions including cancer (Merz et al., 2014), cystic fibrosis (Taylor et al., 2003), post-traumatic stress disorder (van Emmerik et al., 2008), depression (Krpan et al., 2013), high blood pressure (McGuire et al., 2005a) and chronic pelvic pain (Norman et al., 2004).

Several reviews have investigated the effectiveness of FEW in clinical populations. Frisina et al. conducted a meta-analysis of FEW studies (n=9) reporting a larger effect on physical (*d*=0.21), rather than psychological outcomes (*d*=0.07, p<0.05) (Frisina et al., 2004). Similarly, a systematic review in patients with cancer by Merz et al. (n=13) suggested that FEW has a small to moderate effect, in particular on pain and general physical and psychological symptoms (Merz et al., 2014). Although these effect sizes are only modest, any health gains are impressive, given that the intervention is time-limited, inexpensive and easy to administer.

There is no general consensus on the mode of action of expressive writing, nor are the effects likely to result from one mechanism in isolation. A number of different theories have been put forward. It has been suggested that writing offers individuals a “safe space” to create a narrative of events, re-author past experiences and search for new possibilities (Lucius-Hoene et al., 2012; Williams, 1984). Writing provides individuals with the opportunity to express sensitive issues, and bring their thoughts and emotions into conscious awareness allowing for understanding and realisation (Sanders et al., 2011; Howlett, 2004). Through writing, individuals are able to vent their emotions without fear of embarrassment or judgment. Last but not least, writing allows individuals to engage in self-affirming behaviours, which may help to buffer against the effects of stress (Steele, 1988; McQueen and Klein, 2006).

Since the conception of FEW, there has been considerable interest from researchers to refine the original writing instructions based on potential mechanisms of change. This means that, whilst there is an accepted framework (writing for at least 20 minutes, on four separate days), what individuals are asked to write about is often specific to the patient group or outcomes under investigation. For example, greater effects have been observed when individuals are instructed to write about their experiences of illness, as opposed to any distressing event (Craft et al., 2013). What is more, participants have been guided to write about the different aspects of their illness, including impact, treatment, diagnosis, insight gained, self-affirmation, and writing from a different perspective such as letter writing (Arden-Close et al., 2013; Gidron et al., 2002; Graham et al., 2008).

Epilepsy is a disorder of the brain characterised by paroxysmal events associated with a range of motor, sensory and mental manifestations. In addition to managing seizure themselves, individuals with epilepsy are also affected by the underlying pathology causing the seizures, and intimately linked cognitive and emotional problems (Altrup et al., 2005). Psychogenic nonepileptic seizures (PNES) superficially resemble epileptic seizures, but are not associated with epileptiform activity. Instead, most PNES are conceptualised as a dissociative response to distressing stimuli (Brown and Reuber, 2016). Both epilepsy and PNES have a multifaceted impact on daily living, including physical, social and emotional consequences (Taylor et al., 2011; Jones et al., 2016).

Previous research suggests that individuals with epilepsy or PNES are at an increased risk of exhibiting problems with expressing their emotions and thoughts. For example, individuals have described often concealing their diagnosis, or find it difficult to discuss the problems associated with living with a seizure disorder for fear of experiencing stigma, shame or guilt (Rawlings et al., 2017c; Rawlings and Reuber, 2016). What is more, many of the symptoms associated with PNES have been interpreted as resulting from emotional dysregulation (Brown and Reuber, 2016; Brown et al., 2013; Roberts and Reuber, 2014; Novakova et al., 2015) – the exploration of patients’ emotional traits/states is an important process in the treatment of PNES (Quinn et al., 2012).

Due to the close links between both seizure disorders and the additional psychosocial problems, a range of psychotherapeutic techniques have been used in their management. Indeed, guidance to health professional suggest that treatment pathways of epilepsy and PNES should not simply focus on achieving seizure remission, but also address other disabling aspects of these conditions (Rawlings et al., 2017b; Dewhurst et al., 2015; Reuber and House, 2002; NICE, 2015; Lafrance et al., 2013). However, such treatments are time consuming and there is evidence that few individuals get access to these interventions (Dickson et al., 2015).

When developing and evaluating a new, complex (complex because the intervention has multiple working mechanisms) psychological intervention, the Medical Research Council (MRC) has published a framework to help researchers. This framework has since become highly influential and widely cited. As shown in Figure 5.1, the model identifies four main stages, of which, stage A and B will be addressed in the current study – these two stages are seen as the preliminary work that is necessary prior to evaluating a definitive intervention.

B. Feasibility/piloting

1. Testing procedures
2. Estimating recruitment/retention
3. Determining sample size

C. Evaluation

1. Assessing effectiveness
2. Understanding change process
3. Assessing cost-effectiveness

A. Development

1. Identifying the evidence base
2. Identifying/developing theory
3. Modeling process and outcomes

D. Implementation

1. Dissemination
2. Surveillance and monitoring
3. Long term follow-up

Figure 5.1 Stages in developing and testing process - taken from Craig et al., (2008).

Stage A outlines that, best practice is to identify the existing empirical and theoretical evidence to develop the intervention to a point where it can reasonably be expected to have a worthwhile effect (Craig et al., 2008). In line with this, Hollon et al. (2002) suggested in their report of developing a psychosocial intervention for depression, that it is best to use the active mechanisms that undermine the therapeutic effectiveness as a theoretical framework. What is more, establishing and focusing key elements of the intervention on these mechanism of change can help to form the basis for applying empirical knowledge to clinical practice. In light of this, we reviewed the evidence investigating writing therapies (including FEW) in clinical and non-clinical samples with the purpose of devising a series of writing instructions aiming to use a range of potential therapeutic processes to improve the health-related quality of life (HRQoL) of participants.

The next stage in the MRC framework involves piloting and feasibility - this is to make sure that the intervention can be delivered as intended. This is seen as an essential step in the development and implementation of an intervention. The MRC do not provide definitive definitions of these terms, but instead, explain that this stage includes “*testing procedures for their acceptability, estimating the likely rates of recruitment and the retention of subjects* [participants]*, and the calculation of appropriate sample sizes*” (pg. 10, Craig et al., 2008). What is more, a pilot study can help to explore the potential problems or uncertainties that may be associated with the intervention prior to a large-scale, fully powered definitive trial. It is suggest that a range of qualitative and quantitative outcome measures will be needed at this stage to explore different aspects when testing the intervention. A review of the literature investigating the differences between what constitutes a pilot or feasibility study explained that, authors tend to use these terms interchangeably or use pre-defined definitions (some of which are their own) – in other words, there is no consistency across the literature. What is more, the authors conclude by suggesting it is “futile” (pg. 133) to ascribe a particular definition to the term feasibility, however, the term pilot should be reserved for studies that “mimic” (pg. 133) a large scale trial (i.e. those that utilise randomisation and control groups), but which do not solely aim to investigate the effectiveness of the intervention rather to make sure that the full trial delivers maximum benefit (Whitehead et al., 2014).

To this end, the current study was designed as a pilot-randomised controlled trial of a home-based therapeutic writing intervention for individuals living with seizure disorders, specifically epilepsy or PNES. The primary aim was to investigate the feasibility and acceptability of a writing intervention in this population by examining quantitative and qualitative self-reported measures. Notwithstanding the fact that this pilot study was not designed (nor powered) to investigate the effectiveness of an intervention, it was a secondary aim of this study to undertake an exploratory analyses of the effectiveness of the intervention on psychological outcome measures (HRQoL, anxiety, depression and illness perception) with the primary purpose of determining likely effect sizes for group size calculations of future definitive effectiveness studies.

**5.2 Methods**

*5.2.1 Participants*

Participants were approached consecutively and recruited from outpatient neurology clinics at the Royal Hallamshire Hospital, Sheffield (United Kingdom). Participants were also recruited through membership-led organisations for individuals experiencing seizures (see acknowledgements for the list of organisations). Recruitment took place between October 2015 and March 2017. The North of Scotland Research Ethics Committee granted ethical approval for this study (15/NS/0078).

Participants were included if they were over the age of 18 years, had a diagnosis of epilepsy or PNES (individuals thought to have comorbid seizure disorders were excluded from the study), experienced a seizure in the last 12 months, were able to provide informed consent, and complete a demographic and clinical questionnaire without help.

All participants self-reported their diagnosis. The diagnoses of all participants recruited at the Royal Hallamshire Hospital were confirmed by review of their hospital records. Confirmation of the self-reported diagnoses of participants recruited through membership-led organisations was sought from their general practitioner.

*5.2.2 Sample size*

Whitehead et al. explains that, when determining the sample size for a pilot study, due to differences in the purposes of studies, a formal power calculation is not necessary. However, justification for the proposed sample size is required nonetheless. General rules of thumb do exist, which can range from n=12 - 70 (Whitehead et al., 2016). For the current study, a systematic review of 55 studies investigating FEW in clinical samples using a randomised controlled trial design reported that the median number of patients in each study was 66 (range 11 - 507) (Rawlings et al., 2015). As such, we aimed to recruit at least 66 individuals.

*5.2.3 Design*

The method was a 2 (therapeutic writing or control writing) x 3 (baseline, one- and three-month follow-up) research design. Writing interventions similar to the one developed for this study have previously been used in a wide range of non-clinical and clinical scenarios (involving different chronic emotional and organic conditions). The investigation was therefore considered potentially useful to individuals with either epileptic or nonepileptic seizures so both of these participant groups were included. Immediately after participants completed the baseline measures, they were randomly allocated by G.R. using a random number computer generator to the therapeutic or control writing condition. The study was single blinded, and individuals were not aware of the study hypotheses or the fact that there were multiple writing conditions.

*5.2.4 Protocol*

Participants recruited from outpatient neurology clinics were sent a participant information sheet at least 48 hours before their appointment with a Consultant Neurologist. On the day of their appointment, individuals were approached and invited to take part in the study. Those who gave written consent were asked to complete a set of self-report measures. Participants recruited from membership-led organisations replied to an advert for a study of a writing intervention designed to help individuals with seizure disorders. Potential participants then contacted G.R. who gained written informed consent and provided access to an online form allowing participants to complete the self-report measures.

After randomisation, participants were given four writing booklets. Each booklet contained writing instructions, space for writing (four sheets of A4 lined paper) and a link to a website if participants preferred typing to handwriting. Individual had to complete all four sessions in order and within two weeks of completing the first session. Participants had to return their writing; they were informed that their writing would be read by a member of the research team with the aim to deepen our understanding of what it is like to live with a seizure disorder. This also allowed us to control for attrition – any participant who failed to adhere to the writing instructions were classed as *non-completers* (i.e. did not complete all four sessions, wrote about their emotions in the control writing).

Participants were instructed to complete an implementation intention questionnaire stating *when* and *where* they intended to complete each writing session. A similar questionnaire has previously been shown to help individuals with epilepsy adhere to their medication regimen (Brown et al., 2009). Participants were asked to conduct their writings at home, in private and away from distractions. Individuals were instructed to record the time that they began and finish writing.

*5.2.5.1 Therapeutic writing instructions*

The four therapeutic writing topics were adapted from the literature on therapeutic writing (including research investigating FEW) in clinical and non-clinical populations. The four questions were developed with the help from eight individuals who experience seizures, as well as a team of mental health specialists at the Royal Hallamshire Hospital (Table 1).

*5.2.5.2 Control writing instructions*

Participants randomised to the control writing group were asked to focus on their actions and behaviours (Table 5.1). Participants were instructed to be as accurate and detailed as possible. These instructions were based on those most commonly used in previous randomised controlled trials investigating FEW.

Table 5.1 Instructions for therapeutic- and control writing conditions

|  |  |  |  |
| --- | --- | --- | --- |
| Session | Control writing | Therapeutic writing | Adapted from and purpose |
| 1 | I would like you to write about what you did over the last week. | I would like you to write about your very deepest thoughts and feelings about your condition, disorder and/or seizures. In your writing you can write about anything you wish, but it is important that you really let go and explore your thoughts and feelings. | FEW instructions (Pennebaker and Beall, 1986). |
| 2 | I would like you to write about what you did over the last 24 hours. | I would like you to write a letter to your condition, disorder and/or seizures. You can address your letter however you like and discuss anything you wish, for example, Dear Seizures. | Letter writing aimed to help individuals to express difficult emotions and facilitate insight into the emotional role that their symptoms play (Howlett, 2004). |
| 3 | I would like you to write about what you plan to do over the next 24 hours. | I would like you to write a letter to your younger self. This version of you can be any age you wish; they may even be a pre-seizure version of you who has no idea they will be later diagnosed nor have the knowledge of what it is like to live with your condition, disorder and/or seizure. You can address your letter however you like and discuss anything you wish, for example, Dear Pre-Seizure Me or, Dear 16 year old me. | This allowed individuals to notice novel possibilities which can be translated to current problems, recognise what resources and wisdom they have gained to cope, and identify how their past experiences have shaped them into the person they are today and who they may become in the future (Kress et al., 2008). |
| 4 | I would like you to write about what you plan to do over the next week. | I would like you to choose a common personal value and write in-depth about why it is important to you, why it is central to your life and how it makes you feel about yourself? You may want to describe a time in your life when you had the opportunity to really express this value. | This is the values-affirmation task (Sherman et al., 2000). This aimed to help buffer individuals against daily stress and also any negative consequences resulting from the previous writing sessions. |

FEW = Focused Expressive Writing

*5.2.6 Measures*

*5.2.6.1 Demographic and medical information*

This included age, gender, years in education, current seizure diagnosis, duration since seizure onset, and the date of their last seizure.

*5.2.6.2 Anxiety*

The Generalised Anxiety Disorder (GAD-7) is a seven-item scale used as a screening tool and severity measure of mild (score of 5-9), moderate (10-14) and severe anxiety (>15). Participants are asked to report on a four-itemed Likert scale (Not at all, Several days, More than half the days, Nearly every day) how often they have been bothered by anxiety related problems over the past two weeks. The GAD-7 has been validated in those with epilepsy (Seo et al., 2014) and has been used in individuals with PNES (Chen et al., 2014).

*5.2.6.3 Depression*

The Neurological Disorders Depression Inventory for Epilepsy (NDDI-E) is a six-item scale measuring major depression in people with seizures. Participants are asked to report on a four-item Likert scale (Always or Often, Sometimes, Rarely, Never) how best depression related statements describes them over the last two weeks. Scoring above the cutoff of 15 suggests a major depressive episode. The NDDI-E has been validated in those with epilepsy (Cole, 2006) and PNES (Williams and Bagary, 2012)

*5.2.6.4 Illness perception*

The Brief Illness Perception Questionnaire (B-IPQ) is a nine-item scale assessing individuals’ cognitive and emotional representation of illness. Participants are asked to answer questions regarding: consequences, time-line, personal control, treatment control, symptoms, concern, understanding and emotional representation using a ten-item Likert scale. As recommended by the scoring instructions, the relevant items were reverse-coded and the eight items were added to compute a total score representing illness-threat. A higher score represents a more threatening view of the condition. The B-IPQ has been used in patients with epilepsy (Hackett et al., 2011) and PNES (Novakova et al., 2015).

*5.2.6.5 HRQoL*

The NEWQOL-6D (Mulhern et al., 2012)is a six-item HRQoL measure specifically developed for individuals with seizures. Participants are asked questions across six domains: worry about attacks, depression, memory, concentration, control of events, and stigma. Each question has an option of four possible responses. A higher score represents better HRQoL (possible range 0.96 – 0.34).

*5.2.6.6 Seizure frequency and severity*

TheLiverpool Seizure Severity Scale (LSSS-3) is a twelve-item self-report questionnaire asking individuals about their experiences of seizure frequency in the past year, and the severity of their seizures in the last four weeks. It is scored from 0–100 with higher scores reflecting greater seizure severity. The LSSS has been shown to have good internal consistency (*α*=0.72 - 0.96) in those with epilepsy (Baker et al., 1998) and has been used in participants with PNES (Whitehead et al., 2013).

*5.2.6.7 Writing task questionnaire*

Following each writing session, individuals were asked to rate on a one (Not at all) to seven point (A lot) scale, to what extent their writing was personal, meaningful, revealing of their emotions and thoughts, their desire to disclosure the information they have written about and if they have previously disclosed it, the positive effects of writing, felt-distress, and if they feel better after writing. This measure has become a standardised questionnaire in FEW studies (McGuire et al., 2005b; Stanton et al., 2002). Individuals were also provided with a space if they wanted to write about their feelings or thoughts about the writing session. This information was used to explore the acceptability of a writing intervention.

*5.2.7 Data analysis*

A series of two-tailed independent *t*-tests for continuous data and chi-square tests for categorical data was conducted to investigate between group differences. A series of independent *t*-tests were used to compare differences on the time-spent writing, and the items from the Writing Task Questionnaire between individuals in the therapeutic vs. control writing group. Qualitative feedback from the Writing Task Questionnaire was grouped into themes for the therapeutic and control writing group.

To explore the potential effectiveness of therapeutic writing, a mixed ANOVA analysis was conduced in relation to the outcome measures (HRQoL, GAD-7, NDDI-E and B-IPQ) with time of assessment (baseline, one- and three-month follow-up) being the within subjects factors and writing condition (therapeutic writing vs. control writing) being the between subjects factor. A series of paired-samples *t-*test were performed to explore differences within each writing condition. To interpret effect sizes, Cohen’s *d* was used with values of ≤0.2 considered reflecting a small, ≤0.5 a medium and ≥0.8 a large effect size(Cohen, 1988). A two-sided significance level of 0.05 was adopted for all statistical analyses reported. For the sample size calculation to help guide a future definitive effectiveness study, G\*Power was used. This adhered to a repeated measures ANOVA design using a within (therapeutic vs. control writing) and between (baseline, one- and three-month follow up) research method. SPSS 23 was used for all other statistical analyses.

**5.3 Results**

*5.3.1 Recruitment and attrition rates*

Overall, 131 individuals completed the baseline measures and were randomised to either the therapeutic or control writing group (Figure 5.2). More individuals were allocated to the therapeutic than the control writing condition; however, this difference was not significant (X2=1.7, p=0.2). A greater proportion of individuals were recruited from membership-led organisation (X2=16.9, p<0.001).

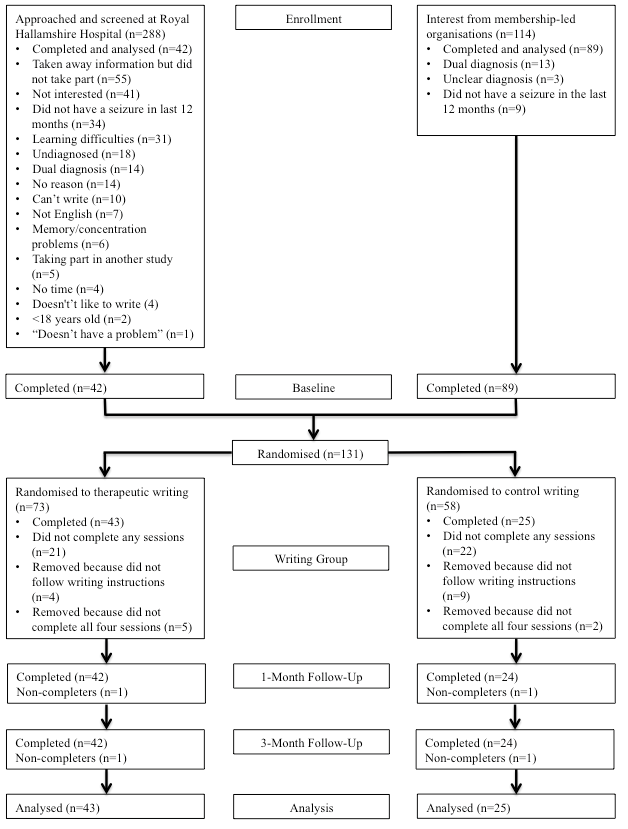


Figure 5.2 CONSORT flow diagram

*5.3.2 Randomisation*

A series of independent-samples *t*-tests or chi-square analyses of the baseline measures for individuals recruited to the therapeutic (n=43) or control writing condition (n=25) demonstrated that participants were closely matched on all of the demographic, condition and psychological variables investigated (Table 5.2). Effect sizes were calculated for the non-dichotomous variables demonstrating that, any differences between the two conditions were approximately small or less.

Table 5.2 Baseline demographic, condition and psychological factors of individuals randomised to the therapeutic- and control writing condition. Means, (standard deviations) unless otherwise stated.

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| Characteristic | Therapeutic writing | Control writing | *p* value | ES |
| Number of participants | 63.3% | 36.7% | 0.29 |  |
| Recruitment method |  |  | 0.38 |  |
| Seizure clinics | 27.9% | 16% |  |  |
| Membership-led organisation | 72.1% | 84% |  |  |
| Demographics |  |  |  |  |
| Age (years) | 41.7 (14.7) | 41.6 (15.4) | 0.97 | 0.009 |
| Education (years) | 16.9 (9.2) | 15 (4.3) | 0.42 | 0.22 |
| Gender |  |  | 0.75 |  |
| Male | 20.9% | 16% |  |  |
| Female | 80.1% | 84% |  |  |
| Condition factors |  |  |  |  |
| Diagnosis |  |  | 1 |  |
| Epilepsy | 62.8% | 60% |  |  |
| PNES | 37.2% | 40% |  |  |
| Duration of condition (years) | 16.9 (15.8) | 17.8 (18.5) | 0.83 | -0.05 |
| Seizures frequency in the last 4 weeks | 30.2 (73.1) | 36.5 (119) | 0.79 | -0.07 |
| Seizure severity | 56.4 (15.2) | 51.3 (16.4) | 0.23 | 0.32 |
| Psychological factors |  |  |  |  |
| NEWQOL-6D | 0.69 (0.13) | 0.71 (0.13) | 0.52 | -0.16 |
| GAD-7 | 8.7 (6.7) | 9 (6.2) | 0.82 | -0.06 |
| None (0-4) | 39.5% | 28% |  |  |
| Mild (5-9) | 20.9% | 40% |  |  |
| Moderate (10-14) | 16.3% | 8% |  |  |
| Severe (>15) | 23.3% | 24% |  |  |
| NDDI-E | 15.6 (4.8) | 16.7 (4.6) | 0.37 | -0.22 |
| <15 cut-off | 48.8% | 24% |  |  |
| >15 cut-off | 51.2% | 76% |  |  |
| B-IPQ threat | 48.4 (13.7) | 50.7 (9.9) | 0.46 | -0.3 |

PNES = Psychogenic Nonepileptic Seizures, NEWQOL-6D = Health Related Quality of Life Measure, GAD-7 = Generalised Anxiety Disorder, NDDI-E = Neurological Disorders Depression Inventory for Epilepsy, B-IPQ = Brief-Illness Perception Questionnaire, ES = Effect Size.

*5.3.3 Baseline measures of individuals with epilepsy vs. PNES*

A series of independent samples *t*-test or chi square analyses were conducted on the baseline measures between the two diagnoses (epilepsy and PNES) to explore whether it was legitimate to combine these groups in the analysis. This demonstrated that there were few significant differences: compared to those with epilepsy (n=42), participants with PNES (n=26) had experienced their condition for a shorter duration, scored higher on the subscales of depression, and perceived their condition as more threatening (Table 5.3).

Table 5.3 Baseline demographic, condition and psychological factors of individuals with epilepsy vs. PNES. Means, (standard deviations) unless otherwise stated.

|  |  |  |  |
| --- | --- | --- | --- |
| Characteristic | Epilepsy | PNES | *p* value |
| Number of participants | 61.8% | 38.2% | 0.052 |
| Writing condition |  |  | 1 |
| Therapeutic writing | 64.3% | 61.3% |  |
| Control writing | 35.7% | 38.7% |  |
| Recruitment method |  |  | 0.57 |
| Seizure clinics | 26.2% | 19.2% |  |
| Membership-led organisation | 32.8% | 80.8% |  |
| Demographics |  |  |  |
| Age (years) | 43.8 (15.3) | 38.2 (13.8) | 0.13 |
| Education (years) | 17.2 (9.7) | 14.7 (2.9) | 0.25 |
| Gender |  |  | 0.34 |
| Male | 23.8% | 11.5% |  |
| Female | 76.2% | 88.5% |  |
| Condition factors |  |  |  |
| Duration of condition (years) | 24 (17.4) | 6.4 (7.3) | <0.001 |
| Seizures frequency in the last 4 weeks | 27.6 (99.7) | 40.9 (78.9) | 0.57 |
| Seizure severity | 54.6 (17.4) | 54.3 (13.3) | 0.93 |
| Psychological factors |  |  |  |
| NEWQOL-6D | 0.72 (0.12) | 0.66 (0.14) | 0.06 |
| GAD-7 | 7.7 (5.8) | 10.7 (7.2) | 0.06 |
| None (0-4) | 42.8% | 23% |  |
| Mild (5-9) | 21.4% | 38.5% |  |
| Moderate (10-14) | 21.4% | 0% |  |
| Severe (>15) | 14.3% | 38.5% |  |
| NDDI-E | 14.7 (4.6) | 18.2 (4) | 0.002 |
| <15 cut-off | 50% | 23% |  |
| >15 cut-off | 50% | 77% |  |
| B-IPQ threat | 46.3 (11.9) | 54 (11.9) | 0.01 |

PNES = Psychogenic Nonepileptic Seizures, NEWQOL-6D = Health Related Quality of Life Measure, GAD-7 = Generalised Anxiety Disorder, NDDI-E = Neurological Disorders Depression Inventory for Epilepsy, B-IPQ = Brief-Illness Perception Questionnaire.

*5.3.4 Baseline measures of seizure clinic vs. membership-led organisation recruits*

As another control measure, differences in baseline measures were analysed between individuals recruited from seizure clinics (n=16) and membership-led organisations (n=52). The analyses demonstrated that a greater proportion of individuals were recruited from membership-led organisations, however, no other significant differences were observed (Table 5.4).

Table 5.4 Baseline demographic, condition and psychological factors of seizure clinic vs. membership-led organisation recruits. Means, (standard deviations) unless otherwise stated.

|  |  |  |  |
| --- | --- | --- | --- |
| Characteristic | Seizure clinics | Membership-led organisations | *p* value |
| Number of participants | 23.5% | 76.5% | <0.001 |
| Writing condition |  |  | 0.38 |
| Therapeutic writing | 75% | 59.6% |  |
| Control writing | 25% | 40.4% |  |
| Demographics |  |  |  |
| Age (years) | 44.8 (13.3) | 40.7 (15.3) | 0.34 |
| Education (years) | 14.7 (2.7) | 16.7 (8.9) | 0.42 |
| Gender |  |  | 0.72 |
| Male | 12.5% | 21.2% |  |
| Female | 87.5% | 78.8% |  |
| Condition factors |  |  |  |
| Diagnosis |  |  | 0.57 |
| Epilepsy | 68.8% | 59.6% |  |
| PNES | 31.2% | 40.4% |  |
| Duration of condition (years) | 22.5 (15.4) | 15.8 (17) | 0.18 |
| Seizures frequency in the last 4 weeks | 25.44 (61) | 34.8 (100.3) | 0.73 |
| Seizure severity | 53.3 (17.2) | 54.8 (15.5) | 0.76 |
| Psychological factors |  |  |  |
| NEWQOL-6D | 0.68 (0.15) | 0.7 (0.12) | 0.59 |
| GAD-7 | 7.9 (7.5) | 9.1 (6.1) | 0.51 |
| None (0-4) | 50% | 30.8% |  |
| Mild (5-9) | 18.8% | 30.8% |  |
| Moderate (10-14) | 6.2% | 15.4% |  |
| Severe (>15) | 25% | 23% |  |
| NDDI-E | 14.9 (5.5) | 16.3 (4.4) | 0.28 |
| <15 cut-off | 50% | 36.5% |  |
| >15 cut-off | 50% | 63.5% |  |
| B-IPQ threat | 48.6 (10.5) | 49.5 (13) | 0.8 |

PNES = Psychogenic Nonepileptic Seizures, NEWQOL-6D = Health Related Quality of Life Measure, GAD-7 = Generalised Anxiety Disorder, NDDI-E = Neurological Disorders Depression Inventory for Epilepsy, B-IPQ = Brief-Illness Perception Questionnaire.

*5.3.5 Completers vs. non-completers*

Overall, 48% of the 131 randomised individuals either dropped out (n=43) or were removed because they did not follow the writing instructions (n=22). There was no significant difference in the number of participants in the two writing arms who completed the writing intervention (n=68) and who did not (n=63). However, those who completed the writing intervention reported having experienced seizures for longer and having less severe seizures (Table 5.5). Compared to the control writing group, a greater number of completers were recruited from membership-led organisations than seizure clinics in the therapeutic writing condition. While not significant, compared to non-completers, a greater proportion of individuals completed the therapeutic than control writing group.

Table 5.5 Baseline demographic, condition and psychological factors of completers and non-completers. Means, (standard deviations) unless otherwise stated.

|  |  |  |  |
| --- | --- | --- | --- |
| Characteristic | Completers | Non-completers | *p* value |
| Number of participants | 51.9% | 48.1% | 0.67 |
| Writing condition |  |  | 0.08 |
| Therapeutic writing | 63.2% | 47.6% |  |
| Control writing | 36.8% | 52.4% |  |
| Recruitment method |  |  | 0.04 |
| Seizure clinics | 23.5% | 43.1% |  |
| Membership-led organisation | 76.5% | 56.9% |  |
| Demographics |  |  |  |
| Age (years) | 41.6 (14.9) | 37.9 (12.9) | 0.13 |
| Education (years) | 16.2 (7.9) | 15.5 (3.4) | 0.59 |
| Gender |  |  | 0.22 |
| Male | 19.1% | 28.6% |  |
| Female | 80.9% | 71.4% |  |
| Condition factors |  |  |  |
| Diagnosis |  |  | 0.59 |
| Epilepsy | 61.8% | 66.7% |  |
| PNES | 38.2% | 33.3% |  |
| Duration of condition (years) | 17.3 (16.8) | 12 (10.8) | 0.04 |
| Seizures frequency in the last 4 weeks | 32.6 (92) | 23.5 (50.3) | 0.5 |
| Seizure severity | 54.5 (15.7) | 61.3 (16.1) | 0.03 |
| Psychological factors |  |  |  |
| NEWQOL-6D | 0.69 (0.13) | 0.7 (0.15) | 0.82 |
| GAD-7 | 8.8 (6.5) | 10.2 (6.5) | 0.21 |
| None (0-4) | 35.4% | 22.2% |  |
| Mild (5-9) | 27.9% | 28.6% |  |
| Moderate (10-14) | 13.2% | 22.2% |  |
| Severe (>15) | 23.5% | 27% |  |
| NDDI-E | 16 (4.7) | 16.6 (6.5) | 0.6 |
| <15 cut-off | 29.7% | 33.3% |  |
| >15 cut-off | 60.3% | 66.7% |  |
| B-IPQ threat | 49.3 (12.4) | 49.6 (11.8) | 0.87 |

PNES = Psychogenic Nonepileptic Seizures, NEWQOL-6D = Health Related Quality of Life Measure, GAD-7 = Generalised Anxiety Disorder, NDDI-E = Neurological Disorders Depression Inventory for Epilepsy, B-IPQ = Brief-Illness Perception Questionnaire.

*5.3.6 Acceptability*

In the two arms of the trial, there was no difference in the number of individuals who choose to type (n=33) rather than handwrite (n=25) their written accounts (X2=1.1, p=0.3). No individual explicitly reported having concerns about returning their writings. Although participants were asked to write for at least 20 minutes per question, the self-reported time taken for all of the sessions was greater. An independent samples *t*-test revealed that the two arms of the trial did not differ on the time spent writing in any of the sessions (Figure 5.3).

Figure 5.3 Self-reported time spent writing in each session.

Overall, compared to individuals in the control writing group, those in the therapeutic writing condition rated their writing as more personal and meaningful, they also reported having engaged in greater emotional and cognitive expression. Although individuals in the therapeutic writing condition reported experiencing greater levels of distress, they perceived that writing had a greater positive effect and they felt better about themselves afterwards (Table 5.6).

Table 5.6 Writing Task Questionnaire. Means (standard deviation).

|  |  |  |  |
| --- | --- | --- | --- |
|  | Therapeutic writing | Control writing | *p* value |
| Session one |  |  |  |
| Personal | 6.7 (.67) | 5.9 (1.4) | 0.002 |
| Meaningful | 6 (1.2) | 4.8 (1.7) | 0.001 |
| Emotional expression | 6 (1.4) | 5.1 (1.35) | 0.01 |
| Cognitive expression | 6 (1.22) | 4.4 (1.4) | <0.001 |
| Desire to disclose | 5.5 (1.7) | 3.7 (2) | <0.001 |
| Actual disclosure | 4.4 (2) | 3.4 (2) | 0.5 |
| Positive effect | 4.9 (1.6) | 4.3 (1.6) | 0.11 |
| Distress | 3 (1.8) | 2.5 (1.9) | 0.19 |
| Feel better about yourself | 4.4 (1.7) | 3.5 (2) | 0.05 |
| Session two |  |  |  |
| Personal | 6.4 (0.9) | 5.8 (1.3) | 0.03 |
| Meaningful | 5.8 (1.3) | 4.9 (1.7) | 0.01 |
| Emotional expression | 6.3 (0.9) | 5.3 (1.7) | 0.003 |
| Cognitive expression | 6.4 (1) | 4.6 (1.8) | <0.001 |
| Desire to disclose | 4.8 (1.9) | 3.7 (2.1) | 0.03 |
| Actual disclosure | 3.6 (1.8) | 3 (1.5) | 0.17 |
| Positive effect | 5 (1.6) | 4.1 (1.9) | 0.03 |
| Distress | 3.4 (2) | 2.3 (1.5) | 0.02 |
| Feel better about yourself | 4.4 (1.8) | 3.7 (1.9) | 0.15 |
| Session three |  |  |  |
| Personal | 6.3 (1.9) | 5.3 (1.7) | 0.009 |
| Meaningful | 5.7 (1.4) | 4.2 (1.8) | 0.001 |
| Emotional expression | 6 (1.2) | 4.4 (2) | <0.001 |
| Cognitive expression | 5.8 (1.3) | 4.5 (1.9) | 0.001 |
| Desire to disclose | 4.4 (2.1) | 2.8 (2) | 0.003 |
| Actual disclosure | 3.7 (2) | 2.4 (1.4) | 0.008 |
| Positive effect | 5.2 (1.6) | 3.5 (2.1) | <0.001 |
| Distress | 3.4 (2.1) | 2.1 (1.6) | 0.01 |
| Feel better about yourself | 4.3 (1.9) | 3.2 (1.9) | 0.02 |
| Session four |  |  |  |
| Personal | 6.4 (0.9) | 5.1 (1.4) | <0.001 |
| Meaningful | 5.8 (1.5) | 4.3 (1.8) | <0.001 |
| Emotional expression | 5.7 (1.3) | 4.6 (2.1) | 0.01 |
| Cognitive expression | 6 (1.2) | 4.6 (1.7) | <0.001 |
| Desire to disclose | 4.4 (1.9) | 3.3 (2.2) | 0.03 |
| Actual disclosure | 3.9 (1.9) | 3 (1.7) | 0.07 |
| Positive effect | 5.2 (1.7) | 3.8 (2) | 0.008 |
| Distress | 3 (2) | 2.2 (1.4) | 0.06 |
| Feel better about yourself | 4.6 (2) | 3.6 (2.2) | 0.05 |
| Average over the four sessions |  |  |  |
| Personal | 6.5 (0.7) | 5.5 (1.3) | <0.001 |
| Meaningful | 5.8 (1.1) | 4.6 (1.3) | <0.001 |
| Emotional expression | 6 (0.8) | 4.9 (1.2) | <0.001 |
| Cognitive expression | 6 (0.9) | 4.5 (1.2) | <0.001 |
| Desire to disclose | 4.8 (1.4) | 3.4 (1.7) | 0.001 |
| Actual disclosure | 3.9 (1.5) | 3 (1.2) | 0.01 |
| Positive effect | 5.1 (1.3) | 3.9 (1.5) | 0.001 |
| Distress | 3.3 (1.3) | 2.3 (1.1) | 0.003 |
| Feel better about yourself | 4.4 (1.3) | 3.5 (1.7) | 0.02 |

In the therapeutic writing condition, 39/43 participants choose to describe how they thought their writing had gone. Individuals reported a range of benefits of therapeutic writing, such as helping them to vent their emotions and thoughts, engage in self-compassion, and improve their understanding or insight into a problem. There were a greater number of reported benefits than unhelpful effects. Individuals often reported wanting more time to write (Table 5.7).

Table 5.7 Reflections from individuals in the therapeutic writing condition

|  |  |
| --- | --- |
| Theme | Illustrative quote |
| **Beneficial effects** | |
| Cathartic/Dump/  Vent | *“I feel a relief when I am writing what I think and it makes me feel better. I feel like I can sleep I have got a lot off my chest.”* |
| Expression of emotions and thoughts | *“I was able to say a lot (not everything) about things troubling me. More personal things I haven’t told anyone.”* |
| Feeling better | *“I never thought of doing that before but it was a great idea-it made me feel really good! Thanks”* |
| Distance | *“I felt really frightened writing it down. It looks better on paper than it did in my head.”* |
| Self-Appreciation/ Compassion/ Affirmation | *“Nurturing myself through the writing, eased up my spirit, some. I forget too, that I need to love myself. I don't have to be perfect to love me.”* |
| Increased Insight/  Enlightening/ Reflection | *“Wow! They should use this idea for depression if they don’t already. I went so deep into my thoughts and feelings and memories and it made everything seem a lot more positive compared to what they were when I was younger. It stirred up some guilt though and it was hard to write just about the epilepsy but overall it’s made me feel better about life in general.”* |
| Empowering | *“It was empowering to see how far I had come in my thoughts and the fact that if I had written this same letter 15 years ago it would have been very dark, but now it is much lighter.”* |
| Be honest and open | *“Think my writing has shown me that I need to be honest and admit why I am struggling.”* |
| Easier than talking | *“I found it easier to talk away about my true feelings”* |
| Self-guidance | *“It has made me realise that I still have a lot in my life to change.”* |
| Regulate emotions | *“I have had a terrible stressful day today and so wasn’t looking forward to writing whilst feeling angry. I now feel much less annoyed at the situation I was put in today.”* |
| Therapeutic | *“Although once or twice I came close to tears, it was actually really therapeutic to get everything out.”* |
| Promote for sharing | *“I'd consider letting my wife, doctors, parent read these writing as I feel it could possibly reveal what it's like for me better than me walking to them.****”*** |
| **Unhelpful effects** | |
| Overpowering | *“It was like opening up a can of worms but I was trying to write about just one thing.”* |
| Looking back | *“I feel sad that I was once so happy and now so miserable.”* |
| Upsetting | *“It made me a little upset (a few tears sneaked out) talking about driving and the things I can’t do.”* |
| Difficult | *“I had lots of things I wanted to say but when found I couldn’t say them all when the pen was in my hand.”* |
| Limited | *“Writing may help me if I can put my written thoughts in front of the doctors. They may actually read it and think about it themselves.”* |

In the control writing condition, 24/25 participants choose to reflect on the writing sessions (Table 5.8). While individuals did report some benefits, the majority of comments reported that the writing was of little benefit or unhelpful. For example, participants described that they found it difficult to remember past events or concentrate on the task, also when describing future events, individuals reported feeling disappointed that their life is “boring”. With the expectation of ‘planning’, individuals in the therapeutic condition reported all of the beneficial effects.

Table 5.8 Reflections from individuals in the control writing condition

|  |  |
| --- | --- |
| Theme | Illustrative quote |
| **Beneficial effects** |  |
| Relaxing | *“It was very quieting, peaceful and relaxing. It made me focus in on something and stay focused.”* |
| Dumping | *“I do see a connection with writing and releasing some of my stress. I really don’t have friends that listen, I am the listener.”* |
| Planning | *“Listing a plan is how I do plans of my days generally. I feel that If I write things down, I may not refer to the list, but I have an image of the list to keep to or I can refer back to it.”* |
| Positive reminisce | *“It showed me that I did a lot for feeling worn out. And that I had many moments of joy playing with my niece and nephew and hearing them laugh. That was probably the best part of my day.”* |
| **Unhelpful effects** |  |
| Not much to write | *“I don't think I realised just how bored and cynical I've become since I ceased to be a student and was just left as unemployed.”* |
| Realisation | *“The writing has helped me, but still makes me realise how different and boring my life is, I don’t do much others would.”* |
| Going over failure | *“The only emotion evoked by the writing was a bitter disappointment that I consumed alcohol over the weekend”* |
| Emotionless | *“The writing wasn't very emotionally stimulating.”* |
| Too rigid | *“I did not find this writing helpful because it is too specific.”* |
| Difficult | *“Memory is a hardship with epilepsy, so a whole weeks events starting 5 days previous is very hard to remember”* |
| Anxiety | *“Feel very anxious and writing what I am about to do has heightened in my head the effects of what my day will bring.”* |
| Concentration | *“My head hurts with focusing now though”* |

*5.3.7 Preliminary evaluation of impact on psychological outcomes*

The exploratory analysis of outcome measures as a function of time (baseline, one- and three-month follow-up) using a series of paired samples *t*-tests in each of the conditions (therapeutic and control writing) revealed that those in the therapeutic writing condition exhibited a significantly greater HRQoL at one-month follow-up *t*(38)=-2.39, p=0.02, effect size=0.65. However, a 2 x 3 mixed ANOVA analysis demonstrated that no significant differences (p>0.05) were found between the two writing conditions as a function of time on any of the outcome measures (Table 5.9).

Table 5.9 Effects of therapeutic vs. control writing condition one- and three-month follow up.

|  |  |  |  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| Outcome | Group | Baseline | | 1-month | | 3-month | | Interaction time x condition | | | | | |
|  |  | Mean | SD | Mean | SD | Mean | SD | F (df) | Partial Eta | *p* | ES | |
| GAD-7 | Therapeutic | 8.6 | 6.8 | 8.4 | 6 | 8.1 | 6.5 | 0.61 (2,122) | 0.01 | 0.55 | | 0.15 | |
|  | Control | 9 | 6.2 | 7.6 | 4.8 | 8.2 | 5 |  |  |  | |  | |
| NDDI-E | Therapeutic | 15.7 | 4.9 | 15.4 | 4.8 | 15 | 5.1 | 0.04 (2,118) | 0.001 | 0.96 | | 0.06 | |
|  | Control | 16.6 | 4.5 | 16.7 | 4.6 | 16.1 | 4.4 |  |  |  | |  | |
| B-IPQ | Therapeutic | 48.5 | 14.2 | 48.5 | 11.4 | 46.9 | 12.2 | 0.09 (2,120) | 0.001 | 0.92 | | 0.06 | |
|  | Control | 50 | 9.8 | 47 | 10.5 | 49.2 | 10 |  |  |  | |  | |
| NEWQOL-6D | Therapeutic | 0.7 | 0.13 | 0.73 | 0.1 | 0.72 | 0.13 | 0.69 (2,118) | 0.03 | 0.5 | | 0.16 | |
|  | Control | 0.71 | 0.12 | 0.73 | 0.1 | 0.71 | 0.1 |  |  |  | |  | |

NEWQOL-6D = Health Related Quality of Life Measure, GAD-7 = Generalised Anxiety Disorder, NDDI-E = Neurological Disorders Depression Inventory for Epilepsy, B-IPQ = Brief-Illness Perception Questionnaire, SD = Standard Deviation, ES = Effect Size, df = Degrees of Freedom

*5.3.8 Sample size calculation for future randomised controlled trials*

Based on the current dataset, a sample size calculation was performed. With an average effect size of 0.11, accepting the alpha level of 0.05, detecting a large effect size (≥0.8), a correlation of 0.53 between the outcome measures at baseline, and a nonsphericity correction of 1 (in the current study Mauchly’s test of sphericity was >0.05, therefore the assumptions of sphericity had been met), a total sample of 128 is required (n = 64 in each writing condition). However, given the reported attrition of 48%, we would predict that the total number of participants who would need be randomised would be at least n = 256.

**5.4 Discussion**

To our knowledge, this single blinded randomised controlled trial is the first to date investigating a therapeutic writing intervention for individuals living with a seizure disorder, specifically epilepsy or PNES. Our results are consistent with previous research suggesting that therapeutic writing is associated with a range of qualitative and quantitative benefits in patients with a clinical diagnosis. The dataset revealed that HRQoL improved significantly one-month after completing the writing sessions in those who were randomised to the therapeutic writing condition (a medium effect size was observed), but not the control group. One of the key aims (and proposed therapeutic processes) of FEW is moving individuals to explore and focus on emotionally charged topics. As such, the intervention was successful as compared to those in the control writing condition, participants in the therapeutic writing condition self-rated their writing as more personal and meaningful, and reported having engaged in greater emotional and cognitive expression.

In terms of feasibility, 14.6% of the potentially suitable patients attending a hospital outpatient seizure clinic who were approached agreed to participate and complete the baseline measures. We are unable to state the number of individuals that were approached by membership-led organisations, however, over two thirds of participants in the current study were recruited in this way. These findings are comparable with other studies reporting rates of attrition during FEW interventions in other clinical populations. For example, in a study investigating the feasibility of FEW in adults with type 2 diabetes who were recruited from local general practitioner’s register and online diabetes support groups, 93.5% of 1,715 individuals who were approached stated that they were ‘not interested’; furthermore, 34.1% of the 41 individuals who were randomised dropped out (Dennick et al., 2015).

It is important to note that, 20 individuals did not adhere to the writing instructions (e.g. they did not complete all four writing sessions or participants in the control condition wrote about their emotions and thoughts). The aim of a pilot study is to explore potential barriers towards adherence, prior to conducting a definitive trial. The findings suggest that individuals need clearer instructions, for example, those in the control condition could be explicitly informed to, “not write about your emotions and thoughts, but instead focus specifically on your behaviour and actions”. Additionally, approaches, other than the implementation intention questionnaire, could be used to help participants to complete all four sessions. For instance, a researcher could contact participants to remind them that they are due to begin a writing session. In the current study, individuals who did not comply with the instructions were removed from the analysis as to not bias the estimates. An intent-to-treat analysis could have otherwise been conducted, this would have ignored factors such as, non-compliance or non-adherence to the protocol (Gupta, 2011). However, as this study was not primarily concerned with determining the efficacy of the treatment, this measure was not taken.

Another observation important in terms of the feasibility and acceptability of the writing conditions used in this study is that individuals in neither group scored significantly higher on any clinical measure (HRQoL, anxiety, depression and illness perception) during follow-up periods than at baseline. Without therapist feedback or supervision, participants could have focused on narratives causing them to feel worse about themselves and their seizure disorder. Our findings suggest that it is safe to administer this sort of home-based writing intervention in this population.

Despite the relatively low recruitment and rate, our findings suggest that therapeutic writing is acceptable to a sizable subgroup of individuals with a seizure disorder. Having said that, it should be noted that participants were informed that another purpose of the current study was that their writings were going to be read to give researchers a deeper understanding of what it is like to live with their seizure disorder. This may not only have influenced what individuals wrote about and how much they decided to disclose, but a proportion of individuals may have been motivated to take part by the opportunity to share their story of illness or to help increase the awareness of their seizure condition. We are unable to control for this, as we did not ask participants their motive for wanting to take part in the current research.

It was interesting that, although individuals were asked to write for at least 20 minutes, the average self-reported time was greater across all sessions. As individuals wrote at home and in private, the risk of any demand characteristics was likely to be minimal. In the Writing Task Questionnaire, compared to the control group, participants in the therapeutic writing condition reported a greater desire to disclose the information that they had written about. This would suggest that there is an unmet treatment need in this population. Indeed, it has been suggested that individuals who experience shame or stigma associated with what they choose to write about are more likely to gain from expressive writing, and there is evidence that many patients with epilepsy or PNES feel stigmatised because of their condition (Jacoby and Austin, 2007; Rawlings et al., 2017a). The provision of an opportunity to write has been used by support groups in other areas, for instance groups for individuals with cancer (Shaw et al., 2006). This form of support could also be beneficial for individuals living with a seizure disorder.

Surprisingly, some individuals in the control condition qualitatively reported that writing about their daily events was beneficial. Individuals with epilepsy or PNES often complain of memory problems (Ponds and Hendriks, 2006; Samarasekera et al., 2015) and writing things down, such as using a diary, has been suggested as being helpful. What is more, mindfulness is a psychological practice that aims to bring ones’ awareness into the present non-judgmentally. This can be achieved in a range of everyday activities including showering, walking and writing. Mindfulness interventions have been associated with reduced depressive and anxiety symptoms, and seizure frequency in those with epilepsy (Tang et al., 2015) and PNES (Baslet et al., 2015). It is therefore possible that writing could be therapeutic even if it is not about emotionally challenging topics.

The qualitative feedback describing the perceived benefits of therapeutic writing mirrored the potential modes of action of FEW suggested previously, such as emotional disclosure, development of a coherent narrative and self-affirmation (Niles et al., 2015). One potential use of therapeutic writing is its use alongside other treatments. Indeed, Graf et al. (2008) combined psychotherapy with expressive writing as homework suggesting that, compared to those in the control writing condition, patients were more open and talkative during sessions, and demonstrated greater insight into their problems. Both patients and therapists found expressive writing contributed to the process and outcome of therapy.

It is easy to see how therapeutic writing could be a useful addition to the range of approaches that therapists use to help individuals with seizure disorders. For example, the written content could be further explored in therapy, help individuals become familiar with expressing their emotions and thoughts in private before they are asked to do it with the therapist, and as a tool to understand their perception and ability to express. In view of the relatively short-lasting effects of therapeutic writing observed in this study (the improvement in HRQoL was not consolidated at the three-month follow up), it is difficult to justify a large scale randomised controlled trial of this intervention in this population. It may be more fruitful to investigate therapeutic writing as a supplement to other approaches that could build on the reported benefits of therapeutic writing that was observed in the current study.

Although participants in the therapeutic writing condition reported a significant improvement in HRQoL, this was not the case when compared to those in the control writing group (i.e. there was no significant interaction between the two writing conditions on HRQoL). There are a number of potential reasons for this finding. As already discussed, the control writing condition may have been associated with therapeutic benefits and at least in this population, the control task chosen may not have been an ideal control condition although our therapeutic writing tasks achieved their aim in the sense that they got participants in this group to write about more emotionally challenging subjects. It is also conceivable that the outcomes measures (GAD-7, NDDI-E, B-IPQ) in the current study failed to capture key concepts of change. Based on individuals’ qualitative feedback, measurements investigating factors such as emotional processing and self-compassion may have been more sensitive.

Resonating with a large proportion of the previous literature investigating FEW in clinical populations, this study was a randomised controlled trial. A control group was intended to test whether therapeutic gains were due to the emotional processing element of writing or the act of writing itself, i.e. a placebo effect of engaging in a therapeutic process (Paudyal et al., 2014; Broderick et al., 2005). Our methodology is also in line with prior studies that chose to blind participants from the study’s objective and randomisation. For example, when investigating the effectiveness of a FEW intervention on health outcomes in 68 patients with ankylosing spondylitis, Hamilton-West and Quine explain that: “*Participants were not informed that the study involved allocation to an intervention or control group, and the randomisation schedule was known only to one researcher (K.E.H.-W.), who did not meet the participants at any stage*” (pg. 644) (Hamilton-West and Quine, 2007). Unfortunately, the PhD student researcher (G.R.) who contacted participants, reviewed patients’ record and collected patients’ writings could not realistically remain blinded in the present project. The design and methodology of the current study was granted ethical approval by a multidisciplinary ethics committee. While the ethics committee was explicitly informed that participants would be randomised in this study, the randomisation process was concealed from patients to ensure that the study was single-blinded. Blinding is a key element in randomised controlled trials as it can help to minimise bias, particularly performance- and ascertainment-bias. If individuals are not blinded, having the awareness that there are different conditions under investigation or group assignment (therapeutic or control) may potentially influence their behaviour (Karanicolas et al., 2010). There are of course ethical implications that need to be taken into consideration when determining whether concealing the aims of a study and methodology from participants are just, for example, if it means withholding treatment (Blader, 2005). In the case of this study, the therapeutic topics were based on psychological theory, but there was no compelling prior evidence that the “therapeutic” topics would actually be more beneficial or less harmful than the control topics. Nevertheless, other approaches could have been used, for instance implementing an ABA or ABBA design.

*5.4.1 Limitations*

A limitation of this study is the potential selection bias i.e. that only participants able and willing to write will have agreed to take part in a study examining a therapeutic writing intervention. Although our intervention provided individuals with the option of typing, which may have helped with some motor problems (i.e. inability to hold a pen), we were unable to make any provisions for individuals with many disabilities, including those with learning difficulties. Although different modes of expression could be used instead of (or in addition to) writing i.e. talking, dance or art, such methods could not have been compared directly with writing and would be more difficult to standardise and analyse.

Whilst we collected a range of clinical and demographic data to characterise our patient groups and made sure that all participants completed the four writing sessions and adhered to the instructions, we did not investigate potential moderating factors for the content of writing. In other writing studies variables such as word count, self-affirmation, affect and narrative structure have been investigated. For example, Niles et al. (2015) analysed 200 written accounts from a FEW study involving 50 healthy participants and found that a very high negative emotional word use in the writings was associated with higher anxiety scores three-months later, and that, conversely, those who engaged in greater self-affirmation and level of detail in their writings reported lower levels of anxiety at follow-up. Future research is needed to investigate the mechanisms of change in expressive writing, and also whether the effectiveness of therapeutic writing could be maximised by providing participants with guidance (and possibly feedback) on their writings.

The heterogeneity of our sample must be acknowledged. As this was a pilot study we only recruited a modest sample size. As such, we are unable to distinguish between the different diagnoses (epilepsy vs. PNES) a well as the different subgroups of epilepsy or PNES. For example, Brown et al. (Brown et al., 2013) suggests that patients with PNES can be subdivided into at least two subgroups based on emotional processing – one group demonstrating emotional dysregulation, while the other report normal emotional regulation but score high on symptoms of somatisation. This raises the possibility that improvements overall may well have been cancelled out or minimised in the statistical analyses by lack of improvements in other subgroups.

As we could not confirm the clinical diagnoses of all individuals who were recruited through membership-led organisation using medical records, there is the potential risk associated with diagnostic certainty. Having said that, this must be balanced against the fact that we have reported on a more varied group making our findings easier to generalise than if we only recruited patients attending hospital services - who may be are more likely to experience severe and refractory seizure disorders.

Randomisation did not result in the same number of individuals being allocated to the therapeutic (n=73) or control (n=58) writing conditions – although it should be noted that this difference was not significant to the alpha level. If a block randomisation method were implemented (i.e. in blocks of ten, five randomised to FEW and five to control), this would have made sure that the randomisation process yielded equally sized participant groups.

*5.4.2 Conclusion*

Therapeutic writing has been proposed as an inexpensive and easy to administer form of therapy in many medical and psychiatric disorders. This study investigated its feasibility, acceptability and potential effectiveness in individuals with epilepsy or PNES. The rates of attrition reported here are consistent with other studies investigating FEW in clinical samples. As such, therapeutic writing is acceptable in those with epilepsy or PNES. The evidence suggests that therapeutic writing (and to a lesser extent control writing) was associated with a range of self-reported benefits based on quantitative and qualitative outcomes. Individuals in the therapeutic, but not control, writing group demonstrated an improvement in HRQoL one-month following the intervention. At this stage, further research is needed into the role that therapeutic writing could play in the treatment of individuals with seizures.

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**Chapter Six**

**Written accounts of living with epilepsy: A thematic analysis**

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**Candidate’s individual contributions:**

G.H. Rawlings was involved in obtaining regulatory approval; methodological design and formulation of the research aims specific to this study; identifying, consenting and confirming the diagnoses of suitable participants; data collection; data analysis and interpretation; drafted, submitted and corrected the published manuscript. The candidate is the first author.

Rawlings, GH:

Signed:

Date: 19th May 2017

Brown, I:

Signed:

Date: 21/05/2017

Reuber, M:

Signed:

Date: 19th May 2017

**Chapter Seven**

**Written accounts of living with psychogenic nonepileptic seizures: A thematic analysis**

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**Seizure: European Journal of Epilepsy 2017; 50: 83-91**

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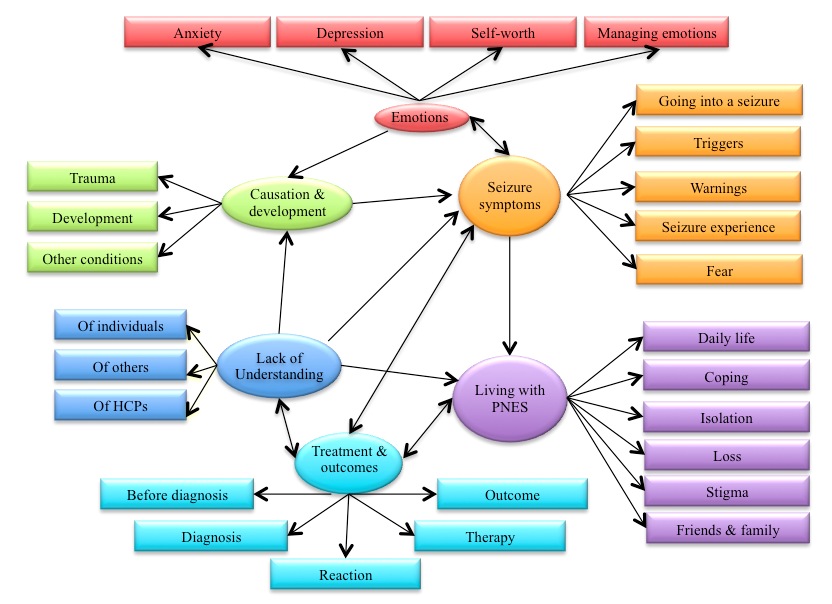


Figure 7.1 Supplement Thematic map showing six key themes (represented by circles) and 26 subthemes (represented by squares). Each colour represents a different theme. HCPs = Healthcare professionals

**Chapter Eight**

**Written accounts of living with epilepsy or psychogenic nonepileptic seizures: A thematic comparison**

**Abstract**

Psychogenic nonepileptic seizures (PNES) superficially resemble epileptic seizures, but are considered a dissociative response. This study examines the subjective experience of living with epilepsy or PNES by thematically comparing individuals’ written accounts of their condition. The current dataset was captured by asking participants to write: about their thoughts and feelings concerning their condition, a letter to their condition, a letter to their younger self, and about a personal value. The writings were thematically compared using a data-driven approach. Individuals with epilepsy and PNES were closely matched on age, gender, education, quality of life and seizure severity. Five key differences emerged between the two conditions. Theme 1: ‘Causation’ revealed those with PNES explored the possible causes of their condition throughout their narratives. Theme 2: ‘Emotions’ demonstrated individuals with epilepsy appeared emotionally stable, whereas the writings of those with PNES reflected anxiety and low mood. Theme 3: ‘Seizure symptoms’ showed differences in ictal experiences and how seizures were conceptualised. Theme 4: ‘Treatment’ explored the differences in the diagnostic journey and experiences of healthcare professionals. Theme 5: ‘Daily life’ revealed that individuals with epilepsy were keen to portray themselves as coping with their condition, whereas those with PNES felt their disorder had destroyed their lives. In epilepsy, the sequelae were conceptualised as something that must be fought, whereas those with PNES tended to describe their seizures as a place they enter. The results demonstrate that qualitative approaches can be used to provide insights into an individuals’ world, as well as offer clinical implications.

**8.1 Introduction**

The manifestations of psychogenic nonepileptic seizures (PNES) superficially resemble those of epileptic seizures. However, whereas epilepsy is a neurological condition characterised by excessive electrical discharges in the brain, PNES are best conceptualised as a dissociative response to potentially distressing stimuli (Brown and Reuber, 2016a; Brown and Reuber, 2016b). As such, PNES are classified as a mental disorder in the current medical nosologies (APA, 2013; WHO, 1992). PNES are not rare, as approximately 20% of patients referred to epilepsy clinics will have the condition (Angus-Leppan, 2008).

Qualitative methodologies have a particularly important role to play if we want to understand how epilepsy or PNES affect individual patients. Qualitative approaches capture and examine individuals’ own words, producing data that is fine-grained and rich in detail. What is more, researchers are able to ask general and open-ended questions. This gives respondents the ability to highlight those issues most relevant to them, and allows individuals to formulate and clarify their responses rather than having to reply by selecting from a limited list of pre-defined categorical statements. This enables readers to gain insights based on *what* individuals say as well as *how* they say it.

Over the last decade there has been an increase in the number of publications using qualitative methodologies to investigate individuals’ perception of living with epilepsy or PNES. Unlike quantitative studies, which have more commonly compared the mean responses of patients with PNES with those with epilepsy, most of this research has investigated these conditions individually. However, there are some exceptions. Using different forms of language analyses to investigate English, German and Italian speaking patients, a series of studies compared how individuals with epilepsy or PNES talk about their problems when they present to a healthcare professional. These studies revealed differences in how individuals’ conceptualise, name, talk about, and describe their seizures, which were clear enough to contribute to the clinical differentiation of epilepsy and PNES (Schwabe et al., 2007; Schwabe et al., 2008; Plug et al., 2009b; Plug et al., 2010; Monzoni et al., 2011; Plug et al., 2011; Cornaggia et al., 2012; Robson et al., 2012).

To date, qualitative research examining PNES has predominantly relied on research interviews or the audio- and video-recording of actual clinic conversations for data collection (Rawlings and Reuber, 2016). Whilst most qualitative studies investigating epilepsy have also utilised interview data, based on the notion that various methodological and analytical approaches are likely to produce different perspectives on a particular problem, other methods have also been used to explore people’s attitudes about living with epilepsy, such as poetry (Featherstone and Sandfield, 2013) and drawing (Stafstrom and Havlena, 2003; Featherstone et al., 2013).

Recently, the subjective experience of living with epilepsy (Rawlings et al., 2017b) or PNES (Rawlings et al., 2017c) has been thematically analysed by examining individuals’ written accounts of their condition. Compared to spoken responses, writing is seen as an individual act allowing for private reflection, exploration and expression of thoughts and feelings (Howlett, 2004). These analyses have helped further to illuminate how individuals are affected by and subsequently manage their condition, as well as to highlight implications for clinical practice. An important finding of this research was that there were clear differences in the emergent themes between individuals with epilepsy and those with PNES. Therefore, using the same datasets, the purpose of the current study was to investigate these differences further using a methodical approach. To this end, a thematic comparison analysis was used to compare and contrast individuals’ writing about their condition.

**8.2 Methods**

*8.2.1 Participants*

The data collection method for this study has been described previously (Rawlings et al., 2017b; Rawlings et al., 2017c). Participants were approached consecutively and recruited from outpatient neurology clinics at the Royal Hallamshire Hospital, Sheffield (United Kingdom). Individuals were also recruited through membership-led organisations for individuals experiencing seizures (see acknowledgements for the list of organisations). Recruitment took place between October 2015 and November 2016. The North of Scotland Research Ethics Committee granted ethical approval for this study (15/NS/0078). Participants were included in the present study if they were over the age of 18 years; had a diagnosis of epilepsy or PNES (individuals with dual-diagnosis were excluded), were able to provide informed consent and complete a demographic and clinical questionnaire without help. The diagnoses of all participants recruited at the Royal Hallamshire Hospital were confirmed by review of their hospital records. When possible, confirmation of the self-reported diagnoses of participants recruited through membership-led organisations was sought from their general practitioner.

*8.2.2 Data collection*

This dataset was collected in the context of a randomised control trial investigating the effects of an expressive writing intervention for individuals with seizure disorders. The current study is based exclusively on data from participants allocated to the *intervention* group. A total of 19 participants with PNES and 20 with epilepsy were included in the study. This is the number of individuals that had been recruited to the writing intervention when the current study was undertaken.

Participants recruited from outpatient neurology clinics were sent a participant information sheet at least 48 hours before their appointment with a Consultant Neurologist. On the day of their appointment, individuals were approached and invited to take part in the study. Those who gave written consent were asked to complete a set of self-report measures. Participants recruited from membership-led organisations replied to an advert for a study of a writing intervention for individuals with seizure disorders. Potential participants then contacted G.R. who gained written informed consent and provided access to an online form allowing participants to complete the self-report measures.

Participants were then given four writing booklets to complete. Each booklet contained writing instructions, space for writing (four A4 sheets of lined paper) and a link to a website if they preferred typing to handwriting. Participants were asked to produce four pieces of writing: 1) their very deepest thoughts and feelings about their condition (Pennebaker and Chung, 2010); 2) a letter to their condition (Howlett, 2004); 3) a letter to their younger self (Kress et al., 2008); and 4) about a personal value (McQueen and Klein, 2006). These topics had been set based on previous studies of writing therapies in other patient groups. Participants were asked to write for at least 20 minutes per question at home and in private.

*8.2.3 Measures*

Participants completed a demographic questionnaire that recorded their age, gender, employment status and years of education. Participants were also asked how long they had experienced seizures and the date of their last seizure. Participants’ health-related quality of life (HRQoL) was investigated using the NEWQOL-6D (Mulhern et al., 2012). This is a six-item HRQoL measure specifically developed for individuals with seizures. A higher score represents a better HRQoL (highest possible score 0.96 – 0.34 lowest possible score). The Generalised Anxiety Disorder (GAD-7) was used to measure anxiety (Spitzer et al., 2006). This is a seven-item scale used as a screening tool and severity measure of mild (score of 5-9), moderate (10-14) and severe anxiety (>15). The six-item Neurological Disorders Depression Inventory for Epilepsy (NDDI-E) was used to screen for likely major depression (Gilliam et al., 2006). A score >15 suggest a current major depressive episode. Seizure frequency and severity were investigated using the Liverpool Seizure Severity Scale questionnaire (LSSS-3) (Baker et al., 1998). This is scored from 0–100 with a higher score representing greater seizure severity.

*8.2.4 Data analysis*

The data was analysed in three main stages:

In the first stage, G.R. transcribed and read all writings to become familiar with the content of narratives. Participants’ answers to each of the four questions were read separately, but as individuals discussed similar events throughout and expanded on experiences in later sessions, it was decided that their writing on all four topics would be considered together for the analysis. G.R. aimed to work reflectively throughout, keeping a journal of his impressions and reflections specific to the diagnosis, as well as differences between the diagnoses.

In stage two, separate thematic analyses were conducted on participants’ writings about epilepsy and PNES (Table 8.1). The analyses were guided by the framework outlined by Braun and Clarke (Braun and Clarke, 2006). A mixed inductive (themes were grounded in the data) and theoretical approach (themes were influenced by the literature, primarily from a recent systematic synthesis of qualitative research into living with a seizure disorder (Rawlings and Reuber, 2016) was used. A coin flip determined that the writings from participants with epilepsy were analysed first, followed by the analysis of the writings from those with PNES. The findings that emerged from analysing the writings from individuals with epilepsy were not intentionally used to structure the analysis of the writings from participants with PNES.

Individuals were informed that a member of the research team would read their writing to gain a better understanding of what it is like to live with their condition, but that they would not be contacted about what they had written. While this meant that participants might have felt that they were able to be more honest and open in their writings, we were unable to achieve member checking. However, expert checking was performed as all the emergent themes were shared between the authors and revisions were made until a general consensus was achieved. M.R. and I.B. are clinicians currently involved in the care of individuals living with seizures. B.S. has extensive experience of analysing written accounts of living with a long-term condition. The thematic analyses have also both been peer-reviewed and published in international journals.

Table 8.1 Stages of thematic analysis.

|  |  |
| --- | --- |
| Stage | Action(s) |
| 1 | G.R. imported and extracted, into NVivo, initial codes. This was a timely and iterative process that involved having to go back through narratives to re-code as new codes emerged. |
| 2 | G.R. compared and collated codes to create main and sub-themes. |
| 3 | Reviewed the themes and codes to define sub-themes. It was at this stage that the themes were shared between the authors allowing for changes. Thematic saturation was not possible as participants were not directed in their narratives and so they could choose to write about anything. However, all narratives were read one final time to make sure no more themes emerged. |
| 4 | Further refinement of sub-themes, assigning clear titles and definitions. |
| 5 | Writing the report, making the explanation of themes and sub-themes coherent. |

In the third stage, any differences in sample characteristics between participants with epilepsy and PNES were examined using chi-square or Mann-Whitney U tests as appropriate. Next, the themes that emerged from analysing the writings from participants with epilepsy and PNES were examined and summarised using a data-driven approach. Key differences were defined, based on the relevance, prevalence and perceived importance of the data (Tang et al., 2009). Themes were shared between the authors allowing for changes.

Throughout this report, in the quotes used (“ ”) [Start] and [End] have been added to represent the beginning and conclusion of citations from narratives. The main themes are presented in the order in which the differences between the two conditions were observed, for example, theme one and two reflect differences in how individuals began their narratives.

**8.3 Results**

Participants were closely matched on age, gender, years of education, HRQoL, seizure severity and number of written words produced. Participants with epilepsy had experienced their seizures for longer and reported fewer seizures in the last four weeks. Participants with PNES had higher scores on self-reported measures of anxiety and depression (Table 8.2).

The thematic analysis in the epilepsy group yielded five key-themes reflecting experiences of (i) seizure onset, (ii) seizure symptoms, (iii) treatment and outcome, (iv) living with epilepsy, and (v) displays of coping [Rawlings et al., 2007b]. In the PNES group, six key-themes were identified: (i) living with PNES, (ii) emotions, (iii) seizure symptoms, (iv) treatment and outcome, (v) causation and development, and (vi) lack of understanding by themselves, others, and healthcare professionals (HCP) [Rawlings et al., Manuscript submitted] (Figure 8.1). The thematic comparison revealed five key differences in terms of (i) causation, (ii) emotions, (iii) seizure descriptions, (iv) treatment, and (v) daily-life. These differences have been summarised in Table 8.3.

Table 8.2 Mann-Whitney-U analysis and chi-square tests. Scores reflect median (Interquartile range) unless stated.

|  |  |  |  |
| --- | --- | --- | --- |
|  | Epilepsy | PNES | *p* value |
| N | 20 | 19 | 0.87 |
| Age | 52.5 (23.5) | 42 (24) | 0.07 |
| Gender (% of female) | 85% | 84.2% | 0.64 |
| Years of education | 14 (6.3) | 16 (5.5) | 0.55 |
| HRQoL | 0.77 (0.12) | 0.67 (0.18) | 0.07 |
| Seizure severity | 51.3 (33.1) | 52.5 (21.8) | 0.55 |
| Years since seizure onset | 25.5 (30.8) | 5 (5) | <0.001 |
| Seizure frequency (last 4 weeks) | 1 (4.8) | 20 (46) | <0.001 |
| Words produced | 1,762 (2,318) | 1,898 (1,964) | 0.55 |
| GAD-7 | 3.5 (5.8) | 9 (16) | 0.007 |
| None (0-4) | 60% | 26.3% |  |
| Mild (5-9) | 20% | 31.6% |  |
| Moderate (10-14) | 10% | 0% |  |
| Severe (>15) | 10% | 42.1% |  |
| NDDI-E | 12 (7.8) | 17.5 (7.5) | 0.001 |
| Score >15 | 25% | 66.7% |  |

PNES = Psychogenic Nonepileptic Seizures, HRQoL = Health Related Quality of Life, GAD-7 = Generalise Anxiety Disorder, NDDI-E = Neurological Disorder Depression for Epilepsy.

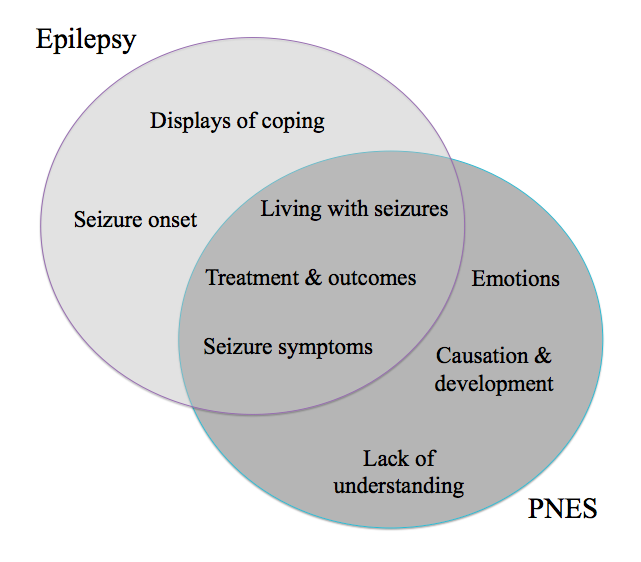


Figure 8.1 Comparative diagram highlighting overlapping and non-overlapping key themes from the thematic analyses investigating written accounts from individuals with epilepsy or psychogenic nonepileptic seizures (PNES).

Table 8.3 A summary of thematic differences between individuals with epilepsy and psychogenic nonepileptic seizures (PNES)

|  |  |  |
| --- | --- | --- |
|  | Epilepsy | PNES |
| 1. Causation | Seizures were described as coming on out of the blue. Participants rarely reported other conditions. Participants did not ruminate over the cause of their condition. | Reports of past trauma were common. Precipitating factors were evident, as participants would discuss past experiences of stress, chaos, and turbulence. Participants often reported other medical diagnoses, which were perceived as being connected to PNES. |
| 2. Emotions | Relatively stable. Mood changes (frustration, anger, depression) were discussed as being associated with their seizures or medication. | Narratives reflected low mood and anxiety - often this was chronic. Notable lack of self-worth and self-appreciation. Discussed problems with emotional processing. |
| 3. Seizure symptoms |  |  |
| 3.1 Description | Seizures were described as external and conceptualised as acting independently of the writer. Seizures would follow participants around, “*stalk*” and “*come on*” or “*hit*” them. Participants projected human characteristics onto their seizures. | Seizures were conceptualised as a state or place that participants enter. Seizures were not discussed as following participants around. Seizure experiences varied greatly. |
| 3.2 Focus | Participants’ narratives focused on living with epilepsy, and how it has interfered with their daily lives. Seizures were described as secondary to other factors. | Participants would focus on their post- and interictal symptoms. Problems associated with seizure events, as well as living with PNES. |
| 3.3 Postictal | Described in detail, some individuals explained that they could get back on with their day as normal. Seizure-related injuries were commonly discussed. | Described in detail, severe and disabling symptoms. Seizure-related injuries were rarely reported. |
| 3.4 Fear | Mainly anticipatory fear. | Anticipatory and ictal fear. It was clear that seizures were a difficult experience. |
| 4. Treatment |  |  |
| 4.1 Diagnosis | An important moment, but a relatively brief event that participants did not expand upon. For some, it changed their identity. | Participants resisted the diagnosis, looked for other diagnoses, asked for other tests. Long process. |
| 4.2 HCPs | A major source of support and seizure management. Trusting the medical doctors. | Participants felt let down, ostracised, unsupported, belittled, disbelieved, and did not trust HCPs. Passed onto different services, little validation even after diagnosis. Negative and at times damaging experiences. |
| 4.3 Treatment | Viewed as the main method of seizure control- albeit side effects were a major source of distress. | Cries for help, not sure of the best approach, skeptical of therapy. |
| 5. Daily-life |  |  |
| 5.1 Coping | Portrayed themselves as coping well and having integrated epilepsy into their lives. Strived to live a normal life. Accepted or resisted limitations posed by epilepsy. Downplayed their symptoms. Found positive gains. Used laughter. Promoted self-autonomy in daily life. Participants described themselves as fighting the condition and winning. | Participants seemed to be struggling to live their lives due to their seizures. Emotional expression of helplessness in opening and ending statements. Maladaptive coping tendencies such as self-harming, dissociation, or not processing their emotions. |
| 5.2 Understanding | Participants seemed to be knowledgeable about their condition and did not explicitly express a wish to know more. | Participants lacked the understanding of their condition e.g. why it started, what the triggers were, how to stop them. Participants asked many questions. Participants described feeling “*lost*” |

PNES = Psychogenic Nonepileptic Seizures, HCP = Healthcare professional

**8.3.1 Theme 1: Causation**

Participants with epilepsy choose to described the events surrounding the onset of their condition at the very beginning of the first writing session, and then would rarely, if ever, write again about the experience or ruminate about the cause. Developing seizures was typically described as an event that had changed their lives or themselves “*forever*”. Individuals explained feeling shocked, frightened, and struggled with altered self-identities as a result.

***Epilepsy****:* [Start]*“I often wonder what my life would have been like without my epilepsy. It was such a shock to me and my family when at the age of 11 I started having seizures…that’s when I became different from my twin sister”* **(Female, 58 years old, last seizure was in 2012)**

Participants with PNES appeared to explore the possible causes of their condition throughout their narratives. Although they had been asked to write about their experiences of living with their condition, 16/19 participants spontaneously reported a past trauma, such as abuse (e.g. physical, sexual, emotional) or a distressing event (e.g. surgery, bereavement). Participants commonly perceived comorbid conditions (emotional or organic) as linked to PNES. For some, it was almost as if their seizures were yet another set of symptoms or problem that they had to manage and overcome. This also gave the impression that participants perceived their seizures as a consequence of other events in their lives, for example, their relationship with another person. In contrast, those with epilepsy depicted the development of seizures as sudden and unprovoked (“*out of the blue*”).

**PNES**: [Start] *“I HATE this condition. My life was great before I got "ill", well most people would call my life hard but it was great to me. I was finally in a stable place, after a string of life events; one alone would test the strongest of people, but I felt "nothing could stop me now". I'd left an abusive relationship I'd had for eight years.”***(Female, 31 years old, 30+ seizures in the last 4 weeks)**

**8.3.2 Theme 2: Emotions**

One of the most prominent differences between the two groups of individuals was the emotional tone of their writing. Participants with epilepsy seemed emotionally stable and fluctuations in mood were often described as associated with postictal symptoms or adverse effects of anti-epileptic medication.

In contrast, the writings of individuals with PNES were very negative, reflecting depressive symptoms and lack of positive emotions, and increased levels of anxiety. These feelings appeared to stem from the development of PNES, but in some cases, they were also discussed as a long-term characteristic. A key example of the difference in emotional tone was how participants began and ended their writing sessions. Participants with epilepsy would often begin their narratives in chronological order, describing events surrounding the onset of their condition and what had happened during their first seizure - their narratives might have also end with a statement of defiance. By contrast, those with PNES tended to begin with an expression of suffering and finish on a negative note.

**Epilepsy:** *“There have been times of course when my positivity has left me in hospital or when I lost a lot of weight due to problems caused by medication, but I usually get my fight back later on.”* [End] **(Female, 57 years old, 1 seizure in the last 4 weeks)**

**PNES:**  “*I just wish there was a way to have it all go away and not have to worry about my condition anymore and get back to normal. I just need some help.”* [End] **(Male, 42 years old, 20 seizures in the last 4 weeks)**

It seemed that participants with PNES were struggling with their condition, and at times, expressed a sense of powerless. Moreover, participants described hating themselves, writing that they are “*worthless*”, “*weak*” or “*useless*”. Such expressions of self-depreciation were almost non-existent in those with epilepsy.

Individuals with PNES were more likely to describe that they suppress their emotions or not “*letting anyone in*”. Some described that this has contributed to the development of their condition. Moreover, following the manifestation of their seizures, those with PNES often described how they might struggle to regulate or control their emotions.

**8.3.3 Theme 3: Seizure symptoms**

*8.3.3.1 Description*

Differences in how individuals wrote about their seizures were observed. Those with epilepsy would tend to describe their seizures as an external, independently (sometimes hostile) acting agent. Seizures were reported as following them around or “*stalking*” them. Similarly, several participants projected human characteristics onto their seizures, such as giving them a name or consciousness.

In comparison, participants with PNES were more inclined to describe their seizures as a place or a state that they enter, and in which, the individual retains agency in the seizure.

**Epilepsy:** *“I call my epilepsy Bob… It’s much easier to say “Bob’s been a bit of a nuisance today’ or ‘Bob just paid me a visit!’”* **(Female, 71 years old, 4 seizures in the last 4 weeks)**

**PNES:** “*I often think I am back in that hell hole, having just relived the whole thing again whilst in* (sic) *seizure.”* **(Female, 43 years old, 120 seizures in the last 4 weeks)**

*8.3.3.2 Focus*

Participants with PNES appeared to focus more on their post- and interictal symptoms. When individuals did describe seizure episodes, it would often be of the situation or circumstances of particular seizures.

Those with epilepsy, however, centered their narratives on how the condition has affected their daily life, for instance, the challenges it has posed to gaining employment. Seizure episodes did not dominate individuals’ experience of living with epilepsy. This gave the impression that “*secondary*” issues such as the side effects of medication, related-stigma, the restrictions imposed because of epilepsy, and the anxiety about having another seizure were the biggest challenges. In contrast, for those with PNES, it was also the actual episodes themselves that were problematic.

**Epilepsy:** *“Currently epilepsy effects my life more due to secondary symptoms, mostly those I have from the anti-epileptic drugs.”* **(Female, 40 years old, 0 seizures in the last four weeks)**

**PNES:** *“I suppose - by now - it is safe to say it [PNES] has ruined me, basically. Having a seizure every one till two/two and a half hours does not leave much room for anything else.”* **(Female, 31 years old, 340 seizures in the last 4 weeks)**

*8.3.3.3 Post-ictal experiences*

Both groups tended to report their postictal symptoms in detail; however, those with epilepsy were more likely to describe themselves as getting back on with their day as normal, or that they would not let it affect them. Seizure-related injuries were described more commonly by those with epilepsy, than those with PNES.

**Epilepsy**: *“I had three seizures before I went to work this morning but so...? I still went to work, did a full day and had lunch in the pub.”* **(Female, 36 years old, 5 seizures in the last 4 weeks)**

**PNES:** *“When I have a seizure I black out and have no idea what is going on and wake up sometimes groggy and really weak and not knowing who or where I am.”* **(Male, 42 years old, 20 seizures in the last 4 weeks)**

*8.3.3.4 Fear*

While both groups described that they were afraid of triggering or having a seizure, individuals with PNES were more likely to state that the seizure itself involved fear or that it was a horrible experience.

**Epilepsy:** “*Epilepsy is something I have learned to live with, some days (particularly when a severe fit rears it ugly head) I feel scared out of control and useless*.” **(Female, 62 years old, 2 seizures in the last 4 weeks)**

**PNES:** *“The way these seizures make me feel is this. Until recently, genuinely I believed that I was indeed dying.”* **(Female, 46 years old, 224 seizures in the last 4 weeks)**

**8.3.4 Theme 4: Treatment**

*8.3.4.1 Diagnosis*

Unsurprisingly, the diagnostic journey was a key part in the narratives of participants with PNES. Some individuals appeared to reject or resist the diagnosis, whilst no participant with epilepsy reported such a reaction – in fact, some individuals with epilepsy explained that their diagnosis came as a “*relief*” as it validated what they were experiencing.

**Epilepsy**: “[letter to younger-self]…*it turns out you have epilepsy. It’ll be scary thinking about it at first but honestly; it’s such a relief. I remember the unknown and it sucks, so don’t worry your answer is coming!”***(Female, 27 years old, 0 seizures in the last 4 weeks but typically averages 1 seizure every other month)**

**PNES:** “*I accept that they may be psychogenic … but I'm not yet convinced that there is no physical link.”***(Male, 69 years old, 7 seizures in the last four weeks)**

*8.3.4.2 Healthcare professionals (HCPs)*

With very few exceptions, participants with epilepsy uniformly described HCPs in a positive manner, perceiving them as helpful, supportive and a valuable source of knowledge.

This was in total contrast to the experiences described by those with PNES. Individuals described past experiences in which HCPs failed to listen and did not believe that their symptoms were real, and beyond their control. Participants described having avoided healthcare services in the past because of previous adverse experiences. Individuals explained feeling let down and ostracised by HCPs.

**Epilepsy:** *“My wonderful, calm, patient neurologist has listened to, heard and prescribed”* **(Female, 59 years old, last seizure was in 2014)**

**PNES:** “*What a life, but at least most days now I don’t end up at that shitty hospital where the doctors treat you like shit and call you a fake. How I have never been arrested there I don’t know.”***(Female, 43 years old, 120 seizures in the last 4 weeks)**

*8.3.4.3 Treatment*

Given the differences in the acceptance of their diagnosis, it follows that participants perceived their treatment differently. Those with epilepsy viewed their medication as key to seizure control, and viewed the side effects as a compromise. In PNES however, individuals described difficulties finding and securing access to psychological or specialised care. Participants seemed skeptical of therapy and perceived it as an approach to treat their seizures opposed to changing other aspects of their life.

**Epilepsy:** *“I have a high dose of the medication that I take and need that high dose to control the seizures as well as is feasibly possible in my body.”* **(Female, 40 years old, 0 seizures in the last 4 weeks)**

**PNES:**  *“I came across a psychologist though, yesterday to be fair and she was amazing. Although she did not have much knowledge of functional neurological disorders apart from what she had to Google, she sat back and listened… So my hopes are raised a little more with the extra help that I may receive (but I wont hold my breath).”* **(Female, 26 years old, 2 seizures in the last 4 weeks)**

**8.3.5 Theme 5: Daily life**

*8.3.5.1 Coping*

The two groups differed greatly in their descriptions of how they depicted themselves as living with their condition. Participants with epilepsy appeared keen to present themselves as having integrated the condition into their lives and would report that they are “*coping*” with the adversity. For the most part, individuals appeared to be fighting, striving to live a *normal* life, and persevering “*despite*” having epilepsy. In comparison, nearly half of those with PNES explicitly stated that their condition had “*destroyed*” their life or produced cries for help.

**Epilepsy:** *“Everyone is running, cycling, on the stepper and there is little old me just walking on the treadmill… At first it bothered me that I couldn’t run like the others but then I just accept that’s how it is and aim for my 3 miles, at least I have my music on and reach my goals, the same as everyone else!”* **(Female, 34 years old, 0 seizures in the last 4 weeks but typically experiences 2 seizures a year)**

**PNES:** *“This condition seems to have taken my life away from me.”* **(Male, 42 years old, 20 seizures in the last 4 weeks)**

Those with epilepsy would downplay and minimise the severity of their condition. Participants would also describe the benefits of laughter and report having gained positive insights from developing the condition, which was rarely observed in PNES.

**Epilepsy:** *“There have been times I’ve screamed at you in frustration …but there’s also things that without you I would probably not have learned. If I am only to have only half a life then I will make it count.”* **(Female, 56 years old, 7 seizures in the last 4 weeks)**

Individuals with PNES would describe and show insight into their coping behaviours that may not be useful, for instance, self-harming, bottling it up and dissociating. Those with epilepsy would tend not to describe specific coping strategies but when they did, it was often practical behaviours such as, keeping a diary to aid with memory and relying on friends and family.

**PNES:** [Letter to younger self] *“When bad things happen to you, do not bottle it all in. This will not help you in the long run. You are better off telling someone, anyone as hard as that may seem it is better option I promise you, it’ll be easier that way.”* **(Female, 26 years old, 35 seizures in the last 4 weeks)**

*8.3.5.2 Understanding*

Those with epilepsy seemed to present themselves as knowledgeable about their condition, for example, they would give advice to their younger selves about how to cope. In contrast, participants with PNES described themselves as “*lost*”, stating explicitly that they wanted to know more about PNES, for instance, why they had developed seizures, what caused them, and how to stop them.

**PNES:** *“Until there is more understanding on the condition and how to explain things to anyone diagnosed then it's a lost world I seem to have been put into and one I'd like to find my way out of.”***(Female, 31 years old, 30+ seizures in the last 4 weeks)**

Differences in outcome expectations were also observed. Participants with epilepsy appeared better able to accept that they had developed seizures, and some seemed content that they would have their seizure disorder for the rest of their lives, while others were determined to be free of it.

**Epilepsy:** *“It defines me it’s part of who I am, my identity. I don’t like that I’ve allowed that to happen”.* **(Female, 27 years old, 0 seizures in the last 4 weeks)**

In contrast, participants with PNES seemed to be confused about the nature of their seizures and unsure of the disease timeline. Although some individuals did express the desire to be seizure free or have their old life back, this appeared to be a “*wish*” rather than an expectation. Participants with PNES came across as wanting to manage or “*beat*” their disorder, however, they did not know how to and experienced almost every day as a struggle.

**PNES:** “*I am now 26 and living with the man I want to spend the rest of my life with…I just hope that my seizures and illness wont get in the way of our life and hopefully our plans to start a family.”***(Female, 26 years old, 35 seizures in the last 4 weeks)**

**8.4 Discussion**

Both epilepsy and PNES were associated with profound social, psychological and emotional difficulties. Although individuals were closely matched on HRQoL, participants appeared to respond to the impact of their seizure disorder differently. Within Western societies there is a growing pressure to be *successfully ill.* For example, those with long-term disorders are expected to accept that their condition maybe chronic and rise to the challenge by finding solutions, champion their story, and gain new insights into their sense of self (Frank, 1997). It has also become a social norm to use metaphors of battle, with illnesses being perceived as a threatening entity that must be fought (Kielhofner, 2008). In the current study, these expectations were, for the most part, reflected in the narratives of participants with epilepsy. Individuals were keen to present themselves as coping with adversity and would often express that they would not let it “*win*”. It seemed important for those with epilepsy to express they are living a “normal” life, however, they did admit that it is occasionally interrupted, very briefly, by events associated with their condition (either by the seizures themselves or their sequelae) – for some this was a challenge while others accepted it with equanimity. A similar finding has been reported in other qualitative studies investigating the consequences of epilepsy on daily life (Raty, et al. 2007; Monzoni & Reuber, 2009). Raty et al., reports young adults described themselves as being able to cope with and accept the issues associated with their condition, and that they feel “normal” and healthy despite having epilepsy – one participant reported, “*there is nothing catastrophic about an epilepsy diagnosis”* (pg. 394). The authors suggest this behaviour may reflect a normalisation process in which, individuals use various strategies aimed at making themselves feel “normal”, for example, by explicitly saying that they are indeed “normal”, striving to keep up the appearance of normality, or construct and live a story of life as a “normal” person.

Among those with PNES, however, this perspective was rarely expressed. That is not to say that individuals might not have wanted to possess this outlook, but it may be they felt that they were unable to conform to these societal norms for a number of reasons. For example, they may experience insufficient control over or understanding of their symptoms, or had not been given the support they needed.

The tendency for those with epilepsy to normalise experiences of their condition has been contrasted with that of individuals with PNES who catastrophise about such events. Using content analysis, Robson et al. (2012) investigated the use of “third party references” - this is where patients divert questions regarding their seizures to witnesses, such as family members - by patients with epilepsy or PNES during clinical consultations with a neurologist. The results demonstrated the two patient groups made a similar number of references as each other, however, 12 out of 13 patients with PNES used third party references to catastrophise about their seizure events whereas, only one patient out of seven with epilepsy did. Six patients with epilepsy made normalising references while only two did with PNES.

The reason for this difference between the two conditions is unclear. (1) It maybe that experientially, non-epileptic seizures are indeed a more negative and difficult event compared to epileptic seizures. In line with this, studies have shown that ictal-panic is more common in PNES than epilepsy (see Chapter 4). (2) In the current study and the one by Robson et al. (2012), the frequency of seizures was greater in those with PNES compared to patients with epilepsy and so seizure events may have dominated and constituted a larger part of their daily life. (3) Patients with PNES may exhibit a greater tendency to engage in cognitive distortions, for example, holding on to negative thinking patterns. Although caution should be used when attempting to draw formulations simply from patient’s written accounts, other forms of negative thinking (in addition to catastrophising) were evident in the writings of those with PNES. For instance, study participants with PNES often used negative self-labeling (I am useless) whilst this was not observed in the written accounts of those with epilepsy. (4) The manner in which others perceive their condition may also influence how patients with PNES present their symptoms. Patients with non-epileptic seizures may feel the need (on some level) to catastrophise their experiences to highlight the severity of the condition and to reinforce that it is not made up. What is more, as a mental disorder, PNES are associated with high levels of stigma. Indeed, it is conceivable that patients are concerned about being diagnosed or labeled as someone who experiences a problem with their mental health. This may contribute to patients with PNES resisting or dismissing psychosocial interpretations of their symptoms, instead conceptualising their problems as physical and very much beyond their control (Monzoni et al., 2001; Whitehead et al., 2013).

How individuals appraise and perceive their condition is of clinical importance. For example, in those with PNES or epilepsy, illness perceptions have been shown to be a greater predictor of HRQoL when compared to clinical factors, such as seizure frequency and severity, and almost as important as symptoms of psychopathology (Rawlings et al., 2017a). In PNES, illness perceptions have also been found to be correlated with the number and severity of somatic symptoms and psychological distress (Novakova et al., 2015). The cognitive model of adjustment to illness suggests how the individual appraises, understands and evaluates their condition determines his or her emotional and behavioural response. This model can help to explain why individuals with the same disorder may react differently or have very different experiences from one another. For example, Moorey and Greer (2002) posit that in cancer, managing the initial appraisal of the diagnosis is one of the key stages. Patients often achieve this by either incorporating the experience into their word-view, or by altering their belief systems. This is a necessary and valuable step because after this period, which is often associated with turmoil and confusion, patients form secondary appraisals considering the wider implications of their condition and what can be done about it (i.e. treatments and recovery). This helps to influence and develop adjustment styles. Greer and Watson (1987) propose that there are five common approaches: fighting, avoidance, hopelessness, anxious preoccupation and fatalism. This stage is seen as an important process as it can have connotations for other areas of daily living; for example, developing an appraisal of hopelessness can manifest a depressive state or trait as negative thinking styles can become generalised to factors other than their illness, such as their self-perception or future events. On the other hand, appraisals associated with a good prognosis can also result in maladaptive behaviours; patients who perceive their illness as a challenge may view healthy and normal feelings (such as anger, sadness or anxiety) as a weakness or defeat, and therefore suppress or ignore these emotions, when in fact, processing and expressing emotions that are associated with an illness can be therapeutic (see chapter 5).

Notwithstanding the fact that this model has primarily been applied to those living with cancer, it may have some relevance to the findings in the current study. A proportion of individuals with epilepsy conceptualised their life with epilepsy as a fight, while those with PNES appeared to feel overwhelmed and helpless as a result of their condition. As individuals with PNES often spontaneously described and ruminated about the perceived cause of their condition, this may suggest that they have not integrated such concepts into their life story, or formed the appropriate primary appraisal. In comparison, those with epilepsy may have achieved this initial stage, which has allowed them to develop secondary appraisals resulting in more beneficial adjustments styles. The findings suggest that promoting the active involvement of patients (and in some cases, family members) in cognitive-based therapies to appraise and manage the diagnosis, symptoms or consequences should be encouraged in those living with seizures.

Self-efficacy is an individuals’ perceived ability to manage difficult situations. In the current study it seemed that those with epilepsy possessed a greater sense of self-efficacy towards coping with their condition compared to individuals with PNES. While this measure has not yet been quantitatively investigated in PNES, Sung et al. reports an association between self-efficacy, and self-esteem and life satisfaction in epilepsy. Moreover, self-efficacy has been found to mediate the relationship between seizure severity and life satisfaction (Sung et al., 2013). In another study, self-efficacy was related to successful self-management and high outcome expectations in epilepsy (Robinson et al., 2008). Further research is needed into the concept of self-efficacy in PNES, as it could be a potential mechanism of change.

Despite the fact that individuals were closely matched on seizure severity, there were notable differences in several characteristics of the ictal experience. Firstly, individuals with PNES were more likely to associate their seizures with fear and explain that they are a horrible experience. The finding that ictal panic is more often experienced by individuals with PNES compared to those with epilepsy is in line with previous findings using quantitative methodologies (Rawlings et al., 2017d). Individuals also tended to differ in how they conceptualised their seizures. In epilepsy, seizures were described as an external agent, impacting upon on the individual, whereas in PNES, seizures seemed to be a state or place that they find themselves in. This difference is consistent with findings from quantitative research (Watson et al., 2002; D'Angelosante et al., 2015), and more specifically, from a series of studies using language analyses to investigate interviews between patients and a member of their medical team (Plug et al., 2009b; Cornaggia et al., 2012; Schwabe et al., 2008).

It is important to gain an understanding of the subjective experiences of seizure episodes as this information could contribute to the diagnostic process (Reuber et al., 2016; Plug et al., 2009a; Reuber et al., 2009) and help to inform how clinicians communicate with their patients. For example, therapists can share such insights with individuals to help them identify and manage bodily and mental manifestations of a seizure. With practice, individuals may become adept at recognising how their emotions, thoughts and behaviours modulate their seizures. This is particularly relevant given the fact that about a third of individuals with epilepsy or PNES report alexithymic tendencies (Myers et al., 2013). Future research could explore novel ways of enabling patients to better understand and name their own feelings about their seizures. There is certainly scope to examine different approaches, such as dramatisations, poetry and artwork.

Differences in the nature, expression and processing of emotions between the two conditions were observed. It is a limitation of this study that individuals were not matched on anxiety and depression. Therefore, it is difficult to draw strong conclusions from the differences in emotional tone. Having said that, it has been reported elsewhere that individuals with PNES tend to report higher levels of anxiety and depression than those with epilepsy (Brown and Reuber, 2016a). Therefore, it could be argued that our samples are representative of these two patient populations. Our findings add to the idea that individuals with PNES exhibit altered emotional functioning tendencies (Brown and Reuber, 2016b; Brown et al., 2013; Pick et al., 2016).

Differences in the reported experiences and perception towards HCPs between the two conditions were striking. In PNES, stories whereby individuals had felt discriminated against due to their diagnosis or let down by HCPs constituted a major part of the narrative. The difference in the perceptions of healthcare providers of those with epilepsy and PNES observed here mirrors the findings of previous studies. Karterud et al. analysed the transcripts from ten individuals who had had their diagnosis changed from epilepsy to PNES reporting that individuals described a ‘transfer of responsibility’ from HCPs to themselves. One patient explained feeling left to deal with the condition on her own, whereas with epilepsy she would be offered a multi-professional team at an epilepsy centre (Karterud et al., 2010). The finding that individuals with PNES report poor experiences of healthcare is unfortunately not uncommon (Tolchin et al., 2016). It is clear that, in addition to improving the understanding and sensitivity towards PNES, greater efforts are needed to help HCPs feel more confident in providing care for patients with this condition and to insure everyone involved in their treatment is working to a consistent model (Worsely et al., 2011). Two qualitative studies reported that patients with PNES expressed a sense of doubt about the diagnosis, which in part, seemed to be related to the HCPs perceptions of their disorders (Wyatt et al., 2014; Thompson et al., 2009).

This comparative study has a number of potential limitations. Individuals were not matched (although representative of the general population) on several factors including, the duration of seizures and seizure frequency. This is likely to have had an effect on individual’s accounts. For example, participants with epilepsy had experienced seizures for a longer duration (median 25.5 years), possibly giving them more time to adapt to living with the condition. Having said that, the median duration of PNES disorders was five years, so most participants in both groups had chronic seizure disorders. On a similar note, there are issues with generalising the current findings to other individuals living with a seizure disorder due to factors such as age, gender and clinical symptoms, or just by the fact that the current dataset was collected in the context of a study that aimed to investigate the effectiveness of a writing intervention for individuals diagnosed with a seizure disorder, and therefore, our sample may only be representative of individuals who value and feel that they need additional support provided by psychological interventions. More specifically, there is a small proportion (about 10%) of individuals with PNES who report seizure cessation following their diagnosis (Reuber, 2008). Although in the current study one participant with PNES did not experience a seizure in the last four weeks, it is unlikely that we have captured and explored the narratives of these individuals. Having said that, it has been argued that generalisability is not a primary purpose of qualitative analysis (Polit and Beck, 2010). Instead, these approaches aim to gain a deeper and richer understanding into the lived experience of illness seeking situational, as opposed to demographical representativeness.

We chose to recruit individuals from both neurology clinics and membership-led organisation. Whilst this means that we were able to gather a more diverse range of experiences than if participants had been recruited from a single centre, we were unable to confirm all diagnoses because we did not have access to all participants’ medical records.

In conclusion, the current study demonstrates that clear and important differences can be observed between lived experiences of epilepsy and PNES based on written accounts of the condition. For those with epilepsy, seizures and the sequelae of epilepsy were both conceptualised as something that must be fought. In contrast, those with PNES rarely used fighting terminology, and instead, were likely to describe their seizures as a place or state they enter. Our findings suggest that, helping individuals to develop a sense of self-efficacy and interventions promoting greater self-monitoring of bodily and mental sensations could be beneficial in both conditions. Future research should look to use different methodological and analytical approaches to further examine subjective phenomena associated with epilepsy and PNES.

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**Chapter Nine**

**Summary and Conclusions**

**9.1 Key findings**

The overall aim of this PhD was to investigate the phenomenology and clinical implications (diagnostic, prognostic and therapeutic) of the lived experience of epilepsy or psychogenic nonepileptic seizures (PNES).

**Chapter 2**

Complementing a similar paper summarising qualitative research involving patients with epilepsy (Kerr et al., 2011), chapter 2 reported the first systematic synthesis of qualitative research investigating the subjective experience of PNES. Based on the inclusion and exclusion criteria, 21 suitable studies were found. These papers were consecutively synthesised using a theory and data driven approach inductively. This means that whilst the themes were largely grounded in the literature (data-driven), external classifications based on theory or clinical relevance were applied to guide the written report of the outcomes. For example, “*Neurologists have distinguished between four different states in the lives of people with seizures: ictal, inter-, pre- and post-ictal (Beyenburg et al., 2005). Cornaggia et al. (2012) reports that patients with PNES are not always able to identify these phases suggesting that, in PNES at least, this division may be artifactual. Notwithstanding this, we are using these phases to organise our synthesis of information from the studies included in this review although these categories are not derived from patients themselves*.” Another example of this is that, not all individuals will have experienced the same diagnostic milestones in the order reported here: getting the diagnostic label, communication of PNES, misdiagnosis, and dealing with the implications of the terminology. However, for the sake of clarity, I decided to describe these findings by structuring the experience of “diagnosis” into themes that followed an order allowing the accounts of different individuals to be compared more easily rather than collating all descriptions into a single more idiosyncratic experience (comprising elements such as, receiving the diagnosis, how it was communicated, the – potential – impact of changing a previous diagnosis (i.e. from epilepsy to PNES) and the affect of all of these elements on the individual).

Five key themes emerged from the synthesis:

* Theme one: ‘seizure events’ revealed the experiential heterogeneity of seizure events. What is more, there appeared to be differences in how individuals described their seizures. Those with PNES would often offer very little information about their experiences of seizures and resist the interviewers attempts to focus on such topics, whereas individuals with epilepsy tended to volunteer this information readily and actively formulate the events. The two diagnostic groups seemed to conceptualise their seizures differently, individuals with epilepsy described their seizures as an external agent/force that impacted them from the outside. In contrast, individuals with PNES were more likely to describe their seizures as a space or place that they enter in which they may have some control over the course of the seizure.
* Theme two: ‘diagnosis’ focused on individuals’ accounts of diagnosis. This was reported as being a lengthy process, which was associated with feelings of limbo, frustration and worry. Many participants shared a sense of uncertainty surrounding PNES, often resisting (either overtly and/or passively) psychological explanations, diagnosis, and treatments.
* Theme three: ‘treatment and management’ revealed that negative experiences with healthcare professionals were common; individuals sought validation, care and an explanation of their experiences from professionals however they often reported feeling ignored or doubted. Moreover, individuals expressed mixed ideas, expectations and experiences of psychological interventions. Some found treatment beneficial while others reported improvements in other areas of their life, but did not report a reduction in their seizures.
* Theme four: ‘emotional events’ revealed that many individuals had experienced past or current distressing events. Some participants demonstrated insight into their methods of emotional processing. The expression of negative emotion was common, while individuals rarely spoke of positive emotions.
* Theme five: ‘impact of PNES on daily life’ indicated that PNES were described as a significant burden, for themselves as well as their friends and family, which was associated with financial and psychosocial losses.

It was concluded that, qualitative studies have produced helpful insights into individuals’ experiences of living with PNES, but many groups (men, young people, elderly, non-Western individuals) are underrepresented in studies carried out to date. Finally, research using other methods of data collection and analysis could help to deepen our understanding of this disorder.

One of the tenets of the bio-psychosocial model is that, to understand and respond to the patients suffering, healthcare services must attend to the biological, psychological and social dimensions of illness. A prominent theme that emerged from the analysis was patients expressed great confusion and uncertainty about the different aspects of their condition. This seemed to result in damaging the alliance or increasing emotional distance between the doctor and patient. The responsibility of a healthcare provider is not simply to make a bio-medical diagnosis, but to also interpret the illness from the perspective of the patient, helping them to understand what their illness means and how it applies to them. Patients need to be given the time, space, and support to formulate and articulate their worries and concerns, and start to form their expectations of illness. Communicating medical information should not simply be about providing answers, but also promoting understanding (Borrell-Carrió et al., 2004).

It has been reported elsewhere that, the approach in which patients with PNES are given their diagnosis could be improved. Dworetzky surveyed 133 epilepsy experts in the United States reporting that, 42% of doctors would tell their patient to make an appointment with mental health services themselves. Only n=56 experts reported giving educational materials about the condition to patients to share with their mental health provider (given the fact that the condition is poorly understood amongst mental health services, it is unlikely that the service provider will know what to do when the patient arrives for their appointment). Overall, less than half of the (n=61) experts asked reported that patients meet with a psychiatrist prior to hospital discharge (Dworetzky, 2015).

It is clear that a more structured pathway is needed to help patients understand their diagnosis, and increase uptake of mental health services. In an attempt to improve attendance at a cardiac rehabilitation programme, Wyer et al. used the theory of planned behaviour to structure the content of letters inviting patients following a cardiac event (Wyer et al., 2001). The theory of planned behaviour suggests that the likelihood of behaviour can be predicted by examining an individual’s attitudes, beliefs and perceived behavioural control (Ajzen and Fishbein, 1970; Ajzen and Fishbein, 1980). These letters: introduced the intervention and reinforced the diagnosis; boosted patient’s perceived control (“you will be offered advice and information about how best to recover” pg. 156); normalised their condition and treatment (“professionals recommend that people who have had a heart attack should attend a cardiac rehabilitation programme” pg. 156); influenced the patients’ attitude towards the behaviour (“those who attend such a programme are more likely to recover sooner and better” pg. 156); and ended the letter explaining that patients can also ask any questions that they may have about their condition. Compared to a control group (n=44) who were given letters that were not influenced by the theory of planned behaviour, those in the experimental group (n=43) were more likely to accept the invitation (p<0.04), and actually attend the intervention (p<0.003). A similar method could be utilised in those with PNES.

**Chapter 3**

This chapter explored the frequency of panic symptoms in PNES, and investigated if such experiences, just before, during or after episodes of transient loss of consciousness (TLoC), can help to distinguish PNES from the other common causes of TLoC, namely syncope and epilepsy. This study was based on a dataset that had already been collected by Reuber et al. (Reuber et al., 2016; Reuber et al., 2011). Individuals with secure diagnoses (made by experts in the disorder and based on the gold-standard procedure for diagnosing the condition) of PNES (n = 98), epilepsy (n = 95) and syncope (n = 100) were identified using clinical databases from three United Kingdom hospitals (Royal Hallamshire Hospital, National Hospital for Neurology and Neurosurgery, and Royal Victoria Infirmary). Using a Likert-scale, participants self-reported the frequency with which they experienced seven symptoms of panic disorder in association with their episodes. These questions reflected the symptomology of panic as described in the current medical nosologies (World Health Organization, 1992; APA, 2013). A composite panic symptom score was calculated on the basis of the frequency of symptoms. The findings suggest that, 8.2% of individuals with PNES, 29.5% in epilepsy and 43% in syncope reported “never” experiencing any of the seven panic symptoms in their episodes of TLoC. Individuals with PNES reported more frequent panic symptoms in their attacks than those with epilepsy (p<0.001) or syncope (p<0.001), however, participants with PNES were more likely “rarely” or “never” to report five of the seven ictal panic symptoms than “frequently” or “always” (see Table 9.1).

|  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
|  | PNES | | | Epilepsy | | | Syncope | | |
|  | Mean | Upper CI | Lower CI | Mean | Upper CI | Lower CI | Mean | Upper CI | Lower CI |
| Feel very frightened | 3.04 | 3.32 | 2.78 | 2.35 | 2.64 | 2.05 | 1.69 | 1.93 | 1.46 |
| Feel something terrible might happen | 2.09 | 2.37 | 1.81 | 1.7 | 1.97 | 1.44 | 1.31 | 1.49 | 1.13 |
| Frightened I am going to die | 2.07 | 2.3 | 1.83 | 1.35 | 1.54 | 1.16 | 1.31 | 1.49 | 1.14 |
| Frightened I will lose control | 2.54 | 2.25 | 2.84 | 1.94 | 2.23 | 1.64 | 1.57 | 1.8 | 1.33 |
| Frightened I will go crazy | 1.87 | 2.11 | 1.63 | 1.32 | 1.5 | 1.14 | 1.07 | 1.14 | 1 |
| Heart pounds, feel shaky and sweaty | 2.97 | 3.23 | 2.71 | 2.05 | 2.35 | 1.76 | 2.16 | 2.45 | 1.87 |
| Feel I have to get out of the situation | 2.24 | 2.53 | 1.96 | 1.9 | 2.18 | 1.63 | 1.67 | 1.9 | 1.43 |

Table 9.1 Means and confidence intervals (CI) of individuals with psychogenic nonepileptic seizures (PNES), epilepsy or syncope in response to the questions about panic symptoms associated with episodes of transient loss of consciousness

We did not report the value of those who responded “sometimes” experiencing the ictal panic symptoms listed, which is why the percentages in Table 2 in the published article do not tally to 100%. However, this percentage of those with responded “sometimes” can be calculated by the sum of the two percentages (“always or frequently” and “rarely or never”) subtracted from 100. As Table 9.2 shows, compared to those with epilepsy or syncope, a greater proportion of individuals with PNES reported “sometimes” experiencing all of the symptoms of ictal panic investigated. This is consisted with the idea that PNES are characterised by a greater degree of experiential heterogeneity whereas, the subjective experience of an epileptic seizure is more homogeneous i.e. there is less intra-individual variability (Reuber et al., 2011). As discussed in chapter 1, based on the bio-psychosocial model we can make the argument that, subjective experiences can provide further context and meaning to symptoms which can, in part, help to improve the therapeutic process. One of the aims in therapy for PNES is to help patients improve their awareness and recognition of seizure warning symptoms – patients can then for example develop coping or control skills to suppress their seizures such as distraction or grounding techniques. The results from this study demonstrate that a proportion of individuals with PNES do not experience a habitual attack and so it is important for therapists to explore a wide range of symptoms that may be associated with their nonepileptic seizures.

Table 9.2 Percentage of respondents who reported “sometimes” to experiencing the ictal panic symptoms listed

|  |  |  |  |
| --- | --- | --- | --- |
|  | PNES | Epilepsy | Syncope |
| Feel very frightened | 34% | 16% | 17% |
| Feel that something terrible might happen | 23% | 11% | 7% |
| Frightened that I am going to die | 26% | 7% | 7% |
| Frightened that I will lose control | 26% | 9% | 7% |
| Frightened that I will go crazy | 18% | 8% | 3% |
| Heart pounds, feel shaky and sweaty | 34% | 13% | 19% |
| Feel I have to get out of the situation | 29% | 14% | 14% |

PNES = Psychogenic nonepileptic seizures

A receiver operating characteristic analysis demonstrated that the composite panic symptom score distinguished individuals with PNES from the other groups (sensitivity 71.1%, specificity 71.2%), but not epilepsy from syncope. Overall, individuals with PNES reported TLoC associated panic symptoms more commonly than those with epilepsy or syncope. Having said that, this value was arrived at using inferential statistics to distinguish between three different groups of individuals who had a pre-determined and clear diagnosis. As such, the clinical validity and reliability of this value as a predictive test in patient groups, including diagnostically less clear cases, has not been explored.

This outcome of this study needs to be interpreted in light of the following limitations. As explained, all patients were diagnosed using the gold standard procedure, which is likely to have introduced a selection bias. Patients with an uncertain or dual diagnosis (e.g. epilepsy and PNES) were not included. This has important implications for the test accuracy of the panic composite, as it is likely to have exaggerated its accuracy, and I am unable to generalise these findings to all patients who have experienced episodes of TLoC. Notwithstanding this, given the observed modest sensitivity and specificity, the application of using symptoms of ictal-panic in diagnostic practice is limited. If differences in the composite score had been greater and the score more promising as a differential diagnostic tool, further tests could have been applied, for example, determining the false positive and false negative rate and testing the measure in a more heterogeneous sample. However, we recommend that it would be more fruitful to add questions about ictal panic symptoms to a range of other questions investigating different aspects associated with TLoC experiences as well as interictal symptomatology.

The outcomes offer implications to help conceptualise PNES, namely that as a proportion of individuals experience cognitive and autonomic symptoms of panic, a single psychopathological mechanism (PNES as “panic attacks without panic”) is insufficient to explain PNES in all patients or at least in all PNES.

A purpose of the bio-psychosocial model is to incorporate patients’ subjective experience and objective bio-medical data to gain a greater understanding of the illness. In epilepsy we can investigate seizure-related changes in the brain objectively, for example capturing events using EEG to monitor epileptiform activity, or report on the semiology of seizures by observing the patient. However, this reductionist stance, focusing on neurological processes, ignores subjective data and as such, it cannot tell us what the patient is actually experiencing – we are on the outside looking in. It is easy to infer that, when epileptiform activity is recorded, the patient is having a seizure, however, the findings from this study demonstrate that, not all patients with epilepsy (or syncope or PNES) experience a total loss of consciousness during their attacks (or at least, for part of it). This is consistent with the literature as Devinksy et al., (1996) reported that when 16 patients with both epilepsy and PNES were asked to remember a word or phrase during a seizure, 15 patients were unable to recall the item after an epileptic seizure, whereas 14 patients were able to recall it after a PNES.

When combined, these findings have clinical relevance in that during medical interventions, patients presenting with TLoC should be cared for and managed as if they are aware or conscious of the events going on around them e.g. they should be informed of what is happening, where they are, and who they are with. In PNES, such an approach may help to reduce the length and/or severity of the seizure, and in both epilepsy or PNES: it could ease the confusion and anxiety that patient’s tend to report experiencing immediately following a seizure, also it might lessen the fear or anxiety that is associated with having a seizure in the future.

**Chapter 4**

Here we investigated the profile, relationship and predictive power of demographic (age, gender, years of education), condition-related (seizure frequency and severity, duration since onset), and psychological factors (depression, anxiety and illness perception) on health-related quality of life (HRQoL) in participants with PNES and those with epilepsy. Individuals with epilepsy (n = 62) and PNES (n = 45) were recruited from the Royal Hallamshire Hospital and from membership-led organisations for individuals living with seizures. This dataset was collected in the context of a randomised control trial investigating a writing intervention for individuals with seizures (see chapter 5). Participants completed a series of standardised self-report questionnaires assessing: anxiety (Generalised Anxiety Disorder 7, GAD-7), depression (Neurological Disorders Depression Inventory for Epilepsy, NDDI-E), illness perception (Brief - Illness Perception Questionnaire, B-IPQ), HRQoL (NEWQOL-6D), and seizure frequency and severity (Liverpool Seizure Severity Scale questionnaire, LSSS-3). A prior sample size calculation was conducted. This was based on a correlation of moderate strength ≥0.5. This figure was grounded in the literature - Novakova et al. found that in patients with PNES (n=50) the B-IPQ was correlated with measures of mental (-0.697, p<0.01) and physical (-0.442, p<0.01, the two averaged 0.57) HRQoL using the Short Form-36 (Novakova et al., 2015). In epilepsy, Shallcross et al. reported in 70 patients that the B-IPQ was moderately correlated (-0.54, p<0.05) with HRQoL – Quality of Life in Epilepsy Inventory-31 (QOLIE-31-P) (Shallcross et al., 2015). The sample size calculation specific to the current study revealed that a sample of 31 individuals with epilepsy or PNES was required. The data from 45 participants with PNES and 62 with epilepsy were included. The findings from the study demonstrated that, individuals with epilepsy reported higher HRQoL and scored lower on measures of depression and anxiety. Participants with PNES perceived their condition as more threatening overall. In both conditions, HRQoL was negatively correlated with more severe illness perception and psychological distress. In epilepsy and PNES, psychological distress (epilepsy: 27%; PNES: 24.8%) and illness perception (epilepsy: 23.1%; PNES: 23.3%) accounted for the largest amount of variance in HRQoL. Clinical factors were found not to be significant predictors, while demographic factors significantly predicted HRQoL in epilepsy (12.6%), but not in PNES. Our findings support the notion that psychological factors are a stronger predictor of HRQoL in epilepsy and PNES, than condition-related and demographic variables. Prior research suggests that anxiety and depression are key predictors of HRQoL (Taylor et al., 2011; Jones et al., 2016); this study demonstrates that the relationship between illness perception and HRQoL is similarly close. These findings highlight the importance of addressing patients’ beliefs and perceptions about their condition.

Of course, the results from this research are somewhat exploratory. Patient reported outcomes (PROs) are a validated and standardised measure of patient’s perceptions of their own health and wellbeing. Given that epilepsy (and PNES) can have a multidimensional impact on daily life, it is argued that a range of PROs should be used to evaluate the different areas of patient’s lives (e.g. social, emotional, employment, stigma-related, psychological, physical) (Nixon et al. 2013). In this study, a range of PROs was utilised and their limitations must be considered. Each component constituting the factor “illness perception” was investigated using only one question. The full Illness Perception Questionnaire for example asks 50 questions, as opposed to eight in the B-IPQ. Using a greater number of questions would have allowed further inferential statistics to be conducted to investigate the internal consistency and reliability (i.e. using Cronbach’s alpha) of the dataset. What is more, this questionnaire has not been validated in those living with epilepsy or PNES. The questionnaires investigating symptoms of psychopathology reported thresholds based on associations i.e. those who score above a certain value are more likely to experience mood symptoms compared to those who score below the cut off. The same criticism can also be applied to the measure of seizure severity. There is no grading currently available for the measure that was used to explore patients’ HRQoL, the NEWQoL-6D. This means that we are unable to differentiate between clinical and statistical significance. For example, if a measure such as the EQ-5D or SF-36 had been utilised, then the value could have been further analysed to identify between group differences on the basis of the likely clinical significance of different levels of HRQoL. What is more, the present study was only correlational in design and therefore we cannot argue causality. For example, an intervention study could be conducted targeting illness perceptions exploring differences between pre and post-scores on HRQoL. Such a study would allow statistical significance to be calculated, but also the use of other methodologies, such as the Leeds Reliable Change Index (Agostinis et al., 2008), which would have permitted clinical relevance to be explored, or a cost-utility analysis to determine whether the treatment is cost-effective. Finally, there is some overlap between specific concepts measured by the self-report questionnaires. For example, the NDDI-E is a screening tool for depression in people with epilepsy and the NEWQoL-6D asks patients how often they “have problems with depression”. As such, it is likely that the scores of these items are going to be highly correlated, which may have affected the findings. Such overlap of concepts can result in collinearity or multicollinearity – whereby one variable in a multiple regression model is closely related to another variable. While measures were undertaken in this study to control for such an outcome (i.e. tolerance and VIF scores were all within acceptable limits), it is still a problem that needs to be acknowledged nonetheless. Using a measurement that asks a greater number of questions would have reduced the likelihood of violating the assumption of collinearity, thus improving the reliability of the findings.

Our health beliefs have an important impact on our subsequent engagement in health behaviours. They can be used to explore reasons why individuals engage, positively or negatively, in treatments. For instance, compared to those with epilepsy, individuals with PNES viewed their treatment as being less effective at controlling their symptoms. This is likely to have a negative reaction on an individual’s motivation to attend and adhere to treatments.

The bio-psychosocial model provides a practical and integrative framework when trying to understand how patients manage living with a seizure disorder. The model helps to identify factors that impact adjustment, which can in turn, be targeted by treatments. By comparing the results between patients with epilepsy or PNES, we can determine which associations are similar across samples, and also investigate factors that may be unique to living with the specific seizure disorder. The findings from this study suggest that psychological variables have an influential role in predicting health in individuals living with epilepsy or PNES. These factors would be overlooked or devalued if for example, the patient was treated bio-medically. This demonstrates the importance of taking into account patients’ beliefs about the different aspect of their condition.

Notwithstanding the limitations associated with the PRO measures used to investigate anxiety and depression in the current study, the findings are consistent with previous research suggesting that, symptoms related to anxiety or depression are typically the most important predictors of HRQoL in both seizure conditions (Taylor et al., 2011; Jones et al., 2016). Given this importance, it must be recognised that a large proportion of individuals with epilepsy or PNES reported higher anxiety and depression scores compared to normative data from the general public (Table 9.3). This highlights the prevalence of symptoms of psychopathology in patients living with seizures. More research is needed to investigate the causal and/or correlational nature of depression and anxiety in these conditions, and how it can best be managed.

Table 9.3 Prevalence of anxiety and depression in those with PNES or epilepsy compared to the general population

|  |  |  |  |
| --- | --- | --- | --- |
|  | PNES (n=45) | Epilepsy (n=62) | Normative data from the general public |
| Anxiety (Generalised Anxiety Disorder- 7) | None (17.8%)  Mild (33.3%)  Moderate (13.3%)  Severe (35.6%) | None (43.5%)  Mild (29%)  Moderate (14.5%)  Severe (13%) | n=5,030  None (70.5%)  Mild (23.5%)  Moderate (4.7%)  Severe (1.3%)  (Löwe et al., 2008) |
| Depression | Risk of major depression episode (75.6%) - (Neurological Disorders Depression Inventory for Epilepsy) | Risk of major depression episode (43.5%) - (Neurological Disorders Depression Inventory for Epilepsy) | Review of 10 high-income countries – lifetime prevalence of DSM-IV/CIDI major depression episode (14.6%)  (Kessler and Bromet, 2013) |

CIDI = Centre for International Disaster Information

**Chapter 5**

This chapter reported a pilot randomised controlled trial of a home-based writing intervention for individuals living with epilepsy or PNES. Writing therapies have been used in clinical and non-clinical samples, and have found to be associated with a range of qualitative and quantitative benefits (Frattaroli, 2006; Merz et al., 2014). The writing intervention was designed on the basis of Pennebaker and Beall’s FEW paradigm (Pennebaker and Beall, 1986), however, the questions were adapted from the literature investigating writing therapies for clinical populations. Individuals were either randomised to the control (n = 25) or therapeutic writing group (n = 43). Participants in the control group were asked to write about their daily events, whereas individuals allocated to the therapeutic condition were instructed to write about:

* Their very deepest thoughts and feelings about their condition (1);
* A letter to their condition (2);
* A letter to their younger self (3);
* About a personal value and why it is important to them (4).

Participants were asked to write for at least 20 minutes per question, at home, in private and on separate days. Individuals were recruited from the Royal Hallamshire Hospital and from membership led organisations. A mixture of qualitative (Writing Task Questionnaire) and quantitative (GAD-7, NDDI-E, B-IPQ, NEWQOL-6D, LSSS-3, Writing Task Questionnaire) measures were used to investigate the acceptability, feasibility and preliminary effectiveness of the intervention at one and three-months follow-up. The baseline scores were analysed in chapter 4. The key findings were that, based on rates of attrition, a writing intervention in individuals with epilepsy or PNES was found to be acceptable. Individuals in both writing conditions, on average, wrote for a longer duration than 20 minutes per session and expressed a desire to disclose such information suggesting an unmet treatment need. Compared to those in the control writing condition, those in the therapeutic condition reported having engaged in greater emotional and cognitive expression, and felt that writing had had a positive effect on them. By analysing qualitative feedback, those in the therapeutic condition reported that writing had a range of benefits (this was also reported in the control condition, although to a lesser extent). Individuals in the therapeutic condition demonstrated a significant improvement in HRQoL one-month following the writing intervention (medium effect size = 0.65). In conclusion, writing is an inexpensive and easily accessible form of therapy. While a number of benefits of writing were reported, future research should aim to investigate the use of writing as a supplement to other psychological therapies in those with a seizure disorder.

Although this was a pilot study and the main objective was not concerned with efficacy of the intervention, some preliminary conclusions came be made. The findings suggest that expressing ones thoughts and emotions about living with a seizure disorder is related with an improvement in HRQoL. Whilst we do not know the specific mechanisms that may be responsible for this association (although a number of different modes of action have been proposed), we do know that suppressing and inhibiting subjective experiences can be harmful resulting in deleterious effects. Cole et al. for example, found in a sample of 222 gay men, those who concealed their homosexual identity exhibited a greater risk of contracting a respiratory infection when compared to men who were more open about their sexual preference (p=0.001) (Cole et al., 1996).

It is important to investigate why individuals living with a seizure may feel the need to suppress their subjective experiences. When reviewing the literature on the link between emotion and pain, Lumley et al. suggest that emotional inhibition may, in part, stem from the negative reactions patients have received when expressing their emotions to other individuals (including healthcare professionals) (Lumley et al., 2011). For example, in patients who are treated bio-medically, their emotional needs that are connected to living with an illness will be ignored or underrated, whereas in the bio-psychosocial model, emotions are recognised as an influential factor in determining the patients’ state of health and should be managed appropriately. Indeed, there is evidence to suggest that, supporting individuals with epilepsy to recognise and accept the emotional aspect of their condition is beneficial. Acceptance and commitment therapy (ACT) is based on the idea that, instead of avoiding or challenging your thoughts and emotions, you develop skills that allow you to accept and be mindful of your experiences (in addition to applying behaviour change strategies). Lundgren et al. demonstrated that, individuals with epilepsy (n=13) who attended two sessions of ACT therapy reported improvements in seizure frequency, seizure duration and HRQoL at pre, post and one year follow-up (Lundgren et al., 2006). While there is no published empirical evidence to suggest the useful of ACT in individuals with PNES, the fact that ACT has been used to treat patients with other somatic disorders supports its use as a potential treatment option in this population (Cope et al., 2017).

**Chapter 6, 7 and 8**

The final chapters investigated the subjective experience of living with epilepsy or PNES by thematically analysing and comparing written accounts of living with these conditions. Qualitative methodologies can produce data that is fine grained and rich detail as participants are able to describe, in their own words, what is important to them. Previous qualitative research exploring the lived experience of epilepsy of PNES has predominantly relied on datasets collected using interviews. In comparison, writing is an individual act allowing for private exploration and expression, and has previously been used to reveal insights into the phenomenology of other conditions (Kleinman, 1988). This dataset was collected in the context of a randomised controlled trial (chapter 5). The written accounts from those allocated to the therapeutic writing group were analysed. In the writings of those with epilepsy (chapter 6), five key-themes emerged from the data:

* Theme one: ‘seizure onset’ demonstrated that the development of seizures and subsequent diagnosis was an important event that could change an individuals’ identity.
* Theme two: ‘seizure symptoms’ revealed that participants externalised their seizures as an intrusive agent with a constant presence in their lives.
* Theme three: ‘treatment and outcome’ reflected medication as an essential means to controlling seizures with subsequent side effects being perceived as a compromise.
* Theme four: ‘living with epilepsy’ explored the consequences of the condition, including restrictions and related-stigma.
* Theme five: ‘displays of coping’ demonstrated that, for the most part, participants were keen to present themselves as living well with epilepsy.

In those with PNES (chapter 7), six main-themes emerged from the data:

* Theme one: ‘living with PNES’ demonstrated that all participants presented the condition as having a debilitating effect.
* Theme two: ‘emotions’ revealed that many individuals were struggling with anxiety, low mood and self-worth.
* Theme three: ‘seizure symptoms’ shown variability was a prominent feature in the description of ictal events.
* Theme four: ‘treatment and outcomes’ demonstrated that individual’s perception towards diagnosis and therapy differed greatly.
* Theme five: ‘causation and development’ revealed that most participants spontaneously reported experiencing a traumatic event in the past.
* Theme six: ‘lack of understanding’ from themselves, the public and healthcare professionals appeared to pose considerable challenges.

In chapter 8, a thematic comparison was conducted between the writings of those with epilepsy and individuals with PNES. Participants with epilepsy (n = 20) and PNES (n = 19) were matched on sample size, age, gender, education, HRQoL (NEWQOL-6D), seizure severity (LSSS-3), and words written. Thematic differences were observed between the two conditions (PNES and epilepsy):

* Theme one: ‘causation’ revealed that those with PNES explored the possible causes of their condition throughout their narratives. In epilepsy, individuals described the onset of epilepsy as coming out of the blue, and did not ruminate about the cause.
* Theme two: ‘emotions’ demonstrated individuals with epilepsy appeared emotionally stable, whereas the writings of those with PNES reflected anxiety and low mood.
* Theme three: ‘seizure symptoms’ showed differences in ictal experiences and how seizures were conceptualised. In epilepsy, seizures were described as being external agents following the individual around, whereas in PNES, seizures were a state or place that participants entered.
* Theme four: ‘treatment’ explored the differences in the diagnostic journey and experiences of healthcare professionals. In epilepsy, healthcare professionals were described as being supportive and a source of seizure management, in contrast, those with PNES felt let down and ostracised by healthcare professionals.
* Theme five: ‘daily life’ revealed that individuals with epilepsy were keen to portray themselves as coping with their condition, whereas those with PNES felt their disorder had destroyed their lives.

Overall, the findings from chapter 6, 7 and 8 add to the growing research applying qualitative methodologies to investigate the phenomenology of epilepsy and PNES. In epilepsy, seizures and the sequelae of the condition were both conceptualised as something that must be fought, whereas those with PNES would rarely use-fighting terminology, and were more likely to describe their seizures as a state or place that they enter. The results demonstrate that differences can be observed between the two diagnoses based on participants’ written accounts. Qualitative approaches can provide insights into an individuals’ inner world, as well as offer clinical implications.

The act of formulating and sharing our story of illness is an important process in recovery. Narrative based approaches are centered on the idea that stories of illness are more than a representation of something pathological; but through telling our story of suffering we are able to heal, restore order and come to terms with what has happened (White and Epston, 1990). The role of the healthcare provider is to share an understanding of the patient’s account, wherein their symptoms are acknowledged, validated and accepted untransformed. To that end, patient’s accounts are a reflection of their life story and trajectory, which can then be used to give meaning to the diagnosis and guide treatments (Epstein et al., 1999). The findings from the narratives of individuals with PNES investigated here are consistent with the previous literature, in that a large proportion of individuals with PNES reported a past trauma (Reuber, 2008b). What is more, compared to those with epilepsy, participants seemed to perceive their condition as a reactive consequence of life events. Notwithstanding the fact that the aim of the bio-psychosocial model is to assess illness rather than to explain it, these results are in line with the framework it offers suggesting multiple and interacting causes could contribute to the development of a medical diagnosis, and that the mind and body are not separate entities as proposed by the bio-medical model because psychosocial trauma could lead to physical manifestations.

As a result of accounting for social and psychological tenets of illness, we can gain a greater understanding of the non-biological consequences and burden of living with a medical condition. Patients with PNES or epilepsy reported experiencing condition-related stigma in the current study. Whilst we await research to investigate stigma in those with PNES, it has been well documented across different cultures that, epilepsy-related stigma is a strong correlate and an important predictor of HRQoL (Taylor et al., 2011; McLaughlin et al., 2008; Jacoby, 2002; Lim et al., 2009). Stigma can operate at different levels. Internalised-stigma is where individuals discriminate against themselves because for example, they come to believe the negative stereotypes that are associated with their condition. In epilepsy, internalised stigma has been associated with increased levels of fear, anxiety and seizure severity, and a greater need for information and support relating to epilepsy (Austin et al., 2014). Externalised-stigma refers to the experience of being discriminated against by others. In 2000, a study investigating over 5,000 adults living with epilepsy in European countries reported that 51% of individuals had previously experienced feelings of stigma (Baker et al., 2000). What is more, qualitative studies exploring the phenomenology of epilepsy have helped to expose the detrimental impact that actual, perceived and feared-stigmatisation can have on individuals, with many reporting that they have been the victim of negative stereotypes, discriminative behaviour and prejudicial acts (Kerr et al., 2011).

Further research is needed into the cause and consequence of stigma in these conditions as it is likely to have an impact on a range of daily activities as well as how individuals engage with medical professionals. For instance, all illnesses, no matter how major or minor, are associated with emotional, psychological and social implications – as suggested by the bio-psychosocial model. However, Epstein et al. make the point that, healthcare professionals seem to focus on the biological factors first, and if they cannot be used to explain the observed clinical manifestations, then they move onto explore emotional or social factors. Conversely, if the exploration of psychosocial issues became standard practice in the management of all conditions (and all patients), not just those that are unexplained, then these disorders would be less stigmatised (Epstein et al., 1999). In the case of PNES for example, from the patients’ perspective, the high levels of stigma associated with mental health disorders (Corrigan and Watson, 2002) may be one of the reasons why individuals present their symptoms in a certain way. Indeed, it is conceivable that the fear of being diagnosed with a mental health condition (i.e. the associated stigma) contributes to patients with PNES resisting or dismissing psychosocial interpretations of their symptoms, and instead conceptualise their problems as physical (Whitehead et al., 2013; Monzoni et al., 2011). In epilepsy, the focus of care on observable factors (seizure frequency) may mean that psychosocial implications of the condition are viewed as less important or devalued which may account for the greater prevalence of symptoms of psychopathology compared to normative datasets (Table 9.2).

**9.2 Limitations**

The bio-psychosocial model includes patients’ subjective experiences in an attempt to provide patients with more *power* and importance in the clinical process. As a result, this has had implications on the patient-doctor relationship helping, in part, to transform the patient’s position in clinical encounters from a passive character (second to their biology) to the leading role, in which the goal of care is to treat the patient, not just the illness (Borrell-Carrió et al., 2004). Whilst the objective of this thesis was to investigate patient’s accounts of their condition, it has failed to explore healthcare professionals’ experiences (attitudes, views, behaviour) and how the two (which can be very contrasting at times) influence one another. For example, although published over 25 years ago, Lambert reported the percent of improvement in patients attending psychotherapy as a function of therapeutic factors, showing variables associated with the therapist (e.g. empathy, understanding, alliance, relationship) accounted for 30% of the outcome (Lambert et al., 1986; Lambert, 1992) (Figure 9.1). It is easy to see from the findings published elsewhere investigating healthcare providers’ views towards PNES, how their input is likely to have an influential factor on the patients state of health. Case and point, Sahaya et al. surveyed 94 healthcare professionals reporting that, only 57 believed that PNES were beyond the patient’s control. While 75% of neurologists believed this (n=12/16), nearly half of the in-patient nurses who were asked believed that PNES were fake (n=16/31) (Sahaya et al., 2012). More research is required to investigate the cause (and effect) of such attitudes and how they can be best addressed. Dworetzky asked 131 epilepsy specialists how they teach residents about PNES - 62 reported that they focus on the wasted resources caused by PNES. The authors suggest that this is likely to teach or reinforce the idea that those with PNES are wasting doctors’ time whilst those with epilepsy are the ones most deserving of their attention (Dworetzky, 2015).

Figure 9.1 Factors associated with improvements in psychotherapy

One of the aims of this thesis was to explore distinguishing features and the profile between unselected cases of PNES and epilepsy (and syncope in chapter 3). However, due to the modest sample sizes, differences in subjective experiences between different subtypes of the same condition were not investigated. For example, it is conceivable that individuals with different types of epilepsy or syncope (or indeed, in those who experience different manifestations or experiences associated with PNES) would report different levels of ictal panic symptoms. What is more, we are unable to make distinctions between non-clinical factors, for example, it is likely that individuals with adult onset epilepsy will have experienced other challenges and require different support needs compared to those with early onset epilepsy, i.e. these individuals will have had to adjust from an adult without epilepsy to one with the condition.

Although we did confirm the diagnosis of all those recruited from the Royal Hallamshire Hospital, and attempted to confirm seizure diagnoses using the medical records for participants living in the UK who were recruited via membership-led organisations, we could not confirm all diagnoses and so the inclusion of individuals with self-reported, rather than proven diagnoses of epilepsy or PNES could be seen as weakness of this thesis – nor can we confirm that all diagnoses were made using a gold-standard approach (Lafrance et al., 2013). However, it has previously been shown that a large proportion of individuals who receive the diagnosis of PNES in the UK never undergo confirmatory tests, such as video-EEG (Mayor et al., 2011). Notwithstanding this risk, while introducing a modest element of diagnostic uncertainty, our mixed (seizure clinics/membership-led organisations) recruitment strategy allowing individuals with self-declared diagnoses to participate is likely to have provided us with a more representative picture of individuals with seizure disorders than a hospital outpatient sample could have done. This may make the findings more generalisable than if individuals had been recruited from a single center.

The dataset analysed in chapters 4, 6, 7 and 8 were collected in the context of a randomised controlled trial that aimed to investigate the benefits of a therapeutic writing intervention (chapter 5). Therefore, it is likely that we will only have captured experiences from a subgroup of individuals, for instance, those who value psychological approaches, feel that they need additional support, and are able to (intellectually, physically and emotionally) write about their experiences of living with seizures. For example, findings from self-reported measures suggest that there is at least a subgroup of individuals with PNES who exhibit marked difficulties in the ability to express and describe feelings and bodily sensations (Bewley et al., 2005; Myers et al., 2013).

The search terms used in the qualitative synthesis (chapter 2) could have been expanded upon. For example, the phenomena “subjective experiences”, “lived experiences”, and “phenomenology” could have been further divided to include terms such as “opinions”, “attitudes”, “views”, “impressions”, “behaviours”, “perceptions” and “emotions”. This would have helped to broaden and specify the search. Also, a greater use could have been made of Boolean operators (AND, OR). The quality of the search strategy will affect the papers included as well as those that have been missed out, ultimately impacting the trustworthiness and outcome of the review. Although three key sources were used (six large databases, the references and citations of accepted journals, and the authors’ knowledge of papers) more resources could have been utilised, for example, a number of experts in the field could have been contacted and asked if they are aware of any suitable papers, given our aims, that we had not otherwise identified, as well as the use of unpublished studies, abstracts, posters and book chapters. The inclusion of such grey literature could have provided a greater breadth of patient’s experiences, however, at the same time it may have reduced the reliability of the reviews findings due to the fact that these studies may not have been subjected to a peer-review process. On a similar note, the papers included in the current review were not assessed or rated based on the quality of their methodology. For example, the Critical Appraisal Skills Programme could have been used to assess the trustworthiness, results and relevance of the studies included in the synthesis. Based on the ratings, greater importance could have been given to findings from studies that scored highly, and less weight to poorer quality studies.

The delivery and improvement of healthcare and policy requires a broad range of qualitative and quantitative evidence that has been collated across different healthcare contexts (Tong et al., 2012). As such, the value of qualitative research is becoming more recognised. While it has become commonplace for reviews to be conducted with the aim to amalgamate quantitative data, methods are much less developed in the area of synthesising qualitative findings. Methods for synthesising qualitative research are still emerging and while there is no consensus on which methodological approaches should be used, it is clear that there is no single approach that is best for all reviews. Instead, the most suitable method will depend on factors such as the review’s aim, the data and ontological perspectives (Thomas and Harden, 2008). In chapter 2, a systematic synthesis was utilised. This means that similarities and differences between the findings in each study in the review were examined and grouped together. Thomas and Harden define this process as “translation”. This data is then synthesised into clear and concise themes that represent the findings. This is a subjective process undertaken by the researcher who should have become immersed in the data. While certain measures can be taken to check for reliability i.e. in the current review, the themes and sub-themes were shared between the authors and any disagreements were discussed until a consensus was formed, the outcome of the analysis is open to interpretation and discussion. For instance, in theme four *emotional events*, emotive events (e.g. past trauma or abuse, current stressors) and emotional experience (e.g. processing emotions and symptoms of depression), although represented by separate sub-themes, were grouped under the same main theme. The reason for this is that, when patients described their experiences of past trauma or current anxieties, they did not simply produce factual narrations of their experience. Instead, as discussed in the review, patients would describe the emotions that are associated with such life events as well as their feelings regarding the association between the event and the development of PNES. However, it could be argued that these experiences (emotions and traumatic events) are deserving of their own theme and therefore should be explored individually despite being linked.

It is a limitation of this thesis that, in chapter 3, a prior sample size calculation specific to investigating the frequency of ictal related panic symptoms in those with epilepsy, PNES or syncope was not performed. This study was a retrospective analysis of the data collected in response to seven questions from the Paroxysmal Experience Profile questionnaire, which were originally gathered for the purpose of another project entitled: *Development of a self-report and seizure witness observation tool – the Paroxysmal Experience Profile and Paroxysmal Event Observation Questionnaires.* A power calculation performed for that study reported at least 87 patients were needed in each group – this analysis was conducted by a statistician from the Sheffield Teaching Hospital Research Department and not by the candidate, G.R. Performing a post-hoc sample size calculation specific to the observed effect size in the mean difference in the panic composite score between individuals with PNES (n=90, mean 16.8, standard deviation, 6.7), epilepsy (n=93, mean 12.5, standard deviation 6.7) and syncope (n=98, mean 10.7, standard deviation 4.9), demonstrated that, using a two tailed hypothesis, adopting the alpha level of 0.05, and a power value of 0.8 (this value is often used in practice), sufficient power to detect a difference between individuals with PNES and epilepsy would have required a sample size of n=40 in each group (Cohen’s *d* = 0.64), a difference between PNES vs. syncope n = 16 in each group (*d=* 1.04), and epilepsy vs. syncope n=165 in each group (*d =* 0.31). These post-hoc calculations suggest that the study was sufficiently powered to find a difference between PNES and epilepsy or syncope.

In the thematic comparative analysis, the researcher, G.R., who was responsible for exploring the emergent themes, was not blinded to the patients’ diagnosis. Although the researcher aimed to work deductively (the themes were grounded in the data) and reflectively (i.e. acknowledging and taking note of any impressions and reflections that emerged due to outside influences), this may still have somewhat biased his interpretation of the findings. Given the parameters and purpose of this thesis, it was not possible to maintain blinding of the researcher - this researcher was responsible for screening patients’ medical records for eligibility and, participants would make explicit references to their condition in their narrative, for example, *“ I am actually very embarrassed to basically sum up how my pseudo seizures have affected me in all sorts of ways.”* Future research however, could look to validate or explore the differences in written accounts that have been reported here using different methodologies. For example, a number of authors could undertake the thematic analysis and *argue* reliability using measures of inter-rater reliability, also researchers blinded to the patient’s diagnosis could explore their written accounts and attempt to predict medical diagnosis based on the outcomes that have been reported here and elsewhere (Rawlings and Reuber, 2016) - in fact, using this approach, Reuber et al. demonstrated that two linguistics blinded correctly predicted the diagnosis of 17 out of 20 patients (PNES or epilepsy) based on a range of interactional and linguistic features (Reuber et al., 2009).

As explained in chapter 2, there is no single best qualitative method, and the most appropriate method for a given question depends on a range of factors including ontology, epistemology and focus of interest (Ritchie et al., 2014). Whilst we could have used a range of different analytical methods (i.e. interpretive phenomenological analysis, framework analysis) it was determined that thematic analysis would be utilised. Not only is this a suitable and appropriate method, and has previously been used to explore the subjective accounts of individuals living with epilepsy (Kerr et al., 2011) or PNES (Rawlings and Reuber, 2016), but it also helped to provide the basis for an additional study that was undertaken based on the written accounts. The candidate (G.R.) has also performed a narrative analysis, using Frank’s approach which aims to identify narrative typologies (Frank, 1995). Narrative analysis is governed by two general principles. Firstly, it does not only segregate or fragment words into discrete categories that can then be coded. Instead or in addition, it takes a holistic approach, interpreting the story as a whole, which generates insights far beyond what can be deduced from single sentences alone. In a narrative, both content (i.e. themes, events) and form (i.e. the way it is told) hold importance and should be explored. Having said that, in order to be in a position to examine the structure of a story I first had to choose a suitable approach to investigate the content. I followed Riessman who suggested that thematic analysis is appropriate for this task (Riessman, 2008).

It is a limitation that in chapters 6, 7 and 8, qualitative data was not collected until theoretical saturation was achieved. This means that I failed to reach a point in the analysis where collating more data would not lead to more information being found, specific to the research question under investigation. Saturation is a method used in qualitative research (especially in grounded theory where the approach is deductive) to increase the reliability of the analysis. For example, it is at the point of saturation where repeated evidence has been provided that supports the emergent themes. The primary reason that saturation was not reached in these studies is because the anchor questions were open and non-directive. For example, individuals were given prompts and instructed that they “could” or “may” want to write about certain topics, and as such, the possible responses were endless. Indeed, some individuals answered the prompt line-by-line in a systematic manner; others began to answer the question but then their own ideas, views and feelings started to emerge in the text (similar to what is observed in a free association narrative interview); while a small proportion focused solely on the most salient aspects of their condition which were unique to their situation largely ignoring the anchor question. In these chapters, the number of individuals who described a certain experience has been reported, for example, *“16/19 participants spontaneously reported a past trauma, such as abuse (e.g. physical, sexual, emotional) or a distressing event (e.g. surgery, bereavement)”.* This provides context to the emergent theme, for instance, demonstrating its (un)commonness. However, the reader should realise that these numbers have to be interpreted with caution. The fact that 16/19 contributors may have mentioned a particular theme does not make this theme more prominent or important than one that was highlighted as most important by 8/19 for example. As long as readers bear this limitation in mind, these values still carry some meaning in a qualitative study. The fact that 16/19 contributors mentioned a particular theme without specific prompting does suggest that this theme was something that had at least some relevance to many contributors.

**9.3 Future research**

Potential areas of future research were outlined in the specific chapters, however some are also presented here.

Firstly, it was reported in the systematic synthesis of qualitative research into PNES that, the methods used to gather data has been limited, predominantly relying on clinical or research interviews – this is also the case (albeit to a lesser extent) in research investigating epilepsy. Other approaches to gather verbal and non-verbal qualitative data may be successfully applied to explore the phenomenology of epilepsy and PNES. Indeed, the lived accounts of epileptic seizures have been investigated using poetry (Featherstone and Sandfield, 2013) and drawings (Stafstrom and Havlena, 2003; Featherstone et al., 2013). In chapters 6, 7 and 8 it was demonstrated that asking individuals to write about their experiences could gather new insights. Furthermore, prompting individuals with epilepsy or PNES about particular ictal symptoms (for instance, those which are difficult to describe, such as symptoms of depersonalisation and derealisation) may produce a more detailed account of seizure experiences than the open questions typically employed in qualitative research. For example, Watson et al. reports that, when interviewing patients who were in Seattle (United States) during an earthquake, 23% of 26 patients with epilepsy spontaneously reported that they thought they were having a seizure during the earthquake, whereas no patient with PNES (n = 22) made this claim – however once directly asked, five patients with PNES thought the earthquake was a seizure (Watson et al., 2002). This approach of course needs to be balanced with the risk of directing or influencing the individual, for example though the use of leading questions.

On a similar note, in addition to the need of different methods to collect data, greater diversity in analytical methods is also required. Findings from the qualitative analyses in the current thesis (chapter 6 and 7) suggested that, sub-groups of individuals could be identified based on their subjective accounts. Narrative investigations have become widely used in other field of medicine (Atkinson and Rubinelli, 2012). Narrative analysis takes a holistic approach investigating transcripts in their entirety in an effort to determine how the narrator has created ordered understandings of events in their life. The focus in narrative analysis is on the content of people’s experiences, as well as the structure - how the narrator frames their experiences (Riessman, 1993; Frank, 2010). Different approaches of narrative analysis have been put forward to examine how illness narratives are expressed and constructed. Frank proposes a method that identifies narrative types, or storylines, that underlie the plot within stories of illness (Frank, 1995). This approach compares commonalities and differences across a collection of illness stories to examine if types of narrative forms can be identified. For example, Frank offers a three-fold typology: *restitution narrative* which bears a resemblance to the sick role - the individual is healthy, becomes ill, seeks treatment, and health is restored; *chaos narrative* is the most difficult narrative to hear, the story lacks structure as the narrator struggles to define a beginning, middle and end, it is an “anti-narrative” because topics do not necessarily lead to another, the individual can be overwhelmed by the distress caused by their illness and express little hope; and last but not least, a *quest narrative* in which the individual conceptualises illness as a metaphorical journey, where the traveler will gain insight and reflect on their experiences, and return with a “boon”. Although Frank only defines three typologies, he explains that others can and should be identified. In fact, different narrative typologies have already been proposed in individual with mental illness (Carless and Douglas, 2008), cancer (Evans et al., 2012), human immunodeficiency virus (HIV) (Mosack et al., 2005), and those who have survived a stroke (France et al., 2013) or self harm (Chandler, 2014). However, this method has yet to be used to investigate individuals with epilepsy or PNES. It is important to recognise that this approach to narrative analysis is not to identify types of people, but rather to understand the plot and tensions of individual stories. Moreover, narratives are transient as people can move between different typologies depending on the moment in time. It is also likely that narratives will overlap, as no storyline will conform seamlessly within a narrative typology. Including narrative medicine into clinical practice can allow care to be more personalised and responsive to the patients’ needs (Atkinson and Rubinelli, 2012). For example, Frank explains that listening to stories of illness can be difficult, as the plot can be disordered and weaved together from multiple stories. As such, clinicians and family members can use narrative typologies as a guide or reference to aid listening to patients’ stories. What is more, narrative typologies can help patients to better understand their experiences, and to distinguish between the story that they are telling and the story that they want to tell (Frank, 2010).

Finally, given that PNES are a mental disorder manifesting with seizures superficially resembling that of epilepsy, it is not surprising that insights from the qualitative study (chapter 7) examining written accounts of living with PNES suggest that many individuals experience distressing levels of stigma attributable to their condition. However, to date, studies that have reported PNES-related stigma (including what is reported in the current thesis) only do so indirectly and stigma has not been the study objective in any published works relating to PNES. There is a notable lack of in-depth qualitative or quantitative investigations intended specifically to explore the stigma attached to PNES. This means that very little is actually known about the nature, prevalence, and perception of stigma associated with this condition. The relative dearth of research is particularly striking when the literature on PNES-related stigma is compared to that on epilepsy or mental health disorders – both found more commonly in individuals with PNES than expected in the general population (Reuber, 2008a; Brown and Reuber, 2016). Looper and Kirmayer demonstrated that individuals with a functional somatic syndrome (chronic fatigue syndrome, fibromyalgia and irritable bowel syndrome) report a greater degree of perceived stigma when compared to those with a “comparable” medical condition with a clear pathology (multiple sclerosis, rheumatoid arthritis and inflammatory bowel disease) (Looper and Kirmayer, 2004). These authors suggested that their results may have been, at least in part, due to the uncertainty or ambiguity about the cause of functional symptoms - there is certainly greater acceptance and understanding of epilepsy than PNES, even within the medical community (Worsely et al., 2011). Extrapolating these findings, it is conceivable that stigma may be more prevalent in those with PNES compared to epilepsy.

**9.4 Conclusions**

In summary, using qualitative and quantitative research methodologies, this thesis explored the subjective experience of living with epilepsy or PNES highlighting the diagnostic, prognostic and therapeutic implications. When reaching the final diagnosis to explain TLoC, all available evidence should be used. To that end, the findings in chapter 3 are consistent with other research suggesting that, in addition to objective and visible manifestations, subjective experiences should also be taken into consideration. The importance of managing individuals’ beliefs about their condition in treatment pathways has been demonstrated in chapter 4. This approach to care would help to address the full spectrum of challenges associated with living with a seizure disorder. In chapter 5 it was shown that writing about experiences of living with seizures was associated with a range of benefits. However at this stage, further research is needed to investigate the role that therapeutic writing could play in treating those with a seizure disorder, such as a phase III RCT design. Finally, qualitative research allows individuals to use their own words, which can give us a deeper insight into their inner world. We have further expanded on such findings in chapters 6, 7 and 8 outlining the relevance of listening to such voices. Such an outcome of their written accounts is no more important than working to eradicate the negative experiences of healthcare services described by individuals with PNES. This group of individuals, as like any other, need and are deserving of a caring, compassionate and comprehensive approach to their disorder

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**Appendices**

## **Appendix 1 Paroxysmal Experience Profile Questionnaire**

1) I am Male

Female

2) I am ….. years old

3) I was ….. years old when I had my first attack/blackout/seizure/turn?

4) My most recent attack was……………………..ago

(Please complete if you have had an attack in the last 12 months)

In the last year I have had an average of………. ….attacks a week

………...……...attacks a month

………...……...attacks a year

5) In my lifetime I have been taken to hospital with an attack….

Once

Between 2 and 5 times

More than 5 times

6) I have been treated on intensive care for at least one attack?

Yes No

7) Members of my close family have had ……..

Attacks Yes No

Blackouts Yes No

Epilepsy Yes No

8) Over the years my attacks have been diagnosed by a doctor as……..

Fainting spells

Epileptic seizures

Caused by low blood sugar

Non-epileptic attacks

Functional seizures

Dissociative Attacks

Pseudoseizures

Panic Attacks

Syncope

(Please tick more than one box if different diagnoses have been made)

9) I think the correct diagnosis is

10) The words I use to describe my attacks/turns/blackouts/seizures are

………………………………………………

11) I think my attacks are caused by

………………………………………………

12) I have had the following health problems in addition to my attacks (please tick if appropriate)?

Febrile seizures in childhood

Meningitis

Head injury with loss of consciousness

Heart attack

Chest pain or tightness

Palpitations

Other heart condition

Stroke

Brain tumour

Diabetes

Brief jerks of my arms and legs when I am drifting off to sleep

Brief jerks of my arms and legs at other times

Blank spells lasting a few seconds

Lightheaded spells

Poor coordination

Breathlessness unrelated to exercise

Hot flushes

13) This is how I would describe my attacks:

For each statement below tick the box numbered 1 to 5 where 1 means that you strongly agree and 5 means that you strongly disagree. If you do not know the answer to the question you can tick column 3 (“I neither agree nor disagree”).

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| No. |  | I very strongly agree  1 | I agree  2 | I neither agree nor disagree  3 | I disagree  4 | I very strongly disagree  5 |
| 1 | My attacks often come on when I am asleep |  |  |  |  |  |
| 2 | I am aware of a trigger for my attacks |  |  |  |  |  |
| 3 | My attacks may be triggered by the site of blood or needles |  |  |  |  |  |
| 4 | My attacks are associated with sitting or standing for a long time |  |  |  |  |  |
| No. |  | I very strongly agree  1 | I agree  2 | I neither agree nor disagree  3 | I disagree  4 | I very strongly disagree  5 |
| 5 | My attacks come on when I pass water or open my bowels |  |  |  |  |  |
| 6 | My attacks are associated with emotional stress |  |  |  |  |  |
| 7 | My attacks come on out of the blue without any warning |  |  |  |  |  |
| 8 | When I feel an attack coming on I try to fight it |  |  |  |  |  |
| 9 | My attacks well up inside me |  |  |  |  |  |
| 10 | My attacks come over me |  |  |  |  |  |
| 11 | In my attacks I seem to be controlled by someone outside me |  |  |  |  |  |
| 12 | My attacks build up gradually |  |  |  |  |  |
| 13 | In my attacks I feel a rising sensation in my body |  |  |  |  |  |
| 14 | In my attacks I sometimes have a sense of feeling as if I’ve seen something before when I know I have not |  |  |  |  |  |
| 15 | In my attacks I sometimes have a sense of feeling as if I’ve never seen something before when I know I have |  |  |  |  |  |
| 16 | In my attacks I feel lightheaded, like I might pass out |  |  |  |  |  |
| 17 | Just before my attacks I feel anxious or nervous |  |  |  |  |  |
| 18 | Just before my attacks I feel irritable or upset |  |  |  |  |  |
| 29 | In my attacks I sometimes feel sick |  |  |  |  |  |
| 20 | I am aware of my heart racing in my attacks |  |  |  |  |  |
| 21 | I feel hot or cold in my attacks |  |  |  |  |  |
| 22 | My head spins in my attacks |  |  |  |  |  |
| 23 | In my attacks I experience tingling or numbness in my skin |  |  |  |  |  |
| 24 | In my attacks my vision goes dim or dark |  |  |  |  |  |
| 25 | In my attacks sounds are distorted |  |  |  |  |  |
| 26 | In my attacks everything seems to move away from me |  |  |  |  |  |
| 27 | During my attacks I see things which are not really there |  |  |  |  |  |
| 28 | During my attacks I hear things which are not really there |  |  |  |  |  |
| 29 | During my attacks I smell things which are not really there |  |  |  |  |  |
| 30 | My attacks cause a bad taste in my mouth |  |  |  |  |  |
| 31 | In my attacks my mouth goes very dry |  |  |  |  |  |
| No. |  | I very strongly agree  1 | I agree  2 | I neither agree nor disagree  3 | I disagree  4 | I very strongly disagree  5 |
| 32 | In my attacks memories seem to flash into my mind |  |  |  |  |  |
| 33 | During my attacks I can see or hear the people around me |  |  |  |  |  |
| 34 | During my attacks I have no idea what is happening  around me |  |  |  |  |  |
| 35 | In my attacks I am conscious but I can’t react to things |  |  |  |  |  |
| 36 | In my attacks I have the feeling that there is something which I am trying to hold onto |  |  |  |  |  |
| 37 | In my attacks I drift in and out of consciousness |  |  |  |  |  |
| 38 | During an attack, I sometimes hear voices inside my head |  |  |  |  |  |
| 39 | During an attack, I sometimes feel as if I am two different people |  |  |  |  |  |
| 40 | During an attack, I sometimes have the feeling that my body is not my own |  |  |  |  |  |
| 41 | I am aware of shaking uncontrollably during an attack. |  |  |  |  |  |
| 42 | During my attacks everything seems as if it’s an imitation of reality |  |  |  |  |  |
| 43 | During an attack, I sometimes feel as if other people, objects, and the world are not real |  |  |  |  |  |
| 44 | During my attacks I feel as if I’m in a stage set |  |  |  |  |  |
| 45 | During an attack, I sometimes do not recognise my friends or family |  |  |  |  |  |
| 46 | During my attacks I feel as if I am outside my body |  |  |  |  |  |
| 47 | During an attack, I sometimes see myself as if looking at another person |  |  |  |  |  |
| 48 | During my attacks I feel as if I am looking at myself from outside |  |  |  |  |  |
| 49 | During my attacks I feel as if I’m unreal |  |  |  |  |  |
| 50 | During my attacks I feel as if I’m not a person |  |  |  |  |  |
| 51 | During my attacks I feel as if I’m not in the living world |  |  |  |  |  |
| 52 | My attacks make me feel like I have lost time |  |  |  |  |  |
| 53 | My attacks make time go in slow motion |  |  |  |  |  |
| 54 | During my attacks I feel as if I’m in a dream |  |  |  |  |  |

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| No. |  | I very strongly agree  1 | I agree  2 | I neither agree nor disagree  3 | I disagree  4 | I very strongly disagree  5 |
| 55 | During my attacks I have memories of a past bad experience which I can’t stop |  |  |  |  |  |
| 56 | During my attacks I have distressing dreams of a past bad experience |  |  |  |  |  |
| 57\* | During my attacks I feel very frightened |  |  |  |  |  |
| 58 | In my attacks I feel like I am choking or very short of breath |  |  |  |  |  |
| 59\* | During my attacks I feel that something terrible might happen |  |  |  |  |  |
| 60 | My attacks feel like someone is trying to attack or kill me |  |  |  |  |  |
| 61\* | During my attacks I am frightened that I am going to die |  |  |  |  |  |
| 62\* | During my attacks I am frightened that I will lose control |  |  |  |  |  |
| 63\* | During my attacks I am frightened that I will go crazy |  |  |  |  |  |
| 64\* | During my attacks my heart pounds and I feel shaky and sweaty |  |  |  |  |  |
| 65\* | During my attacks I feel that I have to get out of the situation |  |  |  |  |  |
| 66 | My attacks feel like a kettle boiling over |  |  |  |  |  |
| 67 | My attacks feel like a fuse blowing |  |  |  |  |  |
| 68 | My attacks are like a burst of electricity in my brain |  |  |  |  |  |
| 69 | My attacks are painful - like a hammer blow |  |  |  |  |  |
| 70 | My attacks feel like a knife through the head |  |  |  |  |  |
| 71 | My attacks feel like my head going under water |  |  |  |  |  |
| 72 | My attacks make me feel very low or empty |  |  |  |  |  |
| 73 | I find the thought of being in a attack disgusting |  |  |  |  |  |
| 74 | I want to know what happens when I have blacked out |  |  |  |  |  |
| 75 | I am glad I don’t know what happens when I have blacked out |  |  |  |  |  |
| 76 | I sometimes wake from my attacks with a cut tongue |  |  |  |  |  |
| 77 | After my attacks I sometimes find I have burned myself |  |  |  |  |  |
| 78 | After my attacks I am often drenched in sweat |  |  |  |  |  |

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| No. |  | I very strongly agree  1 | I agree  2 | I neither agree nor disagree  3 | I disagree  4 | I very strongly disagree  5 |
| 79 | After my attacks I feel relieved |  |  |  |  |  |
| 80 | After my attacks I feel drained or sleepy |  |  |  |  |  |
| 81 | After my attacks my muscles ache |  |  |  |  |  |
| 82 | After my attacks I feel very confused |  |  |  |  |  |
| 83 | Afterwards I have no idea that I have had an attack |  |  |  |  |  |
| 84 | After the attack I feel very upset |  |  |  |  |  |
| 85 | After an attack, I sometimes find things among my belongings which I don’t remember buying |  |  |  |  |  |
| 86 | After my attacks, I sometimes find myself somewhere but don’t know how I got there |  |  |  |  |  |

\* Represents questions used to investigate panic symptoms

## **Appendix 2. Personal Information Questionnaire**

PERSONAL INFORMATION

Please answer the following questions.

Patient ID: ……………………………………… (to be completed by the investigator)

1. Todays date: …………………………
2. Full name: ……………………………………………………………………..

3. Date of birth: ………………………….

4. Gender: O I am male

O I am female

O Other

1. Address including postcode: ……………………………………………………….

…………………………………………………………………………………………

1. Telephone number: …………………………………………………………………
2. Mobile number: …………………………………………………………………….
3. Email address: …………………………………………………………………………
4. Preferred method of contact:

O Phone

O Email

O Post

1. Work: (please tick correct option) O School/college

O University

O Employed O Self-employed O Unemployed O I receive disability benefits O Retired on health-grounds O I receive an old-age pension

1. For how many years in total did you go to school/college/university?

………. years

1. What is your current diagnosis? (please tick)

O Epilepsy

O Non-epileptic Attack Disorder (Psychogenic non-epileptic seizures, dissociative seizures)

O Both

O Not sure

1. For how many months or years have you had seizures?

(for example 6 months or 3 years) ………………. months

………………. Years

1. When did you have your last seizure? Date: …………………………..
2. Are you a current patient under the care of a neurologist at the Royal Hallamshire Hospital, Sheffield UK?

O Yes

O No

O Not sure

1. What is the name and contact details of your GP (General Practitioner)?

………………………………………………………………………………………..

………………………………………………………………………………………..

1. As explained in the Participant Information Sheet, which way would you like to take part in the study?

O Option 1 – Handwrite

O Option 2 – Type using a computer

O Option 3 – Not sure

## **Appendix 3 Generalised Anxiety Disorder – 7 (GAD-7)**

GAD 7

This questionnaire asks for your views about **anxiety.**

Over the last **2 weeks**, please tick how often have you been bothered by the following problems?

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  | **Not at all** | **Several days** | **More than half the days** | **Nearly every day** |
| 1. Feeling nervous, anxious or on edge? | O | O | O | O |
| 1. Not being able to stop or control worrying? | O | O | O | O |
| 1. Worrying too much about different things? | O | O | O | O |
| 1. Trouble relaxing? | O | O | O | O |
| 1. Being so restless that it is hard to sit still? | O | O | O | O |
| 1. Becoming easily annoyed or irritable? | O | O | O | O |
| 1. Feeling afraid as if something awful might happen? | O | O | O | O |

## **Appendix 4 Neurological Disorder Depression Inventory For Epilepsy (NDDI-E)**

This questionnaire asks for your views about **depression.**

For the statements below, please tick the number that best describes you over the last **2 weeks** including today

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  | **Always or often** | **Sometimes** | **Rarely** | **Never** |
| 1. Everything is a struggle | O | O | O | O |
| 1. Frustrated | O | O | O | O |
| 1. Nothing I do is right | O | O | O | O |
| 1. Feel guilty | O | O | O | O |
| 1. Difficulty finding pleasure | O | O | O | O |
| 1. I’d be better off dead | O | O | O | O |

## **Appendix 5 Brief-Illness Perception Questionnaire (B-IPQ)**

This questionnaire asks for your views about **illness**

For the statements below, please circle the number that best describes you.

|  |
| --- |
| 1. How much does your illness affect your life?   0 1 2 3 4 5 6 7 8 9 10  No affect Severely  at all affects my life |
| 1. How long do you think your illness will continue?   0 1 2 3 4 5 6 7 8 9 10  A very Forever  short time |
| 1. How much control do you feel you have over your illness?   0 1 2 3 4 5 6 7 8 9 10  Absolutely Extreme  no control amount of  control |
| 1. How much do you think your treatment can help your illness?   0 1 2 3 4 5 6 7 8 9 10  Not at all Extremely  helpful |
| 1. How much do you experience symptoms from your illness?   0 1 2 3 4 5 6 7 8 9 10 No symptoms Many severe  at all symptoms |
| 1. How concerned are you about your illness?   0 1 2 3 4 5 6 7 8 9 10  Not at all Extremely  concerned concerned |
| 1. How well do you feel you understand your illness?   0 1 2 3 4 5 6 7 8 9 10  Don't Understand  understand very clearly  at all |
| 1. How much does your illness affect you emotionally? (e.g. does it make you angry, scared, upset or depressed?)   0 1 2 3 4 5 6 7 8 9 10  Not at all Extremely  affected affected  emotionally emotionally |
| 1. Please list in rank-order the **three most important factors** that you believe caused your illness.   The most important causes for me:   1. \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ 2. \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_   3. \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ |

## **Appendix 6 Health-Related Quality of Life (NEWQOL-6D)**

This questionnaire asks for your views about seizures (please tick one answer).

1. How worried are you that you might have another attack?

O I am **not** worried at all

O I am a **little** worried

O I am **fairly** worried

O I am **very** worried

1. How often do you have problems with depression?

O I **never** have problems with depression

O I **rarely** have problems with depression

O I **sometimes** have problems with depression

O I **always or often** have problems with depression

1. Thinking about your memory:

O I **never** have problems with my memory

O I **rarely** have problems with my memory

O I **sometimes** have problems with my memory

O I **always or often** have problems with my memory

1. Thinking about your concentration:

O I have **no** problems concentrating for more than a short period of time

OI have **mild** problems concentrating for more than a short period of time

OI have **moderate** problems concentrating for more than a short period of time

O I have **serious** problems concentrating for more than a short period of time

1. How much control do you feel you have over things that happen to you?

OI have **complete** control over things that happen to me

OI have **some** control over things that happen to me

OI have **little** control over things that happen to me

OI have **no** control over things that happen to me

1. How much do you feel people treat you as an inferior person?

O I do **not** feel that people treat me like an inferior person

O I feel that some people **maybe** treat me like an inferior person

O I feel that some people **probably** treat me like an inferior person

O I feel that some people **definitely** treat me like an inferior person

## **Appendix 7 Liverpool Seizure Severity Scale (LSSS-3)**

So we can better understand the frequency and severity of your seizures, please complete the following questionnaire about your seizures.

1. How often have you experienced seizures in the **past year?**

*Please enter “0” if you have not experienced any seizures in the past year. Please give an estimate of the number of seizures you usually had in a day, a week, or a month (for example, “1 seizure per week” or “10 seizures per month”).*

seizures per (day/week/month)

1. How many seizures have you had in the last **4 weeks?**

*Please enter “0” if you have not experienced seizures in the last 4 weeks. If you cannot remember the exact number of seizures you’ve experienced, please estimated based on the number you usually had during a single day or week.*

seizures

Please complete the follow questions thinking about the most severe seizure you experienced during the **past 4 weeks**. Please tick one.

1. I feel that my most severe seizures have mostly been:

O Very severe

O Severe

O Mild

O Very mild

1. Most commonly when I blank out/lose consciousness:

O I blank out for less than 1 minute

O I blank out for between 1 and 2 minutes

O I blank out for between 3 and 5 minutes

O I blank out for more than 5 minutes

O I never blank out/lose consciousness

1. When I have most severe seizures, I smack my lips, fidget, or behave in an unusual way:

O Always

O Usually

O Sometimes

O Never

1. After my most severe seizures:

O I feel very confused

O I feel fairly confused

O I feel slightly confused

O I do not feel confused at all

1. After my most severe seizures my confusion lasts for:

O Less than 1 minute

O Between 1 and 5 minutes

O Between 6 minutes and 1 hour

O 1 to 2 hours

O More than 2 hours

O I never feel confused

1. When I have my most severe seizures:

O I always fall to the ground

O I usually fall to the ground

O I sometimes fall to the ground

O I never fall to the ground

1. After my most severe seizures:

O I always have a headache

O I usually have a headache

O I sometimes have a headache

O I never have a headache

1. After my most severe seizures:

O I always feel sleepy

O I usually feel sleepy

O I sometimes feel sleepy

O I never feel sleepy

1. After my most severe seizures:

O I always find that I have wet myself

O I usually find that I have wet myself

O I sometimes find that I have wet myself

O I never find that I have wet myself

1. After my most severe seizures:

O I always find that I have bitten my tongue

O I usually find that I have bitten my tongue

O I sometimes find that I have bitten my tongue

O I never find that I have bitten my tongue

1. After my most severe seizures:

O I always find that I have injured myself (other than biting my tongue)

O I usually find that I have injured myself (other than biting my tongue)

O I sometimes find that I have injured myself (other than biting my tongue)

O I never find that I have injured myself (other than biting my tongue)

1. After my most severe seizures I can usually return to what I am doing in:

O Less than 1 minute

O Between 1 and 5 minutes

O Between 6 minutes and 1 hour

O 1 to 2 hours

O More than 2 hours

## **Appendix 8 Implementation Intention Questionnaire**

WRITING SESSION TIMETABLE

Please write down the day, time and location where you will complete each writing session, *for example Tuesday at 7pm in my bedroom.*

*Please remember all session need to be completed within two weeks in the correct order*

…………………………………………………………………………………………..

I intend to complete the **1st** writing session on:

Day: ……………………….. Location: ………………………

Time: ………………………

…………………………………………………………………………………………..

I intend to complete the **2nd** writing session on:

Day: ……………………….. Location: ………………………

Time: ………………………

…………………………………………………………………………………………..

I intend to complete the **3rd** writing session on:

Day: ……………………….. Location: ………………………

Time: ………………………

…………………………………………………………………………………………..

I intend to complete the **4th** writing session on:

Day: ……………………… Location: ………………………

Time: ……………………..

## **Appendix 9 Writing Task Questionnaire**

WRITING TASK QUESTIONNAIRE

For the following questions, please cross or circle the number that best describes you

1. To what extent was your writing **personal**?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. To what extent was your writing **meaningful** and **valuable** to you?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. To what extent was your writing revealing of your **feelings** and **emotions**?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. To what extent did you write about your **thoughts**?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. How much have you wanted to talk to others about the information you have written about?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. How much **have** you talked to others about the information you have written about?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |
|  |  |  |  |  |  |  |

1. How much has writing had a **positive effect** on you?

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. Please rate the severity of distress, if any, you experienced **whilst** writing.

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |
|  |  |  |  |  |  |  |

1. Please rate how **better you feel** after yourself after writing.

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Not at all |  |  | Some |  |  | A lot |

1. You may want to describe how you think and feel your writing went.

............................................................................................................................

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## **Appendix 10 Therapeutic Writing Booklet**

SESSION ONE

I would like you to write about your very deepest thoughts and feelings about your condition, disorder and/or seizures. In your writing you can write about anything you wish, but it is important that you really let go and explore your thoughts and feelings. You may want to write about how your condition, disorder and/or seizures makes you feel, and how it affects your day-to-day life. You could write about how it has affected your relationships with others such as family members, friends, neighbours and work colleagues, or how your family, friends, doctors, nurses, therapists, paramedics and even complete strangers have reacted to your condition, disorder and/or seizures. How has it affected your independence and financial stability, your work life and career? How has it affected the way you feel about yourself, your self-esteem and how others see you? This is a chance to tell your story about what it is really like in your inner-world living with your condition, disorder and/or seizures.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

SESSION TWO

I would like you to write a letter to your condition, disorder and/or seizures. You can address your letter however you like and discuss anything you wish, for example, Dear Seizures. In your letter you may want to confront or challenge your condition, disorder and/or seizures. You could write about what you really think of it and how you really feel… be honest and try not to hold back. You can use swear words if you wish. You could write about how your thoughts and feelings towards it have changed over time, for example, how you first felt when you were diagnosed, when you first developed the symptoms or when you were going through a difficult time in your life. What are your thoughts and feelings towards it now? What do you want your thoughts and feelings to be like in the future? In your letter try and recognise what it has taught you about yourself, the good and bad. What have you learned about who you are as a person and what your strengths and weakness are? You may want to thank it for any benefits or insight it has provided you with. This is your opportunity to talk to your condition, disorder and/or seizures, so what do you want it to know?

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

SESSION THREE

I would like you to write a letter to your younger self. This version of you can be any age you wish; they may even be a pre-seizure version of you who has no idea they will be later diagnosed nor have the knowledge of what it is like to live with your condition, disorder and/or seizure. You can address your letter however you like and discuss anything you wish, for example, Dear Pre-Seizure Me or, Dear 16 year old me. In your letter you could write about what you know now, that you didn’t know then. What have you discovered about yourself? Think about the lessons you have had to learn along the way so the younger version of you doesn’t have to make the same mistakes. What should you have done or shouldn’t have done and why? In your letter try and consider what advice or guidance you would give your younger self. You may want to provide instruction on how to cope or manage with your condition, disorder and/or seizure in emotional and also practical everyday terms. When writing your letter try and consider the person who you were then and who you are now, how much you have grown and developed. I want you to show compassion for your younger self, telling them how to take care and nurture themselves drawing upon all the wisdom you have gained.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

SESSION FOUR

Please read the following list of common personal values.

I would like you to think about and circle which **three** matter **most** to you? Which do you most naturally use to guide your choices? Which do you appreciate most about yourself? What is it that makes you, you?

Perhaps what matters **most** to you isn’t listed and is something else (after all this is not a full list), in that case, please think about what does matter to you most.

Your career Membership to a social group

Relationships with friends and family Your happiness

Your faith and religious values Your independence

Sense of humour Your creativity

Your strength/ability to cope with difficulties Your generosity

Your honesty Your enthusiasm

Other(s):

I would like you to choose **one** of your three values and write in-depth about why it is important to you, why it is central to your life and how it makes you feel about yourself? You may want to describe a time in your life when you had the opportunity to really express this value.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

## **Appendix 11 Control Writing Booklet**

SESSION ONE

I would like you to write about **what you did over the last week**. Please focus your writing on your **actions and behaviour**. Be as descriptive as possible focusing on facts and events as they happened.

The most important thing is for you to describe your time over the last week as **accurately** and as **detailed** as possible.

**Date started: ………………………..**

**Time started: ………………………..**

SESSION TWO

I would like you to write about **what you did over the last 24 hours**. Please focus your writing on your **actions and behaviour**. Be as descriptive as possible focusing on facts and events as they happened.

The most important thing is for you to describe your time over the last 24 hours as **accurately** and as **detailed** as possible.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

SESSION THREE

I would like you to write about **what you plan to do over the next 24 hours**. Please focus your writing on your **actions and behaviour**. Be as descriptive as possible, focusing on facts and events as you expect and plan them to happen.

The most important thing is for you to describe your time over the next 24 hours as **accurately** and as **detailed** as possible.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

SESSION FOUR

I would like you to write about **what you plan to do over the next week**. Please focus your writing on your **actions and behaviour**. Be as descriptive as possible, focusing on facts and events as you expect and plan them to happen.

The most important thing is for you to describe your time over the next week as **accurately** and as **detailed** as possible.

**Date started: ………………………..**

**Time started: ………………………..**

**Time finished: ………………………..**

## **Appendix 12 Writing Instructions**

INFORMATION

Please read this page before starting the writing session

Writing therapy is a unique and personal process, which means there is no best way of writing that will work for everyone. However, we have provided some guidance that will help you.

Guidance for writing

* The challenge of writing therapy is **not** to allow outside distractions to disrupt your writing time. So find a time and place where you will not be disturbed, and which is away from your mobile phone and the TV.
* Write continuously for at **least** 20 minutes.
* Try and complete the sessions one day after another (consecutively), although you must complete all four sessions within **two weeks** of completing the first session.
* Do not worry about your writing style, punctuation, spelling or grammar
* No feedback will be given on what you have written about so write only for yourself, this is all kept confidential and no one will be able to identify you
* Write about something personal and important to you
* Deal only with events or situations you feel are manageable to write about now, if you are becoming too distressed change what you are writing about or stop writing.
* You **MUST** complete the writing booklet in order i.e. 1, 2, 3 then 4.
* Do not open the writing session until you are ready to begin writing

INSTRUCTIONS

You can either take part in this study in two ways (please chose the option which you feel most comfortable with, which is most natural and easiest for you):

**Option 1** **-** Handwriting your response

When you are ready to start a writing session please open the booklet. You will first see a question. Please read the question carefully to make sure you fully understand it. You should aim to read it several times. Then spend the next 20 minutes (at least) answering the question. Should you finish writing before the 20 minutes, please keep writing. You may want to go into even more detail about what you have already written about. **Please also write down the time you start and the time you finish.** We have provided you with space to write but please feel free to use more paper if you need. Once you are finished, there is a short questionnaire at the back of the booklet that you should fill out. This will ask you about your experience of writing. Once this is completed, place the booklet back in the free post envelope. Once all four writing sessions are completed, please seal the free post envelope and send it back to us.

**Option B -** Typing your response

When you are ready to start a writing session please access the webpage:

>inset URL here<

When you access the webpage you will see four webpage links that will take you to a writing session. When you click on the link you will see a question. Please read the question carefully to make sure you fully understand it. You should aim to read it several times. Then spend the next 20 minutes (at least) answering the question. Should you finish writing before the 20 minutes, please keep writing. You may want to go into even more detail about what you have already written about. **Please also write down your unique patient ID number, the time you start and the time you finish**. Once you are finished, there is a short questionnaire at the bottom of the web page that you should fill out. This will ask you about your experience of writing. **Once this is complete please press submit.**

…….……………………………………

Please remember we will contact you in **one** and **three months** time to ask you to complete some more questionnaires.

Thank you.

Below you will find a list of contact details of organisations that can offer further help and support should you become distressed or anxious concerning anything you have written about or by the questionnaires you have completed. If you are a patient under the current care of a neurologist in Sheffield Teaching Hospital you can contact the neurologist for advice. Other patients are reminded to contact their general practitioner (GP) for advice. You can also find the researchers contact details should you need further information or help with the writing tasks.

Useful Contacts

NHS Direct

Tel: 0845 46 47

Website: [www.nhsdirect.nhs.uk](http://www.nhsdirect.nhs.uk)

Mind, the mental health charity

Tel: 0300 123 3393

Website: [www.mind.org.uk](http://www.mind.org.uk)

Samaritans

Tel: 08457 90 90 90

Website: [www.samaritans.org](http://www.samaritans.org)

Breathing Space

Tel: 0800 83 85 87

Website: [www.breathingspacescotland.co.uk](http://www.breathingspacescotland.co.uk)

Epilepsy Action

Tel: 0808 800 5050

Website: <https://www.epilepsy.org.uk>

Epilepsy Society

Tel: 01494 601 400

Website: <http://www.epilepsysociety.org.uk>

Researchers Contacts

Gregg Rawlings

Address: N152, Academic Neurology Unit, Royal Hallamshire Hospital, Glossop Road, Sheffield, S10 2JF

Tel: 0114 2711597

Email: [ghrawlings1@sheffield.ac.uk](mailto:ghrawlings1@sheffield.ac.uk)

**Appendix 13 Invitation Sheet - Sheffield Teaching Hospital**

[Sheffield Teaching Hospitals NHS Foundation Trust](http://www.sth.nhs.uk/index.php)

Dear Sir/Madam,

**Re: Developing a writing intervention for patients with seizures (a home-based study)**

We are currently conducting a research study at the Royal Hallamshire Hospital in Sheffield to test whether a writing intervention can help people with seizures feel more in control of their condition, reduce their seizures and improve their quality of life.

You have been identified as someone who could take part in this study because you are currently receiving treatment to help with seizures at the Royal Hallamshire Hospital.

A participant information sheet is enclosed with this letter. We are sending this information sheet to you so that you can find out about the study and think about taking part. Please read the information sheet to help you to understand what the study will involve and to provide you with time to think about what your involvement in the study would mean to you.

Some of the data from this study will be used by a postgraduate student of the University of Sheffield as part of an educational project.

If you have any questions please do not hesitate to contact the research student (Gregg Rawlings) on 0114 2711597.

Your clinical care will not be affected in any way if you do or do not take part in this study. If you do decide to take part in the study you will be free to withdraw at any time.

Kind Regards,

Professor Markus Reuber Gregg Rawlings

Honorary Consultant Neurologist PhD Student

**Appendix 14 Invitation Sheet – Membership led organisations**

**Developing a writing intervention for patients with seizures (a home-based study)**

We are currently conducting a research study at the Royal Hallamshire Hospital in Sheffield to test whether writing can help people with seizures feel more in control of their condition, reduce their seizures and improve their quality of life. The knowledge gained from this study will also further contribute to a better understanding of what it is like to live with seizures.

For this study, we are looking to recruit people who are over the age of 18 and who have a medical diagnosis of "epilepsy", "non-epileptic attack disorder" (also known as psychogenic non-epileptic seizures or dissociative seizures) or both "epilepsy and non-epileptic attack disorder". Participants must also be literate in English, able to write and complete a series of questionnaires without any assistance, and who can provide informed consent. People taking part in this study will have to write four times for twenty minutes about different topics.

Some of the data from this study will be used by a postgraduate student of the University of Sheffield as part of an educational project.

If you would like further information about the study please first read the "Participant Information Sheet", the link for which can be found at the side of this webpage. After reading the "Participants Information Sheet" you are still interested in taking part please complete the two forms on the "Register as a Participant" web page.

If you would like more information about the study, please do not hesitate to contact the research student (Gregg Rawlings) whose details can be found below.

Your clinical care will not be affected in any way if you do or do not take part in this study. If you do decide to take part in the study you will be free to withdraw at any time.

Kind Regards,

Professor Markus Reuber (Honorary Consultant Neurologist)

Gregg Rawlings (PhD student)

Gregg Rawlings, Academic Neurology Unit, University of Sheffield, Royal Hallamshire Hospital, Glossop Road, Sheffield S10 2JF Email: ghrawlings1@sheffield.ac.uk, Telephone: 0114 2711597

**Appendix 15 Participant Information Sheet - Sheffield Teaching Hospital**

# university of sheffield.gif

# [Sheffield Teaching Hospitals NHS Foundation Trust](http://www.sth.nhs.uk/index.php)PARTICIPANT INFORMATION SHEET

**Title of Project: Developing a writing intervention for patients with seizures (a home-based study)**

**Name of Researchers: Gregg Rawlings, Prof Markus Reuber, Prof Brendan Stone & Dr Ian Brown**

*We would like to invite you to take part in a research study. Before you decide whether to take part, you should understand why the research is being done and what it would involve for you. Please read the following information carefully and talk to others about the study if you wish. Please contact us if there is anything that is not clear or if you would like more information. Please take time to decide whether or not you wish to take part. Thank you for reading this.*

**Background**

Epilepsy and non-epileptic attack disorder (NEAD) are chronic, disabling conditions that are characterised by seizures. People living with these disorders are also at an increased risk of developing other symptoms such as depression, anxiety and pain, such as headaches. Research has shown in patients with other chronic conditions (such as arthritis and asthma), that writing may help to reduce symptoms and improve physical, and psychological health. Although writing is a simple and easy form of therapy, the health benefits have yet to be investigated in patients with seizures.

This study is being carried out as part of a PhD research project based at the University of Sheffield.

**What is the purpose of the study?**

The purpose of the study is to test whether a writing intervention can help reduce symptoms of epilepsy and NEAD including: seizure frequency and severity, quality of life, anxiety, depression and illness perception. We are also interested in what you write about in the hope it will generate deeper insights and understanding of living with a seizure disorder.

**Why have I been asked to take part?**

We are approaching people who have experienced seizures and who are receiving treatment to help with seizures at the Royal Hallamshire Hospital in Sheffield. We are asking people with epileptic seizures as well as people with non-epileptic attacks to take part in this study.Right now, we only want to inform you about the study. You do not have to decide whether you want to take part until you go to the hospital for your appointment in the neurology outpatient clinic, where you will have an opportunity to ask a researcher any questions you have about the study.

**Do I have to take part?**

It is your decision whether or not to take part. If you have any questions about this study you can contact us or write them down and ask the researcher on the day of your clinic appointment. If you do decide to take part you are free to withdraw at any time, without giving a reason. This would not affect the standard of care you receive in any way.

**What will happen to me if I take part?**

When you attend your appointment in the neurology clinic at the Royal Hallamshire Hospital, you will have a chance to ask questions. We will then ask you to sign a consent form to record your agreement to take part. At the time of your appointment, you will also be asked to complete a set of questionnaires. This should take no longer than 15 minutes.

You can take part in the study in two ways: option 1 is if you would like to handwrite, option 2 is if you would like to type using a computer. You will receive both sets of material so you can decide which option is best for you at a later date.

**Option 1 (handwriting)**

You will receive four writing booklets. Each booklet will contain a topic (question) you will be asked to write about, space for you to write and a short questionnaire. You will be asked to complete one booklet each day at home, making sure all four are completed within two weeks. We ask that you spend at least 20 minutes writing about each topic.

Once you have finished each booklet, you will be asked to complete a short questionnaire asking how you felt the writing went. Once all four booklets are complete you will be asked to place the booklets back in the free post envelope that we will provide and send them back to us.

We will also provide a set of instructions containing important contact details and guidance to help you with your writing.

**Option 2 (computer typing)**

You will receive an information sheet with the details of a website address. When you access the webpage you will see four webpage links that will take you to a writing session. Each session will contain a topic (question) you should write about, space for you to write and a short questionnaire. You will be asked to complete one session each day at home, making sure all four are completed within two weeks. We ask that you spend at least 20 minutes writing about the topic.

Once you have finished writing you will be asked to complete a short questionnaire at the bottom of the page asking how you felt the writing went, and then press submit.

We will also provide a set of instructions containing important contact details and guidance to help you with your writing.

…………………………

You will also be provided with a writing session timetable asking you to record the time, date and location that you intend to complete the four individual writing sessions/booklets at home.

As part of the study, one month and three months after completing the last writing session we will send you a set of follow-up questionnaires similar to what you completed on the day of your appointment.

**What are the possible benefits of this study?**

This study will help us to evaluate the benefits of a writing intervention for improving quality of life and reducing symptoms associated with seizure disorders. Also, the knowledge gained from patient’s writings will further contribute to a better understanding of what it is like to live with the individual conditions. We hope that the study proves that the writing is helpful and if so, it could be made available to other people experiencing seizures.

**What are the possible risks of taking part in this study?**

There are no significant risks associated with taking part in the study. Although we will provide you with details of services and organisations you can contact for further support. If you feel distressed or anxious during writing you can stop at any time.

**Will my taking part in this study be kept confidential?**

All the information that is collected about you during this study will be kept strictly confidential. We will keep your personal details, such as name, address and telephone number, separate and locked in a secure location. This means that your identity will be kept private. Any personal details held by us will be destroyed once the study has finished.

**What will happen to the results of the study?**

The results of this study will contribute to a PhD Thesis. We will also publish the results of the study in a scientific journal. You will not be identified individually in the write-up. If any of your writings are published they will be done so under a pseudo (fake) name and any names or locations will be changed. If you would like a summary of the results of the study once it is complete, please let us know.

As part of this study, you will be asked to complete some short screening questionnaires about anxiety and depression. If you reported high levels symptoms of anxiety or depression on these questionnaires, we will inform your GP about these findings so they can be addressed if this seems necessary and appropriate. If you are under the current care of a neurologist at the Royal Hallamshire Hospital in Sheffield we will inform your neurologists about high anxiety or depression scores as well.

**What if I change my mind?**

You do not have to take part in this study. If you have agreed to take part, you can stop at any time without giving your reasons. This will have no effect on any services you are receiving.

**Who should I contact if I have a question or need more information?**

Gregg Rawlings

Academic Neurology Unit

University of Sheffield

Royal Hallamshire Hospital

Glossop Road

Sheffield S10 2JF

Email: ghrawlings1@sheffield.ac.uk

Telephone: 0114 2711597

**What if something goes wrong?**

If you have a concern about any aspect of this study, you should ask to speak to the   
researchers who will do their best to answer your questions. If they are unable to   
resolve your concern or you wish to make a complaint regarding the study, please contact Sheffield Patient Services Team (previously known as PALS) on 0114 2712400 or Dr Philip Harvey (Registrar and Secretary, University of Sheffield) on [registrar@sheffield.ac.uk](mailto:registrar@sheffield.ac.uk) or 0114 222 1101.

**Appendix 16 Participant Information Sheet – Membership-led organisation**

# university of sheffield.gif

# [Sheffield Teaching Hospitals NHS Foundation Trust](http://www.sth.nhs.uk/index.php)PARTICIPANT INFORMATION SHEET

**Title of Project: Developing a writing intervention for patients with seizures (a home-based study)**

**Name of Researchers: Gregg Rawlings, Prof Markus Reuber, Prof Brendan Stone & Dr Ian Brown**

*We would like to invite you to take part in a research study. Before you decide whether to take part, you should understand why the research is being done and what it would involve for you. Please read the following information carefully and talk to others about the study if you wish. Please contact us if there is anything that is not clear or if you would like more information. Please take time to decide whether or not you wish to take part. Thank you for reading this.*

**Background**

Epilepsy and non-epileptic attack disorder (NEAD) are chronic, disabling conditions that are characterised by seizures. People living with these disorders are also at an increased risk of developing other symptoms such as depression, anxiety and pain such as headaches. Research has shown in patients with other chronic conditions (such as arthritis and asthma), that writing may help to reduce symptoms and improve physical and psychological health. Although writing is a simple and easy form of therapy, the health benefits have yet to be investigated in patients with seizures.

This study is being carried out as part of a PhD research project based at the University of Sheffield.

**What is the purpose of the study?**

The purpose of the study is to test whether a writing intervention can help reduce symptoms of epilepsy and NEAD including seizure frequency and severity, quality of life, anxiety, depression and illness perception. We are also interested in what you write about in the hope it will generate deeper insights and understanding of living with a seizure disorder.

**Why have I been asked to take part?**

We are approaching people who are over the age of 18 years old, who have a medical diagnosis of either epilepsy or NEAD (psychogenic nonepileptic / dissociative seizures) or both, who are literate in English, able to write and complete a series of questionnaires without any assistance, and who can provide informed consent.

If you agree to take part, we will ask for your consent for us to contact your doctor to confirm your medical diagnosis.

**Do I have to take part?**

It is your decision whether or not to take part. If you have any questions about this study you can contact us on the contact details provided at the end of the Participant Information Sheet. If you do decide to take part you are free to withdraw at any time, without giving a reason. This would not affect the standard of care you receive in any way.

**What will happen to me if I take part?**

You will be asked to sign a consent form to record your agreement to take part. You will also be asked to complete a set of questionnaires. This should take no longer than 15 minutes.

You can take part in the study in two ways: option 1 is if you would like to handwrite, option 2 is if you would like to type using a computer. You will receive both sets of material so you can decide which option is best for you at a later date.

**Option 1 (handwriting)**

You will receive four writing booklets. Each booklet will contain a topic (question) you should write about, space for you to write and a short questionnaire. You will be asked to complete one booklet each day at home, making sure all four are completed within two weeks. We ask that you spend at least 20 minutes writing about the topic.

Once you have finished each booklet, you will be asked to complete a short questionnaire asking how you felt the writing went. Once all four booklets are complete you will be asked to place the booklets back in the free post envelope that we will provide and send them back to us.

We will also provide a set of instructions containing important contact details and guidance to help you with your writing.

**Option 2 (computer typing)**

You will receive an information sheet with the details of a website address. When you access the webpage you will see four webpage links that will take you to a writing session. Each session will contain a topic (question) you should write about, space for you to write and a short questionnaire. You will be asked to complete one session each day at home, making sure all four are completed within two weeks. We ask that you spend at least 20 minutes writing about the topic.

Once you have finished writing you will be asked to complete a short questionnaire at the bottom of the page asking how you felt the writing went, and then press submit.

We will also provide a set of instructions containing important contact details and guidance to help you with your writing.

…………………………

You will also be provided with a writing session timetable asking you to record the time, date and location that you intend to complete the four individual writing sessions/booklets at home.

As part of the study, one month and three months after completing the last writing session we will send you a set of follow-up questionnaires similar to what you completed on the day of your appointment. You will be asked to place them all in a free post envelope and send them back to us once completed (regardless of whether you handwritten or used a computer to write).

**What are the possible benefits of this study?**

This study will help us evaluate the benefits of a writing intervention for improving the quality of life and reducing symptoms associated with seizure disorders. Also, the knowledge gained from patient’s writings will further contribute to a better understanding of what it is like to live with the individual conditions. We hope that the study proves that the writing is helpful and if so, it could be made available to other people experiencing seizures.

**What are the possible risks of taking part in this study?**

There are no significant risks associated with taking part in the study. Although we will provide you with details of services and organisations you can contact for further support. If you feel distressed or anxious during writing you can stop at any time.

**Will my taking part in this study be kept confidential?**

All the information that is collected about you during this study will be kept strictly confidential. We will keep your personal details, such as name, address and telephone number, separate and locked in a secure location. This means that your identity will be kept private. Any personal details held by us will be destroyed once the study has finished.

**What will happen to the results of the study?**

The results of this study will contribute to a PhD Thesis. We will also publish the results of the study in a scientific journal. You will not be identified individually in the write-up. If any of your writings are published they will be done so under a pseudo (fake) name and any names or locations will be changed. If you would like a summary of the results of the study once it is complete, please let us know.

As part of this study, you will be asked to complete some short screening questionnaires about anxiety and depression. If you reported high levels of symptoms of anxiety or depression on these questionnaires, we will inform your GP about these findings so they can be addressed if this seems necessary and appropriate.

**What if I change my mind?**

You do not have to take part in this study. If you have agreed to take part, you can stop at any time without giving your reasons. This will have no effect on any services you are receiving.

**Who should I contact if I have a question or need more information?**

Gregg Rawlings

Academic Neurology Unit

University of Sheffield

Royal Hallamshire Hospital

Glossop Road

Sheffield S10 2JF

Email: ghrawlings1@sheffield.ac.uk

Telephone: 0114 2711597

**What if something goes wrong?**

If you have a concern about any aspect of this study, you should ask to speak to the   
researchers who will do their best to answer your questions. If they are unable to   
resolve your concern or you wish to make a complaint regarding the study, please contact Sheffield Patient Services Team (previously known as PALS) on 0114 2712400 or Dr Philip Harvey (Registrar and Secretary, University of Sheffield) on [registrar@sheffield.ac.uk](mailto:registrar@sheffield.ac.uk) or 0114 222 1101.

**Appendix 17 Consent form**

# university of sheffield.gif

# [Sheffield Teaching Hospitals NHS Foundation Trust](http://www.sth.nhs.uk/index.php)CONSENT FORM - Patient Participant

**Title of Project: Developing a writing intervention for patients with seizures (a home-based study)**

**Name of Researchers: Gregg Rawlings, Prof Markus Reuber, Prof Brendan Stone and Dr Ian Brown**

|  |  |
| --- | --- |
|  | **Please initial box** |
| 1. I confirm that I have read and understood the information sheet for the above study and have had the opportunity to ask questions. |  |
| 1. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without my medical care or legal rights being affected. |  |
| 1. I understand that relevant sections of my medical notes and data collected during the study may be looked at by members of the research team from the University of Sheffield, regulatory authorities or representatives from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records. |  |
| 1. I agree to provide my address to a member of the research team and I give permission to be contacted by the researcher for follow-up assessment. |  |
| 1. I agree to provide my telephone number to a member of the research team and I give permission to be contacted by the researcher to monitor progress and follow-up assessment. |  |
| 1. I agree to take part in the above study and understand that the data will be used as part of a PhD degree thesis. 2. I agree that the research team can inform my General Practitioner about evidence of likely anxiety or depression   8. I agree that the research team can contact my General Practitioner to confirm my medical diagnosis  9. I agree to the use of direct quotations in publications provided that anonymity is preserved so that I cannot be identified. |  |

\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

Name Signature

Todays date Your date of birth

Name of person taking consent Signature

**Appendix 18 Research Protocol**

**STH RESEARCH PROTOCOL**

1. **Project Details**

**Project Title**

Developing a writing intervention for patients with epilepsy and nonepileptic attack disorder

**Investigator Details**

Investigator 1 Prof Markus Reuber

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Tel. No.: 01142711597

**Sponsor Details**

STH Foundation Trust

**STH Project Reference Number**

STH18910

**Protocol Version Number and Date**

Version 1, 15/07/15 Version 2 28/10/2015

**STH Directorate Affiliation**

Neurosciences

1. **Research Question**

This study is a randomised controlled trial that will investigate the effects of a writing intervention in patients with epilepsy and patients with nonepileptic attack disorder

Primary Aims

1. The main aim is to determine whether a writing intervention can improve patients’ health-related quality of life and frequency of seizures.

Secondary Aims

1. Our secondary aims are to investigate the effects of the writing intervention on healthcare usage, levels of anxiety and depression, illness perception and seizure severity
2. The study will also investigate the patients writing using qualitative techniques with the view to generate a better understanding of what it is like to live with epileptic or nonepileptic seizures.
3. **Abstrac**t

Epilepsy and nonepileptic attack disorder (NEAD) are chronic conditions characterised by recurrent seizures and impaired consciousness. Individuals with either disorder are further associated with an increased risk of developing other psychiatric symptoms and reduced quality of life. Previous research has demonstrated in patients with other chronic illnesses (such as, cancer, HIV and fibromyalgia) that writing about thoughts and emotions, compared to writing about emotional devoid topics, can be effective in symptom management and improve physical and psychological health. Although writing is an inexpensive, easy to administer form of therapy there is no work investigating its effects on patients with seizures. The proposed study will develop and evaluate whether a writing intervention can improve the quality of life and reduce the frequency of seizure in patients with epilepsy and patients with nonepileptic attack disorder. In addition we will also use qualitative methods to explore patients’ writings to gain a deeper understanding of the subjective experience of living with seizures.

1. **Originality of Research**

A writing intervention aimed at improving symptoms in patients with seizures has never been tested. The proposed study will also explore patients’ writings using narrative analysis (a qualitative method) with the view to generate deeper insight and understanding of living with the individual conditions. This knowledge is currently lacking for the literature. The project is intended to contribute to a PhD degree.

1. **Background**

Epilepsy is a disease of the brain diagnosed on the basis of a single unprovoked seizure, plus the probability of seizure reoccurrence of at least approximately 60% or after two unprovoked seizures (Fisher et al., 2014). The prevalence of epilepsy is high, estimated between 500 - 1,000 cases per 100,000. The majority of patients can successfully control their epileptic seizures using anti-epileptic drugs (AEDs), however, approximately one third of patients fail to respond to AEDs instead relying on alternative interventions. These include, surgery, vagus nerve stimulation, special diets and psychological intervention (NICE, 2015).

Non-epileptic attack disorder (NEAD) superficially resemble epileptic seizures, however, have been conceptualised as a paroxysmal dissociative response to distress caused by internal or external stimuli. As such, manifestations of NEAD are considered to be associative of psychological processes rather than abnormal electroencephalography (EEG) activity associative of epilepsy (Reuber, 2009; Bowman, 2006). Prevalence is difficult to estimate due to delayed and incorrect diagnosis (the correct diagnosis of NEAD is typically not made for several years after seizure manifestation, (Reuber et al., 2002), but is somewhere between 2 to 33 cases per 100,000 (Benbadis and Hauser, 2000). The current treatment of choice for NEAD is psychotherapy (Lafrance et al., 2013), however, problems in diagnosis mean that most patients receive delayed care or are initially prescribed AEDs. This offers little or no effect, but rather increases the risk of iatrogenic harm (Mayor et al., 2012).

In addition to the direct manifestation of seizures, both epilepsy and NEAD are further associated with an increased prevalence of mental health symptoms. For example, in a sample of 5,835 patients with epilepsy: 18% suffered from depression, 15% neuroses, 11% anxiety and 9% with psychoses. Overall, 41% of patients were diagnosed with a psychiatric disorder (Gaitatzis et al., 2004). Based on more modest sample sizes: Patidar et al., (2013) reported comorbid anxiety (62.3%), depressive disorders (90.2%) and chronic tension type headache (42.9%) in 63 patients with NEAD and epilepsy; and in patients diagnosed with NEAD alone, Alessio et al., (2006) (n=24) reported 54% suffered a history of anxiety, 37.5% somatoform disorders, 29% post-traumatic stress disorder (PTSD) and 25% parasuicide.

In their guidelines for managing epilepsies, the National Institute of Clinical Excellence (NICE) recommend the use of psychological interventions in conjunction with other therapies (NICE, 2015). One potential advantage of combining psychological therapies alongside other treatments is that, patients may learn alternative coping strategies to manage seizure onset and comorbid symptoms. Such psychological therapies potentially exist in the form of self-help manuals, particularly writing interventions. Writing offers an economical, low-intensive and easily integrative method of coping with the emotional and cognitive demands associated with illness. Compared to more traditional psychological services, which may have limited or delayed access, writing therapies can be delivered to and reached by a far greater number of patients. However, there is a need for rigorous and robust research testing the effectiveness of writing therapies in patients with seizures before such interventions can be recommended.

The existing evidence has suggested therapy writing can offer health benefits in a range of illnesses including: cancer (Merz et al., 2014), cystic fibrosis (Taylor et al., 2003), PTSD (van Emmerik et al., 2008) and human immunodeficiency virus (HIV) (Petrie et al., 2004) and in array of comorbid symptoms such as: migraines (D'Souza et al., 2008), depression (Krpan et al., 2013) and high blood pressure (McGuire et al., 2005). FEW has also been suggested to reduce healthcare and medicine utilisation (Rosenberg et al., 2002). Several reviews have been conducted investigating FEW’s effectiveness. Smyth (1998) conducted the first meta-analysis (n=13) reporting improvements of 23% in health outcomes (*d=*0.47) whereas Frattaroli (2006), who undertook the most comprehensive review using 146 studies with a total of 10,994 participants, found only small health gains (*d=*0.15). Frisina et al., (2004) conducted a meta-analysis of FEW studies involving clinical samples (n=9) reporting a larger effect on physical (*d*=0.21) rather than psychological outcomes (*d*=0.07, p<0.05). Similarly, Merz et al., (2014) conducted a systematic review (n=13) exploring FEW in patients with cancer suggesting FEW did have a small to moderate effect when the studies were combined, in particular on sleep, pain, and general physical and psychological symptoms.

The proposed study is novel, as it will assess the effectiveness of a writing intervention specifically designed to help patients with seizures. The results will provide further evidence for the utility of writing as a self-help intervention technique. More specifically, the study will offer an empirical evaluation of the effect sizes of a self-help intervention for patients with seizures as a basis for future studies.

The study will also analyse what patients write about using narrative analysis techniques with the view to generate greater insight into subjective experience of the psychopathology and what it is liked to live with seizure disorders. This evidence may improve our understanding of experiences and how patients adapt or cope to seizure disorders, but also, potentially highlight subtle differences in narratives between epilepsy and NEAD.

If the intervention proves effective, the project will offer a simple self-help tool that could be integrated into existing intervention programmes for epilepsy and NEAD to improve the quality of life, and reduce stress, seizure severity and frequency of the patients.

1. **Plan of the Investigation**

**Design**

The study will be a randomised controlled trial. Patients will be randomised to two groups, (1) a therapeutic writing intervention (writing about emotions and cognitions) and a (2) controlled writing intervention (writing about emotionally devoid topics). A neutral writing condition is necessary to control for potential therapeutic gains for the act of writing itself i.e. affording yourself 20 minutes out from your day for creative and reflective thinking. Patients will be assessed at baseline, at one- month and at three-month follow-up.

**Participants**

Patients will be recruited from the neurology outpatient clinic and psychotherapy services at the Royal Hallamshire Hospital (UK). We will also recruit from social media websites for epilepsy and NEAD self-help (although, unless diagnosis can be confirmed data from these patients will remain separate).

**Recruitment**

*Outpatient clinic recruitment*: Patients attending the neurology outpatient clinic with a confirmed diagnosis of either epilepsy or NEAD will be invited to take part in the study. The diagnosis will be based on the clinical judgment of a consultant neurologist. Patients will receive via post an invitation letter and patient consent form including study purposes and procedure at least 48 hours in advance of their appointment in the neurology outpatient clinic.

In addition to recruitment from the outpatient clinic, we will approach patients with seizures under the current care of a Consultant Neurologist at Sheffield Teaching Hospitals NHS Foundation Trust about this study by letter. We will send information about the study and a return slip. Patients will be encouraged to contact the research team if they want to take part or find out more about the study.

Patients can also be referred to the research team by Healthcare professionals i.e. patients who have undergone psychotherapy at the Royal Hallamshire Hospital, patients attending Epilepsy Specialist Nurse clinics and patients who are attending neurology clinics in satellite clinics served by neurologists based at the Royal Hallamshire Hospital. We would like to inform these patients by advertising the study using a poster placed in clinic and waiting areas (poster is attached). We have also created a “Referral Form” which would allow psychotherapists or neurologists to inform researchers of patients who have expressed an interest in participation. The professional completing the referral form will obtain the patient’s verbal consent to pass on their contact details and for researchers to contact the patient with further information about this study. The researcher will still be consenting the patient for the study should they wish to take part.

*Social media recruitment / recruitment via self-help groups*: An invitation letter and website link to our website will be placed on social media or self-help group webpages. Patients can use the link to find out more about the study, complete the informed consent and register their interest in the study. Patients will then be contact by a member of the research team.

**Inclusion Criteria**

1. Clinically firm diagnosis of epilepsy and/or NEAD

3. Over the age of 18 years.

4. Able to complete the self-report questionnaires and writing without help.

5. Able to give informed consent.

6. Literate in English

**Exclusion Criteria**

1. People who are unable to give informed consent.

2. People who are unable to complete the self-report questionnaires and writing intervention unaided.

3. People who have not experienced a seizure within the last 12 months.

4. Illiterate in English

5. Those receiving are going to receive psychotherapy within the next four months at The Royal Hallamshire Hospital

**Procedure**

The self-help intervention for this study has been developed on the basis of review of available literature of therapeutic writing. Patients who have provided baseline data will receive the writing intervention according to the random allocation to treatment or control group. Patients can complete the intervention by either using pen and paper and sending us their writings by free post, or by typing their writing and submitting it to us using a specific website address.

The writing intervention is time limited. Patients are asked to complete four writing sessions over two weeks, ideally on consecutive days. Patients are asked to write for a minimum of 20 minutes per writing session. Each writing session has a clearly defined set of instructions that the patients should write about. After each writing session patients are asked to record the time and date they started and finished writing. They are also required to complete a questionnaire asking them to reflect on their writing experience (writing task questionnaire). This will monitor progress and adherence. Patients are given a writing intention document asking when and where they intend to complete each writing session. This has been found to enhance the effectiveness of self-help interventions (Kloss and Lisman, 2002) and has previously been used to increase medication adherence in patients with seizures (Mooney et al., 2009). Patients are provided contact details of the researchers and organisations should they experience any difficulties with the intervention.

During the intervention patients will be contacted up to two times by phone, two times by email and one letter to monitor progress and adherence. Patients will be provided contact details of the researchers and organisations should they experience any difficulties with the intervention. Follow-up will be one (T2) and three month (T3) following the last writing session. Patients will be contacted by post, telephone or email depending on consent and preference. The follow-up assessment will involve completing the same outcome measures as the baseline measures. Overall, patients will take part in the intervention for a maximum of 15 weeks.

**Outcome Measures**

*Demographic Questionnaire* This includes: name, date of birth, gender, contact details, employment status, years of education, current diagnosis, years since onset of seizures, date of last seizure, whether they are a current patient at the RHH under the care of a neurologist, and contact details of their GP.

*Healthcare utilisation* see (Mayor et al., 2010; Mayor et al., 2012) Patients will be asked to self-report how many visits they have made to the doctors (General Practitioner), outpatient departments and Accident and Emergency over the last four weeks. Patients will also be asked to provide information about their medication and whether usage has increased or decreased in the last four weeks.

*Anxiety:* The Generalised Anxiety Disorder is a seven-item scale used as a screening tool and severity measure of mild, moderate and severe anxiety. Patients are asked to report on a four-itemed Likert scale (Not at all, Several days, More than half the days, Nearly every day) how often they have been bothered by anxiety related problems over the past two weeks (Spitzer et al., 2006). The GAD-7 has been previously validated in patients with epilepsy, demonstrating excellent internal consistency (Cronbach’s alpha 0.92, (Seo et al., 2014) and its use in patients with NEAD is well cited (Brown et al., 2013; Chen et al., 2014).

*Depression:* The Neurological Disorders Depression Inventory for Epilepsy (NDDI-E) is a six-item scale measuring major depression in people with epilepsy. Patients are asked to report on a four-item Likert scale (Always or Often, Sometimes, Rarely, Never) how best depression related statements (i.e. everything is a struggle) describes them over the last two weeks. The NDDI-E has been validated showing good internal consistency in patients with epilepsy (Cronbach’s alpha 0.85, (Cole, 2006) and NEAD (0.87, (Williams and Bagary, 2012).

*Perception of illness*: The Brief Illness Perception Questionnaire (BIPQ) is a nine-item scale assessing patient’s cognitive and emotional representation of illness. Patients are asked to answer questions regarding: consequences, time-line, personal control, treatment control, identity, illness concern, coherence, emotional representation and perceived causes using a ten-item Likert scale (Broadbent et al., 2006). Although this measurement has not been validated in patients with epilepsy or NEAD, its use for patients with seizures is well documented (Whitehead and Reuber, 2012; Novakova et al., 2015).

*Quality of life:* The NEWQOL-6D(Mulhern et al., 2012)is a six-item health related QoL measure specifically developed for patients with seizures. Although it was originally intended for patients with epilepsy, all questions are also likely to be relevant to patients with NEAD. Patients are asked questions across the following six domains: worry about attacks, depression, memory, concentration, control of events and stigma. Each question has an option of four possible responses, 444444 represents the worst health state, 111111 represents the best. In addition to measuring QoL, the NEWQOL can be transformed into a healthy utility measure QALY (quality-adjusted life year – this measures illness burden), which can then be used in a cost-utility analysis of the intervention.

*Seizures:* TheLiverpool Seizure Severity Scale (LSSS) (Baker et al., 1998) is a twelve-item self-report questionnaire asking patients about their experiences of seizure frequency and severity. The LSSS is scored from 0 - 100; higher scores reflect greater seizure severity. For example, a good outcome for AEDs is accepted if the patient achieves a greater than 50% reduction in seizure severity (this will also be dependent on side effects). The LSSS is a measure of patients’ own perception of seizures, rather than factual assessment. This measure has been shown to have good internal consistency in patients with epilepsy (0.72 to 0.96, (Baker et al., 1998) and although not validated, has been used in patients with NEAD (Whitehead et al., 2013).

*Writing task questionnaire:* This measure has become a standardised questionnaire used in the majority of studies investigating FEW. Patients are asked to rate on a one (Not at all) to seven point (A lot) scale, to what extent was their writings’ personal, meaningful, revealing of their emotions and thoughts; how much have they not talked to others about the information they wrote about and how much have they talked to others; what positive effects has the writing had; did they feel any distress whilst writing; do they feel better after writing; and finally, patients are given a space should they want to write about their feelings or thoughts towards the writing session. Patients are asked to complete this questionnaire following every writing session (a total of four times).

**Setting**

The study is a home-based. Patients recruited from the neurology outpatient clinic at the Royal Hallamshire Hospital in Sheffield will complete baseline measures at time of their appointment.

**Analyses**

1. To test randomisation: Any differences in demographics between conditions (control and therapy writing) and seizure disorders (NEAD and epilepsy) will be verified by a series of t tests for continuous data and chi square tests for categorical data

2. Primary aim: Evaluate the effects of a writing intervention on frequency of seizures and quality of life. Data will be obtained from patients at three time points, T0 (at baseline), T1 (one month following the last writing session) and T2 (three months following the last writing session). A repeated measures analysis of variance (ANOVA) will be conducted in relation to primary and secondary outcome measures with time of assessment (T0 verses T1, T0 verses T2) being the within subjects factors and group (control versus therapy) being the between subjects factor. This will be analysed as patients all patients with seizures, and for individual conditions (epilepsy and NEAD). Analyses will be controlled for by intention to treat carrying the last measurement (T0, T1, T2) forward. A significance level of alpha (0.05) will be adopted for all statistical analysis reported. In addition, as previous research has indicated FEW has moderate but significant effect size, to test clinical relevance Cohen’s *d* will also be reported.

3.Secondary aim: Investigate the patient’s writings using narrative analysis, a qualitative technique with the view to generate understanding of what it is like to live with seizures. This approach focuses the diegetic levels of the texts; use of tense; lexis; pronouns; the construction and presentation of identity; the negotiation of temporality; and the relationship between memory, history, and the present.

**Power Analysis**

The power calculation was based on two meta-analysis of writing therapy. One review used data from 9 clinical samples reporting an effect size of d= 0.19 the other looked at 13 studies of healthy participants reporting an effect size of d = 0.47. In order to achieve 80% power with a sample size of 75 (same subjects) and the significant level of alpha =0.05, the required effect size will be 0.33. Given the time frame of the study and the number of patients attending the clinic and number of registered patients on the Epilepsy Action forum, the researchers anticipate that this is realistic. The researchers anticipate recruiting approximately 90 patients with seizures in each group (control and therapy writing conditions) (180 in total). Based on attrition from previous studies using writing therapy the expected drop out rate is about 20%. This will result in a final sample size of approximately 75 participants in each group, i.e. 150 in total

**Recruitment**

Patients identified as suitable in accordance with the inclusion/exclusion criteria detailed above will be invited to take part in the study. Information concerning the purposes and procedures of the study will be sent to patients at least 48 hours in advance of their appointment in the neurology outpatient clinic (or will be posted on self help websites). The information sheet will also inform patients about the fact that their data will be kept anonymous and that they have the right to withdraw from the study at any point. Patients will have an opportunity to ask questions and revisit the information sheet with a member of the research team when they come for their appointment or via email, post or telephone.

**Subject Withdrawal**

Patients are not obliged to take part in the study and are free to withdraw at any point without giving a reason. Withdrawal will have no effect on the services provided to patients. Patients are clearly told about their right to withdraw from the study at any stage without adverse consequences to their medical care in the Patient Information Sheet.

**Safety Assessment**

This study will not put participants at any risk.

**Quality Control**

Quality of the project will be ensured throughout. We will use standardised questionnaires with well established reliability and validity.

**Project Plan**

|  |  |  |
| --- | --- | --- |
| Year | Month | Process |
| 2014 | October – January | Literature Review |
| 2015 | January – July | Develop writing intervention, finalise study design  Register the research with the research department at Sheffield Teaching Hospital Foundation Trust. |
|  | July – October | Intervention review and ethics approval |
|  | October – November 2016 | Start participant recruitment, Data collection |
| 2016 | December | Data analysis and interpretation |
| 2017 | April – August | Write up and dissemination of findings |

**Statistical Opinion**

The members of the research team have the statistical expertise to evaluate the findings.

**Expertise**

Professor Markus Reuber is an epileptologist with extensive clinical and research experience in patients with seizures. Both have previously supervised research leading to the award of PhDs. Gregg Rawlings is a psychology graduate who will be supervised by Professor Reuber. This project will form the basis of Gregg Rawlings’s PhD thesis.

**Ethical Issues**

Ethical approval will be sought.

Completion of the NDDI-E and the GAD-7 may reveal an existing psychiatric disorder. The patients' consultant (if known) will be informed of any self-reported evidence of relevant mood or anxiety disorder. Patients will be encouraged to contact the research team if they experience any difficulties with the study measures or intervention.

Patients will be made aware that they have the right to withdraw from the study at any point without giving a reason and that this will not affect the services provided to them.

All data will be anonymised through coding the names of the patients by the researcher. Details will be kept in a locked cabinet. All electronic data will be password-protected.

**Service Users Involvement**

As part of the design procedure, we asked patients receiving care at the Royal Hallamshire Hospital to rate the acceptability of seven possible topics that participants could write about for each of the four writing session. The final four was selected based on their responses. Patients were also asked about acceptability of the intervention protocol i.e. length of writing periods, setting of the intervention.

**Dissemination**

Findings of the study will form part of a PhD thesis. The results will be presented at scientific meetings and be submitted for publication in academic epileptology and clinical psychology journals.

**Taking the Work Forward**

Results of the study will provide an evaluation of the possible utility of a writing intervention that could be incorporated into the current treatment regime of patients at the neurology outpatient clinic.

**Costing Schedule**

The PhD student carrying out this research is receiving a stipend from the University of Sheffield. The funding period started on 1 October 2014. Funding will continue until 30 September 2017.

**Funding Arrangements**

The PhD student carrying out this research is receiving a stipend from the University of Sheffield. Funding from Sheffield University will cover the administrative costs associated with this project.

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**Appendix 19 IRAS Form**

**Appendix 20 Ethical Approval**