INVESTIGATING SELF-PERCEPTION OF EMOTION IN INDIVIDUALS WITH NON-EPILEPTIC SEIZURES (NES)

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INTELLECTUAL PROPERTY STATEMENT

The candidate confirms that the work submitted is her own and that appropriate credit has been given where reference has been made to the work of others.

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[All] praise is [due] to Allah, Lord of the worlds. Quran, verse 1: 2

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ABSTRACT

Emotional processing difficulties are hypothesised to be involved in the aetiology and maintenance of non-epileptic seizures (NES). This thesis aimed to explore the relationship between aspects of emotional processing: interoception, alexithymia and executive functioning, in people with NES in comparison with healthy controls and to understand how people with NES experience their symptoms, live with their condition, and perceive the role of life events in relation to their seizures.

Study 1 reviewed the evidence for a relationship between interoception and other key emotional factors in studies which employed heartbeat perception tasks to measure interoception. Study quality was found to be generally poor, with no consistent evidence for significant findings between interoception and emotional factors, including alexithymia, depression, and anxiety.

Study 2 was a cross-sectional, online, study to investigate an interactional model of emotion processing, exploring relationships between interoceptive sensibility, alexithymia, and executive functioning (attentional bias) in NES participants and healthy controls. Measures included the Body Perception Questionnaire (BPQ-VSF), the Toronto Alexithymia Scale-20 (TAS-20) and the emotional Stroop task (eStroop). The NES group, compared to controls, reported higher BPQ-VSF and TAS-20 scores. There were no significant correlations between any of the measures of interest in either the NES or control group. There was no evidence to support the proposed model.

Study 3 was a qualitative study using Interpretative Phenomenological Analysis to explore: how individuals with NES respond emotionally to recent life events; and how these events impact on seizures. Six themes were developed from the analysis which described how NES affected many aspects of people’s lives. Four models captured the different ways in which people perceived the relationship between life stressors, their emotional responses, and their seizures: event->emotional response-> seizure; event-> emotional response -x-> no seizure; no event ->emotional reaction/experience -> seizure; and no event -x->no emotional response->seizure.

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ABBREVIATIONS

FND	Functional neurological disorder
FS	Functional seizures
GET	Group Experiential Theme
HBD	Heartbeat discrimination
HPT	Heartbeat perception task
HCP	Healthcare professionals
ICM	Integrative Cognitive Model
IPA	Interpretative phenomenological analysis
LE	Life event
MTM	Mental tracking method
MUS	Medically unexplained symptoms
NES	Non-epileptic seizures
PET	Personal Experiential Theme
pwNES	people with NES

CHAPTER 1: INTRODUCTION

1.1 Introduction

Non-epileptic seizures (NES) are episodes of abnormal behaviour and experience that typically involve impairment of consciousness, flaccid or rigid collapse and/or jerky or tremulous limb movements (Hubsch et al, 2011). They fall under the group of disorders known as functional neurological disorder (FND) and are referred to by different names in the literature, including functional seizures, psychogenic seizures and non-epileptic attack disorder. Historically, non-epileptic seizures (NES) have also been considered to be symptoms of hysteria (Trimble and Reynolds, 2016) and seen as primarily a woman's condition (Asadi-Pooya, 2016; Goldstein et al, 2019; Myers et al, 2018; Kaplan et al, 2021).

With regards to presentation, the seizures themselves can vary from individual to individual, and are generally described as superficially resembling an epileptic seizure (Hallet et al, 2022; Duncan, 2016). However, unlike epileptic seizures, non-epileptic seizures are not accompanied by the abnormal brain activity characteristic of epilepsy.

The aetiology of NES remains unknown, but several theories have developed over the last hundred years. The main theoretical models have been summarised by Brown and Reuber (2016) and include both cognitive and psychodynamic approaches. A feature of several of the models of NES is a focus on emotion, as difficulties in processing emotion are a plausible driver for maladaptive defensive or coping responses. Modern theories of emotion often have a foundation based on the James-Lange theory (Lacasse, 2017), and this is apparent in the Somatic Marker Hypothesis proposed by Damasio (1994), which postulates that emotional processes guide behaviour, particularly decision making.

The perception of the internal state of the body (e.g. the rate of one's heartbeat) is referred to as interoception. Misrepresentations of the body's internal states, or a disconnect between the body's signals and the brain's interpretation and prediction of those signals, has been suggested to underlie a range of conditions including NES. Interoception is an area that has grown in interest with regards to emotion processing, as has the concept of alexithymia.

Alexithymia can be translated as, 'no words for emotion' (Lesser, 1981) and refers to the inability to identify and describe emotional experience, which has commonly been associated with a tendency to experience and report more physical symptoms. Both interoception and alexithymia will be described in further detail below.

The literature suggests that the NES population is heterogenous, in aetiology, presentation and factors that maintain NES symptoms. For example, a large proportion of those diagnosed with NES have a history of trauma, but this is not true of all individuals. Similarly, there appear to be groups of NES individuals that are characterised by having higher levels of traits, such as alexithymia, whereas other individuals do not.

None of the current aetiological models provide a complete explanation of the aetiology and pathogenesis of NES. The Integrative Cognitive Model developed recently by Brown and Reuber, (2016) is an attempt to address this problem and incorporate previous theory and research findings (to date) into a single model which can provide the basis for future hypothesis-driven phenomenological, psychological and experimental research. The model will be described in more detail below.

The aim of this thesis is not to explore the aetiology of NES, rather the main focus is on factors that may contribute towards maintaining the seizure episodes, once they have begun. The intention is to gain a better understanding of how people with NES (pwNES) process emotion and whether emotional responses to stressful life events may contribute towards maintaining NES.

This chapter introduces NES with regards to diagnosis of the phenomenon, common precipitating and maintaining factors that are associated with NES, and some common co-morbidities. The aim is to provide background and context to the three studies that make up this thesis, and three main areas of interest: interoception, alexithymia and executive functioning.

1.2. Semiology and diagnosis

Approximately 20% of patients are diagnosed with NES after presenting to a doctor with ‘seizure-like’ events (Kotsopoulous et al, 2003). Many individuals diagnosed with NES start with a diagnosis of epilepsy that is subsequently withdrawn (Reuber et al, 2002). The condition is generally regarded as being associated with psychological issues, and the most relevant diagnoses are dissociative neurological symptoms disorder (International Classification of Diseases Eleventh Edition (ICD-11, World Health Organisation, 2015), conversion disorder or functional neurological symptom disorder, according to Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM V: American Psychiatric Association, 2013).

Diagnosis of NES may be based on several stages, including whether a video EEG demonstrates epileptiform activity during a seizure. The seizure manifestations usually differ from epilepsy in certain respects such that experienced clinicians can differentiate between NES and epilepsy with a high degree of accuracy if they are able to witness a seizure or view a recording of a typical event (Chen et al, 2008). Reliable indicators of NES are: long duration, occurrence from apparent sleep with EEG verified wakefulness, fluctuating course, asynchronous movements, closed eyes during the episode, ictal crying, memory recall and absence of post ictal confusion (Avbersek and Sisodiya, 2010; Stone et al, 2020), however NES is still often regarded as a diagnosis made by exclusion.

It has been suggested that a delay in diagnosis can be due to both poor awareness of NES and a lack of access to relevant diagnostic tests (Reuber et al, 2002). Those individuals that present with an acute presentation at hospital, receive a quicker diagnosis and tend to have better long-term outcomes (Duncan et al, 2011). In general, patients with NES have a poor prognosis, with 75% continuing to have seizures on a long-term basis (Duncan et al, 2011).

Prognosis may also be affected by other factors at the presentation/diagnostic stage, such as attitudes of clinicians towards, and a lack of knowledge of, functional disorders. The latter may lead to longer periods of insufficient or inappropriate intervention; for example, studies have reported that it may take up to 7 years for a diagnosis of NES (Lancman et al, 1993; Reuber et al, 2002). In some cases, this has led to misdiagnosis as epilepsy (Reuber et al, 2002), with prescribing of anti-epileptic medication (Bodde et al, 2009), and increased risk of iatrogenic harm. Delays in diagnosis also cause problems for both patients and services: individuals may present frequently to emergency departments (Reuber et al, 2002) when it is not the most appropriate place for them to receive care. Delayed diagnosis may also harm the potential for psychological treatment to have a positive outcome (Bodde et al 2009). It has been noted that on receiving a diagnosis, seizures cease for some patients (Ettinger et al, 1999).

1.3. Epidemiology and prognosis

It has been suggested (Hallett et al, 2022) that FND is found to be both a global phenomenon

(Kanemoto et al, 2017) that has existed across the ages (Stone, 2016). However, studies regarding the prevalence of NES in the general population are limited (Benbadis and Hauser, 2000). In a recent review (Carson and Lehn, 2016), the prevalence for NES is reported to be between 2 and 33/100,000/year (Benbadis and Hauser, 2000). Carson and Lehn (2016) note that the difference in rates reflects that NES may be one of several different symptoms presented by the individual, therefore this may impact on how the condition is defined. NES often co-occurs with other functional neurological disorders (FND) such as movement disorders, and therefore the more inclusive diagnosis of FND may be preferred.

The incidence rate refers to how many new cases occur in a specific population over a period of time (Bowers et al, 2006). Using a 'first-seizure clinic', Duncan et al (2011) conducted a study over three years to identify incidence rates for NES. The authors reported that 68 patients from a population of 367,566 were diagnosed with NES, giving an incidence rate of 4.90/100,000/year. This study was conducted in a Scottish county in the Highlands (Ayrshire and Arran), with a majority White Scottish demographic, and only included people aged 13 years and above, so the findings are less comparable with other areas of Scotland and the UK that have a more diverse population. In a study from Iceland, the incidence was estimated to be 1.4 per 1000,000/year but again young people (below 15 years) were not included (Sigurdardottir and Olafsson, 1998). A further study from the United States provided an estimate of 3.3/1000,000/year, however, this study required evaluation at a referral centre which limits the validity of the findings (Szaflarski et al, 2000). The absence of other population-based studies for NES means that both the prevalence and incidence of NES remains somewhat uncertain. None of the above studies reported on ethnicity and little is known about the prevalence and incidence of NES in the global south (Asadi-Pooya, 2021).

NES are more common in women than men, at a ratio of 3:1 (Duncan et al, 2006), although sex is equally distributed in the preadolescent and older age groups (Duncan et al, 2006; Patel et al, 2007). Non-epileptic seizures occur across the lifespan, but the diagnosis is most common between the ages of 15-30 years (Reuber, 2008).

Compared to patients with epilepsy, those diagnosed with NES are typically described as having a poor prognosis. A systematic review of longitudinal studies of NES (Gelauff and Stone, 2016) reported that most studies found that less than 50% of patients had fully recovered from seizures at follow-up. One of the studies included in the review divided NES patients into an acute and chronic group (Buchanan and Snars, 1993). A large proportion of patients (83%) in the acute group were reported to be seizure-free after a mean of 2.3 years. Of the chronic group, only 28% were reported to be seizure-free on follow up. However, this is a relatively old study and it had a number of methodological issues: it was a retrospective study, both adults and children were included, and over half of the acute group (66.6%) were thought to also have epilepsy. Additionally, the authors comment that the chronic group had more severe psychological problems and were older.

1.4. The Integrative Cognitive Model (ICM)

Reuber and Brown (2017) have proposed that, by consolidating earlier theories into one model, the Integrative Cognitive Model (ICM: Brown, 2004, 2006, 2013) offers a single framework by which to understand NES. Prior theories include the following (Brown and Reuber 2016):

- NES are a ‘sensorimotor flashback’ during a period of stress or intense emotion that causes a loss of involuntary control. This could be due to the individual’s inability to recognise the link with trauma, or not being willing to express the traumatic memories (Bowman, 2006; Kuyk et al, 1996).

- NES are a physical manifestation of emotional distress and part of a defensive process that prevents the individual from becoming overwhelmed by the emotional consequences of adversity (Freud and Breuer, 1895; 1955).

- NES are a hard-wired aberrant reflex similar to other defensive reactions, such as freezing. The response is considered to provide a defensive or protective element which occurs in response to perceived threat (Baslet, 2011; Baslet et al, 2016).

- NES are a product of aberrant learned behaviour maintained by positive and negative reinforcement (Moore and Baker, 1997).

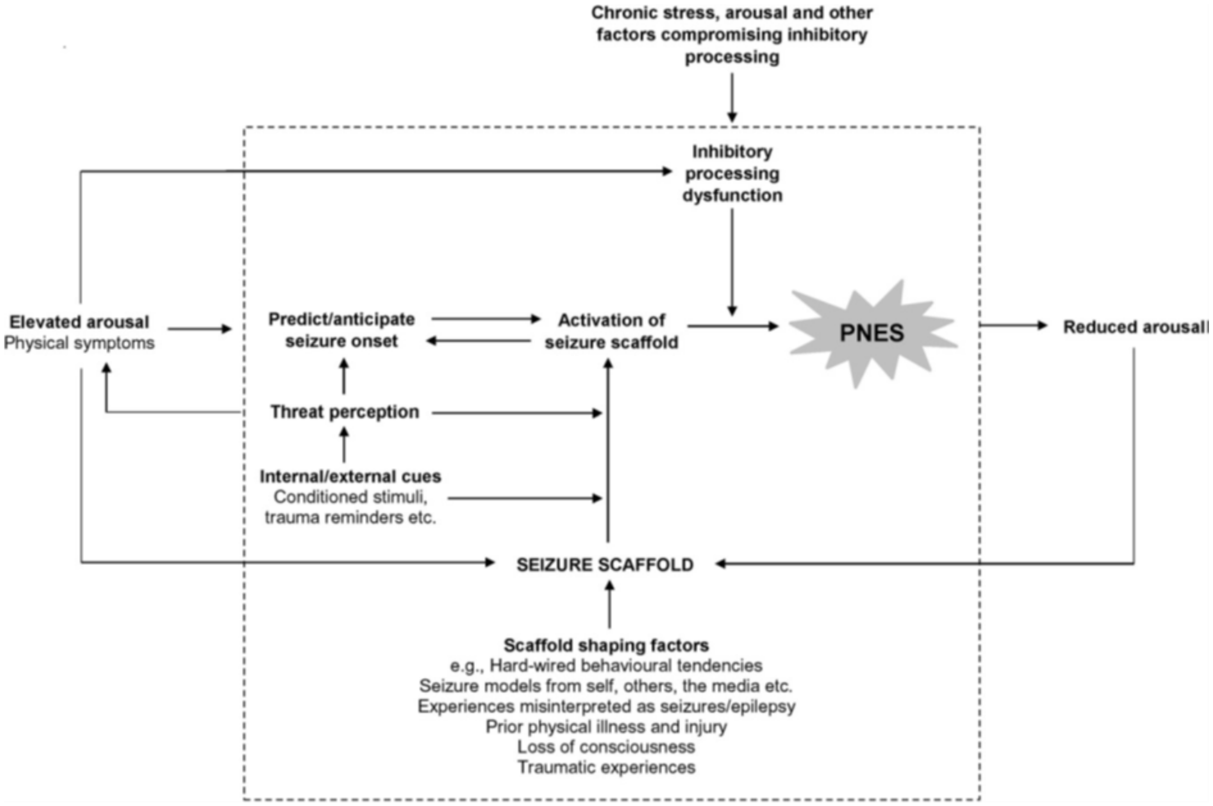
In the ICM, Reuber and Brown (2017) hypothesise that NES result, ‘from the automatic execution of a learnt mental representation of seizures’ ‘(the “seizure scaffold”), typically in the context of a high-level inhibitory dysfunction resulting from chronic stress, arousal and other factors that compromise high level processing’. Figure 1.1 is a diagrammatic reproduction of the model from Reuber and Brown (2017). The ICM is based on the assumption that the images/memories of seizures in the individual’s past lay the foundation for NES. Non-epileptic seizures occur when an automatic and unconscious system is triggered. This starts a process of expectation from the individual that a seizure will occur, and the resulting seizure confirms the individual's expectations. Reuber and Brown (2017) explain that the individual is often unable to control this process, and in most cases the seizure feels involuntary. Chronic stress, arousal and other factors reduce inhibitory processing which increase the likelihood of a seizure being triggered.

Three stages are generally found in the experience of NES (Brown and Reuber, 2016):

1. The first stage is often experienced as a physical, rather than an emotional experience, as the individual undergoes an intense increase in sympathetic arousal corresponding to a variety of autonomic symptoms (such as an increase in heartrate, feeling hot, sweating). At this stage the individual is sensitive to the activation of the misrepresentative hypotheses ('rogue representations'). When this vulnerability is paired with the predication of a seizure occurring, this triggers the seizure scaffold (figure 1.1 below).
2. The second stage is when the seizure is experienced.

3. At the third stage the individual experiences a reduction in arousal. The seizure itself varies from individual to individual. This is due to the stored knowledge and experiences used to build the mental representations, or if based on 'inherent components', such as 'cognitive biases typically associated with emotions such as fear, anger, and/or disgust'.

Figure 1.1: The seizure scaffold from the Integrative Cognitive Model (Brown and Reuber, 2016)



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Fundamental to the ICM is that, beyond the actual stimuli, numerous factors can cause the activation of hypotheses during the ‘perceptual process’. The actual triggers or perceptions may or may not bear a close relationship to actual events or reality. In the case of functional symptoms, the ICM suggests that symptoms arise when the activation levels of ideas or representations of physical illness increase to a point where the system regards them as the best explanation of, or response to, what is happening at that moment. As the process is preconscious and the individual has no access to it, the symptoms are experienced as real. Where there is a great disparity between representations and actual reality, Brown has used the term ‘rogue representations’ (Brown, 2004; Brown and Reuber 2016), because they represent an inaccurate or distorted perception of the symptoms experienced. High level attentional dysfunction may contribute to the development and maintenance of symptoms by impairing an individual’s ability to inhibit discrepant mental representations.

As the authors (Reuber and Brown, 2017) state, the ICM is an attempt to bring together previous theories concerning the aetiology and pathogenesis of NES into one overarching framework. It is beyond the scope of this thesis to test the model in its entirety. Instead, the thesis focuses upon the role of emotional processing in the maintenance of NES examining three related concepts: interoception, alexithymia and executive functioning (attentional bias).

1.5. Emotion processing

The first detailed study of the condition we now understand as NES was carried out by the French psychologist Pierre Janet (1859-1947) who based his ideas on the detailed examination of hundreds of patients who were considered to be suffering from hysteria (Janet 1889). Janet viewed NES as evidence of a breakdown or collapse in function in the face of stress or intense emotion. Central to his ideas was the idea of collapse of normal mental events and fragmentation of psychological systems. He used both the term, disaggregation meaning a failure of mental synthesis and dissociation to refer to compartmentalisation. According to Janet, NES can be understood as the activation of memory fragments (usually traumatic) which are triggered by an event, and relived, although the person is not aware they are reliving an event from their past. The fragments of memories may be pre-verbal and take the form of gestures or actions. They cannot be assimilated into consciousness because of the anxiety associated with them and the degree of ‘disaggregation’ of the psyche. The role of dissociation in NES has been reviewed by Brown and Reuber (2016), who suggest that the evidence at present indicates that dissociation commonly occurs in NES but is not sufficient to explain the phenomenon, as NES can arise in the absence of dissociation. The ‘disaggregation’ described by Janet has received less attention but clearly involves a disruption to mental and emotional synthesis, which in modern parlance could be construed as a collapse of emotional processing.

Interoception

In ‘The Principles of Psychology’, James (1890) postulated that emotions comprise perceptions of bodily changes that occur in response to an event: i.e. a physical sensation before a cognitive appraisal of the situation (Hatfield et al, 2019). James asked the question ‘Do we run from a bear because we are afraid or are we afraid because we run?’. He argued that we are afraid because we run, ‘Our natural way of thinking about …emotions is that the mental perception of some fact excites the mental affection called emotion, and that this latter state of mind gives rise to the bodily expression. My theory is that the bodily changes follow directly the perception of the exciting fact and that our feeling of the same changes as they occur IS the emotion’ (James 1890). The role of bodily changes and cognitions in experiencing emotion was considered controversial at the time it was proposed by James (Lacasse, 2017), and his ideas feel into abeyance until the end of the last century.

Damasio (1994) revived interest in Jamesian ideas after studying patients who had suffered damage to the prefrontal medial cortex brain region. The patients performed well on intelligence tests, but struggled to function in society, making repeated poor life choices. They also showed marked difficulty in expressing emotion and could seem fatuous and uncaring. Damasio (1994) hypothesised that the impaired ability to experience and express emotion in these patients severely impacted their decision making. He went on to hypothesise that emotional processes consciously or unconsciously impact decision making by creating biomarkers, or so-called ‘somatic markers’. Somatic markers are changes in both the body and brain which together make up an emotion. They are triggered by a person’s perception of external or imagined events and can include both superficial body changes (e.g. posture, facial expression, hand gestures) and internal body changes such as endocrine release or changes in heart rate. Damasio (1994) suggested that many of our decisions which we think are based on logical, thoughtful processes are in fact influenced and guided by emotional processes (‘gut feelings’) of which we are often not aware.

According to the somatic marker hypothesis (Damasio, 1994), somatic signals are thought to enable a faster experience-driven integration of decision-making by forcing attention towards the potential negative outcome of an action and provide a sort of unconscious alarm. The medial prefrontal cortex, amygdala, insula, somatosensory cortex and brainstem nuclei are thought to be involved in the relevant emotion circuitry in the brain.

Damasio provided empirical support for his ideas from studies using a gambling task, involving different decks of cards: high risk decks resulting in overall losses for the gambler and low risk decks resulting in a net profit (Bechara, et al, 1994). Normal individuals who complete the gambling task adopt an advantageous strategy very quickly and their disadvantageous decisions are associated with increased skin conductance (signs of a somatic marker). Patients with lesions in the pre-frontal cortex, which is associated with an impairment in somatic signalling, perform poorly on the task (i.e. lose a lot of money) and do not show evidence of a generation of ‘somatic markers’ in the form of changes in skin conductance before making disadvantageous choices. Individuals with particular accurate sensitivity to bodily signals such as temperature, pain, visceral or muscular sensation, or heartbeat show superior performance on the gambling task described above (Werner et al, 2009).

This ability to detect subtle changes in the body’s internal milieu is referred to as interoception. The concept of interoception is important to the Jamesian (1890) theory of emotion – arousal and is a basic component of judging emotional experience (Barrett et al, 2004), and may be a key part of emotional processing (Schandry, 1981; Fustos et al, 2012; Werner et al, 2009). Murphy et al (2017) comment that there is a ‘growing appreciation’ regarding the role of interoception in emotional stability and regulation (Schandry, 1981; Fustos et al, 2012; Werner et al, 2009; Pollatos and Schandry, 2008). Consequentially, if one has awareness of one’s internal bodily activity, one should also be able to perceive one’s emotional state, and Calsius et al (2016) highlight that interoception appears to be a key factor in body and self-awareness in relation to functional symptoms (Craig, 2003, 2011).

The brain used to be regarded as a passive processor of sensory information from the external world, whereas it is now considered as playing a much more active role in inference, generating predictions (based on past experiences) and generating hypotheses about the causes of its sensations (Picard and Friston, 2014). The brain to a certain extent balances top-down interoceptive signals (inferences from prior experience) and bottom-up incoming interoceptive signals (Seth et al, 2011). It is hypothesized that a sense of body ownership is determined by the degree of agreement between top-down and bottom -up signals and that disorders where there is a breakdown in the sense of body ownership (e.g. dissociation) result from pathologically imprecise interoceptive predictive signals and an increased discrepancy between top-down and bottom-up signals (Ainley et al, 2016). In Chapter 2, there is a more detailed discussion of interoception and research on NES.

Alexithymia

Alexithymia is described as difficulty in verbally expressing an emotional experience (Uliaszek et al, 2012). It is hypothesised that this results in psychological distress being expressed in physical terms, and emotions being described as physiological reactions (Taylor et al, 1997). The recognition of several deficits in emotion processing in those with NES, has led to the conjecture that such deficits make individuals vulnerable towards developing functional symptoms (Baslet et al, 2016). Being able to identify and express an emotional experience is in itself an aspect of processing emotion (Uliaszek et al, 2012).

Alexithymia has been reported to be higher in NES populations than controls (Bewley et al, 2005; Subic-Wrana et al, 2005; Uliaszek et al, 2012). Kaplan et al (2013) found differences between NES and epilepsy controls on the Difficulty Identifying Feelings sub-scale of the Toronto Alexithymia Scale (TAS-20: Bagby et al, 1994). Baslet et al (2016) state that problems with recognising emotion may also extend to difficulty with recognising that seizures may possibly be triggered by emotional factors. They (Baslet et al, 2016) also suggest that the identification of fear sensitivity (Hixson et al, 2006) and avoidance (Goldstein and Mellers, 2006) in patients with NES are factors that point to a ‘potential psychopathological mechanism’, where an increase in responses to autonomic activity and a biased response style leads to poor use of behavioural strategies when the individual is challenged with an emotional situation (Baslet et al, 2010).

Brown and Reuber (2016) likewise suggest that NES may result from either misattributing emotion as a physical disorder, or alternatively NES represent a mechanism by which ‘unrecognised emotional tension’ is released. Others (Urbanek et al, 2014; Novakova er al, 2015) have noted higher levels of feeling overwhelmed by emotion (Urbanek et al, 2014) or high scores on emotional processing (Novakova et al, 2015) when NES participants are compared to healthy controls.

Alexithymia is commonly measured using the Toronto Alexithymia Scale (TAS-20: Bagby et al, 1994). Multiple studies have reported that approximately one third of people with NES have alexithymic traits (Tojek et al, 2000; Myers et al, 2013; Wolf et al, 2015). Although these rates of alexithymia are higher than the general population, they are similar to rates in patients with epilepsy and other medical conditions (Brown and Reuber, 2016). O’Brien et al (2015) also found higher levels of depressive and anxiety symptoms, dissociative symptoms, alexithymia, spatial working memory and attention in pwNES than in healthy controls. The high prevalence of alexithymic traits in pwNES has led some authors to argue that this infers a pattern of chronically abnormal emotional processing (O’Brien et al, 2015) (Saarijavi et al 2006).

Brown and Reuber (2016) suggest that comparing pwNES with controls, or those with epilepsy, has possibly undermined the level of importance alexithymia may play in some pwNES. The heterogeneity that has been reported in studies using cluster analysis (Brown et al, 2013) suggest there may be distinctly different groups of NES, and for some subgroups of NES participants, alexithymia may be an important aetiological and maintaining factor.

It has also been suggested that it is possibly difficult for patients with deficits in emotional processing to be able to reliably report alexithymia, and that other methods may be more effective at demonstrating problems with expressing feelings (Brown et al, 2016). For example, Reuber et al (2011) found that carers of pwNES were more likely to consider an association between emotional stress and seizures, than the patients themselves.

Many of the studies discussed above have methodological concerns. The original work by Damasio has received a considerable degree of criticism (Dunn et al, 2006). Although there is support for the somatic marker hypothesis in terms of the areas of the brain involved in decision making, emotion and body-state representation, it is unclear how these interact at a psychological level (Dunn et al, 2006). Nearly all of the experimental studies investigating the role of emotional processing and alexithymia in pwNES consist of small unpowered, sample sizes and mixed populations. Given the heterogeneity in NES populations suggested by studies using cluster analysis, many findings of studies comparing pwNES with a control condition will be influenced by the proportion of NES participants with different psychological profiles in the study group. Equally, 'normal controls' and epilepsy groups can be affected by selection bias depending upon recruitment methods which are rarely explained in full.

Executive functioning (attentional bias)

Janet (1889) considered dissociative symptoms to be related to attentional deficits caused by trauma or stress; recent theorists such as Brown (2004) have reported similar hypotheses. It has been highlighted (Brown and Reuber, 2016) that about 40-50% of patients with NES perform poorer on 'neuropsychological test batteries (Cragar et al, 2002; Reuber et al, 2002)' when compared against controls (Binder et al, 1998) or those with epilepsy (Strutt et al, 2011), suggesting that pwNES indicate difficulties with aspects of executive functioning such as attention. The evidence indicates that those with NES have difficulties filtering out unimportant sensory stimuli and demonstrate patterns of heightened vigilance towards such stimuli when compared with those with epilepsy (Baslet et al, 2016). It could be that deficits in higher-order attention and executive functioning lead to alternative expressions of dysfunction in NES: patterns such as dysfunction with verbal memory and fluency

(Willment et al, 2015). Paradigms designed to measure abilities across classic cognitive domains that require emotion processing simultaneously are known as 'dual cognitive-emotion paradigms' (Willment et al, 2015). Such tasks measure performance relating to domains such as attention, executive functioning, and memory; for example, under conditions requiring emotion processing. Using such tasks, it has been demonstrated that pwNES may have deficits across several domains (Willment et al, 2015).

However, research into emotion processing in NES is limited (Uliaszek et al, 2012), and evidence towards hypotheses relating to attentional bias in NES is conflicting. For example, several studies have indicated that those who experience functional symptoms may demonstrate hypervigilance towards physical symptoms and physiological arousal. Studies using the emotional Stroop task have shown that individuals with functional symptoms indicate more emotional interference (Afzal et al, 2006; Lim and Kim, 2005), and this has led to the suggestion that the motivation for this may be threat avoidance (De Ruiter and Brosschot, 1994). Although, Bakvis et al (2009) reported no difference between pwNES and healthy controls on the Stroop (non-emotional) paradigm. Almis et al (2013) reported that NES patients responded slower when compared against healthy controls' performance of the Stroop task, whilst also reporting no differences in inhibition processing, based on similar scores on cognitive interference.

Bakvis et al (2009) investigated attentional processing in pwNES, and whether there was an association between 'interpersonal trauma' and stress. The study was interested in whether patients with NES pre-consciously assigned attention towards (socially) threatening stimuli. They used a version of the Stroop test referred to as the 'masked emotional Stroop test'. This task presents participants with facial expressions as stimuli, e.g. happy, angry and neutral, that are presented for a set period of time. In this study (Bakvis et al, 2009), stimuli were presented for 14 ms, and then the emotional stimuli were followed by a 'masking' stimulus. They found that NES patients demonstrated an attentional bias for angry faces (or hypervigilance), compared to controls, and this correlated with self-reports of sexual trauma.

Cognitive vigilance is considered to be a coping style when an individual is faced with a threat (Calvo and Eysenck, 2000). This coping style involves focussing on the threat at an excessive/disproportionate level, with avoidance of 'threat processing and high physiological arousal' (Calvo and Eysenck, 2000). In the Bakvis et al study (2009), people with NES appeared to show a preconscious attentional bias, by demonstrating increased vigilance towards threat stimuli, with a significant correlation with sexual trauma. Other work has also found a similar ‘hypervigilance’ for threat stimuli and trauma by participants with Dissociative Identity Disorder (Hermans et al, 2006).

Another study by Bakvis et al (2010) used a different paradigm (N-back test) with pwNES and healthy controls. They reported that at baseline, pwNES showed more interference from distracting stimuli (neutral and emotional). Under the stress condition, the interference effect continued even when no distracting stimuli were present. The interference effect (relating to stress) was significantly associated with cortisol levels in NES patients but not in controls. Brown and Reuber (2016) suggest that this response is indicative of general attention deficits, rather than emotion specific deficits. They also suggest that an alternative explanation may be that pwNES struggle /fail to filter out any unimportant stimuli.

A study by Gul and Ahmad (2014) using a task that required switching attention, suggested that pwNES compared to controls showed difficulty with switching attention from emotional stimuli. Brown and Reuber (2016) offer another explanation: that is, participants may have been demonstrating a quicker engagement with the stimuli requiring their attention, i.e. more attention to, or difficulties with disengaging from the emotional stimuli. Brown and Reuber (2016) also comment that, because individuals with co-morbid mental health problems were excluded, the generalisability of the results are somewhat limited.

Baslet et al (2016) suggest that pwNES are found to ‘perform outside normal limits on objective cognitive measures’, such as those that test executive functioning, and that a consistent finding is a problem with attention regulation (Willment et al, 2015). However, such deficits are found in other psychopathologies and neurologic conditions, so are not specific to NES, and not necessarily a defining feature of the condition.

Emotion regulation

Successful emotion regulation is important to maintaining good health. Gratz and Roemer (2004) have described emotion regulation as the ability to control behaviour when an individual is experiencing intense emotions. That is, emotion regulation does not simply refer to controlling one’s emotions, but rather the ability to control *intense* emotions. They say that, based on this model, efficient or adequate regulation is that which enables the individual to complete their goals (short and long term) by minimising the subjective psychological distress in response to an emotional experience (i.e. affective state). They further describe the emotion regulation model proposed by Gratz and Roemer (2004) as including four key areas:

1. Awareness and understanding of emotions.
2. Acceptance of emotions.

3. When experiencing negative emotions, having the ability to control impulsive behaviours and act appropriately to achieve goals.

4. Having the ability to use emotion regulation strategies appropriate to the situation to regulate behaviour.

Therefore, poor emotion regulation may be a seminal process underlying several conditions (Campbell-Sills et al, 2006; Linehan, 1993).

Emotion dysregulation refers to difficulties with recognition and regulation of emotional states. Emotion dysregulation may in part be responsible for the advancement and maintenance of NES (Brown et al, 2013). Uliaszek et al (2012) explain that the different profiles within NES samples suggest there is a minimum of two aberrant emotion regulation profiles susceptible to NES. The two types that have been identified suggest "undermodulation" and "overmodulation" of affect in NES patients (Willment et al, 2015; Uliaszek et al, 2012). Undermodulation refers to individuals demonstrating 'emotional reactivity, poor arousal tolerance, and difficulties controlling affect'. Overmodulation is demonstrated by 'emotional avoidance, excessively controlled behaviour, and a tendency to use physical symptoms as a way of expressing emotional conflicts' describing "somatic defense" (Cragar et al, 2005; Brown et al, 2013).

Brown et al (2013) investigated how pwNES performed on a number of self-completion measures that included levels of emotion dysregulation (Difficulties in Emotion Regulation: DERS, Gratz and Roemer, 2004); alexithymia (Toronto Alexithymia Scale: TAS, Bagby et al, 1994); attachment (Relationship Scales Questionnaire: RSQ, Griffin and Bartholomew, 1994); anxiety (Generalized Anxiety Disorder: GAD, Spitzer at al, 2006); and dissociation (Somatoform Dissociation Questionnaire: SDQ, Nijenhuis et al, 1996). Participants were those with either epilepsy or NES. A cluster analysis was used to determine whether any patterns of emotion dysregulation were apparent within the NES sample. The authors reported that two clusters were found within the NES group: one group was characterised by reporting difficulties with most aspects of emotion regulation and higher levels of somatisation, alexithymia and anxiety, when compared with participants with epilepsy, and the second group reported high levels of somatisation and depression, but not high alexithymia or emotion dysregulation when compared with participants with epilepsy. Similarly, a study by Uliaszek et al (2012), again using a cluster analysis, found two clusters, one with higher overall scores on the DERS questionnaire compared to the other cluster.

Life events and chronic difficulties

Life events (LE) are discrete events, such as a death of a close relative or a car accident, which happen over a short period of time whereas chronic difficulties are stressors than last longer than a month, such as an ongoing housing problem. Life events and chronic difficulties have frequently been studied together, along with childhood experience (e.g. childhood trauma). Childhood experience is often considered to act as a predisposing factor for a variety of both mental and physical health problems, whereas life events are regarded as potential triggers for a specific episode of illness. The clinical picture is much more complex than this, however, as childhood or early experience, influences the nature and number of life events people are exposed to as adults.

Some studies have reported that individuals with FND report more LE than controls (Bowman, 1999; Tojek et al, 2000; Nicholson et al, 2016); whereas others report that pwNES do not report more LE when compared to controls (Roelofs et al, 2005; Testa et al, 2012). Ludwig et al (2018) conducted a systematic review of stressful life events and maltreatment in FND. The 34 studies included in the review were case-control studies and 24 included pwNES. All 34 studies assessed stressors retrospectively and several methodological limitations were recognised by the reviewers including: a lack of blinding in most studies, failure to use a gold standard instrument to measure life stress in all but three studies, and problems with power. There was considerable variation in the overall quality of the studies. The reviewers (Ludwig et al, 2018) found higher rates of reported stressors, both past and present in patients with FND than controls, with a higher risk for reported childhood factors than adult life, and a higher risk for childhood emotional neglect than physical or sexual abuse.

An earlier review by Sharpe and Faye (2006) included 34 studies which had examined the relationship between reported child sexual abuse and NES. Although the overall findings supported a link between child sexual abuse and later onset of NES, the authors cautioned drawing any definitive conclusions because of widespread limitations in research design of the included studies.

It has been hypothesised that the onset and maintenance of NES are associated with stressful life events in the context of poor coping strategies. When compared to the scores from participants with epilepsy or healthy controls, pwNES have higher levels of distress in relation to a variety of factors, such as work, social functioning, health, and legal difficulties (Testa et al, 2012).

Lived experience of NES

There have been several studies which have used qualitative methods to better understand the experience of pwNES. Rawlings and Reuber (2016) carried out a systematic synthesis of 21 qualitative studies. The majority of the studies were UK based (14) and overall 220 patient accounts were included the review. A wide range of different approaches to analysing participants' accounts were employed including thematic, content and linguistic analysis, and two studies used Interpretative Phenomenological Analysis (IPA), the preferred methodology in Study 3 in this thesis.

The reviewers identified five key themes which reflected experiences of seizure events, diagnosis, treatment and management, emotional events and impact on daily life. Many of the studies focused on the lived experience of NES and people's understanding of the diagnosis and their perceived treatment needs. Only one study explored the understanding and perceptions of emotional functioning, emotional contributors to NES and alexithymia in individuals with NES (Pick et al, 2016). None of the studies explored the perceptions of links between life events, stressors and their seizures, in those with NES. Further detail of this area of work will be provided in the introduction to study 3.

1.6. Other relevant factors

Other relevant factors, which are not the direct focus of this thesis will be described briefly below.

Organic and functional disorders are often found to co-exist, presenting a complex 'clinical picture', whereby the foundation of the condition may be organic, but functional symptoms may present alongside (Roelofs et al, 2019). Functional disorders are also found to co-exist with psychological conditions such as depression, anxiety, and post traumatic stress disorder (PTSD). Co-morbid neurological conditions include traumatic brain injury, chronic pain, and intellectual disability (Baslet et al, 2016). Additionally, conditions, such as chronic conditions like migraines or asthma, or conditions such as fibromyalgia, chronic fatigue syndrome, and irritable bowel syndrome also often co-exist (Perez et al, 2016).

Somatisation

Somatisation is a broad concept referring to the presence of symptoms that are not attributable to an organic cause (De Gucht and Heiser, 2003) that involves the tendency to express distress in the form of physical symptoms. A fairly consistent finding in research on NES is that pwNES tend to report more non-NES physical symptoms than controls: either patients with epilepsy or the normal population (Brown and Reuber, 2016). However, there is limited evidence that people are 'converting' or expressing emotional problems through their physical symptoms, and other explanations are equally plausible e.g. somatosensory amplification (hypervigilance and the interpretation of sensations as threatening or implying illness) (Rief and Barsky, 2005). Brown and Reuber (2016) suggest that it is not enough to assume the presence of physical symptoms suggest pwNES are using somatisation or defensive processes, and argue it is an interpretative 'leap'. Stone et al (2004) found that patients with recent onset NES were more likely to attribute somatic rather than psychological causes for their problems, compared to epilepsy patients. They also found that pwNES reported more difficult life events prior to seizure onset, but also demonstrated a greater tendency to dismiss stressful events. Stone et al (2004) suggest there may be two ways this may be interpreted. One explanation could be that those with NES focus on physical symptoms and attribute their NES to a physical cause, which could indicate avoidance of emotional difficulties. Or the alternative explanation could be they are concerned that symptoms will be 'dismissed as 'all in the mind' by doctors' (Stone et al, 2004), so under report any emotional issues. This latter point may be related to the stigma that is often associated with having a psychiatric diagnosis.

Epilepsy

Approximately 10-15% of NES patients with persistent/chronic symptoms are found to have epilepsy (Duncan et al, 2011; Benbadis et al, 2001), and this is higher (30%) in those who also have a learning disability (Duncan and Oto, 2008). Although the two seizures disorders are commonly co-morbid, NES and epilepsy are considered as distinct disorders. The prognosis for patients with epilepsy is better than for patients with NES. Additionally, attitudes from healthcare professionals (HCPs) are at times more compassionate towards those with epilepsy since some physicians conceptualise NES to be pseudo or feigned events (Worsley et al, 2011; Rawlings et al, 2017).

Anxiety

Anxiety disorders have been reported to be present for 11-50% of pwNES (Baslet et al, 2016). Although a number of studies have reported high rates of comorbid anxiety, this has not been replicated in other studies (Brown and Reuber, 2016). The prevalence of anxiety appears to be slightly higher in pwNES than those with epilepsy with scores in the moderate range for both conditions. Anxiety may therefore reflect a general worry related to having seizures rather than a specific reaction to NES (Brown and Reuber, 2016), or a specific aetiological factor.

Only a small number of studies have investigated whether anxiety symptoms are present before or during a seizure in pwNES (Brown and Reuber, 2016). A postal survey by Reuber et al (2011) of 100 patients found that 17% of participants reported that seizures were “always” or “frequently” preceded by ‘emotional stress’; and 26% of respondents reported that seizures were “rarely” or “never” preceded by stress. Twenty six percent of participants reported ‘always/frequently’ feeling irritable or upset before a seizure. However, only a minority of participants were ‘regularly aware’ of physical symptoms of anxiety during a seizure.

Autonomic arousal appears to be commonly present before, during and after NES with associated physiological symptoms which may or may not be accompanied by feelings of fear and/or distress. It has been hypothesised that some seizures may be produced by dissociative responses to intense anxiety or they may be actual panic attacks that are misinterpreted by the patient or an inexperienced physician as a seizure (Brown and Reuber, 2016).

Depression

Depression is the most common co-morbid condition in pwNES (Abubakr et al, 2003; Lesser, 1996), with depressive disorders found in 57-85% of patients (Baslet et al, 2016). A recent systematic review reported that pwNES have a higher prevalence of depressive symptoms compared to patients with epilepsy, although not necessarily higher rates of diagnosable depressive disorders (Walsh et al, 2018). Depression in patients with both NES and epilepsy has been found to be associated with seizure frequency (LaFrance et al, 2010) and quality of life (Lehrner et al, 1999).

1.7. Summary and conclusions

Emotional processing appears to be an important factor in both understanding why NES develops and the reoccurrence of the phenomenon (NES). However, the current evidence is based upon experimental work that has a low to moderate methodological quality and there is, as yet no definitive evidence to support a mainstream theory in the field.

Key aspects of emotional processing have been described, including interoception, alexithymia, emotional regulation and executive functioning. Although all these factors have been studied in pwNES, it is unclear whether they are specific to NES manifestations or to the experience of seizures in general. Studies of pwNES, which have included a comparative group of patients with epilepsy, often report little difference between NES and epilepsy participants, whereas studies employing a control group of normal healthy controls are more likely to report differences with pwNES showing greater levels of emotional dysfunction than controls.

Comorbid conditions common in NES include organic, functional, and psychological conditions, including epilepsy, anxiety and depression. There is some evidence from a small number of studies that have employed a cluster analysis that suggest that there are subgroups of pwNES with distinctly different psychosocial profiles.

Emotional processing is of interest to those trying to understand how to treat patients with NES more effectively. Qualitative methods can be particularly useful in helping to understand how pwNES experience the phenomenon, the healthcare system and the impact of NES on their lives. Although various forms of life stress have been implicated in the aetiology and maintenance of NES, very few studies have explored how people themselves view the relationship between life stressors and their seizures.

The aim of this thesis was to better understand the relationship between interoception, alexithymia and executive functioning in pwNES and each factor’s role in the maintenance of NES phenomena. That is, this thesis investigated components considered to be key in emotional processing: detection (interoception), recognition (alexithymia), regulation and attention. The original aim included the intention to assess the specificity of emotional processing in NES by comparing NES patients, recruited from a NHS neurology outpatient clinic, with controls (study 2). This aim had to be modified and study 2 redesigned to comply with COVID-social distancing guidelines which meant it was not possible to include patients as originally planned. A further aim of the thesis was to gain a better understanding of how pwNES experience emotion and how they perceive the relationship between stressful life events and their seizures.

The thesis comprises three studies:

Study 1: A systematic review to determine the evidence for a relationship between interoception and other key aspects of emotional processing in studies which have used a heartbeat perception task to measure interoception (interoceptive accuracy). A further aim was to identify which commonly used heartbeat perception task would be most suitable for use in a clinical population.

Study 2: A cross-sectional study to compare interoceptive sensibility (self-report), alexithymia and executive functioning in pwNES versus healthy controls, and to explore the relationship between these three factors of emotional processing, and to test an interactional model between them. This study was originally designed to measure interoceptive accuracy as opposed to interoceptive sensibility and address some of the methodological concerns identified in the systematic review (Study 1). However, the original study had to be abandoned due to the COVID lockdown and a new study 2 designed to comply with COVID guidelines. This meant that some of the planned methodological improvements to design could not be incorporated into study 2 as originally hoped. Further details are provided in Chapter 3.

Study 3: A qualitative cross-sectional study using Interpretative Phenomenological Analysis to explore how pwNES experienced and understood their seizure events in the context of their emotional responses to recent life events.

CHAPTER 2: STUDY 1

A systematic review of the association between emotion and interoception, when measured using a heartbeat perception task

1.1. Introduction

The self-perception of internal bodily activity (visceral activity) is integral to some theories of emotion (Herbert et al, 2007), such as the James-Lange theory (James, 1884; Lange, 1885) and the somatic marker hypothesis (Damasio, 1994). Such theories stress the importance of the interaction between physiological arousal and cognitive processes (Moors, 2009). As Seth (2013) describes, emotions are psychological states presenting a fusion of behaviour, experience and visceral activity – laying the foundations for theories such as the somatic marker hypothesis (Damasio, 1994).

The Autonomic Nervous System (ANS) refers to 'a set of neural pathways connecting the brain and body. These pathways send information from the body about the status of organs and tissues' (Porges et al, 1993; 2015). The ANS contains two branches: the sympathetic, involved in activation (fight and flight responses) and the parasympathetic involved in relaxation (rest and digest responses) (Mauss and Robinson, 2009; Garfinkel and Critchley, 2016). These two branches tend to be in opposition to one another: meaning that if the sympathetic response is high then the parasympathetic will be low, and vice versa.

The ANS conveys information regarding physiological states such as heart rate, hunger and pain (Craig, 2002; Critchley et al, 2004; Pollatos et al 2007). Theories such as Damasio's Somatic Marker Hypothesis (1994) propose that our experience of emotion is associated with our perception of changes in our bodily state, and therefore the foundation of the emotional experience (Eshkevari et al, 2014). Meaning that interoception relates to how the individual perceives their body in relation to information from the ANS (Eshkevari et al, 2014; Kleckner et al, 2015).

Interoception involves the detection or perception of internal bodily changes, such as blood pressure or heartrate (Fustos et al, 2012) and therefore it is postulated that the better the individual is at detecting bodily changes, the greater their ability to detect their emotions (Pollatos and Schandry, 2008; Garfinkel and Critchley, 2016). That is, those who have an impaired sense of their physiological state will also 'have a correspondingly reduced experience of emotions (Critchley et al., 2004; Pollatos et al., 2008)' suggest Tanaka et al

(2021). It has been suggested that interoception relates to ‘bottom-up’ and ‘top-down’ processes (Seth, 2013) meaning that it is a process incorporating information not only from the current body state (e.g. heartrate) but also previous experience (Barrett, 2017). The role of interoception in the experiencing and processing of emotion is still a matter of debate (Domschke, et al, 2010) and methods used to investigate interoception are inconsistent (Garfinkel et al, 2015).

The various tests used to measure interoception are based on detection of visceral activity. Therefore, interoceptive tasks generally utilise methods based on cardiac, respiratory or gastric activity; although some studies have chosen to use questionnaires to explore awareness of bodily sensations (Muir et al, 2016, Betka et al, 2018, Longarzo et al, 2015). Cardiac perception tests are commonly referred to as heartbeat perception tasks (HPTs) (Murphy et al, 2017). They take two main forms – the mental tracking method (MTM) or the heartbeat discrimination task (HBD). The two tests differ: MTM requires the participant to perceive and count their heartbeats without taking their pulse. Heartbeat discrimination tasks require the participants to listen to tones, or flashes of light, and decide whether they are congruent or incongruent with their own heartbeat. Both tests are described in more detail below.

Several different aspects of interoception have been described in the literature, and these are listed in Table 1, together with their definitions and ways in which they can be measured. The table shows that interoception has been defined and investigated, using different concepts and paradigms. Heartbeat perception tasks refer to all tasks used to measure interoception, via heartbeat perception, regardless of whether the MTM or the HBD is utilised.

Table 1: Definitions of interoception found within the literature

Concept	**Definition**	**Measured using**
Interoception	Umbrella term referring to the detection or perception of internal bodily changes (Ceunen et al, 2016).	Heartrate; blood pressure; respiration; gastric load test.
Interoceptive accuracy	Accuracy with which the individual has counted/identified their heartbeats (Garfinkel et al, 2016).	Mental tracking method/ heartbeat discrimination task.
Interoceptive sensitivity	Degree to which an individual perceives 'the state of their body' (Murphy et al, 2017).	Mental tracking method/ heartbeat discrimination task.
Interoceptive sensibility	How accurate the individual believes their interoceptive ability to be (Murphy et al, 2017; Garfinkel et al, 2016; Mehling et al, 2012)	Questionnaires or visual analogue scale.
Interoceptive awareness	How accurate an individual's judgement of their performance on an interoceptive task (sensibility) is predictive of their interoceptive sensitivity (Garfinkel et al, 2015; Murphy et al 2017) – also referred to as metacognitive awareness.	Heartbeat discrimination task and visual analogue scale.

Garfinkel and Critchley (2016) highlight that whilst heartbeat perception is usually considered to be a 'stable constitutional trait of the individual', one's heartbeat may be affected by different factors such as exercise (Schandry et al, 1993) or stress (Schulz et al, 2013). Interoception has been investigated in relation to a variety of different medical conditions and mental phenomena, including: pain (Craig, 2003); medically unexplained symptoms (Schaefer et al, 2014); anxiety (Domeschke et al, 2010); emotions (Damasio, 1994); emotion regulation (Fustos et al, 2012); and Autistic Spectrum Disorder (Shah et al, 2016).

It has been noted that in some conditions, such as autism and Functional neurological disorder (FND), facets of ANS activity are altered. For example, people with autism tend to demonstrate a higher resting heart rate, compared to those without Autism, which means that individuals with Autism exhibit a higher activation of the sympathetic response at rest. In individuals with FND (Maurier et al, 2016) and non-epileptic seizures (Ponnusamy et al., 2011), it has been reported that individuals with these conditions have been found to demonstrate a lower resting heart rate when compared against healthy controls.

Additionally, pharmacological interventions may also have an impact on the ANS. For example, a study by Livermore et al (2022) reported that an increase in serotonin levels improved interoceptive accuracy of participants. That is after a selective serotonin reuptake inhibitor (SSRI) was administered, participants increased in their confidence of their judgement of their interoceptive accuracy. They explain that whilst SSRIs are used to treat depression and associated with blunting of ‘interoceptive processing’ (Pollatos et al, 2009), a single dose was associated with ‘enhanced symptoms of anxiety’ which has been associated with better interoception (Dunn et al, 2020). Alcohol has also been reported to be associated with reduced interoceptive accuracy in male participants’ performance on the MTM, at both high and low levels of physiological arousal (Abrams et al, 2018).

These studies highlight that other factors that may not be considered, such as medication, an individual’s resting heart rate, depression/anxiety may also have an impact on the individual’s interoceptive accuracy. Therefore, if one’s emotions are influenced by interoceptive information, difficulties with emotion processing may be demonstrated in those with altered ANS responses.

Domschke et al (2010) provided an overview of the use of heartbeat perception tasks. The authors identified three main methods that have been used to investigate heartbeat perception: mental tracking paradigms; tasks relating to ‘signal detection’; and ‘intra-individual correlation tasks’. These are each described in more detail below.

The premise of heartbeat perception tasks is based on the James-Lange theory of emotion (1884, 1885) and they have been used by numerous studies as a test for interoception; and are still widely used. The Mental Tracking Method (MTM) (Garfinkel et al, 2016), is also referred to as the Schandry task - named after Ranier Schandry (Schandry, 1981). The MTM requires the individual to silently count their heartbeat, without taking their pulse, for a set period of time, typically 25, 35 or 45 seconds. The participant then reports the number of heartbeats counted to the researcher, once the time period is over. The researcher monitors the participant’s heartrate during the counting phase and obtains an interoceptive accuracy score by comparing the reported and actual heartbeats.

The Heartbeat Discrimination method (HBD) relates to signal detection theory (Ehlers et al, 1988). This task was developed by Whitehead et al (1977). The task requires the individual to listen to a set of tones. The participant then reports to the researcher whether the tones are congruent or incongruent with their own heartbeat.

Intra-individual correlation tasks are used to calculate a correlation based on the participant's self-report on how accurate they believe their performance to be against the actual heartbeat recorded (Pennebaker et al, 1982; Tyrer et al, 1981). This method does not appear to specify whether the MTM or HBD is used, which is left to the choice of the researcher. Metacognitive awareness is described (Garfinkel et al, 2015; Murphy et al, 2017) as the association between the individual's confidence in their judgement of their heartbeat accuracy and the individual's actual performance on the heartbeat perception task. That is, intra-individual correlation tasks essentially refer to metacognitive awareness of interoceptive sensibility. In the study by Garfinkel et al (2016), this was calculated by conducting a Receiver Operating Curve (ROC) analysis and reporting the area under the curve as the measure of accuracy.

In summary, heartbeat perception tasks are frequently used as a measure of interoception. The literature, however, highlights differences in the way in which interoception may be conceptualised, measured or reported, although, there have been efforts towards standardising methods and definitions by a number of authors - see table 1 for examples.

Interoception is a complex phenomenon of increased interest, especially in relation to emotion. To the author's knowledge, no reviews, thus far, have explored the relationship between interoception and emotional states, when interoception has been measured using a heartbeat perception task.

2.2 Aims and objectives

The aim of this review was to determine whether there is a relationship between interoception and a range of emotional states/conditions – based on an association between emotion and interoception measured using heartbeat perception tasks. A secondary aim was to identify which is the most commonly used heartbeat perception task and the most suitable for clinical populations.

Objectives:

1. To determine the relationship between interoception and a range of emotions/emotional traits or states, when interoception has been measured using heartbeat perception tasks.

2. To identify which heartbeat perception paradigms have been used most frequently as measures of interoception in studies of emotion and interoception.

3. To identify which of the heartbeat perception paradigms may be the most useful in investigating interoception in clinical groups.

2.3. Methods

2.3.1 Literature search

The databases EMBASE, MEDLINE and PsychINFO were used to search for relevant publications. The time period for the EMBASE database was 1947 to July 14, 2018; the MEDLINE database was searched for the time period 1946 to July 14, 2018; and PsychINFO 1806 to July 14, 2018. Abstract and title searches were carried out using the key concepts interocep*; emo*; alexithymia; heartbeat percep*; Schandry*; mental track*; heartbeat discrimination, in the three databases. The databases were also searched using the terms (interocep* adj accuracy); (heart adj beat adj3 detect*); (heart adj beat adj3 percep*); (heart adj beat adj3 discrimination); (Schandry adj task) to identify suitable papers.

2.3.2 Inclusion and exclusion criteria

The inclusion criteria were: studies conducted with adult populations; published in the English language. The definition for the adult age group varied between databases: EMBASE defined adults as those being between 18-64 years of age; Medline had a cut-off for adult participants at least 19 years old; PsychINFO <18 + years were considered as adults. All studies were required to have measured interoception using a heartbeat perception task, and to have included a recognised measure of emotion or a measure of a clinical/affective condition (e.g. anxiety, alexithymia, depression).

The exclusion criteria were: studies published in a language other than English; studies not using human participants. Studies of children or adolescent populations were excluded on the basis that the study that would be subsequently designed, based on the findings of this review, would be with an adult population.

2.3.3 Screening

Duplicate papers were identified and deleted from the combined search results. Remaining articles were screened by title and abstract to identify papers that met the inclusion criteria. Papers were included if they referred to the use of a heartbeat perception task and referred to the measurement of emotion or an affective state (anxiety or depression) or alexithymia.

Studies were rejected if, for example, they focussed on a task other than the heartbeat perception tasks, they did not refer to investigating interoception, or the study was concerned with empathy. This was because, although empathy is concerned with the recognition of emotion, it relates to recognising emotion in others whereas this review is focussed on how the individual perceives their own emotion.

2.3.4 Quality assessment of studies

The “Quality Assessment Tool for Quantitative Studies” was used to rate study quality (Effective Public Practice Project, 1998). Individual studies were rated on a range of areas: selection bias; study design; confounders; blinding; data collection methods; withdrawals and dropouts; intervention integrity; analysis. The authors state that the tool offers a standardised measure by which study quality can be assessed. It has been developed to be used for systematic reviews. This tool is suitable for use with non-randomised studies and so was relevant to the studies returned by the search. It is a tool that has been used in multiple systematic reviews and has been validated for use in health research (Armijo-Olivio et al, 2012). Study quality is rated as ‘strong, moderate or weak’ over all categories. The overall rating is based on five of the eight categories.

2.3.5 Data extraction

Descriptive data were collected from the included studies. These were items such as study design; sample size; age; interoceptive dimension of interest; the heartbeat perception task used; and measurement of emotions/emotional states.

Effect sizes were reported from those studies that reported associations relevant to this review (see table 6).

2.4. Results

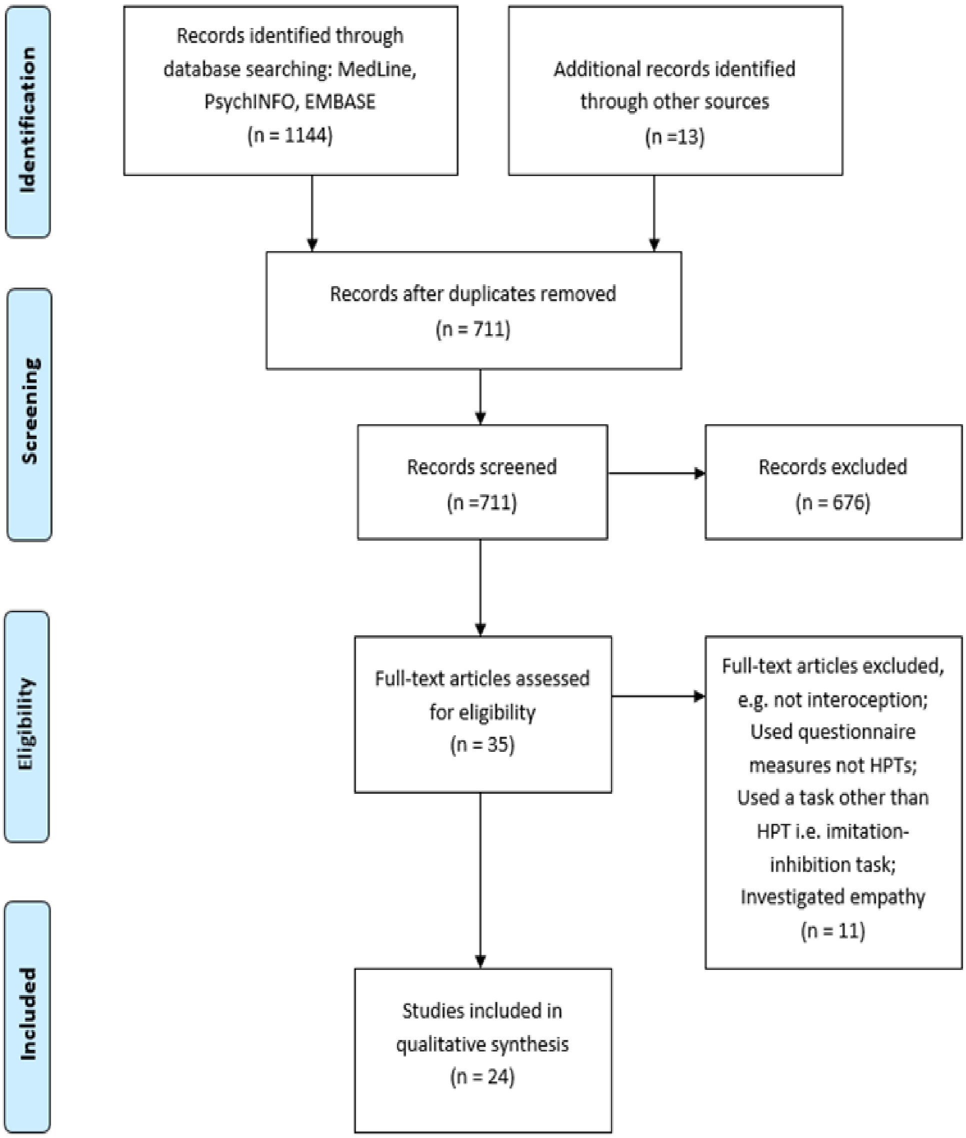
2.4.1. Study selection

The search returned a total of 1144 papers. After exclusion criteria were applied and duplicates removed, 711 papers remained (see figure 2.1 for flow chart showing selection of eligible studies). After screening titles and abstracts 22 papers were retained. Another 13 papers were identified and added from hand searches and in total 35 papers were downloaded in full. After assessing the papers for eligibility, 24 papers were found to meet the aims of the review and included in the final analysis. The included studies were published between 1981 and 2018.

2.4.2. Study characteristics

Table 2 summarises the main results from the studies under review. The table reports the study characteristics, sample characteristics and the method of investigating interoception - i.e. MTM or HBD.

The overall sample size was N=2762, of which 1835 participants were female (66%). The largest sample size was 998 (Zamariola et al, 2018) and the smallest was 20 participants (Pollatos et al, 2007). The mean age of participants was 34 years based on data from 21 studies (88%), with a range of 22 to 61 years (Wiens et al, 2000; Ricciardi et al, 2016b). One third of the studies (n=8) involved studies of people with alexithymia. Depression and anxiety and their association with interoception were investigated in four and six studies, respectively. The most common study design employed was a cross sectional study (N=21, 88%), with a cohort design being used by two studies (8%). One study used a case-control design (Schaefer et al, 2014).



From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

Figure 2.1: flow chart of study selection based on PRISMA diagram for systematic reviews

Table 2: Study characteristics: summary table presenting study characteristics, heartbeat perception tasks used and relevant findings. Age was reported differently across studies and is reported in the table based on the information available in the papers.

Study	Design	Sample size	F	M	Age (range/ mean)	BMI measured?	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable	Questionnaires
Schandry (1981)	Cross sectional	39	16	23	18-40	N	Accuracy	Mental tracking	N	Anxiety	STAI[1]
Aim		To test whether good heartbeat perceivers will report higher levels of emotional experience, i.e. anxiety.									
Relevant findings		Those with accurate heartbeat perception also show higher State Anxiety and that emotional experience is associated with interoception.									
Weins et al (2000)	Cross sectional	52	33	19	18-41 21.8	N	Accuracy	Signal detection	N	Valence	N/A
Aim		To investigate the relationship between self-report of emotional experience and interoception.									
Relevant findings		Reported higher emotional intensity associated with more accurate heartbeat perception.									
Barrett et al (2004)	Cohort	109	76	33	Not reported	N	Sensitivity	Signal detection	N	Arousal Focus	N/A
Aim		To investigate the association between interoception and arousal focus.									
Relevant findings		Study 1 reported that Arousal Focus was correlated with IS (r=0.23; p<0.05). Study 2 reported the same relationship.									
Herbert et al (2007)	Cross sectional	37	18	19	20-43 Good:25.2 Poor: 27.2	N	Sensitivity	Mental tracking	N	Valence and Arousal	N/A
Aim		To investigate whether accurate heartbeat perceivers rate emotional experience with more intensity.									
Relevant findings		Reported a positive correlation between being an accurate heartbeat perceiver and experiencing emotional stimuli with more intensity i.e. arousal.									
Pollatos et al (2007)	Cross sectional	20	0	20	21-34 26.8	N	Awareness	MTM	N	Anxiety	STAI[1]
Aim		To assess whether interoception and cardiovascular arousal are processed by same brain regions, and whether that are affected by emotion.									
Relevant findings		Reported a significant association between anxiety and interoception (R=0.58; p,0.05)									

Table 2 cont.

Study	Design	Sample size	F	M	Age (range/ mean)	BMI measured?	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable	Questionnaires
Dunn et al (2010)	Cross sectional	58	35	23	45	N	Accuracy	Mental Tracking	N	Arousal and valence	None
Aim		To investigate the level to which interoception is related to arousal and valence.									
Relevant findings		Higher interoceptive accuracy associated with higher arousal ratings.									
Herbert et al (2011)	Cross sectional	155	87	67	28.65 Women: 28.50 Men: 28.97	Y	Awareness	Mental tracking	N	Depression and alexithymia	TAS[2] BDI[3]
Aim		To investigate whether interoception and alexithymia are associated.									
Relevant findings		Reported that interoceptive awareness was negatively correlated with alexithymia. That is, poorer interoceptive awareness is associated with higher levels of alexithymia.									
Fustos et al (2012)	Cross sectional	28	18	10	25.5	N	Awareness	Mental tracking	N	Emotion regulation	None
Aim		To investigate whether interoception associated with better ability to regulate negative emotions.									
Relevant findings		Higher interoceptive awareness is associated with better emotion regulation towards negative affect.									
Terhaar et al (2012)	Cross sectional	32	25	7	Patients: 41.75 Controls: 39.81	N	Awareness	Mental tracking and Signal detection	N	Depression	HAMD[4] BDI[3]
Aim		To investigate whether heartbeat perception reduced in individuals who are depressed.									
Relevant findings		Reported that depressed participants demonstrated poorer interoception when compared to controls (d=0.85; p=0.011), and heartbeat evoked potentials also reduced in depressed participants.									
Kindermann et al (2014a)	Cross sectional	40	22	18	High: 23.75 Low: 23.90	Y	Accuracy	Mental tracking	N	Anxiety	STAI[1]
Aim		To investigate whether individuals with good heartbeat perception report more negative emotions during stress.									
Relevant findings		Reported that under stress conditions, those with better heartbeat perception report more negative emotions compared to participants that demonstrated low heartbeat perception.									

Table 2 cont.

Study	Design	Sample size	F	M	Age (range/ mean)	BMI measured?	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable	Questionnaires
Kindermann et al (2014b)	Cross sectional	50	24	26	23.92	N	Accuracy	Mental tracking	N	Good Bad mood	N/A
Aim		To investigate whether individuals with more accurate heartbeat perception report more negative emotions when experiencing mental stress.									
Relevant findings		Having more accurate heartbeat perception is associated with experiencing more negative emotions during stress.									
Mallorquí-Bagué et al (2014)	Cross sectional	36	20	16	24.1	N	Sensitivity	Mental tracking and Signal detection	Y	Anxiety	STAI[1]
Aim		To investigate whether interoception associated with anxiety in individuals with sub-clinical hypermobility.									
Relevant findings		Reported a positive correlation between state anxiety and interoceptive sensitivity.									
Schaefer et al (2014)	Case control	52	38	14	Training group: 40.07 Control: 45.26	Y	Accuracy	Mental tracking	N	Depression	BDI[3]
Aim		To investigate in improving interoception leads to a decrease in symptom severity in somatoform disorders.									
Relevant findings		Reported that patients with somatoform disorders demonstrate lower interoceptive accuracy.									
Pollatos et al (2015)	Cross sectional	116	89	27	25.6	N	Sensitivity	Mental tracking	N	Emotion regulation	ERQ[5]
Aim		To investigate whether interoception is associated with coping with social exclusion, and better emotion regulation.									
Relevant findings		Interoceptive sensitivity associated with emotion regulation.									
Demartini et al (2016)	Cross sectional	60	48	12	FMS: 45.7 PNES: 45.9 Controls: 43.1	Y	Sensitivity	Mental tracking	N	Alexithymia	TAS[2]
Aim		To investigate the phenomenology of patients with functional neurological disorders									
Relevant findings		Reported no significant difference on interoceptive awareness between the groups.									

Table 2 cont.

Study	Design	Sample size	F	M	Age (range/ mean)	BMI measured?	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable	Questionnaires
Garfinkel et al (2016)	Cross sectional	40	4	36	ASD: 28.06 Controls: 27.81	N	Accuracy	Mental tracking and Signal detection	Y	Anxiety	STAI[1]
Aim		To investigate whether individuals with Autistic Spectrum Disorder showed impaired interoception, and show higher levels of subjective interoceptive perception than healthy control participants.									
Relevant findings		Individuals with Autistic Spectrum Disorder have reduced interoceptive accuracy.									
Pollatos and Georgiou (2016)	Cross sectional	46	46	0	Patients: 24.0 Controls: 25.1	Y	Accuracy	Mental tracking	Y	Anxiety	STAI[1]
Aim		To investigate whether individuals with bulimia nervosa show impaired interoception compared with controls									
Relevant findings		Partial correlation between interoceptive accuracy and anxiety in control group, when BMI controlled for (*r*=-0.42, *p*<0.05).									
Ricciardi et al (2016a)	Cross sectional	33	24	9	Patients: 41.5 controls: 33	Y	Sensitivity	Mental tracking	N	Depression	MADRS[7] TAS[2]
Aim		To investigate whether individuals with functional motor disorder show lower interoception than healthy control and whether it is associated with alexithymia and depression.									
Relevant findings		Reported that reduced interoception was negatively correlated with depression when participants pooled (i.e. patients + controls).									
Ricciardi et al (2016b)	Cross sectional	40	19	21	Patient: 61.4 Control: 56.5	N	Sensitivity	Mental tracking	N	Alexithymia	TAS[2] HAMD[4]
Aim		To investigate if individuals with Parkinson's disorder show impaired interoception, and whether it is associated with symptoms such as depression and anxiety.									
Relevant findings		No significant correlations reported between interoceptive sensitivity and alexithymia, or with depression or anxiety.									

Study	Design	Sample size	F	M	Age (range/ mean)	BMI measured?	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable	Questionnaires
Shah et al (2016)	Cross sectional	76	30	46	Study 1: 29.1 Study 2: patient: 32.9 Controls: 32.9	N	Accuracy	Mental tracking	N	Alexithymia	TAS^{2} BDI^{3} STAI^{1}
Aim		Investigate the association between interoception, autism and alexithymia.									
Relevant findings		Interoceptive accuracy was reported to be associated with alexithymia.									
Bornemann and Singer (2017)	Cohort	318	187	131	20-55 40.8	N	Accuracy	Mental tracking	N	Alexithymia	TAS^{2}
Aim		To investigate the impact of interoceptive training on emotional awareness.									
Relevant findings		Alexithymia and interoceptive accuracy are correlated when individuals undergo training designed to make them more aware of their body.									
Christensen et al (2018)	Cross sectional	40	40	0	Dancers: 23.25 Controls: 25.35	N	Accuracy	Mental tracking	N	Alexithymia	TAS^{2} BVAQ^{6}
Aim		To investigate whether interoception is associated with alexithymia, and whether more accurate interoception is linked to being in the arts.									
Relevant findings		No significant correlation between interoceptive accuracy and alexithymia.									
Murphy et al (2018)	Cross sectional	287	201	86	18-90 38.07	Y	Sensitivity	Mental tracking	N	Alexithymia	STAI^{1} BDI^{3} TAS^{2}
Aim		Investigate whether impaired interoception associated with alexithymia.									
Relevant findings		No significant correlations reported between interoceptive accuracy and alexithymia.									
Zamariola et al (2018)	Cross sectional	998	735	263	19.86-22.25	N	Accuracy	Mental tracking	Y	Alexithymia	TAS^{2}
Aim		To investigate whether different dimensions of interoception related to alexithymia.									
Relevant findings		Reported that there was no significant correlation between alexithymia and interoceptive accuracy.									

Questionnaires used by the studies: 1 STAI = State-Trait Anxiety Inventory; 2 TAS = Toronto Alexithymia Scale; 3 BDI = Beck Depression Inventory; 4 HAM-D = Hamilton Depression Rating Scale; 5 ERQ= Emotion Regulation Questionnaire; 6 BVAQ=Bermond-Vost Alexithymia Questionnaire; 7 MADRS = Montgomery Äsberg Depression Rating Scale

2.4.3. Studies with normal population samples

Sixteen studies had non-clinical (i.e. normal population) samples with an overall sample size of 2383. Table 3 reports the demographic data from these studies.

Out of the 16 studies, 10 studies (63%) recruited students either for the whole study sample or students made up the majority of the study participants. Three of the 10 studies also recruited some participants who were employed. Six studies (38%) recruited healthy volunteers with no further details supplied. The study by Christensen et al (2018) compared undergraduate students with those being trained as professional dancers. The whole sample (N=40) could be essentially described as a student sample.

Age of participants in studies using normal samples

Mean ages in the studies of non-clinical populations ranged from 21.8 to 45.0 years of age. Three studies did not report mean age: that is, two studies reported the age range for participants (Schandry, 1981; Herbert et al, 2007) and one study did not report age at all (Barrett et al, 2004).

Sex reported in studies using normal samples

Approximately two thirds of the overall sample was female (N=1602; 67%) compared to male participants (N=781; 33%); although some individual studies were found to include more male participants in their samples (Schandry, 1981; Herbert et al, 2007; Kindermann and Werner, 2014a).

One study recruited female participants only (Christensen et al, 2018). And one study recruited male participants only (Pollatos et al, 2007).

The largest sample size reported was 998 by Zamariola et al (2018). The sample was made up of 735 women and 263 men.

Study location of studies which included normal population samples

Nine studies were conducted in Germany; two were carried out in North America; four in England and one study was conducted in Belgium.

2.4.4 Studies with clinical populations

Approximately a third of the total number of the studies in the review (N=8; 33%) reported using clinical samples, with an overall sample size of 379 participants. All eight of these studies included a control group with which patient participants were compared. Across all eight studies 158 participants (42%) were included as controls and 221 participants (58%) as patient participants (Table 4).

The different clinical conditions included depressive symptoms (Terhaar et al, 2012); somatoform disorder (Schaefer et al, 2014); functional neurological disorders (Demartini et al, 2016; Riccardi et al 2016a); Autistic Spectrum Disorder (Garfinkel et al, 2016; Shah et al, 2016); eating disorder (Pollatos and Georgiou, 2016); and Parkinson's disorder (Ricciardi et al, 2016b).

Age of participants in studies using clinical samples

Mean ages in the studies of with clinical samples ranged from 24.0 to 61.4 years of age.

Mean ages for control samples across these eight studies were reported as ranging from 25.1 to 56.5 years of age. Higher mean ages for both patient and control participants were found in the study investigating Parkinson's disease (Ricciardi et al, 2016b), a condition that typically affects adults over 50 years of age.

Sex of participants in studies using clinical samples

Participants in the eight studies with clinical populations were predominantly reported as female (N= 244). Over 60% of the overall sample was female (64%) compared to male participants making up 36% (N=135) of the sample. That is, similar to studies with normal samples, there were almost twice as many female participants compared to male participants.

However, half of the eight studies (N=4) in this section were found to include more male participants in their samples (Schaefer et al, 2014; Garfinkel et al, 2016; Riccardi et al, 2016b; Shah et al, 2016).

Two of the four studies with higher male participants were investigating Autistic Spectrum Disorder and Parkinson's disease. These are conditions that are commonly reported/detected more in men than women.

One study recruited female participants only (Pollatos and Georgiou, 2016). This study investigated interoception in those with Bulimia Nervosa. Eating disorders are typically reported and diagnosed in more women than men.

Study location of studies which included clinical populations

Reporting of the location of where the study was conducted, varied across studies.

Six studies reported the institute from where participants were recruited, indicating the country in which the study was held (Germany: Terhaar et al, 2012 Schaefer et al, 2014; England: Garfinkel et al, 2016; Italy: Demartini et al, 2016; Ricciardi et al 2016a; Ricciardi et al 2016b;).

Two studies did not report the location and this detail was extrapolated based on the location reported for authors (Pollatos and Georgiou, 2016; Shah et al, 2016).

Table 3: studies using normal population samples.

This table displays demographic data available in studies concerned with non-clinical populations (N=16).

Study	Country	Mean age (years)	Sample size (N=)	Female (N=)	Male (N=)	Students (N=)	Employed (N=)
Schandry (1981)	Germany	Not reported	39	16	23	29	10
Wiens et al (2000)	North America	21.8	52	33	19	52	0
Barrett et al (2004)	North America	Not reported	109	76	33	109	0
Herbert et al (2007)	Germany	Not reported	37	18	19	37	0
Pollatos et al (2007)	Germany	26.8	20	0	20	Not reported	Not reported
Dunn et al (2010)	England	45	58	35	23	Not reported	Not reported
Herbert et al (2011)	Germany	28.65	155	88	67	155	0
Fustos et al (2012)	Germany	25.5	28	18	10	28	0
Kindermann and Werner (2014a)	Germany	23.92	50	24	26	46	4
Kindermann and Werner (2014b)	Germany	High: 23.75 Low: 23.90	40	22	18	36	3
Mallorqui- Bagué et al (2014)	England	24.1	36	20	16	Not reported	Not reported
Pollatos et al (2015)	Germany	25.6	116	89	27	Not reported	Not reported
Bornemann and Singer (2017)	Germany	40.8	318	187	131	Not reported	Not reported
Christensen et al (2018)	England	Students: 24.25 Dancers: 25.35	40	40	0	40	0
Murphy et al (2018)	England	38.07	287	201	86	Not reported	Not reported
Zamariola et al (2018)	Belgium	21.27	998	735	263	998	0
Total			2383	1602	781	1530	17

Table 4. Studies using clinical samples.

This table displays demographic data available in studies compared with clinical populations (N=8).

Study	Country	Mean age (years)	Sample size (N=)	Female (N=)	Male (N=)	Students (N=)	Employed (N=)
Terhaar et al (2012)	Germany	Patients: 41.8 Controls: 39.8	32	25	7	16	16
Schaefer et al (2014)	Germany	Patients: 40.07 Controls: 45.26	52	48	4	23	29
Demartini et al (2016)	Italy	Patients: FMS 45.7 PNES 45.9 Controls: 43.1	60	48	12	20	40
Garfinkel et al (2016)	England	Patients: 28.1 Controls: 27.8	40	4	36	20	20
Pollatos and Georgiou (2016)	Germany	Patients: 24.0 Controls: 25.1	46	46	0	23	23
Ricciardi et al (2016a)	England	Patients: 41.5 Controls: 33	33	24	9	17	16
Ricciardi et al (2016b)	England	Patients: 61.4 Controls: 56.5	40	19	21	20	20
Shah et al (2016)	England	Study 1: Patients: 29.1 Study 2: Patients: 32.9 Controls: 32.9	76	30	46	19	57
Total			379	244	135	158	221

2.5. Assessment of study quality and potential sources of bias

Bias occurs when a systematic error in the study methodology leads to incorrect or inaccurate reporting of results. The Quality Assessment Tool for Quantitative Studies (Effective Public Health Practice Project, 1998) was used to rate several areas typically considered to have the potential for bias and the findings are reported below. Table 5 shows the individual ratings by study and category and provides the overall quality rating for each of the studies.

2.5.1 Overall quality assessment rating for all 24 studies.

Overall two thirds of studies were rated as having a 'weak' level of quality (N=16), one third of (N=8) studies were rated as 'moderate' and no studies were rated as 'strong' (see Table 5).

The assessment tool rates studies with respect to the management of withdrawals/ dropouts within studies. In this review, this was not relevant to most studies as they required participants to perform a heartbeat perception task on one occasion only and therefore no follow up was required by the study. Five studies, however, were rated under this category since they either required participants to return and complete further tasks (Barrett et al, 2004; Schaefer et al, 2014; Bornemann et al, 2017), or reported the number/reasons for participants being withdrawn such as equipment failure (Dunn et al, 2010) or 'invalid' heartbeat evoked potential results (Terhaar et al, 2012).

2.5.2. Individual category ratings

Selection bias

Selection bias occurs when the characteristics of participants selected to take part in the research do not represent the general population from which they were drawn (Henderson and Pahe, 2007).

In this review, over half of the studies were rated as 'weak' (N=14; 58%) in regard to selection bias.

Normal population studies (non-clinical)

Of the 16 studies investigating interoception in normal populations, 14 (88%) were rated as being a 'weak' study with regards to study quality.

These studies recruited participants from universities and so samples consisted largely of students (e.g. Schandry et al, 1981; Wiens et al, 2000; Herbert et al, 2011; Zamariola et al, 2018).

Clinical population studies

A quarter of the studies (N=2) investigating interoception in a patient population were rated as ‘weak’, with the majority of the studies (N=6; 75%) achieving a ‘moderate’ rating of overall quality.

Seven studies with patient populations reported recruiting participants from clinics. One study reported the use of separate databases to recruit both patients and controls (Shah et al, 2016). One study (Terhaar et al, 2012) did not report how participants were recruited.

According to the rating tool, studies rated as ‘medium’ indicate that samples are only ‘somewhat likely’ to be representative of the target population.

Information on how control participants were recruited in studies using a case-control design, were not provided.

2.5.2.1. Study design

Normal population studies

Of the 16 studies testing interoception in normal populations, the majority of studies used a cross-sectional study design (N=15). They were therefore assessed to be ‘weak’ using the assessment tool rating.

Clinical population studies

Similar to the studies using normal populations, the majority of studies investigating clinical populations were rated to have a ‘weak’ study design. That is, seven of the eight studies were rated as ‘weak’. The study by Schaefer et al (2014) achieved a ‘moderate’ rating.

2.5.2.3 Data collection

All studies in this review used standardised measures except for the study by Barrett et al (2004). They used a ‘paper and pen’ exercise that required participants to use a Likert scale to rate emotion related adjectives taken from the Positive and Negative Affective Schedule (Watson and Clark, 1994).

2.5.2.4 Confounding variables

Confounding factors may be variables that appear to be inconsequential however could still influence the outcome in some way – e.g. sex or age. Matching of participants in case control studies, i.e. matching cases and control participants on key characteristics can be a method used to reduce confounders. Regression analyses may also be conducted as a way in which to ‘control’ for certain co-factors.

With regards to how studies dealt with confounding factors: a third of the all the studies included in this review (N=8; 33%) were rated as ‘weak’ and over half (N=13; 54%) of studies were rated as ‘strong’.

Studies rated as strong were likely to have matched participants on common characteristics, usually age, sex, and BMI. There is evidence that these factors may influence heartbeat perception and this explains why these participant characteristics were matched or accounted for in a regression analyses.

Normal population studies

Half of the studies (N=8; 50%) were assessed as ‘weak’ under this item. Three studies were assessed as being ‘moderate’ in their rating. Therefore, five studies (31%) achieved a ‘strong’ rating in relationship to managing confounding variables.

Clinical population studies

All eight studies investigating interoception in clinical populations were assessed to be ‘strong’ using the rating scale. This is based on matching controls to patients on specific characteristics. Age and sex were variables used to match controls to patients, with one study matching on BMI too (Ricciardi et al, 2016a).

2.5.2.5 Power and effect

Power is the ability of a test to detect a relationship or a difference between two samples. Power should be estimated a priori and relates to the sample size required to provide sufficient power to reject the null hypothesis when it is false.

Three out of all the studies in this review (N=24) reported a sample size calculation. Two studies were using normal populations (Mallorquí-Bagué et al, 2014; Christensen et al, 2018). One study with a clinical sample reported a sample size calculation (Shah et al, 2016).

Most studies were found to report small sample sizes (N=17), meaning they may not have had enough power to report a true effect if found.

Table 5: Assessment of risk of bias in included studies

Study (*investigated clinical samples)	Selection bias	Study design	Confounders	Data collection	Withdrawals/ dropouts	**Overall rating**
Schandry (1981)	moderate	weak	weak	moderate	N/A	**Weak**
Wiens et al (2000)	weak	weak	weak	moderate	N/A	**Weak**
Barrett et al (2004)	weak	moderate	weak	weak	weak	**Weak**
Herbert et al (2007)	weak	weak	moderate	strong	N/A	**Weak**
Pollatos et al (2007)	weak	weak	weak	weak	weak	**Weak**
Dunn et al (2010)	weak	weak	weak	moderate	strong	**Weak**
Herbert et al (2011)	weak	weak	moderate	strong	N/A	**Weak**
Fustos et al (2012)	weak	weak	weak	weak	N/A	**Weak**
Terhaar et al (2012)*	moderate	weak	strong	weak	strong	**Weak**
Kindermann and Werner (2014) a	weak	weak	strong	moderate	N/A	**Weak**
Kindermann and Werner (2014) b	weak	weak	strong	weak	N/A	**Weak**
Mallorquí-Bagué et al (2014)	weak	weak	strong	moderate	N/A	**Weak**
Schaefer et al (2014)*	moderate	moderate	strong	strong	weak	**Moderate**
Pollatos et al (2015)	weak	weak	weak	moderate	N/A	**Weak**
Demartini et al (2016)*	moderate	weak	strong	moderate	N/A	**Moderate**
Garfinkel et al (2016)*	moderate	weak	strong	moderate	N/A	**Moderate**
Pollatos and Georgiou (2016)*	moderate	weak	strong	strong	N/A	**Moderate**
Ricciardi et al (2016a)*	moderate	weak	strong	moderate	N/A	**Moderate**
Ricciardi et al (2016b)*	moderate	weak	strong	moderate	N/A	**Moderate**
Shah et al (2016)*	moderate	weak	strong	weak	N/A	**Weak**
Bornemann et al (2017)	weak	moderate	strong	strong	strong	**Moderate**
Christensen et al (2018)	moderate	weak	moderate	moderate	N/A	**Moderate**
Murphy et al (2018)	weak	weak	strong	moderate	N/A	**Weak**
Zamariola et al (2018)	weak	weak	weak	moderate	N/A	**Weak**
Total number of studies with overall rating of:					Weak **16**	
					Moderate **8**	
					Strong **0**	**Total =24**

2.6. The relationship between interoception and emotional states/conditions.

The first objective of this review was to determine the relationship between interoception, and emotions/emotional states based upon studies of interoception which have used heartbeat perception tasks. In this review, it was found that reporting of associations between interoception and emotional variables was inconsistent. For example, although a study may have included a measure of anxiety, the study may not have explored an association between the two variables. Table 6 shows the studies which investigated associations between interoception and emotional states: and if reported by the study, the strength of the correlation and whether found to be significant. The table also includes details of the questionnaires used to measure emotional state e.g. (depression/anxiety) or emotion recognition (alexithymia).

From this point onwards, interoception will be used as a unifying term to include and refer to all the interoceptive experiences measured and reported by the studies included in this review.

2.6.1. Emotion

The relationship between basic emotions and interoception was investigated explicitly by only one study - Wiens et al (2000). Basic emotions are those that we may consider as universal or innate (Allport, 1924; Darwin; 1965; Ekman, 1972; Panksepp, 1998), such as sadness, anger, fear.

The study by Wiens et al (2000) investigated emotional experience and used the emotional descriptors of amusement, fear and anger. The authors were interested in the relationship between heartbeat perception and individual emotional experience. They hypothesised that those with more accurate heartbeat perception would report a more intense emotional experience, when compared to those with less accurate heartbeat detection. This was explored using the heartbeat detection test in 52 participants. Film clips were used as the emotional stimulus and valence and intensity ratings were recorded. The authors reported that ‘good heartbeat detectors’ reported a more intense experience of emotion whilst watching the film clips, compared to the ‘poor heartbeat detectors’, and this was across all three film categories: amusement/anger/fear. The authors found a significant relationship between intensity ratings of emotion and heartbeat detection.

Table 6. Table showing the relationship between interoception, and emotional variable investigated by studies.

Relationship	**Study**	**Sample size**	**Questionnaire**	**Effect size**		**Significant**
Interoception and depression	Herbert et al (2011)	155	BDI^1	SMALL^c	$r=-0.21$, $p<0.05$	Y
	Terhaar et al (2012)	32^2	HAMD^2/BDI^1	LARGE	$d=0.85$, $p=0.011$	Y
	Ricciardi et al (2016) a	33	MADRS^3	MEDIUM^c	$r_s=-0.47$, $p=0.02$	Y
	Ricciardi et al (2016) b	40	HAMD^2	Not reported	----	N
Interoception and anxiety	Schandry (1981)	39	STAI^5	Not reported	------	Y
	Pollatos et al (2007)	20	STAI^4	LARGE^c	$R= 0.58$, $P<0.05$	Y
	Kindermann et al (2014) a	40	STAI^4	SMALL^a	$d=0.02$, $p=0.96$	N
	Mallorquí-Bagué et al (2014)	36	STAI^4	SMALL^c	$r=0.284$, $p=0.046$	Y
	Garfinkel et al (2016)	40	STAI^4	SMALL^c	$r=0.26$, $p=1.00$; $r=-0.22$, $p=1.00$	N
	Ricciardi et al (2016) b	40	HAM-A^4	Not reported	----	N
	Pollatos et al (2016)	46	STAI^5	SMALL^c	$r=0.14$, $p=0.36$	N
Interoception and alexithymia	Herbert et al (2011)	155	TAS^6	MEDIUM^c	$r=-0.37$, $p<0.01$	Y
	Ricciardi et al (2016) a	33	TAS^6	MEDIUM^c	$r_s=-0.30$, $p=0.28$	N
	Ricciardi et al (2016) b	40	TAS^6	Not reported	----	N
	Shah et al (2016)	76	TAS^6	MEDIUM^c	$r=-0.36$, $p=0.025$	Y
	Bornemann et al (2017)	318	TAS^6	SMALL^a	$r=0.207$, $p=0.002$	Y
	Murphy et al (2018)	287	TAS^6	NONE^c	$r=-0.079$, $p=0.182$	N
	Zamariola et al (2018)	414	TAS^6	SMALL^c	$r=-0.02$, $p>0.05$; $r=-0.03$,	N
		160		MEDIUM^c	$p>0.05$	N
Interoception and intensity of emotion	Wiens et al (2000)	52	N/A	Not reported	----	Y
	Barrett et al (2004)	109	N/A	SMALL^c	$r=0.23$, $p<0.05$	Y
	Herbert et al (2007)	155	N/A	MEDIUM^c	$r_s= 0.32$, $p<0.05$; $r_s= 0.55$,	Y
				LARGE^c	$p<0.001$	Y

	Dunn et al (2010)	58	N/A	Not reported	---	N
	Kindermann et al (2014) a	40	PANAS[7]	LARGE[a]	*d*=0.84, *p*=0.02	Y
	Kindermann et al (2014) b	50	MMQ[8]	SMALL	η_p^2 η =0.05, *p*=0.11	N
Interoception and emotion regulation	Fustos et al (2012)	28	SAM[9]	SMALL[c]	*r*=0.48, *p*<0.01	Y
	Pollatos et al (2015)	116	ERQ[10]	MEDIUM[c]	*R*=.38, *p*<0.05	Y

Effect size reported using either Cohen's d (*d*), Partial Eta squared (η^2_p), correlation coefficient (*r*):

[a] Cohen's d reported: *d*= 0.2 small effect size; *d*=0.5 medium effect size; *d*=0.8 large effect size.

[b] Partial Eta squared: η_p^2 =0.01 small effect size; η_p^2 =0.06 medium effect size; η_p^2 =0.14 large effect size.

[c] Correlation coefficient: *r*=0 no relationship; *r*=0.10 to 0.29 small relationship; *r*=0.30 to 0.49 medium relationship; *r*=0.50 to 1.0 large relationship.

Questionnaires used by the studies:

1 BDI = Beck Depression Inventory;

2 HAM-D = Hamilton Depression Rating Scale;

3 MADRS = Montgomery Asberg Depression Rating Scale;

4 HAM-A = Hamilton Anxiety rating Scale;

5 STAI = State-Trait Anxiety Inventory;

6 TAS = Toronto Alexithymia Scale;

7 PANAS = Positive and Negative Affective Scale;

8 MMQ= Multidimensional Mood Questionnaire;

9 SAM= Self-Assessment Manikin;

10 ERQ= Emotion Regulation Questionnaire.

2.6.2. Emotional regulation

Two studies examined the relationship between effective emotion regulation and heartbeat perception (Fustos et al, 2012; Pollatos et al, 2015).

Fustos et al (2012) investigated whether interoception was associated with a better ability to downregulate negative emotions. The study, with a sample of 28 participants, used pictures from the International Affective Picture System (Lang et al, 1997) as emotional stimuli. In order to judge emotional regulation, participants were asked to reappraise either neutral or unpleasant pictures, and to use cognitive re-appraisal techniques. They had been taught to down regulate their responses to a random selection of the unpleasant pictures. Reappraisal is considered useful to developing healthier responses to affect, and better well-being (John and Gross, 2004). The authors found the highest emotional responses were to the unpleasant pictures and the lowest to the neutral pictures. Cognitive reappraisal reduced negative emotional response to the unpleasant pictures with responses midpoint between the other two groups. The authors found that the perceived downregulation of arousal caused by cognitive reappraisal was significantly correlated with interoceptive awareness ($r=0.48$, $p<0.01$). The findings suggest that the more aware a person is of ongoing bodily processes, the more successful this person’s emotion regulation, in response to negative affect, will be.

Pollatos et al (2015) investigated the relationship between interoception and an individual’s ability to cope with social exclusion through effective regulation of emotion. From the regression analysis, they reported that interoception was significantly associated with emotion regulation ($R=0.38$, $p<0.001$). That is, more accurate interoception (i.e. higher interoceptive sensitivity) was reported to be associated with the emotion regulation strategies of reappraisal and suppression.

2.6.3. The relationship between interoception and affective states or alexithymia

Thirteen studies explored the relationship between interoception and depression, anxiety or alexithymia. These studies are described in sections 2.6.3.1 to 2.6.3.3.

2.6.3.1 Depression and interoception

Four studies investigated the association between interoception and depression (Herbert et al, 2011; Terhaar et al, 2012; Ricciardi et al, 2016 a&b). The questionnaires used to measure depression varied amongst the four studies. Two studies (Herbert et al, 2011; Terhaar et al,

2012) used the Beck Depression Inventory (BDI: Beck and Beck, 1972) which is a widely used self-report measure of depressive symptoms. Three studies chose to use clinician rated measures of depression: Ricciardi et al (2016a) used the Montgomery Asberg Depression Rating Scale (MADRS: Montgomery et al, 1979); and two papers (Terhaar et al, 2012; Ricciardi et al, 2016b) cited the Hamilton Depression Rating Scale (HAM-D: Hamilton, 1960). Terhaar et al (2012) was the only study to use both a self-rated (BDI) and the clinician rated (HAM-D) questionnaire.

Study populations varied. Ricciardi et al (2016a) studied patients with functional neurological disorders, Ricciardi et al (2016b) studied patients with Parkinson's disease, Terhaar et al (2012) studied patients with depression alone and Herbert et al (2011) used a healthy student sample.

All studies reported a negative correlation between interoception and depression or depressive symptoms, except for Ricciardi et al (2016b). That is, those participants with higher levels of depression showed poorer interoception.

Herbert et al (2011) reported a negative association between interoception and depressive symptoms (r=-0.21; p<0.05) in a healthy student sample. Ricciardi et al (2016a) found that patients with Functional Motor Disorder reported more depressive symptoms, i.e. scored higher on the depression rating scale than controls, however, they reported no significant correlation between interoception and depression, by individual group (i.e. patients or controls). When participants were combined (patients + controls) to increase statistical power, they reported a significant correlation between interoception and depression. Terhaar et al (2012) compared depressed and non-depressed participants on interoception and reported that patients demonstrated significantly reduced interoception (d=0.85; p=0.011).

The findings from these studies suggest that reduced interoception may be associated with depression, but the number of studies was small, and the study populations varied, which makes comparison difficult.

2.6.3.2. Anxiety and interoception

Six studies examined the relationship between interoception and anxiety (Schandry, 1981; Kindermann and Werner, 2014a; Mallorquí-Bagué et al, 2014; Ricciardi et al, 2016b; Garfinkel et al, 2016; Pollatos and Georgiou, 2016). The State-Trait Anxiety Inventory (STAI:

Spielberger et al, 1983) was used to measure anxiety in five studies and one study (Ricciardi et al, 2016b) used the Hamilton Anxiety rating Scale (HAM-A: Hamilton, 1960). The STAI is a self-report questionnaire that reports both state and trait anxiety levels.

As with depression, the populations of the six studies varied: Kindermann et al (2014a) and Schandry (1981) recruited healthy volunteers; Ricciardi et al (2016) patients with Parkinson's Disease; Mallorqui-Bagué et al (2014) healthy volunteers with joint hypermobility; Pollatos and Georgiou, 2016) bulimia nervosa; and Garfinkel et al (2016) autistic spectrum disorder.

Mallorquí-Bagué et al, 2014 investigated the association between anxiety, enhanced affective reactivity and interoception in people with joint hypermobility (N=36). They reported a small positive correlation between state anxiety and a better performance on the heartbeat perception (HBP) task i.e. interoception (r=0.284, p=0.046).

State anxiety was also found to be higher amongst 'good heartbeat perceivers' in healthy volunteers in the study by Schandry (1981). Respiration and skin conductance level was also measured in this study, and the authors reported that there was no difference in these parameters between good or poor 'heartbeat perceivers'. They concluded that the higher levels of reported anxiety by the 'good heartbeat perceivers' was explained by 'better perception of physiological processes' and not to higher levels of autonomic arousal. Pollatos et al (2007) also found elevated levels of anxiety were associated with better interoception (r=0.58; p<0.05) in patients with bulimia nervosa.

Kindermann et al (2014a) used the STAI to assess trait anxiety amongst healthy participants and found a small, non-significant effect between interoception and anxiety (r=0.15; p=0.35).

Garfinkel et al (2016) in a study investigating interoception, in individuals with Autistic Spectrum Disorder, found anxiety was associated with reduced interoception. They compared 20 participants with Autistic Spectrum Disorder (ASD) with 20 healthy controls, on their performance on two Heartbeat perception tasks: the mental tracking method (MTM) and the heartbeat discrimination task (HBD).

Participants with ASD had higher scores of state [t (34.35) = −4.81, p < 0.001; d = −1.52], and trait [t (34.2) = −5.00, p < 0.001; d = 1.57] anxiety, compared to healthy controls, and demonstrated significantly lower interoception compared to controls. Trait anxiety was found to be negatively associated with performance on the heartbeat perception (r=-0.47, *p*=0.02) but not significantly associated with performance on the MTM (*r*=-0.17, *p*=1.00); and state anxiety was not associated with performance on either task.

Overall, the findings of the above studies are mixed. There is some suggestion that in healthy participants, anxiety may be associated with more accurate interoception. However, the number of studies is small. Studies involving clinical samples do not report consistent findings in either direction for the association between anxiety and interoception. Again, the number of studies is small, and the clinical samples vary, making any comparison difficult.

2.6.3.3. Alexithymia and interoception

The relationship between interoception and alexithymia was explored by eight studies. Half of these studies investigated the relationship between interoception and alexithymia in non-clinical samples (Herbert et al, 2011; Bornemann et al, 2017; Murphy et al, 2018; Zamariola et al, 2018).

Alexithymia was measured using the self-report questionnaire the Toronto Alexithymia Scale (TAS-20: Bagby et al, 1994) in every study. The TAS is a measure commonly used to record levels of alexithymia within both patient and non-patient groups.

Non-clinical samples

Four studies investigated the association between interoception and alexithymia with 'healthy' participants (Zamariola et al, 2018; Herbert et al, 2011; Bornemann and Singer, 2017; Murphy et al, 2018). All these studies had larger study populations than the four studies involving clinical samples (see section 2.4.4.).

In the study by Zamariola et al (2018) the MTM and a variety of questionnaires (Body Awareness Questionnaire: Shields et al, 1989; Multidimensional Assessment of Interoceptive Awareness: Mehling et al, 2012; Interoceptive Awareness Questionnaire: Bogaerts et al, in preparation) were used to measure interoception. The TAS-20 was used to measure alexithymia.

This study had the largest sample (N=998) and this was achieved by combining 10 studies that were conducted by the authors.

The mental tracking method was used to test for interoceptive accuracy in six of their 10 studies. Interoceptive questionnaires were used to investigate interoceptive sensibility in nine studies. That is, the study aimed to test both objective (MTM) and subjective approaches (self-report questionnaires) to investigate the link between alexithymia and interoception.

The six studies that used the mental tracking method to investigate interoception were combined, by Zamariola et al (2018) into two groups. This gave sample sizes of 414 and 160. They reported no significant associations between interoceptive accuracy and alexithymia r= -0.02 and -0.03 with $p>0.05$.

In another study, Murphy and colleagues (2018) collected data over a period of two years to investigate the link between interoception and alexithymia. The investigators recruited 287 individuals but achieved this by including the same participants in more than one study, although duplicate values for participants who participated in more than one study were removed prior to analyses. The authors excluded 12 outliers based on various characteristics such as BMI and extreme depression scores from their main analyses.

The authors did not find a significant correlation between the heartbeat task and alexithymia (r (285) =-0.079, p=0.182) on univariate analyses. However, when all potential co-founders were controlled for in a series of regression analyses, there was a small but significant association between alexithymia and poor interoception (r= -0.192, p=0.021).

Herbert et al (2011) conducted the mental tracking task with 155 healthy participants who were asked to complete the TAS-20 and the depression questionnaire the BDI-2. They reported that over three quarters of the sample (76.7%) reported minimal depressive symptoms, and no participant scored high enough to indicate a depressive disorder. They also reported that the interoception scores and the TAS-20 scores were comparable to their previous study which also recruited healthy volunteers (Herbert et al, 2007). They reported a significant negative correlation between interoception and alexithymia (r= -0.37, $p<0.01$) on univariate analyses, and, when they conducted a regression analysis, controlling for depression, the correlation remained (F (2, 152) =22.96, p=0.0001, R^2=0.23).

In another study the effects of meditative training (breathing and attention focussing exercises) on heartbeat perception in healthy individuals was investigated by Bornemann and Singer (2017). Four groups of participants were included: three cohorts underwent different types of ‘contemplative mental practice training’ and a fourth cohort acted as a control group. In addition to interoceptive accuracy, alexithymia was also measured. They reported that at baseline – that is, before any training, there was no significant relationship between interoception and alexithymia across all groups (*r*=0.08, *p*=0.159). A significant negative correlation was reported between changes in interoception and changes in levels of alexithymia, between baseline and the first training module (*r*=-0.207, *p*=0.002).

Clinical samples

Demartini et al (2016) compared participants with psychogenic non-epileptic seizures (PNES; N=20) and functional motor symptoms (FMS; N=20) with healthy controls (N=20). The relationship between interoception and dissociation, alexithymia, depersonalization, depression and anxiety was explored to see if there were differences between the clinical and non-clinical population. The study found no significant differences between the groups with regards to interoception (FMS: 0.58; PNES: 0.59; controls: 0.63; *p*=.818); although the FMS and PNES groups were found to score higher on alexithymia items than the controls.

A study by Shah et al (2016) investigated interoception in individuals with Autistic Spectrum Disorder (ASD). The authors conducted two experiments: in the first study they established the relationship between interoception and alexithymia in healthy controls; and then compared participants with ASD to the healthy controls in the second study. In the first experiment they recruited participants without a diagnosis of ASD (N=38) and found that interoception and alexithymia were negatively correlated (*r*=-0.36, *p*=0.025).

Experiment 2 compared adults with ASD (N=19), to those without ASD (N=19), matched on age, gender, and IQ. No group differences for interoception (*d*=0.26, *p*=0.43) were found. As in experiment 1 with healthy controls, in the ASD group alexithymia was negatively correlated with interoception (*r*=-0.64, *p*<0.001).

In a regression analysis, alexithymia was the only factor independently associated with interoception (*β*=-0.56, *t*=-3.12, *p*=0.004) for participants with ASD. Shah et al (2016) concluded that it was alexithymia, and not autism, that was associated with deficits in interoception.

Two studies by Ricciardi et al (2016 a&b) reported no significant correlation between interoception and alexithymia. Ricciardi et al (2016a) conducted a study in which individuals with a functional motor disorder (FMD) were compared with healthy controls (HC), matched for age, gender and BMI (*Rho*=-0.30; *p*=0.28). Regression analyses, with depression controlled for, did not show interoception to be independently associated with alexithymia (*b*=10.50, *SE*= 22.18, *p*=0.636, 95% *CI* [-32.98, 53.98]). In the second study by Ricciardi et al

(2016b), they investigated interoception in those with a diagnosis of Parkinson's disease. This study also reported no association between interoception and alexithymia (*r*=0.05; *p* =0.8).

In summary, studies investigating the relationship between alexithymia and interoception present contrasting findings. Five report no significant associations between alexithymia and interoception (Demartini et al 2016; Murphy et al 2018; Zamariola et al, 2018; Ricciardi et al 2016 a & b), and three report a negative correlation between interoception and alexithymia (Herbert et al 2011; Shah et al, 2016; Bornemann and Singer, 2017).

There is, therefore, no compelling evidence that alexithymia is associated with interoception. All the studies differed in terms of study populations, sample sizes, and baseline cofounders that were controlled for in regression analyses. In addition, although the TAS-20 is widely used to measure levels of alexithymia, it requires the individual to reflect and report on their own styles of detecting/recognising emotion. Which, in the case of those populations suspected of having higher levels of alexithymia, may find such reflection problematic.

2.6.4. Emotional intensity

The association between interoception and the intensity of the emotional experience for the individual was assessed by six studies. The studies below either created their own measurement to express how emotion is experienced (Barrett et al, 2004), measured valence (Herbert et al, 2007; Wiens et al, 2000; Dunn et al, 2010); or the effect of stress on emotional experience (Kindermann and Werner, 2014 a&b). All of these studies were rated to be of 'weak' quality, with a high likelihood of selection bias with respect to the samples included in the studies.

Barrett et al (2004) chose to investigate the relationship between interoception and Arousal Focus (AF). AF is an emotional concept constructed by the authors, to report the activation and deactivation in experienced emotion.

Two studies were conducted using the heartbeat discrimination method. Participants were required to report emotional experiences over 28 and 60 day periods. Participants were asked to rate their emotional experiences throughout the day. The authors hypothesised that interoception would be positively associated with AF. That is, they expected those with greater interoception to report higher levels of emotional feeling (AF) in their reports of emotional experiences. Their findings suggested that interoception was significantly associated with AF (*r*=0.23, *p*<0.05). The authors stated that this indicates that those individuals better at discriminating when tones are coincident, or not, with their heartbeat, also experience more feelings of activation and deactivation (i.e. arousal) in the moment they have an emotional experience.

The aim of the study by Herbert et al (2007) was to investigate the association between interoception and the emotional experience related to the processing of emotional pictures. Participants were divided into good (N=17) and poor (N=20) heartbeat perceivers. Scores achieved above 0.85 were allocated to the good heartbeat perception group, and scores below this were considered to show poor heartbeat perception. The authors found that there was no significant effect with regards to the heartbeat perception group and valence (η^2_p =0.05, p>0.05). With regards to arousal ratings, group was associated with a significant effect (η^2_p = 0.83, p<0.05). Good heartbeat perceivers were found to demonstrate significant, albeit slightly higher arousal ratings, compared to poor heartbeat perceivers.

Two very similar studies were carried out by Kindermann and Werner (2014 a&b). In the first study, the authors were interested in the impact of heartbeat perception on emotional experience in the situation of mental stress (N=40). And their second study (N=50) explored whether participants with good heartbeat perception would report more negative emotions when experiencing mental stress. In the first study 20 participants with high heartbeat perception were recruited, and 20 participants with low heartbeat perception. Kindermann and Werner reported that stress did not have a significant effect on heartbeat perception on either good or poor heartbeat perceivers (η^2= 0.2, p=0.51). The experimental condition (stress or rest period) was reported to have a significant effect on reporting of negative emotions. Post hoc *t* tests revealed that more negative emotions were reported under the stress condition by participants with good interoception (M=15.11) compared to those with poor interoception (M=13.17), t (38) = 0.15, p=0.02, d=0.84. No significant differences were found in reporting of negative emotions during the rest period (d=0.05, p=0.88).

In the second study, when under stress, participants with good heartbeat perception reported lower mood scores (M=27.92), compared to participants with low heartbeat perception (M=30.79); but the effect of good/poor interoception on mood (good/bad) was not significant (η^2_p=0.05, p=0.11).

In summary, all the studies that explored the relationship between interoception and emotional experience using a heartbeat perception task were rated ‘weak’ based on their methodology. It is possible that those individuals with good heartbeat perception, i.e. good interoception, may also report higher levels of arousal or a more intense experience of emotion – when compared to those participants rated as having poor interoception. This would have to be confirmed, however, by studies with better methodology.

2.6.5 Summary of findings of associations between emotions and interoception

This review of studies investigating the relationship between interoception and emotional states, using a heartbeat perception task is unable to come to any clear conclusions. It is possible that individuals with higher levels of depression/depressive symptoms show poorer interoception than healthy controls, but there is insufficient evidence at present to substantiate this. Equally, although some studies have found that individuals with higher levels of state anxiety have better interoception than controls, the evidence is weak and equivocal. A similar, inconsistent picture emerged from the studies investigating interoception and alexithymia. Five reported no significant association and three studies reported significant findings. Most of the studies included in the review were under-powered and involved such diverse study populations that comparison between and across studies is difficult.

2.7. Heartbeat perception tasks

The second objective of this review was to identify which heartbeat perception tasks have been most frequently used as measures of interoception in the current literature.

2.7.1. Heartbeat perception paradigms

Schandry (1981) popularised using the mental tracking method (MTM) as a measure of self-perception of emotion. The MTM was adapted by Whitehead et al (1977) into the HBD by using tones as the stimulus of detection.

2.7.2. Mental tracking paradigm (MTM) vs signal detection (HBD)

Bornemann and Singer (2017) describe the heartbeat as a ‘discrete signal’ and the MTM as a ‘non-invasive’ task. When looking across all 24 studies, the MTM was found to be used as the interoceptive task, in the majority of studies (N=22; 92%). Included in these 22 studies, three studies used both the HBD and the MTM (Terhaar et al, 2012; Mallorquí-Bagué et al, 2014; Garfinkel et al, 2016).

Two studies used the HBD only (Wiens et al, 2000; Barrett et al, 2004).

2.7.3. Characteristics of heartbeat perception tasks

An attempt was made to highlight the characteristics of heartbeat perception tasks reported in the studies included in this review. However, based on the data available in the papers, this was limited to features such as how interoception was calculated (accuracy score or binary outcome: incorrect/correct or high/low accuracy); how reliable and valid the tasks are considered to be; and the interoceptive scores/outcome.

Accuracy scores are calculated by the difference in actual and reported heartbeats; or participants were rated with a binary outcome response: correct/incorrect (Kleckner et al, 2015). The latter is determined by using a cut-off score used to decide which scores fall within the correct range, with those below the cut-off falling into the incorrect range.

From the 24 studies in this review, mean heartbeat perception scores were reported in 12 studies only, i.e. 50%. From these 12 studies, the scores from 10 studies are reported here.

Two studies (Schandry, 1981; Murphy et al, 2018) were excluded because they reported heartbeat perception scores using a scale different to the majority of the studies.

Murphy et al (2018) calculated heartbeat perception scores on a scale of 0 to 400; where better heartbeat perception scores were based on achieving a high score. Mean heartbeat perception scores was reported as 192.77. Schandry (1981) calculated an error score and reported high perception accuracy as being closest to a score of 0; that is an error score of 0 would depict perfect heartbeat perception.

The mean heartbeat perception scores from the remaining 10 studies were calculated such that a score closest to 1 indicates high accuracy (see section 2.7.3.3.). From these studies the mean heartbeat perception score was 0.70, the median heartbeat perception scores score reported was 0.64 with a range 0.50 to 0.95. Those studies that were conducted with 'healthy' (or with a control group of) participants reported an average heartbeat perception score of 0.64 with a range of 0.54 to 0.72. Studies with patient samples reported an average heartbeat perception score of 0.58, ranging from 0.50 to 0.68. This suggests that healthy/control participants detected their heartbeats more accurately than patient participants; that is, controls showed better interoception than patients.

2.7.3.1 Reliability and validity of heartbeat perception tasks

Reliability

Reliability of a test refers to the test demonstrating that it consistently measures the same concept/factor under investigation – under different conditions (Heale et al, 2015). The mental tracking method has been shown to have good test-retest reliability (Bornemann and Singer al, 2017; Demartini et al, 2016).

The study by Bornemann and Singer (2017) was concerned with training effects on the MTM task. Within the study by Bornemann and Singer (2017), the control group was used to test the reliability of the task by performing the MTM at different time points. The test-retest reliability coefficients were reported as r=0.776, r=0.710, r=0.749, at a significance level of p<0.001 for all points.

Validity

The validity of a test refers to the ability of the test or tool to accurately measure that which it is designed to measure (Heale and Twycross, 2015). Therefore, in this review the validity of the heartbeat perception tasks refers to the ability of the task to report how accurately the individual perceived their heartbeats.

Studies have employed different methods to report the validity of the heartbeat perception tasks. In the studies by Garfinkel et al (2016) and Murphy et al (2018) participants were asked about their knowledge about the average resting heartrate. Shah et al (2016) chose to use a time estimation test and another study used heartbeat evoked potential (HEP) scores as an objective measure with which to test the MTM against (Terhaar et al, 2012). HEPs refer to sections of brain activity from an electroencephalogram (ECG) that are synchronised with heartbeats and are therefore considered to be a method by which to determine heartbeat activity during emotion (MacKinnon et al, 2013). That is, the change in HEP amplitude is interpreted as an indication of an increase/decrease in signals from the heart to the brain (McCraty, 2003). Terhaar et al (2012) reported that performance on the HEP was not correlated with performance of either the MTM or HBD. Therefore, HEPs could be considered as unbiased markers of interoception.

The time estimation task (Shah et al, 2016) has been used to demonstrate whether performance on the MTM is actually a reflection of the participant estimating how much time has elapsed during the counting period, rather than how many heartbeats they have been able to perceive. They compared the mean results on the MTM (*M*=69.12%, *SD*=19.78%) and the time estimation task (*M*=73.3%, *SD*= 22.81%), and concluded that the distributions indicated that the two tasks were matched in difficulty and therefore the time estimation task was appropriate to be used as a control measure. No correlation between the MTM and the time estimation task was found (*r* = -0.001). From this it could be inferred that participants were not guessing the number of heartbeats by counting the time passed. If participants were counting the seconds passed then it would be assumed that a strong correlation would have been reported, rather than the very week/negligible correlation found. Christensen et al (2018) used the MTM and reported a linear trend that showed that the shortest counting period had the highest level of accuracy. This could suggest that there may be some ceiling effects to the level of accuracy that can be reported using the MTM. Bornemann and Singer (2017) commented on this, suggesting that the MTM is more prone to ceiling effects.

2.7.3.2. Potential confounding variables effecting heartbeat accuracy

It has been suggested that heartbeat perception accuracy may be influenced by several factors such as age, sex and body mass (BMI). In a study with 318 participants (Bornemann and Singer, 2017), no association was reported between heartbeat perception accuracy and changes in heartrate (*r*=-0.250, *p*<0.001); nor age (*r*=-0.081, *p*=0.152). Heartbeat perception accuracy was noted to differ according to gender with male participants reporting more accurate scores (*M*=0.665, *SD*=0.238) compared to female participants (*M*=0.566, *SD*=0.256), *t*(316) =3.426, *p*<0.001.

2.7.3.3. High vs low heartbeat perception accuracy

Several studies divided participants into poor and good heartbeat perceivers, however the methodology used varied amongst studies. For example, in the study by Schandry (1981) participants were ranked as being good/poor based on reporting the number of counted beats being ± 2 from the actual number of heartbeats recorded. The study by Schandry calculated accuracy (reported as an error score) by subtracting the participant's reported heartbeats from the absolute difference between the actual number of heartbeats and the reported number of heartbeats, divided by the actual number of heartbeats: Σ (|actual number heartbeats – reported number heartbeats| / actual number heartbeats). Whereas, in both studies by Kindermann et al (2014 a&b), participants were classified as having good or poor perception using a cut-off of 0.85 when scores were measured between 0-1. Scores closest to 1 were considered as more accurate, i.e. a smaller difference between the perceived and recorded heartbeat. These studies calculated the accuracy score by taking the error score (as calculated by Schandry, 1981) and subtracting this from 1 e.g. 1/3 Σ (1 − (|recorded heartbeats − counted heartbeats|) / recorded heartbeats).

Schaefer et al (2014) also rated accuracy as being closest to 1 but chose to use a continuous scale rather than to divide participants into good/poor or high/low perceivers - illustrating that dichotomous and continuous scoring methods are used with the MTM. Garfinkel et al (2016) chose to calculate a ratio of the perceived heartbeats to the actual number of heartbeats recorded: 1 − |nbeatsreal−nbeatsreported|/ (nbeatsreal+ nbeatsreported)/2, the average of which was taken and reported as the perception score.

The calculation of the score was based on the difference between the heartbeats reported and the actual heartbeats that were recorded during the counting period. The equations used to calculate the heartbeat perception scores score varied between studies. No study reported replicating the equation used by Schandry (1981), which produced error, rather than accuracy scores.

2.7.4. Intra-individual correlation tasks

Intra-individual correlation tasks refer to a comparison of the individual’s performance on an HPT against a subjective measure of interoception (Ceunen et al, 2016), that is the individual’s judgement of their own performance. Intra-individual correlation tasks may also be referred to as metacognitive accuracy or interoceptive awareness (Garfinkel et al, 2016).

It has been suggested that these tasks report an accurate assessment of interoceptive ability (Murphy et al, 2018) or a reflection of one’s interoceptive awareness (Garfinkel et al, 2016). Such tasks usually comprise of either using a visual analogue scale and a heartbeat perception task. The visual analogue scale requires the participant to rate their confidence in their interoceptive accuracy using a scale for example rating from ‘total guess’ to ‘complete confidence’ (Garfinkel et al, 2016). These tasks and were included in a minority of studies (N=2: Garfinkel et al, 2016; Christensen et al, 2018).

Murphy et al (2018) chose to ask participants to report their knowledge of average resting heartrate. They claim this is more favourable than requesting participants to judge their own performance on the heartbeat perception task.

Garfinkel et al (2016) used a visual analogue scale with which participants rated the level of confidence in their performance on each heartbeat perception task, on a trial by trial basis to measure interoceptive awareness. Additionally, they included the awareness section of an interoceptive questionnaire (Porges Body Perception Questionnaire, 1993) as a subjective measure of interoception.

2.7.5 Section summary of study quality and heartbeat perception tasks

Assessment of study quality in this review found study quality to be poor with most of the studies rated as having a weak level of quality (N=16, table 5). Forty two percent (N=10) of studies reported recruiting participants from universities.

The heartbeat perception tasks used by the studies varied between the mental tracking task and the heartbeat discrimination task, with the mental tracking method being the task most used. Based on the data reported in the papers, mean heartbeat perception scores could be calculated on fewer than half of the studies (42%). That is, mean heartbeat perception scores were reported in 10 studies. Mean heartbeat perception scores were found to be higher in control participants, compared to patients – indicating that patients had poorer interoception in general.

It was reported that heartbeat perception tasks have good reliability and validity. Reliability was reported using test-retest coefficient and validity investigated using time estimation tasks, for example. The test-retest reliability coefficient reported correlations of r=0.776, r=0.710, r=0.749, at a significance level of p<0.001, on the mental tracking task measured at the different time points (Bornemann and Singer, 2017). The different time points showed comparable correlations, leading to the conclusion that the mental tracking task is a reliable measure of heartbeat perception. A time estimation task was used to investigate the validity of the mental tracking task. No correlation was found between the two tasks (r=-0.001), from which it could be inferred that participants were not counting the time passed in order to guess their heartbeats and therefore suggesting that the mental tracking task is a valid method of heartbeat perception. Although these studies suggest that heartbeat perception tasks are valid and reliable measures of interoception, this evaluation of heartbeat perception tasks was investigated by seven studies only. Furthermore, one study reported that participants demonstrated the highest level of heartbeat perception scores accuracy during the shortest counting period – which could be taken to indicate the mental tracking method is susceptible to ceiling effects.

Overall, the studies in this review reflect that methodology towards investigating interoception: MTM, HBD or intra-individual tasks; and calculation of heartbeat perception accuracy varies. Such differences make exact comparisons of accuracy, reliability and external validity of the task difficult to conclude from the studies in this review.

2.8. Discussion

There were three aims of this review. The first was to examine the association between interoception and emotion in studies which have used a heartbeat perception task to measure interoception. The second was to identify which is the most commonly used heartbeat perception task and the third, to identify which task is the most suitable for use with clinical populations.

There is great interest between the potential link between interoception and associated states, such as anxiety, depression and alexithymia. This review found that studies had investigated interoception and associations with emotion/emotional states in both normative and clinical populations. Although it is not known how representative the studies with normal samples are of the general population due to the paucity of the demographic data reported.

What is noted from this review is that anxiety and depression indicate a different relationship with interoception. In the literature higher levels of anxiety are associated with better interoception (Domeschke et al, 2010) and higher levels of depression are associated with poorer interoception (Murphy et al, 2017). However this generalisation was not upheld by the studies in this review.

Studies in this review presented a mixed picture with some studies reporting a significant association between interoception and anxiety (Schandry, 1981; Pollatos et al, 2007; Mallorquí-Bagué et al, 2014), and five studies reporting non-significant correlations between anxiety and interoception. Therefore the studies in this review do not support the view that individuals with higher levels of anxiety will perform better on interoceptive tasks. And although the differences in study samples (clinical v normal) may lead to the assumption that there is another factor that has a role, this cannot be inferred from the studies in this review.

Similar results were found for the studies investigating depression and interoception, although more studies reported a significant association between depression and interoception (Herbert et al, 2011; Terhaar et al, 2012; Ricciardi et al, 2016a); than a non-significant relation between the two variables (Ricciardi et al, 2016b).

Two studies were found to report a negative relationship between interoception and both sub-clinical depression (Herbert et al, 2011) and major depression (Terhaar et al, 2012). That is experiencing higher levels of depressive symptoms is associated with a poorer perception of one’s internal bodily activity. These studies appear to support the conclusion by Ricciardi et al (2016a) that poor interoception may indicate problems with emotional processing.

Terhaar et al (2012) offer explanations as to why a correlation is found between depression and reduced interoception: it could relate to an impaired awareness of one’s body which ‘leads to mental exhaustion and reactive depression’ (Herbert et al, 2007b), or it could relate to alexithymia (Herbert et al, 2007a, 2010). However, such conclusions cannot be established with confidence when sample sizes are small, and the results are not replicated by other studies.

With regards to alexithymia and interoception, it has been suggested that atypical interoception may be characterised by alexithymia (Murphey et al, 2017). However, the findings from the studies in this review did not support this, with only three studies reporting a significant association between alexithymia and interoception (Herbert et al, 2011; Shah et al, 2016; Bornemann and Singer, 2017), and five studies reporting a non-significant finding (Ricciardi et al, 2016a&b; Murphy et al, 2018; Zamariola, 2018).

Consequently, the findings from this review suggest that an inconsistent relationship between interoception and alexithymia is found in the literature. This also raises the question of measurement validity. Although the TAS-20 is widely used to measure levels of alexithymia, it requires the individual to reflect and report on their own styles of detecting/recognising emotion. This may be problematic for individuals from populations suspected of having higher levels of alexithymia.

In this review, studies investigated interoception in both normal and clinical populations. As has been noted earlier, studies with normal samples were found to rely on student participants, raising the question of how representative such study samples are. When clinical samples are considered, there are similarly additional factors to consider with regards to how individuals may perform of interoceptive tasks.

Ricciardi et al (2016a) suggest that maybe individuals with FND pay more attention to external bodily experiences/symptoms because of a difficulty with being able to detect internal bodily states. This may explain why a non-significant relationship was reported.

Overall, the findings in this review reported that studies in general were very heterogeneous in nature: differing in focus and investigating different populations. Furthermore, most studies were rated to be of a poor quality, so the findings from the studies need to be handled with some caution. Therefore making generalisations from these studies is made difficult, since no studies were convincingly robust in their design.

In this review, the mental tracking method (MTM) was found to be the most frequently used measure to assess interoception. This may be because the MTM is easy to use, requires minimal training on the part of the investigator, and in fact from the perspective of the participant, is simple to explain since everyone has perceived their heartbeat at some point such as when feeling anxious or after running. It has also been commented that performance on the MTM may be considered as reporting a 'pure' form of interoception since it does not rely on exteroceptive signals (Garfinkel et al, 2016). That is when compared to tasks such as the HBD, which require the individual to focus on both internal activity and external stimuli.

However, whilst the mental tracking method and the heartbeat discrimination task have been found to be the most common methods of measuring heartbeat perception, it could be that the tasks vary in how accurate they measure interoception. For example, Wittkamp et al (2018) suggest that performance on the tasks may be influenced by psychological processes. Stress has been found to improve accuracy on the MTM and decrease accuracy on the HBD. From this, it could be inferred that stress may cause the individual to be more aware of their heartbeat, and less attentive to external stimuli.

Although the heartbeat perception task is widely used, Murphy et al (2018) have been critical of its utility as a measure for investigating interoception. They suggest that whilst it is quick and easy to use, it has a number of fundamental problems such as the potential for other influencing factors to affect the reported outcome, such as age or stress (Khalsa et al, 2009) or discrepancies in the way the task is administered.

With regards to the studies in this review, although the MTM was most commonly used to investigate interoception, the number of counting periods varied across studies. It is feasible that those studies using more counting periods – or in the case of the HBD, more tone detection, may have allowed practice effects to influence accuracy scores.

Khalsa and colleagues (2009) comment that interpretation of correlations made with heartbeat perception tasks may be difficult if confounding factors, such as fatigue, are not considered. They mention that the use of time estimation tasks can act as a control task and may be particularly useful with participants who have ASD or populations high in alexithymia (Shah et al, 2016). They further state that even after confounding factors are controlled for, the heartbeat perception task may not be a good measure of interoception, as 40% of individuals report no heartbeat awareness (Khalsa et al, 2009). A measure to counteract influencing factors is to collect from participants what their knowledge of their own heartrate is. This method was utilised by three studies in this review (Garfinkel et al, 2016; Murphy et al, 2018; and Pick et al, 2020).

Schandry (1981) on the other hand argues that if the MTM was a poor measure of interoception and reported participants counting/guessing their correct heartbeat rhythm, then scores would be consistent across all the counting periods. In order to validate this claim, in his study a correlation between standard deviations of heartrate was conducted against standard deviations of the scores from the good heartbeat perceivers. It was reported that a small, positive but non-significant association was found. This was taken to suggest that participants were not estimating their heartrate. Other studies have employed the use of a time estimation task to similarly validate the findings when using a heartbeat perception task (Shah et al, 2016)

Wiens et al (2000) suggest that HBD have been developed to provide a valid method because of the methodological problems with the MTM – such as guessing heartbeats based on prior knowledge of the average heartrate. Although as mentioned earlier, it has been indicated that both interoceptive and exteroceptive cues are required in the HBD (Couto et al, 2015).

Ceunen et al (2013) argue that caution should be applied in taking performance on one interoceptive task as evidence of interoceptive ability overall. They suggest it could be considered that those studies focussing on one aspect of interoception, such as heartbeat perception, provide ‘specific information on a subcomponent of interoception’. This can then be contrasted against other interoceptive tasks in order to be able to make generalised conclusions.

Strengths and limitations

A strength of this review is that it included all studies in which an objective measure of heartbeat perception has been used to assess interoception. The mental tracking method was found to be the most frequent measure employed probably because of its ease of use.

A limitation of this review is that all studies on clinical samples used the mental tracking method as the paradigm to investigate heartbeat perception. Therefore, it was not possible to make comparisons with other heartbeat perception tasks and establish whether one method would be more accurate than the other.

From the studies in this review, it has not been possible to establish clear relationships between interoception and emotion related variables such as depression, anxiety, and alexithymia. A major limitation of this review is that most studies included in the review were of poor quality. Selection was a major problem.

Under half of the studies (N=13; 43%) were concerned with normal populations, i.e. non-clinical populations. Most of these studies (N=10) recruited university students, who may not be representative of the general population. Age has been identified within the literature as a potentially influencing factor on interoception. That is older age groups are more accurate in estimating their heartbeat due to experience. Those studies investigating interoception in patient populations had participants with higher mean ages than non-clinical populations. This would suggest that these samples would perform more accurately, if age influences performances.

Sample size is another area which could have potential consequences for the results reported by the studies in this review. A high proportion of the studies had a small sample size suggesting that these studies may be in fact underpowered, and none of the studies reported a power calculation. Larger sample sizes would increase confidence in the conclusions formed from the relationships reported by studies.

Additionally, the interoceptive dimension under investigation varied across studies, as did the method by which accuracy was calculated. Meaning that comparisons across studies were not equal.

Recommendations

Based on the findings of this review, there are still deficits within the literature with regards to interoception and self-perception of emotion. Further studies, with better methodology need to be carried out to determine whether there is a relationship between interoception as measured by the heartbeat perception task and emotion. That is, standardising the definitions and names used for interoceptive domains investigated. This would reduce the use of different terminology being used across studies to investigate the same aspect of interoception and provide clarity of which aspect of interoception is being investigated.

Studies should avoid using exclusively undergraduate student populations, control for potential biases including age and BMI, and include populations that are representative of the patient group or population under study. This diversity of study populations needs to extend to ethnicity also. The studies in this review did not report ethnic backgrounds and therefore it can be assumed that participants were white. The failure of studies to comment on this as a limitation indicates that researchers either consider their studies to be representative of the general population, or that it is a factor that is not considered important.

More studies on clinical populations would be helpful so that clearer conclusions can be formed. That is, there needs to be more studies investigating interoception in FND populations, and with larger samples. This is so that it can be established whether interoceptive deficits are found in those with specific FND conditions, for example less prominent in movement disorders, and perhaps more present in those with non-epileptic seizures.

If one paradigm to investigate heartbeat perception was agreed upon, this would improve the accuracy of comparisons across studies. The MTM is non-invasive, easy for the researcher and participant to learn, and focusses on internal activity. It is potentially a more precise measure of heartbeat perception, compared to the HBD.

Finally, better powered studies are required. This review highlighted that many studies were underpowered and therefore susceptible to Type 1 errors.

2.9. Conclusion

This review highlights that interoception and its association with emotion perception appears to be a growing area of research, particularly in some conditions such as FND. However, there needs to be some areas of agreement with regards to what cardiac interoception reflects and the tasks used to investigate this. There is an indication that efforts are being made to offer clarity on interoceptive dimensions and how to investigate these (e.g. Garfinkel et al, 2016).

The merits of using the MTM have been highlighted and Daubenmier et al (2013) state that heartbeat perception tasks have come to be considered as the “gold standard” with regards to investigating interoception. This may indicate why there appears to be an overreliance on this paradigm in the literature.

Following on from this review, it was planned that a study with both healthy controls and participants with non-epileptic seizures would be conducted. This study had to been abandoned in the light of the COVID situation, when face to face research no longer became viable. Therefore another study was designed and is discussed in chapter 3. The circumstances therefore required an update to be made to the literature. In doing so, several new studies were found that had been published since the literature search was conducted. These studies are presented in the section below.

2.10. Recent studies with FND populations

Due to the consequences of the COVID pandemic, the completion of the PhD was delayed, and an entirely new study had to be designed to replace a study which had to be abandoned before completion, as it involved face to face research. The new study involves participants with non-epileptic seizures, a form of functional neurological disorder. The systematic review was, therefore, updated in July 2021 with a focus upon studies involving functional neurological disorder and these new studies are included in an extra section below.

Functional Neurological Disorder is an umbrella term that encompasses several diagnoses. For example, FND includes those with functional movement/motor disorder, functional/

dissociative/ non-epileptic seizures, functional cognitive disorder, functional speech disorder (Hallett et al 2022).

2.10.1. Study characteristics with FND

Six studies were identified as investigating interoception with FND populations. The overall sample size from all six studies was N=265. The study with the largest sample size was 71 (Koreki et al, 2020) and the smallest was 16 participants (Demartini et al, 2019).

See table 7 below for details of these six studies. The table reports the study characteristics such as study design and method of investigating interoception - i.e. mental tracking method or heartbeat discrimination task. Sample characteristics such as sample size, the numbers of female/male participants, and age are also reported. The table also reports the overall quality rating of the study.

2.10.2. Age of participants with FND

All studies reported age by group: patients and controls. Based on the five studies that reported the mean, the mean age of participants with FND was found to be 39.9 years. One study reported a range of 24 to 62 years for the patient sample (Williams et al, 2021). The mean age of control participants was 36.9 years. Pick et al (2020) reported the median age and so the participants from their study are not included here.

2.10.3. Sex reported in studies with FND

One hundred and eighty-eight participants were female (71%). This is a slightly higher proportion than the proportion of women participants in the original review (see table 2). One study did not report the sex of control participants (Jungilligens et al, 2020).

Table 7. Study characteristics of recent studies with FND populations investigating interoception

Study	Design	Sample size (F/M)	Age (range/ mean)	BMI measured	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable/ questionnaires	Study quality rating
Demartini et al (2019)	Cross sectional	16 (14/2)	Patients: 48.22 Controls: 44.86	N	Sensitivity	Mental tracking method	N	Alexithymia: TAS[2] Depression: HAM-D[5] Anxiety: HAM-A[6]	MODERATE
Aim		To explore whether several aspects of perception and attention would show effects after a session of Transcranial direct-current stimulation (tDCS).							
Relevant findings		Patients demonstrated poorer interoception compared to control participants.							
Jungilligens et al (2020)	Cross sectional	40 Patients: (15/5) Controls: (not reported)	Patients: 32.9 Controls: 29.4	Y	Sensitivity	Mental tracking method	N	Depression: BDI[3] Anxiety: BAI[4] Alexithymia: TAS[2] ERQ[7]	MODERATE
Aim		Investigated emotion recognition, regulation, inhibition and whether associated with impaired interoception, and individual's perception of interoceptive abilities.							
Relevant findings		No significant differences between the patients and control groups on interoceptive scores							

Study	Design	Sample size (F/M)	Age (range/ mean)	BMI measured	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable/ questionnaires	Study quality rating
Koreki et al (2020)	Cross-sectional	71 (66/5)	Patients: 32 Controls: 32	N	Accuracy & sensibility	Mental tracking & signal detection	Y	Depression: BDI[3] Anxiety: STAI[1]	MODERATE
Aim	To investigate whether interoceptive accuracy would be impaired in individuals with functional seizures, linked to dissociation at trait level and seizure frequency.								
Relevant findings	Patients significantly lower on heartbeat accuracy, compared to controls.								
Pick et al (2020)	Cross-sectional	39 (31/8)	Patients: 44 Controls: 27 median	Y	Accuracy, awareness & sensibility	Mental tracking method	Y	Depression: PHQ-9[11] Anxiety: GAD-7[12] MAIA[8]	MODERATE
Aim	To investigate whether dissociation has an impact of interoceptive processing.								
Relevant findings	Patients showed lower interoceptive accuracy after dissociation task, suggesting dissociation affects interoceptive ability.								

Study	Design	Sample size (F/M)	Age (range/ mean)	BMI measured	Interoceptive dimension measured	Mental tracking method or Signal detection task	Interoception questionnaire	Emotional variable/ questionnaires	Study quality rating
Ricciardi et al (2021)	Cross sectional	46 (33/12)	Patients: 44.8 Control: 44.8	Y	Accuracy Sensibility Metacognitive interoceptive awareness	Mental tracking	Y	Alexithymia: TAS[2] Anxiety: HADS-A[9] Depression: HADS-D[10]	WEAK
Aim		Investigated whether interoceptive processing differs in individuals with and without FND							
Relevant findings		Patients demonstrated lower interoceptive accuracy and sensibility compared to controls.							
Williams et al (2021)	Cross sectional	53 (44/9)	Patients: 24-62; 41.6 Controls: 19-58; 33.3	N	Sensitivity	Mental tracking	N	Depression: PHQ-9[11] Anxiety: GAD-7[12]	STRONG
Aim		Investigated whether stress would have an effect on interoception in participants with FND.							
Relevant findings		Patients showed significantly lower interoception, compared to controls, when depressive symptoms and age controlled for.							

Questionnaires used by the studies: 1 STAI = State-Trait Anxiety Inventory; 2 TAS = Toronto Alexithymia Scale; 3 BDI = Beck Depression Inventory; 4 BAI = Beck Anxiety Inventory; 5 HAM-D = Hamilton Rating Scale Depression; 6 HAM-A = Hamilton Rating Scale Anxiety; 7 ERQ= Emotion Regulation Questionnaire; 8 MAIA = Multidimensional Assessment of Interoceptive Awareness; 9 HADS-A Hospital Anxiety and Depression Scale - Anxiety; 10 HADS-D Hospital Anxiety and Depression Scale - Depression; 11 PHQ-9 = Patient Health Questionnaire; 12 GAD-7 = General Anxiety Disorder Assessment.

2.10.4. Study location in studies with FND

Four studies were conducted in England; one study was conducted in Italy (Demartini et al, 2019) and one study was carried out in Germany (Jungilligens et al, 2020).

2.10.5. Assessment of study quality of studies with FND

Studies were rated using the same assessment tool mentioned is section 5 (The Quality Assessment Tool for Quantitative Studies: Effective Public Health Practice Project, 1998). Four studies were rated to be of a ‘moderate’ quality overall, and one study achieved a ‘weak’ rating (Ricciardi et al, 2021). One study achieved a ‘strong’ rating (Williams et al, 2021).

See Table 18 (Appendix U) for assessment of risk of bias and study quality ratings achieved.

Selection bias

Four studies were rated as ‘moderate’ with regards to how participants were recruited. Two studies were rated as ‘poor’ on selection bias (Pick et al, 2020; Ricciardi et al, 2021). These two studies were rated as ‘poor’ because patients were recruited via advertisements (Pick et al, 2020), or it was not described (Ricciardi et al, 2021). Based on the Quality Assessment Tool for Quantitative Studies, participants that self-refer are less likely to reflect being representative of the ‘target population’.

No study achieved a ‘good’ rating.

Study design

All six studies employed a cross-sectional study design and used both patient and healthy control participants. Participants were required to participate on one occasion only in all the studies.

Data collection

All studies in this section used standardised questionnaire measures. More details are available in table 7.

Neuropsychological tasks were used by two studies (Jungilligens et al, 2020; Ricciardi et al, 2021). Jungilligens et al (2020) used an emotional go/no go task. The study by Ricciardi et al (2021) used the Stroop task to assess executive functioning.

The Cold Presser Test is a physiological task used as a physical stress induction task by Williams et al (2021).

Confounding variables

Four studies matched patients and healthy controls on age and sex. The study by Pick et al (2020) also matched participants on IQ and their own perception of their knowledge of their heartbeat.

Two studies (Jungilligens et al, 2020; Williams et al, 2021) did not match participants on characteristics. Although in the statistical analyses conducted, Jungilligens et al (2020) controlled for BMI and blood pressure, and Williams et al (2021) controlled for age and depressive symptoms.

All six studies were rated as ‘moderate’ with regards to how confounding variables were managed.

2.11. The relationship between interoception and emotional states/conditions.

2.11.1. Emotion

All six studies in this section used questionnaires that either measured levels of psychopathology (anxiety and depression), or emotion recognition (alexithymia). The different questionnaires used by studies are noted in table 7.

2.11.2. Depression and interoception

All six studies asked participants to complete a depression measure. The questionnaires varied between the studies: one study used the Hamilton Rating Scale for Depression (Demartini et al, 2019); two studies used the Beck Depression Inventory (Jungilligens et al, 2020; Koreki et al, 2020); two studies used the Patient Health Questionnaire (Pick et al, 2020; Williams et al, 2021); one study used the Hospital Anxiety and Depression Scale (Ricciardi et al, 2021).

Only one study (Ricciardi et al, 2021) reported investigating whether a correlation between interoception and depression would be found. Ricciardi et al (2021) reported that they did not find a significant relationship between interoception and depression (ρ = 0.211, p=0.385). The other five studies measured depression to record the levels of psychopathology present in the samples.

2.11.3. Anxiety and interoception

Two studies used the General Anxiety Disorder Assessment to measure anxiety symptoms in participants (Pick et al, 2020; Williams et al, 2021). Other studies used a variety of different questionnaires to measure anxiety (see table 7).

The study by Ricciardi et al (2021) examined the correlation between interoception and anxiety. They reported that there was no significant correlation between interoception and anxiety for the patients with functional disorders (p=0.012, p=0.96), or across the whole sample of participants (p= -0.054, p=0.736).

None of the other five studies explored the relationship between interoception and anxiety.

2.11.4. Alexithymia and interoception

Three studies measured alexithymia (Demartini et al, 2019; Jungilligens et al, 2020; Ricciardi et al, 2021). They all used the Toronto Alexithymia Scale (TAS-20: Bagby et al, 1994).

Demartini et al (2019) used Transcranial direct-current stimulation (tDCS) with both patients and healthy controls. The tDCS technique is designed to administer a weak electrical current to the brain, using a non-invasive method. After receiving one session of tDCS, they found a negative correlation between interoception and alexithymia scores for both patient and healthy control participants (p=0.024; p= -0 .597). This correlation was significant for the FND participants only.

Ricciardi et al (2021) investigated interoceptive processing in those with and without Functional Motor Disorder (FMD). They reported that there was no significant correlation between alexithymia scores and interoceptive accuracy for the FMD participants (ρ = 0.142, p=0.561), or for the whole sample (ρ = 0.054, p=0.734).

2.11.5. Heartbeat perception

All six studies used the mental tracking method to investigate interoception. The study by Koreki et al (2020) additionally used a heartbeat discrimination task (signal detection) and an interoception questionnaire also. The studies by Pick et al (2020) and Ricciardi et al (2021) included interoception questionnaires too.

2.11.5.1. Interoception and FND

Studies in this section were interested in the interplay between interoception and FND. Five studies reported that patient participants, i.e. those with FND demonstrated lower or poorer interoception compared to healthy controls. The only study that did not report this relationship was Jungilligens et al (2020).

Demartini et al (2019) reported that compared to controls, patients demonstrated poorer interoceptive sensitivity [$F[1, 12] = 15.875, p=0.002$)]. Although in an earlier study (Demartini et al, 2016) they had reported that no differences in interoception were found between groups on interoception (see section 2.6.3.3.).

Koreki et al (2020) used both heartbeat perception paradigms: the mental tracking method and heartbeat discrimination. On the mental tracking task, patients showed significantly lower heartbeat accuracy compared to controls ($P< 0.001$, $d=1.12$). On the heartbeat discrimination task, patients showed impaired interoception also, compared to controls, but it was not a significant result (FND= 0.52 correct; controls= 0.56 correct).

Pick et al (2020) used both the mental tracking method and the Multidimensional Assessment of Interoceptive Awareness questionnaire (MAIA: Mehling et al, 2018) to investigate interoception. The mental tracking task was used as a measure of interoceptive accuracy and the MAIA of interoceptive awareness. A mirror gazing task was used to induce a dissociative state in both FND and control participants. At baseline, there was no significant difference between the groups on interoceptive accuracy ($p=0.967$, $r=0.009$). However, after the dissociation task, the FND group demonstrated poorer interoceptive accuracy compared to controls ($p=0.021$, $r=0.379$).

Ricciardi et al (2021) reported that participants with FND showed poorer interoception compared to controls (p=0.03). They reported that interoceptive accuracy was positively correlated with Body Awareness on the 'predict body reaction' subscale, as measured by the Body Awareness Questionnaire (Porges, 1993). This was reported for the whole group (p=-0.51, p=0.004). Interestingly, in the FND group, Ricciardi et al (2021) reported that severity of symptoms was found to be correlated with interoceptive awareness (p=0.888, p<0.001).

Williams et al (2021) used a stress induction task (Cold Pressor Test) to investigate the differences before and after physiological stress on interoceptive ability in those with and without FND. In this study, when patients were compared to controls, patients were reported to demonstrate significantly lower interoception, when depression and age were controlled for [F(1, 48) = 5.42, p= .024, η_p^2= .10]. After the stress task, a significant effect on interoception was reported for all participants [F(1, 48) = 8.91, p= .004, η_p^2= .16]. This shows that interoceptive accuracy improved following the stress test for all participants. When these scores were examined by group, the improvement in interoception was found for those with FND, and not the controls – although it was a non-significant effect [F(1, 48) = 1.11, p= .30, η_p^2= .02].

Williams et al (2021) suggest that since the differences in interoceptive ability were found after controlling for depressive symptoms, anxiety, and age, and those with FND were found to show interoceptive deficits, this suggests that those variables do not explain the interoceptive impairments in participants with FND.

2.11.5.2. Beliefs about interoceptive accuracy

In addition to using the mental tracking method to measure interoceptive accuracy, Koreki et al (2020) investigated beliefs about interoceptive accuracy using the Body Perception Questionnaire (BPQ: Porges, 1993). This questionnaire is focussed on how often the individual perceives bodily sensations. The results from the BPQ were found to be significantly higher for participants with FND (P<0.001, d=-1.14). That is, the patient participants indicated a discrepancy between the beliefs they had about their interoceptive perception and the interoceptive accuracy they demonstrated.

Pick et al (2020) reported that participants with FND rated themselves as having lower confidence in their interoceptive ability, compared to controls. Pick suggests that this could indicate a weakened metacognition.

The study by Ricciardi et al (2021) however, reported that metacognitive awareness was not significantly different between the FND and control groups (*p*=0.2). That is, patients did not show a significant difference between their reported interoceptive accuracy versus how accurate they judged themselves to be in perceiving their heartbeat. This is different to that which was reported by Koreki et al (2020) and Pick et al (2020).

2.11.6. Section summary

These studies were added to this review because, although not available when the original search was conducted in 2018, they have since been published and are relevant to this PhD.

The studies in this section generally replicate the findings reported in the main review.

The results from the six studies in this section provide some support that that interoceptive deficits are found in clinical populations, here specifically FND samples. But all six studies in are cross-sectional studies, with small sample sizes and subject to bias. Also, their design means it is not possible to comment on causality. Therefore, it cannot be assumed that interoception is an impairment that contributes to the aetiology of disorders such as FND, because it could be that it is deficit as a result of the disorder (Williams et al, 2021).

The Quality Assessment Tool for Quantitative Studies (Effective Public Health Practice Project, 1998) was used to rate the quality of the studies included in this review. The dictionary that accompanies the tool was used to decide the level to which studies met the criteria for ‘poor’, ‘moderate’ or ‘weak’ across each of the criteria used to establish study quality. It is possible that by the strict adherence to the tool and the criteria set out in the dictionary, this led to some studies being rated more harshly because studies were rated on the judgement of one researcher. Using a second rater may have allowed discussion around whether criteria needed to be used with some flexibility. Had this approach been adapted some studies may possibly have achieved better overall ratings.

The mental tracking method remains to be the most commonly used paradigm to investigate interoception and was used in all six of the studies in the section above.

CHAPTER 3: STUDY 2

Investigating self-perception of emotion in individuals with non-epileptic seizures: using self-report measures and an emotional stroop task.

3.1 Introduction

It has been hypothesised that difficulties with processing emotion may be involved in the pathogenesis and maintenance of non-epileptic seizures (Brown et al, 2016). Aspects of emotional processing include interoception, alexithymia and executive functioning /attentional bias. The findings of the systematic review described in Chapter 2 suggest that there is insufficient evidence at present to support a clear association between interoceptive accuracy, measured by heartbeat perception tasks, and other commonly studied emotional factors in people with NES (pwNES). One of the main reasons for a lack of compelling evidence may be related to methodological shortcomings in studies which have been conducted to date in the field. Better understanding of the relationship between these factors in pwNES in comparison with controls may help understand potential mechanisms involved in the development and maintenance of seizures.

Study 2 was designed to investigate the association between interoception, alexithymia and executive functioning in patients with NES, and to address some of the methodological concerns identified in the studies included in the systematic review (Study 1). The original study 2 employed a recognised, independent measure of heartbeat perception and standardised questionnaires to measure emotional symptoms (including alexithymia); was fully powered and intended to recruit a clinical population of NES patients (with a confirmed diagnosis of NES). It also included an emotional Stroop (eStroop) task to investigate executive functioning/attentional bias.

Unfortunately, the study was halted in March 2020 as the country went into national lockdown because of the COVID-19 pandemic. At this point in time, the study had received ethical approval and an adequate number of control participants had been recruited, but patient recruitment had only been open for a few months. The intention had been to recruit participants with NES from outpatient neurology clinics and more widely from neuropsychology and liaison psychiatry. All NHS services were severely affected by the pandemic and although they re-opened in autumn 2020, the throughput of patients was diminished, and some services operated remotely. As one of the study tasks involved measurement of heartbeat perception, this necessitated patients to consent to attend in person and the researcher to wear full PPE. After lengthy discussions and attempts to try to open recruitment post October 2020, a decision was made to abandon the study. A new 'COVID'-proof study, that could be conducted completely online, had to be designed and executed and the methods for this new study are described below. The aim of this new study 2 was to investigate the association between interoception, alexithymia and executive functioning, the same concepts that were the focus of the abandoned study, but modifications to the design were undertaken to ensure that all aspects of the study could be conducted remotely. The study and the relevant literature are detailed below.

A proposed model of dysfunctional emotion processing in non-epileptic seizures

A common feature of models of NES is the hypothesis that dysfunctional emotional processing likely plays a role in the aetiology and maintenance of the seizures. In the literature there is an emerging area of interest focussing on the role of interoception in emotion processing and the importance interoception may play in the aetiology and maintenance of seizures in pwNES.

Interoception refers to the process that occurs when subtle internal bodily signals and activity are detected and interpreted by the body (Dunn et al, 2010; Khalsa et al, 2019). It is postulated that interoception is linked to emotional processing and decision making (Critchley and Harrison, 2013; Seth, 2013; Khalsa et al, 2018). A common method to investigate this psychophysiological process is to use a heartbeat perception task which measures interoceptive accuracy. Questionnaire measures may also be used instead, or alongside a heartbeat perception task, to measure interoceptive sensibility- the person's perception of their awareness of internal bodily function. A subset of the studies in the systematic review (Study 1) explored interoceptive ability in participants with functional neurological disorder (FND) (Jungilligens et al, 2020; Ricciardi et al, 2021; Koreki et al, 2020; Pick et al, 2020; Demartini et al, 2019; Williams et al, 2021). Results from these studies were mixed. Some studies reported that patients with FND had poorer interoceptive accuracy when compared to controls (Demartini et al, 2019; Koreki et al, 2020; Pick et al, 2020; Ricciardi et al, 2021; Williams et al, 2021; and others reported no difference (Jungilligens et al 2020). No studies reported finding better interoception in FND participants compared to controls.

Alexithymia has been described as a trait relating to an inability to recognise or express emotion (Calsius et al, 2016; Uliaszek et al, 2012). It is hypothesised that this can result in psychological distress with emotions being expressed in the form of physical symptoms (Taylor et al, 1997). Alexithymia is commonly measured using a self-report questionnaire, the Toronto Alexithymia Scale (TAS-20; Bagby et al, 1994), and this measure has been employed to measure alexithymia in many studies of clinical populations, including those with non-epileptic seizures (Bewley et al, 2005; Tojek et al, 2000; Kaplan et al, 2013; Myers et al, 2013).

Alexithymia has been associated with dissociation (Mason et al, 2005), and has been relatively frequently investigated in NES and FND studies. Studies have reported that alexithymia is higher in those with FND, when compared with controls (Urbanek et al, 2014), or when compared with people with mild-Major Depressive Disorder (mild-MDD; Martino et al, 2018). Although, other studies have not demonstrated higher levels of alexithymia in patients with NES in comparison with those with epilepsy (Tojek et al, 2000; Myers et al, 2013).

Executive functioning is used to refer to several processes, such as the ability to plan ahead and meet goals, display self-control, follow multiple step directions and stay focused despite distractions (Anderson et al, 2002) and is thought to be involved in decision making (Dunn et al, 2006). It is suggested (Viviani et al, 2021) that executive functioning relates to the ‘higher-order cognitive processes’ that are involved in regulating and managing ‘lower-level cognitive operations, especially in novel or complex circumstances’ (MacPherson et al, 2019). That is, the attention allocated to stimuli, particularly in new or unusual situations, and the resulting behaviour (Anderson et al, 2002).

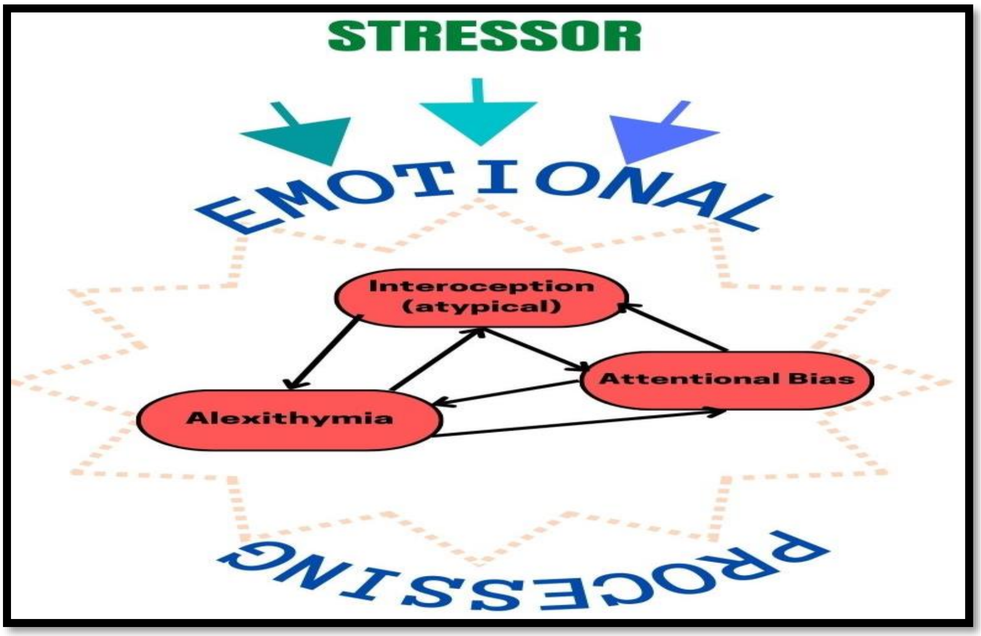
Problems with attention, working memory and executive functioning have been reported in a substantial proportion of patients with NES (Strutt et al, 2011). The Stroop task is one of the most common ways of evaluating selective attention, cognitive flexibility and executive functioning (Stroop, 1935). The original task requires participants to respond to words presented in coloured text. In some instances, the ink colour and word meaning are the same (congruent), and in some instances the ink colour and word meaning do not match (incongruent). The task is designed to show how the individual responds and processes clashing information, known as the “Stroop effect” (Price et al, 2012). The response latencies are based on the length of time taken to respond to the stimuli presented. Williams et al (1996) popularised the ‘emotional Stroop task’, referred to in the present study as the eStroop task, which uses emotional stimuli. The eStroop task is designed to test the individual’s ability to inhibit a pre-potent response (i.e. executive functioning) when faced with emotional stimuli.

From the Stroop task, attentional bias can be demonstrated in a positive or negative manner. That is, a positive attentional bias score on the Stroop task indicates vigilance; and a negative attentional bias score is taken as indicating avoidance. Positive bias, i.e., vigilance is demonstrated by slower response times for emotional stimuli, when compared to neutral stimuli. Negative bias, i.e., avoidance, is indicated by quicker response latencies towards emotional stimuli when compared to response times towards neutral stimuli (Mathews and MacLeod, 1994).

Existing studies in NES have investigated executive functioning using a Stroop task (Bakvis et al, 2009; Almis et al, 2013) or a task-switching paradigm (Gul et al, 2014). Bakvis et al (2009) used a ‘masked’ emotional Stroop task with 39 participants with NES, and Almis et al (2013) used a Stroop task with 44 NES participants. Both studies reported that NES participants reported a bias towards stimuli, shown by slower response latencies. Bakvis et al (2009) concluded that NES participants demonstrated an attentional bias towards negative stimuli. Gul and Ahmad (2014) reported that when participants completed a task-switching task, when compared against controls (N=72), NES participants (N=72) showed difficulty with switching attention between tasks that included emotional stimuli. They concluded that NES participants were more likely to be using dysfunctional coping strategies (emotion suppression), demonstrating impaired emotional processing.

Study 2 was redesigned to test a proposed interactional model between interoception (interoceptive sensibility) alexithymia and attentional bias in NES. The model hypothesises that emotional processing is compromised in pwNES due to atypical interoception, alexithymia (emotion recognition) and attentional bias towards emotional stimuli (executive functioning). The model is shown below in figure 3.1. The arrows between interoception, alexithymia and attentional bias indicate that the relationship between each of the components is augmented by the dysfunction of the others, contributing to atypical emotional processing. The model does not seek to explain how NES may develop or what factors may pre-dispose to its development. The main aetiological theories pertaining to NES have been reviewed recently by Brown and Reuber (2016) together with supporting evidence for each of the different theoretical approaches. The proposed model is more simplistic with the intention of determining interactions between aspects of psychological function which may contribute to the persistence of symptoms once they have been established.

Figure.3.1. the proposed model of dysfunctional emotion processing in NES



Anxiety and Depression

As noted in the systematic review (Study 1), high and low interoception have been found to be associated with higher levels of anxiety, depression and alexithymia – although no clear conclusions could be drawn from the studies included in the review. Higher rates of psychopathology have been reported in several studies with NES samples (Anxiety: Holman et al, 2008; Preuter et al, 2002; Testa et al, 2011; Novakova et al, 2017 Depression: Kanner et al, 2012; Martino et al, 2018; Sojka et al, 2018; Pick et al, 2020). Comparisons of anxiety symptoms in patients with NES and epilepsy have shown a mixed picture with only nine out of 28 studies finding comparatively higher levels of anxiety in NES participants (Brown and Reuber 2016). Although anxiety and depression were not core features of interest in Study 2, it was decided to include measures of anxiety and depression to be able to characterise the populations included in the study.

3.2. The present study

The study was designed as a cross-sectional study comparing participants with NES with normal controls, with all measures completed online. A patient sample of NES with a confirmed diagnosis (by a consultant neurologist) could not be used due to COVID restrictions, so participants with NES were recruited via a well-known FND charity. Instead of using a task to measure heartbeat perception, a questionnaire measure, the Body Perception Questionnaire (BPQ-VSF: Body Perception Questionnaire Body Awareness Very Short Form, Porges, 1993; 2015), was used to measure interoceptive sensibility.

Interoceptive sensibility is described as an individual's perceived accuracy of their ability to recognise changes in their internal body state (Murphy et al, 2017; Garfinkel et al, 2015). The BPQ-VSF has been used in studies with several clinical populations (Gaggero et al, 2021; Betka et al, 2018; Limmer et al, 2015), including FND (Sojka et al, 2020; Ricciardi et al, 2021) and NES (Koreki et al, 2020).

The BPQ-VSF was chosen over other interoceptive questionnaires such as the Multidimensional Assessment of Interoceptive Awareness (MAIA: Mehling et al, 2018). This is because the psychometric properties of the MAIA include attention regulation and emotional awareness. It was felt that this measure would potentially overlap with two other measures in the proposed study; The Toronto Alexithymia Scale (TAS-20, Bagby et al, 1994) and Difficulties in Emotion Regulation (DERS, Gratz and Roemer, 2004). The BPQ-VSF is brief, at 12 questions, compared to some interoception questionnaires, so may also reduce the level of participation fatigue that participants could potentially face from having to concentrate on a screen to complete the study tasks.

3.3. Research aims and hypotheses

The aim of the current study was to compare emotional processing, via interoception, alexithymia and executive functioning/attentional bias, in people with non-epileptic seizures compared with healthy controls, and to compare the associations between measures of interoceptive sensibility, alexithymia and executive processing within the two groups.

Hypotheses

1. It was hypothesised, that compared to healthy controls, participants with NES would report lower interoceptive ability (determined by lower scores on the BPQ-VSF) and poorer executive functioning (slower response times on the eStroop), and higher rates of alexithymia (determined by the TAS-20) and emotion dysregulation (determined by the DERS).

With reference to the proposed model the following hypotheses were posited:

2. Scores on the Toronto Alexithymia Scale-20 and reaction times on the eStroop task to emotional stimuli would both independently predict scores on the Body Perception Questionnaire-Very Short Form
3. Scores on the Body Perception Questionnaire-Very Short Form and reaction times on the eStroop task to emotional stimuli would both independently predict scores on the Toronto Alexithymia Scale for NES participants but not controls.
4. Scores on the Body Perception Questionnaire-Very Short Form and the Toronto Alexithymia Scale would be independent predictors of reaction times (emotional interference) on the eStroop task to emotional stimuli for NES participants but not controls.

3.4. Methods

3.4.1. Study design

This study was an online experimental study using a mixture of self-report questionnaires and a neuropsychological paradigm. This design was a cross sectional between-groups study. All study activities were completed by study participants using Gorilla (www.gorilla.sc).

The electronic study platform was created using the Gorilla Experiment Builder (Anwyl-Irvine, Massonnié, Flitton, Kirkham and Evershed, 2018). The experiment was tested in it's entirety by four volunteers before the study went 'live'. Volunteers were asked to use different web browsers to complete the experiment from start to finish, and report any problems encountered to the researcher. Any problems were rectified before advertising the study.

3.4.2. Participants

<u>NES participants</u>

Inclusion criteria:

Adult ≥18 years of age.

Has received a diagnosis of NES.

Has capacity to provide informed consent.

Able to read and understand English.

Have access to the Internet.

Resides in the UK.

Exclusion criteria:

≤17 years of age.

No diagnosis of NES.

Unable to provide informed consent.

Co-morbid diagnosis of epilepsy, dementia or other neurological disorder.

Unable to read English.

Does not reside in the UK.

<u>Control participants</u>

Inclusion criteria:

No diagnosis of NES.

Adult ≥18 years of age.

Able to provide informed consent.

Able to read and understand English.

Have access to the Internet.

Resides in the UK.

Exclusion criteria:

≤17 years of age.

Unable to provide informed consent.

Co-morbid diagnosis of epilepsy, dementia or other neurological disorder.

Unable to read English.

Does not reside in the UK.

3.4.3. Power calculation

Based on the primary research questions, a priori calculations using G*Power (v3.1.9; Faul et al, 2007) indicated that a total of 92 participants should be recruited: 46 participants with non-epileptic seizures and 46 participants without. It was calculated that a sample of this size would have 95% power to detect a large between groups effect (r=0.6).

The effect size is the strength of difference or relationship between groups and has an impact on the power of the statistical test (Donovan, 2016). The larger the effect size, the more power is assumed of the significance test, and the smaller the probability of reporting a Type 2 error (Sullivan and Feinn, 2012). In the literature regarding interoception, studies ranged from small to large effects, with more studies reporting medium to large effect sizes. A study using the BPQ, the same interoception measure used in the present study, reported a large effect size (Koreki et al, 2020). Therefore, a large effect size was chosen for this study.

3.4.4. Measures

All measures were self-report questionnaires and are detailed in the sections below. See appendices E-I for questionnaires.

3.4.4.1. Interoception

Body Perception Questionnaire- Very Short Form (BPQ-VSF: Porges, 1993; 2015)

This study assessed interoceptive sensibility using the Body Perception Questionnaire Body Awareness Very Short Form (BPQ-VSF: Porges, 1993; 2015).

The questionnaire is designed to report the subjective awareness an individual has of their internal bodily activity. The BPQ-VSF is the shorter version of the Body Perception Questionnaire.

The BPQ-VSF was chosen for this study because participants were required to complete several questionnaires as well as the eStroop task. Therefore, in addition to the justifications outlined earlier, and to achieve completion of all the study tasks in around one hour, the very short form version of the BPQ was selected.

The BPQ has been used in studies with several populations (Gaggero et al, 2021; Betka et al, 2018; Limmer et al, 2015), including FND (Sojka et al, 2020; Ricciardi et al, 2021) and NES specifically (Koreki et al, 2019). The Body Perception Questionnaire Very Short Form questionnaire (BPQ-VSF) is a subscale from the larger 122 item Body Perception

Questionnaire (Porges, 1993, 2015). Internal consistency of the BPQ-VSF has been reported as Cronbach's alpha =0.83 American college sample; =0.86 Spanish internet sample; =0.91 American internet sample (Cabrera et al, 2017). Test-retest reliability was reported as 'excellent' (ICC=.97).

3.4.4.2. Alexithymia

Alexithymia was measured using the Toronto Alexithymia Scale (Bagby et al, 1994). The TAS-20 contains 20 questions designed to measure high or low alexithymia. The scale provides a total score and individual scores for each of its three subscales: *difficulty identifying feelings*; *difficulty describing feelings; externally orientated thinking*. See Appendix G. On the TAS-20, a cut-off score of ≥61 is used to determine whether individuals meet the threshold for alexithymia or not, based on the total score achieved on the questionnaire (Taylor, 2000).

Internal consistency reported as Cronbach's alpha = 0.81 (Bagby et al, 1994).

3.4.4.3. Emotion regulation

The Difficulties in Emotion Regulation Scale (Gratz and Roemer, 2004) was included as a measure to investigate emotion regulation. This questionnaire is designed to give an overall score of emotion dysregulation, as well as scores on individual subscales. The higher the score, whether on total or subscale, the higher the level of emotion dysregulation.

There are 36 questions associated to six subscales relating to emotion regulation: *nonacceptance of emotional responses; difficulties in engaging in goal-directed; impulse control difficulties; lack of emotional awareness; limited access to emotion regulation strategies; lack of emotional clarity.*

The scale creators report the DERS to have high internal consistency (Chronbach's alpha = 0.93; Gratz and Roemer, 2004).

3.4.4.4. Depression

The Patient Health Questionnaire (PHQ-8, Kroenke et al, 2001) was used to measure depressive symptoms in participants.

The PHQ-8 is an eight-item diagnostic tool designed to measure severity of depressive symptoms in the general population. The overall score is interpreted as indicating mild, moderate or severe depression in the respondent.

The PHQ-8 has been reported to be comparable to the PHQ-9 for detecting major depression when a cut-off ≥10 is used; sensitivity of 88%, specificity of 88% (Kroenke and Spitzer, 2002). Internal consistency of the scale has been reported as Cronbach's alpha = 0.892 (Shin et al, 2019).

A meta-analysis reported that scores on the PHQ-9 and PhQ-8 were closely correlated (r = 0.996, CI 95%); with the PHQ-8 demonstrating a small reduction in sensitivity, but not specificity when 10 is used as a cut-off as indicating current depression in research and general populations (Wu et al, 2020).

3.4.4.5. Anxiety

This study used the Generalized Anxiety Disorders scale (GAD-7: Spitzer et al, 2006). This is a seven-item scale used in both clinical and non-clinical settings.

The GAD-7 has been reported as being an effective tool to assess the severity of generalised anxiety disorder in clinical and research settings (Spitzer et al, 2006), although it has been recently found to have limited diagnostic utility with pwNES (Goldstein et al, 2023). A score of 10 is used as the cut-off to identify general anxiety disorder; with additional cut points to indicate mild (score of 5-9), moderate (score of 10-14) or severe (score of 15-21) anxiety. Respondents can score from 0-21 (Spitzer et al, 2006).

The GAD-7 has been reported as showing a good internal consistency (Chronbach's alpha = 0.866) with a community sample (Johnson et al, 2019), and high internal consistency with pwNES (Chronbach's alpha = 0.92; Goldstein et al, 2023).

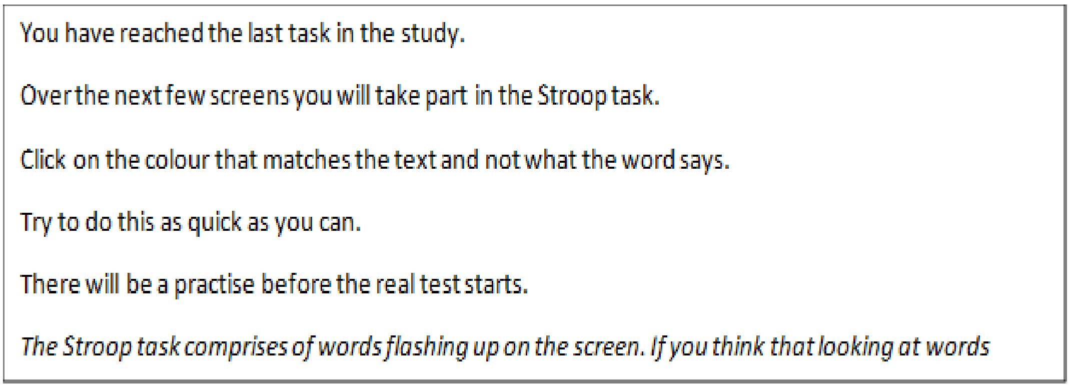
The measures of depression and anxiety were included to add contextual information about the two experimental groups.

3.4.4.6. Experimental paradigm

Emotional Stroop task (eStroop)

This paradigm is designed to investigate attentional bias towards stimuli, used to indicate emotional interference.

Text on the introductory screen instructs participants on how to perform the task (see text box below, fig 3.2). The text included a trigger warning:



You have reached the last task in the study.

Over the next few screens you will take part in the Stroop task.

Click on the colour that matches the text and not what the word says.

Try to do this as quick as you can.

There will be a practise before the real test starts.

The Stroop task comprises of words flashing up on the screen. If you think that looking at words

Fig 3.2 Introductory screen to the Stroop and eStroop tasks

Practice trials

Participants saw a fixation cross on the screen, displayed for 6000ms (Dresler et al, 2009). Participants were then given a practice run to orientate them towards that which the task required. An example of one of the practice screens is shown here (fig 3.3):

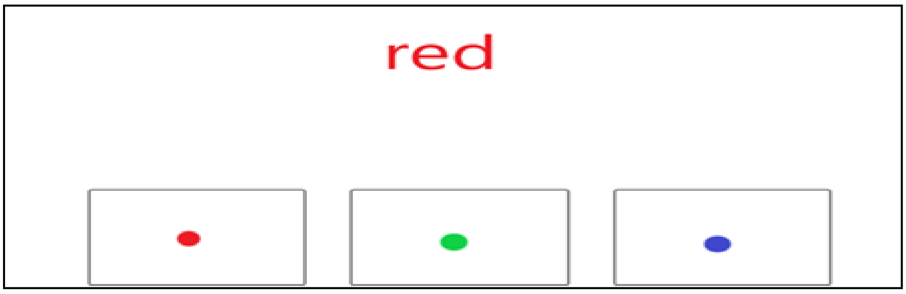


Fig. 3.3. This is an example of a congruent word. Both the meaning and the colour of the text match.

Participants were given two practice blocks, based on the classic Stroop task (Stroop, 1935). That is 10 words in each block consisting of congruent and incongruent words. The task required participants to respond to the colour of the text by click on the corresponding colour from the buttons presented on the screen. For example, the word ‘green’ may be presented in red text. Using the allocated button, the participant should choose ’red’ in response to the word presented on the screen.

3.4.4.6.1. Experimental stimuli

It has been argued that response times towards stimuli may be affected by several factors (Larsen et al, 2006). Stimuli, presented in the form of words should be matched for all lexical components (such as familiarity, frequency and length) because these are features that contribute to word recognition suggest Larsen et al (2006).

This means that word length should be the same in all word lists: control (neutral) and the emotional (threat) word lists because otherwise reaction times may be affected by word length, rather than meaning (Larsen et al, 2006). A list of words has been developed by Larsen et al (2008) that has been matched for length and valence. This list of 66 words (22 negative; 22 positive; 22 neutral) were used for the eStroop task in the present study.

eStroop trials

During the main experimental task, participants were presented with negative (threat), neutral (control) and positive words (Larsen et al, 2008). The baseline response time was taken from reaction times (RT) to the neutral stimuli to measure interference. An example is shown below of a negative word from the list developed by Larsen et al (2008).

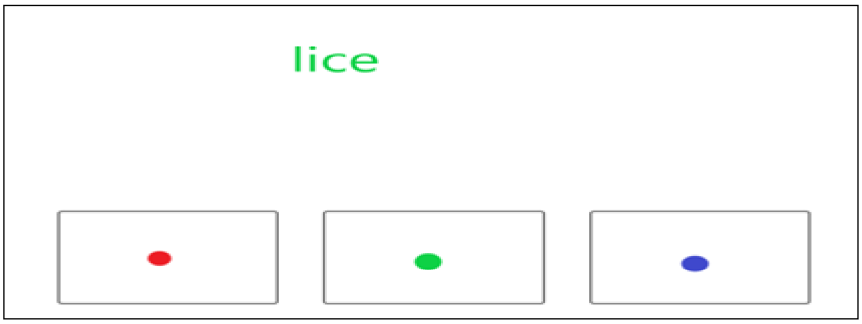


Fig.3.4. Example of negative stimuli.

Participants carried out the task twice: once with neutral and negative words, and neutral and positive words. Stimuli, i.e. words remained on the screen until the participant pressed a button in response.

At the halfway point, participants received a message to tell them they were halfway and to continue.

The task outcome was recorded as the participant's response time to stimuli - taken as a measure of emotional interference experienced.

3.5. General procedure

3.5.1. Ethical considerations

To reduce the potential impact of spreading coronavirus, the study was designed to be conducted online. Had the research been carried out face to face, the researcher would have to have used personal protective equipment (PPE). This raised the additional factor of using

PPE when it was known that some NHS services were struggling to obtain enough PPE for NHS staff. The researcher would have been using potentially crucial resources for 'non-essential' research and it was also felt unethical to expect participants to attend face to face meetings solely for the purposes of research. There were also practical limitations due to a lack of room space in hospitals.

Although it was not anticipated that the research would induce distress for participants, the Patient Health Questionnaire-8 (PHQ-8: Kroenke et al, 2001) was used in the present study rather than the PHQ-9. The difference between the two versions of the Patient Health Questionnaire is that the PHQ-8 omits the question: *Thoughts that you would be better off dead or of hurting yourself in some way*. During the planned face to face study, the researcher would have had the opportunity to check with participants if they needed support with their mental health. But the present study was designed to be to be completed anonymously online. This meant that there was no input from the researcher at the time of participation because participants could choose to participate whenever they wanted to. Therefore, the PHQ-8 was considered more appropriate for this study.

3.5.1.1 Ethical approvals

Ethical approval was sought from the University of Leeds School of Psychology Research Ethics Committee (SOPREC, Ref: PSYC-332). Approval was received after a short delay on the 20th October 2021. Ethical approval confirmation email available in Appendix A.

Delays to advertising the study and recruitment resulted after the researcher became unwell with Coronavirus.

3.5.2. Recruitment

Potential participants were recruited via a variety of methods. The study was advertised by FNDHope UK, a charity specifically set up to provide advice and help for people with FND. The research was also publicised using social media and via the School of Psychology, University of Leeds, participant database. The advert outlined the main inclusion/exclusion criteria that participants should meet, and what individuals would be required to do if they chose to participate in the study. (See Appendix J).

Those individuals that were interested in the research followed a link to the study page for their group: control or NES. Alternatively, for those participants who wished to see the information before visiting the research site, the participant information sheet and consent form was available to view on the researcher's university webpage,

Data was collected from 20th December 2021 to 21st February 2022. See diagram 3.5.

3.5.2.1. NES sample

The study was advertised over social media by the researcher and FNDHope UK. The study was advertised by the UK arm of the international charity FND Hope over their multiple social media accounts, as well as on the research page of their website. Their assistance was instrumental to recruitment.

3.5.2.2. Control participants

The control group was mainly recruited from the distribution list held by the School of Psychology. This is a database of individuals interested in taking part in research. The database includes students and staff at the University, as well as the general public who have registered their interest in being research participants.

Adverts of new studies are sent out to these potential participants via email. The study advert contained the links so that participants could access the study directly. The researcher's contact details were included for those participants who had questions or encountered problems.

Recruitment flow chart showing how NES and control participants were recruited to Study 2

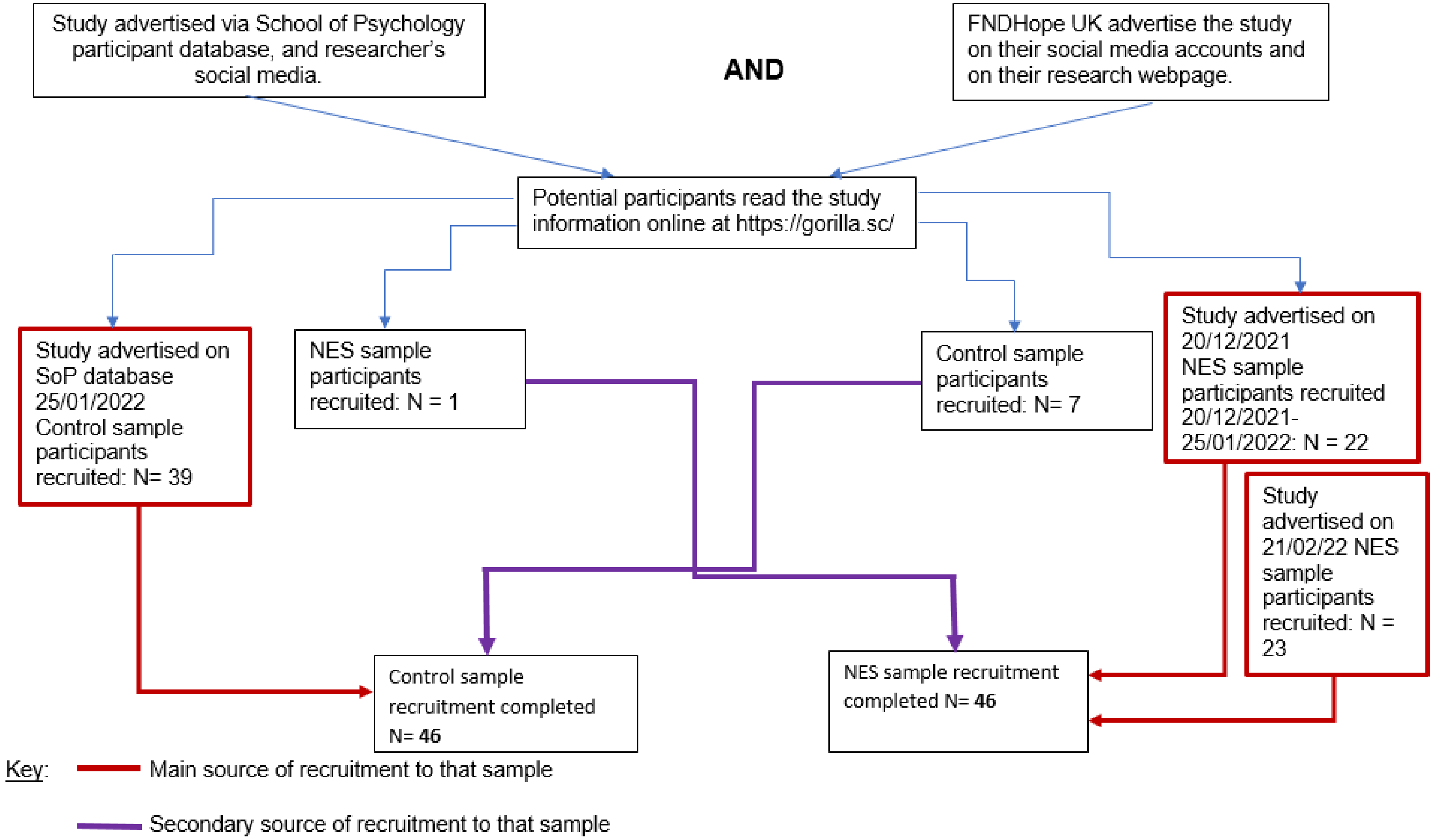


Diagram 3.5

3.5.3. Research activities

The online platform Gorilla (www.gorilla.sc; Anwyl-Irvine, Massonnié, Flitton, Kirkham and Evershed, 2018) acted as the host for all research activities. That is, recruitment, consent, completing the questionnaires and the eStroop test occurred remotely via this platform.

The study was designed such that participants were required to complete all activities before they could progress to the eStroop task. That is, no item could be left unchecked or unanswered. Once the projected sample size was achieved, the experiment was designed to automatically stop recruiting participants.

3.5.4. Consent

Interested individuals accessed the study information via the study pages on Gorilla, or on the researcher's university webpage.

Once participants accessed the study using the link for the Gorilla site, they first viewed an introductory video, summarising what was expected from them as a participant (see Appendix K for video transcript). This video was followed by the participant information page (Appendix B).

After the information page, participants were directed to an online consent form to provide written consent before completing any questionnaires or taking part in the eStroop task. Participants were only able to progress to the questionnaires and task, if they provided consent to all items listed in the consent form (see Appendix C).

One of the items on the consent form stipulated that the participant should use a laptop or desktop computer to participate. Although participants ticked this box to report that they were using a desktop/laptop computer, around a fifth of the sample (N=18; 20%) used tablets or mobile phones to complete the study tasks.

The requirement to use a laptop/desktop computer for the study had been added to ensure that participants would have a large enough screen on which to view the stimuli for the eStroop task. This is because it was assumed that screen size may have an impact on response times, i.e. the stimuli may be harder to see on a smaller screen and therefore response times may be considerably slower. This is one such variable that could have been controlled if the researcher had set the parameters of the study so that only those using a laptop/desktop computer could participate, however this was not the case and was not known until data had been collected.

3.5.5. Questionnaires

After the consent process, participants first completed a demographic form. The demographic questionnaire comprised a list of questions such as age, sex, ethnicity, occupation and education.

The only difference between the versions, of the demographic questionnaires, was that the NES group completed questions regarding their seizures. Participants with NES were asked when they experienced their first seizures, and when they were diagnosed NES. All other questions were the same for both groups.

3.5.6. 'Thank you' voucher

Participants were given the option to receive a voucher of the value of £10, as a 'thank you' gesture for their participation. Those participants that wished to receive the voucher were asked to provide their email address at the end of the experiment. The voucher was sent to them electronically.

3.6. Data analysis

3.6.1. Reducing bias

It is suggested that steps are taken to reduce bias, such as using more robust methods to analyse data by using bootstrapping (Field, 2013).

A common method used when dealing with reaction time data is to remove those values found to be ±2.5 standard deviations above or below the mean reaction time (Ratcliff, 1993). In the present study, instead of excluding eStroop latencies considered as outliers, i.e. ± 2.5 standard deviations from the mean, data were analysed using more robust methods. Field (2013) indicates that Spearman's correlation coefficient can be used to 'minimize the effects of extreme scores'.

3.6.2. Inferential statistical tests

To test the hypotheses set out at 3.2., total scores from questionnaire measures and emotional interference scores (attentional bias) were used in the analyses.

The mean emotional interference (EI) score was calculated using mean reaction times (RT) from performance on the eStroop task (Dresler et al, 2009; Pratto and John, 1991):

EI Negative = RTnegative – RTneutral

EI Positive = RTpositive – RTneutral.

After screening the data for normality, bootstrapped *t*-tests and non-parametric correlational analyses were conducted. In place of conducting non-parametric tests to compare means, data were subjected to bootstrapping to use a more robust method of analysis (Field, 2013). *T*-tests were used to test for differences between the groups, across all measures. Using total scores, Spearman's correlation coefficient analyses were conducted to test the strength and direction of the correlation within groups, with absolute values of *d,* i.e., effect size reported.

To test the hypotheses 2-4, data were transformed by using a reverse score transformation followed by a log transformation to base 10, so that a linear regression could be carried out. The regression analyses were conducted separately for both groups: NES and control participants. The measure of interoception was used as the dependent variable in the first instance, with alexithymia (TAS-20) and reaction times to emotional stimuli (eStroop test) as the independent variables (hypothesis 2). This was repeated with alexithymia (TAS-20) as the dependent variable, and interoception (BPQ-VSF) and reaction times (eStroop) as the independent variables. Finally, reaction times on the eStroop were used as the dependent variable with alexithymia (TAS-20) and interoception (BPQ-VSF) as the independent variables (hypothesis 4). Demographic variables and measures of depression and anxiety and emotional regulation were not included in the regression analyses.

The Data was analysed using the software package IBM SPSS 27.

3.7. Data storage

Data consisted of scores from completed questionnaires, response times from performance on the eStroop task and information collected from the participant demographic forms. This has been stored in accordance with the University of Leeds' data storage policy, which stipulates that OneDrive-University of Leeds is appropriate for the storage of anonymised data.

3.8. Participant withdrawal from study

Participants were free to withdraw from the study at any point. This was made clear in the Participant Information Sheet and consent form (Appendix B and C) in which it was stipulated that participants were able to withdraw from the study at any point, up to the point that the data had been collected. Once data was submitted via the online system (www.gorilla.sc; Anwyl-Irvine, Massonnié, Flitton, Kirkham & Evershed, 2018) data could not be requested to be removed by participants.

3.9. Assessment and management of risk

From an ethical perspective, the PHQ-8 was used to mitigate the risk of highlighting distress that the individual may have had or be experiencing at the time they participated in the study, however, the risks were considered to be minimal.

Although none of the research activities were designed to induce seizures, a ‘trigger warning’ was included before the eStroop task. This was to draw attention to individuals with NES in case their seizures may be induced via flashing stimuli. This was also mentioned in the Participant Information Sheet so that potential participants were made aware of this before continuing through the questionnaires to the task.

3.10. Results

3.10.1. Recruitment

Ninety-two participants were recruited to the study, 46 in the NES group and 46 controls. One participant stated on the feedback form that they experienced functional symptoms, although not non-epileptic seizures. This participant was removed from the control sample before statistical analyses were conducted and is only included in the demographic data in table 9.

3.10.2 Data exploration and assumption checks

3.10.2.1. Data exploration

Data for age, BMI, questionnaire scores, and reaction times on the eStroop, were visually inspected to check for any obvious inputting errors. Any errors found were corrected.

Histograms, p-p and q-q plots of reaction times on the eStroop indicated that data were not normally distributed in both groups of participants. Tests for skewness on data from the NES group indicated that reaction time data for negative stimuli (skewness= 4.277) and the first set of neutral stimuli (skewness= 1.117) were likely not normally distributed and indicated a positive skew, with scores gathering to the left of the distribution. Reaction time data for the positive stimuli and second group of neutral stimuli were likely to be normally distributed (skewness: positive = -0.343; neutral= 0.408).

Reaction time data from the control group suggested that scores were not normally distributed for any of the eStroop task stimuli (skewness: negative = 1.589; neutral= 1.945; positive = 4.238; neutral = 4.238). The data demonstrated a positive skew, meaning that reaction times were grouped to the left of the distribution.

3.10.2.2. Kolmogorov-Smirnov test for normality

Tests for normality of the data, using the Kolmogorov-Smirnov test, showed that several of the scores for some of the measures were not normally distributed. The results were:

- NES group data for the interoceptive sensibility score *D* (46) = .192, *p*< .001.
- Control group data for the interoceptive sensibility score *D* (46) = .140, *p*=.025.
- NES group data for alexithymia scores *D* (46) = .207, *p*<.001.
- Control group data for alexithymia scores *D* (46) = .131, *p*=.045.
- NES group data for negative reaction time on the eStroop task *D* (46) = .173, *p*=.01; neutral reaction times *D* (46) = .163; *p*=.004; and positive reaction time *D* (46) = .150; *p*=.011.
- Control group reaction times to negative stimuli on the eStroop task *D* (46) =.084, *p*=.200; neutral stimuli *D* (46) = .100, *p*=.200; positive stimuli *D* (46) =.206, *p*<.001 were significantly non-normal.

3.10.2.3. Homoscedasticity

For the interoception scores, Levene's test reported that variances were found to be unequal for NES participants and controls, *F* (1, 86) = 8.63, *p*=.04.

However, Field (2013) warns that in larger samples, Levene's test can be significant when groups may not be considerably different.

3.10.2.4. Demographic characteristics

Demographic characteristics for NES and controls can be found in Tables 8 and 9 respectively. A comparison of the characteristics for the two groups (NES and controls) is presented in table 10. The demographic data presented in tables 8 and 9 are based on the raw data, i.e. 92 individuals participated: 46 participants with NES and 46 participants acting as controls, prior to the one control participant being withdrawn.

There were fewer complete responses for ethnicity and employment. It is presumed that there was a malfunction with the online platform used for the study that resulted in this data loss for these demographic questions.

There was no difference in age between the groups but the NES group had more female participants (89.1%) than the control group (67.4%) (*p*=0.008).

Significant differences between the groups were found on ethnicity (*p*<0.001), with a greater proportion of participants from a white background in the control group (76.2%)

versus controls (54.5%). However, there was also a substantial percentage of participants for whom this data was missing: 41.3% NES and 6.5% controls.

The groups were significantly different regarding employment status (*p*=0.031), with more NES participants being in employment than controls.

Participants with NES were asked an additional question about how long it took for them to receive a diagnosis; for half the sample (50%), this took between six months to one year to receive the diagnosis and a fifth (19.6%) of participants received a diagnosis of NES in less than six months. A similar proportion (17.4%) of participants received a diagnosis between one-two years. A minority of participants (8.7%) reported that it took between five-nine years to receive a diagnosis. For some participants (4.3%) this data was not available.

Table 8. Non-epileptic seizures participants' demographic data

Non-epileptic seizures group characteristics	**Number (%)**							
Sex	**Female**	**Male**				**Did not want to answer**		
n=46	41 (89.1)	4 (8.7)				1 (2.2)		
Age (years)	**18-25**	**26-30**	**31-35**	**36-40**	**41-45**	**46-50**	**51-55**	**Missing**
n=45	9 (19.6)	24 (52.2)	2 (4.3)	6 (13.0)	1 (2.2)	2 (4.3)	1 (2.2)	1 (2.2)
Ethnic origin	**Asian**	**Italian**	**White**[1]			**Missing**		
n=27	1 (2.2)	1 (2.2)	25 (54.4)			19 (41.3)		
Employed	**Employed**	**Student**				**Unemployed**		
n=46	28 (60.9)	5 (10.9)				13 (28.3)		
n= 46	0 (0)	4 (8.7)		5 (10.9)		5 (10.9)		32 (69.6)
Highest qualification	**High school qualification**[2]	**Bachelor's degree**		**Master's degree or postgrad qualification**		**PhD**	**Other qualification**[3]	**Missing**
n=25	9 (19.6)	5 (10.9)		3 (6.5)		2 (4.3)	6 (13.0)	21 (45.7)
Time taken to receive diagnosis	**>6 months**		**6 months – 1 year**		**1-2 years**		**5-9 years**	**Missing**
n=44	9 (19.6)		23 (50.0)		8 (17.4)		4 (8.7)	2 (4.3)

[1] Includes participants that answered 'British', 'White', 'White British', White Scottish'.

[2] Includes participants that answered 'CSE', 'GCSE', or 'A Levels'.

[3] Includes participants that answered 'college', 'national 5', 'NVQ Level 2', GNVQ', 'BTEC diploma', SQA higher'.

[4] Includes participants that answered 'physio exercises' or exercises specific to a condition

Table 9. Control group demographic table

Control group characteristics	**Number (%)**										
Sex	**Female** (n %)	**Male** (n %)						**Did not want to answer**			
n=46	31 (67.4)	14 (30.4)						1 (2.2)			
Age (years)	**18-25**	**26-30**	**31-35**	**36-40**	**41-45**	**46-50**	**51-55**	**56-60**	**61 +**	**Missing**	
n=45	16 (34.8)	6 (13)	4 (8.7)	6 (13.0)	2 (4.3)	4 (8.7)	3 (6.5)	1 (2.2)	4 (8.7)	1 (2.2)	
Ethnic origin	**Chinese**[1]	**Dual heritage**[2]		**Indian**				**White**[3]		**Missing**	
n=43	4 (8.7)	3 (6.5)		1 (2.2)				35 (76.2)		3 (6.5)	
Employed	**Employed**	**Student**						**Unemployed**			
n=46	26 (56.5)	14 (30.4)						6 (13.0)			
Highest qualification	**High school qualification**[4]	**Bachelor's degree**		**Master's degree or postgrad qualification**				**PhD**		**Other qualification**[5]	
n=46	15 (32.6)	14 (30.4)		11 (23.9)				2 (4.3)		4 (8.7)	

[1] includes participants that answered 'British Chines' and 'Chinese'

[2] Includes participants that answered 'mixed', 'mixed race', or 'mixed white and black Caribbean'.

[3] Includes participants that answered 'British', 'White', 'White British', White English', or 'White European'.

[4] Includes participants that answered 'high school', 'CSE', 'GCSE', 'A Levels', or 'HND'.

[5] Includes participants that answered 'vocational level 2', 'Level 4 diploma', 'college', 'NVQ Level 3'.

[6] Includes participants that answered 'jogging' or 'running'.

[7] Includes participants that answered 'physio exercises' or exercises specific to a condition.

[8] Includes participants that answered 'walking' or 'hiking'.

Table 10. Summary of participant demographics by group (non-epileptic seizure or control participants)

	NES	**Controls**	**Test outcome**
Age (years) (n=90)	**Mdn (IQR)**	**Mdn (IQR)**	**Mann Whitney U test**
	30 (7)	31 (23)	U (89) =1066, p=.533
Sex (n = 90)	n (%)	n (%)	**Chi square test**
Female Male	41 (45.5) 4 (4.4)	31 (34.4) 14 (15.5)	χ^2 =6.944 p=.008
Ethnicity (n = 90)	n (%)	n (%)	
White Other ethnicities Missing	26 (28.9) 1 (1.1) 19 (21.1)	35 (38.9) 8 (8.9) 3 (3.3)	χ^2 = 18.454, p<.001
Employment (n =92)	n (%)	n (%)	
Yes No Student	28 (30.4) 13 (14.1) 5 (5.4)	26 (28.3) 6 (6.5) 14 (15.2)	χ^2 = 6.916, p=.031

Mdn=median IQR = interquartile range

3.10.3. Questionnaires

Table 11 shows the scores for the questionnaire measures used in the study and the eStroop task. Higher scores across all questionnaires were reported by NES participants, compared to controls. This was true for both total and sub-scale scores on questionnaires.

Bootstrapped independent *t*-tests were used to report group differences on alexithymia, interoception, depression and anxiety scores. Bias corrected and accelerated bootstrap 95% confidence intervals are reported in square brackets in the text below.

Table 11. Bootstrapped *t*- tests for questionnaire and eStroop responses

Variable (Method of measurement)	**Group**	**Mean (SD)**	***t statistic***	**Effect size (*d*)**
Interoceptive sensibility (BPQ-VSF)	**Non-epileptic seizures**	39.00 (8.882)	-5.105*	0.91
	Control	26.84 (13.342)		
Alexithymia (TAS-20)	**Non-epileptic seizures**	61.22 (13.303)	-4.710*	1.00
	Control	48.24 (12.965)		
Emotion dysregulation (DERS)	**Non-epileptic seizures**	116.00 (23.460)	-5.159*	1.11
	Control	91.16 (22.455)		
Depressive symptoms (PHQ-8)	**Non-epileptic seizures**	14.13 (5.158)	-6.187*	1.19
	Control	6.67 (6.281)		
Anxiety symptoms (GAD-7)	**Non-epileptic seizures**	11.52 (4.938)	-5.863*	1.01
	Control	5.33 (5.130)		
Positive Emotional Interference (eStroop)	**Non-epileptic seizures**	217.929 (26.134)	-2.102	0.84
	Control	39.449 (212.092)		
Negative Emotional Interference (eStroop)	**Non-epileptic seizures**	260.007 (164.865)	-1.787	2.30
	Control	93.527 (72.502)		

SD= standard deviation

*significant at *p*<0.001 level (2-tailed)   ** significant at p<0.05 (2-tailed)

Cohen's (1988) *d*= 0.2 small; 0.5 medium; 08 large.

Interoception

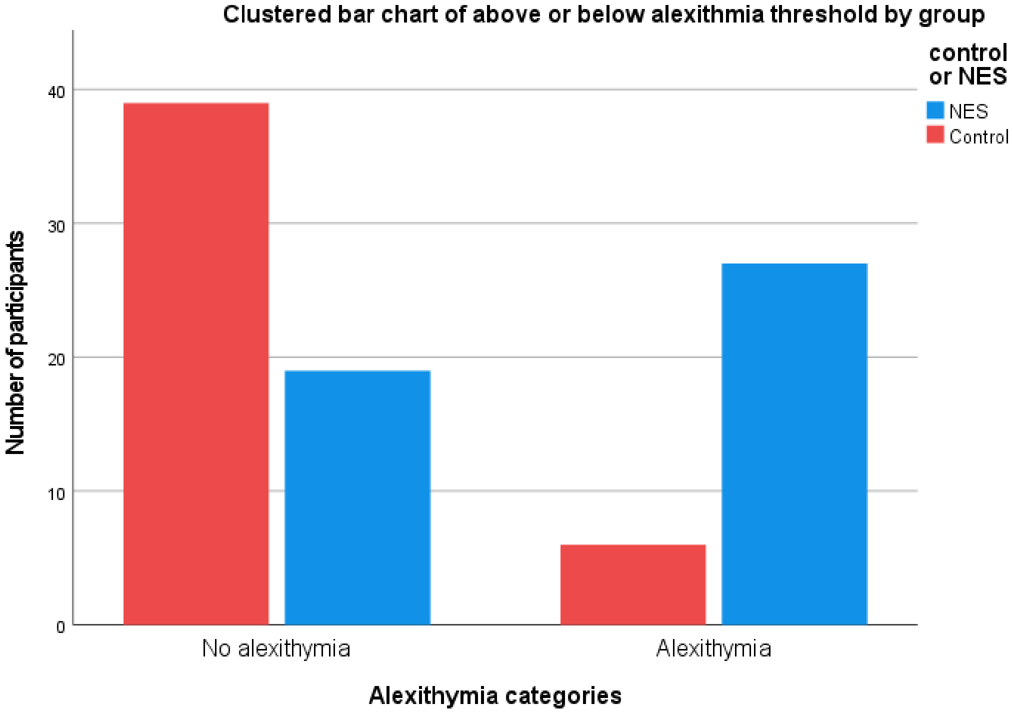
Participants with NES (*M*=39.00, *SE* = 1.31) reported higher BPQ-VSF scores, compared to control participants (*M*=26.84, *SE*= 1.99). This difference -12.156, BCa 95% CI [16.898, -7.413] was significant *t* (76) = -5.105, *p*<.001; represented by a large-sized effect, *d*= 0.91

Alexithymia

More participants with NES (n=27) scored above threshold (61 or above) on the TAS-20, compared to six control participants. See figure 3.6

NES participants reported higher levels of alexithymia (*M*= 61.22, *SE* = 1.961) compared to healthy controls (*M*=48.24, *SE* = 1.933). This difference, -12.973, BCa 95% CI [-18.446, -7.413] was significant *t* (89) = -5.105, *p*<.001; with a large sized effect *d*= 1.00.

Figure 3.6. The proportion of participants in each group (NES and controls) who were above cut off on the Toronto Alexithymia Scale (TAS-20)



Emotion dysregulation

Participants with NES reported higher levels of emotion dysregulation on the DERS than controls. This difference, -24.844, BCa 95% CI [-34.413, -15.276] was significant *t* (89) = -5.159, *p*=<.001; represented by a large sized effect, *d*=1.11.

Depression

Participants with NES had higher scores on the PHQ-8 (*M*= 14.13, *SE* = .761), than control participants (*M*= 6.67, *SE*=.936). The difference between the groups -7.464, BCa 95% CI [-9.862, -5.065] was significant *t* (85) = -6.187, *p*<.001; with a large sized effect, *d*= 1.19.

Anxiety

NES participants had higher scores on the GAD-7 (*M*= 11.52, *SE* = .728), than control participants (*M*=5.33, *SE*=.765). This difference, -6.188, BCa 95% CI [-8.286, -4.091] was significant *t* (89) = -5.863, *p*<.001; represented by a large sized effect, *d*= 1.01.

3.10.4. Neuropsychological test: Emotional Interference

Reaction time scores were used to produce the emotional interference scores (EI): negative and positive. Figures 3.7 and 3.8 shows the mean positive and negative EI scores, respectively, by group.

The mean emotional interference score was calculated using mean reaction times (RT) (Dresler et al, 2009; Pratto and John, 1991):

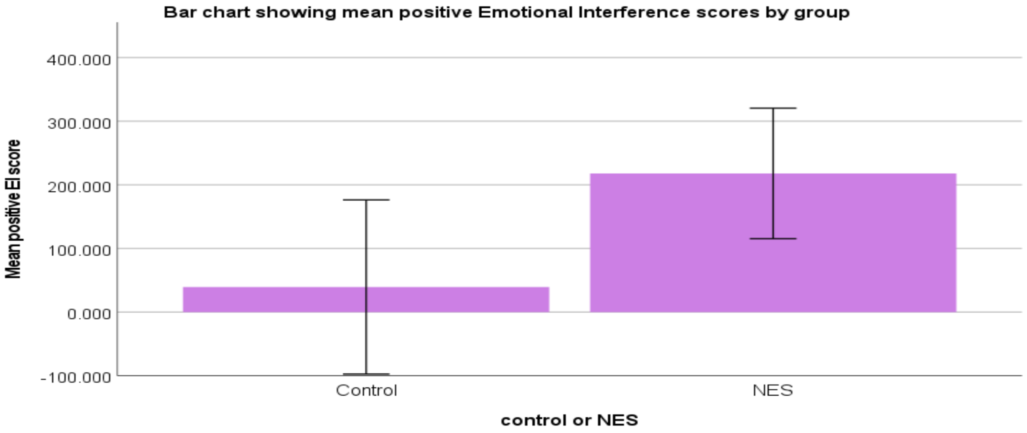
EI (Emotional Interference) = RTnegative – RTneutral and RTpositive – RTneutral

Only correct response times are included to calculate emotional interference scores (Dresler et al, 2009). Twice as many NES participants made incorrect responses (N=22) compared to control participants (N=11). The higher the number of incorrect responses, the greater level of attentional bias. This suggests that a higher level of interference was experienced by participants with NES.

Table 11 displays the mean positive and negative emotional interference scores, and the results are also reported here. There was no difference between participants with NES and healthy controls in scores for negative emotional interference (*t* (64) = -1.787, *p*=.079) on the eStroop task.

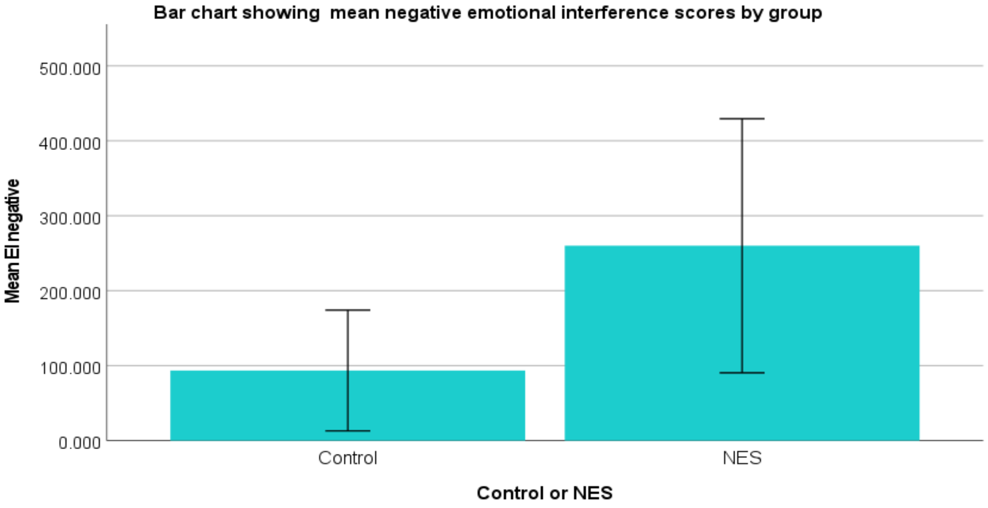
On emotional interference towards positive stimuli, NES participants were found to react slower (*M*= 217.929, *SE* = 51.526), compared to control participants who reacted quicker (*M*= 39.449, *SE*= 66.791). Although an independent *t*-test found that this difference, -178.480, BCa 95% CI [-320.199, -20.930] was not significant *t* (82) = -2.102, *p*=.072.

Figure 3.7.



*Error bars represent 95% confidence intervals.

Figure 3.8



* Error bars represent 95% confidence intervals.

3.11. Correlation analyses

To investigate associations between interoception, alexithymia and attentional bias, Spearman's correlation coefficient analyses were conducted. Bias corrected and accelerated bootstrap 95% confidence intervals are reported in square brackets for significant results (table 12). Scores on the PHQ-8, GAD-7 and DERS are also included in the table.

Table 12. Spearman's correlation coefficient matrix

Non-epileptic seizures n= 46	**BPQ-VSF**	**Negative EI**	**Positive EI**
BPQ-VSF	-	.093	.267
TAS-20	.184	.154	.313* [.041, .555]
DERS	.217	0.007	-.082
PHQ-8	.360* [-.001, .690]	.069	-.023
GAD-7	.395** [.067, .663]	.086	.120
Negative EI	.093	-	.393** [.134, .592]
Positive EI	.267	.393** [.134, .592]	-
Controls n= 45	**BPQ-VSF**	**Negative EI**	**Positive EI**
BPQ-VSF	-	.053	-.110
TAS-20	.119	.003	.022
DERS	-.053	.007	.056
PHQ-8	.328* [.029, .607]	.151	.166
GAD-7	.545** [.306, .724]	.150	-.016
Negative EI	.053	-	.038
Positive EI	-.110	.038	-

*Correlation was significant at the .05 level (2-tailed) **Correlation was significant at the.01 level (2-tailed). No correlations retained significance after a post-hoc Bonferonni correction (*p*<.008) was applied.

All associations were either very weak or weak, for both NES and control participants. The exception being the association between interoception and anxiety for controls, this was reported as a modest positive correlation (r_s=.545). Several significant correlations were reported in both NES and control groups. However, these associations did not survive post-hoc Bonferroni correction for multiple comparisons (*p*<.008).

3.12. Regression analyses

In order to further explore the model set out in figure 3.1, bootstrapped simple liner regression analyses were carried out. Data were reversed scored and then log transformed to correct for the negative skew in the variable data.

Hypothesis 2. Interoception was set as the dependent variable and the independent variables were alexithymia and emotional interference (attentional bias). No significant results were reported (*p*>.05). That is, alexithymia and emotional interference (positive or negative) were not found to be independent predictors of interoception for either NES or control participants.

Hypothesis 3. When alexithymia was set as the dependent variable and the independent variables were interoception and emotional interference, the positive emotional interference scores were found to be independently associated with alexithymia scores for the NES sample b=-114.372, p=.024, BCa 95% CI [-214.96, -27.23] but not for the controls.

Hypothesis 4. When the eStroop scores were set as the dependent variables (response to positive stimuli in one regression and response to negative stimuli in the other regression) and the independent variables were interoception and alexithymia, no significant results for either group (NES or controls) were reported for positive emotional interference scores (p>.05) or for negative emotional interference scores (p>.05). That is, interoception and alexithymia were not found to be independent predictors of emotional interference (positive or negative) for either NES or control participants.

3.13. Discussion

The aims of the present study were to determine whether there were differences between NES participants and healthy controls on key aspects of emotional processing: interoception, alexithymia and executive functioning; and to investigate the associations between interoception, alexithymia and attentional bias within NES participants and healthy controls. Participants with NES scored significantly higher on measures of interoception, alexithymia, emotion dysregulation and depression and anxiety, when compared to healthy controls. Results for the eStroop task showed no difference between the groups in reaction times to negative or positive stimuli.

There were remarkably few associations of note either in the correlation or regression analyses between the BPQ-VSF, TAS-20 and eStroop results. Most of the correlation coefficients were very small and none of the correlations were significant after Bonferroni corrections were applied for either NES participants or controls. The regression analyses showed a significant relationship between the TAS-20 scores and the interference to positive emotional stimuli on the eStroop task which was independent of scores on the BPQ-VSF for NES participants. However, as this was the only significant finding, it is difficult to be certain of its importance.

Many studies have demonstrated that the emotional effect of negative stimuli will produce a delay (interference) in Stroop response times (Quan et al, 2020). This is because negative stimuli in whatever form trigger a threat response, which occupies an individual’s attention and implies hypervigilance. The reasons for interference with reactions to positive stimuli on the Stroop are less clearly formulated, and results from studies are less consistent. It has been hypothesised that there may be differential responses depending on the kind of positive stimuli. From an evolutionary perspective, positive stimuli that are linked in some form to survival (e.g. appetising food) may be prioritised and gain attention in comparison with positive stimuli which have a low biological imperative (e.g. beautiful flowers).

Several previous studies involving NES patients or participants have used the Stroop test but the results are difficult to compare because of differences in the study paradigms, study populations and kinds of Stroop tests used. For example, Bakvis et al, 2009 used an eStroop test which involved responses to angry and happy faces in patients with NES and healthy controls, with and without being subjected to stress. They reported evidence of positive attentional bias to angry faces in NES participants compared to controls under resting but not stress conditions. Interestingly, they also tested latency using the eStroop colour word test (similar to that used in the present study) and did not find any major differences using this form of the test. Other studies (Karaaslan and Hamamci, 2020) have also found similarities in performance on the eStroop Color-Word test for patients with NES and epilepsy, with both groups taking longer than normal controls. The NES participants who participated in the present study were not recruited from health clinics, due to problems with COVID-19 restrictions and so are not directly comparable to patients with NES, which may explain some of the differences in results with other studies.

The NES participants reported higher scores on the BPQ-VSF, compared to the scores reported by controls. This was surprising because it was expected that participants with NES would report poorer interoception, when compared to controls. Previous studies have reported mixed findings with regards to individuals with NES and interoception (Jungilligens et al, 2020; Ricciardi et al, 2021; Koreki et al, 2020; Pick et al, 2020; Demartini et al, 2019; Williams et al, 2021). With some studies reporting that participants with FND demonstrated poorer interoceptive accuracy when compared to controls (Koreki et al, 2020; Demartini et al, 2019; Williams et al, 2021). However, it should be noted that the present study used a self-report measure to assess interoception and there is a difference between the objective ability people have to perceive their own bodily activity (interoceptive accuracy) and their own estimations of how good they are at perceiving their internal bodily states (interceptive sensibility). In a study using the BPQ-VSF and a heartbeat perception task, Koreki et al (2020) were able to demonstrate that participants with NES perceived themselves to have better interoception, based on scores on the BPQ-VSF, compared to their actual performance on heartbeat perception tasks. Koreki and colleagues (2020) found the greater the discrepancy between a person’s beliefs about their awareness of their internal body milieu and their objective accuracy in judging interoceptive signals, the greater their reported levels of dissociation and the higher their seizure frequency.

The results of the present study suggest that people with NES have a tendency to hold interoceptive predictions which may be imprecise and exaggerated compared with healthy controls. If people with NES also have poorer interoceptive accuracy as suggested by some of the above studies (Koreki et al, 2020, Demartini et al, 2019, Williams et al, 2021), it may suggest that people with NES are unable to moderate or update their beliefs about their interoceptive awareness and the normal bottom up top down processing of sensation which relies on error signals to adjust expectations is impaired (Koreki et al, 2020).

As expected, the current study found a significant difference between the groups (NES and controls) on alexithymia. These findings are consistent with studies by Urbanek et al (2014) and Demartini et al, (2016) that reported that alexithymia was significantly higher in participants with NES, compared to healthy controls. Similarly, another study found participants with NES scored significantly higher on the TAS-20 compared to those with mild-Major Depressive Disorder (Martino et al, 2018). Brown et al (2013) also reported that in their study they found a subgroup of patients who had higher levels of alexithymia after performing a cluster analysis on the participants with NES.

However, other studies which have included participants with epilepsy reported finding no differences on alexithymia scores between NES and epilepsy participants (Tojek et al, 2000; Myers et al, 2013; Kaplan et al, 2021). This suggests that alexithymia may not be specific to NES and those with seizure disorders in general struggle with expressing feelings.

In the present study, when sample characteristics were compared, significant differences were reported on sex, ethnicity and employment. With regards to sex differences between the groups, male participants made up around a fifth of the sample (19.9%). Studies report that women are more commonly diagnosed with NES, compared to men. Most of the sample (82.1%) with NES were between the ages of 18-40 years old, with a mean age of 30.4 years. This was similar to the mean age of participants reported by other studies (32.6 years: Jungilligens et al, 2021; 34.7 years: Sojka et al, 2020). Participants with NES also scored higher on the measures of depression and anxiety which again was expected, based on findings reported in previous studies with NES samples (e.g. Testa et al, 2011; Novakova et al, 2017; Sojka et al, 2018; Pick et al, 2020).

Turning to the associations explored in the study. The correlation between alexithymia and interoception was a very weak non-significant association for both NES and control participants, and there were no significant associations between the key measures of interest for either NES participants or controls. This rather surprising finding suggests that the proposed model of interactional relationships between interoception (sensibility), alexithymia and executive functioning is not supported. In fact, according to the results of the study, the three aspects of emotional processing (interoception, alexithymia and executive functioning) appear to be relatively independent of each other.

Other studies, however, have found associations between alexithymia and interoception in populations other than NES participants. Murphy and colleagues (2018) have found that higher scores on the TAS-20 are associated with less accurate perception of interoception in healthy participants, and this is supported by other research with similar findings (Brewer et al, 2016; Longarzo et al., 2015; Shah et al., 2016). Interestingly, Murphy and colleagues (2018) used several different measures of interoception, including interoceptive sensibility and interoceptive accuracy in two different physical domains, whereas Brewer et al (2016) used two measures of interoceptive sensibility, an Interoceptive Confusion Scale and a State-Emotion Similarity Questionnaire.

The differences between findings of the present study and other findings can be explained by differences in methodology, including differences in study populations and measures used to assess interoception. It is also possible that there are distinctly different groups of NES with differing psychosocial profiles (Brown et al, 2013) within the NES population as a whole, which would influence the degree to which different psychological measures would be correlated. Unfortunately the present study was not designed to conduct a cluster analysis as the sample size in both groups is at the smallest margin deemed acceptable for such an analysis (Dalmaijer et al, 2022).

The current study used the Difficulties in Emotion Regulation Scale to measure general levels of emotion regulation between the two study groups. NES participants reported higher levels of emotion dysregulation, than controls. Although emotional regulation was not included in the proposed model which was tested in Study 2, it is of interest that there were no significant relationships between interoceptive sensibility and emotion dysregulation for either NES or control participants. Williams et al (2021) have reported that FND participants report more emotion processing deficits compared to controls, although they found that participants with FND demonstrated lower interoception when this was measured using a heartbeat perception task.

3.13.1. Strengths and limitations

There are several strengths of this study. First, this is the first study to the author's knowledge to explore associations between key aspects of emotional processing in participants with NES. It was noted in Study 1 that many studies have measured alexithymia and interoception in participants with either FND or NES in comparison with controls, but none have explored the association between them.

Second, the study used recognised self-report instruments with established psychometric properties to measure the psychosocial variables and neurocognitive variables of interest.

Third the study was fully powered to detect differences between the two groups in terms of the main measures.

Finally, using an online method to recruit and conduct the current study was also a strength. It made the study more accessible to those that may struggle with fatigue, mobility, or have other constraints on their time such as work or childcare. Hosting the study online allowed participants to take as long as they wished to consider participation, and when they did participate, they were not limited by the day or time of day. This may have removed concerns participants may have had about cancelling a research appointment if they were unwell on the arranged day.

Using a completely online method, whereby participants access the study information themselves, also reduced the potential for participants to feel pressured or coerced into participating in research, compared to participants being recruited from a clinic for example.

The study inevitably had several limitations in part due to its redesign because of the COVID-19 pandemic and the requirement to complete all the measures online.

First, interoception was assessed using a self-report measure as opposed to a heartbeat perception task. As discussed above interoceptive sensibility is not the same as interoceptive accuracy, and it is difficult to compare the results of the present study with other studies that have measured interoceptive accuracy.

Second, all the measures employed in the study were self-report. Although the TAS-20 has been widely used to measure alexithymia, it can be argued that people with pronounced alexithymic traits may have difficulty in self-awareness and may under report on a self -report measure. The Toronto Structured Interview for Alexithymia (Bagby et al, 2006) has been developed as an objective measure of alexithymia but it was beyond the scope of this study to employ such a detailed method.

Third, NES participants were recruited via a variety of methods including via a well-known charity. It was not possible, therefore, to confirm diagnosis of NES in the study group. The original plan had been to recruit from neurology clinics where the diagnosis would have been confirmed by a consultant neurologist. It is possible, therefore, that some participants in the study may have had symptoms that they attributed to NES but could in fact be anxiety /panic related.

Fourth, whilst the study was designed such that participants had to complete every field before progressing, there was still some loss of data. Not having the researcher present when participants completed questionnaires meant that the researcher was not able to manually input any data that was missed. A malfunction with the online platform used to provide the research activities presumably resulted in the data lost.

Fifth, hosting the study online may have reduced access for those who may not have the access to a computer. This could be the reason behind why some participants completed the research using a mobile phone or tablet, because this may have been the device available to them. Conducting the study in a psychology lab would have reduced the potential for other distractions and would have standardised the approach with regards to the device used to participate.

Sixth, a substantial proportion of the data on ethnicity was missing for the participants with NES. It is known from the literature that NES is diagnosed cross-culturally. However from the data available in the current study, most participants reported being from a White background. It could be inferred that the way in which the study was advertised meant that it did not reach a diverse population. This could be because those that access the FND charity are mostly from White backgrounds, or those diagnosed with NES in the UK are likely to be from White backgrounds. However, such conclusions cannot be established based on the data here.

Finally, a further possible limitation is that participants were not asked about co-morbid conditions. It could be that some other co-morbid conditions have an impact on an individual's bodily awareness and co-morbid conditions are common in those with NES (Dixit et al, 2013).

3.14. Conclusion

A model was proposed that a bi-directional relationship between interoception, alexithymia and attentional bias would be found in pwNES. No substantial evidence was found to support such a model either in people with NES or healthy controls. The study supports previous research which suggests that people with FND/ NES tend to overestimate their ability to perceive their internal bodily state in comparison with healthy controls. Although, it may also indicate that pwNES are more sensitive to their internal bodily states due to additional factors, such as higher rates of anxiety. The study also supports previous research which suggests people with NES report higher rates of self-reported alexithymia, greater difficulties with emotional regulation, and higher depressive and anxiety symptoms.

With regards to clinical implications, the findings suggest that each of these different aspects of emotional processing maybe of relevance in NES participants but may not be specific to NES. People with NES may require help with processing emotion managing psychological distress and possibly require support to learn how to distinguish their different emotional states better, and how they may experience these in their bodies, to lead them to regulate emotions more effectively.

CHAPTER 4: STUDY 3

A phenomenological exploration of emotional responses to life events in the context of non-epileptic seizures

4.1 Introduction

Life events research has an established interest in both physical and psychological illness. It has been suggested that even in those without diagnosed psychopathologies, life events influence how individuals respond to daily events (Myin-Germys et al, 2003). The Integrative Cognitive Model (ICM: Brown and Reuber, 2016) is a model in which traumatic life events are proposed as one of several factors that may continue to cause chronic stress and arousal responses towards daily events in people with NES (pwNES). In a study by Bowman et al (1999) it was stated that stressful life events were found to be reported by 92% of their sample. Such findings suggest that understanding how pwNES respond to life events may help to understand the role they play in the experience of NES.

In Study 2, it was investigated whether pwNES would show different patterns of emotion processing when compared to controls. The present study was a qualitative study aimed to provide a more nuanced understanding of whether pwNES perceived their emotions to have an impact on their seizures, in the context of life events experienced recently.

Life events research in NES

Life events studies in NES/ FND have been focussed on reporting psychological stressors around symptom onset. From these studies, it is stated that individuals with FND reported more life events than controls (Bowman et al, 1999; Tojek et al, 2000; Nicholson et al, 2016); and other studies reporting that pwNES do not report more life events than controls (Roelofs et al, 2005; Testa et al, 2012, although, a systematic review of quantitative FND studies did report higher rates of stressors when compared against controls (Ludwig et al, 2018). This suggests that beyond the number of life events individuals experienced, there may be additional factor(s) for those with FND. For example, other elements that have been identified as perpetuating factors in NES are family difficulties, depression, and anxiety (Reuber et al, 2007). Therefore, if stressful factors, such as life events, are considered to be one of the perpetuating factors of NES, it seems important to investigate this especially if poor emotional processing is implicated by models of NES.

A study by Roelofs et al (2005) investigated life events in those with FND (reported as conversion disorder). They focussed on investigating recent life events that may be considered as ‘symptom eliciting factors in the light of retraumatization’ in those individuals with a history of abuse in childhood. Recent life events were defined as those experienced in the 12 months before symptoms began. They found that participants with FND (N=54) did not report more life events when compared to reports from controls (N=50). However, recent life events were reported to be significantly associated with more intense ‘conversion symptoms’. From this it could be postulated that recent life events may act more as maintenance factors, with historical life events acting as causal factors toward developing NES. Roelofs et al (2005) also found that certain life events were associated with the number and severity of FND symptoms. Life events related to work or close relationships were associated with experiencing a high level of symptom associated distress. It was also reported that a ‘relation between recently experienced life events and the number of’ FND symptoms was found.

A qualitative study explored the life experiences of individuals with NES in South Africa (Pretorius and Sparrow, 2015). The study highlighted that psychotherapy may be a path towards improving the quality of life in those with NES (Brown et al, 2011; Myers et al, 2012). It was also highlighted that most of the psychological literature is based on studies conducted in the West, of which the findings are ‘transferred directly to the people of South Africa, with little consideration for the cultural and historical implications’ (Pretorius and Sparrow, 2015). For example, a theme reported in this study related to the use of religion and spirituality in coping. This is not a theme that has been reported by other studies in the literature around NES and life events. Such findings demonstrate that inclusion of research in different contexts can strengthen the evidence base towards understanding how individuals cope with living with NES. However, this study is not without criticism. Whilst Pretorius and Sparrow (2015) included four (out of 10) participants from a dual heritage background, the study authors used outdated terminology to describe these participants (‘coloured’). This raises the question of how the authors themselves consider patients from Black backgrounds or other ethnicities, and how representative this study is of the population of South Africa, based on who may be diagnosed and treated for NES in South African clinics.

Life events have been suggested as one factor that may contribute to maintaining NES, based on the speculation that individuals with NES employ poor coping strategies when faced with stressful life events. For example, when those with NES are compared to those with epilepsy or healthy controls, individuals with NES were found to report higher rates of distress connected to ongoing stressful life factors pertaining to work, social functioning, health and legal difficulties (Testa et al, 2012).

Due to theories of NES indicating that traumatic experiences are likely to have a predisposing and precipitating role in the condition, the life events literature in NES is concentrated mostly on life events in early life (e.g. Alper et al, 1993), or life events around the onset of symptoms (e.g. Bègue, et al, 2021). Therefore, from the existing literature of life events research in FND and NES, it is difficult to gain a clear understanding of how life events (LE) may act as perpetuating components in the disease.

Emotional experiences and non-epileptic seizures

The way in which an individual responds and copes with stress may dictate whether a stressor acts as a perpetuating or contributing component to an individual’s psychopathology (Myers et al, 2013). A study by Myers et al (2013) investigated coping strategies in pwNES using a battery of questionnaires including the self-rating scales the Coping Inventory for Stressful Situations (CISS, Endler and Parker, 1990), and the Toronto Alexithymia Scale (TAS-20, Bagby et al, 1994). It was hypothesised that pwNES would use more avoidant type strategies, which would be linked to ‘maladaptive psychological features’ (Myers et al, 2013). Results from the study indicated that a third of the sample reported using ‘self-oriented stress reduction approaches’. Such strategies are thought to be inadequate to deal with stressful situations. These are approaches that refer to emotional reactions which are focussed on the individual, such as angry outbursts, fantasising and self-preoccupation. Myers et al (2013) describe task orientated approaches towards stress to be the ‘healthiest’ strategies to cope with stressors. Their findings (Myers et al, 2013) of decreased use of task orientated strategies was consistent with the findings from other NES studies (Testa et al, 2012; Frances et al, 1999; Goldstein et al, 2000). It was also reported that task-oriented strategies were negatively associated with depressive symptoms, and they suggest that the reduction of symptoms may be the effect of resolving problems (Myers et al, 2013). From such studies it can be hypothesised that pwNES may be more likely to employ unhelpful or ineffective strategies to manage stress or emotion, compared to the general population (Myers et al, 2013). Another such strategy that is considered ‘dysfunctional’ is rumination (Aker et al, 2014). This emotion regulation strategy is reported to be a common method used by pwNES and is considered to denote difficulties with emotion regulation and poor coping in stressful situations (Tojek et al, 2000). It has been reported that compared to controls, pwNES report higher levels of rumination and this indicates poor coping with stress (Tojek et al, 2000). A study by Whitfield et al (2020) compared pwNES to participants with epilepsy on levels of rumination, investigated in the form of ‘repetitive negative thinking (RNT)’, catastrophising, anxiety and depression. The study reported that pwNES reported higher levels of catastrophising, RNT, anxiety and depression when compared to participants with epilepsy. Associations between RNT and catastrophising were found to be correlated in both groups, although a stronger association was found for participants with NES. Also, a diagnosis of NES was found to independently predict RNT. Therefore, Whitfield et al (2020) highlight that rumination and catastrophisation should be considered as potential areas of significance with regards to making psychological interventions more focussed to the needs of pwNES.

A review of the qualitative literature, pertaining to experiences of living with NES (Rawlings and Reuber, 2016), reported that participants used unhelpful or dysfunctional strategies (Fairclough et al, 2013) to regulate emotion through avoidance or suppression. The review also found participants made links between their ‘emotional states and the manifestation of symptoms’ (Fairclough et al, 2013; Pick et al, 2016), and participants showed ‘insight into their methods of processing emotions’ (Pick et al, 2016). Rawlings and Reuber (2016) highlight that there appears to be an ‘over-reliance on escape-avoidant coping’ in pwNES (Frances et al, 1999; Testa et al, 2012; Myers et al, 2013; Dimaro et al, 2014). They (Rawlings and Reuber, 2016) suggest that psychotherapy may be useful for this, since it has been shown to be a successful intervention in helping individual’s develop better strategies to cope with emotions (LaFrance et al, 2013).

One of the several themes reported by the review was regarding the seizure experience (Rawlings and Reuber, 2016). Participants described different experiences of emotion at the time of the seizure. Some individuals described experiencing no emotion during the seizure, whereas others were clear that the episodes had a ‘negative “horrible” emotional impact’ for them (Pick et al, 2016; Thompson et al, 2009). Studies indicated that pwNES spoke differently about seizure episodes, compared to those with epilepsy (Rawlings and Reuber, 2016). This was demonstrated by the different metaphors used by the two seizure groups. Participants with NES reported themselves to be an ‘actor’ in the seizure experience, with some control or influence on their behaviour during the episode, whereas those with epilepsy described the seizure to be an external force (Plug et al, 2009) and a ‘threat’ against which they had to fight (Cornaggia et al, 2012). The review by Rawling and Reuber (2016) also found that in relation to their seizures, participants described feelings of ‘shame, stigma or embarrassment’ (Fairclough et al, 2013; Karterud et al, 2010), although not all participants demonstrated this (Wyatt et al, 2014).

Pick et al (2016) investigated the emotional experiences of pwNES and whether they identified a link between the onset and continuation of seizures with their emotional functioning. Interviews were carried out with 15 participants to investigate how individuals perceived their emotions in relation to their seizures. Interpretative Phenomenological Analysis (IPA) was used to analyse the data. The IPA method is focussed on interpreting the individual experience to explore the meanings ‘implicit within the data’. In exploring how participants considered their emotions to be linked to seizures, several themes were developed (Pick et al, 2016). These included: ‘inhibited experience and expression of negative affect’; ‘abuse’; ‘emotions linked to physical feelings’; ‘stressful/adverse life experiences or circumstances’. The findings were interpreted to suggest that individuals with NES showed some insight into their emotion processing styles and expressed difficulties with managing emotion. Pick et al (2016) posited that emotion triggers a reaction at a preconscious level and this triggers ‘previously neutral cues that have been associated with trauma related affect.’ This could be conceptualised as a ‘somatic marker’ (Damasio, 1994), or a mistaken hypothesis as in the Integrative Cognitive Model (Brown and Reuber, 2016), which leads the individual to experience a seizure.

Qualitative research in NES

Qualitative research in NES has been limited in its focus. Studies have focussed on how patients perceived the diagnosis and treatment of NES (Carton et al, 2003; Baxter et al, 2012; Fairclough et al, 2013; Dickinson et al, 2011; Wyatt et al, 2014), or have explored the experience of living with NES (Pretorius and Sparrow, 2015; Rawlings et al, 2017). One study with a NES sample investigated individuals’ perceptions of emotions and stressful experiences (Pick et al, 2016).

As mentioned earlier, a review of the qualitative research in NES was conducted by Rawlings and Reuber (2016). The review focussed on studies that described the experience of living with the phenomenon and reported several themes. The themes included those that related to patients’ experiences of the seizures, diagnosis, and support from healthcare professionals (HCP), and how NES affected their everyday lives (Rawlings and Reuber, 2016). Rawlings and Reuber (2016) suggest that if NES are considered from a biopsychosocial perspective (such as the Integrative Cognitive Model: ICM, Brown and

Reuber, 2016), the current evidence indicates a multitude of predisposing, precipitating and perpetuating factors. As noted in an earlier chapter, within the mix of these factors some pwNES are reported to show higher levels of alexithymia and other emotional processing deficits (Brown et al, 2013; Novakova et al, 2015).

The review (Rawlings and Reuber, 2016) found that stress and trauma were linked to past events, and for some patients these were considered to be linked to the aetiology of NES (Carton et al, 2003; Karterud et al, 2010; Pick et al, 2016; Wyatt et al, 2014). However, this was not replicated by all studies in the review, because it was also reported that some patients did not respond positively to a psychogenic explanation for NES. This was based on their perception of not having faced difficulties in their past that would have caused a stress/trauma-based development of NES, although, Fairclough et al (2013) highlighted that these beliefs were mismatched for some participants – based on a comparison with their responses to self-report distress and trauma questionnaires. Similarly, some patients 'made contradictory remarks making both psychological and organic references when discussing aetiology' (Green et al, 2004; Wyatt et al, 2014). Ongoing 'psychosocial difficulties' were expressed as difficulties with relationships, work and health (Fairclough et al, 2013; Pick et al, 2016; Thompson et al, 2009). The studies here perhaps illustrate the complex nature of seizure experiences, which makes it difficult for individuals to identify and make sense of which factors may contribute to –'trigger' seizure events.

The theme relating to diagnosis highlighted that upon receiving a diagnosis, participants described feeling vindicated and relieved (Thompson et al, 2009). After receiving the diagnosis feelings of 'disbelief' and 'confusion' were also reported frequently by the studies (Green et al, 2004; Wyatt et al, 2014). Additionally, following diagnosis there were experiences that made individuals feel that they were left in "limbo", feeling that they had been abandoned by services (Thompson et al, 2009; Wyatt et al, 2014). Both these studies (Thompson et al, 2009; Wyatt et al, 2014) were conducted in the UK, indicating that participants had been left with a poor impression of healthcare services. Whilst these examples are not of emotions per se, they highlighted the reactions of participants to events associated with their diagnosis of NES. Such feelings may have a longer-term effect on individuals if negative impressions of healthcare services are held, possibly impacting their motivation to seek support for NES if needed.

Experiences with HCPs were found to be both positive and negative. Positive participant experiences, such as professionals providing patients with the opportunity to ask questions, or professionals being ‘approachable, understanding and attentive’ were expressed by several studies (Dickinson et al, 2012; Fairclough et al, 2013; Karterud et al, 2015; Pretorius et al, 2015; Thompson et al, 2009; Wyatt et al, 2014). However, it was also reported (Rawlings and Reuber, 2016) that negative experiences with healthcare professionals were common (Thompson et al, 2009; Green et al, 2004).

The ‘impact of PNES to daily life’ was also highlighted by the studies reviewed (Rawlings and Reuber, 2016). Here aspects considered to influence one’s quality of life were described. There were multiple issues highlighted, for example, feeling that the seizures were burdensome to them and those around them, or grieving over not being able to work because the value of work was not limited to financial income (Fairclough et al, 2013; Pick et al, 2016). A loss of independence and freedom was a common experience and relinquishing one’s driving licence contributed to these experiences (Baxter et al, 2012; Green et al, 2004; Pretorius, 2016; Pretorius and Sparrow, 2015). Loss also in relation to ‘privacy’ was reported, due to some participants having to always have someone with them as a safety measure (Pretorius, 2016; Pretorius and Sparrow, 2015). A study that investigated the lived experienced of NES via written accounts (Rawlings et al, 2017), reported themes that included ‘self-worth’ and ‘managing emotions’. In the theme ‘self-worth’, participants indicated ‘a lack of compassion’ by describing themselves as ‘“useless”, “pathetic” and a “waste of space and money”’. In relation to ‘managing emotions’, participants expressed using similar strategies to those mentioned by Rawlings and Reuber (2016) above. That is, supressing or avoidance of emotions. This was because participants felt overwhelmed by emotions or that they may be “harmful” (Rawlings et al, 2017). Difficulties with expressing and regulating emotion was also described.

Rawlings and Reuber (2016) commented on the limitations of the studies included in their review. That is, the studies were conducted in Europe, North America and South Africa, making generalisability limited. They similarly commented in relation to study samples being predominantly female. With these criticisms in mind, they suggest that studies in different cultural/societal contexts and with more male participants are needed.

As highlighted by the studies above, recent life events may have a maintenance role in NES From the existing studies on life events research and qualitative research with NES samples, it appears that there is a gap in the understanding of how pwNES perceive the impact of recent life events, and whether these events are contributory factors towards the maintenance of the condition. To our knowledge, no qualitative study has been conducted to explore the impact of recent LE in adults with NES, and the emotional responses to such events. Since there is a concern with understanding the impact and meaning for individuals, IPA appears to be an appropriate method to explore this.

4.2. Aims and objectives

This study aimed to explore how individuals with NES responded emotionally when they experienced a recent life event. The study aimed to provide an opportunity for participants to express in their own words, how they responded to the life event, and the extent to which they perceived their emotional response to have an impact on seizure episodes.

The aim was to investigate:

How do individuals describe and make sense of their emotional responses to a recent life event and to what extent do they perceive this to have an impact on their episodes of NES.

4.2.1. Objectives

To explore how individuals with NES perceived their response to recent life events; by investigating:

- How did individuals with NES experience and understand their emotional response to a specific life event that occurred over the past 12 months?
- To what extent did individuals with NES perceive any connection between their emotional response to a life event and seizure episodes?
- How did individuals with NES express their emotions at the time of a life event?

4.3. Methodology

Interpretative Phenomenological Analysis

This study used interpretative phenomenological analysis (IPA). This is a qualitative and experiential approach with its epistemological foundations in three approaches: phenomenology, hermeneutics, and idiography (Smith et al, 2022). The three approaches are briefly introduced below to outline how IPA is constructed.

Phenomenology refers to the study of the lived experience of the individual: how they exist and how they experience a phenomenon (Larkin and Thompson, 2012). Phenomenology is not only concerned with the experience of the matter under investigation but is also concerned with meaning. The role of the researcher is to highlight the main features of the phenomenon, as experienced by participants (Peoples, 2020). The researcher aims to understand how the participant experiences the world, their reality (Willig, 2012). The final aim, therefore, is to ‘bring together the objective and subjective dimensions of experiences as lived’ (Peoples, 2020).

Idiography and hermeneutics are also foundational concepts in IPA. Heidegger and Merleau-Ponty are the main philosophers to which hermeneutics is attributed. Idiography is focussed on detail, the specific, the individual, in order to analyse the phenomenon being researched in depth (Smith et al, 2022). This does not mean that idiography is not concerned with generalisations explain Smith et al (2022), ‘but rather prescribes a different way in establishing those generalizations (Harre, 1979)’.

Heidegger and Merleau-Ponty (Larkin et al, 2006) shared the perspective that individuals are completely immersed in the world and relationships; that individuals are ‘embodied’ in their worlds (Merleau-Ponty, 1962). These are factors that influence how the individual perceives their world (Larkin et al, 2012). Larkin et al (2012) explain that this allows the approach to move from being descriptive (phenomenology) towards being interpretative (hermeneutic).

Following on from the philosophical approaches highlighted above, IPA, therefore, is proposed as ‘phenomenological psychology’. IPA is focussed on meaning, exploring the individual’s experience and perception of the phenomenon (Eatough and Smith, 2006). The phenomenon or causes of events are not the focal point, but rather the meaning attributed to the phenomenon (events, objects and such) in the participant’s life, and the significance of these phenomenon for the individual. In this way, IPA demonstrates a ‘commitment’ towards being idiographic in its approach. IPA concerns itself with the individual, rather than the general aspects of the experience (Larkin et al, 2012).

Eatough and Smith (2006) highlight that IPA is most definitely a psychological approach given its concern with meaning, understanding the motivations behind behaviours, and the way in which participants think about events. The researcher focuses on the language used by participants to convey their meanings and experiences. It is suggested that the individual’s accounts of their emotional experience, are not simply expressions of emotion, but these accounts reflect the way in which individuals make sense of their relationships and connections between the different time points in their lives (Eatough and Smith, 2006). The participant is embodied in the expression and meaning making of their life. The language, emotions, objects and such used to express themselves hold meaning. Smith and Osborn (2003) explain that the researcher is involved in a two-fold process whereby the researcher is not only concerned with capturing how the participant conveys their understanding of their world, but the researcher is also making sense of how the participant understands their experiences. This is referred to as the double hermeneutic. To not only try to understand how the participant experiences and understands their world, but to interpret the meaning the participant is trying to convey. The researcher aims to understand the intricacies of the meaning being conveyed by the participant, rather than focusing on frequencies. That is, IPA looks beyond the superficial meaning that may be present in the data.

Interpretative phenomenological analysis shares features with other qualitative methods such as thematic analysis. Thematic analysis seeks to code data into groups/ categories/ patterns that bring the pieces together under some common theme, and in this way a thematic approach is not dissimilar to IPA (Figgou et al, 2015). The IPA methodology is useful for those research questions in which emotion is key (Eatough and Smith, 2017). The IPA approach is focused on the ‘personal lived experience’ and whilst it is not possible to capture the ‘pure experience’, the aim is to get as close as possible (Nizza and Smith, 2021).

It has been highlighted that IPA is analytic and interpretative in its approach (Eatough and Smith, 2006; Larkin and Thompson, 2012). For this reason, IPA has been described as an ‘integrative approach’ (Smith et al, 2009) because whilst the ‘analytic interpretation’ has its foundation in the participants’ account, it looks beyond how the participants’ understand their experiences.

Smith et al (2009) outline three principles that an IPA study should have at its foundation:

1. The study should be interested in the participant’s engagement with phenomenon, i.e., ‘the participant’s relationship with something that is of importance to them in their world, as they experience and understand it.’
2. The data undergoes a deep interpretative analysis.
3. The researcher considers the data for each participant, within its context, in depth during the analysis.

By adhering to these three principles, IPA fulfils the requirements of phenomenology, hermeneutics and is idiographic (Smith et al, 2009).

IPA is descriptive, as well as interrogative and questioning – pulling out multiple meanings from the data, allowing the researcher to maintain the ‘richness’ of the individual’s experience (Smith et al, 2009). Therefore, IPA was considered to be an appropriate method for the current study.

4.4. Methods

4.4.1 Study design

A semi-structured interview is most commonly used to collect data appropriate for IPA research (Smith et al, 2009). This format is thought to allow the researcher the opportunity to collect data that is ‘rich, detailed and reflective’, and quality rather than quantity is key in IPA (Smith et al, 2012). It is suggested that transcribed transcripts of the participant’s account are used for the analysis (Smith et al, 2012). The researcher, therefore, used one to one, semi-structured recorded interviews to collect the data.

4.4.2. Sample size

Smith et al (2009) suggest that when using IPA, a sample size of around 3-6 participants is sufficient. Studies investigating NES, that have also used IPA, have varied in sample size from 6-15 participants (Green et al, 2004; Pick et al, 2016; Thompson et al, 2009; Wyatt et al, 2014).

Clarke (2010) suggests that sample sizes of around three participants are unlikely to provide the relevant experience for students to ‘reflect on and develop’, and therefore advises for those completing a study as part of a doctorate, to aim for a sample size of 4-10 participants. The researcher aimed to recruit 8-10 participants. The final sample consisted of 10 participants.

4.4.3. Sampling strategy

A purposive sampling approach was taken. This is described as choosing those participants that will generate appropriate data (Green and Thorogood, 2006); meaning that participants were selected based on having characteristics that met the aims of the research. Therefore, participants with NES were selected on the basis that they had experienced a life event in the last 12 months.

4.4.4. Inclusion/exclusion criteria

Inclusion criteria:

An adult (≥18 years of age), in the UK with a diagnosis of NES;

Must have experienced at least one life event in the last 12 months;
Able to provide informed consent;
Able to read and speak English, Urdu or Punjabi. These languages were specified because these are the languages that the researcher speaks fluently.

Exclusion criteria
≤17 years of age;
Did not have a diagnosis of NES;
Had not experienced any life events in the last 12 months;
Expressed that they had recently self-harmed or expressed suicidal plans or intent;
Were unable to provide informed consent;
Had a co-morbid diagnosis of epilepsy, dementia or other neurological disorder;
Were unable to read or speak English, Urdu or Punjabi.

4.4.5. Ethical considerations

Advertising of the study included the researcher using her own social media accounts to promote the study. For the purposes of the study advert, the post was made ‘shareable’, and the researcher’s Facebook account was set so that only accepted accounts could view Facebook activity, to maintain a level of privacy. The researcher’s Twitter account was also used to advertise the study. The researcher’s private contact details (telephone or email) were not available on either of the social media accounts. Potential participants were able to contact the researcher via her university email address, or direct message through Twitter. Due to the Covid-19 lockdowns requiring the researcher to work from home, she was unable to use her office telephone. Therefore, when the researcher was required to contact a potential participant using her personal mobile, the researcher would explain to the individual that she would withhold her telephone number.

Although the participants were not interviewed face to face, there was the possibility that individuals could have a seizure during the interview. For this reason, a risk protocol was designed. The questions were not designed to induce seizure activity, but the unpredictable nature of NES meant that there was a possibility that a participant would have an episode during the interview. Therefore, at the start of the interview the researcher asked the participant if they had any warning signs before a seizure. Those participants that could describe warning signs were asked to let the researcher know if they began to experience them. The plan was to cease the interview if the participant began to feel that a seizure was imminent. Participants were also asked if there were other people in their house at the time of the interview, in case the individual did have a seizure and then the family member/partner/carer would be aware they needed supervision. In instances where participants lived alone, the risk protocol indicated that an ambulance would be called by the researcher.

The interview was not designed to induce distress and so the risks were considered to be minimal. However, it was considered a possibility that participants may report feeling upset when answering questions relating to their health. Although one or two participants showed that they were upset at some points when talking about difficult events, no participant expressed feeling distressed and wanting to stop the interview, which of course was an option available to them.

Data was stored in accordance with the University of Leeds data storage policy. Demographic forms, consent forms, interview audio recordings and transcripts were stored on the secure OneDrive. Pseudonyms replaced participants' real names, regional locations were used rather than exact towns and cities, and jobs or course names were generalised to offer some level of anonymity and maintain the confidentiality of participants. Where participants mentioned others (e.g. partners/family members/friends) by name, the name was replaced with the relationship.

A final ethical consideration was remuneration. Participants were offered the option to receive a gift voucher after participating in the study. This information was included in the study advert and offering a voucher as a 'token of appreciation' to participants is within the university's ethical guidelines.

4.4.6. Ethics approvals

Ethical approval was obtained from the University of Leeds, School of Psychology Ethics Committee (SOPREC): Ref PSYC-51, Date 19-05-2020.

4.4.7. Data collection

Participants were required to complete a demographic questionnaire. This was used to collect sample characteristics, such as: age, ethnicity, occupation, onset of NES and when they received a diagnosis. (Appendix O). Due to the COVID pandemic, all interviews took place remotely. Nine interviews were conducted online, and one over the telephone. Interviews were audio recorded and transcribed by the researcher at a later date.

A semi-structured interview was used to interview participants (Appendix P). The researcher used a series of prompts to aid the participant to express themselves, share their experiences and their perception of how they responded at the time of the LE. The schedule had been developed through discussion with supervisors with the focus being how best to phrase questions that would generate discussion and avoid using closed questions. The interview scheduled was tested with a friend before the first interview was conducted. This was to test the flow of questions and whether the prompts were helpful. After the first interview proper was conducted, the researcher transcribed the interview and shared the transcript with her supervisors. This was to allow for a discussion about the appropriateness of the data collected and is a step suggested by Smith et al (2009). No refinements were made to the schedule at this stage.

In addition to the interview schedule, a timeline was used. This acted as an aide-memoire to help participants recall a life event(s) that occurred over the last 12 months, and whether any of these events coincided with times that they experienced a non-epileptic seizure. Each life event was plotted on the timeline and discussed in turn, starting with the most recent event that had occurred. The interview therefore progressed by the researcher referring to an event on the timeline and exploring the event using the questions from the interview schedule. This procedure was followed for each event, with the researcher highlighting that the participant may find the questions repetitive.

At the end of the interview participants were thanked for their time and asked if they consented to the researcher contacting them again, should any follow up questions arise. This was required for one participant only. This was the first participant interviewed. After the interview transcript was shared with supervisors, the researcher identified follow up questions with regards to a LE that was not discussed.

The final task was to ask participants which voucher they would like to receive. A £20 voucher was emailed to participants as a ‘thank you’ for their participation.

4.8. Analysis

This study used Interpretative Phenomenological Analysis (IPA: Smith et al, 2009) to analyse the interview data.

The focus of the approach is to learn about the individual’s perceptions and beliefs: their ‘psychological world’. Therefore, IPA is a two-stage process whereby the participant attempts to make sense of their world, followed by the researcher attempting to understand the participants' views (Smith et al, 2022).

The analysis itself is multi-layered, starting with the individual themes (Personal Experiential Themes: PETs) and ending with a list of themes representing the whole sample (Group Experiential Themes: GETs). Steps outlined by Smith et al (2022), combined with guidance from a consultant experienced in IPA, were used to analyse the data. The process is outlined below:

1. The researcher examined each transcript in detail, conducting a line-by-line analysis. The first reading was carried out whilst listening to the audio of the interview. The next reading was to look for repetition, emphasis on certain words, metaphors, tone. The transcript was annotated with comments from the researcher.
2. Further readings of the transcript allowed for annotations to be turned into themes. That is, moving from the descriptive towards looking for meaning for the participant.
3. The next stage involved a discussion with the consultant about impressions and ideas.
4. This led to developing the themes informed by the discussion.
5. Next, similar themes were clustered together.
6. Data relating to life events was extracted from the transcript, to see if anything pertinent was missed such as patterns in the data that may show understanding of events related to seizure experience, current events and future plans.
7. The groups of clusters were then named/labelled to reflect the participant's experience, developing the Personal Experiential Statements (PETs) for the participant.
8. Steps 1-7 were repeated for each participant. A table of PETs and subthemes was constructed for each participant. As part of the iterative process, these themes were refined and restructured as the analysis progressed.
9. All 10 tables of PETs, that is one table from each participant, were examined without any quotes. This was to identify any patterns, common themes, or divergence.

10. PETs were separated so that clustering could be achieved of similar themes across the sample.
11. These clusters were populated with extracts from participants and developed into Group Experiential Themes (GETs). This was an iterative process that involved steps such as those taken at 4 and 5: discussion and refinement.
12. The GETs were ordered to reflect the overall narrative that the sample appeared to tell.

At stages 2 and 3, several strategies were used to explore the data and improve the analysis, for example using spider diagrams to identify the different aspects and evidence for a specific theme such as anxiety. See appendix Q for examples.

4.8.1. Rigour in analysis

It is suggested that it is ‘good practice’ to try to achieve rigour in the analysis. This can be attained by taking steps to improve the reliability and validity of the analysis (Green and Thorogood, 2006). The researcher attended two workshops. The first was a one-day course designed to introduce the theoretical background of IPA, as well as providing exercises to gain practical experience of using IPA (Getting Great Data: introduction to IPA). The second workshop was an in-depth workshop which was described as an ‘advanced’ workshop for IPA analysis.

4.8.2. Internal reliability

It has been suggested that in qualitative research reliability is difficult because it is often interpreted to mean ‘replication’ (Spencer and Rithcie, 2012), and therefore ‘consistency’ may be more appropriate. That is, whether other researchers would describe/pinpoint the same themes/concepts, or whether different categories are identified (Spencer and Ritchie, 2012).

To ensure internal reliability, i.e. consensus between the researcher and supervision team, the researcher attended supervision meetings as analysis progressed, and had regular meetings with a consultant, well-versed in IPA, for feedback on the process. Once the first version of GETs were developed, the researcher shared the analysis, and discussed with the whole supervision team.

4.8.3. External validity

It has been suggested that joint working between those who experience the phenomenon/use services, and researchers/practitioners, can lead to a 'joint agreement about the meaning of the data' and may improve the validity of findings (Faulkner, 2012). Possible strategies include seeking feedback from participants or similar samples. IPA researchers do not always support the use of this strategy because the nature of IPA, with its focus on researcher interpretation, sometimes leads to the development of themes that do not resonate with participants (Smith et al, 2009) and, in the circumstances, it was decided against.

4.8.4. Reflexivity

The researcher considered their position within the process of conducting the study. A reflexive statement is shared in Appendix S.

4.9. Results

In this section, first details about the sample are presented: demographics, seizure types and associated symptoms experienced by participants (table 13 and 14). A table of life events is also presented (table 15). Italicised words show where a participant's own words have been used in the text. Where a participant paused during speech, this is shown by ….. Where an extract has been reduced for clarity and brevity, this is illustrated using […].

A vignette, in which pertinent information about each participant is presented for each of the 10 participants. The timeline of events for each participant are presented in each case study. The timeline illustrates the events that occurred for the participant over the 12 months before the interview. These events are signposted using green arrows → . In cases where participants mentioned events that occurred outside of the 12 months of interest, these are depicted using blue arrows → .

The group analysis makes up the remainder of the results presented here; beginning with the table of Group Experiential Themes (GETs: table 16) for the sample. Each of the GETs, and the group level subthemes, are presented with the associated evidence from participants in the text. This is followed by a section illustrating the different models that participants demonstrated, when describing seizures and their emotional responses to an event around the time of the seizure(s).

It is not a requirement in IPA for data from all participants to be relevant to all GETS because the group analysis is not focussed on presenting an 'average' of the phenomenon but rather it is about highlighting the shared aspects expressed by participants, as well as those features that are divergent (Smith et al, 2022).

4.9.1. Sample recruitment

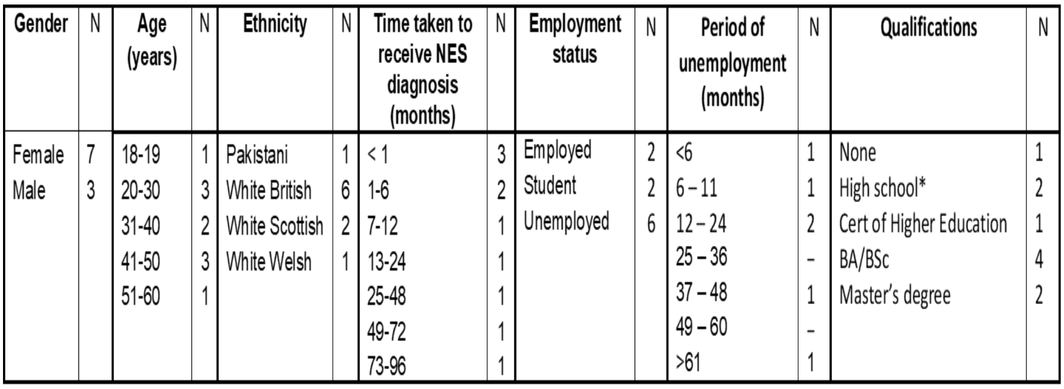
Due to the COVID pandemic, an amendment to the study protocol was made so that all research activity took place remotely. Once ethics approvals were obtained, recruitment took place from June 2020 to 31st October 2020.

Ten participants were recruited. Participants were recruited mostly via the FNDHope UK social media channels (N=5), and then from participants seeing the study on the research page of the FNDHope website (N=3). The remainder of the participants were recruited through the researcher's social media channels (Facebook and Twitter N= 2).

4.9.2. Sample demographics

The sample was made up of 10 participants from the UK. The majority of the sample was female (N=7), White (N=9), between the ages of 20-50 years old (N= 8) and unemployed (N=6). Sixty percent of the sample (N=6) received a diagnosis within one year. For the remainder of the sample it took between seven months to eight years (96 months) to receive a diagnosis. Table 13 contains demographic data for the sample.

Table 13. Participant demographic information



Gender	N	Age (years)	N	Ethnicity	N	Time taken to receive NES diagnosis (months)	N	Employment status	N	Period of unemployment (months)	N	Qualifications	N
Female	7	18-19	1	Pakistani	1	< 1	3	Employed	2	<6	1	None	1
Male	3	20-30	3	White British	6	1-6	2	Student	2	6 – 11	1	High school*	2
		31-40	2	White Scottish	2	7-12	1	Unemployed	6	12 – 24	2	Cert of Higher Education	1
		41-50	3	White Welsh	1	13-24	1			25 – 36	-	BA/BSc	4
		51-60	1			25-48	1			37 – 48	1	Master's degree	2
						49-72	1			49 – 60	-		
						73-96	1			>61	1		

*GCSE or A Levels

4.9.3. Seizure and symptoms

All participants described seizure episodes that involved involuntary movements. Most participants (N=8) described experiencing different types of seizures. These seizure types varied amongst the sample. Dissociation was a common experience (N=8) and half the sample described paralysis during the event.

Over the course of the interview some participants mentioned other physical and mental health conditions that they experienced. Table 14 (Appendix T) displays seizure symptoms and co-morbid conditions described by participants.

4.9.4. Life events

Participants were provided with a list of life events from which they could identify those experienced over the past 12 months, including an option to identify any other life events not mentioned in the list. This list was influenced by the checklist of events used by the Life Events and Difficulties Schedule (Brown and Harris, 1989). In the present study, a life event was considered to a specific or discrete incident (Brown and Harris, 1989) that occurred during the period of interest.

The period under investigation was the previous 12 months from when the interview took place. This time period was selected because the literature indicated that the way in which an individual managed stress may have an impact on whether the stressful factor contributed towards an individual's illness (Myers et al, 2013), and Roelefs et al (2005) had stated that there appeared to be an association between recent life events and the intensity with which functional symptoms were experienced.

In the current study, when completing the life events checklist, some participants identified events that had occurred outside of the period of interest. These are depicted on the timelines as taking place before the study period. The main purpose of the timeline during the interview was to aid recollection of events for the participant.

The life events reported in table 15 demonstrated that participants reported a range of events that they, or those close to them had, experienced over the last 12 months of their life. Overall participants reported 25 common life events versus 26 life events specified by participants themselves. The list was separated to demonstrate the number of 'other events' identified by participants, that is those events that were not present in the preprepared list of events presented to participants. This was to examine whether the type of events specified by participants themselves would indicate a pattern, such as discrimination or health related events. No such pattern emerged.

There was also the possibility that an event may occur multiple times over the timeline for participants. This happened with one participant, Ashley, with regards to attending hospital and the two voluntary roles that she relinquished during the study period.

Table 15. Events experienced by participants over the last 12 months

Life event	**N**
Moved house/ flat	4
Hospitalised	3
Received a new diagnosis	5
Had an accident	1
Family member/someone close to you had an accident	3
Family member/someone close to you hospitalised	2
Family member/ someone close to you die	2
Pet died	1
Made redundant	1
Started a new educational course	3
Other events:	**N**
Started a new job	1
Lockdown	3
Stopped volunteering	2
Surrendered driving licence	1
Traumatic event for family member	1
Change in religion	1
Became a grandparent	1
Began treatment (psychological/physical)	2
Discharged from service	3
Hospital visit	3
First seizure/ seizure after long time/ last seizure	5
Exams cancelled	1
Meeting with manager	2

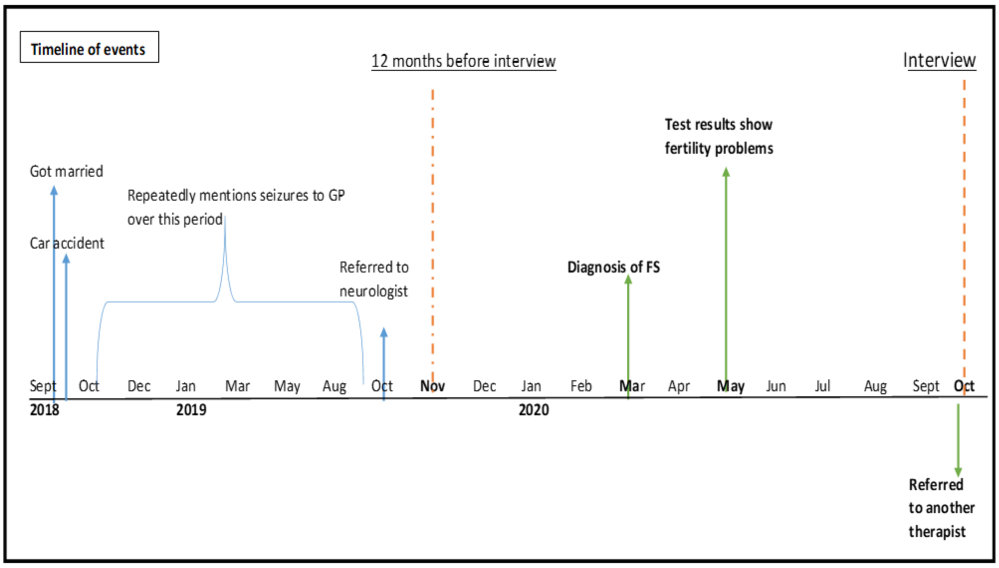
N= number of times the event occurred across the sample

4.9.5. Participant case studies

This section contains a brief introduction, for each of the 10 participants, and includes information pertinent to provide some context for each participant.

Abigail:

Abigail was a White thirty-one-year-old woman from Wales. Abigail had a certificate in higher education and been unemployed for 23 months. She was married in 2018 but had been in a relationship with her husband for over 13 years. Although Abigail wanted to have children, her husband had wanted to have children earlier in the relationship before she felt that she was *ready* to.



Abigail first experienced a seizure 24 months before the interview and received a diagnosis of NES and FND in March 2020. Abigail described different types of seizure events. She considered *drop attacks* to occur in response to her body no being able to manage with emotion but was unsure what precipitated the other types of episodes.

Abigail identified receiving the diagnosis of NES as a life event. During the interview, two other events were discussed: receiving test results relating to fertility and being referred to a new therapist (see the timeline above). Abigail had a car accident (RTA) in September 2018, which placed this event outside of the study period. She referred to this as *her trauma.* Abigail had not felt able to return to the site of the accident. She did not want to talk about the incident during the interview. The accident had left Abigail unable to walk unaided; she now used a mobility scooter outside of the house and sticks inside of the house to walk. Abigail was the only participant in this sample that mentioned having a carer.

Abigail began experiencing seizures in October 2018 and mentioned them to her GP in October 2018. She continued to mention them on multiple occasions to the GP until, she says, the GP acknowledged them – which was a year later. The referral to the neurologist was made outside of the study period (October 2019), but the consultation and diagnosis took place during the study period (March 2020). Abigail also mentioned that since the accident she had been experiencing anxiety and depression and had been receiving treatment for this. Abigail was trying to come to terms with the changes that had occurred in her life due to the RTA, but the information about the impact on her and details about the event are limited. Abigail was involved in a personal injury claim indicating that the RTA was not her fault, or another party was also to blame for the accident.

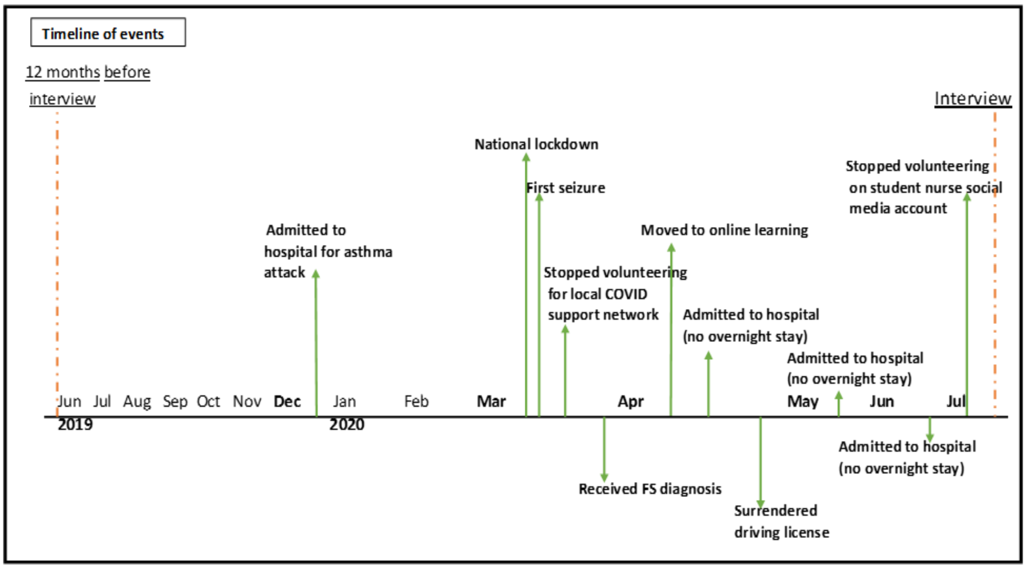
Abigail was diagnosed with fibromyalgia around 5 years ago. She highlighted that this diagnosis was made by a rheumatologist. This seemed an important detail to Abigail, it meant her diagnosis was more definite or authentic compared to those individuals who have been diagnosed with fibromyalgia by a GP.

In emails to the researcher, Abigail mentioned that she had childhood epilepsy. It was established that she was not having epileptic seizures and she said that she had not taken medication for 25 years. This would suggest she had not taken medication, nor had an epileptic seizure since she was 6 years old.

Ashley:

Ashley was a White British female, 28 years old, married and living in northeast England. Ashley was a fulltime student, completing a degree to become a healthcare professional.

Ashley was diagnosed with NES a month after experiencing her first seizure. However, during the interview it became apparent that she had been experiencing minor seizure-related events leading up to the first ‘big’ seizure. Life events identified by Ashley were hospitalised; received a new diagnosis; lockdown; stopped volunteering; surrendered driving license; and moved to online learning for the nursing degree (see timeline). Ashley spoke about grieving after giving up her driving license. She and her husband had also begun to think about moving house so that commuting to her student placements would be easier for her.



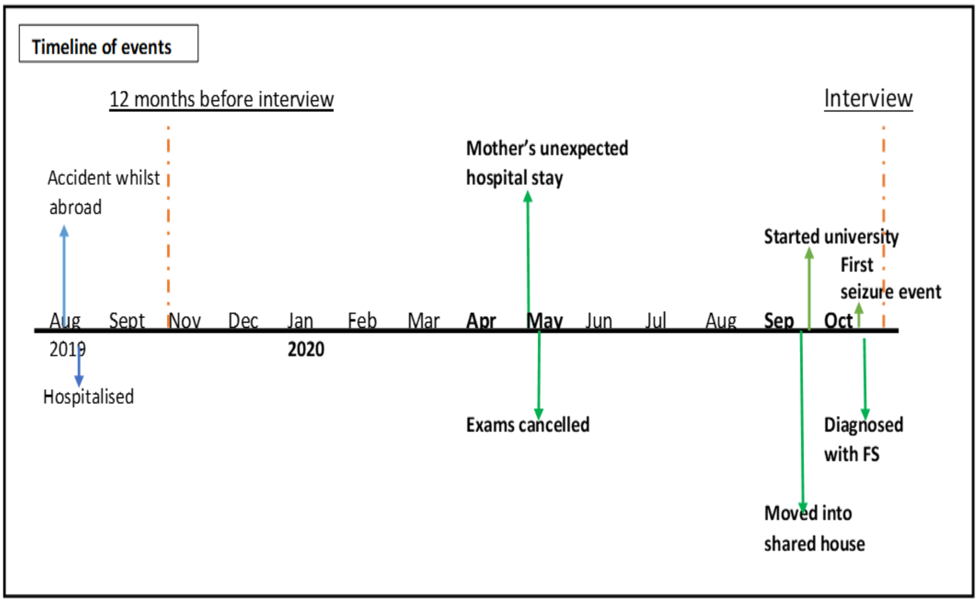
In trying to understand the way in which she reacted to events, Ashley explained that her experiences at school had shaped this. That is, Ashley felt that she needed to prove to others that she was worthy, and this meant having to achieve top marks in her academic work. Ashley described how her husband often helped to offer a different perspective to help her feel prouder of her accomplishments, instead of criticising herself for not doing even better.

Ashley was not typical of the sample because she was both a pwNES and a HCP. Therefore, the impact of NES on her identity was apparent in the way that the life events were relevant to her as a patient and as a HCP. That is, the participant had two identities and therefore perhaps two realities running in parallel. The two realities may reflect how the participant expected her life and training to evolve, versus the actuality of how her life and training were, especially now that she had been diagnosed with NES.

Bryony:

Bryony was a White British, 18-year-old woman. She was a university student studying a humanities degree. Bryony lived with her parents and two siblings (f 10; m 17) until she moved away for university. She had shared a room with a sibling in the family home.

In addition to NES, which was her newest diagnosis, Bryony had several other diagnoses: Ehlers Danlos Syndrome (EDS), Fibromyalgia, asthma, Postural Tachycardia Syndrome (POTS) and Functional Neurological Disorder (FND). These had all been diagnosed over the past two years. Bryony experienced her first seizure episode a couple of weeks before the interview took place. In that time period she had another seizure, meaning these episodes were new experiences for her. She suspected that the seizures were triggered by stress but was still trying to understand if there was a clear pattern to them.



Bryony lived in a shared house with 12 other students. She felt well supported by her housemates; describing how they had been attentive in checking she was ok, and that they were *lovely*. She lived at a commutable distance, so that if she needed to or wanted to go home it was not too far a distance (1 ½ hour journey). She described often calling home to speak to her family, although she contradicted herself by stating that she only called once a week and had not been very homesick.

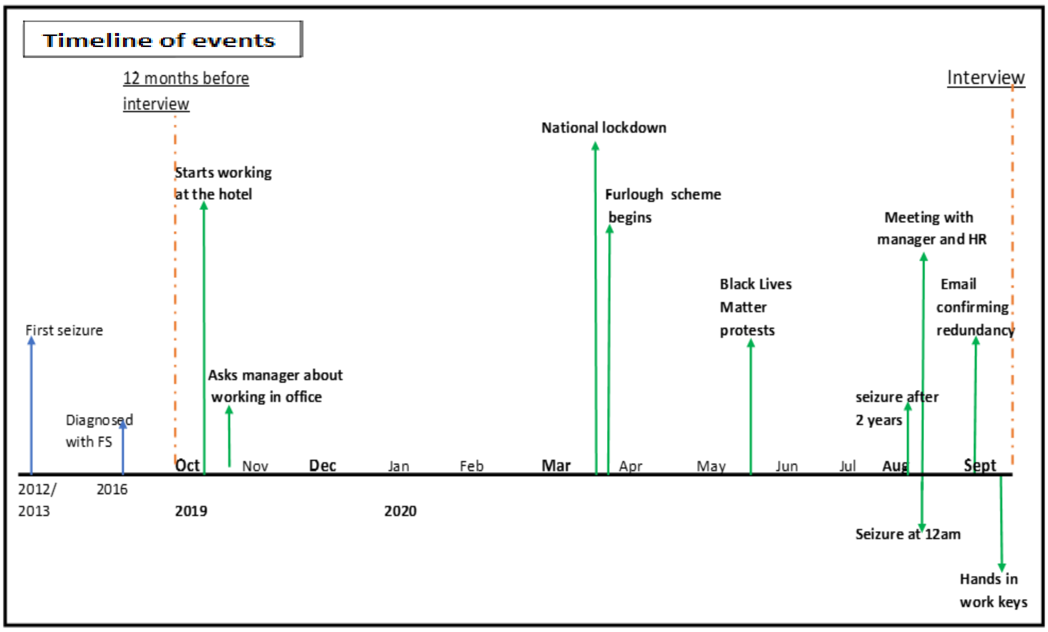
The past two years had brought a number of changes in Bryony's lifestyle and health. An accident occurred whilst she was abroad; she shattered her hip as she was getting out of bed. Bryony used a wheelchair and had spent the last year learning to walk again. This had been impacted by the COVID pandemic due to no face-to-face clinics being offered for some time. The diagnosis of FND came after the accident, as did the diagnosis of fibromyalgia.

Bryony was softly spoken but there were times, especially at the beginning of the interview where she appeared to be speaking quietly. This was possibly because she lived in a shared house and may have been concerned that she would disturb others or be overheard talking about personal incidents. Additionally, the video often froze whilst the audio continued. At times the audio slowed down so that it was difficult to tell if Bryony was speaking slowly or the audio was slowing down.

In emails to set up the date for the interview, Bryony had mentioned that she would find it helpful to take a short break. Therefore a short break of 8 or 9 minutes took place around 40 minutes into the interview.

Chloe:

Chloe was a 24-year-old, White Scottish woman. At the time of the interview she was an unemployed university graduate. She lived in a large city in West Central Scotland but grew up in the countryside. She lived with her partner (m) in a one-bedroom flat. Her family lived around 150 miles away and she did not see them very often.



Chloe mentioned that she had epilepsy as a child but had not had an epileptic seizure since she was 15 years old. These seizures occurred at times when she experienced stress. Chloe described the time between the epileptiform seizures ending and the NES beginning as *blurry*. Chloe highlighted that her mother had observed both types of seizures and described them being different to each other. Chloe also referred to traumatic events in her past. These were when her family were made homeless after living in the same house for 10 years. This coincided with her mother, a single parent, requiring an operation.

Chloe was diagnosed with NES in 2016 but had been experiencing seizures for 3-4 years before this. This suggested that the NES started around the age of 16/17 years old. Chloe referred to the seizures as dissociative seizures because her earlier seizures were dissociative experiences. The seizures that occurred recently had been different and Chloe described remaining conscious throughout. Chloe suggested that stressful events in her life contributed to seizure activity. She explained that whilst she may feel that she was managing the situation, and felt *normal*, a seizure would occur, and this acted a reminder that she had not dealt with the emotions related to the event.

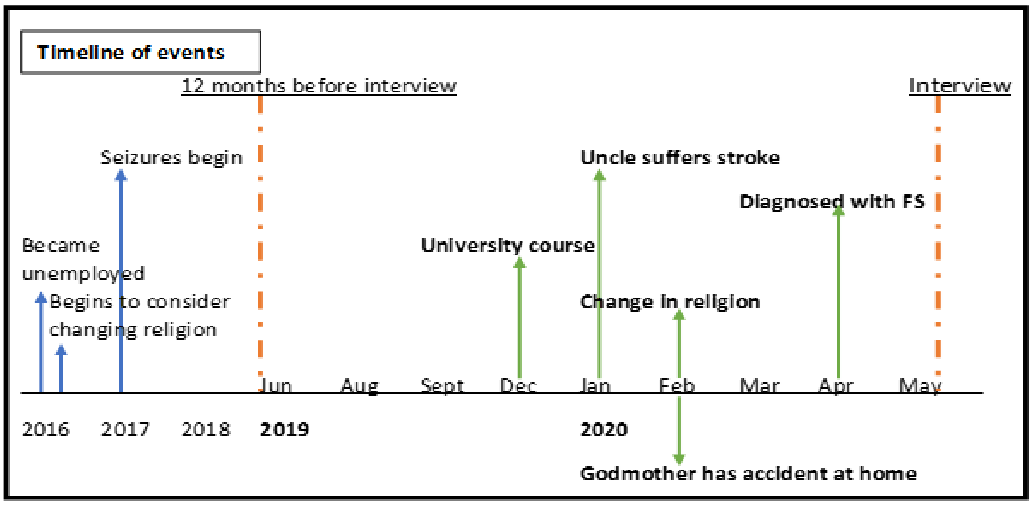
Chloe had been working in a hospitality role for the past year. Because of the COVID situation, Chloe worked for six months before she was put onto the Government furlough scheme. Chloe was made redundant from this job when the scheme reduced the financial help to employers. Being made redundant was the life event identified by Chloe. Other events identified by the researcher, during the time period discussed, related to work events, and the COVID pandemic. Towards the end of the interview, Chloe made reference to the Black Lives Matter movement and the impact that it had had for her.

Chloe also had comorbid diagnoses of asthma, fibromyalgia and borderline personality disorder (BPD). She described herself as being unable to regulate her emotions and experiencing strong and intense emotions. She also mentioned that in the past she had experienced dissociative amnesia for three years.

Craig:

Craig was a 40-year-old, White male from the Midlands. He lived alone with his parents living close by. Craig had been unemployed for four years. He identified himself as a student but was vague about the course he was studying.

Craig was diagnosed with NES in April 2020, although he did not identify this as a life event on the checklist. Craig had stated that he first experienced a seizure around 1-2 years ago; however, during the interview it transpired that he had been experiencing seizures for around 4-5 years. Craig was undertaking his own research to understand the seizures and was convinced that the evidence he had from MRI data indicated that there was likely a structural issue that was causing the pain/pressure in his head and seizure events.



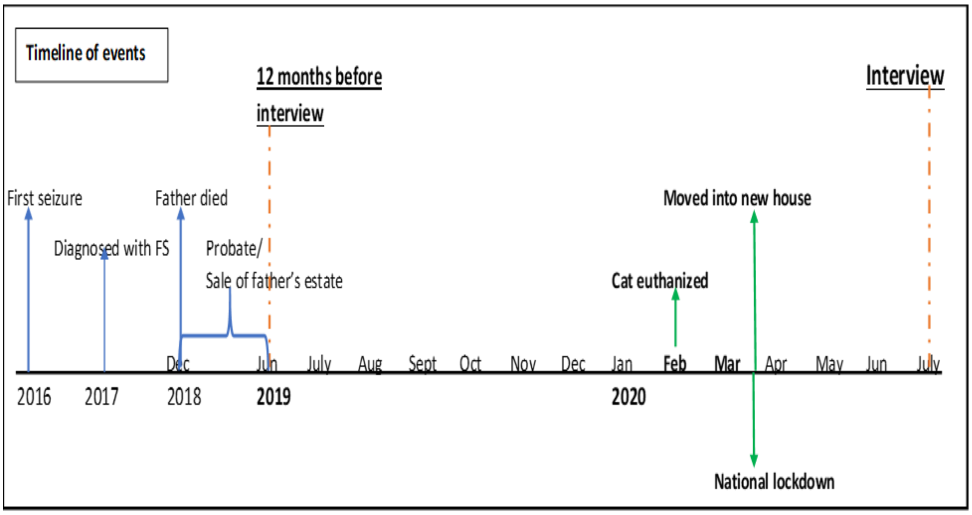
Craig identified four life events that had occurred during the last 12 months: he started a new educational course; his maternal uncle suffered a stroke; his 80-year old godmother had to attend the Accident and Emergency department after an accident in her home; he changed his religion. On discussing the change in religion, it became known that this had been going on for some years. Craig did not change his religion but his denomination. It was not clear whether Craig had become unwell before or after becoming unemployed, or whether his health may have been a contributing factor.

Craig had requested a phone call before the interview was arranged. During this call he shared the warning signs he was aware of before a seizure. He described his face feeling tingly, turning red and his eyes looking bloodshot. He described experiencing a surge of energy during the seizure and then having no energy afterwards, the episode left him feeling exhausted.

During the interview, Craig appeared evasive or angry towards answering some questions around his family. After transcribing the interview, it became clear to the researcher that the event of being diagnosed with NES, had not been discussed. Therefore, a follow-up interview was requested by the researcher to discuss this event.

Mark:

Mark was a White male aged 42 years old. He lived in the South of England with his wife and two daughters under the age of 10. Mark had a postgraduate qualification and was employed in a managerial role. Both Mark and his wife had family living close by.



In addition to a diagnosis of NES, Mark had several other health conditions: chronic fatigue, sleep apnoea and a diagnosis of Generalised Anxiety Disorder.

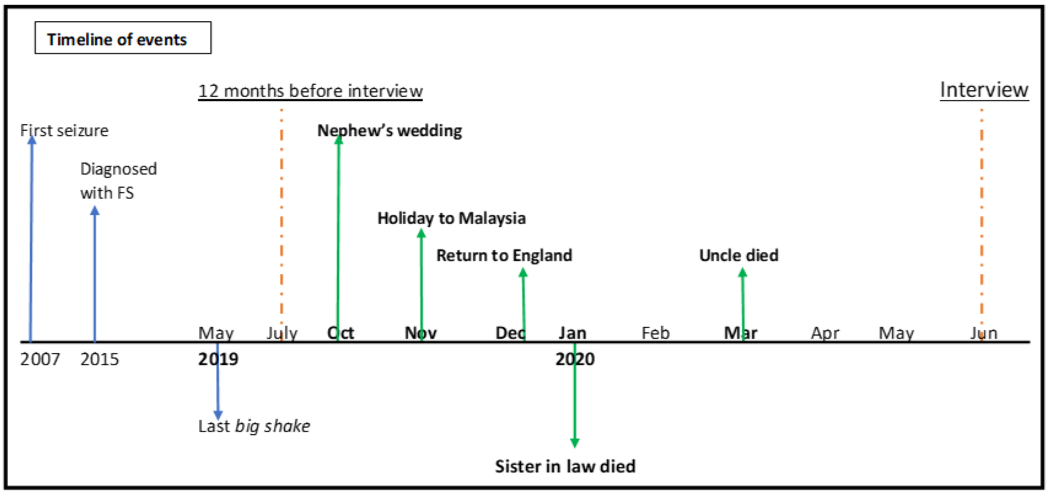
Mark experienced his first seizure in 2016 but was not diagnosed with NES until a year later. Mark expressed that his seizure episodes would not reflect the seizures described by others with NES. He understood NES and FND to be based on a model of *miswiring* in his brain. He described having different types of seizures. One type was characterised by temporary paralysis which could be broken by either Mark managing to move, or by someone else coming and touching his arm. He also had another type of episode in which his legs began to collapse from underneath him, but without falling to the ground. He found it difficult to speak at times and attributed this type of episode to his FND and not as a seizure episode.

His seizure activity had been worse since moving into a new house and had begun to stabilise around the time of the interview. He described moving from a house, in which they had lived for six years, as feeling stressful. Life events that were identified by Mark were: moving to a new house and having a pet euthanised. A third life event – lockdown, was also discussed.

The interview took part during the first national lockdown and his wife and youngest daughter came into the room at different points in the interview. Another life event was identified by Mark– his father's death 18 months ago. This was not discussed because it fell outside of the study period. However, reference was made to this because the house move was made possible partly due to the inheritance received after his father died. Mark spoke about sorting out the will from the perspective of the administrative tasks that had become more complicated than expected. He did not mention any emotion attached to managing this as a result of his father dying.

Parveen:

Parveen was a sixty-year-old, British Pakistani woman from West Yorkshire. She was married and had three grown up children: two sons and a daughter. Parveen lived with her husband and one of her sons. Her other children, both married, lived close to the family home.



Parveen stopped working in the family business six years before the interview, although she was identified as a 'housewife' on the demographic form completed by her husband. She had never been to school but had tried to attend some courses after moving to live in England.

Parveen had spent time thinking about why her seizures occurred but had not been able to formulate an explanation for herself. She described having several tests and discussions with doctors to try and understand why she experienced seizures. She explained that the seizures did not relate to times of stress/anxiety – they occurred *suddenly* at times when she may be anxious, sad or when she felt *normal*. She sometimes experienced some warning signs and referred to a seizure episode as a *shake*.

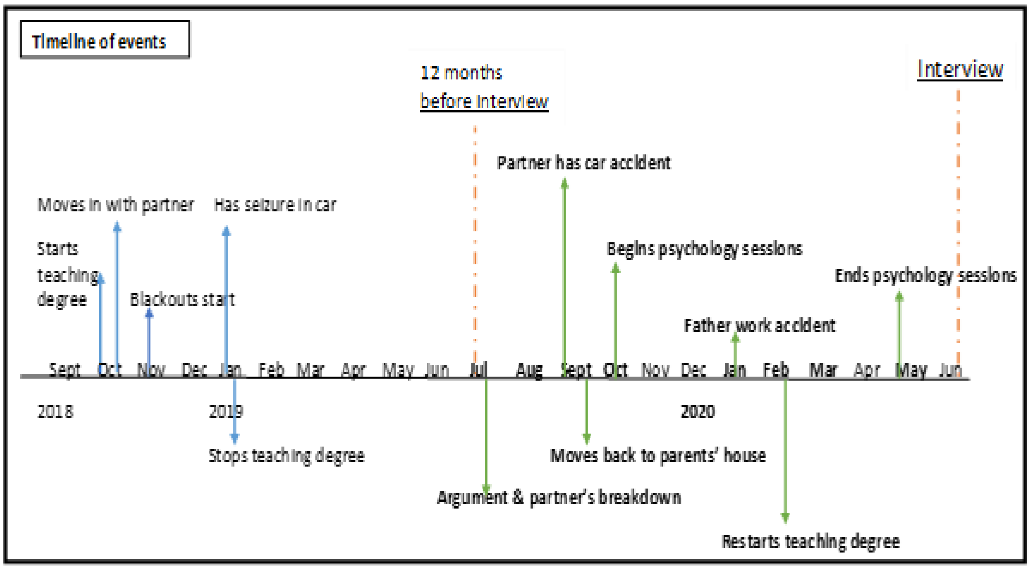
At the time of the interview, Parveen had not experienced a *big shake* for over a year (14 months). She was not sure why the last seizure had happened at that time and said that the doctors could not say why it had occurred either. The seizure took place when Parveen was in hospital being treated for pneumonia. Parveen describes being in hospital all day and waiting to be discharged. It was whilst she and her husband were waiting that she began to feel unwell. She predicted that a seizure was about to occur and then it did.

Parveen spoke about seizures in a manner that suggested that she accepted them as part of her life. The seizures were in the background but were not a defining feature of her. They did not appear to form part of her identity as they did for some participants. This acceptance of the seizures as being part of life could relate to experiencing the seizures for such a long time; or it could be informed by her faith, and she may have considered the seizures as a test to endure in life.

Parveen was the participant with the longest experience of NES, and the participant for whom it took the longest time to receive a diagnosis. Parveen first experienced a seizure 13 years ago (2007) but did not receive a diagnosis until eight years later (2015). Parveen had also been treated with medication but was no longer taking this.

Parveen did not speak or read English, although she did use some English words during the interview. Therefore the interview was conducted in Punjabi, Parveen’s first language. The study information was sent to a family member who shared this information with the participant. The researcher arranged a telephone call before the interview was set, so that she could speak with Parveen to ensure that Parveen had understood that she was not under any obligation to participate. It was during this conversation that the participant mentioned that she had worked, but not in the last six years. It is not known whether Parveen enjoyed working in the shop or why she stopped working.

Samantha:
Samantha was a White British woman, 22 years old from the Northwest of England. Samantha had been unemployed for 18 months at the time she was interviewed. She was in a long-term relationship with her partner from high school.



Samantha experienced her first seizure in January 2019 and was diagnosed with NES two days after the first seizure. Even though she had received this diagnosis, Samantha sought out confirmation using a private clinic. Samantha understood her seizure events to relate to stressful situations and factors in her life. Using this model, she tried to manage stress levels by making coursework more manageable so that it did not trigger episodes for her.

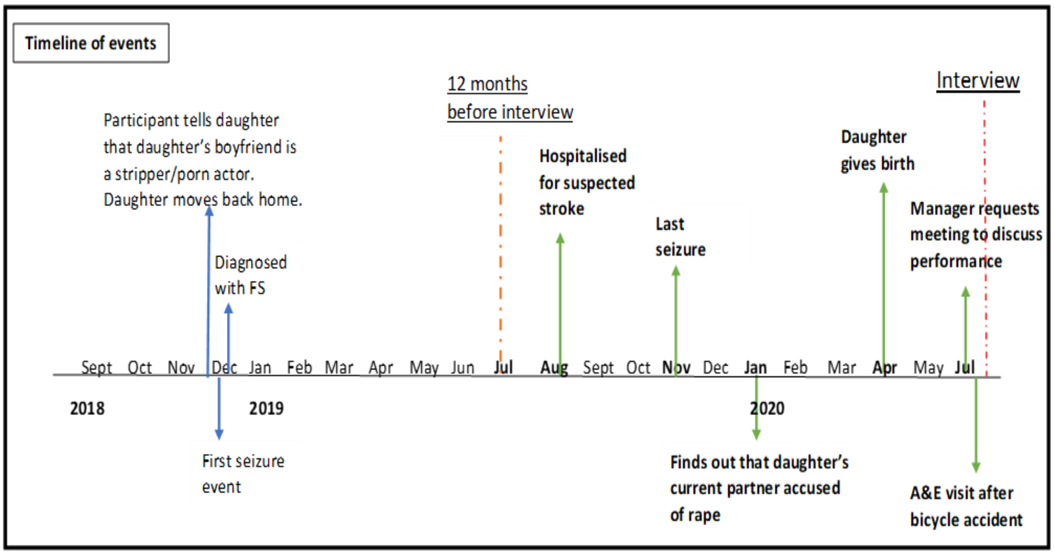
Samantha was currently living with her parents and her younger brother. Of the life events identified, this included moving from living with her partner into her parents' house. Other life events identified were: her partner had a car accident; Samantha began (and completed) psychological therapy; her father had an accident at work; and Samantha restarted her teaching degree (see timeline).

The timeline also shows events that occurred before the time period of interest, i.e. before the 12-month period under investigation. These events were found to be relevant to the discussion: Samantha moved in with her partner; she began to have seizure events; she had a seizure in her car; she stopped attending university.

At the time of the interview, Samantha appeared open and happy to answer questions. However, on listening back to the interview there was an indication in the tone of her voice that she perhaps was not completely comfortable doing so. This was apparent on two occasions during the interview. Once when Samantha was asked if she would be comfortable to talk about the sessions with the psychologist. The second point was when Samantha was asked if she would be happy to answer any follow-up questions from the researcher, if needed.

Sheila:

Sheila was a White Scottish, 47-year-old woman. Sheila was from the Northeast of Scotland. She had a degree and had been working in community nursing for five years. She lived with her husband, and had two grown-up children, a daughter and son. They did not live with her. Sheila first experienced a seizure in December 2018 and was diagnosed with NES a week later.



Sheila had formed a model to understand which triggers provoked seizure events and functional symptoms, for her. She considered stress to be the triggering factor and believed that having to disclose to her daughter that her daughter's boyfriend was acting in pornographic films, was the trigger for her first seizure. Sheila had not experienced a seizure event for 10 months at the time of interview.

Sheila attributed current speech difficulties to the revelations of her daughter's current partner being accused of rape. There was no seizure activity that coincided with this life event. There were however ongoing stressful factors at work; the first seizure also occurred in the workplace.

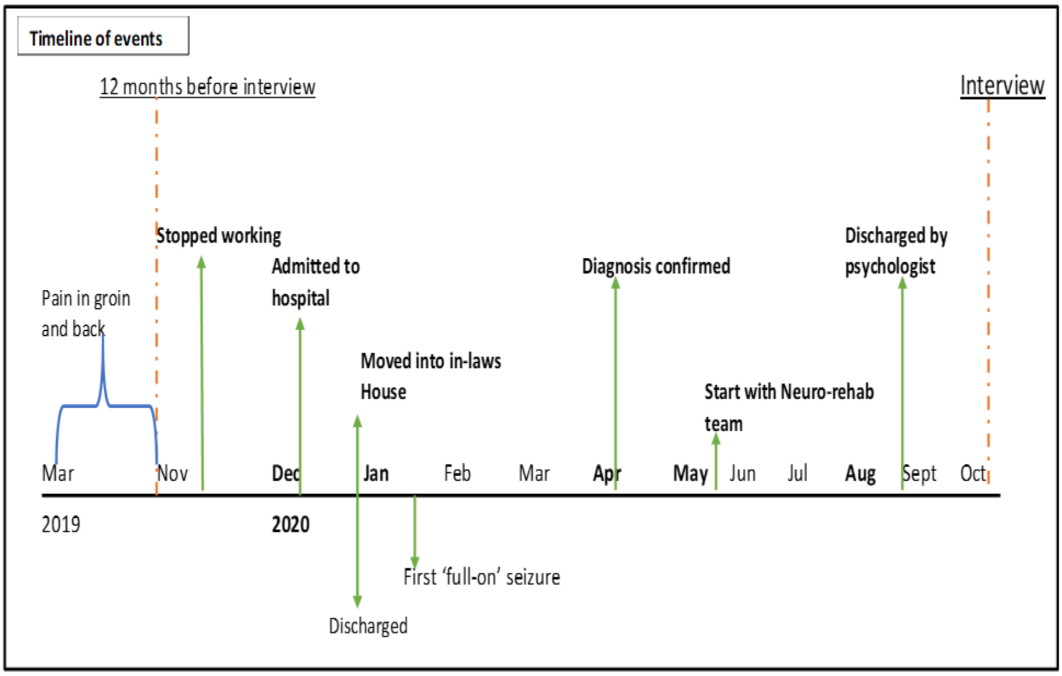
Sheila was sexually assaulted when she was 13 years old. Sheila made the link between the sexual assault and events in her life now, as a way of explaining the long-term impact it has had on her. This was apparent when she spoke about the relationship with her daughter, and the consequences that events in her daughter's life had on Sheila's life. Sheila did not mention the perpetrator of the sexual assault. However, Sheila appeared to attribute blame to her mother for not protecting her (*my mum never protected me*). Sheila was aware that she was not *responsible* for the assault but mentioned that the *baggage* remained behind.

She described finding it difficult to *defend* herself, although she could *stand up* for others. She described herself as an *easy target*, someone who gets *walked over, always trying to please everyone* and *always says yes*. An incident in which Sheila was asked to meet with her manager may be an example of the impact of the *baggage*. Sheila's manager raised concerns regarding Sheila's decision making at work. Sheila began to question whether the criticisms about her work were legitimate. She *started doubting* herself when faced with criticism from her manager, although Sheila said that the comments seemed unfair based on the previous feedback, she had received in her supervision meetings.

Sheila was seeing a psychologist for treatment for an eating disorder. She considered the eating disorder as a method by which she avoided dealing with emotions linked to the trauma. She talked about pushing the emotions down, shoving them away by using eating to comfort her. Sheila considered herself to be a typical case of childhood trauma leading to disordered eating as a form of coping. She had considered that she may have replaced one *crutch* with another, i.e. non-epileptic seizures replaced the eating behaviour. However, she appeared on some level to query whether NES was an accurate diagnosis and whether it may be epilepsy instead.

But so I don't know. Maybe there is a bit of epilepsy in there as well. Err, [sighs] who knows? But the only way to rule them out is to catch it on the EEG isn't, so. Sheila, p.47

Simon:
Simon was a White British male, 44 years old, married and living in Southeast England. He was self-employed and had been unable to work for 11 months. He and his family currently lived in his wife's parents' house. This was because their own house had not been suitable for adaptation to meet Simon's physical needs. He first experienced a seizure in January 2020, was kept in hospital overnight and diagnosed with NES the following morning. The diagnosis was confirmed with a video call with a neurologist some months later. Simon was clear that his seizures were not psychogenic in nature, although having the experiences had affected his mental health.



Simon explained that his wife had become adept at preparing him when they noticed a seizure beginning – usually indicated by his leg twitching. This preparation involved feeding Simon and taking him to the toilet because his seizure episodes could go on for some hours.

Simon identified his life events in the last 12 months as: moved house; hospitalised; and received a new diagnosis (see timeline). Simon described a specific incident that occurred on Christmas day whilst he was in hospital as an inpatient. The event was traumatic – it caused a lot of distress for him. Whenever he thought about his time in hospital, his thoughts would go to that incident. This one incident had also meant that future interactions with health services were judged against this.

Simon described feeling guilt for the way in which the family (wife and children) had been affected by his illness. He talked about being to blame for the change in their circumstances. Pain and mood seem to be linked for him, and he noticed this had an impact on his behaviour with his children.

At the beginning of the interview, he highlighted to the researcher that he sometimes had difficulty with his speech. During the interview difficulties with his speech appeared at times when he spoke of emotive situations. It could also be that speech difficulties were a result of the participant becoming more tired, as the interview went on for some time. Simon also mentioned noticing a decline in his attention and concentration, although this did not seem to have an impact on his performance during the interview.

Simon described having a good relationship with his wife’s family but did not mention any type of relationship with his own family. He did not refer to his own family at all. Simon insisted that he did not have any difficult/ traumatic events in his past.

4.10. Group analysis

The group analysis revealed six themes that were relevant across the sample. These themes are discussed in more detail, with examples in the section below.

Group Experiential Themes

The Group Experiential Themes (GETs) reflect the themes that were common across the group. The GETs are presented in uppercase, and the group level subthemes in lowercase. The table below show which themes were present for each participant.

Table 16. Participants present at each GET and subtheme.

THEME	Abigail	Ashley	Bryony	Chloe	Craig	Mark	Parveen	Samantha	Sheila	Simon
A: STRUGGLING TO LIVE WITH NES										
The journey to diagnosis		*	*		*		*		*	*
Perplexing feelings about the diagnosis	*	*	*		*					*
Suspecting that brain is not a typical NES brain					*	*				*
Making connections to try and understand what happens to me	*	*	*	*		*	*	*	*	
Seizures make me feel helpless		*		*	*			*	*	*
B: BEING LABELLED WITH NES FEELS DEGRADING										
Not being believed by others brings feelings of shame	*	*							*	*
Feeling ashamed of not being able to do more		*				*				
Impact of being stigmatised with NES		*	*	*		*	*		*	*
C: THE LACK OF SUPPORT MAKES ME FEEL ALONE IN MANAGING THIS CONDITION										
Feeling frustrated because do not feel equipped to manage this condition without support	*								*	*
Experiences of loss and feeling lost when support ends								*		*
Punitive experiences have a lasting impact		*							*	*
D: LIVING WITH NES IS CHALLENGING										
Non-epileptic seizures makes the transition from child to adult more challenging			*					*		
Feeling like a failure as a parent						*			*	*
Working is harder with non-epileptic seizures		*		*		*		*	*	*
E: HOW DO I LIVE BETTER WITH THIS?										
Raising awareness helps me to fight back		*							*	*
Having non-epileptic seizures has helped with understanding others better									*	
Feeling empowered and confident about fighting off seizures									*	
Distracting self at times of emotion or difficult thoughts			*							
Spending time in the remembrance of God brings blessings and healing							*			
F: LIVING WITH NES DURING COVID										
Access to healthcare was limited	*	*	*							*
Feeling lonely during the lockdown				*	*	*				
Lockdown feels hard			*	*		*				
Reflecting on the experiences of others				*		*				
Under the cloud of COVID						*				

A. STRUGGLING TO LIVE WITH NON-EPILEPTIC SEIZURES

The journey to diagnosis

The time taken to receive a diagnosis varied amongst the participants, with some receiving a diagnosis in a matter of days (Bryony, Samantha, Sheila, Simon); whereas for others it took longer (Abigail, Ashley, Chloe, Craig, Mark, Parveen). See table 13 for details. For some participants it seemed that there was a period of uncertainty before diagnosis that began when they started to experience symptoms. For example, participants may have noticed that they were having symptoms but either did not pay much attention to them (Simon) or explained them away as being related to other factors, such as fatigue, after work a long shift (Ashley). Craig mentioned waiting for five months to be seen by a neurologist, however from his interview it was clear that he had been experiencing seizures for some years.

Waiting for a diagnosis was experienced as a difficult time for some participants because it was a time of uncertainty, frustration and anxiety (Simon). For others (Craig) it was *horrendous* and *awful* period of time when waiting to be seen by a neurologist; it felt burdensome, that he was carrying *an awful weight*.

I was having mini seizures. Very, very frequent. Very energetic. Erm, like I said kind of similar to an electrocution. And that was, you know, very depressing. […] and that was a horrendous period. That was an awful weight because I had to wait five months… before I could speak to the neurologist. And during this time it was, it was ebbs and flows. Craig, p.30

Simon’s experience mimicked the experience of others in some ways – he experienced panic when the first seizure occurred, and confusion because he did not know what was happening to him. However, his feelings towards the experience before and after the diagnosis were the same: he felt uncertain, frustrated and anxious at both points in time.

Err…I remember thinking ‘what the hell is going on?’ because my body was uncontrollably – you know, that had never happened before. I was like ‘why can’t I stop this?’. Err, I was out, I remember being out of breath, I think I was panicking. Simon, p.18

I remember, I remember the word ‘FND’ and he [hospital neurologist] explained it was Functional Neuro, Functional Neurological Disorder. [...] no he said something along the lines of…he was sort of half joking, to lighten the, something along the lines of ‘it’s not nice *but it’s not going to kill you’ or something like that. So ‘it’s very unpleasant, it’s very painful but it’s not going to kill you’. Which I think is his way of putting my mind at ease. But I remember him saying that, but I still don’t remember the explanation. Err, that was it I think, and I can’t really. […] I don’t really remember …more than that. Except that it was like ‘what the hell, what is that?’ you know. Like I said, it’s not going to kill but still what does that mean? Going forward. Err, is it going to happen again because to me it happened out of the blue. Err, is it going to be a regular thing? I don’t remember him saying anything about that. Err…. whether it was going to happen again, what other symptoms.* Simon, p.18

Other participants (Parveen, Sheila, and Mark) did not share any feelings or details about receiving the diagnosis. Although Parveen and Sheila did talk about the impact of the diagnosis. For Parveen the journey to receiving a diagnosis took eight years, the longest of any of the other participants. She was relieved that the diagnosis meant that she no longer needed to attend hospital after a seizure. Possibly because this may have reduced the anxiety that she felt about going to hospital. For Sheila, although she had received a quick diagnosis, she was still waiting on the help that she needed to get better.

It’s been a long time since they’ve been happening, and I’ve spoken to the doctor a lot. I used to be very worried about it before but I’m grateful to God that now I recover more quickly, and the pain goes away. […] I used to get really anxious when I used to have them and have to go to hospital. But thank God I don’t get that worried when I have them now. […] I just stay at home and get better – I’ve never called the doctor. Parveen, p.14

And I’m also very aware that people keep telling you that I was very lucky I got a quick diagnosis, and they keep saying that if you can get all that you need really quickly, you’ve got a better chance of recovery. And all this time you’re waiting you’re thinking [sighs] ‘come on, hurry up. Fix this. I want to be well’. Sheila, p.28

Perplexing feelings about the diagnosis

After receiving a diagnosis, participants (Abigail, Ashley, Simon) referred to experiencing contradictory feelings or frustration around the event; and other participants described feeling ambivalent (Craig, Samantha, Ashley, Sheila). The ambivalence about the diagnosis seemed to relate to doubting whether the diagnosis was accurate. This was expressed by a few participants (Craig, Samantha, Ashley, Sheila), that they queried whether the diagnosis was accurate. One participant, who was not typical of the sample (Bryony) still described experiencing a range of emotions around receiving a diagnosis.

Craig was not satisfied with his experience of receiving a diagnosis. He was convinced that the pain and pressure that he felt, in his head, indicated something other than NES. His dissatisfaction was in relation to the method of diagnosis being based on exclusion, and doctors not paying attention to Craig's research. Craig referred to the struggle between him and the NHS because he considered their intentions, behind the test and diagnosis, to be different to his own. Craig expressed that the NHS was focussed on showing that there was nothing serious causing the seizures, whereas his focus was to understand what was causing the pain and putting a stop to it.

Because in the NHS, it just says 'patient is ok', 'test okay'. […]I had to actually get that data myself from the NHS. I requested the data. And I went through it, and I could see that, from the CD rom, I could see what the regions of the brain were that were hurting me at the time. […] And I was, I was quite concerned about a deep pressure, err that I could feel on the left-hand side of the, the cranium - in the parietal lobe. […] For me it was a bit of an ongoing battle… with the NHS[...] once I could see what was there, what was not there, it made a huge difference, that I can actually start working on the rest, of what I thought were the likely causes. Cause one of the issues that I was heavily concerned about was not being able to find something that could guarantee some freedom or some relief. Craig, p.31

Sheila questioned her diagnosis because there was, at some point, a query about whether her seizures were epileptic in nature. Although scans and EEGs indicated that the seizures were not epileptic, she still seemed to have a question lingering about the seizures being NES because she did not perceive herself as experiencing dissociation during the seizure.

Feeling *relief* was common amongst some participants (Abigail, Ashley, Simon) but the manner in which it was expressed varied. The amount of relief Abigail underwent upon receiving a diagnosis was described as similar to someone being told that they were *free of cancer*. The relief was associated with feeling that finally someone had listened and validated her experiences. However, although the diagnosis brought a sense of vindication for Abigail - that she was not to blame for her symptoms, it was still difficult to *fully* accept the new diagnosis. Abigail explained that this related to understanding the condition as being lifelong and not curable. Therefore, if she was to accept this it was as though she was resigning herself to having these symptoms for the rest of her life.

*I **felt** that the world's been lifted off my shoulders you know. I **felt** so relieved. I suppose someone who's had cancer, if they've been told that they're clear of cancer, I probably felt a bit like how they feel you know? And then I know, alright, it's not good news, to have this condition and things.... Err, and I probably haven't accepted it fully, but at least it's an answer. At least I can say to people now this is what's wrong with me. This is caused from the accident. Like I've got an answer… Yeah.... that's what I was more relieved about. So yeah.* Abigail, p.13

Ashley also spoke about the relief experienced when she was diagnosed with NES. The diagnosis made her feel hopeful because in contrast to Abigail's understanding, Ashley was told that there was the potential to recover completely. Similar to Abigail's reaction, Ashley also felt relieved that she had been diagnosed with something that was not life threatening. She was *quite hopeful* at that start; however, she described that *disbelief* crept in when she thought about the diagnosis. She struggled to comprehend how the symptoms that she lived with were not caused by something *serious*.

I remember feeling at the time like…quite hopeful because he'd said, err, it's a good thing we diagnosed it so early. [...] because it means that there's less undoing to do. And there's potential that you'll make a full recovery, err, and you'll be okay, err. So yeah, I felt quite hope- and a bit of relief as well. Because he'd explained that it meant there is no structural problems, you know it's not gonna get worse, and worse. It's not going to kill me or anything like that. So it kind of took away a lot of the worries that I've been having. Thinking like 'can I die from this?'[...]. Err, but then at the same time it was like [blows raspberry] mind blowing! I was like 'no! I don't understand how all of these things that have been happening to me'. How there's not something really serious going on in my body! So there was a little bit of like disbelief – I suppose. It's like, I just can't believe. That's just, 'how?'. I still feel a little bit like that but I do accept it. Ashley, p.23.

Simon also indicated that whilst initially he experienced *quiet relief* on receiving a diagnosis, because he could *finally tell people what was wrong*. This positive feeling was short lived. The frustration came soon after, when it was realised that there was not an obvious treatment plan that would be put in place for him, and this feeling replaced the hope he initially found. That is, the expectations following diagnosis did not match his experience of what transpired after diagnosis.

Bryony represented a different type of experience when it came to her diagnosis of NES; she was not typical of the sample of participants. She was the youngest at 18 years old and had only recently begun to experience NES. From her perspective, developing NES was considered an extension of the poor health and symptoms that she had been diagnosed with over the past two years. Bryony indicated that she was relieved to find out that she had NES, but the relief was mostly about the ease of diagnosis and not having to attend hospital for tests. Beyond this relief, she described feeling weary. This was because this was another diagnosis to add to the list of conditions that she already managed (fibromyalgia, Ehlers Danlos Syndrome, FND, asthma). Bryony described receiving the diagnosis as *very nice*. This language reflects the difference in her experience compared to other participants. She expected to receive a diagnosis – there was not the same level of frustration in her experiences compared to others in this study. She did not indicate that experiences with health services had been tiresome or difficult, or punitive like others have described.

Suspecting that brain is not a typical NES brain

Some participants (Craig, Mark and Simon) indicated that they did not consider themselves to fit with the archetypal NES/FND model. All the participants relevant to this theme were male. From the extracts below it appears that the participants were not only trying to show their understanding of the research in this area, but there was an emphasis on their seizures not being psychological in nature. That their seizures related to their biology in some way. The implication of this being that this conceptualisation made their seizures more real, not feigned or depicting mental illness.

There were several quotes from Mark that reflected how he had come to understand his seizure events. He started by explaining to the researcher that he suspected that his presentation would not be as the researcher *expects*. He believed his experiences would be different to others with NES. His understanding was that his seizures were based on neurological dysfunction (*miswiring*) and not psychological.

I'm not totally sure if my experience of like functional seizures is quite what - what you're expecting because I don't lose consciousness. Mark, p.1.

And I also suspect that, you know, it's not - what I'm telling you perhaps isn't what other people have told you. Mark, p.19

It makes sense to me, but it's really hard to vocalise and describe the fact that in my body those things are definitely misswired. Or have become misswired when I got FND. Suddenly it doesn't work properly. Mark, p.38

Mark also used words such as *inside of my head* to show that the experience was real and associated with the way his brain did not function correctly. Mark explained that external events often triggered an emotional response, which ended with a seizure episode. However, Mark indicated that this was different to others: his seizures were linked to *his weird condition*; *his* FND.

Mark tried to establish himself as an authority on his condition, and this was based on how the experiences felt for him. Additionally he stated that he had looked up the information, meaning that his understanding had been corroborated by science. This presentation was not dissimilar to how Craig or Simon presented themselves. Craig emphasised his level of knowledge, although he too showed that he was overwhelmed by trying to understand the diagnosis. Simon was more subtle in the way that he expressed knowing better than the information that he had been given.

Simon also emphasised that his experiences were real; they are not *in his head*. In this context meaning that they were not psychological, and the seizures were connected to his physiological experience (i.e. pain). Simon was concerned by the stigma that can accompany a mental health diagnosis and alluded to the judgements made by others that the seizures were *fake*. This suspicion from medical professionals and family/friends was the aspect that has had an impact on his mental health.

There was an anger underneath the surface that came across in Craig's account. This reflected that he was managing the situation himself because those assigned to him, such as the psychiatrists and psychologists, did not know how to help him, in his opinion.

they're [healthcare professionals] mostly concerned with structural malformities, rather than what I've spoken about .... I've already started with psychology and I've spoken to two or three psychiatrists on the subject. But they're not relevant to what I would consider to be, the. They can't help me. Craig, p.24

So actually, I, I wrote to the to the GP and the neurologist at the time and said you are essentially wasting my time, because you've actually deleted quite a lot of information that would have helped make a more certain diagnosis. So, what they're working through is, is a way of elimination. So working through a set of most probable diagnosis and doing tests to eliminate - one by one, till eventually they come down to a shortlist. Which is not how I'd do it. [...] And they've ignored a great deal of information that I've provided. Craig, p.29

Making connections to try and understand what happens to me

In making sense of their journey, some participants had to look beyond their recent experiences (Chloe, Samantha and Sheila). These participants explained how incidents in their past had, in their opinion, acted as causes towards developing seizures.

In the quote here, Chloe is describing how difficulties growing up (homelessness and financial difficulties) meant that she had lived with anxiety and stress for as long as she could remember. She began to have dissociative experiences as a teenager but did not receive a diagnosis until adulthood. Since receiving a diagnosis, she could now conceptualise these events as NES. Chloe suggested that to deal with being in a constant state of hyper arousal, her body *developed other symptoms* i.e. dissociation/seizures as a way of managing the distress.

*I think the anxiety was always there and the stress was always there. I didn't have any time to relax, and my body just developed other... other symptoms err…so, I think, I remember the first. The first thing that was happening was that I was dissociating **a lot**. And I didn't know what dissociation was. And then there'd be times that I felt like my hearing would go but nothing else would happen. I just... couldn't hear, because of the anxiety I assume, err. And then things like blackouts and seizures started to happen again but they were... less, they were more subtle so it would just be my jaw and my lip, or it would just be my legs and err.* Chloe, p.19

Sheila had come to understand NES as a response to the sexual assault in her childhood: developing low self-esteem and an eating disorder, and now NES in reaction to current stressful experiences with her daughter and work-related issues. That is, in trying to explain how seizures had developed for her, she started with a general overview and then made reference to how specific stressful life factors had an influenced symptoms and seizures for her. What is reflected in the quotes below is that the sexual assault was something that Sheila made reference to in different parts of the interview. The quotes are brought together here to illustrate how Sheila understood the development of NES, for her.

And I think everything, the whole spark off, definitely, has been around my daughter and the traumas. And then it was, you know, err, I was sexually assaulted when I was 13 and then trying to deal with this eating disorder and then finding out that the partner was doing all of this stuff. It was just like 'poof!'. Sheila, p.53.

And I can see the horrible pattern that I fit into this stereotyped. You know, the assaults when I was little. And you, know, the overeating as comfort, you know which spirals into obesity, which spirals into low confidence. You know, I can see the patterns. Sheila, p.29

Samantha also looked at the events that led up to her having the seizure that she describes as the factor that *kickstarted* her decline in health.

Just as I started a new job, err my education… and in the middle of all of that we decided to move in together! Cause we thought, you know, we didn't have enough on our plates! .... Err, so we moved in together and then I had – I blacked out. This is when it all started basically. Samantha*,* p.30

For other participants (Abigail, Ashley, Bryony, Mark), reflections were based on more recent life events and how they considered these to be triggers for seizure episodes. And these have been illustrated in the section describing the models participants used to understand the impact on their seizures.

So I think like, with the ***emotion*** *I think it was too much for my body to handle like.... The devastation and all the emotions, I think it with the.... the seizures, I think I was getting them a lot more than perhaps you know previous then*. Abigail, p.5

Bryony had several co-morbid conditions and described being *used to her body giving up.* So, although she was still trying to understand whether there was a pattern, her understanding of her body leads her to the conclusion that this was her body's response to stressful events.

I'm quite used to my body giving up whenever anything major happens, or even anything relatively minor happens. Err because it's like that! But during that kind of time I was just like 'right! My grades are bad, my future's a bit murky and my body has given up, that's sounds par for the course!'. Bryony, p.25.

For one participant (Mark), seizure events were conceptualised as a vicious cycle paired with an element of prediction, the belief that certain events will end with a seizure. He suggested it was like a *self-fulfilling prophecy*.

There's definitely a degree of self, err how do I put it? Yeah, I went into it thinking 'we're about to have our cat put down. I will almost certainly have a seizure whilst it's happening […]. If I have a day where I can get through the day, without any symptoms at all. And then the next day I think, 'well I didn't have any symptoms yesterday.' And it kind of builds on itself. Mark, p.34

Seizures make me feel helpless

Some participants (Chloe, Craig, Samantha) talked about the seizures as though they were a separate entity to them; something which they were unable to control. It was evident from the extracts below that participants felt out of control or wanted to express that it was an event in which they had no control. Although Samantha talked about a form of negotiating with the NES.

Samantha described bargaining with her body so that she was allowed to reach a place of safety before the seizure occurred. That is, when she experienced warning symptoms, she began communicating with her body, although the body/FND had the final decision which she expresses in the second quote as: *this is what needs to happen*.

And then kind of my body saying 'okay, we'll let you have this. We'll let you get to safety. You've now got safety so we're not going to make, we're making sure that you know you *can't open your eyes. You're going to have to pass out again', you know just to recoup, well not, it was…I can't think of the word but it was kind of like to justify opening my eyes during a seizure.* Samantha*,* p.26.

Another aspect that contributed to feeling helpless was that the seizures were unpredictable. The disbelief that the event is happening again without warning was expressed by Chloe. It occurred at times when she felt *normal*, and this caused her to panic as the seizures appeared *out of nowhere.* This suggests that she had limited control over the seizure, and the seizure was inevitable in order to reset her emotional state.

I think it's why I do panic because they do come out of ***nowhere*** *and like I felt normal, I felt fine, I didn't feel that stressed either. Err, and in my* ***mind*** *I thought I was managing the whole losing my job thing okay, err.... but apparently my body had a different idea.*__*.....*__ *[Sighs] I think my subconscious is always going. I think far too much, and I just struggle to turn that off. And when I do relax ... it's like my body or my brain doesn't want me to relax and my body or my brain goes into a panic mode or something. [...] It's when I least expect it. When I think I'm relaxed….. and I think I've got everything under control and my body's just like 'nope'! [Laughs]* Chloe*,* p.12.

Craig described his experience with the seizures as not only something he had no control over, but also incredibly painful. He likened it to *torture*. He wished to be able to press a button and liberate himself from the pain, although he seemingly chose another form of torture – *a pit of snakes*. The reference to torture could be based on Craig's experiences of not being listened to and not getting tests and such when he wanted them to be conducted. For example, the GP did not make the referral for an EEG when Craig requested it; the neurologist conducted tests to exclude epilepsy, rather than tests that would confirm a diagnosis of NES. So, whilst Craig did receive a diagnosis, it did not occur in a way that was satisfactory to him. Therefore, not only was the physical experience of the seizure *torturous,* but so was the process of receiving a diagnosis, to which it felt there is no satisfactory outcome.

Sheila was different to the above participants. She had managed to stop a seizure from occurring when undergoing an EEG to check for the type of seizures she was having. That is, she appeared to be demonstrating a form of control, but she also indicated that she feared that she may not be strong enough to ‘defeat’ the seizure.

Sheila: But I, oh gosh. But I’m able to, but I was able to find a way to fight it off when it was happening. If that makes any sense?
Researcher: What was going through your head when you were trying to fight it off? Do you remember what thoughts you were having?
Sheila: ‘Breathe, breathe, breathe, breathe. Do not, do not, do not, do not!’ [Laughs] [Clears throat]
Researcher: And that worked?
Sheila: Yeah, I was just. I was so scared. So, so scared. Sheila, p.50

Summary:

The above extracts demonstrate that the journey is personal for each participant. That the sense making is something that they are doing for themselves. There is evidence that individuals are using both scientific research and evidence from their own experiences to form their opinions about how they come to understand seizure events.

Receiving the diagnosis was an event for some individuals. Participants mentioned feelings of relief and confusion. No participant associated the event to an increase or cessation of their seizures. Part of the NES journey was to make sense of how NES were triggered. Here participants reflected using different models to show which factors they perceived to contribute to seizures, such as stress or pain.

Seizure episodes were events that left participants feeling panicked and not in control. Seizures were also considered to act as ‘reboot’ when stress or emotion was too much for the individual to cope with.

B. BEING LABELLED WITH NES FEELS DEGRADING

Not being believed by others brings feelings of shame

Experiences described by participants showed that participants recalled more negative experiences with healthcare professionals (HCP), although two participants (Abigail and Bryony) described experiences with HCP that were helpful. Abigail described how a neurologist had travelled to a hospital closer to where she lived so that attending the clinic was made easier for her. As described in GET A, the appointment with the neurologist was a positive and vindicating experience for her.

Err, when I rung to see where I was on the list. I told the booking office people that I could go to see him short notice, you know, because obviously I don't work and things. Err, but I said obviously I was disabled and obviously I needed disabled access and things. […] They said he […] would run a clinic, or come to my local hospital, err which five minutes up the road from me, err possibly. And he did see me up there. So I didn't have to travel due to the circumstances, so that was good. Abigail, p.15

Bryony mentioned that she struggled to grasp information if she was unable to see the speakers face. Therefore, a phone appointment with a neurologist made her anxious that she would miss information and not know that she had missed it. The neurologist managed to address this by following up the phone appointment with information in an email to Bryony. This was reassuring to Bryony.

they don't have a zoom facility set up and because I'm asthmatic, she said it was probably best if we tried a phone call, see how it went instead of coming in, at the minute.[…]So I said 'okay' but I was very much psyching myself up for it then.[…] Seeing as I was at the stage where I really had no idea what was really going on, I was worried that she would tell me something really important and I would miss it entirely. […] Err, I wouldn't know that I missed it would be the issue rather than asking her to say it again.[…] I would just…not process that entire section of that conversation. But it was okay. She sent me the links of a lot of the things that she mentioned in an email afterwards, err, which helped a lot. Bryony, p.11

Participants (Abigail, Ashley, Sheila, Simon) shared experiences that made them feel that family, friends and/or HCP did not believe that the seizure events were real. Participants believed that others thought that the seizure events were *all in their head*, meaning they were feigning the episodes, because they were considered to have a psychogenic basis.

Abigail perceived that there was a judgement being made, that she pretended to have mobility problems and seizures to claim money. The words used to express this showed Abigail's perception was that people were minimizing the impact that the traumatic event, a car accident, had on her. This perhaps conveyed that she may have been personally struggling with how long it was taking her to *get on with life*.

Everybody was telling me there was nothing wrong with me. Err... everyone was just telling me that it was all psychological issues there was nothing wrong with me – I just needed to get over the trauma, and then I'd be able to walk again […] Everybody told me that it was all in my ***mind*** *that I was making myself think that I couldn't walk […] I was thinking that I was having these seizures. It was like 'nothing wrong with you - just get on with life!' [….] Because people were saying to me 'well what's wrong with you then?'. And then I'd say to them 'I don't know'. And I knew people were thinking 'oh she's making it up, she wants money'.* Abigail, p.12

Sheila perceived her parents as disbelieving that she had a real illness. She interpreted their behaviour to indicate that they were annoyed at the inconvenience of having to visit her in hospital. And that it was only upon seeing the severity of her symptoms did her father accept that she may be genuinely unwell.

And my folks came over to the hospital and they were like 'oh what have you done now?'. And my dad was actually very annoyed [...]. He thought I was just dramatizing it until he saw it and went 'oh, oh, right, ok'. Because when you actually see it, it's not just me going 'well I can't speak' [puts on funny voice] [laughs]. Sheila, p.24

This hospital admission was due to the suspicion that Sheila had experienced a stroke. Sheila perceived a change in the attitude towards her from healthcare staff once the tests and investigations led symptoms to be attributed to FND. Sheila explained that this was reflected by the words used to communicate to her that she had not had a stroke but that the symptoms were *just your FND*. This, to Sheila, indicated that she was not being taken seriously by staff once they related the symptoms to FND.

But also you just feel like such a waste a time. You feel like such a burden. And you just feel like - the shame. You just think 'I have wasted all that money', you know. The [sighs] .....'It's just your FND.' It's like 'ok'. So then you feel, I feel like so ashamed. Sheila, p.52.

Ashley described different experiences with HCP, and most had been negative. Ashley shared an example of being mistreated, a traumatic experience where she was physically abused by a HCP inflicting pain on her. She described this as a tactic used to bring her out of the paralysed state that she was in when admitted to hospital. She could hear the HCP talking around her. The physical abuse was to illicit a reaction, to make her respond, presumably based on the belief that she was faking her symptoms. She indicated that she was thinking about how her experiences may have an impact on future experiences. That is, she was concerned that she would be judged by potential colleagues to be someone not to be trusted or taken seriously (*wasting time*).

Err, so yeah 'this one's refusing to come around' and I remember thinking 'I'd love to!' [Laughs] 'Like I'd really love to!'….. Err, yeah so then - they had to try like different things to get me to come around. So, they like, they were like […] pinching my ears and things. And I could feel it. I could definitely feel it. […] Err, so I remember this one nurse was trying to get me to come around. And then I heard her go to this man, err, called [male name]. And then she said 'oh we've got a good one for you. This one's really, this one's really difficult. You're gonna enjoy this one!'….. Err, 'I know you've got a knack for it!'. So, he came and yeah, kind of, like put two fingers behind my ears - like in that little groove bit. And then like pushed in really hard and kind of like lifted me up by my head from like the laying down position. And I remember it being like one of the top five most painful experiences in my life! Err. And I wanted to cry. I wanted to scream. In my head I was like ***screaming*** *but I couldn't….And, and I know, even at the time he was 'huh, that's weird. She's not even, she's not even shouting out or anything!' and he was like, 'and I know that hurt'. Err, and all I could hear was like my heart rate thing that was attached to me - was just, shot up. Err, obviously. Because, I was absolutely like… horrified. Err, but yeah - they tried that and that didn't work. So they were just like 'oh we'll have to admit her then'. And they sort of left me, left me in that bed for another maybe hour? And, then I just kind of came around by myself. Err, I think kind of having that. I don't know if it was cause nobody was around me, poking and prodding me kinda making me feel more stressed….. Because when he did that thing - I had like another, err convulsive type seizure. Err, and I think I had another one, a few minutes later - but then after that I was left, left to myself. It was quiet and I just eventually just came out of it on my own.* Ashley, p.8

There was a sense of disappointment from participants that their experiences with healthcare services had been *bad*. Although Ashley laughed whilst talking about her experiences in the hospital (p.8), the laugh conveyed a sense of disbelief at what she had experienced, being mistreated when she was in a vulnerable state. This was perhaps more acute for Ashley because, as a trainee healthcare professional, she was poorly treated by those that could potentially be her future colleagues, and from people she expected to show compassion and understanding. Ashley stated that the thought of this made her feel embarrassed. This suggests that the lasting impression on her, from her interactions with healthcare staff, had been that she should feel ashamed for having to use healthcare services. Ashamed to take up space and use resources that they felt that she was not entitled to.

Like it's so frustrating as well cause you kinda just wanna shout out like 'I want to stop, but I can't! I can't control this'. I haven't got time to be like pretending to have seizures - I've got things to do! […] 'Oh what if I end up working with these people. And they think, oh that's the girl that comes in with the seizures all the time'. So there's that element of it as well…. Err, yeah mostly it just makes me feel just embarrassed. Because I kind of have got an idea of how - the people that I've met so far - like how they seemed to feel about it. And it just feels like I'm wasting their time and I don't deserve to be there, kind of thing. Ashley, p.14.

After these experiences, Ashley was now cautious towards using healthcare services to the extent that she did not want to access healthcare unless vital (*dying*). For example Ashley was reluctant to engage with even primary care services, unless she desperately needed to. Her experiences left her feeling unable to access them without being able to justify why she needed healthcare. From the extracts here, it suggests that if she suspected symptoms to be FND related she would not contact the GP because she expected to be told to manage the situation herself.

Ashley: I definitely don't want to go back again unless I'm dead! [Laughs]. […]. It was just a really– just a really hard day. The fact that I felt like I was so out of control of my own body, and I couldn't do anything. And the fact that I felt like, I wasn't…. like I wasn't believed and. Err…The fact that nobody seemed to understand what was going on. And some of them even found it funny, and it was just embarrassing. And, yeah. Ashley, p.17

Err, [blows raspberry] even ringing the doctors. Ringing the doctors now, I just feel like, err, I have to have a really good, good reason to ring them now. And I don't know what that reason is yet [laughs] [...]. I'm questioning every time now that I think, 'oh maybe I should ring my doctor because I'm not sure what's going on. Or, you know I'm struggling', but then

I think: ‘oh, they're just going to think that I'm wasting their time.’ They’ve got really sick people that they could be seeing […] [sighs] I don't know. ***Even if*** *it got to the point where I couldn't walk anymore - I kind of feel like I still wouldn't ring the doctors and I'd kind of be like ‘this is FND. I've just gotta deal with it. This is my life!’ [Laughs]. Because I just think that’s how they would be. They’d be like ‘well it's FND – that’s it. Go and read … this website and do some grounding techniques.* Ashley, p.25

Simon shared an experience in hospital that he described as being punitive, and as though he was being admonished for being there. He described that in the subsequent days after the incident, he *dreaded* seeing the doctor again; he was terrified of this possibility. At a surface level, it may not appear that Simon is describing feelings of embarrassment or shame, however in the context of what happened and how Simon described the incident, the emotion at the foundation of this experience is shame. This experience with the doctor seemed to have confirmed the guilt that Simon was feeling, that he was to blame for his situation, and this left him feeling ashamed. That he is was not a man, because he was not fulfilling his duties as a father and provider for his family.

Because obviously - pain, and I didn’t want to be away from my family. And he came round, really abrupt. He didn’t introduce himself, he was just doing a ward round, and he went err ‘Mr [participant], I don’t know why you’re here. You’re just taking up a bed, err, it’s just chronic pain, you’ll just have to live with it. You need to man up’. So I was devastated. I was low and depressed anyway but that just ***wrecked*** *me for the day. I was so upset. So, you know you don’t expect to hear that from a doctor, especially when, you’re going through a lot and possibly your mental health is not 100% at that time.* Simon, p.5

And I, for those three days, I was dreading, you know proper filled with dread and terror. Because I didn’t want to see him again. What. I don’t know. I really was scared. Thankfully I didn’t, you know. [...] So now that I’ve been discharged from it, I’m not going to thankfully, shouldn’t, ever cross his path again…. Err, but for those three days – yeah, I was really dreading it. Or every time there was a ward round, because it was a side ward, you know I was quite close to the double doors that went out to the main ward. And every time that door opened, even like subconsciously I was like you know, I was dreading it was going to be him, you know. Err. So yeah, those three days were horrendous. It was a great relief to be discharged, just to get out of there [laughs] just in case I was going to see him again. Simon, p.12.

Similar to Ashley’s hospital experience, this incident above was traumatising for Simon. From the extract, it can be understood that Simon felt that the doctor was dismissive of how much pain he was in. The experience left him feeling devastated (*wrecked*) and low. At a basic level, this interaction demonstrated a lack of empathy from the doctor towards a patient. At a deeper level it could be construed that the doctor was perpetuating ideas of toxic masculinity: that a real man is one who does not need help; he should get on with his life regardless of pain. Simon explained that his low mood was exacerbated by this comment to the point where he was inconsolable. Simon suggested the impact was for that day, but it evidently had a lasting negative impact on his self-esteem.

Feeling ashamed of not being able to do more

For some participants (Mark and Ashely), the impact of NES on their lives included not being able to engage in more work (Mark) or extra activities (Ashley). That is, it appeared that they struggled with this because of the standards that they perhaps had set for themselves.

Mark related this to his role as team manager. He was not able to work longer hours but was aware that members of his team were, and this made him feel inadequate in his role. Mark expressed feeling guilty despite telling the researcher that he felt supported by his own manager. For Mark, the guilt could relate to having a supportive work environment, an understanding boss, but not being physically able to work any more hours to indicate gratitude or compensate the company for showing him compassion towards his situation. The feeling of guilt was associated with the physical feelings of fatigue, and *occasionally* experiencing brain fog, because these were reminders that he was working to the best of his perceived ability or capacity.

I know my work is incredibly supportive but it's still, it’s still… difficult. You know I still feel guilty that I'm not putting in the hours and you know - particularly there are people in my team that don't have kids. Or you know…have different sorts of situations that mean they're able to work longer hours than I am. Yeah, you know as the boss you sort of feel the pressure to put the hours in. But you get stuck. You know, three o’clock I’ve got to look after the girls for a couple of hours. Then five o’clock I’m just stuck at my computer staring at my screen, just like, aaah. So that, yeah. ……So yeah. Lockdown is tough! Mark, p.29

Ashley conveyed that she was upset about having to acknowledge that she had to make changes because of her health. This was possibly made harder because she was starting out in her career. She was at an age where one would not expect to have to make health related decisions (28 years old), and also, she was in a physically demanding role working for the NHS.

And I just kind of felt like because it was to do with being unwell, that's one thing that I struggle with. I don't like the thought of it being ill would get in the way of doing things I enjoy. Err, because it and kind of goes against, do, do you know what I mean? Err. I just wouldn't like to think that - I'm not ready to admit that it's ***that*** *bad, that I need to stop doing things. So it was having to admit that to myself. And say actually, 'you are struggling' and it was probably the hardest part.* Ashley, p.5.

Impact of being stigmatised with NES

Four participants (Ashley, Mark, Simon and Sheila) talked about the stigma associated with having a diagnosis of NES. The stigma they referred to was about being *labelled* with a mental illness. Other participants (Bryony and Parveen) did not refer to any perception of the diagnosis being stigmatising.

Bryony and Parveen, therefore, were not typical of the sample. Bryony was the youngest participant in the group and had experienced a seizure for the first time only a few weeks before being interviewed. Bryony's experiences were described as informative and reassuring. Her experiences with healthcare services – compared to other participants, were neither punitive nor dismissive. This could have been facilitated by the fact that her age (18) meant that her mother appeared to be at the forefront of understanding the condition and helping Bryony to understand.

She [neurologist] gave me a lot of time to ask questions which, err, I think is what I wanted. It was pretty quick, but I'm not sure I would like it to be much longer. Err, I got a lot of the information that I kind of needed to hear immediately, mostly like what had happened and what I was supposed to try and do next. And then a lot of the kind of secondary information like the causes and further information, she sent me later. Err, so I did get everything that I needed from the conversation, err, but in a way that meant that I would take it in, which I really do appreciate. Bryony, p.12

Similarly, Parveen did not describe her dealings with healthcare services to be negative or dismissive. However, it should be noted that Parveen had the longest timeline in terms of receiving a diagnosis for NES. It could be that in the relief of being finally diagnosed, Parveen had not reflected on how difficult this journey may have been for her. She indicated experiencing anxiety around the seizures when she had to attend hospital. What is not known is whether this anxiety was around not being able to communicate whilst in a seizure, or anxiety about how she would be treated in the hospital.

it's been a long time since they've been happening, and I've spoken to the doctor a lot. I used to be very worried about it before but I'm grateful to God that now I recover more quickly and the pain goes away. […] I used to get really anxious when I used to have them and have to go to hospital. But thank God I don't get that worried when I have them now. […]. It used to happen a lot. […]I couldn't speak. The doctors would be speaking to me and I'd shake my head, but I'd say I can't speak. […] And then the doctor said she's paralysed. The doctor would sit next to me for hours at a time talking and calling me. And then the ambulance they would do the same. They'd say 'we don't know what's wrong, she can't speak'. Parveen, p.13

Some participants (Mark and Simon) made a point to clarify that they did not believe the aetiology of their non-epileptic seizures to be linked with trauma or abuse. Simon expressed that FND was misrepresented in the media that he had come across, as a completely *psychological* condition.

I just…because there's a great lack of understanding, or…. misunderstanding of FND…… And there's been a few programmes on telly recently which … have, I personally felt, misrepresented it. There seemed to be a stigma associated with it….and … a lot of people, I'd say majority of people, assume it's all psychological…. And I get that for some people, absolutely. But…I'm 100% sure, and there are cases of it, mine is pain/injury related.... ***But****, on all these FND forums and everything you see that are people in the same boat, but automatically, and you see it in medical professions there seems to be this stigma and it seems to be, they assume it's, it's a mental health. Yeah. Well obviously, it has affected my mental health. But they think it's caused by that, and they think it's all psychological. We've come across that a lot, and we think that's* ***wrong****, and that also has a negative effect on* *me. Because that makes me feel sometimes - when everyone tells you 'it's all in your head, it's all in your head'. It's* ***not****!* Simon, p.36

On the surface, Mark did not appear to be referring to stigma in the extract below. He expressed that perhaps his current condition was *payback* for having a healthy and relatively untroubled life in the first 30 years of his life. However, he continued to mention that he had no history of *emotional trouble*, *abuse* or *trauma*. This was an attempt to distance himself from his seizures as being labelled as a mental health disorder.

But yeah never really had to deal with anything particular - but you know fairly, humdrum of life. No particular problems or stresses at all really, you know. So yeah, it is really only the last three years where that slight sense of… [Laughs] payback for the luck of the previous 20/30 years before that, you know. Err, yeah, no like, the FND stuff was, it's all completely out. It was completely out of the blue. There's no, you know. I don't have, I don't have any err, history of emotional troubles. I've never been a victim of any kind of ***abuse****. I've never had, err, I've never had any traumatic experience really, at all. You know, no, no more than, yeah, err, err. One vaguely unsettling experience when somebody died, you know, near me. I certainly don't have PTSD or anything like that. Err, you know no kind of history of emotional problems, no health problems really.* Mark, p.35.

And other participants indicated that they were querying the diagnosis still (Sheila and Ashley), perhaps looking for a more acceptable physical explanation for their symptoms. Sheila mentioned the internal struggle that she had with receiving the diagnosis. She disliked the diagnosis because in her experience it brought with it negative experiences and perceptions from others. However, she also described herself as a typical presentation of NES based on the trauma she had faced in her life. This internal and external struggle manifested as her *fighting* against the *stigma* and feeling *impatient* with the battle she was in.

Whereas some people I've met with it, just accept it easy. Whereas I just fight it the whole time like [blows raspberry]. I get really impatient with it like, ugh, yeah, I don't like the stigma. I don't like the label.' Sheila, p.38

Two participants (Chloe and Sheila) expressed that they felt discriminated against, at work, for having a disability. Sheila was apparently more aware of the consequences that this may have for individuals with NES; that unfortunately individuals may struggle to keep or continue to be in employment.

Sheila: Yeah, unfortunately, you know a lot of people have said that a lot of people with FND never get back to their job, and I've never fitted into the team that well. And I just thought….she's just using this as an excuse to sort of move me onwards. But I actually really love my job, but, errm. [Sighs]. [….] I mean she does it all the time. She kept talking about my disability. […]
Researcher: And it's had no impact on your work?
Sheila: Well, it has! But we've worked around it. Sheila, p.13

Chloe also indicated that at the time redundancies were made, although she expected that she may be made redundant, she felt that the reasons behind the decision provided to her were not a full account. Having to manage with multiple conditions meant that Chloe had to take time off sick at times. And although the focus here was not on the seizures, the impact of having a debilitating illness was essentially the same as that which was described by Sheila.

And although they were very accommodating – and you know they were saying maybe I could work in the office instead of standing and being at the front office. They didn't implement those things fast enough for me to feel... comfortable or that I could…. It felt more like a, a nuisance than anything – me asking for extra support.[…] Even though they said, when they hired me, that they'd give me all the support that I needed. Err.... but on the score scale thing that they gave me they didn't mark my attendance at all. So, they said that my attendance was fine.....which I don't, which I don't honestly think is what. I think my attendance has an issue to do with why I was let go [...]. Chloe, p.17

Summary:
Although experiences with health services were not negative for all participants, more negative experiences were recalled by participants. Some participants described events and interactions that made them feel negatively judged and that having a mental health condition was stigmatising. One participant in particular had an experience where they

were not believed that their symptoms were genuine. Such experiences had a longer-term impact on individuals, and the examples shared above suggest that these individuals have been left with a level of trauma from the attitudes and mistreatment.

C. THE LACK OF SUPPORT MAKES ME FEEL ALONE IN MANAGING THIS CONDITION

Feeling frustrated after being left to manage this condition without support

There is a sense that participants (Abigail, Sheila, Simon) were disappointed that even after receiving a diagnosis it was left to them to find a way to manage living with NES. Participants described that there was a lack of information provided at the point of diagnosis, and this meant that they felt they were having to manage a condition without being equipped with the information on how to manage. Simon indicated a sense of disappointment; that although this experience did not feel punitive (see subtheme below), the outcome was essentially the same. He was disappointed and frustrated to be told that he must live with this condition, and because the information given to him was *vague*. Sheila expressed that she had been told that it was positive that she had received a quick diagnosis, however she was still waiting for the support to help her improve.

It was almost like back to the other doctor where he went: 'you're going to have to live with the pain'. Okay. Pfft! I'm going to have to live with these seizures, great! You know err…. A little bit more information or clarification would, would be good. For whatever reason I didn't get that, at the time it just seemed very. I mean He was a much nicer doctor - very pleasant compared to previous experience. But again it was very vague. And of course FND is very vague, I get that. […] but you'll just have to… manage it. Okay. But they didn't tell us how to manage it…. So how can you manage something if a) if you don't know what it is, I mean how, how. So, so we didn't know what you do...... In the end we felt lost, and we were a bit sort of bereft at 'right, okay. Great! Here we are again!' [Laughs]. Simon, p.21

Abigail's experience was one of contrasts. She began by describing that she was desperate to be heard and this journey finally led to a *wonderful examination* which resulted in a diagnosis which felt like vindication.

And no one was listening to me, and I was ***begging*** *for help. I'm begging all these people 'please help me, refer me to someone. I just want help. I just want an answer. Help to try and get better!' You know 'I don't want to be like this, my whole life has been turned upside* *down!' I was* ***begging*** *them for help. Even my mum was in the appointments saying, 'look we're begging for help, can someone please give her something, send her somewhere.' And they were like 'there's nothing wrong with her'. And they kept saying 'her scans are clear, she didn't break her back', 'her scans are clear, there's nothing wrong with her.' And we kept saying, 'there's something, something is not right somewhere. There's something is wrong with her'. And then we went to him. And I was an hour and a half in with the neurologist he gave me a wonderful examination, done lots of tests. Err...And he said, then he gave me the diagnosis.* Abigail, p.14

In some ways, Abigail's experience was similar to Simon's. Both Abigail and Simon were supported by family members. For some participants, family members were the participant's advocate and support outside and inside the clinic setting, providing the practical and emotional support also. However, whilst Abigail did not express the frustration that Simon and Sheila did about not having support, she indicated that she was aware that certain steps must be taken before she could receive the support that she thought would benefit her. She was prepared to take part, in what sounded like a performative task, so that she can access or be referred to the service that was assumed to be better for her.

Like he said he can't do much with me now because we're in lockdown. [...] he said like they've got to be evidenced based - they've tried other avenues first. So he said like he's going to do this video like call me for a couple of weeks, course thing, to show that we've tried. He said once he knows it's not going to benefit me. It's not going to work for me. […] then I can go straight on that list then for that one-to-one. So, I said 'yeah okay'. But he thinks an inpatient place will benefit me and he would support that as well. […] cos I feel like there's a plan going forward. A temporary plan, err... to try. Like I said we'll try anything. Abigail, p.23

Experiences of loss and feeling lost when support ends

Participants shared experiences of when they had received support (Bryony, Ashley, Samantha, Simon), although only a minority (Abigail, Bryony and Samantha) expressed that these were not negative experiences.

The extracts here demonstrate how participants described either a sense of loss (Samantha) or feeling lost (Simon) when the support that they were receiving came to an end either in an expected (Samantha) or unexpected way (Simon).

In the case of Samantha, there was a sense that she had lost the one person who understood what she was going through. Although Samantha knew the therapy would end after 12 sessions, she struggled to process the emotions. She stated that other people considered the therapy ending as positive, but Samantha had focussed on the loss. She spoke of *grieving* over the relationship ending during her interview. Samantha was concerned that she would not be able to manage without the support of her psychologist. In fact Samantha shared that in relation to this, before going to sleep, she often ruminated over how she would manage in future situations, and this left her feeling anxious.

I was upset. I was sad. I was very, very numb - all at the same time. I couldn't really express how I was feeling. Err, I tried to express it to [partner]. He could see that I was feeling really distraught and stressed. Err, and he said you know, 'what's the matter?' And I said, you know 'I've finished with my psychologist'. And when I've been saying that to people they've gone 'oh, that's brilliant! Well done you! Well done you!' And I think my mind wasn't ready for it. Err, so it's taking me a long time to adjust. Err, and it's a case of trying to figure out, you know, what's going to happen next. You know I'm on my own now. What, what's going to happen! Samantha, p.19

In fact it was on the Friday I had one of my worst sei...seizures. So, the Friday aft…so like I had my appointment on the Monday and on that Friday I had a really, really bad seizure. Err, and it was worse than one of my first ones actually.[....] I was feeling really… anxious about the whole situation because [...] I just finally understood, you know, I'm not going to see her again. Err, and, I got….very emotional that day, err for some reason, err I was just really numb. Just emotional, upset. There was a couple of times when I ended up crying! I was just really, really down in the dumps. And, I think that must have triggered it. Samantha, p.21

Simon related that his experiences with healthcare services left him feeling that he/his condition was not taken seriously enough to warrant support. Simon stated that he still thought about the time he received the diagnosis, perhaps refreshing the sense of disappointment he felt after the initial feeling of hope faded. It seemed that Simon was unsure about what he expected would happen after receiving a diagnosis of FND, but he described some expectation of there being a treatment plan. His disappointment appeared to relate to receiving some care but then being discharged from services. This repeated experience for Simon left him feeling let down by healthcare services time after time. He described the feeling of being *left in limbo*, without support.

I'll remember it in a sense that I've finally got that diagnosis – that's great. ***But*** *[...] the way we were just again…. 'there's a website, have a look at it'. Err we, we've done all the research since, and you know. Spoken to the GP and they've helped us get the neuro-rehab team. [...] You know there wasn't a treatment plan. You know I don't know what I expected. You know with some diagnosis you get, err you know like with cancer you get, have a like you get chemotherapy – there's a treatment route. Isn't' there? [...]. I guess because everyone's so different. There's different reasons for FND, there's psychological, physical, emotional, there's physical injury, different causes, so. But you would have still thought that there would still be, I don't know… some form of...initial treatment plan. [...]. But we had to instigate that so…. that was a bit disheartening.* Simon, p.23

Punitive experiences have a lasting impact

Three participants (Sheila, Ashley, Simon), described some healthcare experiences as punitive, leading to a reluctance to use services again.

There were experiences of feeling unworthy of receiving care (Sheila). Sheila perceived the neurologist to have judged her for having made the wrong decision to attend hospital for a suspected stroke. This perhaps felt unfair to Sheila because her description of how she felt suggests that she was judged as acting inappropriately. Sheila had followed the advice of a family member who is a HCP, and also after calling NHS 24. Sheila had initially been reluctant to go to the hospital. She had not called an ambulance, as suggested by NHS 24, and her husband had taken her to the hospital instead. Despite this, she felt that she had been chastised by the neurologist for presenting at the hospital as an emergency case, instead of calling the neurologist for advice.

You know I felt like I was a naughty girl that was getting a telling off for not phoning her first. It was like I should have known better. [Laughs][...]. Made me feel like I'd wasted NHS' *time and money. And I think 'you know what, the way that I was presenting if it was anybody else, you know'. Yeah..... And I think I only got that because I was so able to articulate that. I think anyone else would have got 'no, you did the right thing'. [Laughs].* Sheila, p.33

An incident in the hospital between a doctor and Simon, left Simon feeling berated. He described the situation as feeling punitive. It could be that the imbalance of position (doctor vs patient) meant that Simon felt he had no power. In this extract, Simon talked about how difficult the episode with the doctor was for him, it was like a trauma for him. He does not like to think on or talk about it because it is still upsetting for him.

It's a bit upsetting. I don't like, I don't like talking about it. It, well no [sighs] [laughs]. The ***whole*** *time was unpleasant,* ***but*** *the time that really affected me was the Christmas day episode that I told you about…. With the doctor. And even now I'm getting a bit, emotional. Because it was – I was so low anyway, and for him to say that you know. It just. Yeah, that really upset me.* Simon, p.11

Ashley's experiences were unique because she was not only a patient, but she was also a HCP too. She conveyed a sense of disappointment and of being misrepresented. There may also have been a feeling of betrayal too. That as a HCP, one may perceive another healthcare professional to act with compassion, but when that individual no longer seemed to be on your 'side' their demeanour and tone changed, to suggest they no longer supported the patient. Ashley indicated that she found herself ruminating on some of the past experiences such as being treated as a difficult patient that was 'pretending' to have a seizure. That is, the incident left a lasting impression on her, to the extent that only death would be the circumstances in which she perceived it would be acceptable to attend hospital (above). Talking about the incident was difficult for Ashley and she became tearful when recalling what happened.

Ashley described that whilst the first experience was positive, this changed when Ashley met this individual again. Ashley assumed this to be because the HCP had judged Ashley to have ignored the advice that was given to her. That because Ashley did not act on the advice, the consequences for Ashley were that the HCP considered Ashley to be disingenuous. That Ashley must not be interested in getting better because she had not followed the advice. Ashley's disappointment related to the change in the manner that she perceived from the HCP, that Ashley was misunderstood and considered a *mess about.*

> *And I remember she being really lovely and being really understanding. But she kind of decided from what training she'd had that, err, FND is caused by PTSD. So that was her, that was her understanding of FND. So I remember the time before this time, her sort of talking* ***to*** *me, about, err, how you know it usually PTSD and I should get treated for that. And she had PTSD before so she kind of had that, err, attachment to that idea – I suppose. […] And I think she really wanted to help me. But this time when she came, she just [sighs and laughs] she did not have time for me! She actually sounded annoyed. I remember her saying to my husband 'did she go to the PTSD person that I told her about?'. And my husband was like 'no, but she's actually got other things that her neurologist has suggested, so, you know she's, she's accessing support. But not this one - that you wanted her to do!'.* Ashley, p.14

Summary:

In this theme participants reflected on experiences with healthcare services. These experiences were punctuated by the attitudes of healthcare professionals encountered by participants, and how negative attitudes had a lasting impact on the individual. Beyond feeling unsupported, disappointed or frustrated, participants spoke about specific events that felt punitive or traumatising. Participants (Samantha and Ashley) indicated ruminating over the events, and the emotional impact this had on them, causing one individual (Samantha) to experience seizures related to the emotions brought about from the event.

D. **LIVING WITH NON-EPILEPTIC SEIZURES IS CHALLENGING**

Several participants (Bryony, Samantha, Sheila, Mark, Simon, Ashley, and Chloe) indicated that living with non-epileptic seizures made different aspects of life more challenging for them. For these participants these factors related to their identity as an adult, a parent or an employee**.** These challenges often appeared to relate to the standards that participants had set for themselves.

Non-epileptic seizures makes the transition from child to adult more challenging

Two participants (Bryony and Samantha) in the sample were at the point where they were looking forward to moving in with a boyfriend (Samantha) or moving away from home for the first time (Bryony). These signalled adult events for them.

This subtheme focussed on a developmental life stage. That is, this subtheme related to a transition from one part of their life to another that was not only to do with their chronological age but the activities that highlighted the move from child to adult for these individuals. For Samantha it seemed there had been a setback in their adulthood, whereas for Bryony, she was moving forward.

Samantha described that several events occurred around the same time, and she experienced her first seizure soon after (1-2 months). This change in her health became a disruptive force in her life and relationship with her partner, resulting in the decision to move back to their respective parental homes.

Just as I started a new job, err my education… and in the middle of all of that we decided to move in together! Cause we thought, you know, we didn't have enough on our plates! [...]. Err, so we moved in together and then I had – I blacked out. This is when it all started basically. Samantha*,* p.3

Samantha indicated that the decision for her and her partner to move back to their respective family homes was not easy. In the quote above Samantha indicates that the decision to move-in together was made by her and her partner, however elsewhere in the interview Samantha had explained that her father had initially encouraged her to move out of the family home. Below, Samantha explains she had concerns that asking to move back would be viewed unfavourably by her parents, particularly her father it seemed. She described being *scared* that her parents would not agree to her moving back home and the thought of the conversation with them made her *really anxious*. She had concerns that she and her partner would have to continue to manage with the situation, on their own – possibly meaning that she perceived there to be no alternative. That as an adult, one had to deal with the outcome, there would be no help. Samantha referred to being *defeated* by the situation. She saw this outcome as a failure. That not being successful in their attempt at being independent adults meant that they had been defeated by the challenges that adults face. However, her parents agreed to her moving back home and the disruption to their plans was seen as a temporary setback. The period after moving back home was one in which Samantha experienced multiple seizures.

And my bedroom, my dad used as like a little retreat. [...] so it became his second bedroom. Err, so, so that that week I was sleeping in my dad’s bedroom, not my bedroom. [...] and I *had a lot seizures. I passed out quite a lot because I felt like I didn't belong anymore. And, err, I wasn't part of the house. Err, I was missing [partner]. And there was just a lot of emotional factors in there. And when I finally started to move back – [...] putting everything back in the house and back in my bedroom. It made me feel a bit more at home. And it is only until recently that I've redecorated it. So it's…more me for now… basically. Instead of when I was teenager, or when I just came back from uni.[...] it's adult me in my bedroom now. So it's, it's a lot, a lot [sigh]. It's a lot easier now compared to when I first moved in. Err, I mean seizure and my passing out are a lot less frequent. Err, but it was really, really rocky, rocky start.* Samantha, p.39

We can look back at this as a learning curve. We know what to do. And we now know – we was discussing it the other day [...] so we've used as a learning curve. Err, and it was a relief. Err, we were upset but - it's just life. Samantha, p.38

Bryony, the youngest participant in the sample, moved away to live in university halls. She expressed relief in being able to achieve *adult* tasks, i.e. independent living tasks. Her experiences with FND and NES were relatively new and so she was still learning about the condition and how to manage it. She was also in a situation where she was learning how to live independently for the first time. In both instances the common feature for Samantha and Bryony was moving out of the parental home to live independently and developing seizures after this event, but the outcome and meaning of events was different.

Feeling like a failure as a parent

Four participants were parents (Parveen, Sheila, Mark and Simon). Three participants (Mark, Sheila, Simon) mentioned how interactions with their children, whether young or grown, had changed, although the impact was different for each participant. For example, Mark and Simon's children were younger and lived with them. Mark and Simon indicated that they felt they were lacking as fathers; their parenting style had changed (Simon), or was inadequate (Mark), based on their judgement.

Simon expressed that having FND had an impact on how he was able to interact with his children. The cognitive effects of FND, combined with chronic pain had an effect on his mood and levels of tolerance.

I get very frustrated easily. So I want to do stuff with my kids. But if we're playing a board game or something and I just – again I can't follow it, so I just get ***frustrated****. You know I don't want to take it out on them, but I get snappy sometimes. They're aware of it. They see it as part of my symptoms, but I don't like it. […] I'm CBT training to try and not do it….But I can't get out of that. It's such a big thing for me.* Simon, p.10

Whilst Simon had mentioned trouble managing his frustration, it could be that this was fuelled not only by not being able to do what he used to, being in constant pain, but also the guilt that he felt. In the extract below, Simon describes ruminating over his thoughts about his health and the impact that it has had on his family. His guilt was associated to his family no longer being able to live in their own home, and that he *feels helpless,* and *completely useless* because he was limited in what he could do - comparing his life to how it used to be before NES. His frustration and guilt related to how he felt that he had failed as a father (and husband). He blamed himself and this is likely why the doctor's words (*man up*) had such a hard impact on Simon. A stranger had verbalised his thoughts and feelings, and this perhaps felt like confirmation that he should feel guilty for being unwell.

I don't sleep well, so I'm often awake at night. And again that's the time again when you think things and churn over your thoughts. So yeah, for me it's, I'm ***always*** *thinking about that. Whether it's consciously or subconsciously, but I am. It's always there. Or I'll see my wife get upset about something, or, I'll feel so helpless, or so useless that I can't help her. [...]. And everyone tells me that it's not the case, but it keeps coming back to me. It's like we're only in this situation because of me. We've only had to move house because of* ***me****. We've only, you know the kids are upset because they're not in their house, because of me. It's not, not my fault but I take that guilt on…. Completely. I struggle with that. And I do feel that it's genuinely my fault. Err, and none of this would ever, you know we'd be at home, you know [sighs] happy family unit if it wasn't for my situation. [...] and I do feel completely useless – even with all the counselling etc, etc. that's still ongoing and hopefully that'll get better but that's the key thing for me – [quietly] that I'm struggling with.* Simon*,* p.10

Mark was clear that the physical limitations experienced by managing with FND made him feel inadequate as a parent.

My FND makes it worse because I'm not. Because I can look after the kids by myself, but I find it quite difficult to, to look after the kids… because if they start shouting at me, I tend not to be able to talk or I can't walk to. It's just, just hard to manage them. And when you got, you know, crushing fatigue as well at the end of the day. Mark, p.28.

Mark referred to not doing his *best*. Although he did not quantify what this meant, how one would know they were being the *best*, the point he was making was that he felt that he was not achieving this standard. This appeared similar to Sheila, in that this was a standard that they had set for themselves by which they were judging how adequate their parenting was.

And then [sighs] there's a lot of guilt. You just feel, sort of, like a crap parent because you're not, you know. You're not doing your best for, err. So yeah, that's, that's err pretty rubbish. Mark, p.29.

Sheila and Parveen's children were grownup, and both participants had children living independently, in their own relationships. They both appeared to provide emotional support for their children, although Sheila mentioned that she had to keep the relationship with her daughter at a distance at times. Sheila explained that life choices made by her daughter, had an impact on Sheila's health. Relationships that her daughter had in the past and present had been with men who either were involved in sexually exploitative work or had been accused of a sexual crime. Sheila explained that having been subjected to sexual assault as a teenager, had meant that exposure to details from her daughter about her partners had an impact on Sheila's symptoms, and the relationship with her daughter.

Sheila: […] it had a massive impact again. More damage to the relationship. More damage to, sort of flashbacking again with me and…then with part of the FND issues I have. I have really bad problem with speech at times and so that really flared up big time again […] Err, it doesn't happen immediately. It happens within a few of days of it. ... And of course, you know, within a couple of days my daughter's been in touch and then she realises that I can't talk. And then she feels guilty […]. It's not at the moment of the stress, it's as I'm coming down from the stress that it becomes really apparent. That I become quite inarticulate, […], that's the reason I ended up in hospital – the one back in August […].
Researcher: And how does thinking about the event make you feel?

Sheila: Angry and disappointed. Sheila, p.10

And although Sheila described a difficult relationship with her daughter, above she also highlighted that her daughter was concerned about her health and Sheila perceived her to feel guilty knowing that she had contributed to her mother becoming unwell. Sheila also mentioned not feeling *protected* by her mother. In trying to separate herself from her daughter's situation, Sheila felt that she was being a poor parent. Perhaps feeling guilty that she failed her own daughter by not being able to prevent her from being in unhealthy relationships. This indicated to beliefs around what it meant to feel protected and being a 'good' mother, that were not being fulfilled on her part, because even though she accepted that she was not responsible for the decisions made by her daughter, those decisions had an impact on Sheila's life and health – Sheila would have to deal with the aftermath.

And I felt like my mum never protected me and therefore I felt I wasn't protecting my daughter. And I think it was all of that, you know that was coming back on me. I think I've got quite good insight into that and err, I know what I'm responsible for and I'm not responsible for those things that have happened. But you're still left with the baggage. Sheila, p.53

Parveen did not indicate feeling inadequate as a mother or difficulties in the relationships with her children. In fact she mentioned that her son that lived with her supported her when she experienced seizures. A life event that Parveen spoke about was when her son-in-law's mother died. She mentioned wanting to support her son-in-law and daughter when she found out the news. Parveen had a close relationship with her son-in-law's mother, but her focus was on being there for him.
He, my son, will get me whatever I need. He tells me to sit down so that I don't fall and to tell him what I need, and he'll get it for me. Then after an hour or so I'm usually fine. Parveen, p.9

I was quite upset, err, even though…..my kids were very shocked. My daughter was really shocked. We went to their house. To console them, it was his mother [that died], my son in law's. [Inaudible]. No I didn't have a shake [meaning seizure], thank God – obviously I was upset about [son-in-law's mother]. Parveen, p.10

Working is harder with non-epileptic seizures

Several participants (Simon, Ashley, Mark, Samantha, Sheila and Chloe) alluded to the way in which NES/FND had affected their ability to achieve work related activities. There was a sense that they had either relinquished certain activities (Simon/Ashley) or had modified their approach (Mark/Samantha), so that they could manage with NES/FND. Sheila and Chloe felt that they had faced discrimination because of their ill health.

Simon was self-employed, and this had been both positive and challenging in his situation. Being self-employed had allowed him to choose jobs that he felt were manageable for him, but the work he could do became more limited over time and had an impact on his financial situation. Simon referred to his life changing *overnight*. The dramatic change was perhaps not only referring to becoming physically incapacitated, but that overnight his whole life changed because neither he, nor his family, returned to their home and he was no longer able to work once discharged from hospital. So, whilst Simon's focus in this quote was about having an active/physical job, there was a sense that this contributed to his idea of being a provider, as a husband and father – a masculine identity.

*Yeah. No, we talked to each other about and I think because you know we'd been, as a family, living through it for the previous – at that point nine, nine months or so.... And seeing me get err…. progressively worse – symptom wise. Because I was very active. [...] for a few years, self-employed. So active, very physical. And I was getting to the point where I was having to reduce work to the amount or being very picky about the work that I was choosing because I knew [...] that would aggravate the pain more […]. So that was really frustrating and then we got to December. Well, yeah literally overnight I went from being, okay slightly less functional than normal, to…**overnight** I feel like my life changed. I went from being active to – bedbound.* Simon, p.10

When Samantha was interviewed, she had begun to prepare herself to return to her teaching degree. Whilst Samantha did not talk about teaching in a way that indicated it was important to her identity, it was clear that her decision to leave her training was because she became disabled. Samantha had tried to use strategies to reduce the burden she felt, to reduce the impact on her levels of stress. However, she struggled to stick to this approach when she felt the urge to get work completed, and then suffered the repercussions of doing so.

I was feeling really anxious, and I had to break everything down. So, I'd have to do like maybe two hours a day on my assignment, and just leave it at that. Otherwise, I would have got too stressed out. And there were days when I was getting close to the deadline when I thought, 'you know what, I'm just going to do it all day.' […] And when I did it all day then I'd end up having a seizure at the end of it [....]. It was causing me a lot anxiety and stress, so I had to reign it back. Samantha, p.4

Chloe was managing with fibromyalgia and NES/FND, and although she had been told that she would be supported at work, in her opinion the 'reasonable accommodations' were not made fast enough to allow her to work without having to take time off sick. She expressed a sense of feeling inadequate, because she had been unable to fulfil her role due to being restricted by her work environment.

I felt like I couldn't do the job that she hired me for if I'm honest... I felt like it was a nuisance to ask…err [6 second pause]. I dunno. She made me feel supported in the, in the moment and she said that would be fine, it would be completely okay with everyone and with the team. But that was another thing, she, she never explained to the team what was happening I don't think. I mean people I spoke to in the team, I told them I had a disability, but I don't think anyone fully knew, what the severity was or what, if I had a bad day and I'd spoken to her and she said 'I'll let the team know that you need to be in the office today, or whatever, because your legs are sore' but that just never.... really got through [...]. So, if I'd come into work and say 'do you mind if I sit in the office today? I'm having a really bad pain day'. People that would have to go stand out would feel well 'why is she getting to go sit down?'. […] There was no communication between the manager and the rest of the team, and I didn't feel like that was my place to do that. Err, so yeah, I felt a bit of, like a grudge or …. Err, judged a wee bit, from the rest of the team. Chloe, p.23.

Sheila had also experienced difficulties with management. In her account she described a mismatch between feedback in supervision meetings, and a sudden revelation from her manager telling Sheila that she had been underperforming in her role.

I just think that's so unjust – really unjust. Err.[…] And I don't think it's me being naïve at all. I, [sigh] even the union and the occupational health were like 'what is this actually about?

*There seems to be a hidden agenda there.'. and I was like 'I'm glad you're seeing it because I've been accused of been quite overthinking things and reading into things too much'. […] So I started doubting myself thinking 'am I? Maybe I am.' Err 'maybe I am' [laughs]. 'If a few people are saying this maybe I should listen to that' and err…yeah. But then when I started showing my husband the supervisory and the things that were being said he was like '**No**! No you're really not' and I was like 'well I'm glad you're seeing that and it's not just me imagining it'. Because when enough people say it to you, you start believing it*. Sheila, p.12

There was absolutely no prior warning. It was a video conference. She just phoned me up and said 'can we have a chat'. And I said 'yep, no problem'. […] And she said 'well I'm really concerned about your memory and your cognitive abilities. She said 'some of your decision-making skills have not been good'. [...] And because I was so distressed by this – because it felt like such a personal attack. […] I was inconsolable. You know and I was like ' I just don't get it. Where's this come from?'. And you know she brought up different examples and at the time I just couldn't defend myself because I was just feeling so overwhelmed. And I wrote back, err, within the next day and put down my sort of thoughts and questions about it. […] And I says 'well, actually that was my professional, clinical decision and I still stand by that. [...]'. Sheila, p.12

Ashley identified relinquishing her driving license, a direct consequence of developing NES, as a loss. She described grieving over this, something that she had worked hard to achieve, and this had an impact on her independence, work opportunities and daily life. Ashley explained that the impact was not just on her, but on her husband also. She now relied on him to achieve some tasks, and the consequences of not being able to drive brought the stress and pressure of finding somewhere else to live. She was also faced with the possibility of reduced work opportunities for her because using public transport limited the distance she could travel. Whilst Ashley reflected on how this may be unfair to her husband, it may also be that she felt it was unfair on her – having to surrender her license (independence) for something outside of her control (NES).

And I rang my mother-in-law and just had a good cry down the phone [...] Yeah had a good cry - let it all out and then I was ok. Like I still sometimes think 'you know what, I really fancy a trip out to [shopping centre] and a McDonalds.' And then I'm like 'nope!'. [Laughs]. Err, which is frustrating, but [...] I don't think about it all the time. But it's just things like,

[...]. We like live in a little town that's quite a way out from most places. [...] now we are looking at moving to be closer to the university, err because I can't. Because I can't drive there now. So that's like a bit str, a bit of pressure – because like we've kind of got to get this move sorted before September… and. And that's on my mind quite a lot. And err, and then there's that worry of not having the freedom to go on as many different placements. [...] but, just little things like I was saying earlier. I used to just get up and think 'do you know what, I fancy going shopping. Or I fancy going to see my friend'. And now it's like 'oh' – I can't just do that. [...] Even like doing the food shopping [...] before I could do that. And now it's all on [husband] which feels a bit unfair. Ashley, p.28

Researcher: Do you remember how you felt physically at that time?
Ashley: [pause] just really sad. Just really like, err, like a shell of a person really.... yeah… I don't know really how to describe it. [...] Yeah I think loss. Yeah it was kind of like – a bit like grief. You know like I'd lost this big – you know it was quite important to me that independence. It took me like five attempts to pass my test [...] [laughs] so to have that gone in - just like that. Err. It was just like a bit of a shock yeah – that feeling of grief. Ashley, p.29

Summary:
Participants described events that had an impact on them emotionally, and on seizure episodes in some instances. Participants expressed how some events has made them feel scared, anxious and relieved (Samantha), angry and disappointed (Sheila), frustrated and helpless (Simon), guilty (Simon and Mark), judged (Chloe), sad (Parveen).

Some participants reflected on how everyday events had an impact on aspects of their identity, such as being a parent. This indicated a longer term feeling of inadequacy that participants related to their condition. This was also reflected in work identities for some individuals – acting as an ongoing stressor perhaps.

Therefore, using different examples, some participants highlighted how NES had affected their lives beyond individual events, and the impact was not just on those around them (children, partners, colleagues), but the impact was on their sense of self – an aspect of their identity.

E. HOW DO I LIVE BETTER, WITH THIS?

Five participants (Ashley, Bryony, Parveen, Sheila, Simon) conveyed there had been positive experiences arising from NES. Experiences varied amongst participants, with Sheila contributing most.

Raising awareness helps me to fight back

Participants (Simon, Ashley, Sheila) described using social media to raise awareness of FND. On the surface this appeared to be an altruistic act, helping others because their experiences had not been positive. However, it was also of benefit to participants themselves. It provided a platform for participants to express themselves, perhaps a form of catharsis, and was also an opportunity for participants to fight against the misinformation about FND that they perceived to be around.

Ashley and Simon indicated similar motivations towards writing blogs about their *journey* with the condition. They both mentioned an interest in *raising awareness* and Simon mentioned that he and his wife wanted to act as *advocates for FND.* In the extract below, Ashley speaks about how the act of writing has also become a coping strategy for her, showing that the task has two purposes. It helped her to gain a sense of *perspective*: that what had happened was out of her control or in the past.

My wife and I have become real advocates for FND……We just want to make more people aware, so anything we can do – like this. We've started our own Facebook blog recently…. to document our journey. Just because we want to make people aware of it, so anything we can do. Simon, p.40

It gives me a bit of perspective. Err. So when I write it all down then I'm kind of in more in a problem-solving mode rather than panic. Err, so when I see it all on paper, I think 'okay. That's happened. There's nothing that I can do about it now. Err, so put it away'. Or, if there is something that I can do about it…Like, so now I've set up a blog about FND to kind of raise awareness. And I think it needs raising, I think awareness needs to be raised. I think more people with it need to get their voices heard. Ashley, p.35

Using different platforms was also a way to *fight back*. Here Sheila talked about fighting FND and helping others too. That is, empowering one with information allows one to challenge misconceptions they may have about the condition. And empowering oneself with techniques to cope with seizures, allows one to fight against the experience of non-epileptic seizures.

Nobody taught me anything. Nobody was helping me with them. Nobody said like, how to fight it. I was just like 'I need to find something'. Or hold on to because I felt like my sanity was just, just going. […]. And I think that's where my, my err, support comes in with the group. Because I am so lucky that I can access, and I know how to access, and I'm really committed to finding a way through this. I don't just want to accept that this is inevitable. Err, I'm trying really hard to point. And sometimes it wins, sometimes it doesn't but I like that I can try and empower others to give them that little belief in themselves to try. To fight this. Sheila, p.44.

Having non-epileptic seizures has helped with understanding others better

Sheila mentioned positive factors had also arisen due to having NES. For example, the relationship that Sheila described with her work colleagues contrasted with the relationship that she had with the children she worked with through her professional role. She did not feel welcome in the team, describing a *clique* that she was not part of. She acknowledged that dealing with the child protection aspects of the work was unpleasant, but despite this was positive that she *loves* working with the children. Interestingly Sheila did not highlight that child protection issues were a trigger for seizures. Although her first seizure had occurred at work.

Sheila tried to see the positive side to her difficulties, and although she would rather not have experienced NES, she believed that her experiences had placed her in a better situation to empathise with others. It was as though she was expressing that there was some comfort in knowing that others had similar experiences.

Even though I still wish it had never happened but, but it has. So try and make the best out of it. It's like experiencing a panic attack or experiencing anxiety. It's like, now I can really empathise with people - when they're telling me these things.... I kind had an understanding of it before but. To actually walk that and experience it - in yourself. Phew! It's humbling. Sheila, p.31

Feeling empowered and confident about fighting off seizures

Sheila shared that she had developed *coping mechanisms* to deal with seizures. She had managed to use breathing techniques to fight having a seizure when she was undergoing a diagnostic test. These strategies gave her some confidence that if she was to feel a seizure starting, she would be able to stop or stall it from progressing. This was important not only because she hoped to be able to help others do this too, but because in another part of the interview she had indicated that she lacked confidence in herself.

But I'm hopeful, I'm hopeful that I've got some tools in my toolbox now to sort of, maybe fight them. I know with the seizures I've managed to ground myself with, err, basically doing the err, breathing exercises that I would do with the ladies in labour. Sheila, p.44.

Distracting self at times of emotion or difficult thoughts

Bryony focused on the fact that she had become more accomplished at bread making, and this illustrated that she had used practical activities to distract herself. She used such methods at times when she felt upset or confused. These activities diverted her focus to the activity, and not the emotional turmoil she experienced when she was unsure of what the outcome of a situation. For example, when her mother was admitted to hospital or when she did not receive her predicted grades after the cancellation of high school exams in the UK during the COVID situation.

I've gotten really good at baking. Because every time I had a thought that upset me, like, you know 'I've not seen such and such person', or 'now what am I supposed to do with my future?'. Or when I got my grades back and they were really poor…Err, and every time that happened, I just made bread. Err… Yeah, my brain copes with things in very strange ways: admin and bread making. But it did mean that I got through and I was fine. Bryony, p.23.

Spending time in the remembrance of God brings blessings and healing

When Parveen spoke about her seizures, she was neither negative nor complimentary about them. Her seizures were experiences that she had been managing for years, they were part of life for her. However, Parveen shared that she had not had a *big* seizure for over a year. She was humble in her analysis of why this may have been.

Parveen shared that she had been spending time in private prayer and supplication. This was a strategy that had been developed, after conversation with a group facilitator, from a group that Parveen attended to learn and reflect on her beliefs and religion. This is an example of how strategies that align with an individual's values and beliefs can make the activity feel less like a coping strategy, and more like a beneficial way to utilise their time. At a time when Parveen was not able to attend the religious classes because of the lockdown, this was an activity that Parveen was able to keep up with.

And it's been lots of years now - about 1 ½ to 2 years since she [group facilitator] suggested it. And I pray two voluntary units [of prayer] with every obligatory prayer. And I supplicate – nothing else really….. God gives us healing…….. So only God knows if it's the prayers or, and there's blessings in saying prayers. Parveen, p.15

Summary:

Participants expressed that not all aspects of living with NES were negative. For some it had meant that they had developed ways of coping with stressful times or had found meaningful activities to occupy their time with. Some participants had also started to etch out a new role for themselves based on raising awareness; and facilitating learning for those who may not be as equipped to find out the information pertinent to living with NES. In doing so, they also gained some perspective on the difficulties that they had faced.

The examples in this theme highlighted that whilst participants were talking about practical activities (writing blogs, breathing exercises, prayer) to cope or manage with aspects of NES, participants were alluding to how they conducted their everyday lives with NES. That is, these tasks brought a sense of accomplishment (baking) or control and confidence (breathing) at times when individuals felt they were potentially losing control over their daily lives because of seizure episodes.

F. LIVING WITH NON-EPILEPTIC SEIZURES DURING COVID

All interviews took place after the COVID pandemic had started, although the impact of the pandemic did not feature prominently for all individuals. In the extracts below it is clear that Chloe and Mark were most articulate about how the national lockdown had affected them. Socio-political events that occurred during this time period included the Black Lives Matter movement, and the impact of this was referred to (Chloe).

Feeling lonely during the lockdown

Three participants (Craig, Chloe, Mark) indicated that the lockdown situation had made them feel lonely, they missed spending time with friends and family. This was an issue faced by many people during the lockdown, not only those with chronic illnesses. In this subtheme, the only participant that lived alone was Craig. Chloe lived with her partner and Mark lived with his wife and children. Craig was happy to speak with the researcher and described this as a welcomed interaction since he was unable to visit family at that time. The quote here shows that he was looking forward to the time that he would be able to visit his family.

It's still very much the lock-in condition, isn't it. And my family, on my mother's side are basically, are both, err, what do they call…are err… [...]. High-risk. So one of the incidents that I've had is not being able to see my family [....]. Anyway, the point being that, [says a name], my mother, as I said has this high, high risk categorisation because she is over 70 and she has one or two illnesses, along with my sister. So it's not been possible to do the usual commute and family…rounds, essentially [....].So, yeah. I'm queuing them up though. I've got a few, I've got a few holiday plans lined up for when things get eased off. Craig, p.26

In the quote below, Chloe clearly states that she felt lonely. She and her boyfriend lived together but the pandemic meant that at first, she was furloughed by her workplace, and then made redundant. Her interview gave a sense that going out to work was more than receiving an income; it also had a social aspect – an opportunity to be around others since she had few friends where she lived and her family lived over 100 miles away. The type of relationship Chloe had with her colleagues was not obvious, but it was clear that she enjoyed her work. This was in part based on the interactions with customers. Chloe also indicated that the nature of the pandemic, an infectious disease, made her feel anxious about going outside. Her own health condition meant that she was in a risk category.

But then obviously as the months go on, it's just [sighs] lonely. It's very lonely. I mean I don't have family here, err, very few friends here. Err.... yeah, it's weird. Like I don't know why but I didn't think the pandemic would count as like a life event because it's happening to everyone kind of thing.... But yeah, it's had a huge impact. Err...Especially not being able to *get out and. Like I've got asthma and a couple other illnesses that make me prone to illness, err, so going* ***outside*** *has been very difficult.* Chloe, p.25

Mark referred to the lack of change during the pandemic lockdown. That being in the same environment everyday meant there was no break from the house or his family. Whilst he did not say that he felt alone, he indicated this by mentioning that he *misses friends*. The *whatever* may refer to colleagues or the time spent alone commuting. So in this way, it was not only about being around people- but the opposite too, having time to oneself.

But the lack of change in environment - I think it's actually… quite a subtle thing. It's not as simple things as, you know people say like you miss your friends, or whatever. I miss… the changes of environment – the breaking up of the rhythm. Every day is very much like the next day. And that's a real problem……. You can you lose track of time. Mark*,* p.31.

Under the cloud of COVID

The feeling of being trapped in the situation was expressed clearly by Mark. He described finding it difficult to differentiate between events that had occurred, not only because they happened close together, but because they occurred within the setting of COVID. Mark highlighted that moving house, something that was supposed to be a positive event, became less positive under the cloud of lockdown. He felt stuck in the house, even though he was able to go outside for walks with his children. He described how events seem *compressed* and *low grade*, rather than individual major events for which emotions could be clearly identified. The events were hard for him to separate out, and so too were the emotions associated with these. Altogether they made him feel trapped and *burdened* by the situation.

It's all a continuum - it's all kind of like again, you know my cat died, then we moved house and then we went into lockdown. It is all been compressed into, you know. It's not like they were, you know separate life events, err, you know spaced out. […]. It's not like these little, individual kind of events - with stuff spaced out. It's these ongoing, low grade, burden stuff. Mark*,* p.33.

Impact on health from delays in accessing healthcare

Access to healthcare services was limited for everyone during the lockdown, this was not a specific issue for those with NES. However the impact on participants in the sample (Bryony, Ashley, Abigail, Simon) was referred to. For example, appointments with HCP were either cancelled or held remotely. In some ways this made services more accessible because there was no travelling involved for those with mobility difficulties.

The diagnosis of NES was made over the telephone/video call for Bryony and Ashley. Ashley described how her diagnosis was made using videos her husband took and sent to the neurologist, but the pandemic meant that she was unable to undergo the planned MRI. Whereas other participants (Abigail) were not limited by the pandemic and were able to receive a diagnosis face to face.

For participants like Simon, having appointments cancelled meant that he missed out of treatment that may have helped improve his quality of life. Abigail had experienced problems with accessing other healthcare services, unrelated to NES. And the stress caused by waiting had resulted in Abigail experiencing an increase in seizure events. These experiences may have led to fears about not being able to gain the advantage of time back in acting quickly to treat health problems. In the case of Abigail, this was felt acutely. It was a situation that she thought about *all the time.*

I can't remember the exact timeline but at that point we were waiting for a further pain clinic appointment. [...] And then with COVID, that's just …. knocked a lot of appointments out of the window. Simon, p.14

Oh the waiting, just the waiting plays on my mind a lot. Err, does make me very anxious which I would say does lead to more of my... drop attacks, or the non-epileptic, it's all playing on my mind. I'm just conscious with time going on and I'm thinking 'God this situation we're in, it's not, its how much longer? My age is going to.' I do panic a lot about it, and I think that does, because I panic, my mind is constantly. I don't think it's helping with the, the non-epileptic seizures, [...] Cos I feel in limbo. Like I'm waiting to go back on the NHS. And it's I, like when I'm ringing the NHS, they're telling me that the department's closed because they're closed because of COVID. And [...] I'm going round in circles, no one is doing anything for me you know. I feel like I'm going to be at the bottom of the list [...]. Abigail, p.6

Lockdown made it harder to achieve tasks and events

For some participants (Mark, Chloe, and Bryony) it appeared that lockdown made it harder to achieve certain tasks. Chloe indicated that her motivation to exercise, something that was beneficial to her health, had been affected. This illustrated a domino effect for her: having to stay indoors meant less space to exercise, not enough exercise made her body ache more, having an aching body made her less motivated to go out for exercise.

Err, and also with us both been stuck in the house – we only live in a one-bed flat and it's quite small so there's no room to do any exercise or err. I try and do yoga as much as possible but up and till maybe a couple of days ago, I hadn't done any since lockdown. And it's just ***ruined*** *– ruined my body. Err [sighs] just really achy.......... I have zero motivation to do anything.* Chloe*,* p.26.

For both Mark and Chloe, the pandemic was to blame for the way that they felt. Mark stated that the situation had made him less motivated (less *positive*) or inclined towards carrying out the DIY tasks in his new house. The lockdown may also have made it harder to carry out these tasks if the hardware shops were not open. In this way, Mark too was stuck in a loop, similar to Chloe.

One of the main life events for Bryony during the study period, was that she was due to sit her A Level exams. Due to the COVID pandemic, the exams were cancelled, and Bryony described being left in a state of limbo and anger. It felt like a *weird* and *confusing* time for her because she did not sit the exams she had been preparing for. Bryony then faced a situation whereby her expected grades were downgraded. It was a frustrating and stressful time for her because she was unsure of what the future would look like. The work that she had put in was not being reflected in the new grades that had been allocated to her. The situation was resolved for Bryony when a new system of calculating grades was used and she secured her place at the university of her choice. She had not mentioned goals and aspirations beyond university and so it is not known what the impact may have been for her, had she not been able to attend. However, to Bryony it felt like an unfair, unbelievable and *undeserved* situation to be in because she had worked hard to achieve good grades all year. Bryony was the youngest participant in the sample, and this experience may have felt like her whole future was at jeopardy because she was just beginning her journey in further education.

My postcode was in fact was given preferential entry, err to some of the unis that I applied to. Which worked in my favour there. But when I got my grades, obviously the algorithm wasn't on my side. I ended up with a C, a D and a U…. And obviously after the weekend, on the Monday I ended up with three A. Err, so…... That Friday/Saturday/Sunday and then up to Monday morning was,* ***not*** *very fun. Obviously, I'd missed all my offers for university. Err, so I had to work out what I was doing next because clearing wasn't running. [...], no one knew what was going on or where anyone was going, [...] I was just, I was at a complete loss….* Bryony, p.24

Reflecting on the experiences of others

Participants in general had indicated how the pandemic had affected their lives at a personal level. However, two participants (Mark and Chloe) had considered the experiences of others during this time. Mark reflected on the loss of experiences for his children and feeling particularly regretful for his eldest daughter. Mark showed that he was empathetic towards the multiple experiences that his eldest daughter had missed out on. These were circumstances outside of his control, caused by the Government lockdown. And whilst he expressed that the situation was *unfair* for his children, it may be that he felt it was unfair on them all. The repercussions of the situation were felt by all, including him and his wife who were unable to fulfil their planned activities. This added to the guilt that he felt about being a poor father.

So she missed out on loads of you know all the stuff they do at the end of term. Or she was meant to be going on err, her Christmas present, we were going to take her to a Harry Potter studio tour. That didn't happen. We were mean to be going camping around the time of her birthday. That didn't happen. Just loads of stuff, you know. Yeah. She's not been to school for months, err. So yeah that's just been, again, rubbish for her [….]. Mark, p.28

Other than COVID having an impact on individuals, Chloe spontaneously talked about how the recent awareness of the Black Lives Matter (BLM) movement. The events in America in May 2020 had an impact on her and her partner. The recent BLM events (George Floyd's death/protests across the USA) took place during the pandemic and at the time Chloe had been furloughed from work. This allowed Chloe and her partner to spend time researching and trying to understand the events that occurred. Chloe shared a sense of disbelief that some individuals denied that discrimination occurred based on skin colour. She did not understand why some people did not want equality for all. It could be that this movement had an impact on her because she shared a sense of being discriminated against. From the interview there was no indication that the movement has caused Chloe to reflect on her privilege as a White person, but it had led her to want to understand what had activated the events in spring 2020. At the time of the interview, it appeared that she was still processing the impact and what it meant.

And other things like the Black Lives Matter movement had a huge impact and still is having a huge impact on me. Err, just seeing everything that's going on in America. p.26

I just…. [Sighs] I dunno. I think. Err. I dunno, it just makes me really sad and there's people arguing that, this movement isn't needed, and that Black people and ethnic minorities have got the same rights as everyone else and I'm sitting here watching these things happen and I'm like 'are people ***blind****?'. Like 'cannot you not see what's happening?'....... I just wish. I just wish everyone was* ***nicer****. Especially during this time... It happening during COVID, it blows my mind right now. Like everyone is just trying to survive right now and then people are getting killed for no reason.* Chloe, p.26

Summary:

All participants were interviewed after the COVID pandemic had begun and lockdown in the UK was relevant to most times when participants were interviewed. However the impact of the lockdown and pandemic were not referred to by all participants.

The areas in which an impact was experienced varied; in most instances the reflections were related to the individual – such as access to health services being limited, but others (Mark and Chloe) also considered how events affected those around them. It was also clear that the impact of the pandemic had been similar to the way in which other individuals with a chronic illness may be affected; that is, the issues presented here did not seem to be specific to those with NES.

4.11. Participant's perceived impact on seizures from emotional responses to events

The present study aimed to investigate how pwNES experienced and understood their emotional response to a specific life event that had occurred over the past 12 months. With an aim to understanding the extent to which pwNES perceived a connection between their emotional response to a life event and their seizure episodes.

Participants described a range of events that are reported in table 15. In describing events and the impact of those events, four models emerged. Examples are provided below to illustrate how the models were described by participants. Table 17. illustrates the models participants presented when speaking about events they experienced around their seizure episodes.

Model 1: An event occurred → an emotion/ reaction was experienced → a seizure occurred.

Participants mentioned a variety of events that induced stress and/or anxiety for them. These were events such as moving away from home (Bryony), completing coursework (Samantha), giving up extra-curricular activities (Ashley), stress related to going to hospital (Simon), or a redundancy interview (Chloe). In the extract below, Abigail illustrates how learning about fertility difficulties had a triggering effect for seizures.

Oh the waiting, just the waiting plays on my mind a lot. Err, does make me very anxious which I would say does lead to more of my... drop attacks, or the non-epileptic, it's all playing on my mind. I'm just conscious with time going on and I'm thinking 'God this situation we're in, it's not, its how much longer? My age is going to.' I do panic a lot about it, and I think that does, because I panic, my mind is constantly. I don't think it's helping with the, the non-epileptic seizures, you know. Err...Cos I feel in limbo. Like I'm waiting to go back on the NHS. And it's I, like when I'm ringing the NHS, they're telling me that the department's closed because they're closed because of COVID. And I'm just like, I'm going round in circles, no one is doing anything for me you know. I feel like I'm going to be at the bottom of the list and there's lots of people in front of me. Abigail, p.6

Based on the information that participants shared in their interviews, it is likely that for some individuals (Abigail, Ashley, Bryony, Chloe) this formulation was influenced by their interactions with clinicians and the explanations they received regarding their diagnosis of

NES. For example, Ashley indicated that the ‘hardware/software’ explanation for NES had been used in her appointment with a neurologist. Sheila also indicated having knowledge of this theory when she referred to ‘this whole software issue’ but did not expand on how she had become of this explanation for NES. Abigail, Bryony and Chloe did not use the brain as a computer analogy, but it was clear from their interviews that their understandings of NES were based on that which their respective neurologists had told them about NES. Conversely, Mark and Simon gave the impression that their understanding of NES had been formed through their own research, even though Simon referred to his ‘brain sending the wrong information’ and Mark spoke of his brain being ‘misswired’.

<u>Model 2</u>: An event occurred → an emotion/ reaction was experienced -x→ no seizure occurred.

This model was relevant to participants Craig and Parveen. A life event that Craig had identified was his godmother having to attend the hospital emergency department following an accident at home. In the extract below he described his reaction to finding out the news after the event had occurred. In the example from Craig, he did not consider the emotion relating to the event to be associated with seizure episodes that may have occurred around that time. He perceived any episodes at that time would have been coincidental, and not in response to the emotional reaction he had.

Craig: Yes, so there was an emotional reaction. I sympathised with her. So, I knew what happened. I know her well enough to know what had happened. [….] A genuine accident.
Researcher: Yeah, sure. And, what about, do you remember what your seizures were like around that time when you’d found out about her having to go to A&E?
Craig: [....] I didn't consider my seizures either at the time that it was mentioned to me. So I wasn't actually thinking of the two things. [….]. I wouldn’t say there was a difference in pattern [audio breaks up]. A coincidence yeah. I do believe it relates to coincidence, yes.
Craig, p.14

Parveen had experienced two bereavements of close family and friends during the study period. Parveen described sadness and grief but also explained that she had not experienced seizure events at those times. Her emotional responses to the events had not resulted in seizure episodes, for her.

Model 3: No event occurred → an emotion/reaction was experienced → a seizure occurred.

This model was described by two participants, Ashley and Craig. They gave examples of experiencing a seizure and emotion, but not being able to anchor the emotion to a specific event. Craig explained that the emotions (guilt, anger, hate) at the time of the seizures were misplaced. From his understanding, these were coincidental factors. In this extract Craig explains that the guilt he feels is an involuntary reaction, much like the physical aspects of the seizure. He describes the emotions he experiences at the time of the seizure to be detached from the seizure – that there are two mechanisms operating alongside each other.

The physiological responses which appeared to be somewhat involuntary have resulted in a guilt response, a blushing that is not the same as the irritation that I mentioned. Actually two subtle different reactions. In addition to this guilt which is an important part there is some considered hate or anger that [audio breaks up]. […] I have experienced them so many times that for the fact that they are essentially basically the same there appears to be no. It is a complete detachment, there's no, erm, sensitivity or calming of the process it appears completely disassociated. Like a. like running a car on a broken ramp. There's just two different mechanisms going on at the same time. And, so there's literally a complete loss of, complete detachment. So it feels like that. Craig, p.15 & 22

In the extract below, Ashley describes how she experienced seizures at a rare time of rest. She explained that in hindsight she was able to see that she had experienced a busy, stressful and demanding period leading up to the seizures occurring. Ashley states that her seizures occur when she feels *relaxed*. That is, in the absence of stress or an event, the system was derailed, and seizures were triggered.

So… [Sighs] I personally feel like - especially because usually my seizures happen when I'm relaxed. That I'd had such a full on month and I hadn't stopped, and I'd been so busy. I'd been doing all this volunteering. I was doing all this uni work stuff, […] I was having some problems with some of my friends. Err, they were needing quite a lot of my time as well. And I just had so many things going on. And then it's almost like I, I had that one day to relax which very rarely happens and it was just like 'oh! This is what happens when you *stop. It's nice!'. [Laughs] Err, do you know what I mean? It's like… almost like a shock to the system to have been doing. You know, if you were to be driving in a car and you pulled the handbrake – it'd kind of veer off and it's kind of a similar thing.* Ashley. p.38

Model 4: No event occurred→ no emotion/ reaction was experienced → a seizure occurred.

Three participants, Mark, Parveen and Simon, described seizures that occurred when they were not aware of any specific emotions being experienced. Mark described experiencing seizures that woke him from sleeping, and Parveen (see below) explained that she experienced seizures when she was in a *normal* state. Simon described seizures occurring that were not related to any event. The warning sign was usually when he or his wife noticed his leg begin to twitch.

It doesn't happen because of happy things or worrying things. It just happens 'normally', it just happens in my body, and I end up [experiencing] it. I have no idea. Nothing happy, no worries, nothing. When it happens, I'm 'normal' and it just happens in my body – not related to anything happy or sad. I can just be sitting down, and I can't tell, and it happens. Parveen, p.4

Table 17. Model relevant to each participant.

	Model 1	**Model 2**	**Model 3**	**Model 4**
ABIGAIL	X			
ASHLEY	X		X	
BRYONY	X			
CHLOE	X			
CRAIG		X	X	
MARK	X			X
PARVEEN		X		X
SAMANTHA	X			
SHEILA	X			
SIMON	X			X

From the table above it is apparent that most participants (n=8) gave examples consistent with model 1. That is, experiencing an emotion or reaction to the event and perceiving this to have an impact on seizure episodes, was the most common understanding that participants had of a link between their emotional response and their seizures. Model 4 was the other model that was relevant around a third of the sample (n=3). This model referred to those seizures that occurred when there was no clear event or emotion experienced by the individual. Both models 2 and 3 were referred to by two participants.

Models 3 and 4 included examples from participants where they experienced a seizure at a time of relaxed (Ashley) or felt normal (Parveen). A possible explanation for these seizure types could be the sympathetic activation experienced during the build-up to a seizure, is replaced by the parasympathetic response that provides reprieve from the increased levels of arousal (Brown and Reuber, 2017). This corresponds to findings from studies that have reported seizures to be 'associated with parasympathetic activation' (van der Kruijs et al, 2016), as commented by Reuber and Brown (2017).

Summary

The examples in this section demonstrate how individuals made sense of their emotional responses that they did (or did not) have around the time of a life event, and the extent to which they perceived this to have an impact (or not) on seizure episodes.

4.12. Discussion

The present study aimed to investigate how people with NES (pwNES) described and made sense of their emotional responses to a recent life event, and whether this was perceived to have an impact on their episodes of non-epileptic seizures.

Summary of findings

The process of receiving a diagnosis was a different journey for each participant, and it was an event in itself for some. Those participants that spoke of this expressed that the diagnosis bought about several feelings for them. There were feelings of relief upon receiving a diagnosis; but then confusion and disbelief after the initial feelings of relief dissipated. Even for those participants that appeared to accept, rather than fight against seizures, the seizure itself was described as a disruptive entity.

Experiences with healthcare were mixed; several participants had negative experiences, and two participants gave examples of positive experiences with healthcare services. In the latter case, participants shared details of events that indicated that HCP had gone out of their way to make the service more accessible or had considered the needs of the individual when conveying the diagnosis. For those that recalled negative experiences, attitudes and treatment from HCP made participants feel that they were perceived to be feigning seizures and wasting the time of healthcare services. Experiences related to healthcare services were traumatising or punitive events for some individuals.

The impact of the emotional responses on seizure episodes was clearer in some instances (cat euthanised, poor treatment in hospital), but in other instances there appeared to be a long-term impact from the event(s), that possibly acted as a maintenance factor for seizures. For example, ongoing difficulties at work, or difficult family relationships may indicate ongoing stress/anxiety for individuals. Some participants indicated that their seizures appeared at times of rest that is, after a period of hyperarousal.

For many participants, NES was a condition that permeated most if not all aspects of their lives. The condition had an impact on their identity, achieving their work and fulfilling goals they had for the future. However, some participants were able to express the positive changes they had noticed in relation to developing seizures, or ways they had developed to cope with or control seizure events.

All participants were interviewed after the COVID pandemic had begun and lockdown in the UK was relevant to most times when participants were interviewed. The impact of the pandemic was not referred to by all individuals, but for those for whom it was relevant, the affect appeared to have similarities to the ways in which others with a chronic illness may be affected.

A novel finding from the current study was that four models were apparent in the way in which participants made connections between their emotional responses around events and the impact on their seizures. Both female and male participants were represented across the different seizure conceptualisations. Although, male participants appeared to separate their experiences from that which they considered to be typical for pwNES. Male participants appeared to distance themselves from those for whom trauma or abuse may have been precipitating factors in aetiology.

Main findings

There were three objectives that the present study set out to investigate. Each objective is considered in turn, with the evidence from the themes that developed.

The first objective aimed to investigate how pwNES experienced and understood their emotional response to a specific life event that had occurred over the past 12 months.

This led to a novel finding that participants demonstrated using four different models to express their sense-making of emotion around events and seizures that occurred (see section 5.9). In two models emotion was not relevant to seizure outcome. Five participants demonstrated different models in relation to different events they experienced.

The subtheme *making connections to try and understand what happens to me* (GET A), provides examples of how participants demonstrated that incidents in their past (Chloe, Samantha, Sheila) may have predisposed them towards NES, and how these influenced their response towards recent events they had experienced. Other participants described seizures as a way in which the body reacted when overwhelmed by emotion (Abigail, Ashley, Samantha), a way of resetting when stress and anxiety were too much to cope with. For some participants there was a level of anticipation that their body was poor at managing stress and so seizures were inevitable progressive decline in their health (Bryony), or if a difficult situation was expected (pet euthanised) then a seizure was likely to occur after the event (Mark).

The second objective aimed to investigate the extent to which individuals with NES perceived a connection between their emotional response to a life event and seizure episodes.

This objective was concerned with the impact that individuals perceived their emotional responses had on seizure episodes. GET D (*LIVING WITH NES IS CHALLENGING*) brought attention to how the emotional impact from events may surpass the event itself. That beyond the emotional response to an event, the impact of the emotion felt permeated other aspects of an individual's sense of self. For example events related to not being able to work or drive had a lasting impact on some individuals. Losing a sense of independence or one's income not only brought financial stress but had an impact on an individual's identity, and self-esteem. This theme illustrated that connections between emotional responses to live events did not always have the impact on seizures at the time of the event, but that there was a link between the event and the emotion/ feeling that remained or developed for the participant. In this way the emotional impact may be seen acting as a maintaining factor in NES. Participants may not have made these connections in this way for themselves, but the lasting impact of emotion was seen in examples when they indicated experiencing stress in relation to work difficulties or feeling inadequate in their role as a parent.

The third objective aimed to investigate how individuals with NES described their emotions at the time of a life event.

The results from the present study found that when participants were called to reflect on recent events over the past 12 months of their life, emotions recalled were mostly negative such as sadness, loss, anxiety. It was also noted that events that were expected to be positive or happy occasions (ending therapy, moving house) were unexpectedly experienced as difficult by participants (Samantha/ Mark). However, there were some events when participants spoke of feeling hope. This was particularly relevant to the time of diagnosis. For one participant it was also in relation to being told that there may be some treatment available (Abigail). Some participants (e.g. Ashley, Chloe, Samantha) considered that feelings of stress or anxiety associated to an event were often triggers for seizure episodes.

The final theme *LIVING WITH NON-EPILEPTIC SEIZURES DURING COVID* showed that the impact of lockdown was more relevant to some participants than others. GET F highlighted that individuals may experience loneliness even when living as part of a household. Participants that spoke of loneliness did not indicate that there was a direct impact on their seizures. However for Craig, being at home was a factor that increased seizure activity, and lockdown ensured that he was unable to leave the house regularly. This highlighted that participants did not always make these connections between their reactions to an event (feeling isolated/lonely) and their seizures. In the subtheme *under the cloud of COVID* Mark spoke of feeling burdened by *low-grade*, on-going stressors that brought his mood down. That is, the general feeling was not of intense specific emotions, but that the situation was one in which the individual felt trapped.

Study findings in relation to the wider literature

In addition to exploring how the data answered the research question, the six themes developed through the analysis were further considered in the context of the wider literature available.

Experiences of healthcare

Diagnosis brought conflicting feelings for participants. It was expressed that there was relief in receiving a name for the condition and being told it was not life-threatening (Abigail, Ashley, Simon); however there was also confusion and disbelief because there was no cure or treatment plan described by neurologists (Ashley, Simon). Similar feelings have been reported by others with functional neurological disorder (Carton et al, 2013; White, 2016). Participants in the present study also described feeling hope and then frustration with the lack of information and support (Craig, Sheila, Simon). The feelings expressed in the present study by participants, were replicated by HCP in a study by McMillan et al (2014), indicating that both practitioners and patients find the diagnosis and treatment of NES/FND to be one of conflicting emotions. This is possibly due to the lack of information and clarity around how to treat NES.

Three participants expressed that being given a website to look up was not sufficient. For example, a participant described feeling they were *in limbo* after being discharged from a service because the service could not help or support them in a manner appropriate to their needs (Simon). This was expressed in the same manner by Thompson et al (2009) and Wyatt et al (2014). In contrast to this, Bryony reported an example of a good/helpful practise when she received her diagnosis. The consultation with the neurologist was followed up with a written account (email) of the meeting. This allowed her to refer to the discussion she had had with the neurologist and consider the information at a suitable pace to her, as a patient. Reuber (2019) outlines this as a method that he uses with his patients, and in addition to the letter, he includes a leaflet to the CODES website (https://www.codestrial.org/INFORMATIONBOOKLETS) so that the patient can access further information if they would like to.

Additionally, Reuber (2019) outlines how HCP can communicate the diagnosis to patients using a more collaborative approach by information sharing, rather than presenting the diagnosis in a punitive, dismissive or patronising manner. Based on the findings of the present study, this may be an approach amenable to some patients. For example, pwNES may feel that there is more opportunity to ask questions and understand the impact on their lives. From the present study it is clear that the condition has had a long-term impact for some. For example, Ashley spoke about the impact on independence (giving up driving); and two others (Simon, Chloe) indicated the financial implications of having to stop, or make adjustments for, working. If pwNES have a clearer understanding of such matters, they may feel more equipped with what to expect from living with NES.

There were two participants (Chloe, Parveen), however, that seemed to be more accepting of the NES diagnosis. These participants (Chloe, Parveen) did not describe questioning or fighting the diagnosis, therefore the lack of reported conflict against the diagnosis may indicate acceptance on their part. Both participants described feeling relieved when they received the diagnosis, but feeling relief on receiving a diagnosis was a feeling shared by other participants also. Therefore it is not known what was unique about the experience of relief to Chloe and Parveen, but it has been reported that individuals that are accepting of the diagnosis of NES have better outcomes (Mayor et al, 2012). It could therefore be speculated that perhaps in not facing a conflict, against the diagnosis, this may have contributed to the reduction in their seizure episodes. That is, there may have been a reduction of ongoing stress by virtue of having a name/reason for the phenomenon. Although this reduction in seizures for these participants was not immediate, as has been reported in other studies that described pwNES ceased experiencing seizures after receiving a diagnosis (Duncan et al, 2011).

Theme E (HOW DO I LIVE BETTER, WITH THIS?) provided examples of how individuals found ways to make the situation more manageable for themselves. This may be seen as demonstrating resilience in the face of adversity. But based on the experiences shared by individuals it seems more accurate to frame this as individuals were forced to find ways to carry on, due to a lack of support from services. That regardless of expressing feelings of failure, and having to make compromises in some areas, individuals strived to find ways to live with the condition. This highlights, that whilst the literature (Testa et al, 2012) speaks of individuals demonstrating poor coping strategies, the issue may be complex. If individuals are not provided with adequate support from services regarding evidenced based ways to cope, individuals may develop their own strategies – as has been evidenced by the participants in the current study. This GET perhaps also points to the practical coping methods individuals developed in response to the impact of events on their lives.

Outside of the diagnosis experience, in the present study, participants shared experiences of other interactions with HCP. Unfortunately several experiences were negative and had a lasting impact for more than one participant in this sample (Ashley, Sheila and Simon). O'Sullivan et al (2007) reported that in their study HCP in emergency care reported beliefs that NES was due to the patient's own behaviour. Similar experiences were expressed by three participants (Abigail, Ashley, Sheila) in the present study. These participants spoke about HCP assuming that symptoms were being feigned, or a change in attitude from staff when symptoms were endorsed as functional. Another participant spoke of the treatment received when admitted to the accident and emergency department, the change in attitude from the paramedic and the physical abuse that she experienced, because she was thought to be pretending. Those participants that did have negative experiences in the present study also made positive comments about HCP but they described negative experiences in more detail, perhaps suggesting that they focussed more on these events and positive interactions were part of the expected experience with healthcare services. Rawlings et al (2017) also found participants expressed experiences related to poor understanding of FND, particularly from paramedics and emergency departments. Therefore, the findings from the present study and the current literature does suggest that there needs to be more awareness and training so that HCP feel better informed and confident in treating and supporting those with NES and FND.

Shame and guilt

Shame and guilt were referred to by participants either explicitly or were inferred from their descriptions and experiences of events. Five participants referred to feelings of shame (Abigail, Ashley, Mark, Sheila, Simon) and four indicated feelings of guilt (Craig, Mark, Sheila, Simon). Three participants were present across both emotions (Mark, Sheila and Simon).

A review by Reuber et al (2022) highlighted feelings of shame, in those with NES, is not a new finding and although it is not relevant to all pwNES, it is a common factor that can be burdensome in its impact on the individual. Reuber et al (2022) explain that in order to understand the role of these self-conscious emotions, 'an appraisal based process model' (Tracy and Robins, 2004), is helpful towards understanding how the two emotions are based on different attributions related to the individuals sense of private and public self. Shame is experienced when a negative outcome is attributed to an internal cause, a perceived inherent characteristic of the individual that cannot be controlled, such as ‘I am bad at my job because I am stupid’, whereas guilt is felt when the cause of the negative outcome is attributed to an external cause, that the individual can control, such as ‘I am in debt because I spent too much money’.

An example was provided by Abigail in which she spoke of feeling judged by others. She perceived people to judge her to be acting as disabled because she wanted money for being unwell. This is similar to a finding by McMillan et al (2014) who reported that HCPs considered patients to lack motivation towards becoming better because they wanted to receive disability benefit.

The burden of feeling shame and being a *mess about* may act as an ongoing stressor in relation to living with NES. Ashley indicated not wanting to seek help for new symptoms based on the negative experiences with healthcare. In describing feeling shamed by HCP, Ashley may have been expressing that she did not feel worthy of using healthcare services. For example she described a reluctance towards using primary and secondary care services in the future unless she had a good reason to, a reason which she had yet to discover she said.

Guilt was expressed in experiences relating to parenting (Mark), or not being able to provide for one’s family (Simon). Participants appeared to attribute the cause to having NES/FND, rather than it being an inherent deficit when the impact affected their children/spouses. This may have related to ideas around masculinity for these male participants. An interesting area to explore with male participants may be the models they use to understand seizures and the impact the seizures have on their sense of self. It has been reported that men often struggle with accepting the diagnosis of NES and are more likely to attribute aetiology to biological causes such as epilepsy (Oto et al, 2005). In the current study male participants attributed some seizures to being triggered by experiences of pain. Further investigation may offer interesting insights regarding their emotional response to the pain, especially since it has been highlighted that men with NES may have experience of ‘silencing expression of strong emotion’ (Griffith et al, 1998). This refers to individuals silencing themselves as a safety measure in the case of abuse for example (Herman, 2001). Although, both Mark and Simon were empathetic that they had not experienced abuse or trauma of any kind. Nevertheless, if the pain is considered to be an external factor, and is an event that makes the individual feel angry and frustrated, it may help HCP to understand whether there is a perceived link between these emotions and seizure episodes, or whether the emotional response is perceived as a consequence of the episodes – so that psychological therapy can be effective in helping the pwNES cope.

Loss and grief

Attachment theory is considered at the foundation of how the loss of a relationship has an impact on the individual (Worden, 2010). To understand the nature of the loss, the meaning of the attachment is key, and this is linked to the type of relationship and how it develops. For example, Bowlby (1977) argued that attachment is based on a desire for safety and security, and attachment is an enduring part of the life cycle (Worden, 2010). In the current study two participants referred to grieving over a loss of a relationship (Samantha) and independence (Ashley).

Both participants indicated that they had worked hard on developing the relationship (Samantha) or achieving their driving license (Ashley). In describing the experiences, the participants referred to feeling *sad*, *empty*, *anxious*, helpless, and feeling *numb*. These emotions are highlighted as common feelings during the grieving process, and as part of this the grief may be experienced as a 'somatic or bodily distress' (Lindeman, 1944). Samantha may have been indicating a sense of feeling abandoned when she spoke about not knowing how to manage in the future, and this was something that caused anxiety for her. She described the impact on her seizures around this event and this is perhaps the expression of the distress felt in the body.

Insecure attachments can manifest in different ways, for example an individual may be overly self-critical, feel anxious or have perfectionist tendencies (Mikulincer and Shaver, 2012). The experiences shared by Ashley may illustrate this. Ashley also spoke about having to prove herself academically, and related this to experiences at school with her peers. She gave current examples in which she was particularly critical of herself if she did not achieve high marks. Ashley's seizure episodes occurred at times when she felt over-stimulated and at times of rest. She spoke of seizures occurring as a way of her body taking control when she was overwhelmed or not coping well, that is, she recognised that she was not managing with the levels of stress that she experienced. This highlights area in which she could be supported to develop ways of managing stress beyond the techniques that she currently employed (grounding exercises and distraction).

A third participant also indicated that they were grieving over the loss of their identity (Simon). This participant spoke about how their physical and cognitive capabilities had changed after developing NES/FND. Here aspects of masculinity may be relevant, the way in which he perceived to provide safety and security to his family. The loss of identity as a physically strong male head of the family, was replaced by physical difficulties that required him to be supported by his wife, physically and financially. Aspects of masculinity were undertones in the accounts from all three male participants and may have influenced the way in which they conceptualised NES. In trying to explore the importance and nature of the relationship a participant had with their mother (Craig) this was met with defensiveness and was a topic not open to exploration in the research interview.

The evidence from the current study suggests that this is an area that could be explored in therapy with pwNES, to help them understand how their responses to events in their lives may be influenced by the core beliefs they hold about themselves. It also highlights that there is a need for experienced psychologists to support pwNES to be able to do this work.

Alexithymia

Alexithymia has been associated with several psychiatric conditions (Feldman Hall et al, 2012), including NES. Some participants indicated struggling to describe emotions, however, participants were not asked to complete an alexithymia scale so it cannot be asserted with any certainty that participants were indicating alexithymic traits when they spoke of struggling to identify emotions. And it is possible that this could relate to struggling with finding the words (*brain fog*) at times.

Some participants clearly identified emotions around events when explored with them. For example, Simon spoke about feeling devastated following an interaction with a doctor during a lengthy period in hospital and being inconsolable after the event. It has been suggested that crying may be an expression of different emotions such as frustration or being unable to express anger; and relates to perceiving oneself to be helpless (Miceli et al, 2003). In this instance the participant's response may have indicated both the frustration of being told that he had to live in this way for the rest of his life, and possibly the feeling of injustice and anger that it had not been resolved. The participant expressed that he often became angry around his children. It has been suggested that anger manifests as a result of an individual feeling unable to act in a situation (Stevick, 1971; Eatough and Smith, 2006), and may be an indication of a maladaptive stress response that indicates poor coping

(Myers et al, 2013). This is an area in which psychological intervention may benefit pwNES by helping them to recognise and manage their emotions better.

Positive coping strategies

In the theme, *how do I live better with this?* participants (Ashley, Bryony, Parveen, Sheila, Simon) demonstrated that they had tried to find ways of coping with living NES. Participants demonstrated using task-orientated ways of coping. Task-orientated strategies have been described as effective in reducing the impact of stress on the individual and are considered a 'healthier' emotion regulation strategy (Myers et al, 2013). For example, Ashley and Simon had been using writing as a way of raising awareness about the condition, and Bryony had used bread-making as a distraction technique when she was faced with stressful times. Sheila had developed a breathing technique that she described as *grounding* her and allowing her to *fight* against seizures when she felt a seizure was about to start.

One participant (Parveen) appeared to speak about her seizures from a position of acceptance, in contrast to others who perhaps appeared to be fighting against the diagnosis and physically trying to resist in some way. It could be that in relation to Parveen's faith, as a Muslim, she considered the seizures to be a test in her life. Abrahamic faiths have the concept of being tested and that God does not test one with more than they are able to cope with. Those from faith backgrounds have been found to use religious coping methods when faced with stressful situations (Pargament et al, 2004; Utz, 2011). This may be considered as engaging in task-orientated activities, such as prayer and supplication.

Utz (2011) suggests that difficult events in one's life can often revive an interest in one's spiritual/religious beliefs. If the tribulation is considered as a means of becoming closer to God, then this may lead to a form of acceptance by the individual; illness is not considered a hardship for example, and is part of their condition (Utz, 2011). Craig also spoke about being engaged in the pursuit of learning and understanding religion, however unlike Parveen he did not indicate that this was a way of coping with the condition.

This theme perhaps illustrates the creative techniques that pwNES may employ to make their lives manageable and gain some sense of control back. It also raises the possibility that strategies may be more effective if they are based on the beliefs and interests of pwNES and could be a place of collaboration between the patient and therapist.

4.12. Strengths and limitations

Using online interviews was both a strength and a limitation.

Some participants had mobility difficulties and for some individuals travelling was a trigger for seizures. Therefore, being able to be interviewed remotely meant that participants did not have to travel to meet with the researcher. Additionally, it reduced potential costs for participants, because they did not incur any travel costs. Which can be difficult for those on limited incomes.

Using remote interviewing was a more protective method for the researcher, since participants were not known to her. However, it did mean that for those participants who lived alone, if they were to have a seizure episode the risk protocol would have meant calling an ambulance. From the interviews it has been established that calling on healthcare services for NES can have a detrimental effect for future use of services for some individuals.

Additionally, using online interviewing meant that there was data lost when the connection was poor. With one participant it was difficult to ascertain whether the internet connection was causing his speech to be delayed, or whether the participant was having a dissociative experience.

Participants were not asked to provide information about their family or history, and so contextual information, that could have informed understanding better, was not captured. It was attempted with one participant, because it was relevant to understanding issues raised, but the participant was not very receptive to this and was suspicious of the researcher’s motives for asking. Contextual information about family background would have been particularly useful with some individuals. For example, an understanding of certain relationships within a family may give important context to understanding reactions to certain events, but the lack of presence in accounts is the only fact that can be commented on.

Including a participant that did not speak English was a strength for this study. Having a multilingual researcher allowed the participant to speak for herself and avoided the use of an interpreter. Hearing the voice of the participant allows the researcher to gauge nuances that may be lost when using written material. For example, the tone of speaking can convey frustration, anger or humour. Beyond the language capabilities, having a shared background with the participant meant that the participant did not have to explain concepts or phrases – the interview was more fluent.

The idiographic approach used in IPA allows for the individual experience to be present, as well as the group being represented. That is, the analysis presents both converging and diverging examples to evidence the experiences that participants shared. A criticism of the approach may be the interpretative nature of IPA. The double hermeneutic whereby the researcher is interpreting and looking beyond that which the participant says. This may be particularly problematic when researchers do not have experience that relate to specific communities, whether this is chronic illness or ethnicity for example. The researcher did confer with supervisors for feedback during the analysis process, as a method to counter this.

The present study also demonstrated that regardless of the focus of the research, for some individuals it was opportunity for them to tell their ‘story’. A form of catharsis perhaps, after feeling unheard by those around them.

4.13. Recommendations

Feelings of confusion have been reported by patients after receiving a diagnosis of NES and there is no clear treatment pathway for HCP to follow (Mayor et al, 2011; White, 2016). In the present study, three participants expressed being dissatisfied with the amount of information and follow up after receiving the diagnosis. Based on the experiences of participants, and the current literature, it appears that the way in which information is presented can have lasting negative consequences. Healthcare professionals may need to consider how information will be processed by the patient, and what is most important for the patient to know about the diagnosis. One participant shared how receiving written information after a consultation was helpful for her, allowing her to peruse the information when she was feeling less anxious. Such strategies have been recommended by Reuber (2019). Therefore, following up consultations, especially at diagnosis, with a written account afterwards may be a strategy that HCP can adopt. This may help with patients retaining and absorbing useful information.

There may also be a place for raising more awareness of NES/FND through information campaigns across the NHS. This may aid HCP in becoming more confident in recognising and supporting those with FND and break down some of the misconceptions there may be in relation to the condition.

Feeling shame is a common experience in those with FND (Reuber et al, 2022). In the current study participants expressed feelings of guilt, shame, anxiety and depression. Additionally there were examples of rumination by a few participants, especially after poor experiences or delays caused by COVID with healthcare services. Psychological services may be able to work more effectively in helping individuals cope with and manage NES by understanding the lasting emotional impact from daily stressors such as healthcare interactions, family and relationship dynamics, as well as unexpected obstacles such as difficulties with other health issues unrelated to NES. There is also some evidence that by working on core beliefs, using cognitive behavioural therapy or self-acceptance group therapy, individuals may demonstrate a reduction in shame (Swee et al, 2021).

Exploration around ideas of masculinity may also have a place in future research. That is, more research with men with NES may help with understanding the impact the phenomenon has on them and their sense of self. This also leads to the issue of representation in the literature and services. There needs to be more work towards improving the representation of minority groups in research. This also needs to extend to access to services.

4.14. Conclusion

The present study reported a novel finding regarding how individuals with NES understood the impact of emotions and events on their seizures. Four models were described through which participants reported whether emotional reactions to events had, or did not have, an impact on their seizure episodes. Most participants described experiences in which they perceived a connection between events and emotional experiences to be linked and contributed to seizure episodes. In doing so these participants gave examples of NES consistent with the integrative cognitive model of NES (Brown and Reuber, 2017).

Themes found in the existing literature on NES, such as experiences with healthcare, shame, loss and grief were also represented by participants in the present study. Experiences with healthcare appeared to have a lasting impact for some, suggesting that not all life events are equal in their impact. From this sample, it can be inferred that the impact of life events was not always intense emotion, but instead there appeared to be an ongoing impact from some events, such as redundancy or a new diagnosis, that continued to act as a stressor for some individuals.

CHAPTER 5: GENERAL DISCUSSION

My work in this thesis set out to investigate the role of three key aspects of emotion processing (interoception, alexithymia and executive functioning) in people with non-epileptic seizures (pwNES) and to better understand how pwNES react to life stress, and how stress may have an impact towards maintaining their seizures. I conducted three studies with the following aims: to review the evidence for an association between interoception and other key aspects of emotional processing including alexithymia, in studies which had employed an objective measure of heartbeat perception to measure interoception (study 1); to investigate the association between interoception, alexithymia and executive functioning in pwNES and healthy controls (study 2); and understand how individuals with NES perceive and understand their emotional responses to recent stressful life events and the effect of those stressors on their seizures (study 3).

The following chapter will begin with a summary of the main findings from each of the three studies. Followed by a discussion of these findings in the context of the relevant literature.

Summary of main findings

Study 1: *A systematic review of the association between emotion and interoception, when measured using a heartbeat perception task.*

The findings from the systematic review were inconclusive. Overall, the studies included in the review were very heterogeneous: differing in their focus and choice of study populations. Furthermore, most studies were rated to be of a poor quality, and no studies were convincingly robust in their design. Findings were inconsistent, with some studies reporting significant associations between interoception and emotional/psychological variables and others reporting non-significant findings.

A further aim of the review was to determine which of the heartbeat perception tasks (mental tracking or discrimination) was most suitable for use with clinical populations. However, it was not possible to determine this from the available studies. The mental tracking method was the most commonly used paradigm to investigate interoception in studies that explored associations with emotion/emotional states, in both clinical and non-clinical populations.

Study 2: *Investigating self-perception of emotion in individuals with non-epileptic seizures: using self-report measures and an emotional Stroop task.*

This study explored the association between interoception, alexithymia and attentional bias in pwNES and healthy controls. It addressed some of the methodological concerns which were identified in the systematic review (study 1) but was also subject to compromises in its design because of the restrictions of undertaking research during the COVID-19 pandemic. In contrast to many of the studies included in the systematic review, the study was fully powered but a questionnaire measure of interoception had to be substituted for an independent psychophysiological measure of heartbeat perception, because of COVID-19 restrictions on face-to-face contact. Also, pwNES were recruited to this study online via a well-known FND charity, so a diagnosis of NES could not be confirmed. A prior study had been designed to compare the association between interoception and emotional processing in patients with NES (confirmed diagnosis from a neurology specialist) recruited from NHS neurology clinics compared with controls. This study, which addressed many of the methodological concerns, identified by the systematic review had to be abandoned halfway through data collection, due to the shutdown of face-to-face neurology clinics because of the pandemic.

The revised study was designed to explore an interactional model of dysfunctional emotional processing with hypothesised associations between self-reported interoception, alexithymia and executive functioning/attentional bias. If supported by the study results, the model would suggest a potential mechanism for the maintenance of symptoms, in pwNES. The NES participants reported significantly higher scores on measures of self-reported interoception, alexithymia, emotional dysregulation, depression and anxiety in comparison with controls. Differences in attentional bias between the two groups were less clear cut. Despite the clear differences between NES participants and healthy controls on many of the measures, there were no significant associations between interoception, alexithymia and attentional bias, in either the NES group or healthy controls. In fact, many of the correlations between the total scores of the three main measures were very small, and overall, there was very limited evidence from the study to support the proposed model.

The study itself was designed to be conducted within the constraints and aftermath of the COVID-19 pandemic so there were several limitations that may have contributed to the equivocal findings. Firstly, an independent measure of interoception could not be used due to COVID restrictions. The self-reported interoception questionnaire (BPQ-VSF: Porges, 1993, 2015) which was used in the study, records an individual's perception of their ability to recognise their inner bodily sensations (interoceptive sensibility) rather than a more objective heartbeat perception test which focuses upon the ability to detect the body's physiological internal milieu (interoceptive accuracy). Secondly, the sample of NES participants in study 2 were recruited via an online charity, so their diagnoses of NES could not be confirmed, neither could the presence/absence of epilepsy or panic symptoms be excluded. Finally, the control group were in the main recruited via a university/student population, including members of the public from the psychology research volunteer bank, so they could not be said to be entirely representative of the general population.

Another reason for the lack of findings to support the model from study 2 is that the study design necessarily treated NES participants as being part of a homogenous/continuous group in which the proposed model would apply to the majority of participants. As discussed previously in this thesis at least two prior studies, in which a cluster analysis was employed, reported two distinct subgroups of pwNES (Brown et al, 2013; Uliaszek et al, 2012). One group is characterised by high levels of psychopathology and symptomatology and the other by relatively low levels of psychological symptoms (Brown et al, 2013). Were the validity of such groups to be established by further confirmatory studies, it may explain the diverging pattern of results thus far reported from studies exploring interoception and its associations with other key variables in NES, as there may be different developmental pathways for NES symptoms in people with differing psychosocial profiles, and different factors which maintain symptoms.

The sample size of Study 2 was too small to conduct a cluster analysis in the two study groups but future work in this area may consider conducting a cluster analysis in NES participants prior to any further model testing and treat the identified clusters as separate groups for the purposes of analysis. The lack of supporting evidence for the proposed interactional model in Study 2 and the possibility of different clusters of NES participants with differing psychosocial profiles, suggests more in-depth understanding of people's experiences of NES and their perception of how their emotional responses to life stressors may interact with their seizures, is required.

Study 3: *A phenomenological exploration of emotional responses to life events in the context of non-epileptic seizures (NES).*

The third study sought to understand the lived experiences of pwNES. In-depth interviews were conducted with pwNES and analysed using interpretative phenomenological analysis (IPA). The focus was on pwNES' emotional responses to life events in the last 12 months of their lives, and whether they perceived the events to have had an impact on their seizure episodes. The impact of the emotional responses on seizure episodes was apparent in some instances (cat euthanised, poor treatment in hospital), but in other instances there appeared to be a long-term impact from the event(s), that possibly acted as a maintenance factor for seizures. Some participants indicated that their seizures appeared at times of rest, that is, after a period of hyperarousal.

Six group experiential themes (GET) were identified. The themes were ordered to present a narrative demonstrating that a timeline was often relevant to participants' accounts. Nizza and Smith (2021) state that the narrative presented should give a 'sense of coherence to the analysis and is an expression of the hermeneutic circle (Smith, 2007)'. The themes have been ordered to demonstrate that, in describing and reflecting on events, participants indicated a journey from first experiencing symptoms, receiving a diagnosis and the impact the condition had on their life and sometimes on those around them.

STRUGGLING TO LIVE WITH NON-EPILEPTIC SEIZURES: reflected the journey that participants described from receiving a diagnosis to their present lives. Feelings of relief and hope were expressed, by some, at the point of diagnosis, but these feelings were replaced by confusion when a clear treatment pathway was not presented. Part of the journey for participants had been to try to make sense of the episodes by looking for triggers. Some participants described seizures to have a resetting function for when they felt overwhelmed by emotion. The seizure episodes were described as making participants feel anxious and not in control.

BEING LABELLED WITH NON-EPILEPTIC SEIZURES FEELS DEGRADING: negative experiences with healthcare services, friends and family were voiced by some participants, and some participants felt the diagnosis was stigmatising. Two participants highlighted positive experiences with HCP. These were examples of how HCP had made efforts to make services more accessible to their needs.

THE LACK OF SUPPORT MAKES ME FEEL ALONE IN MANAGING THIS CONDITION: some participants described that they felt unsupported, and had to manage the condition themselves, without guidance from services. In instances where participants had received support, this was often short in duration because services did not feel equipped to support pwNES. Poor experiences with HCP made some participants reluctant towards using healthcare in the future.

LIVING WITH NES IS CHALLENGING: living with NES affected multiple aspects of people's identity. Participants described themselves as failures, being ashamed and feeling guilty in their roles as spouses, parents, and co-workers. Feelings of grief over the loss of independence, and a relationship was expressed by some.

HOW DO I LIVE BETTER, WITH THIS? Participants described ways in which they had developed strategies to cope with living with NES. These were task-orientated approaches such as raising awareness of the condition, breathing exercises, and prayer.

LIVING WITH NON-EPILEPTIC SEIZURES DURING COVID: the impact of the pandemic was not unique to participants in the sample; that is the challenges faced by participants were similar to issues that may impact those with chronic illnesses, and the general population during this time. All participants were interviewed after the pandemic had started; however it was not expressed as a challenge for all participants in the study.

The perceived impact of emotional responses to events on seizures

A novel finding from the third study in this thesis, was that four models were apparent in the way in which participants spoke about their emotional responses around events and the impact on their seizures. These connections were at times made by participants themselves, e.g. feeling overwhelmed with stress leads to a seizure; and at other times, the connection was made by the researcher, such as a participant describing that the car journey to the hospital always triggered a seizure. Both female and male participants were represented across the different seizure conceptualisations. Interestingly, male participants in the sample focussed on pain and did not consider stressful factors to be related to their seizures.

The four models that emerged from participants accounts were:

1. The participant experienced an event, to which they had an emotional reaction, and this resulted in a seizure. For example, Chloe had a meeting related to being made redundant from her job. Chloe experienced seizures before and after the meeting.
2. The participant experienced an event, to which they had an emotional reaction, but no seizure occurred around this time. For example, Parveen was informed of

the death of a family friend who was like a father figure to her. She was very sad upon hearing the news but did not experience any seizures around this event.

3. There was no event that occurred, but an emotional reaction was experienced, and a seizure occurred. For example, Ashley described a day in which she had engaged in enjoyable activities, and was feeling relaxed, and then towards the end of the day she began to experience a seizure.
4. There was no event that occurred, and no emotional reaction was experienced, but a seizure occurred. For example, Mark explained that soon after he had fallen asleep, often he would wake up and a seizure would occur.

Findings in the context of the available literature

Interoception

In the literature there is considerable interest in the role of interoception in relation to emotional detection and processing. However, the findings from the systematic review (study 1) were inconclusive and inconsistent. Several previous studies report participants with NES demonstrate poorer interoception on heartbeat perception tasks when compared to controls (Pick et al, 2020; Koreki et al, 2020; Demartini et al, 2019; Williams et al, 2021). The Body Perception Questionnaire employed in Study 2, consisted of a range of items about different internal bodily activity including heartbeat perception. Study 2 found that NES participants had a heightened or exaggerated sense of their awareness of their internal body milieu in comparison with healthy controls. Koreki et al (2020) used the Body Perception Questionnaire (Porges, 1993; 2015), and found that participants with FND reported significantly higher scores on the measure than controls. This response pattern has also been noted in other populations that are expected to demonstrate poor interoceptive accuracy. For example, Garfinkel et al (2016) found that participants with autism spectrum disorder perceived themselves to have better awareness of their inner bodily experience (as measured by the awareness subscale of the Body Perception Questionnaire (Porges, 1993, 2015)), than their actual performance on an objective heartbeat perception test would suggest. It has also been reported that the difference between interoceptive facets (i.e. enhanced interoceptive sensibility) and diminished interoceptive accuracy (objective performance) is greatest in those individuals with the poorest interoceptive accuracy (Garfinkel and Critchely, 2013). Theoretically, this may mean that people with NES have a problem in being able to tune in to their internal bodily sensations, so are less aware of internal emotional signals from the body (somatic markers); the subtle signals which according to Damasio (1994) help guide decision making. Emotional processing involves complex interactions between ‘top down’ and ‘bottom up’ processes in the brain which may be disrupted in certain pwNES.

In study 3, during in-depth interviews, some participants were able to describe how they felt in their body in relation to physical changes and emotions around seizure episodes. It is not possible however from the qualitative nature of the study to determine the accuracy of these accounts in relation to actual physiological changes.

Alexithymia

In Study 2, more participants with NES reported above threshold scores on the TAS-20 than controls. This finding is supported by other studies reporting higher levels of alexithymia in NES compared with non-patient controls, when using the TAS-20 (Urbanek, et al, 2014; Martino et al, 2018; Brown et al, 2013) and is consistent with the findings of a recent systematic review (Sequeira and Silva 2019). However, NES patients and those with epilepsy report similar scores on the TAS-20 (Sequeira and Silva, 2019) which suggests that problems with identifying and describing feelings may be common in pwNES but the presence of such problems are not sufficient to explain the phenomenon.

Study 3 was a qualitative study, so there were no formal measures of alexithymia, but some participants clearly described struggling to articulate their feelings in a manner consistent with alexithymia. However, there are several reasons why people may struggle to describe their feelings to a researcher whom they have met for the first time, which may or may not be related to alexithymia. These may include reticence when meeting someone new, cognitive problems such as brain fog - which is a common complaint of people with NES, or poor recall since events were discussed retrospectively. Some participants in study 3, when asked about their feelings in response to life events, were able to articulate their feelings in a clear and meaningful way. For example, one participant spoke about feeling devastated following an interaction with a healthcare worker that he perceived to be punitive. The interaction was during a lengthy period of time the participant spent in hospital and he expressed feeling dejected and distressed after the event.

Qualitative interviews with participants with and without alexithymia have reported that participants with possible alexithymia may demonstrate ‘emotional understanding’, which refers to a comprehension of why they perceive themselves to feel a certain way in relation to experiences (da Silva, 2018). In study 3 this was possibly demonstrated by a participant that spoke about angry outbursts and frustration. That is, he was beginning to understand or give meaning to this emotional experience. He was recognising that his emotional response of anger was connected to frustration, however the further link of why he felt frustrated was not achieved yet. This is perhaps where psychological therapy may help the participant to work on understanding and exploring this.

It has been suggested that therapists may struggle to work effectively with alexithymic patients because they struggle to communicate their feelings and, this may impair the therapeutic process (Ogrodniczuk et al, 2005). The therapist may hold negative views about an alexithymic patient because the patient may speak about vague difficulties and struggle with developing goals (Ogrodniczuk et al, 2005). Another participant in study 3 demonstrated difficulties with being clear about possible connections between emotions and events and spoke in vague or unclear terms in general when reporting the impact of emotion on seizure episodes. Interestingly this participant had received some psychological therapy but had expressed that the psychologist was unable to help him. This is perhaps an example of a difficulty with ‘description of problems’ that da Silva et al (2018) reported as a theme in their study. It has also been reported that a focus on external change, rather than an interpersonal focus emerged as factors in relation to the theme of ‘affective change focus’ (da Silva et al, 2018). In study 3, two male participants focussed on external change, such as work and how this had an impact on their lifestyle or their family. Whereas two female participants demonstrated how external changes (giving up driving/ending therapy) had an impact on themselves (intrapersonal focus) such as independence, levels of confidence.

Participants in study 3 did not speak about ‘bottling up’ or supressing emotions as has been reported in other studies in pwNES (Rawlings and Reuber, 2016). Overall, some participants in study 3 were able to describe their emotional responses to life stressors and others were not. So even in such a small sample there were marked differences in emotional response patterns between different participants, perhaps illustrating the heterogeneity of pwNES.

Executive functioning

The proposed model that was tested in Study 2, included attentional bias as one of the hypothesised contributing components to a cyclical pattern of emotional processing dysfunction in NES. There was unconvincing evidence from the results of study 2 to support the model and, in particular, problems with attentional bias in people with NES. There were clear differences between the NES participants and healthy controls on most of the measures included in study 2 (TAS-20, DERS, BFQ-VSF, GAD-7, PHQ-8), and response times on the eStroop task showed NES participants showed slower response times to both positive and negative stimuli. However, the groups were not significantly different on either positive or negative emotional interference scores.

Other studies have reported that participants with NES show more difficulties with attentional bias (Bakvis et al, 2009; Almis et al, 2013) and emotion processing (Gul and Ahmad, 2014). Likely differences between the results of the current study and those in the NES literature (Almis et al, 2013; Bakvis et al, 2009; Gul and Ahmad, 2014) include differences in study populations, paradigms used, and type of controls.

Life events

Study 3 was able to deliver a rich and individualised account of the ways in with people with NES experience life stresses and their understanding of the relationship between stress and their seizures. The NES literature highlights the heterogeneous presentation of NES, and the Integrative Cognitive Model (Brown et al, 2016) reflects this by encompassing several different factors and processes that can contribute towards the development and precipitation of seizure events. This too was demonstrated by participants who described different types of seizure experience within the sample. In Study 3, when talking about seizure events, participants often described common features of the seizures they experienced: dissociation, paralysis, and anxiety before, and after a seizure event.

Participants described a range of events that were either specific life events or that would be considered as ongoing difficulties. Emotions and emotional responses featured in these events at times as a direct or indirect consequence of the event. For example, a participant described the sadness and upset at the time their much-loved pet was euthanised. But in other instances participants indicated feeling guilt and shame. These latter feelings perhaps were the result of individuals reflecting and ruminating on how events in their lives had affected them and those around them in intimate relationships, with co-workers and interactions with HCPs. Rumination has been reported to be associated with difficult emotions or experiences (Pick et al, 2016). An example of rumination from study 3 is of a participant that expressed sleeping difficulties, and that this was a period of time when he would *churn over* his thoughts. This participant also experienced a traumatic experience in hospital that left him feeling *wrecked,* and thinking about the incident still made him feel upset. Another participant spoke of an abusive experience in hospital and became upset when talking about the event, expressing that she did not want to go back to the hospital unless in dire need. This perhaps indicates that participants were ruminating but were not expressing this as intrusive thoughts as reported in another study (Pick et al, 2016).

Other qualitative studies with pwNES, using different methodologies or with a different focus have found similar results those in study 3 including themes such as rumination, shame and stigma, stress and trauma (Carton et al, 2003; Green et al, 2004; Fairclough et al, 2013; Karterud et al, 2010; Pick et al, 2016; Rawlings and Reuber, 2016; Rawlings et al, 2017) and negative interactions with healthcare (Green et al, 2004; Mayor et al, 2011; Wyatt et al, 2014). This perhaps indicates that these are areas of importance and common experience for pwNES.

Impact on seizures from emotional responses to events

A novel finding of the study was that participants described four models that described the relationship between emotional responses to life events and their impact on seizures. The models were either referred to by participants as they explained the impact on them e.g. stress led to a seizure(s), or the connection was made by the researcher. In the latter case this was mostly in describing the models that referred to those seizures that occurred when there was no emotion or event that was perceived to be linked to the seizure episode (Models 2 and 4) but was also relevant to some examples of model 1. Some participants also indicated that seizures were a way of resetting; that the seizure was the body taking control when they were overwhelmed with emotion. A similar finding was reported by Pick et al (2106) that pwNES spoke of having a seizure episode after a ‘build up’ of emotion and this brought a feeling ‘relief’, and this has been reported in other qualitative studies with pwNES (Carton et al, 2003; Wyatt et al, 2014).

Participants looked for triggers for their seizures, factors that they could anchor to the episodes and identify as being to blame for the outcome (seizure). Some participants expressed how past trauma had a continued impact on their life and likely led to the development of NES. Male participants spoke of pain being a precipitating factor, whereas female participants more likely viewed stress as being a trigger for their seizures. Individuals also reported that when a seizure occurred at times of rest it had been preceded by a stressful or hectic period. This may be the experience of ‘panic without panic’ that has been proposed by models that consider NES to be a post traumatic type response (Brown and Reuber, 2016). That is the feelings of panic/anxiety are experienced some time after the arousal/stress has passed, a delayed reaction of sorts or the anxiety is not experienced at all. An alternative perspective is that the 'panic without panic' explanation may misrepresent the participant's experience. That is, the participant describes having a seizure during a period of rest and relaxation, and not in a delayed state of hyperarousal. This has been reported by other studies that have reported that individuals may experience seizures during times of rest (van der Kruijs et al, 2016).

Experiences of healthcare

Experiences with healthcare included both negative and positive experiences. Participants shared more negative experiences, and, in some instances, these were described as traumatic. The impact, of the latter type of event, on a participant was that they described a reluctance to use healthcare services in the future. Other participants indicated that their expectations of healthcare services had been disappointing or had not transpired. Such experiences have been shared by pwNES in other studies also (O'Sullivan et al, 2007; Rawlings et al, 2017). A recent survey of 2187 people in the UK regarding their satisfaction with mental and physical health services (Kirkham et al, 2020), reported that the study findings were similar to another pre-COVID study that reported that 60% of the general public were satisfied with the NHS (Appleby et al, 2020). Kirkham et al (2020) also noted that 'the presence of depression, anxiety' and other conditions were correlated with lower satisfaction for the first contact with a NHS mental health service. This may offer some explanation as to why some participants in study 3 focussed more on the negative experiences that they had with healthcare services, rather than reflecting on any positive experiences that they encountered in the NHS. In study 3, two participants shared positive experiences where a HCP had made an effort to make the service more accessible for the individual (Abigail), or the information discussed at diagnosis had been sent in a written format after the appointment (Bryony). The example of providing written information is a strategy that has been recommend by Reuber (2019), since concentration and retention of information may be comprised if patients are feeling anxious at the time of the appointment. Reuber (2019) outlines ways in which the process can be made more collaborative. This may have a more positive impact, not only on how patients feel about the diagnosis, but also about how pwNES perceive HCP will respond to them if they require medical attention.

Shame and guilt

Feelings of shame and guilt have already been briefly discussed. In study 3, participants gave examples of referring to themselves as *useless,* having to prove themselves, or feeling they were a poor parent. These expressions of low self-esteem have also been reported in pwNES by Rawling et al (2017). They noted that pwNES demonstrated 'a lack of self-compassion' and indicated using poor emotional coping via suppression or avoidance of emotions. Feelings of shame and guilt are not new findings in pwNES (Reuber et al, 2022) and such self-directed negative feelings are also common in people with epilepsy (Keikelame and Swartz, 2018), suggesting that this may be related to living with seizures, rather than NES specifically. Although pwNES and their carers have been found to perceive higher rates of stigma than patients with epilepsy (Karakis et al, 2020), which may contribute to maintaining such feelings. The findings from study 3 highlight that shame and guilt may be areas that psychological therapy may be able to help individuals to address.

In study 3, there was also the issue of challenges to masculinity that appeared to be relevant to two male participants. These participants spoke about failing (guilt) in their roles as fathers and husbands. The impact of being unwell may challenge beliefs around the male identity as a being 'strong' and a 'provider'. This struggle has also been noted in an IPA study with men with NES undergoing psychotherapy (Quinn et al, 2012). Factors such as ideas around masculinity may be important to explore in therapy, in order for therapists and patients to understand the impact it may have on their self-esteem, expressions of emotion and identity.

Loss and grief

The emotions of grief and loss were apparent for some participants in study 3. For example, a participant experienced grief and loss after relinquishing their driving license. This individual indicated that it was not only a loss of independence, but also had longer term consequences for her and her husband. This event was a direct consequence of developing NES, and had financial implications, a possible social impact and also an effect on the individual's sense of self. The example of giving up ones driving license is found as a common experience associated with loss in pwNES (Baxter et al, 2012; Green et al, 2004; Pretorius et al, 2009; Pretorius and Sparrow, 2015). The participant, in study 3, that reported surrendering her driving license spoke of grieving over the loss of independence.

This possibly highlights how an event may result in an ongoing form of stress and arousal, and this may act as a maintenance factor for seizure episodes.

Coping with living with NES

In a more general way, participants expressed feeling frustrated and alone in trying to manage their life with NES. Frustration and disappointment are legitimate feelings when individuals do not receive adequate information about how to live with a new diagnosis, or when they do not receive support from healthcare services or employers. It has been reported that HCP have also described feeling confused and frustrated due to the lack of a clear treatment pathway for NES/FND (Mayor et al, 2011; White 2016). Therefore, whilst some people with NES may have difficulties with emotional processing or expression, which may impair goal setting in therapy for example, the feelings of frustration may be confounded by a lack of appropriate support. Participants in Study 3 described developing their own strategies to cope, some of which may be considered dysfunctional or unhelpful in the longer-term but demonstrated a motivation to make their situation better. Additionally, not all coping was dysfunctional. Five participants mentioned task-orientated strategies as ways of living better with NES. In particular two participants demonstrated that using breathing techniques or a strategy in keeping with their religious beliefs had reduced seizure episodes for them. The use of coping strategies is not novel (Pick et al, 2016) and although the examples differed to those reported by Pick et al (2016), the purpose was the same – that is attentional and behavioural distraction from difficult situations, including impending seizures. These findings highlight how pwNES may develop their own ways of managing their symptoms, in the absences of support from services, in order to improve their quality of life.

Ongoing stressful factors

In study 3, some participants spoke about the difficulties that they faced at work (e.g. discrimination), or the guilt that they felt because they were not living up to the standards that they had perhaps set for themselves (e.g. giving up extracurricular activities). In this way they indicated that these negative feelings had a longer-term impact on them. Pick et al (2016) also reported that participants spoke of ‘generally stressful’ lives in which they referred to several stressors or traumatic experiences. This was expressed by participants in study 3 that spoke of how they had to manage relationships that were particularly stressful, or how the impact of the pandemic was a *low-grade burden*, meaning it was not specific events that were stressful but how multiple events resulted in a continued level of stress experienced. This perhaps demonstrates that the consequences of life events may be ongoing and therefore potentially perpetuating factors for NES.

Events in the workplace, or work performance being questioned, based on the disability of the individual also featured in Study 3. The life events literature has indicated that pwNES have demonstrated higher levels of distress in relation to a variety of factors such as work, social functioning, health, and legal difficulties (Testa et al, 2012). This suggests that it may not be the number of life events experienced, but rather the impact that recent life events have on 'the severity' of functional neurological symptoms - of which NES might be considered one such symptom (Roelofs et al, 2005). Therefore perpetuating factors may relate to the level of stress experienced through ongoing difficulties in the workplace.

Strengths and Limitations

The strengths and limitations of each of the three individual studies have been discussed in the relevant chapters. The findings from the systematic review (study 1) were meant to feed into the design of study 2 and to some extent study 3. However, the impact of the COVID 19 pandemic meant that study 2 had to be redesigned without some of its planned methodological strengths.

All the studies included in the systematic review were Eurocentric and demographic data did not include the reporting of ethnicity. This raises the question of how representative such study samples are and whether studies are recruiting sufficiently diverse samples that reflect the populations within which the studies are conducted. The lack of reporting of ethnicity may imply that researchers themselves fail to reflect on what a representative sample is. Additionally, the way in which ethnicity is reported is not always appropriate, such as reporting the study sample in a dichotomous manner: white and non-white (Jalilianhasanpour et al, 2018).

In study 3, using IPA the focus of the analysis was about capturing the meaning that the phenomenon has for the participant, by understanding the individual's interpretation of their experiences. A different methodological approach, such as thematic analysis would have allowed potential coding themes to be identified from the existing literature, and the focus of analysis different because it would have been more about identifying incidents that indicated difficulties with processing emotion. Both approaches have strengths and limitations.

Recommendations

Participants were clear that better information at diagnosis is needed. There was confusion after the initial diagnosis when the hardware/software analogy was often used to explain the condition to participants. This explanation needs to include ‘emotional software’ so that individuals do not perceive the diagnosis as being told that it is ‘all in their mind’. Reuber (2019) has addressed how healthcare practitioners may be able to communicate the diagnosis more effectively, more compassionately and collaboratively with patients.

Better information about NES/FND for healthcare practitioners is required. This awareness is required across primary and secondary care. This is based on participants describing experiences in which they felt they were judged by healthcare workers to be wasting time and resources.

A multidisciplinary approach is required for those with multiple functional neurological symptoms. That is, individuals may need psychological support to adjust to the diagnosis and learn how to recognise/manage emotions better, but some individuals may also need physiotherapy or speech therapy if their condition includes gait/mobility or speech problems.

The voices of underrepresented individuals are important. This means that using interpreters where required is key in research to allow the inclusion of those for whom English is not their first language, or one in which they cannot express and communicate confidently. The current literature is centred on White Western samples. If research is to be representative, then it should aim to be more inclusive. Often individuals from ethnic minority backgrounds are only represented when the study is concerned with a specific community or population, rather than being seen as part of the general population in Western countries.

The novel finding of different models regarding how pwNES understand their seizure experience, indicates that psychological therapy may be helpful for those that perceive clear links between events, emotions and their seizures. Different types of interventions may be required for people who do not recognise a connection between life stress, emotion and their seizures.

Further research should focus upon identifying different psychosocial profiles of pwNES and model testing within these different groups. This may lead to more consistency in research results.

There needs to be some agreement with regards to whether heartbeat perception tasks are considered a good measure of general interoception. It is possible that use of multiple tasks to investigate different types of perception of internal bodily experiences, such as breathing/gastric tasks, could provide a more comprehensive approach to objective measurement of interoception (Garfinkel et al, 2015; Murphy et al, 2018).

Additionally, NES studies need to be clearer about the comparison groups chosen.

Conclusions

Emotion is pervasive in all aspects of one's life, how we relate to others, make decisions, and has been the matter of research and interest for centuries. Theories relating to the development and maintenance of NES highlight that understanding how individuals process emotions may be key, and this has led to an interest in interoception as an area of investigation.

The findings from the systematic review suggest, however, that the current literature is contradictory and inconsistent, most likely due to widespread methodological problems and poor study quality. The work from study 2 was mainly confirmatory in that the findings suggest that pwNES report more psychosocial difficulties and symptoms than healthy controls. People with NES also perceive themselves to be better than controls at being able to detect their inner bodily symptoms, which paradoxically may suggest they actually struggle more with interoceptive abilities as they overestimate their abilities in this area. There was little support for an interactional model between interoceptive sensibility, alexithymia and emotional processing, but the limitations imposed on the design of Study 2 due to COVID-19 restrictions may have impacted on the results.

One of the most striking and novel findings from study 3 was the diversity with which pwNES perceived the relationship between life stress, emotional responses and their seizures. Within the small group of pwNES studied 4 different models were identified. These findings support other research that suggests different subgroups of pwNES may exist with differing psychosocial profiles and ways in which seizures develop and are maintained. This heterogeneity of the NES group may be a major explanation for the lack of consistency of research findings in this area, in addition to methodological issues discussed above.

Study 3 demonstrated that regardless of the focus of the research, for some individuals it was opportunity for them to tell their ‘story’. A form of catharsis perhaps, when individuals with NES feel that they have not been listened to by HCP.

Future studies need to be more inclusive. The current literature is largely unrepresentative of people from ethnic minority backgrounds.

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*papers returned by the systematic search

** papers included after the systematic search was conducted

APPENDIX A

Ethical approval confirmation email study 2

From: Microsoft Power Apps and Power Automate <microsoft@powerapps.com>
Sent: 20 October 2021 16:11
To: Jac Billington <J.Billington@leeds.ac.uk>
Subject: Your ethics application result - Approved

Dear Alscient Developer

Re your ethics application, Investigating self-perception of emotion in individuals with functional seizures: using self-report measures and an emotional Stroop task., ethics reference number: PSYC-332.

I am pleased to inform you that the above research application has been reviewed by the School of Psychology Research Ethics Committee and the desision is

Approved.

If the reviewers have left any comments they will appear below.
Primary reviewer comments (if applicable) :
Secondary reviewer comments (if applicable) :

Please note that this approval only relates to the particular version of documentation supplied in this specific application (ethics ref no: PSYC-332).

If you wish to make any amendments to the approved documentation, please note that all changes require ethical approval prior to implementation.

Please note: You are expected to keep a record of all your approved documentation, as well as documents such as sample consent forms, and other documents relating to the study. This should be kept in your study file, which should be readily available for audit purposes.

You will be given a two week notice period if your project is to be audited. There is a checklist listing examples of documents to be kept which is available at http://ris.leeds.ac.uk/EthicsAudits.

Yours sincerely,
School of Psychology Research Ethics Committee

If you want to unsubscribe from these emails, please use this form.

APPENDIX B

PARTICIPANT INFORMATION SHEET (NES)

Investigating self-perception of emotion in individuals with functional seizures (FS): using self-report measures and an emotional Stroop task.

Invitation to take part in research:

You are being invited to take part in an online research study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. If anything is not clear, or you would like more information, please contact the lead researcher (Shaista Meer).

What is the purpose of the research?

The main aim is to gain a better understanding of how people with functional seizures detect and process emotion. We hope that this will inform future therapeutic approaches, so they are more effective for people with functional seizures.

You might know functional seizures by another name such as dissociative seizures, psychogenic non-epileptic seizures, non-epileptic seizures or non-epileptic attack disorder.

Why have I been chosen?

You have been chosen because we need people who have functional seizures to take part.

To qualify for the study you should also:

- Be at least 18 years old.
- Be able to read and understand English.
- Be able to decide whether you would like to take part.
- Be in the UK.

You cannot take part if you:

- Are under 18 years old.
- Have any major health conditions such as stroke, a cardiac condition, epilepsy, dementia, or other neurological conditions.
- Cannot read English.
- Are not in the UK**.**
-

Do I have to take part?

No. It is entirely up to you whether you choose to take part, and you are free to withdraw from the study at any point.

What is involved if I decide to take part?

If you decide to take part, then you need to choose a time that is suitable to you. You need access to the internet and a computer (laptop or PC) to participate in the study. Taking part in the study is likely to take around an hour of your time.

First you will be asked to sign a consent form. Giving your consent means that you agree to take part in the research based on the information that has been provided to you about the study.

You will then complete six questionnaires. The first questionnaire contains basic questions such as your age, occupation, height and weight. The rest of the questionnaires will ask questions about your emotions, mood and how you detect changes in your body. Everyone who takes part is asked to answer the same questions. Being given these questionnaires does not mean that you have low mood or anxiety.

After this, you will be asked to take part in an experiment called the Stroop task. This task involves responding to words that will be displayed on the screen. This task requires matching the colour of the text on the screen to one of the options. You will be given a chance to practice and there will be prompts on the screen to tell you what to do.

You will receive a £10 ‘thank you’ voucher after completing the tasks. To receive this, you need to add your email address when prompted at the end. Shaista Meer will send you the voucher by email. Your email address will not be kept with any of your test data, it will be in a separate spreadsheet away from any of your responses.

What are the possible disadvantages and risks of taking part?

There are no major disadvantages or risks expected to be experienced by anyone taking part. It is possible that answering questions about your emotional wellbeing might inform you or remind you that your mood is low or anxious. If you find yourself being upset or worried by any of the questions, please talk to your GP about this. There are also some numbers at the end of this leaflet that you might find helpful.

There is a small risk that anyone taking part could have a seizure during this time. If you think this may be a possibility, then please let someone in your household know so that they can check in with you whilst you complete the questionnaires/task.

The Stroop task comprises of words flashing up on the screen. If you think that looking at words repeatedly flashing may cause a seizure – you may decide that you do not want to take part.

What are the benefits of taking part?

Although this research may not directly benefit you, we hope that this research will help us to have a better understanding of how people with functional seizures experience their emotions. This will help us to inform future therapeutic interventions.

Will my taking part be kept confidential and what will happen to the data I provide?

All information that is collected during the study will be treated confidentially. No one is asked to add their name on any of the questionnaires. This anonymises your responses. The data collected (scores on the Stroop task and answers to the questionnaires) will be kept for three years in this anonymised format. If the researcher leaves the University before the end of the three years, the main supervisor **(Prof Guthrie**) will look after the anonymised data. The anonymised data will be stored on a secure network at the University of Leeds.

The only time you will supply your any contact details/identifiable information, will be if you provide your email address to receive the electronic voucher. A list of names will also be provided to the University of Leeds for the purposes of showing that participants have received a voucher. This information will be stored on a secure network at the University of Leeds and will not be associated with your responses to the questionnaires.

Who will know that I am taking part in the study?

No one will know that you have taken part in the study unless you choose to tell them. Your name will not be published anywhere.

Who is responsible for looking after my information?

The University of Leeds is the sponsor for this study based in the United Kingdom. This means that the University of Leeds is responsible for looking after your information. We will be using information from you to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly.

To safeguard your rights, we will use the minimum personally identifiable information possible. This means that after the analysis has been completed, we may remove some details from the data, such as your occupation and ethnicity to further anonymise the information. The anonymised data will be stored for three years after the data has been published.

Who has access to my identifiable data?

The lead researcher will keep your name confidential and will only pass this information to The University of Leeds to show that a voucher has been sent out. Your contact details (email address) will be destroyed once the study has ended.

Certain individuals from The University of Leeds and regulatory organisations may look at your research records to check the accuracy of the research study. The University of Leeds will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

Can I access the information that you have about me?

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. You can find out more about how we use your information http://www.leeds.ac.uk/secretariat/data_protection.html.

What will happen to the results of the study?

Due to the data being analysed as a group, it will not be possible to provide any specific information about your own data. It will not be possible to identify participants from any of the data that will be published and the anonymised data may be used to support further ethically approved research.

This study is part of a PhD and the results of the research will be presented at academic conferences and published in academic journals**.** If you are interested in the findings then you can contact the lead researcher for more information.

Who is organising and funding the research?

This research is funded by the University of Leeds.

Further information: If you have any questions or would like to discuss taking part, then please contact the lead researcher:

Miss Shaista Meer um08sm@leeds.ac.uk; or Prof Else Guthrie (main supervisor) e.a.guthrie@leeds.ac.uk.

If you have been affected by the research then please discuss this further with the researcher.

If you feel distressed then you can access support through:

- IAPT (Improving Access to Psychological Therapies) https://www.leedscommunityhealthcare.nhs.uk/iapt/home/ online assessment or call 0113 843 4388
- The Samaritans on Freephone 116 123

If you want to make a complaint related to this research then you can speak to Clare Skinner, Faculty Head of Research and Innovation Support, Email: governance-ethics@leeds.ac.uk, tel: 0113 343 4897 Or the independent organisation: Information Commissioner's Office on 0303 123 1113.

Ethics approval:

This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee, University of Leeds (SoPREC ref: PSYC-332 date: 20/10/2021).

PARTICIPANT INFORMATION SHEET (controls)

Investigating self-perception of emotion in individuals with functional seizures (FS): using self-report measures and an emotional Stroop task.

Invitation to take part in research:

You are being invited to take part in an online research study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. If anything is not clear, or you would like more information, please contact the lead researcher (Shaista Meer).

What is the purpose of the research?

The main aim is to gain a better understanding of how people with functional seizures detect and process emotion. We hope that this will lead to informing future therapeutic approaches to be more effective for people with functional seizures.

You might know functional seizures by another name such as dissociative seizures, psychogenic non-epileptic seizures, non-epileptic seizures or non-epileptic attack disorder.

Why have I been chosen?

You have been chosen because we need people who do not have functional seizures to take part too, so that we can compare against the responses we receive from people who do have functional seizures.

To qualify for the study you should also:

- Be at least 18 years old.
- Be able to read and understand English.
- Be able to decide whether or not you would like to take part.
- Be in the UK.

You cannot take part if you:

- Are under 18 years old.
- Have any major health conditions such as stroke, a cardiac condition, epilepsy, dementia, or other neurological conditions.
- Cannot read English.
- Are not in the UK.
-

Do I have to take part?

No. It is entirely up to you whether you choose to take part, and you are free to withdraw from the study at any point.

What is involved if I decide to take part?

If you decide to take part, then you need to choose a time that is suitable to you. You need access to the internet and a computer to participate in the study. Taking part in the study is likely to take around an hour of your time.

First you will be asked to sign a consent form. Giving your consent means that you agree to take part in the research based on the information that has been provided to you about the study.

You will then complete six questionnaires. The first questionnaire contains basic questions such as your age, occupation, height and weight. The rest of the questionnaires will ask questions about your emotions, mood and how you detect changes in your body. Everyone who takes part is asked to answer the same questions. Being given these questionnaires does not mean that you have low mood or anxiety.

After this, you will be asked to take part in an experiment called the Stroop task. This task involves responding to words that will be displayed on the screen. This task requires matching the colour of the text on the screen to one of the options. You will be given a chance to practice and there will be prompts on the screen to tell you what to do.

You will receive a £10 ‘thank you’ voucher after completing the tasks. To receive this, you need to add your email address when prompted at the end. Shaista Meer will send you the voucher by email. Your email address will not be kept with any of your test data, it will be in a separate spreadsheet away from any of your responses.

What are the possible disadvantages and risks of taking part?

There are no major disadvantages or risks expected to be experienced by anyone taking part. It is possible that answering questions about your emotional wellbeing might inform you or remind you that your mood is low or anxious**.** If you find yourself being upset or worried by any of the questions, please talk to your GP about this. There are also some numbers at the end of this leaflet that you might find helpful.

What are the benefits of taking part?

Although this research may not directly benefit you, we hope that this research will help us to have a better understanding of how people with functional seizures experience their emotions. This will help us to inform future therapeutic interventions.

Will my taking part be kept confidential and what will happen to the data I provide?

All information that is collected during the study will be treated confidentially. No one is asked to add their name on any of the questionnaires. This anonymises your responses. The data collected (scores on the Stroop task and answers to the questionnaires) will be kept for three years in this anonymised format. If the researcher leaves the University before the end of the three years, the main supervisor **(Prof Guthrie**) will look after the anonymised data. The anonymised data will be stored on a secure network at the University of Leeds.

The only time you will supply your any contact details/identifiable information, will be if you provide your email address to receive the electronic voucher. A list of names will also be provided to the University of Leeds for the purposes of showing that participants have received a voucher. This information will be stored on a secure network at the University of Leeds and will not be associated with your responses to the questionnaires.

Who will know that I am taking part in the study?

No one will know that you have taken part in the study unless you choose to tell them. Your name will not be published anywhere.

Who is responsible for looking after my information?

The University of Leeds is the sponsor for this study based in the United Kingdom. This means that the University of Leeds is responsible for looking after your information. We will be using information from you to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly.

To safeguard your rights, we will use the minimum personally identifiable information possible. This means that after the analysis has been completed, we may remove some details from the data, such as your occupation and ethnicity to further anonymise the information. The anonymised data will be stored for three years after the data has been published.

Who has access to my identifiable data?

The lead researcher will keep your name confidential and will only pass this information to The University of Leeds to show that a voucher has been sent out. Your contact details (email address) will be destroyed once the study has ended.

Certain individuals from The University of Leeds and regulatory organisations may look at your research records to check the accuracy of the research study. The University of Leeds will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

Can I access the information that you have about me?

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. You can find out more about how we use your information http://www.leeds.ac.uk/secretariat/data_protection.html.

What will happen to the results of the study?

Due to the data being analysed as a group, it will not be possible to provide any specific information about your own data. It will not be possible to identify participants from any of the data that will be published, and the anonymised data may be used to support further ethically approved research.

This study is part of a PhD and the results of the research will be presented at academic conferences and published in academic journals**.** If you are interested in the findings, then you can contact the lead researcher for more information.

Who is organising and funding the research?

This research is funded by the University of Leeds.

Further information: If you have any questions or would like to discuss taking part, then please contact the lead researcher:

Miss Shaista Meer um08sm@leeds.ac.uk; or **Prof Else Guthrie (main supervisor) e.a.guthrie@leeds.ac.uk.**

If you have been affected by the research then please contact the researcher to discuss.

If you feel distressed then you can access support through:

- IAPT (Improving Access to Psychological Therapies) https://www.leedscommunityhealthcare.nhs.uk/iapt/home/ online assessment or call 0113 843 4388
- The Samaritans on Freephone 116 123

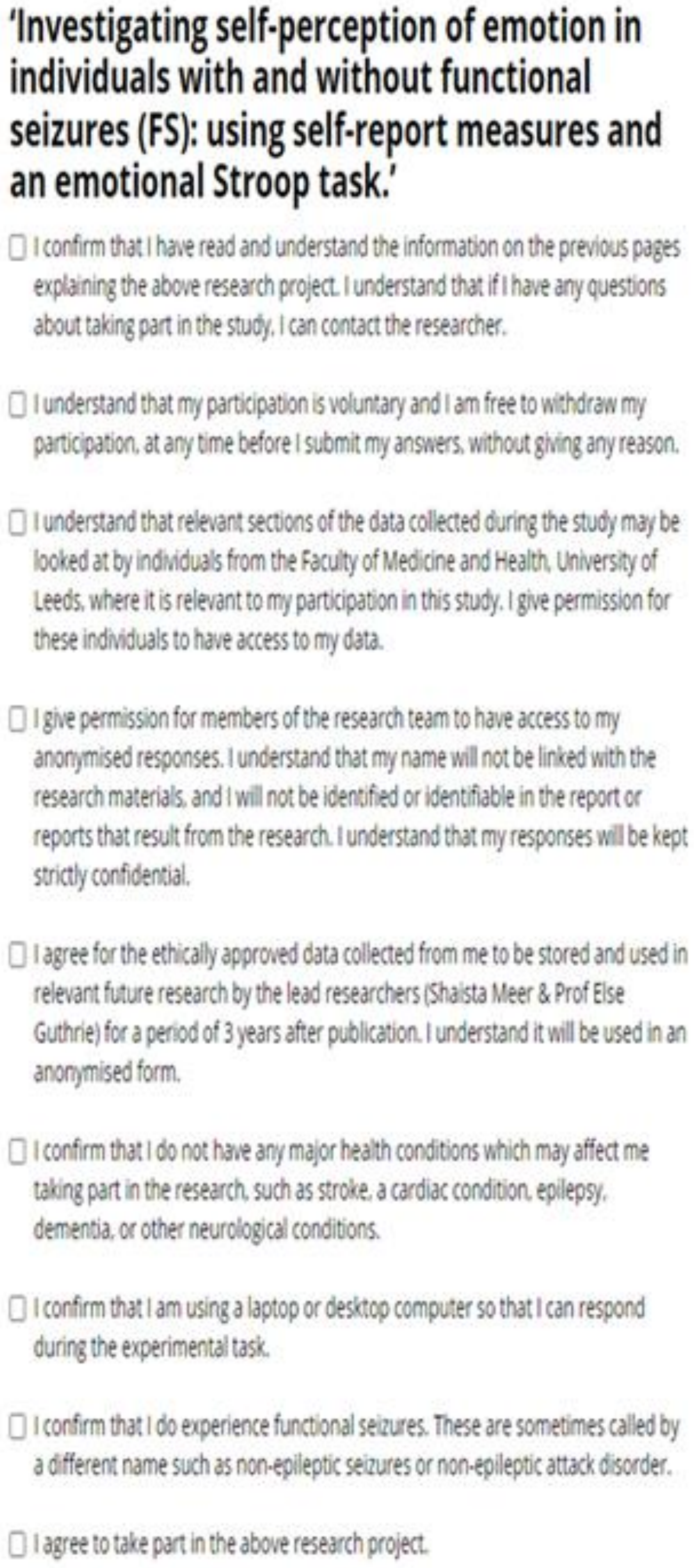
If you want to make a complaint related to this research then you can speak to Clare Skinner, Faculty Head of Research and Innovation Support, Email: governance-ethics@leeds.ac.uk, tel: 0113 343 4897 Or the independent organisation: Information Commissioner's Office on 0303 123 1113.

Ethics approval:

This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee, University of Leeds (SoPREC ref: PSYC-332 date: 20/10/2021).

APPENDIX C

Consent form for NES group



'Investigating self-perception of emotion in individuals with and without functional seizures (FS): using self-report measures and an emotional Stroop task.'

☐ I confirm that I have read and understand the information on the previous pages explaining the above research project. I understand that if I have any questions about taking part in the study, I can contact the researcher.

☐ I understand that my participation is voluntary and I am free to withdraw my participation, at any time before I submit my answers, without giving any reason.

☐ I understand that relevant sections of the data collected during the study may be looked at by individuals from the Faculty of Medicine and Health, University of Leeds, where it is relevant to my participation in this study. I give permission for these individuals to have access to my data.

☐ I give permission for members of the research team to have access to my anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in the report or reports that result from the research. I understand that my responses will be kept strictly confidential.

☐ I agree for the ethically approved data collected from me to be stored and used in relevant future research by the lead researchers (Shaista Meer & Prof Else Guthrie) for a period of 3 years after publication. I understand it will be used in an anonymised form.

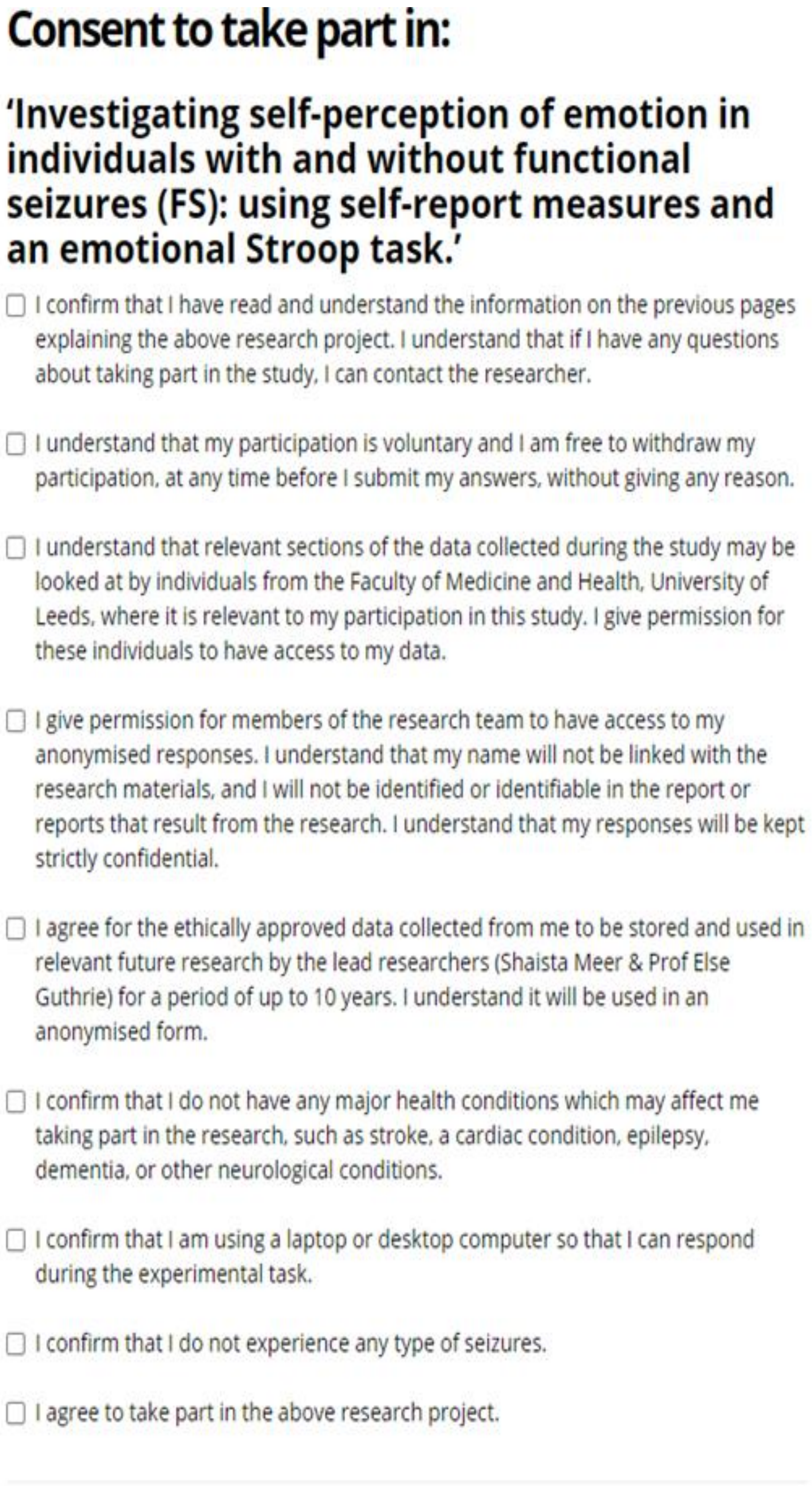
☐ I confirm that I do not have any major health conditions which may affect me taking part in the research, such as stroke, a cardiac condition, epilepsy, dementia, or other neurological conditions.

☐ I confirm that I am using a laptop or desktop computer so that I can respond during the experimental task.

☐ I confirm that I do experience functional seizures. These are sometimes called by a different name such as non-epileptic seizures or non-epileptic attack disorder.

☐ I agree to take part in the above research project.

Consent form for control group



Consent to take part in:

'Investigating self-perception of emotion in individuals with and without functional seizures (FS): using self-report measures and an emotional Stroop task.'

☐ I confirm that I have read and understand the information on the previous pages explaining the above research project. I understand that if I have any questions about taking part in the study, I can contact the researcher.

☐ I understand that my participation is voluntary and I am free to withdraw my participation, at any time before I submit my answers, without giving any reason.

☐ I understand that relevant sections of the data collected during the study may be looked at by individuals from the Faculty of Medicine and Health, University of Leeds, where it is relevant to my participation in this study. I give permission for these individuals to have access to my data.

☐ I give permission for members of the research team to have access to my anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in the report or reports that result from the research. I understand that my responses will be kept strictly confidential.

☐ I agree for the ethically approved data collected from me to be stored and used in relevant future research by the lead researchers (Shaista Meer & Prof Else Guthrie) for a period of up to 10 years. I understand it will be used in an anonymised form.

☐ I confirm that I do not have any major health conditions which may affect me taking part in the research, such as stroke, a cardiac condition, epilepsy, dementia, or other neurological conditions.

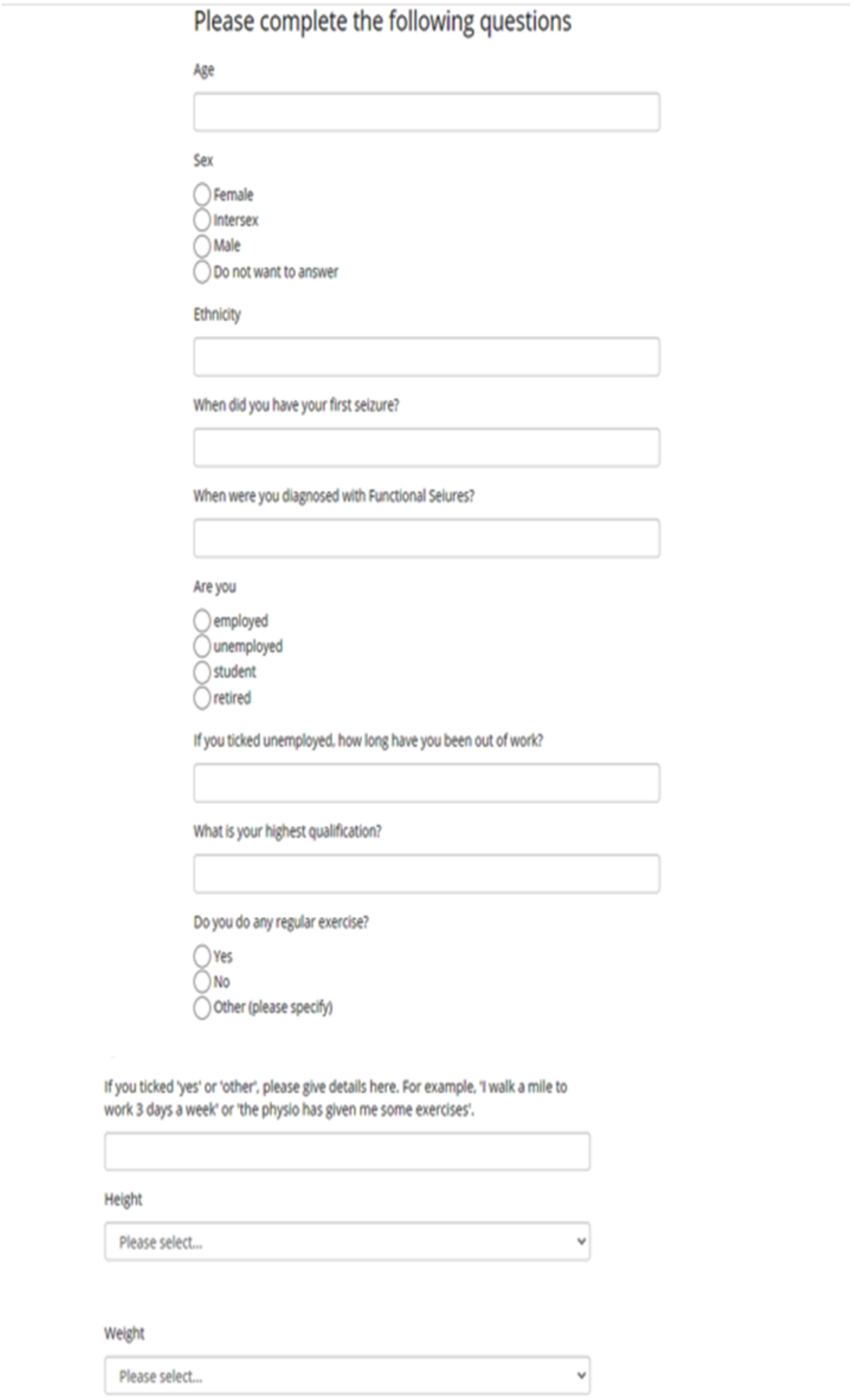
☐ I confirm that I am using a laptop or desktop computer so that I can respond during the experimental task.

☐ I confirm that I do not experience any type of seizures.

☐ I agree to take part in the above research project.

APPENDIX D

Demographic questionnaire (NES)



Please complete the following questions

Age

Sex

○ Female
○ Intersex
○ Male
○ Do not want to answer

Ethnicity

When did you have your first seizure?

When were you diagnosed with Functional Seiures?

Are you

○ employed
○ unemployed
○ student
○ retired

If you ticked unemployed, how long have you been out of work?

What is your highest qualification?

Do you do any regular exercise?

○ Yes
○ No
○ Other (please specify)

If you ticked 'yes' or 'other', please give details here. For example, 'I walk a mile to work 3 days a week' or 'the physio has given me some exercises'.

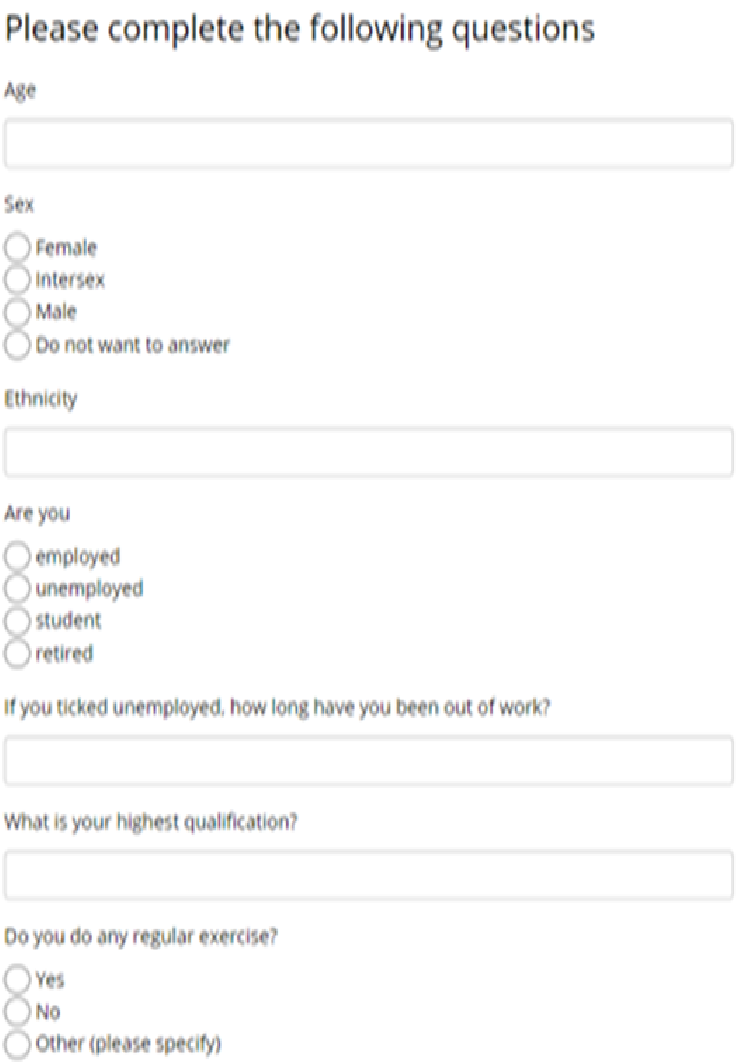
Height

Please select...

Weight

Please select...

Demographic questionnaire (controls)



Please complete the following questions

Age

Sex

○ Female
○ Intersex
○ Male
○ Do not want to answer

Ethnicity

Are you

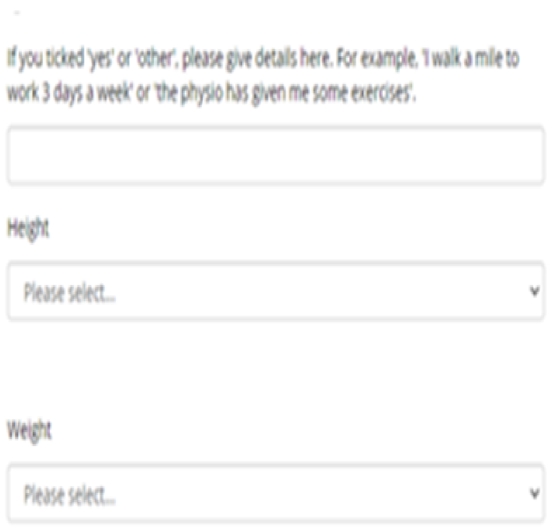
○ employed
○ unemployed
○ student
○ retired

If you ticked unemployed, how long have you been out of work?

What is your highest qualification?

Do you do any regular exercise?

○ Yes
○ No
○ Other (please specify)



If you ticked 'yes' or 'other', please give details here. For example, 'I walk a mile to work 3 days a week' or 'the physio has given me some exercises'.

Height

Please select...

Weight

Please select...

APPENDIX E

Interoception questionnaire

Body Perception Questionnaire Body Awareness Very Short Form
Stephen W. Porges © 1993, 2015

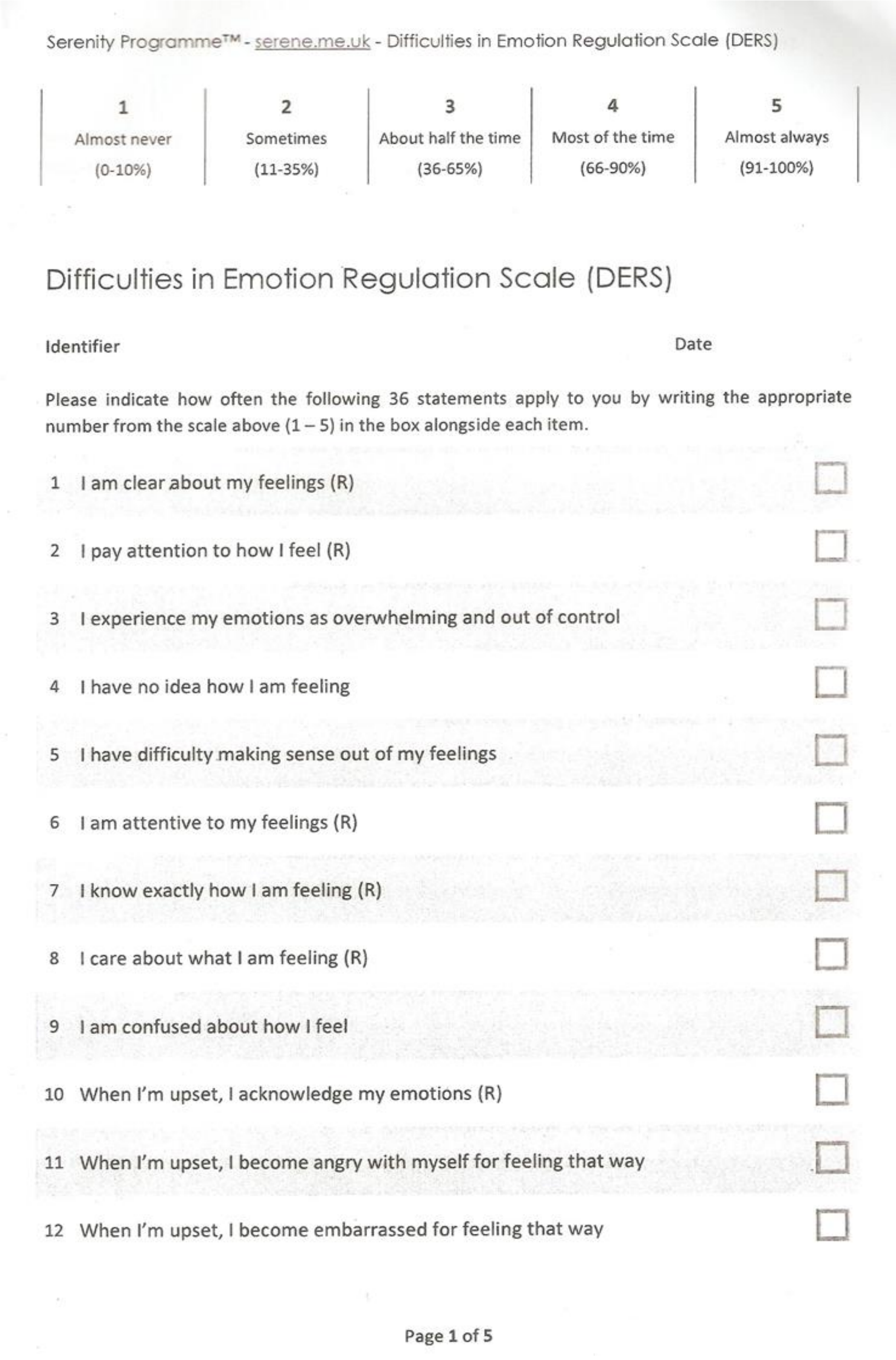
Please rate your awareness on each of the characteristics described below. Select the answer that most accurately describes you.

During most situations I am aware of:

		Never	Occasionally	Sometimes	Usually	Always
1	My mouth being dry	○	○	○	○	○
2	How fast I am breathing	○	○	○	○	○
3	A swelling of my body or parts of my body	○	○	○	○	○
4	Muscle tension in my arms and legs	○	○	○	○	○
5	A bloated feeling because of water retention	○	○	○	○	○
6	Goose bumps	○	○	○	○	○
		Never	Occasionally	Sometimes	Usually	Always
7	Stomach and gut pains	○	○	○	○	○
8	Stomach distension or bloatedness	○	○	○	○	○
9	Tremor in my lips	○	○	○	○	○
10	The hair on the back of my neck "standing up"	○	○	○	○	○
11	An urge to swallow	○	○	○	○	○
12	How hard my heart is beating	○	○	○	○	○
		Never	Occasionally	Sometimes	Usually	Always

APPENDIX F

Emotion regulation questionnaire



1	2	3	4	5
Almost never	Sometimes	About half the time	Most of the time	Almost always
(0-10%)	(11-35%)	(36-65%)	(66-90%)	(91-100%)

Difficulties in Emotion Regulation Scale (DERS)

Identifier

Date

Please indicate how often the following 36 statements apply to you by writing the appropriate number from the scale above (1 – 5) in the box alongside each item.

1 I am clear about my feelings (R) ☐

2 I pay attention to how I feel (R) ☐

3 I experience my emotions as overwhelming and out of control ☐

4 I have no idea how I am feeling ☐

5 I have difficulty making sense out of my feelings ☐

6 I am attentive to my feelings (R) ☐

7 I know exactly how I am feeling (R) ☐

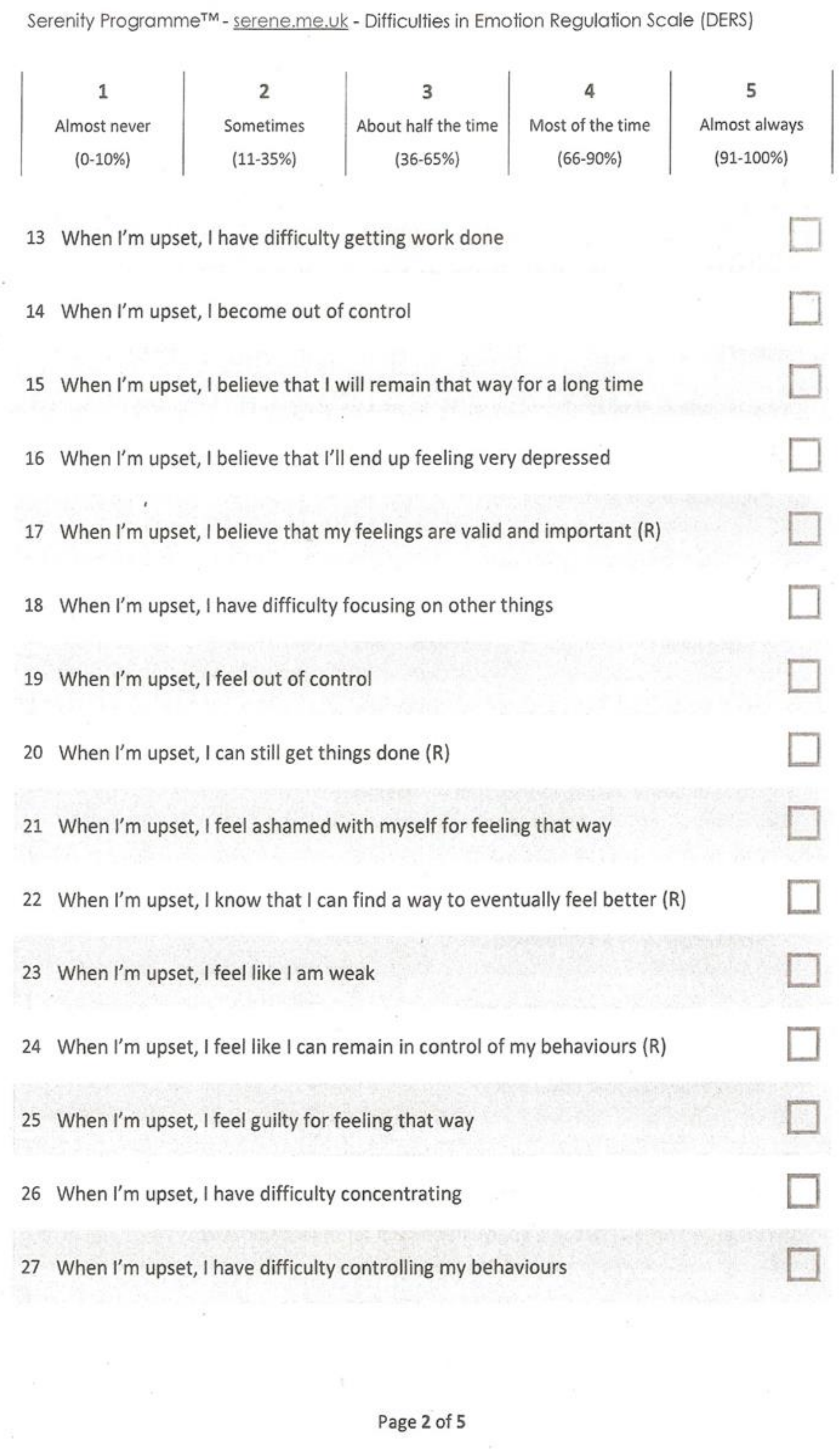
8 I care about what I am feeling (R) ☐

9 I am confused about how I feel ☐

10 When I'm upset, I acknowledge my emotions (R) ☐

11 When I'm upset, I become angry with myself for feeling that way ☐

12 When I'm upset, I become embarrassed for feeling that way ☐



Serenity Programme™ - serene.me.uk - Difficulties in Emotion Regulation Scale (DERS)

1	2	3	4	5
Almost never (0-10%)	Sometimes (11-35%)	About half the time (36-65%)	Most of the time (66-90%)	Almost always (91-100%)

13 When I'm upset, I have difficulty getting work done ☐

14 When I'm upset, I become out of control ☐

15 When I'm upset, I believe that I will remain that way for a long time ☐

16 When I'm upset, I believe that I'll end up feeling very depressed ☐

17 When I'm upset, I believe that my feelings are valid and important (R) ☐

18 When I'm upset, I have difficulty focusing on other things ☐

19 When I'm upset, I feel out of control ☐

20 When I'm upset, I can still get things done (R) ☐

21 When I'm upset, I feel ashamed with myself for feeling that way ☐

22 When I'm upset, I know that I can find a way to eventually feel better (R) ☐

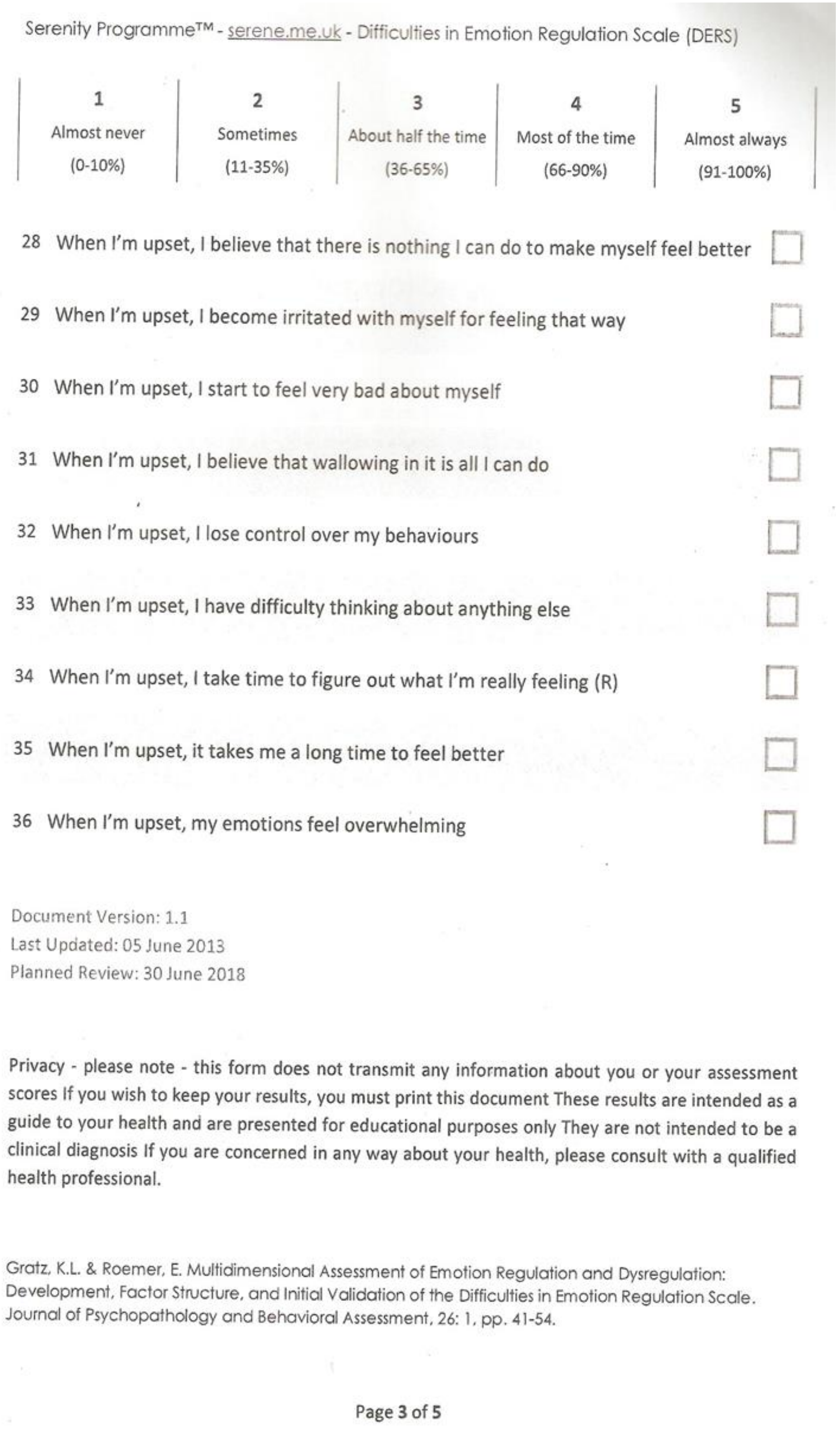
23 When I'm upset, I feel like I am weak ☐

24 When I'm upset, I feel like I can remain in control of my behaviours (R) ☐

25 When I'm upset, I feel guilty for feeling that way ☐

26 When I'm upset, I have difficulty concentrating ☐

27 When I'm upset, I have difficulty controlling my behaviours ☐



Serenity Programme™ - serene.me.uk - Difficulties in Emotion Regulation Scale (DERS)

1	2	3	4	5
Almost never	Sometimes	About half the time	Most of the time	Almost always
(0-10%)	(11-35%)	(36-65%)	(66-90%)	(91-100%)

28 When I'm upset, I believe that there is nothing I can do to make myself feel better ☐

29 When I'm upset, I become irritated with myself for feeling that way ☐

30 When I'm upset, I start to feel very bad about myself ☐

31 When I'm upset, I believe that wallowing in it is all I can do ☐

32 When I'm upset, I lose control over my behaviours ☐

33 When I'm upset, I have difficulty thinking about anything else ☐

34 When I'm upset, I take time to figure out what I'm really feeling (R) ☐

35 When I'm upset, it takes me a long time to feel better ☐

36 When I'm upset, my emotions feel overwhelming ☐

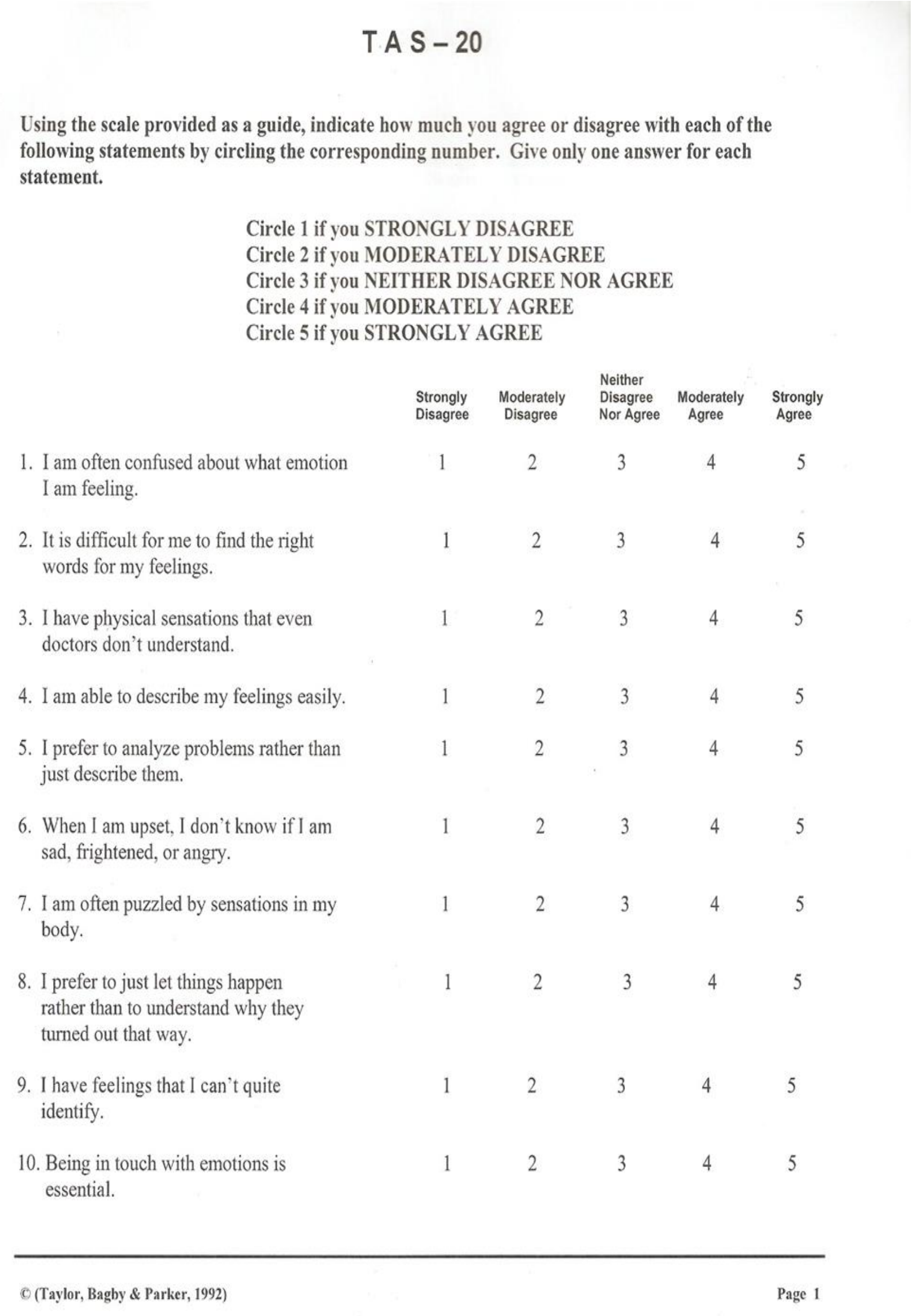
Document Version: 1.1
Last Updated: 05 June 2013
Planned Review: 30 June 2018

Privacy - please note - this form does not transmit any information about you or your assessment scores If you wish to keep your results, you must print this document These results are intended as a guide to your health and are presented for educational purposes only They are not intended to be a clinical diagnosis If you are concerned in any way about your health, please consult with a qualified health professional.

Gratz, K.L. & Roemer, E. Multidimensional Assessment of Emotion Regulation and Dysregulation: Development, Factor Structure, and Initial Validation of the Difficulties in Emotion Regulation Scale. Journal of Psychopathology and Behavioral Assessment, 26: 1, pp. 41-54.

APPENDIX G

Alexithymia questionnaire

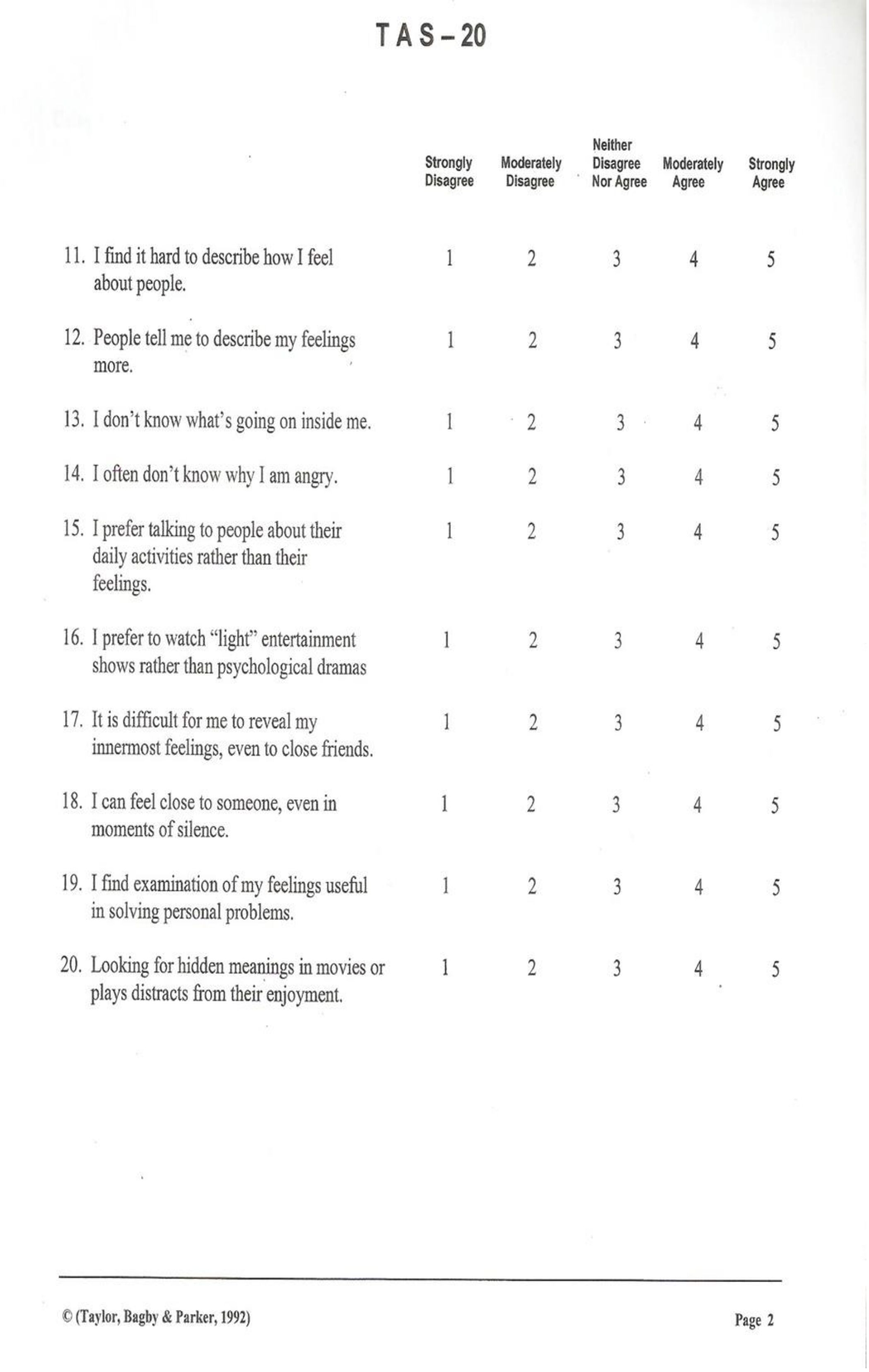


T A S – 20

Using the scale provided as a guide, indicate how much you agree or disagree with each of the following statements by circling the corresponding number. Give only one answer for each statement.

Circle 1 if you STRONGLY DISAGREE
Circle 2 if you MODERATELY DISAGREE
Circle 3 if you NEITHER DISAGREE NOR AGREE
Circle 4 if you MODERATELY AGREE
Circle 5 if you STRONGLY AGREE

	Strongly Disagree	Moderately Disagree	Neither Disagree Nor Agree	Moderately Agree	Strongly Agree
1. I am often confused about what emotion I am feeling.	1	2	3	4	5
2. It is difficult for me to find the right words for my feelings.	1	2	3	4	5
3. I have physical sensations that even doctors don't understand.	1	2	3	4	5
4. I am able to describe my feelings easily.	1	2	3	4	5
5. I prefer to analyze problems rather than just describe them.	1	2	3	4	5
6. When I am upset, I don't know if I am sad, frightened, or angry.	1	2	3	4	5
7. I am often puzzled by sensations in my body.	1	2	3	4	5
8. I prefer to just let things happen rather than to understand why they turned out that way.	1	2	3	4	5
9. I have feelings that I can't quite identify.	1	2	3	4	5
10. Being in touch with emotions is essential.	1	2	3	4	5

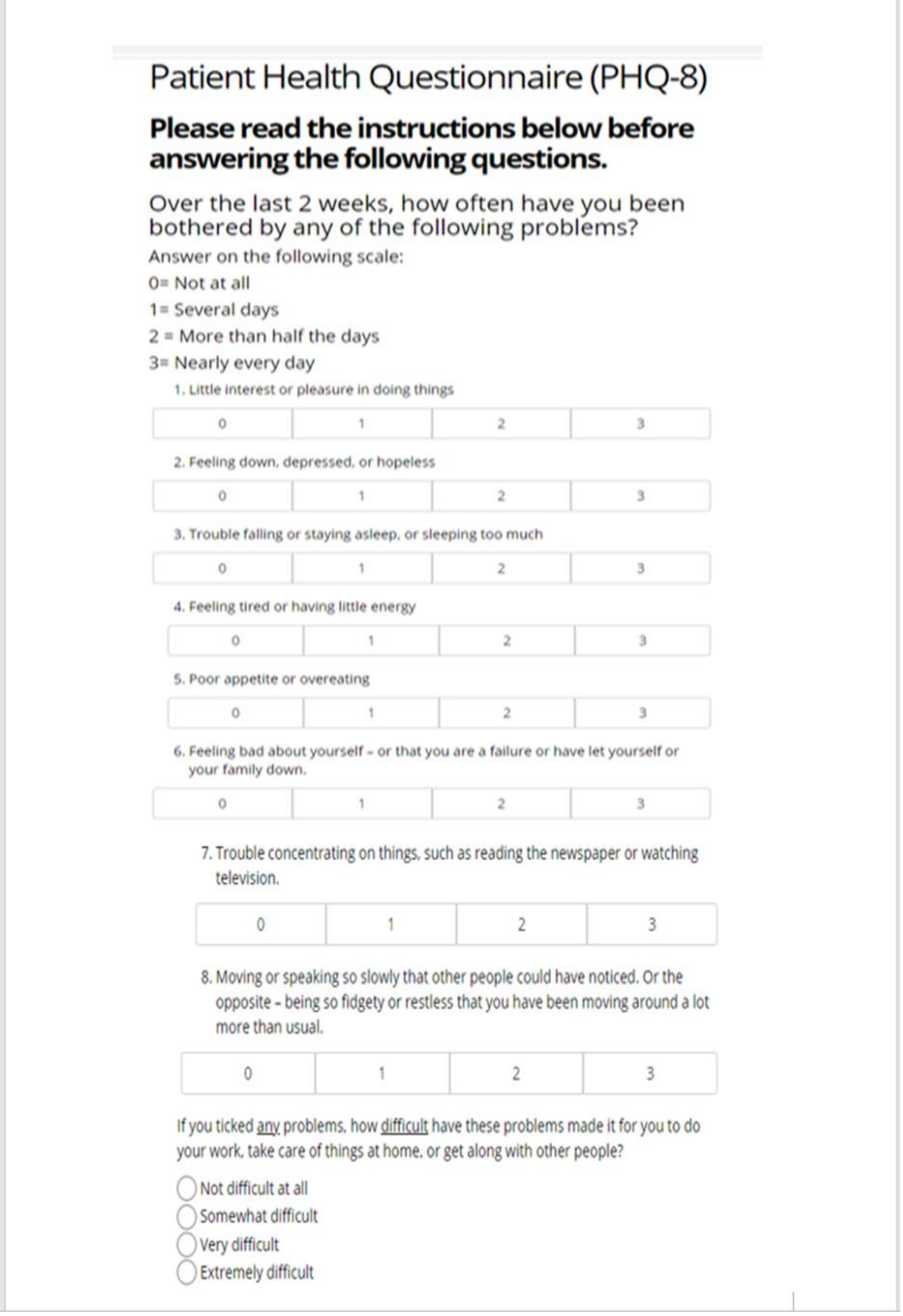


T A S – 20

	Strongly Disagree	Moderately Disagree	Neither Disagree Nor Agree	Moderately Agree	Strongly Agree
11. I find it hard to describe how I feel about people.	1	2	3	4	5
12. People tell me to describe my feelings more.	1	2	3	4	5
13. I don't know what's going on inside me.	1	2	3	4	5
14. I often don't know why I am angry.	1	2	3	4	5
15. I prefer talking to people about their daily activities rather than their feelings.	1	2	3	4	5
16. I prefer to watch "light" entertainment shows rather than psychological dramas	1	2	3	4	5
17. It is difficult for me to reveal my innermost feelings, even to close friends.	1	2	3	4	5
18. I can feel close to someone, even in moments of silence.	1	2	3	4	5
19. I find examination of my feelings useful in solving personal problems.	1	2	3	4	5
20. Looking for hidden meanings in movies or plays distracts from their enjoyment.	1	2	3	4	5

APPENDIX H

Depressive symptoms questionnaire



Patient Health Questionnaire (PHQ-8)

Please read the instructions below before answering the following questions.

Over the last 2 weeks, how often have you been bothered by any of the following problems?

Answer on the following scale:

0= Not at all
1= Several days
2 = More than half the days
3= Nearly every day

1. Little interest or pleasure in doing things

0	1	2	3

2. Feeling down, depressed, or hopeless

0	1	2	3

3. Trouble falling or staying asleep, or sleeping too much

0	1	2	3

4. Feeling tired or having little energy

0	1	2	3

5. Poor appetite or overeating

0	1	2	3

6. Feeling bad about yourself – or that you are a failure or have let yourself or your family down.

0	1	2	3

7. Trouble concentrating on things, such as reading the newspaper or watching television.

0	1	2	3

8. Moving or speaking so slowly that other people could have noticed. Or the opposite – being so fidgety or restless that you have been moving around a lot more than usual.

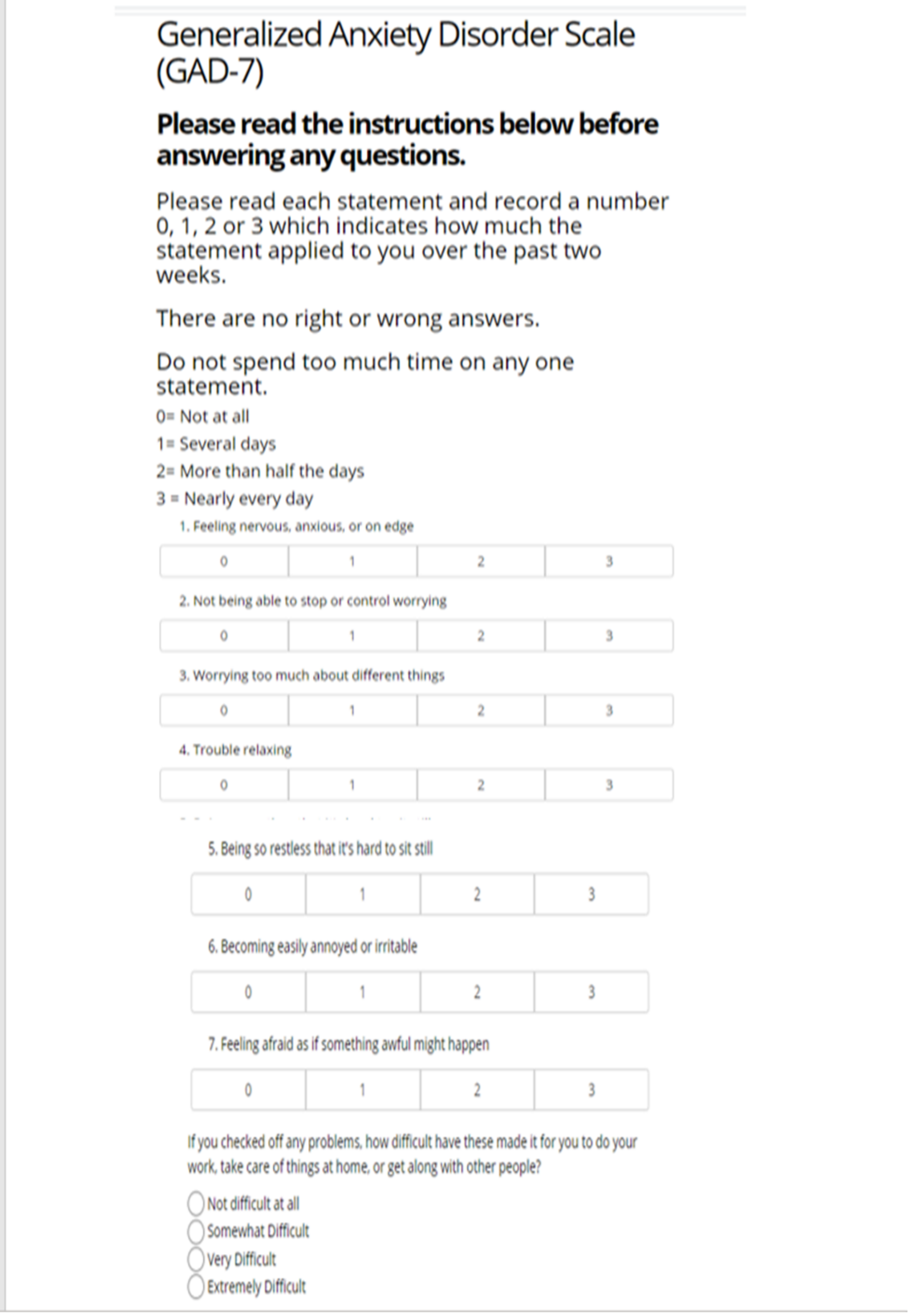
0	1	2	3

If you ticked any problems, how difficult have these problems made it for you to do your work, take care of things at home, or get along with other people?

○ Not difficult at all
○ Somewhat difficult
○ Very difficult
○ Extremely difficult

APPENDIX I

Anxiety questionnaire



Generalized Anxiety Disorder Scale (GAD-7)

Please read the instructions below before answering any questions.

Please read each statement and record a number 0, 1, 2 or 3 which indicates how much the statement applied to you over the past two weeks.

There are no right or wrong answers.

Do not spend too much time on any one statement.

0= Not at all
1= Several days
2= More than half the days
3 = Nearly every day

1. Feeling nervous, anxious, or on edge

0	1	2	3

2. Not being able to stop or control worrying

0	1	2	3

3. Worrying too much about different things

0	1	2	3

4. Trouble relaxing

0	1	2	3

5. Being so restless that it's hard to sit still

0	1	2	3

6. Becoming easily annoyed or irritable

0	1	2	3

7. Feeling afraid as if something awful might happen

0	1	2	3

If you checked off any problems, how difficult have these made it for you to do your work, take care of things at home, or get along with other people?

○ Not difficult at all
○ Somewhat Difficult
○ Very Difficult
○ Extremely Difficult

APPENDIX J

PARTICIPANTS NEEDED FOR AN ONLINE RESEARCH STUDY

Investigating self-perception of emotion in individuals with functional seizures (FS): using self-report measures and an emotional Stroop task.

We need two groups of people: 1. people with functional seizures, and
2. people without seizures of any type.

The research is investigating how people with and without functional seizures detect and process emotion.

You will be required to fill out six questionnaires and take part in a short experimental task. This usually takes up to an hour to do.

Everything will take place online and you can choose to receive a £10 voucher once you submit your answers.

GROUP 1	GROUP 2
If you experience functional seizures and want to take part, you should also:	If you do not experience functional seizures and want to take part, you should also:
• Be in the UK. • Be at least 18 years old. • Be able to understand and read English. • Have access to the Internet and a laptop or PC.	• Be in the UK. • Be at least 18 years old. • Be able to understand and read English. • Have access to the Internet and a laptop or PC.
You cannot take part if you have a diagnosis of epilepsy, dementia, or other neurological disorder.	You cannot take part if you have a diagnosis of epilepsy, dementia, or other neurological disorder.
If you meet the criteria above, you can take part by clicking on the link here: https://research.sc/participant/login/dynamic/74B7E7FD-A3D4-43BE-9BB6-008A217FD8A6	If you meet the criteria above, you can take part by clicking on the link here: https://research.sc/participant/login/dynamic/D0AC781F-C271-4883-AF4D-CA41754B4CA0

If you have any questions about the research or would like some more information, please email Shaista Meer at um08sm@leeds.ac.uk .

*This study has been reviewed and given favourable opinion by the School of Psychology research Ethics Committee (SOPREC), Ref: PSYC-332, Date 20/10/2021.

APPENDIX K

TRANSCRIPT OF VIDEO

Hello, I'm Shaista Meer and this study is part of my PhD research. So, thank you for taking an interest. Unfortunately, we can’t meet face to face at the moment, so I've added this short video to explain, to you, what you can expect during the experiment.

After I stop talking, you’ll be taken to some information. I'm sorry, I know it's long but please do read it. It will explain to you a little bit about why I'm doing the research. And also, how we treat the information that you give me.

After you agree to take part in the research, you’ll be taken to some questions. There are six questionnaires for you to complete. There will be a break between the sections.

After you’ve completed the questionnaires, you then take part in a short task. There will be some instructions, and a chance for you to practice. Please do respond as quickly as you can.

The whole process usually doesn’t usually take longer than an hour, but you have up to two hours to complete it. If you don’t complete everything after the two hours then the experiment will time out. And your responses won’t be included in the research.

To say ‘thank you’ to everyone who completes the tasks, I would like to send out a small voucher. So, if you’d like to receive the voucher, add your email address when prompted – so that I know who to send it to.

Finally, I'd like to reassure you that everything you provide – all your data, will be treated confidentially. If you have any questions about the research before you start, please email me at **um08sm@leeds.ac.uk.**

Thank you.

APPENDIX L

Exploring emotional responses to life events in individuals who experience functional seizures (FS).

PARTICIPANTS NEEDED

- Individuals, in the UK, with functional seizures are invited to take part in online interviews.
- The interview will be carried out using Zoom, so you will not have to travel anywhere.
- The interview can be audio only, if you do not want to turn the camera on.
- All interviews will be confidential.
- After the interview, you will be sent a voucher as a 'thank you' for giving up your time.

To take part you need to be:

- In the UK.
- At least 18 years old.
- Have functional seizures.
- Have experienced a life event in the past 12 months. A life event is usually something major or important like moving house, changing jobs, having an operation, getting married.

The interviews are being carried out as part of PhD research. The research is interested in how people with functional seizures experience their emotions. This is based on some theories about FS that mention responses to emotions as being important.

The interview will concentrate on a life event that you have experienced in the last 12 months.

The interviews will be arranged at a time that is suitable for you.
If you have any questions about the research or would like to take part, please email Shaista Meer at um08sm@leeds.ac.uk to send you some more information.

*This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee (SOPREC): Ref PSYC-51, Date 19-05-2020.

APPENDIX M

PARTICIPANT INFORMATION SHEET

Exploring emotional responses to life events in individuals who experience functional seizures (FS).

Invitation to take part in research:

You are being invited to take part in a research study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. If anything is not clear, or you would like more information, please contact the lead researcher (Shaista Meer).

What is the purpose of the research?

The main aim of this research is to gain a better understanding of how people with functional seizures/non-epileptic seizures/dissociative seizures describe and experience life events, and how they describe feeling at the time of the event.

Life events are usually referred to as major or important events, and examples of these include moving house, changing jobs, having an operation or unexpected hospital stay, getting married. We are interested in people who have experienced at least one life event in the past 12 months.
We hope that this will give individuals the opportunity to express, in their own words, their experiences. This will help us better understand the experiences of people with the condition and also help us to investigate whether this complements or is contrary to theories about why people experience FS. We hope that having an insight into individual's experiences and thoughts may lead to future therapeutic approaches being more informed so that they can be more effective for people with this condition.

Why have I been chosen?

You have been chosen because we need people who have FS to take part. To qualify for the study, you should also:

- Have experienced a life event in the past 12 months;
- Be at least 18 years old;
- Be able to read and understand English, Urdu or Punjabi;
- Be able to decide whether or not you would like to take part;
- Be well enough to take part in the study.
-

You cannot take part if you:

- Have any major health conditions such as stroke, epilepsy, dementia, or other neurological conditions;
- Are currently feeling distressed, self-harming or having thoughts about harming yourself.

Do I have to take part?

No.

It is entirely up to you whether you choose to take part, and you are free to withdraw from the study at any point. If you decide that you no longer wanted your data to be included, you can request this to be withdrawn up until the point it has been analysed.

What is involved if I decide to take part?
If you decide to take part, then the researcher will arrange a suitable date and time for the interview. You will be interviewed about your life event and your experience of this. This could take up to 1 ½ hours. The interview will be via Zoom and can be audio only if you prefer not to turn on the video. Before anything begins you will be given a chance to ask any questions that you may have. You will be asked to sign a consent form and provide some information such as your age, employment and education. Everyone who takes part is asked to answer the same questions.

The interview will involve the researcher asking you questions about the recent life event that you experienced and how you felt around that time. The interview will be recorded so that your answers can be analysed against the responses from other individuals with the FS.

You will receive a £20 'thank you' voucher after completing the interview.

What are the possible disadvantages and risks of taking part?
There are no major disadvantages or risks expected to be experienced by anyone taking part. It is possible that answering questions about how you felt at a particular time might remind you that you had low mood or felt anxious, so some people might feel uncomfortable or upset talking about it now**.** If you find yourself being upset or worried by any of the questions please talk to the researcher about this.

There is a small risk that anyone taking part could have a seizure during this time. If you think this may be a possibility then please let the researcher know.

What are the benefits of taking part?
Although this research may not directly benefit you, we hope that this research will help us to have a better understanding of how people with FS respond when going through a life event. This will help us to inform future therapeutic interventions to support people with FS better.

What happens if I have a 'flare up' on the day we are supposed to meet?
We understand that how you feel can change from day to day. If you are unwell or feel unable to talk on the day – just let the researcher know and another date will be arranged. We will try to be as flexible as possible.

Will my taking part be kept confidential and what will happen to the data I provide?
All information that is collected during the study will be treated confidentially. All participants will be assigned a unique code that will anonymise their

responses. The data collected (your responses to the interview questions and demographic information) will be kept for ten years in the anonymised format. If the researcher leaves the University before the end of the ten years, the main supervisor **(Prof Guthrie**) will look after the anonymised data. The anonymised data will be stored on a secure network at the University of Leeds.

Only the lead researcher (Shaista Meer) will have access to your personal information such as your name and address. These details will be kept on a password protected computer at the University of Leeds and will be destroyed once recruitment has finished.

Who will know that I am taking part in the study?
We will not tell anyone else about your participation unless you were to tell the researcher that you are thinking of or have plans to harm yourself. In this situation, after discussing this with you, the researcher would call NHS 111 to get advice and may also speak to her supervisor. This is to make sure that you get some support and would be done in your presence and with your knowledge.

The researcher will also let her supervisor know where she will be meeting you so that the supervisor can be aware of the researcher’s location.

Who is responsible for looking after my information?
The University of Leeds is the sponsor for this study based in the United Kingdom. This means that the University of Leeds is responsible for looking after your information. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly.
To safeguard your rights, we will use the minimum personally-identifiable information possible. This means that after the analysis has been completed, we will remove some details from the data, such as your occupation or ethnicity to further anonymise the information. The anonymised data will be stored for ten years after the study has finished.

Who has access to my identifiable data?
The lead researcher will keep your name and contact details confidential and will not pass this information to The University of Leeds. The lead researcher will use this information to contact you about the research study. Your contact details will be destroyed once recruitment to the study has ended. Certain individuals from The University of Leeds and regulatory organisations may look at your research records to check the accuracy of the research study. The University of Leeds will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

Can I access the information that you have about me?

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. You can find out more about how we use your information http://www.leeds.ac.uk/secretariat/data_protection.html.

What will happen to the results of the study?
Due to the data being analysed as a group, it will not be possible to provide any specific information about your own data. It will not be possible to identify participants from any of the data that will be published and the anonymised data may be used to support further ethically approved research.
This study is part of a PhD and the results of the research will be presented at academic conferences and published in academic journals**.** If you are interested in the findings then you can contact the lead researcher for more information.

Who is organising and funding the research?
This research is funded by the University of Leeds.

Further information: If you have any questions or would like to discuss taking part, then please contact the lead researcher:

Miss Shaista Meer um08sm@leeds.ac.uk; tel: 0113 3430808 or **Prof Else Guthrie (supervisor) e.a.guthrie@leeds.ac.uk; tel: 0113 3432442.**
If you have been affected by the research then please discuss this further with the researcher.

If you feel distressed then you can access support through:

- Your local IAPT (Improving Access to Psychological Therapies) service
- The Samaritans on Freephone 116 123
-

If you want to make a complaint related to this research then you can speak to Clare Skinner, Faculty Head of Research and Innovation Support, Email: governance-ethics@leeds.ac.uk, tel: 0113 343 4897 Or the independent organisation: Information Commissioner's Office on 0303 123 1113.

Ethics approval:
This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee (S0PREC): Ref PSYC-51, Date 19-05-2020.

APPENDIX N

Consent to take part in 'Exploring emotional responses to life events in individuals who experience functional seizures'	Add your initials next to the statement if you agree
I confirm that I have read and understand the information sheet dated **19th May 2020** explaining the above research project, and I have had the opportunity to ask questions about the project.	
I understand that my participation is voluntary and I am free to withdraw at any time without giving any reason, and without there being any negative consequences. In addition, should I wish not to answer any particular question or questions, I am free to decline. If I choose to withdraw from the study I will contact the lead researcher Shaista Meer (um08sm@leeds.ac.uk / 0113 3430808), or Prof Else Guthrie supervisor:e.a.guthrie@leeds.ac.uk / 0113 3432442).	
I understand that relevant sections of the data collected during the study may be looked at by individuals from the Faculty of Medicine and Health, University of Leeds, from regulatory authorities, where it is relevant to my participation in this study. I give permission for these individuals to have access to my data.	
I give permission for members of the research team to have access to my anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in the report or reports that result from the research. I understand that my responses will be kept strictly confidential.	
I understand that if I decide to withdraw from the study, any data that I have provided will be used in the research unless I request that my data be removed. I understand that I can request this up until the point the data has been analysed.	
I agree for the ethically approved data collected from me to be stored and used in relevant future research by the lead researchers (Shaista Meer & Prof Else Guthrie) for a period of up to 10 years. I understand it will be used in an anonymised form.	
I confirm that I do not have any major health conditions which may affect me taking part in the research, such as stroke, a cardiac condition, epilepsy, dementia, or other neurological conditions. I also confirm that I am not currently self-harming.	
I agree to the research interview being recorded so that the data (i.e. what I say in the interview) can be analysed.	
If I report any plans to harm myself, I understand that the interview will stop. I agree to speak to my GP or contact NHS 111 for advice. I understand that the researcher may contact her supervisor (Prof Else Guthrie) also to talk about what I have told her.	
I agree to take part in the above research project.	

Name of participant		
Participant's signature		
Name of lead researcher		
Researcher's Signature		

Once this has been signed by all parties the participant should receive a copy of the signed and dated participant consent form, the letter/ information sheet and any other written information provided to the participants. A copy of the signed and dated consent form should be kept with the project's main documents which must be kept in a secure location.

This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee (S0PREC): Ref PYSC-51, Date 19-05-2020.

APPENDIX O

Participant information

Participant ID:

Age: **Gender**: **Ethnicity**:
Are you: employed / unemployed / student / retired (circle answer)
If you are employed what is your occupation?
If you are unemployed, how long have you been out of work?

____

If you are retired, how long have you been retired?

____

What is your highest qualification?

When did you first experience a seizure?
When did you receive a diagnosis of non-epileptic seizures?
Have you experienced any of the following over the last 12 months? Please tick all that apply:

☐ Moved house/ flat
☐ Got engaged/married
☐ Relationship breakdown
☐ Had an accident
☐ Hospitalised
☐ Received a new diagnosis
☐ Pregnancy
☐ Gave birth/ adopted a child
☐ Family member/someone close to you had an accident
☐ Family member/someone close to you hospitalised
☐ Changed jobs
☐ Made redundant
☐ Started a new educational course
☐ Family member/ someone close to you die
☐ Pet died
☐ Been assaulted
☐ Victim of a burglary/ robbery
☐ Any other event not mentioned above. Please define the event:

This study has been reviewed and given favourable opinion by the School of Psychology Research Ethics Committee (SOPREC): Ref PSYC-51, Date 19-05-2020.

APPENDIX P

Interview schedule exploration of life events and emotional responses in the context of non-epileptic seizures (NES).

- You've mentioned that you went through/ experienced (mention the event highlighted on the demographic form) recently. Can you tell me more about what happened?

If more than one event mentioned – ask participant to start with the most recent.

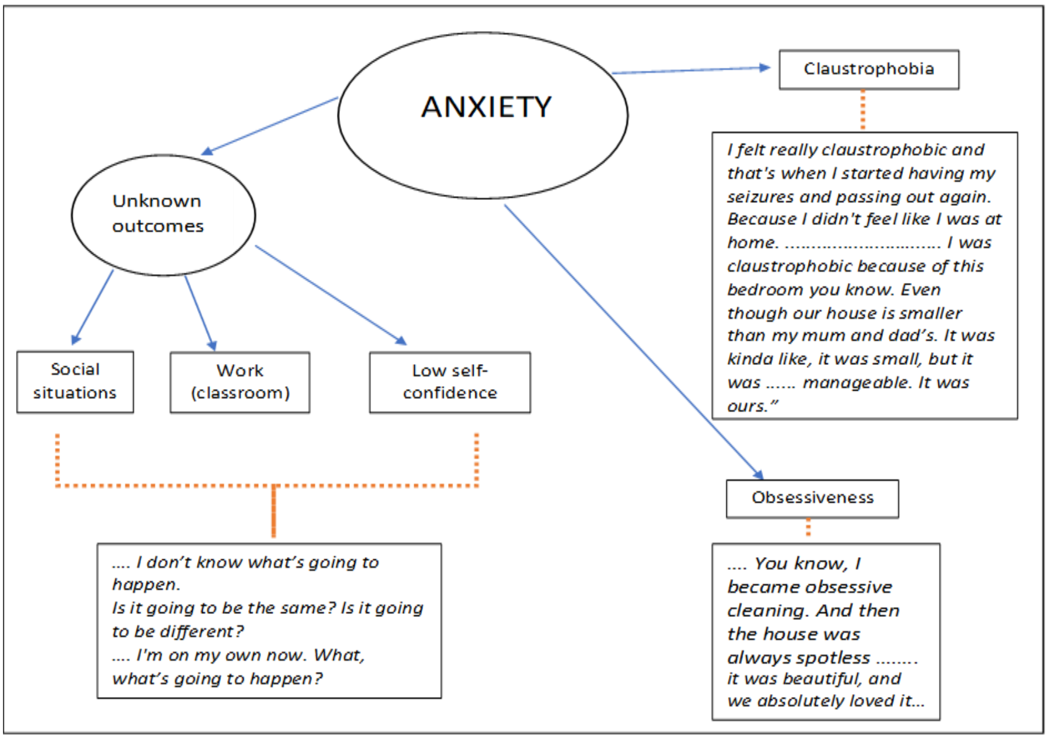
  - What happened?
  - Circumstances leading up to the event?
  - Details about the event e.g. moved to a better/worse neighbourhood.
  - Anyone else involved?
  - Was it your choice
- I'd like to know more about (the event), can you describe how you felt at that time?
  - Do you remember how your body felt at that time?
  - Can you describe if you experienced any emotions at that time?
  - What was going through your mind? Did you remember having any thoughts about what was happening?
  - Did people around you know how you felt?
- I'd like to get a better understanding of what it was like for you. What kind of effect do you think (the event) had on you?
  - No impact? Positive? Negative?
  - Did you find yourself thinking about it a lot?
  - Do you still think about it?
  - How does thinking about the event make you feel?
- Are you comfortable to talk about your seizures? At the time of (event) you said that you felt (mention emotion/physical feelings/specific thoughts) did you experience any seizures around this time?
  - Why do you think this happened? Related to emotional/physical response? Coincidence? Always happens at that time?
- In your opinion, is there any connection between how you felt about (mention event) and your seizures?
  - What makes you say that?
  - Do you feel any emotions around the time you have a seizure?
  - Any triggers? For you/others?
  - Is there anything that always happens before a seizure?
  - Do they always happen at the same time? Same place? Around same people?
  - Is there anything that always happens before a seizure?
  - Did it happen then?
  - Was there anything different about that times and how your seizures were?

- Is there anything that I haven't asked you but you think is important for me to know about your experiences related to your seizures?

- Would you be ok with me contacting you again to talk to you about how I have made sense/interpreted what you have told me today?

APPENDIX Q

Figure 4.1 This diagram illustrates the different ways in which anxiety manifests for Samantha, in relation to her life events mentioned during her interview.

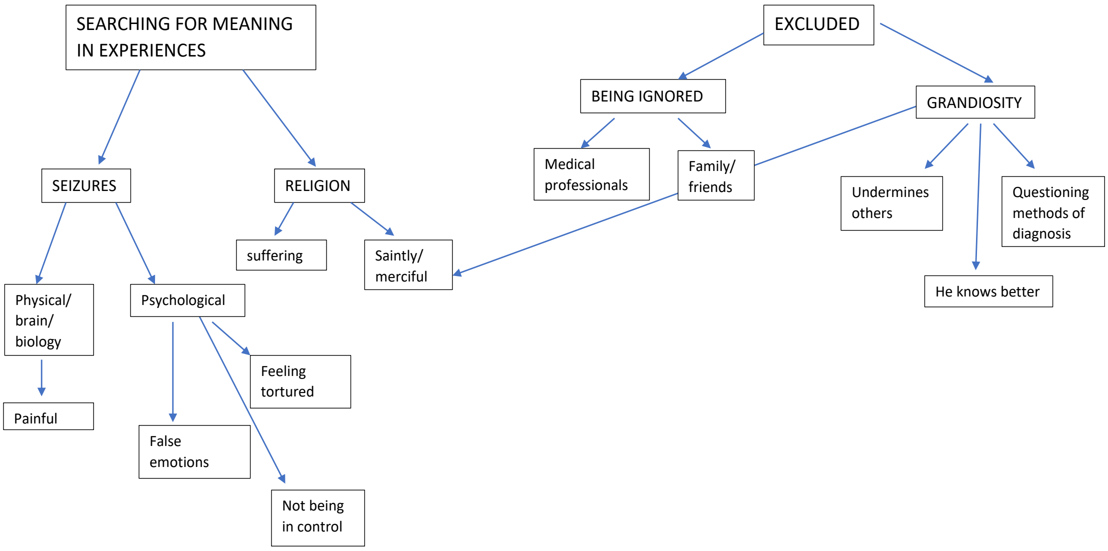


I felt really claustrophobic and that's when I started having my seizures and passing out again. Because I didn't feel like I was at home. ………………………… I was claustrophobic because of this bedroom you know. Even though our house is smaller than my mum and dad's. It was kinda like, it was small, but it was …… manageable. It was ours."

…. I don't know what's going to happen.
Is it going to be the same? Is it going to be different?
…. I'm on my own now. What, what's going to happen?

…. You know, I became obsessive cleaning. And then the house was always spotless …….. it was beautiful, and we absolutely loved it…

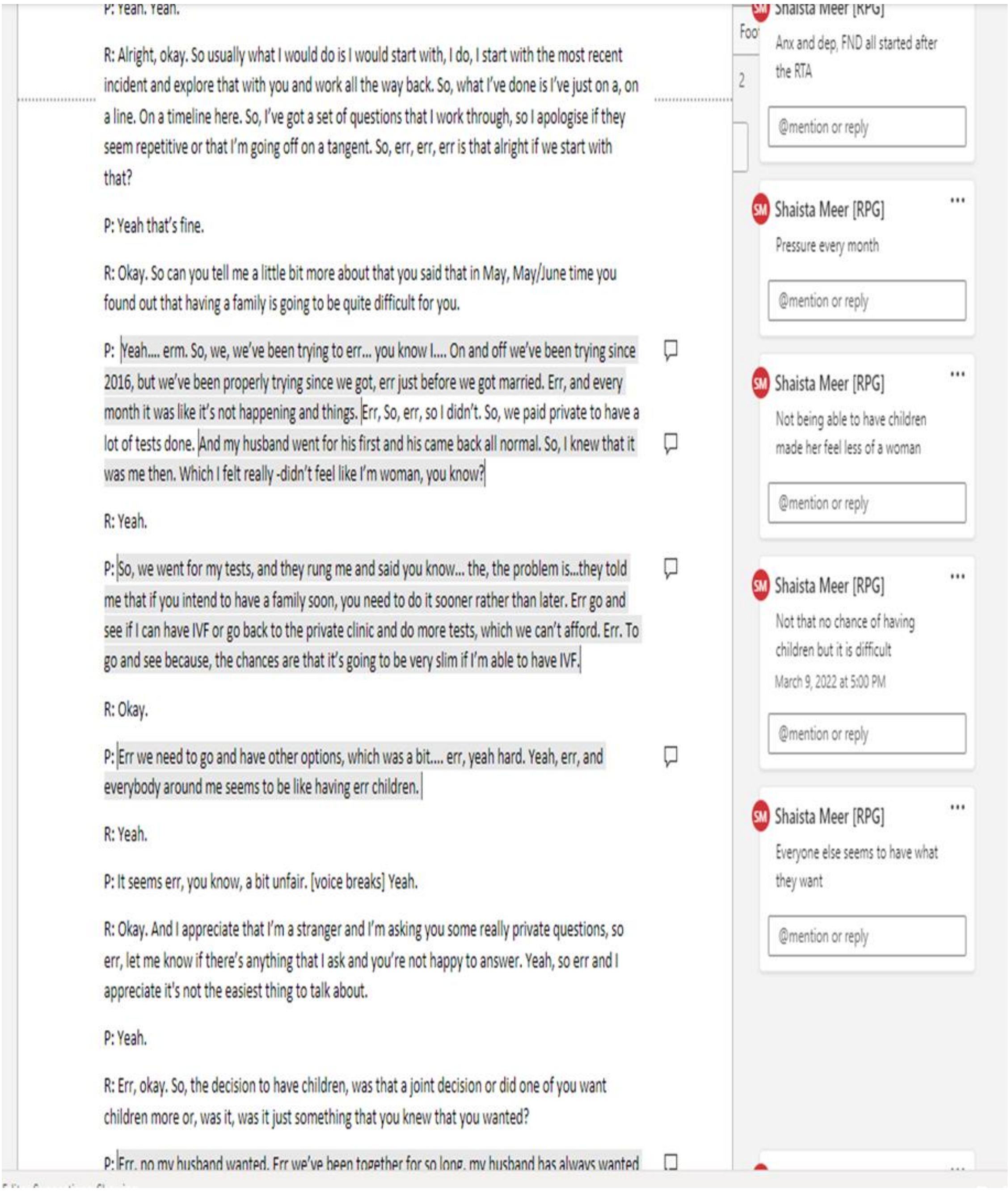
Figure 4.2 This figure shows how a tree of themes can be developed by clustering minor themes and examples, under overarching/umbrella themes. This example is taken from Craig's analysis.



APPENDIX R

Examples of the different stages of analysis

Figure 4.3 Annotations on individual participant transcripts



P: Yeah. Yeah.

R: Alright, okay. So usually what I would do is I would start with, I do, I start with the most recent incident and explore that with you and work all the way back. So, what I've done is I've just on a, on a line. On a timeline here. So, I've got a set of questions that I work through, so I apologise if they seem repetitive or that I'm going off on a tangent. So, err, err, err is that alright if we start with that?

P: Yeah that's fine.

R: Okay. So can you tell me a little bit more about that you said that in May, May/June time you found out that having a family is going to be quite difficult for you.

P: Yeah.... erm. So, we, we've been trying to err... you know I.... On and off we've been trying since 2016, but we've been properly trying since we got, err just before we got married. Err, and every month it was like it's not happening and things. Err, So, err, so I didn't. So, we paid private to have a lot of tests done. And my husband went for his first and his came back all normal. So, I knew that it was me then. Which I felt really -didn't feel like I'm woman, you know?

R: Yeah.

P: So, we went for my tests, and they rung me and said you know... the, the problem is...they told me that if you intend to have a family soon, you need to do it sooner rather than later. Err go and see if I can have IVF or go back to the private clinic and do more tests, which we can't afford. Err. To go and see because, the chances are that it's going to be very slim if I'm able to have IVF.

R: Okay.

P: Err we need to go and have other options, which was a bit.... err, yeah hard. Yeah, err, and everybody around me seems to be like having err children.

R: Yeah.

P: It seems err, you know, a bit unfair. [voice breaks] Yeah.

R: Okay. And I appreciate that I'm a stranger and I'm asking you some really private questions, so err, let me know if there's anything that I ask and you're not happy to answer. Yeah, so err and I appreciate it's not the easiest thing to talk about.

P: Yeah.

R: Err, okay. So, the decision to have children, was that a joint decision or did one of you want children more or, was it, was it just something that you knew that you wanted?

P: Err, no my husband wanted. Err we've been together for so long, my husband has always wanted

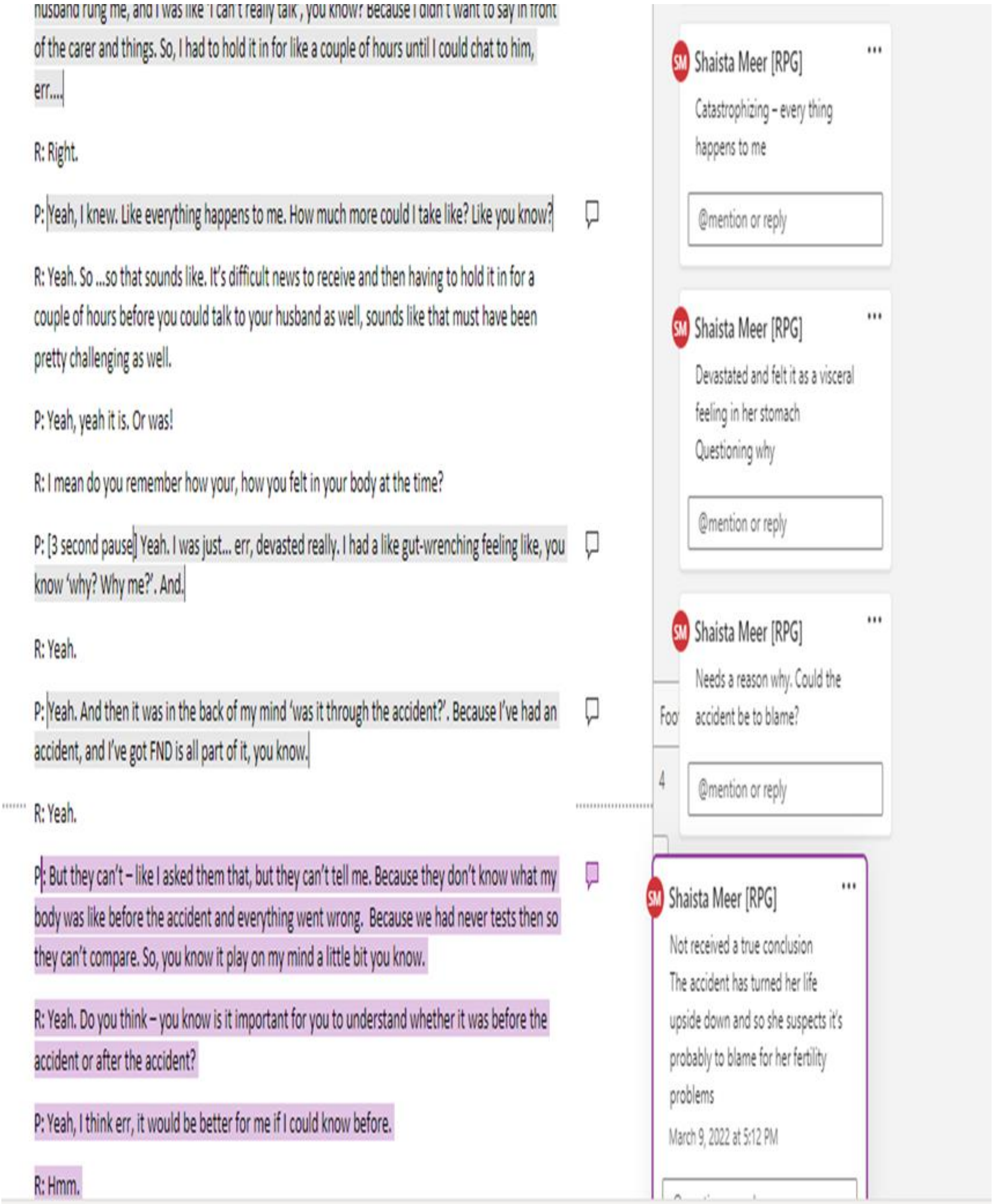
Shaista Meer [RPG]
Anx and dep, FND all started after the RTA

Shaista Meer [RPG]
Pressure every month

Shaista Meer [RPG]
Not being able to have children made her feel less of a woman

Shaista Meer [RPG]
Not that no chance of having children but it is difficult
March 9, 2022 at 5:00 PM

Shaista Meer [RPG]
Everyone else seems to have what they want



husband rung me, and I was like 'I can't really talk', you know? Because I didn't want to say in front of the carer and things. So, I had to hold it in for like a couple of hours until I could chat to him, err....

R: Right.

P: Yeah, I knew. Like everything happens to me. How much more could I take like? Like you know?

R: Yeah. So ...so that sounds like. It's difficult news to receive and then having to hold it in for a couple of hours before you could talk to your husband as well, sounds like that must have been pretty challenging as well.

P: Yeah, yeah it is. Or was!

R: I mean do you remember how your, how you felt in your body at the time?

P: [3 second pause] Yeah. I was just... err, devasted really. I had a like gut-wrenching feeling like, you know 'why? Why me?'. And.

R: Yeah.

P: Yeah. And then it was in the back of my mind 'was it through the accident?'. Because I've had an accident, and I've got FND is all part of it, you know.

R: Yeah.

P: But they can't – like I asked them that, but they can't tell me. Because they don't know what my body was like before the accident and everything went wrong. Because we had never tests then so they can't compare. So, you know it play on my mind a little bit you know.

R: Yeah. Do you think – you know is it important for you to understand whether it was before the accident or after the accident?

P: Yeah, I think err, it would be better for me if I could know before.

R: Hmm.

Shaista Meer [RPG]

Catastrophizing – every thing happens to me

Shaista Meer [RPG]

Devastated and felt it as a visceral feeling in her stomach

Questioning why

Shaista Meer [RPG]

Needs a reason why. Could the accident be to blame?

Shaista Meer [RPG]

Not received a true conclusion

The accident has turned her life upside down and so she suspects it's probably to blame for her fertility problems

March 9, 2022 at 5:12 PM

Figures 4.4 - 4.5 show the different strategies used to group PETs to explore and develop GETS.

Figure 4.4

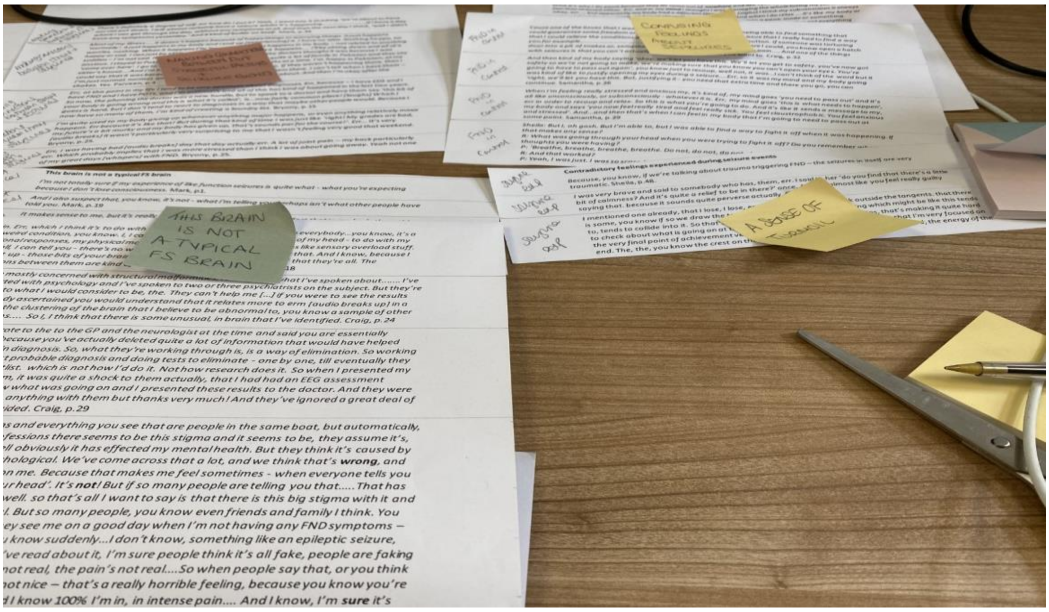
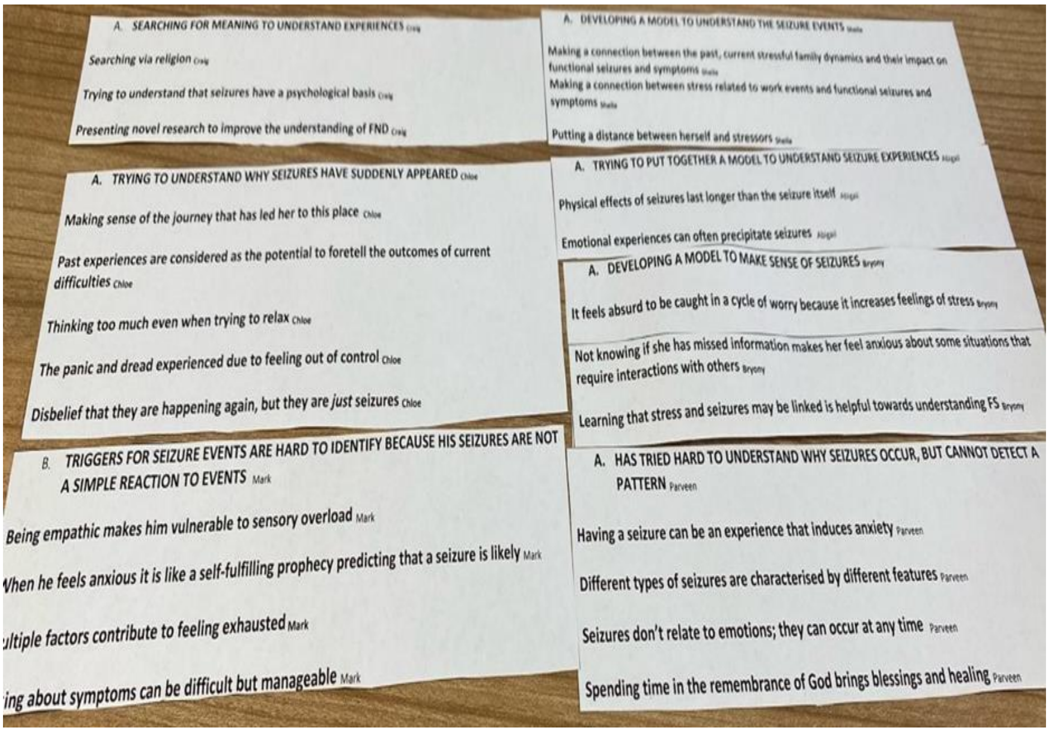


Figure 4.5



A. SEARCHING FOR MEANING TO UNDERSTAND EXPERIENCES Craig

Searching via religion Craig

Trying to understand that seizures have a psychological basis Craig

Presenting novel research to improve the understanding of FND Craig

A. TRYING TO UNDERSTAND WHY SEIZURES HAVE SUDDENLY APPEARED Chloe

Making sense of the journey that has led her to this place Chloe

Past experiences are considered as the potential to foretell the outcomes of current difficulties Chloe

Thinking too much even when trying to relax Chloe

The panic and dread experienced due to feeling out of control Chloe

Disbelief that they are happening again, but they are *just* seizures Chloe

B. TRIGGERS FOR SEIZURE EVENTS ARE HARD TO IDENTIFY BECAUSE HIS SEIZURES ARE NOT A SIMPLE REACTION TO EVENTS Mark

Being empathic makes him vulnerable to sensory overload Mark

Vhen he feels anxious it is like a self-fulfilling prophecy predicting that a seizure is likely Mark

ultiple factors contribute to feeling exhausted Mark

ing about symptoms can be difficult but manageable Mark

A. DEVELOPING A MODEL TO UNDERSTAND THE SEIZURE EVENTS Sheila

Making a connection between the past, current stressful family dynamics and their impact on functional seizures and symptoms Sheila

Making a connection between stress related to work events and functional seizures and symptoms Sheila

Putting a distance between herself and stressors Sheila

A. TRYING TO PUT TOGETHER A MODEL TO UNDERSTAND SEIZURE EXPERIENCES Abigail

Physical effects of seizures last longer than the seizure itself Abigail

Emotional experiences can often precipitate seizures Abigail

A. DEVELOPING A MODEL TO MAKE SENSE OF SEIZURES Bryony

It feels absurd to be caught in a cycle of worry because it increases feelings of stress Bryony

Not knowing if she has missed information makes her feel anxious about some situations that require interactions with others Bryony

Learning that stress and seizures may be linked is helpful towards understanding FS Bryony

A. HAS TRIED HARD TO UNDERSTAND WHY SEIZURES OCCUR, BUT CANNOT DETECT A PATTERN Parveen

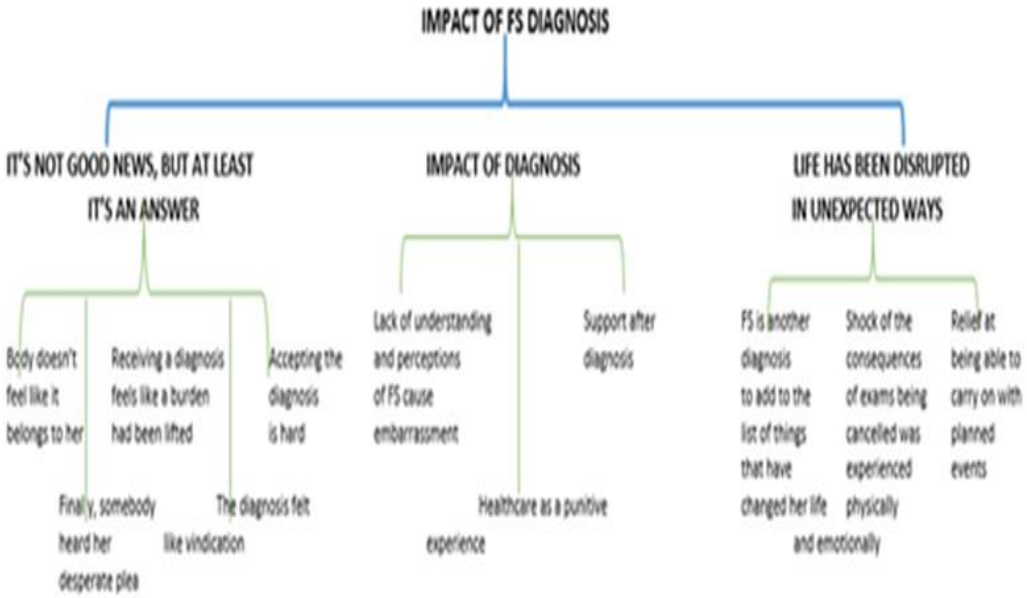
Having a seizure can be an experience that induces anxiety Parveen

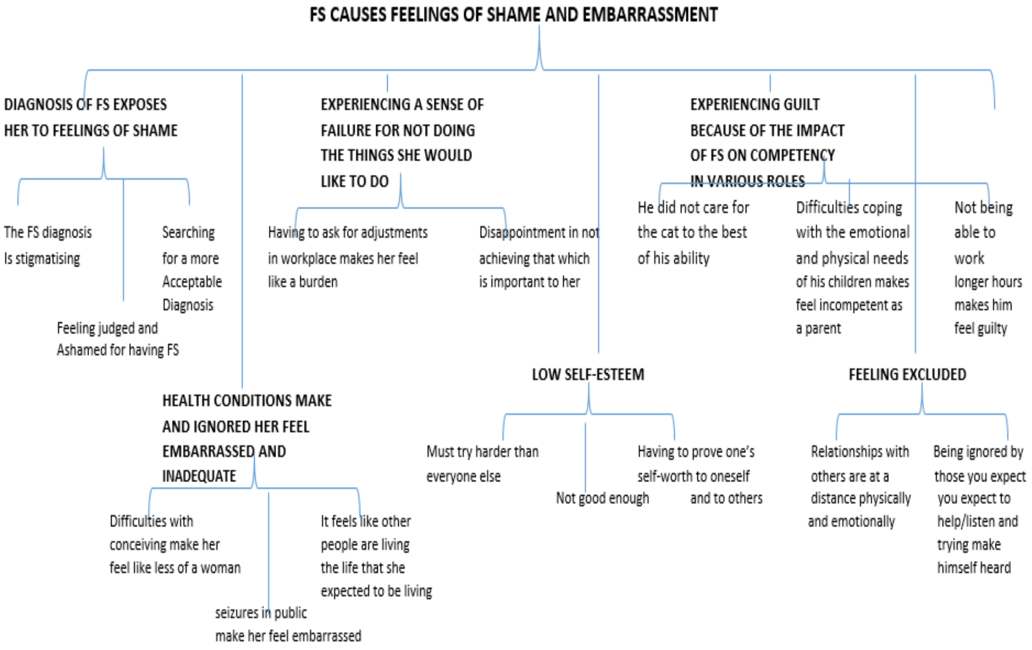
Different types of seizures are characterised by different features Parveen

Seizures don't relate to emotions; they can occur at any time Parveen

Spending time in the remembrance of God brings blessings and healing Parveen

Figure 4.6 Theme trees illustrating the GETs and minor themes. Clustering under the minor themes illustrate the PETs used from participants to develop the themes.





APPENDIX S

Reflexive statement

My approach to this study was one of curiosity, to learn more about individual’s experiences. I wanted to offer individuals the opportunity to describe their experiences in their own words. To give the NES literature another dimension, a richness based on lived experiences. I felt a strong sense of responsibility, which grew as the study progressed. I felt this especially when participants shared experiences that demonstrated inequality based on their recollection of events. This sense of responsibility followed me into the analysis, because I did not want to lose the participant's voice, particularly being aware that some individuals had indicated they felt unheard. I did not want to replicate this with this research. Therefore, at times I struggled to move beyond the descriptive part of the analysis. This was commented on during the analysis meetings, but it was not until I reflected on this that I was able to understand the basis for this struggle that I was experiencing.

I tried to bracket my own biases and to try and focus on what was being said, or not, by the participants. However, my own experiences were at times helpful when interpreting what participants expressed, and at that time I felt my personal understanding of similar experiences provided me with a path to look beyond the superficial meanings in the data. For example, being aware of the judgements sometimes made about individuals who do not speak English. It was important for me to have this participant’s voice in this thesis because she perhaps represents those women who are overlooked for being uneducated and therefore judged as lacking intelligence.

I also reflected on the ethical aspects of working with a small sample of participants. It is a small community and I attempted to anonymise the data. This was also a concern for the one South Asian participant in my sample. I faced the dilemma of whether I should remove details of her ethnicity and use an English pseudonym for her, but decided against this because her ethnicity was important and relevant to her experiences.

APPENDIX T

Table 14. Seizures and symptoms described by participants.

	PARTICIPANT									
	1	2	3	4	5	6	7	8	9	10
COMMON FND SYMPTOMS										
Seizures	X	X	X	X	X	X	X	X	X	X
Drop attacks					X	X				
Absences		X				X		X	X	
Paralysis		X	X	X	X	X				
Dissociation	X		X			X	X	X	X	X
Involuntary movements	X	X	X	X	X	X	X	X	X	X
Fatigue		X	X	X	X	X	X	X	X	X
Chronic pain			X			X	X	X	X	X
Mobility problems				X			X	X	X	X
Speech problems	X		X	X		X				X
COGNITIVE DEFICITS										
Memory	X					X				X
Attention	X									X
MOOD DISORDER										
Anxiety		X	X	X		X	X	X	X	X
Depression	X	X	X					X		X
HELP WITH SELF-CARE		X						X		X
CO-MORBID CONDITIONS										
Asthma					X		X		X	
Borderline personality disorder							X			
Chronic fatigue				X						
Eating disorder						X				
Ehlers Danlos Syndrome									X	
Fibromyalgia							X	X	X	
Postural Tachycardia Syndrome									X	
Sleep apnoea				X	X					

APPENDIX U

Table 18. Assessment of risk of bias in recent studies with FND populations

Study	Selection bias	Study design	Confounders	Data collection methods	Withdrawals	Overall
Demartini et al (2019)	*Moderate* Outpatient clinic/ Hospital staff and family	*Moderate* Cross-sectional	*Moderate* Participants matched on age and sex	*Moderate*	*Good*	**Moderate**
Jungilligens et al (2020)	*Moderate* Inpatients/via adverts	*Moderate* Cross-sectional	*Moderate* Controlled for BMI and BP	*Good*	*Poor*	**Moderate**
Koreki et al (2020)	*Moderate* Outpatient clinic/adverts	*Moderate* Cross-sectional	*Moderate* Participants matched on age and gender	*Good*	*Moderate*	**Moderate**
Pick et al (2020)	*Poor* Adverts	*Moderate* Cross-sectional	*Moderate* Participants matched on IQ, gender, handedness, knowledge of heartrate	*Good*	*Good*	**Moderate**
Ricciardi et al (2021)	*Poor* Not described/visitors and hospital staff	*Moderate* Cross-sectional	*Moderate* Matched participants on age and gender	*Poor*	*N/A*	**Weak**
Williams et al (2021)	*Moderate* Clinic/adverts/university mailing list	*Moderate* Cross-sectional	*Moderate* Controlled for age and depression	*Good*	*N/A*	**Strong**

Overall rating:

STRONG = no ‘weak’ ratings; MODERATE = one ‘weak’ rating; WEAK = two or more ‘weak’ ratings.