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Doctorate in Clinical Psychology Thesis

Exploring the experience of stigma in functional neurological disorder and mindfulness for
functional seizures

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Thesis Abstract

Overview: This thesis explored the concept of stigma as experienced by people with functional neurological disorder (FND) and assessed a mindfulness-based intervention for people experiencing functional seizures (FS).

Systematic Literature Review: The systematic literature review used a qualitative meta-synthesis approach to explore the experience of stigma amongst people experiencing FND. The review identified four themes: delegitimization; excluded, isolated and abandoned – the social cost of stigma; the cost of attempts to manage stigma and; threats to identity and the meaning of mental health. Stigma appears to be a central experience for those with an FND diagnosis.

Empirical Paper: The empirical paper comprised of a single case experimental design (SCED) which assessed the effect of a mindfulness training intervention delivered through a smartphone app for people experiencing FS. Four participants completed the study and a fifth completed it partially. Results found that two participants showed reliable and clinically significant changes on the outcome measure of quality of life and distress as well as process measures of experiential avoidance and mindfulness. However, clinically significant outcomes were not observed for the remainder. The implications of these results are discussed.

Critical Appraisal: This paper shared reflections on the process of this research and provided additional background to some of the key decisions taken over the course of the study while offering a critical analysis of the project.

Declaration

This thesis was undertaken for the Doctorate in Clinical Psychology at Lancaster University, within the Division of Health Research. The work presented here is the author's own, except where due reference is made. The work has not been submitted for the award of a degree elsewhere. The author has no competing interests to report.

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Section 1 Systematic Literature Review

A meta-ethnographic synthesis of the experiences of stigma amongst people with FND

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Abstract

This meta-ethnography explored the experience of stigma amongst people with a diagnosis of functional neurological disorder (FND). Whilst stigma is regularly reported by this group in qualitative research, there are a limited number of studies that focus explicitly on this experience. The aim of this review was to develop a further understanding of how people with FND experience stigma. Five databases were searched (PsycINFO, Web of Science, CINAHL, MEDLINE and EMBASE) and following a screening process and a critical appraisal using the CASP tool, 13 papers were included in the final synthesis. Four major themes emerged; delegitimization; excluded, isolated and abandoned – the social cost of stigma; the cost of attempts to manage stigma and; threats to identity and the meaning of mental health. The results identified negative, stigmatizing attitudes towards people experiencing FND symptoms in a variety of contexts including healthcare and other social institutions. Additionally, the effects of stigma led to further exclusion for participants and appeared to trigger coping styles that led to additional difficulty. This review identifies stigma as a key part of the illness experience of FND and highlights the need for stigma towards this group to be addressed.

Keywords; Functional neurological disorder, stigma, meta-ethnography, functional movement disorder, functional seizures,

1. Introduction

1.1 Functional neurological disorder

Functional neurological disorder (FND) describes the experience of neurological symptoms which cannot be completely explained by existing understandings of neurological pathology. The symptoms of FND can resemble neurological conditions such as epilepsy, stroke or Parkinson's. However, FND doesn't present with similarly identifiable biomarkers as these organic illnesses. FND has an estimated incidence rate of 4-12 per 100,000 per annum worldwide [1] and a large-scale study in Scotland found that FND was the second most frequent complaint in first-time presentations to outpatient neurology after headache [2].

The symptoms of FND are wide ranging and can include seizures, tremor and spasm among others [3]. Research on the prognosis for people with FND has shown that for many people these difficulties are enduring, with follow up studies highlighting that a large proportion of people diagnosed with this condition continue to experience difficulty, or indeed an increase in the severity of the condition over time [4].

In addition to the primary symptoms of FND, people with this diagnosis experience a range of wider difficulties including in their family, work and social lives as a result of their condition [5]. Furthermore, many people with the condition experience mental health difficulties such as anxiety and depression [1]; with these being more common than for individuals with a corresponding organic diagnosis [6]. For many with the condition, the combination of FND and associated difficulties cause a degree of disability and impact on participation in everyday activities. For example, a case-control study reported a significant

drop in employment levels in a sample of 322 people with an FND diagnosis, falling from 87.5% before experiencing symptoms to 24.5% post diagnosis [7].

1.2 History & current understandings of FND

FND has a long history, throughout which understandings of the condition have been in a state of flux. Early descriptions date back to ancient Greece [8]. At this time, symptoms associated with this condition were labelled as “hysteria” which at the time was perceived to be caused by a range of abnormalities of the womb such as “wandering of the womb” or “suffocation of the womb” [8]. The middle ages led to associations between FND symptoms and witchcraft resulting in severe consequences for individuals with the condition [8].

Subsequently, understandings of FND symptoms moved away from earlier supernatural and womb-based explanations to attribute a greater role to the brain and the mind in their origin. Charcot proposed a variety of understandings of FND including the role of “dynamic lesions” on the brain [9], a hereditary component [10] and later noted psychological aspects of the condition such as a correlation with past trauma [9]. Freud was greatly influenced by Charcot and went on to develop psychoanalysis positing that FND symptoms were a physical representation of unconscious struggle, coining the term “conversion” to describe this [9].

To this date, no universally accepted explanation of FND is available. More recent explanations of FND have built on the understandings that the brain and the mind play a key role in their development and maintenance. Neurobiological investigations have found that while no specific brain lesion or abnormality can explain FND, differences in activation and connectivity in specific brain areas have been observed [11]. For example, hypoactivation has been observed in the motor cortex of those with functional tremor [12] and strong functional

connectivity has been observed between areas associated with emotion, movement and executive function in those experiencing functional seizures [13].

Psychological explanations build on those proposed by Charcot and Freud. Modern psychodynamic explanations posit the role of FND symptoms in the unconscious management of feelings, interactions or memories that are unbearable in some way [14]. However, this explanation is contested due to the lack of a clear traumatic event in many cases of FND [15]. Further psychological explanations focus on links between the attentional system, illness beliefs and FND symptoms [16]. Beyond this, cognitive explanations highlight schemas or frames of understanding that underpin FND and result in symptoms when activated [17].

Biopsychosocial explanations of illness seek to integrate these various understandings into a meaningful whole, integrating the impact of each factor upon the development and maintenance of the condition. Models encompassing cognitive, attentional and affective processes alongside social and biological vulnerabilities [18] facilitate the integration of different understandings of FND and allow for the heterogeneity that exists amongst those who develop these symptoms.

1.3 Stigma

Due to the lack of specific biomarkers of disease, stigma is a common experience for those with FND [19]. The term stigma originated in ancient Greece where it was used to describe a physical trait or characteristic that denoted an individual as somehow tainted, immoral or

someone who should be avoided [20]. Current understandings of stigma have been heavily influenced by Goffmann [21] who defined stigma as a social construct as a devaluation of social identity which prevents individuals from attaining full social acceptance. Link & Phelan [22] further emphasised the impact that stigma has in highlighting difference between people or groups, developing a “them” and “us” narrative which ultimately leads to discrimination, power imbalance and loss of status for those affected.

There is growing interest in stigma in the field of health research. Stigma has an impact on health outcomes for those affected. For example, access to life chances such as employment and effective treatment are limited to those affected by stigma and the stress caused by the experience of stigma can exacerbate existing health difficulties [22].

Highlighting the extent of stigma for people with this condition, FNDHope, an international patient-led charity providing support and advocacy for people with FND, conducted a survey of people with FND in 2020 [23]. This survey found that 81% of participants reported being treated poorly due to stigma related to their diagnosis. A large proportion (61%) of the sample also reported that they experienced trauma as a result of their illness journey. Further questions showed that a large number of respondents (61.8%) believe that having an FND diagnosis had affected the healthcare provided in the past and more (64%) were concerned about how it would affect future healthcare. The results of this survey are reflected in research on stigma in one particular FND, functional seizures (FS), where an exploratory study found that 82.7% of a UK based sample of 47 individuals with a diagnosis of FS experienced perceived stigma [6]. The same study compared those with the FND diagnosis of FS to the corresponding organic condition of epilepsy finding that those with the FND

diagnosis were 42% more likely to experience perceived stigma. Furthermore, quantitative research has highlighted the role that stigma plays in exacerbating difficulties with quality of life in the domains of seizure worry, emotional wellbeing and social functioning for those experiencing FND symptoms [24].

Considering the experience of healthcare professionals, research has highlighted stigmatizing beliefs towards people experiencing FND [25]. Clinicians have expressed stigmatizing beliefs including the idea that those with FND symptoms are “faking” their illness in order to benefit from secondary gain [26]. A synthesis of studies on the views of healthcare providers working with FND highlights a vicious circle of stigma [25]. In this explanation, the review identified struggles that healthcare professionals experience in managing the complexity of the presentation of people with FND leading to them “passing the buck” [25 p. 7] on to another discipline without providing adequate support.

As well as the quantitative research into stigma in FND, stigma has been reported in a variety of qualitative studies on FND (e.g. [27, 28]) which describes some of the experiences of stigma encountered by people with FND. Qualitative research in health serves an incredibly important function of identifying and giving a platform to the voice of individual patients and professionals operating within this system. This research methodology often amplifies the voices of both those delivering services and those receiving services in a healthcare environment. In doing so, this type of research has a humanizing effect [29], giving service users an active voice in a sphere that affects them greatly. When considered relative to the negative and potentially dehumanizing effect of stigma, qualitative research itself may contribute to the opposition of stigma through this process of humanization. Furthermore,

reviews of qualitative research play an important role in healthcare and serve functions such as consolidating research on the lived experience of individuals and supporting practitioners [30] as well as influencing policy [31].

Consequently, the aim of the present review is to examine the unique experiences of stigma described by individuals in qualitative research to gain a greater understanding of the impact of stigma upon their experience which would inform the provision of better support. In doing so, the review proceeded with the broad research question; how do people with FND experience stigma?

2. Method

2.1 Database searches

In consultation with two subject librarians, five databases were chosen for the search: PsycINFO, Web of Science, CINAHL, MEDLINE and EMBASE. These databases were chosen for their focus on health sciences research, psychology, healthcare and medicine. The search strategy sought to identify all of the qualitative papers available on FND. In doing so, it adopted qualitative research and FND as two core concepts. Both free text terms and subject headings were utilised which incorporated a variety of terms that have been used to describe FND. The specific FND terms were informed by previous reviews in the area [5, 18, 32]. These terms were combined using the AND operator with a variety of subject headings and free text search terms to cover qualitative research methodologies. See Table 1 for a sample of the search run in PsycINFO. For the remaining databases, this was translated to use their specific subject headings and syntax (see appendix 1 - E for a copy of the full search strategy).

Searches were conducted in February 2021. A search strategy test was conducted whereby five known key papers relevant to the review were identified. All five papers were present in the search results indicating that the search strategy was effective in finding papers relevant to the review.

Table 1 PsycINFO search

	Search terms
	<p>FND terms (DE Somatization) OR (DE "Somatization Disorder") OR (DE "Somatoform Disorders") OR (DE "Conversion Disorder") OR (DE "Movement Disorders") OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) N3 dissociative) OR ((seizure* OR convulsion*) N3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) N3 seizure*) OR ((conversion OR psychogenic OR functional) N3 paralysis) OR ((conversion OR psychogenic OR functional) N3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional N3 paralysis"</p>
AND	<p>Qualitative research terms (DE "Qualitative Measures") OR (DE "Qualitative Methods") OR (DE "Focus Group") OR (DE "Focus Group Interview") OR (DE "Interview Schedules") OR (DE Interviewers) OR (DE Interviewing) OR (DE Interviews) OR (DE "Interpretative Phenomenological Analysis") OR (DE "Mixed Methods Research") OR DE ("Grounded Theory") OR ((personal OR patient OR subjective OR lived OR subjective) N3 experience) OR interview OR qualitative OR "focus group" OR phenomenological OR "phenomenolog*" OR "conversation analysis" OR "thematic analysis" OR "discourse analysis" OR "grounded theory" OR "narrative analysis" OR "mixed method*"</p>

Note: DE is a psycINFO subject heading search term

2.2 Inclusion Criteria

Inclusion criteria for the review were: qualitative methodology used (defined as any method that involved participants' descriptions of their lived experience); focused on the perspective

of individuals with an FND diagnosis; peer-reviewed and available in English. Furthermore, papers must have reported a theme or similar portion of text relevant to stigma. For the purpose of this review, any theme which detailed participants being treated differently to others, facing discrimination, or being treated in a way which prevented them from attaining full social acceptance due to their illness was deemed relevant.

A total of 9,116 papers were identified from the databases searched. After duplicates were removed, 7,114 remained. The title and abstract of these papers were screened for relevance and 7,063 were removed at this stage. Following this, 51 full text papers were assessed for eligibility. Reference lists of these 51 papers were screened and one additional relevant paper was identified and assessed for eligibility. At this stage, 38 papers were excluded as they did not meet inclusion criteria. This left 13 papers remaining. See Fig. 1 for a PRISMA diagram of the screening process and Table 2 for the details of papers included in the review.

Fig. 1 – PRISMA flow diagram

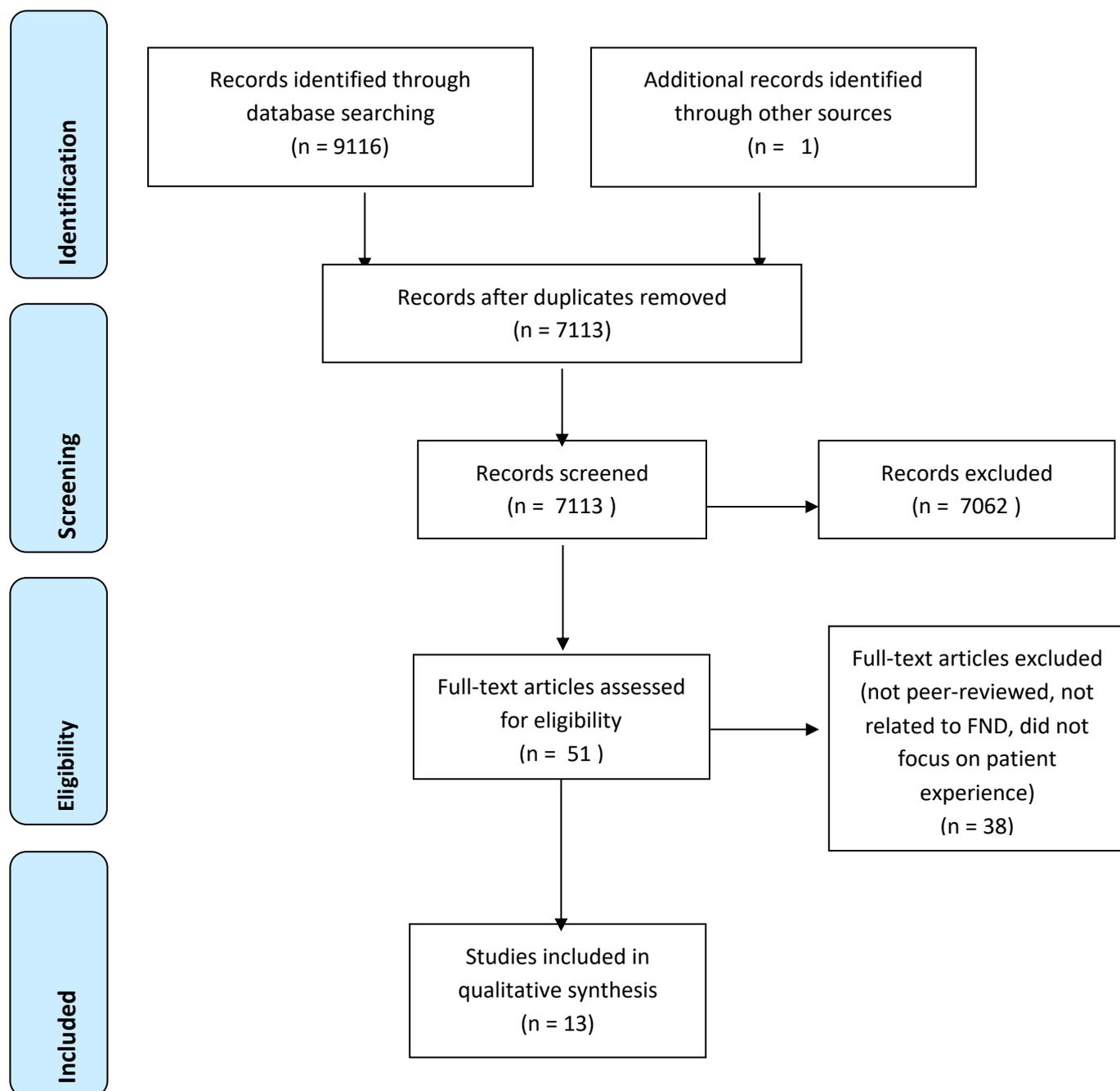


Table 2 - Papers Included in the review

Paper	Aim	Methodology	Participants	Data Collection	Major Findings	CASP
Dosanjh et. al. (2020) [28]	To gain insight into how individuals with FMD make sense of their experience from symptom onset through medical evaluation and diagnosis to post-diagnostic adaptation.	IPA	8 (FMD)	Semi-structured interview	Participants experiences of FMD had a significant impact on relationships with themselves and others. This in turn led to difficulties in well-being.	22
Karterud et. al. (2010) [33]	Explore the experience of participants who experience a change in diagnoses from epilepsy to FS.	Interpretative analysis based on systematic condensation	10 (FS)	In-depth interview	The manner in which diagnosis is communicated can have an impact on how people understand and adjust to it. More cooperation between services such as neurology any psychiatry would bridge people's needs and lead to less feelings of being abandoned.	20
Karterud et. al. (2015) [34]	Explored the impact of using a biopsychosocial approach to explain FS diagnosis	Interpretative analysis based on systematic condensation	11 (FS)	Semi-structured interview	Being believed was a key factor in coping with the condition.	22
Karterud e. al. (2016) [35]	Explored how legitimacy is managed by young people with an FS diagnosis	Thematic analysis	11 (FS – aged 14-24)	Interview and follow up	Discovered a relationship between the legitimacy of the illness that participants experienced and the extent to which they participated and recreated socially. Those with more meaningful illness perceptions engaged in more social participation.	22
McWilliams et. al. (2016) [36]	Characterised the experience of children and families with FS.	Thematic analysis	29 families 10 young people (FS)	Focus groups and telephone interviews	Young people and families with experience of FS experience significant impairment. Pathways to diagnosis are unclear. Found that framing the diagnosis of FS as “good news” was not experienced positively by children and families.	21

Paper	Aim	Methodology	Participants	Data Collection	Major Findings	CASP
Nielsen et. al. (2020) [37]	Explored the experience of people with FMD	Inductive thematic analysis	11 (FMD)	Semi-structured interviews	Participants described a lack of understanding in their interactions with healthcare professionals. They experienced significant emotional and physical burden through their condition. Psychological explanations offered by professionals were not satisfactory.	22
Pretorius and Sparrow (2015) [38]	Explored the experience of people in South Africa with an FS diagnosis	Thematic analysis	10 (FS)	Semi-structured interviews	Described the challenges faced by people with a FS diagnosis and identified some resources that can act as a support.	22
Rawlings et. al. (2017) [39]	Explored written accounts of people with an FS diagnosis	Thematic Analysis	19 (FS)	Written accounts – participants were asked to write; their thoughts and feelings about the condition, a letter to their condition, a letter to their younger selves and about their personal value	Findings highlighted the stigma experienced by people with a diagnosis of FS.	22
Rawlings et. al. (2018) [27]	Explored written accounts of people experiencing seizures	Narrative analysis	29 (epilepsy) 20 (FS)	Written accounts – participants were asked to write for 20 minutes detailing their deepest thoughts and feelings about their condition	Found a difference in narrative between those with epilepsy and those with FS. Noted that those with FS used a “feeling lost” typology when describing their illness.	22
Robson & Lian (2017) [40]	Explored how people with FS experience healthcare encounters.	Thematic analysis	135 (FS)	Free-text survey responses	Highlighted the negative experiences people with FS have in healthcare encounters. These were explored through a stigma framework	21
Sarudiansky et. al. (2017) [41]	Explored the thoughts of people with an FS diagnosis in Buenos Aires, Argentina	Thematic Analysis	5 (FS)	Semi-structured interviews	Found that these participants frequently adopted somatic frameworks to understand their diagnosis.	21

Paper	Aim	Methodology	Participants	Data Collection	Major Findings	CASP
Thompson et. al. (2009) [42]	Provided insight in to the experience of receiving a diagnosis of FS from a patient perspective	IPA	8 (FS)	Semi-structured interviews	The results indicated that some patients experience distress when the diagnosis is communicated. An ability to integrate the diagnosis into a personal narrative was seen as beneficial.	17
Wyatt et. al. (2014) [43]	Explored the experience of receiving a diagnosis of FS and engaging in therapy following this	Thematic analysis	6 (FS)	Semi-structured interviews	Highlighted the impact of receiving this diagnosis and how this can have a subsequent effect on how people engage with therapy.	18

Note: FMD – Functional Movement Disorder; FS – Functional Seizures, IPA – Interpretive Phenomenological Analysis; CASP – Critical Appraisal Skills Programme, qualitative research appraisal tool

2.3 Quality rating system

The quality rating system used in this review was the Critical Appraisal Skills Program (CASP) qualitative research checklist [44]. This tool comprises ten questions designed to assess the quality of qualitative research. The first two questions act as screening questions to determine whether proceeding with the appraisal is worthwhile. The remaining eight questions were used to rate the quality of papers used in the review. In doing so, the three-point rating system advocated by Duggleby et. al. [45] was utilized. This used the CASP questions and applied a score for each question. Scores ranged from a weak score (one point) for articles that did not explain or justify the issue being examined by the CASP question, a moderate score (two points) where the issue was mentioned but not expanded upon, or a strong score (three points) where the issue was both justified and sufficiently expanded upon. The scores from the eight items were added to give a total score for each article with a maximum possible score of 24. Using this system, the lead researcher and a peer reviewed the papers independently and reached a consensus on scores afterwards. Rating disagreements were discussed further in an effort to reach a consensus. Where this was not possible, the lead researcher had the final say on the score applied. Consequently, the papers achieved ratings that ranged from 16 to 22. While CASP scores were not used as inclusion or exclusion criteria, following the analysis the sources of each theme were checked to ensure that the higher quality papers were well-represented in the results and no theme depended solely on the weaker papers.

2.4 Meta-ethnography approach

The method adopted for this review was Noblit and Hare's [46] seven-step method of meta-ethnography. Stigma is a frequently recurring topic in the literature on functional seizures and

for many is a central part of their illness experience. While this has formed a part of many qualitative studies, this information has not yet been synthesized. A meta-ethnography approach allowed for this to be synthesised alongside the additional interpretations to provide a fuller understanding of this experience with a view to developing results that will inform practice [47]. This approach was suitable in investigating the conceptual and theoretical understandings of stigma towards this group, compared with other approaches to qualitative analysis that are more descriptive in nature [48]. The first two steps of “getting started” and “deciding what is relevant to the initial interest” are detailed in the introduction to the topic and the search strategy. Following this, the papers were read and coded line by line, notes were made of key concepts completing the “reading the studies” phase. Then, in the “determining how the studies are related” phase the codes were compared to identify relationships between the concepts covered in each study. After this, the “translating the studies into one another” phase involved translating the codes and themes together in order to develop an understanding of stigma. Following this, “synthesising translations” brought the translations together into themes that represented common translations and new interpretations. Finally, the synthesis was expressed through the writing of this paper. See appendix 1 - B for a demonstration of the process.

2.5 Reflexivity

Given the interpretative nature of this review, it should be recognised that researchers bring an a priori understanding of the topic at hand to the research process. Indeed, the researcher approached this meta synthesis as a trainee clinical psychologist having had direct encounters with people who have experience of FND in a research capacity. While this served as inspiration for the study, it also led to an expectation that stigma would be prevalent in the studies covered by the review. Furthermore, the researcher is a male from a white, Irish,

working class background. As a result, a professional, Western worldview may have influenced this synthesis. However, the potential impact of bias arising from this is somewhat mediated through the use of published research and systematic search methods. Furthermore, consulting with supervisors throughout the process of recognising and reporting the themes emerging from the review served as an opportunity to reflect on this and reduce any potential bias.

3. Results

The review incorporated 13 qualitative papers. These studies included a total of 264 participants with an FND diagnosis. Geographically, samples were recruited from the UK in 7 papers, Norway for 3 papers and one each from South Africa and Argentina. A further paper [40] recruited internationally with the majority of participants living in the UK or North America. As a result, there is bias in the sample included towards White, Western countries. In terms of gender, where reported there was a majority of over 80% female participants across all of the studies. This is consistent with the epidemiological profile of FND [7].

The review identified four themes through which the synthesis of these papers can be expressed. These were: Delegitimization; excluded, isolated and abandoned – the social cost of stigma; the cost of attempts to manage stigma; and threats to identity and the meaning of mental health. These themes will be described in detail.

3.1 Theme 1. Delegitimization

This theme reflects the experience of individuals across the studies whereby they were treated with less legitimacy than those with other illnesses and represents external stigma towards the individual. Delegitimization featured consistently in the accounts of participants across the papers in this review. In the descriptions provided by participants this experience was complex and wide ranging and included being dismissed by others, not being believed [27, 28, 33-43] or of being left with the impression that their illness is less valid than one with a clear organic cause [28, 33, 34, 36, 39, 40, 42]. This experience was felt across a range of contexts including medical settings [28, 38, 40, 42], at work [35] and in education [35, 36].

The felt experience of delegitimization for many came in the form of not being believed [27, 28, 33-43], for example: “I was pretty much told that my condition didn’t really exist and that I was just hysterical and an attention seeker” [40 p. 7]. The lack of belief in the experience of participants’ symptoms was frequently accompanied by a level of judgement “[they] told me I am going crazy and I have conscious control over everything and nothing is wrong I am just lazy and go to the hospital frequently because I have nothing better to do [40 p. 8].” For many participants the experience of not being believed was extended to situations where it was implied that they were feigning their symptoms [28, 33, 35, 39, 40, 43], encountering attitudes such as “either you are making it up or it isn’t a real condition [39 p. 88].”

Alongside disbelief, a degree of blame was present [28, 34, 37, 39-42]. It appeared that where the people and institutions in the lives of participants did not understand the condition, they blamed the person experiencing FND. At times it appeared that where a system could not

account for the experience of a person with FND, it led to that system rejecting the idea that the individual could possibly have that experience “Well none of it makes sense so you can’t be experiencing all these [28 p. 9].”

Interactions with healthcare services served as a location for many delegitimizing experiences [27, 28, 33-40, 42, 43]. Participants spoke about their journeys towards a diagnosis which often took a significant amount of time [27, 28, 37-40, 42, 43] and then even when a diagnosis was given participants felt that medical professionals saw their condition as less valid than one with an identifiable organic cause. For example, a participant with a diagnosis of FS commented:

One neurologist treated me like I wasn’t important because I didn’t really have epilepsy. He just wanted to push me onto another neurologist to get me out of his office. He didn’t really seemed to care about my feelings or what I had to say. I felt very ashamed walking out of his office, because I wasn’t a real epilepsy patient [40 p. 9].”

Experiences of delegitimization were also experienced beyond healthcare and present throughout the lives of the participants. Individuals described similar attitudes in educational settings; “[a teacher] said that I played her just to get out of having a test and I faked having a fit [36 p. 132].” Further examples of this were found in the workplace [28, 35].

On the other hand, there were a minority of occasions where individuals had a more positive experience [28, 34, 37, 38, 42, 43]. Where a sense of legitimacy was present, this changed the nature of interactions for the better. For instance, participants feeling believed and accepted by the professionals that they were encountering had a range of positive implications such as: “I’m thinking, ‘yes! yes! somebody believes me.’ It just made me feel . . . a genuine person

[42 p. 511].” These examples appeared to be rare moments of legitimacy and understanding in a process where the predominant experience was one of being dismissed and delegitimized. For many participants it appeared that the experience of legitimacy was more frequent in closer personal relationships than in contact with professionals and institutions “At school and in working life, the participants said that they felt more respected and better understood by peers and friends than by teachers and employers [35 p. 25].”

3.2 Theme 2. Excluded, isolated and abandoned – the social cost of stigma

This theme relates to the social cost of stigma for participants. Whereas the previous theme highlighted the experience of being stigmatised by professionals, by having an illness that was appraised as invalid, this theme highlights the impact of this external stigma on the public lives of those affected. It describes the ways in which social forces act upon them in a stigmatizing fashion. Within this theme, participants gave descriptions of being abandoned, isolated and pushed to one side by friends and wider society.

A loss of status or damage to their social role was a consequence of stigma in FND [28, 33, 35, 36, 38, 40]. Participants spoke about losing their independence, friends and education [36]. While some participants spoke of the isolation experienced as they missed out on social relationships [42], a rejection from major social institutions was central to this experience. Other participants described a sense of subtle exclusion from normal activity whereby they were cushioned from harm to an extent but ultimately treated differently from their peers; “At school they still treat me like a little kid as well. They say you can't do this, you've got to be careful because you'll over exert yourself [36 p. 130].”

For many, the social cost of stigma came in the form of being abandoned by healthcare services [27, 28, 33-40, 42, 43] “I was discharged again without any explanation and just left [42 p. 511].” This left individuals feeling frustrated with services “refer me on! Do something. Don’t just allow me to stay at home and do nothing [37 p. 2046].” These difficulties in finding appropriate care were frequent with participants feeling that they were alone in managing the condition, that support was not available to them [27]. This feeling was very much present in the journey towards diagnosis that participants described as long and arduous [27, 28, 37-40, 42, 43]. It appeared that a lack of understanding on the part of healthcare played a role in participants’ experience here [27, 28, 33-39, 41-43]. Furthermore, a secondary cost of stigma was identified in that healthcare services often paid less attention to other non FND health concerns raised by participants meaning that participants’ other health concerns were ignored:

“My GP blamed my ear and sinus pain on my condition, even though after prompting her to look in my ears she found that I had a sinus and ear infection [...] Even when visiting my dentist with tooth pain I was asked whether it could be due to clenching my jaw together when I am anxious! [40 p. 7]”

Other participants experienced direct physical harm as a result of their FND diagnosis [38, 40]. Participants spoke of dangerous situations where healthcare workers sought to prove that the condition was being feigned in some way:

The nurse [...] put me in a wheelchair with force and started shouting at me and pushing my shoulder and head back into the chair. I was very woozy and didn’t understand what was happening. My body started to shake, my eyes were open so I was clearly awake, the nurse went to do the sternum rub and instead punched my collar bone and started to rub her knuckles hard on that, she then pushed the wheelchair back into the wall and my head hit. She threatened to call the mental health down and said that ‘she couldn’t watch me all night have a fit’ and that I was taking up everyone’s time and I was wasting the NHS resources and money [40 p. 9].

Such instances indicate a clear breach of ethical practice and display how an experience of social stigma within an institution can manifest in direct physical harm. Indeed, situations where medical professionals appeared to act in these ways seemed to be underpinned by a lack of understanding. For example, when encountering a patient could not be understood, an assumption was made that the patient must be acting in a deceitful manner.

Where participants encountered social systems that were supportive and inclusive, this appeared to act in the opposite fashion, helping them to adjust to their symptoms and diagnosis [33-35, 40]. The support of friends and family counteracted some of the effects of stigma in some cases [38].

3.3 Theme 3. The cost of attempts to manage stigma

This theme related to the personal costs of stigma and represents individuals' efforts to cope with external stigma. Often, participants were presented with a position where they had to manage stigma in addition to their illness. In these situations, participants seemed to respond by seeking to protect themselves against further harm. Where the theme above related to external social stigma, this theme captured some of the internal experiences triggered by stigma.

In order to manage the stigma that they encountered, some participants sought to carry on as normal, as if they did not have FND symptoms [28, 35]. In doing so, participants wore themselves out "I felt exhausted from trying to continue to be normal [28 p. 9]." Similarly, controlling the information available to others about their FND symptoms was a key strategy.

“I was furious if they (the teachers) said something while I was away... (...) ... I'll sue them for breach of confidentiality. I do not want to tell the teachers this ... (...) Really, I want to say that they diagnosed epilepsy. [35 p. 25]”

Telling people that they had epilepsy was a common strategy as this was more readily understood and felt less stigmatising for these participants [33, 35, 36]. This approach links with the idea in theme 1 whereby medical professionals and perhaps wider society have more familiarity with epilepsy as an organic disease and treat it in a less stigmatizing manner.

Many participants resorted to self-isolation as a result of the stigma that they encountered [28, 35, 37, 38, 40, 42]. It appeared that individuals expected a degree of stigma in daily life and as a result avoided normal activity “I went out less. . . I just didn't want to do anything that was going to embarrass me [28 p. 9].” In doing so, participants described withdrawing from social relationships [28] and work [35]. This avoidance of public situations extended to healthcare for fear of further adverse experiences and judgement;

“I felt deeply misunderstood and offended and it has affected me hugely [...] I now have difficulty trusting healthcare professionals [...] I fear hospitals, almost to a phobic extent [...] It has affected me massively [...] when you don't trust that you'll be treated appropriately by others when completely unable to explain or defend yourself, it's terrifying [...] I don't think health professionals realise the potential consequences of their actions [40 p. 10].”

Through this strategy, individuals often appeared to experience compounded difficulties as they did not receive appropriate healthcare as they did not seek it. Furthermore, this approach resulted in participants experiencing further exclusion “you hear less and less from people [37 p. 2045]” and it reduced the potential for participants to experience positive validation [35].

Underlying some of these actions were a variety of fears such as a fear of being judged [27, 28, 33-36, 39-43] “if someone saw me what would they be thinking [28 p. 9]” and participants spoke widely of feeling that others were judging them in some way “I worry that they will think lots of negative things. They’ll think I’m a complete lunatic [34 p. 25].” It appeared that for some, the experience of stigma led to expectation of further judgement from others in many participants “They surely think that I am faking the seizures now as there is no organic reason for them [33 p. 42].”

Again, participants described less frequent moments where they were treated with a degree of legitimacy and respect [28, 34, 36, 38, 42, 43], and this led them to feel that they could participate as normal in society.

“I know I have seizures, and I know that I have problems. It has taken me a long time ... But I’m happy that I have reached where I am today. When I get seizures in public.... I care... not. What happens, happens [35 p. 26].”

Where this was the case, individuals appeared to arrive at this opinion after a significant period of adjustment [34 p. 42].

3.4 Theme 4. Threats to identity and the meaning of mental health

This theme describes interactions that participants found particularly stigmatizing, where they were confronted with opinions and explanations for their difficulties that contrasted greatly with their view of themselves and the world. This theme represented a resistance to internalising stigma where it clashed with their core beliefs about themselves and others.

Many of the stigmatizing responses towards people experiencing FND appeared to cause particular discomfort and pain where they led to a perceived threat to the identity of the individual. For instance, where they were not believed as described above, participants highlighted the feeling that they had been judged as somebody who would set out to deceive others [27, 28, 36, 37, 40-42]. This was often accompanied by some implied judgement that they were in control of their symptoms or chose to have them. In turn, this led participants to feel that their identity was under attack e.g. “I do not want people to think I’m a bad person because I suffer from seizures [34 p. 110].”

A significant threat to participants’ identity presented through the internalisation of blame [27, 28, 33-40, 42, 43]: “loser, just pull yourself together. [39 p. 87]” Participants described themselves as feeling “weak”, “useless”, “pathetic” and a “waste of space and money [39 p. 87].” While the blame identified through theme 1 above was external and directed towards individuals, here the blame was internalised with participants blaming themselves for their difficulties and the impact that they have had on others “I feel guilty, I feel like, why do I have this? I made everyone feel angry and upset and I’m making the family fall apart [36 p. 130].” Statements such as this appeared to show that participants felt a degree of self-blame for stress in their family caused by their symptoms and stigmatizing understandings about the degree of control that they held over them. Additionally, the experience of external stigma for some participants led to an internalisation of stigmatising ideas that their symptoms may not be real “Am I actually getting these symptoms or is it all in the head? [28 p. 9].” This sense of internalised blame for their difficulties often led to intense emotional experiences such as anger “I’d get right frustrated, start crying. . . chuck things and get right angry with meself. [28 p. 8].”

Across the studies, psychological explanations of FND appeared closely linked with notions of identity for individuals in the studies. For some, psychological explanations added to the sense of delegitimization discussed above (theme 1) “Because that’s what it feels like, psychological feels like it should mean, it’s literally you are making it up. It’s all in your head, there’s nothing wrong with you at all [36 p. 2046].” However, for many the description of experiencing a mental illness presented as a greater threat to their identity [28, 33-35, 37-40, 43]. For example, a key concern for many individuals covered by the studies in this review related to being diagnosed with a mental health issue [28, 33-35, 37-40, 43]. Some participants completely rejected the idea of experiencing a mental health difficulty, seeing it as abnormal in some way, and expressed that such an explanation was a challenge to how they viewed themselves “I am normal and don’t have any mental health issues [39 p. 88].” Participants felt misunderstood when referred to psychology or psychiatry for support “I can’t see how talking to somebody is going to fix it [42 p. 510]”

Some of the negative perspectives towards psychological explanations for FND came through healthcare interactions whereby they had been treated well until they had been diagnosed with a functional disorder implying psychological cause.

Negative attitudes of HCPs towards what they perceived as psychogenic problems (i.e. having a psychological basis), may have played some role in the participants’ dissatisfaction with receiving psychological explanations for their problem. It was common for participants to describe experiences of poor treatment and negative interactions with HCPs only after a psychogenic diagnosis was made [37 p. 2046].

This response to psychological explanations of their experience was not universal to participants across the studies. Contrasting with the threats to identity that some explanations resulted in, some participants highlighted the benefits of viewing their condition from this perspective [28, 34, 37, 41, 42]. For example, providing psychological explanations that recognised the interactions between life events and physical responses such as stress and the subsequent impact on FND symptoms appeared more acceptable to some with FND e.g.:

It made a lot more sense when they explained what it was about in a little more depth. That it's all about connecting links that trigger a seizure and that it comes from feelings and thoughts that you have. For example, if you are afraid . . . if you think too much that you're afraid, then a seizure happens. So in that way you get a little more understanding of why seizures occur and what they really are. In comparison with just hearing that you have psychogenic non-epileptic seizures . . . so it was very useful (. . .) It is more about that it is stress reactions that trigger a seizure from the brain [34 p. 110].

Indeed, explanations that accounted for the individual experience and identity of the person experiencing FND appeared to be more readily acceptable. It appeared that these explanations recognised the complexity that the participants faced and sought to understand that as opposed to informing them that they are experiencing a psychological difficulty that they somehow should have control over [28, 34, 37, 41, 42].

4. Discussion

This systematic review provides the first synthesis of studies exploring the experience of stigma for people with FND. In doing so, it adds to the growing body of literature that overtly recognises stigma towards people with FND [6, 24]. For many people who receive a diagnosis of FND, stigma is a central part of their experience. The results of this review identify that this is experienced in all aspects of their lives from their own psychological experience to their interactions with wider society [19].

Returning to Goffman's [21] definition of stigma concerning the devaluation of social identity, the descriptions here show that people with an FND diagnosis experience this on a large scale. The theme of delegitimization described here represents a sense of being treated as "lesser than" or "less valid" than other people. Through this, a sense of "othering" creates distance and presents those with FND with a somehow less valid social identity, the effects of which are clearly visible in themes 2, 3 and 4 of this review. On top of this social experience, individuals described the phenomenon of internalised stigma whereby they have taken this message delivered by society and applied it to themselves, visible in themes 3 and 4. Indeed, participants identify clearly a sense of enacted or external (themes 1 and 2) and felt or internal (theme 3 and 4) stigma identified by Scambler and Hopkins [49]. Research into mental health stigma [50] has highlighted the additional impact of internalised stigma on symptom severity and treatment adherence, signalling the potential additional difficulty that this causes for those with FND symptoms. Additionally, a central tenet of the theory of stigma centres around a power imbalance. Indeed, Link and Phelan's [22] theory on stigma in healthcare notes a power imbalance as a pillar without which stigma cannot operate. As can be seen in the experience of these participants, particularly in themes 1 and 2, stigma has arisen in traditional institutions where a power imbalance often exists – school, work and healthcare.

Furthermore, the results of this review show how participants experience compounding difficulties as a result of the stigma they face. Link and Phelan [22] highlight how stigmatising experiences such as those described by participants are likely to affect their life chances through increased barriers to accessing appropriate education, employment and

healthcare. Indeed, difficulty accessing these have been noted in the FND population [7] and are described by the participants in this review in individuals' difficulties maintaining their pre-illness life in the face of stigma around their condition.

Additionally, the notion of stress in stigma creates a further challenge for individuals affected. As has been observed in theme 3, naturally, people make efforts to manage stigma such as carrying on regardless of the difficulties that they face or carefully managing the perceptions of others. This additional stress may act as an exacerbating factor making the original difficulties faced worse [22], another mechanism through which stigma exacerbates the difficulties faced by those experiencing FND. Indeed, this effect of stigma exacerbating psychological distress and causing additional damage through the erosion of social supports has been noted in the literature [51].

Considering the delegitimization that participants described in this review, a key factor underpinning this experience appears to be the view that FND is less valid due to the lack of established biomarkers for its diagnosis. The idea that healthcare has a positive bias towards conditions that can be observed and counted has been commented on in healthcare literature [52] identifying neoliberal ideals in health. This perspective encourages values in healthcare and society that promote an attitude of personal responsibility. This viewpoint disregards difficulties that cannot be clearly observed as a "disease of the will" [53] creating a belief that those who suffer from them have a degree of control over them. This preference for the physical and observable can be seen on a wider scale in the funding of services. For example, mental health services faced more cuts during the austerity era in the UK than their physical counterparts [54]. Indeed, the feelings of being abandoned expressed by individuals in this

review may relate to this lack of recognition of and subsequent provision for illness that is not socially accepted. Examples of this in this review were noted in the change in healthcare professional's attitudes towards patients when they were deemed to be experiencing a mental health difficulty and in the views of participants not wishing to be viewed as being part of this stigmatized group.

The above point links the results of this study with the experience of stigma faced more widely by people experiencing medically unexplained symptoms (MUS). Whilst MUS are more common than FND, a similar stigma towards people experiencing them exists due to the lack of a medical explanation [55]. Similar to FND, those with MUS have reported greater levels of perceived stigma compared to those with the corresponding medically explained diagnosis [56] which has been shown to have a negative impact on patient outcomes [57]. However, the experience of FND is distinct from MUS in the visibility of symptoms to others, which can attract a greater degree of external stigma.

4.1 Clinical Implications

This review identified stigmatizing interactions with healthcare professionals led people with FND to limit their contact with services. It has been well-documented that people experiencing FND often do not attend follow-up appointments, particularly with psychiatric services [56]. In these cases, addressing stigma would likely improve trust between services and those with FND and increase attendance at treatment. The results in this study compare with a review of practitioner experiences [25]. In the present review, participants reported feeling frequently misunderstood or dismissed leading to stigmatizing experiences.

Interestingly, when practitioners spoke of their experiences, they reflected similar themes,

reporting feelings of being out of their depth, not understanding the presentation of FND [25]. The review of healthcare professionals identified the idea of a vicious cycle whereby individuals diagnosed with FND were passed from one service to the next without receiving an appropriate service. This closely matches participants' descriptions of being abandoned by healthcare services. Thus, it may be that this lack of understanding or ability to provide support for FND patients presents practitioners with a degree of symbolic threat which leads to a stigmatizing response. Indeed, threat has been identified as a key process in the development of stigmatizing beliefs [57]. Therefore, increasing awareness and knowledge of FND is crucial in combatting stigma. Seeking to achieve this, recent research [58] has identified positive changes to practitioners' views of FND following a training programme. Evidently, further training and education for healthcare practitioners is required in order to reduce stigma towards FND.

Furthermore, for some people with an FND diagnosis, a coping style characterised by avoidance is core to their experience [32, 59]. Therefore, for this group encountering the attitudes outlined in themes 1 and 2 may compound this further, leading to the self-isolation and avoidance of services seen in theme 3. Due to the potential for an interaction between stigma and such coping styles to compound the difficulties of those experiencing FND, there is a clear clinical imperative to develop an early understanding of patient difficulties to support their adjustment and to prevent future disengagement due to stigma.

The findings in this study are consistent with those frequently reported in studies on perceptions of people who experience mental health difficulties [60]. Participants in this review rejected the idea of experiencing a mental health difficulty, perhaps recognising the

stigma towards mental health difficulties that exist in society and do not wish to join this stigmatized group in the explanation of their own difficulties. However, it has widely been acknowledged that psychological factors play an important role in the development, maintenance and indeed treatment of FND for many [17]. Therefore, this raises the question: how can people with an FND diagnosis access explanations for their condition that incorporate important psychological factors without the associated stigma and threats to their identity? Some hints are to be found in the responses from participants in this review who identify the utility of explanations of the condition that connect with their unique experience and incorporate biological, psychological and social factors as has been advocated elsewhere [18]. Further research [61] has identified that the manner in which the diagnosis is explored can have an impact on how people adjust to the condition which in turn affects their prognosis. While there is evidence that psychological factors play a central role in understanding FND [17], this review highlights the need to provide psychologically informed explanations in a way that does not result in further stigma.

4.2 Limitations

A potential limitation of the literature which makes up this review is publication bias. As this review focused solely on peer-reviewed literature, it is possible that it failed to identify important concepts that have not been published. Furthermore, as noted in the results section, of the studies published on this topic, a large majority came from Western countries. Thus, the review was limited to the experience in these countries.

In addition, all of the research that makes up the review has been led by clinicians and academics working in the area of FND research or clinical practice, as has this review. By its

very nature, qualitative research involves a level of active participation and interpretation on the researchers' part. As a result, despite their focus on the experience of individuals with FND, in the process of research, this has been filtered through the perspectives of researchers and clinicians who form part of the institutions through which stigma towards this population has been perpetuated. In future research it may be beneficial to seek greater involvement from those affected by the issues covered in this review in the process of design, and analysis of research in this area.

A further limitation of the review process was the use of a sole researcher for much of the project. However, the review did make use of a peer in the quality rating of papers. Additionally, the themes developed were discussed in detail in supervision throughout the study.

4.3 Future research

The presence of stigma throughout the studies reviewed here shows that stigma is a central concern for people with a diagnosis of FND. While research is currently focused on developing further understanding of the neurobiological and psychological features of the difficulty [17, 18, 62], this review highlights a key social aspect of the condition. While there is no specific evidence that stigma plays a causal role in FND, through its actions on those with the condition and the efforts made to cope with the associated judgement and distress, stigma may form a key part of understanding the condition. Despite the descriptions of stigma in the papers in this review, only one had an explicit focus on stigma. Therefore, further qualitative research into the stigma experienced by this group is warranted.

Furthermore, given the impact that stigma has on the lives of individuals affected by it, further research is required in this field to address stigma towards people with FND. Research investigating the efficacy of initiatives designed to reduce stigma would be useful to tackle this problem in healthcare settings and on a wider social scale.

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Appendix 1 – A - Table of CASP scores

Paper	Research Design	Sampling	Data Collection	Reflexivity	Ethical Issues	Data Analysis	Findings	Value of Research	Total Score
Dosanjh et. al. (2020) [28]	3	3	3	2	3	3	3	2	22
Karterud et. al. (2010) [33]	2	3	3	1	2	3	3	2	20
Karterud et. al. (2015) [34]	3	3	3	1	3	3	3	3	22
Karterud e. al. (2016) [35]	2	3	3	2	3	3	3	3	22
McWilliams et. al. (2016) [36]	2	3	3	1	3	3	3	3	21
Nielsen et. al. (2020) [37]	2	3	3	2	3	3	3	2	21
Pretorius and Sparrow (2015) [38]	2	3	3	3	3	2	3	3	22
Rawlings et. al. (2017) [39]	3	3	3	2	2	3	3	3	22
Rawlings et. al. (2018) [27]	3	3	3	2	3	3	3	2	22
Robson & Lian (2017) [40]	3	3	2	2	3	3	3	3	22
Sarudiansky et. al. (2017) [41]	2	3	3	2	3	3	3	2	21
Thompson et. al. (2009) [42]	2	3	2	2	1	2	3	2	17
Wyatt et. al. (2014) [43]	2	3	2	1	3	2	3	2	18

Note: Items in columns 2-9 relate to questions 3-10 in the CASP tool. See appendix 1 - E for a copy.

Appendix 1 - B Sample of meta-ethnography process

Theme	Translation	Code	Papers	
Delegitimization	Dismissed, less valid	Nothing can be wrong with you	Dosanjh et. al. (2020), Pretorius & Sparrow (2015), Robson & Lian (2017), Nielsen (2020)	
		Not believed	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), McWilliams et. al. (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Sarudiansky et. al. (2017), Thompson et. al. (2009), Wyatt et. al. (2014).	
		Not listened to	Dosanjh et. al. (2020), Robson & Lian (2017), Wyatt (2014)	
		Dismissing medical interactions	Dosanjh et. al. (2020), Pretorius & Sparrow (2015), Robson & Lian (2017), Thompson et. al. (2009)	
		Dismissing school interaction	Karterud et. al. (2016), McWilliams et. al. (2016)	
		It's under your control – feigning and blame	Dismissed in the workplace	Karterud et. al. (2016)
			Making it up	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud (2016), Rawlings (2017), Robson & Lian (2017), Wyatt (2014)
			Being blamed	Dosanjh et. al. (2020), Karterud et. al. (2015), Neilsen et. al. (2020), Rawlings et. al. (2017), Robson & Lian (2017), Sarudiansky et. al. (2017), Thompson et. al. (2009),
		Less than organic	Not organic – not valid	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), McWilliams et. al. (2016), Rawlings (2017), Robson & Lian (2017), Thompson et. al. (2009)
		Refutations		Opposite of dismissed
Positive diagnosis	Dosanjh et. al. (2020), Karterud (2015)			
Believed	Dosanjh (2020), Karterud et. al. (2015), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Thompson et. al. (2009), Wyatt (2014)			

Theme	Translation	Code	Papers
Excluded, Isolated, abandoned – the social cost of stigma	Journey to treatment, what treatment?	Seeking answers	Dosanjh (2020), Nielsen et. al. (2020) Pretorius & Sparrow (2015), Rawlings (2017), Rawlings (2018), Robson & Lian (2017), Thompson et. al. (2009), Wyatt et. al. (2014)
		No knowledge or understanding	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al., Karterud et. al. (2016), McWilliams et. al. (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Sarudiansky et. al. (2017), Thompson et. al. (2009), Wyatt et. al. (2014).
		Abandoned by healthcare	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), McWilliams et. al. (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Thompson et. al. (2009), Wyatt et. al. (2014).
	Loss of status	Demoted & rejected in the workplace	Dosanjh et. al. (2020), Karterud (2010)
		Role change	Dosanjh et. al. (2020), Karterud (2016) McWilliams et. al. (2016), Robson & Lian (2017), Pretorius & Sparrow (2015) Thompson et. al. (2009)
		Loss of independence	Dosanjh et. al. (2020), McWilliams et. al. (2016), Pretorius & sparrow (2015), Thompson et. al. (2009)
	Excluded and isolated	Social exclusion	Karterud (2016), McWilliams et. al. (2016), Nielsen (2020), Pretorius & Sparrow (2015), Rawlings (2018), Robson & Lian (2017), Thompson et. al. (2009),
		Relationship strain	Dosanjh (2020), McWilliams (2016), Nielsen et. al. (2020)
	Physical harm and unethical treatment	Conflict	Dosanjh et. al. (2020), Nielsen et. al. (2020), Sarudiansky et. al. (2017),
		Physical harm & attempts to find feigning	Robson & Lian (2017), Pretorius & Sparrow (2015)
Refutations	Understanding (rare occasions)	Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), Robson & Lian (2017)	

Theme	Translation	Code	Papers
The costs of attempts to manage stigma	Masking and seeking legitimacy	Carry on as normal and conceal the diagnosis Control information/pretend its epilepsy	Dosanjh et. al. (2020), Karterud (2016) Karterud (2010), Karterud (2016), McWilliams (2016)
	Avoiding and withdrawing	Self-isolate	Dosanjh et. al. (2020), Karterud (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Robson & Lian (2017), Thompson et. al. (2009).
	Beliefs about belief	Avoid healthcare and mistrust	Karterud (2010), Robson & Lian (2017), Wyatt et. al. (2014)
		Perceived judgement	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), McWilliams et. al. (2016), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Sarudiansky et. al. (2017), Thompson et. al. (2009), Wyatt et. al. (2014).
	Fear reaction of others	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), McWilliams et. al. (2016), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Sarudiansky et. al. (2017), Thompson et. al. (2009), Wyatt et. al. (2014).	
Refutations	Connection & legitimacy leading to participation	Dosanjh et. al. (2020), Karterud et. al. (2015), Karterud et. al. (2016), Pretorius & Sparrow (2017), Thompson et. al. (2009), Wyatt et. al. (2014)	

Theme	Translation	Code	Papers
Threats to identity and the meaning of mental health	Seizures mean I'm a bad person	Nothing medically wrong with you, what is the problem?	Dosanjh et. al. (2020), Rawlings et. al. (2018), McWilliams et. al. (2016), Nielsen et. al. (2020), Sarudiansky et. al. (2017) Robson & Lian (2017), Thompson et. al. (2009)
		Dishonest/attention seeker	Karterud et. al. (2016), McWilliams et. al. (2016) Nielsen et. al. (2020), Rawlings (2017), Robson & Lian (2017),
	Internalised blame and stigma	Self-blame/Am I making this up?	Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), McWilliams et. al. (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Rawlings et. al. (2017), Rawlings et. al. (2018), Robson & Lian (2017), Thompson et. al. (2009), Wyatt et. al. (2014).
		"I'm not crazy" – mixed responses to psychological explanations	Not mental illness
	Psychogenic implying control		Dosanjh et. al. (2020), Karterud et. al. (2010), Karterud et. al. (2015), Karterud et. al. (2016), Nielsen et. al. (2020), Pretorius & Sparrow (2015), Rawlings et. al. (2017), Robson & Lian (2017), Wyatt et. al. (2014).
	Refutations	Meaningful understanding	Dosanjh et. al. (2020), Karterud et. al. (2015), Nielsen et. al. (2020), Sarudiansky et. al. (2017), Thompson et. al. (2009)

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[2] Van der Geer J, Hanraads JAJ, Lupton RA. The art of writing a scientific article. *Heliyon*. 2018;19:e00205. <https://doi.org/10.1016/j.heliyon.2018.e00205>

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[3] Strunk Jr W, White EB. *The elements of style*. 4th ed. New York: Longman; 2000.

Reference to a chapter in an edited book:

[4] Mettam GR, Adams LB. How to prepare an electronic version of your article. In: Jones BS, Smith RZ, editors. *Introduction to the electronic age*, New York: E-Publishing Inc; 2009, p. 281–304.

Reference to a website:

[5] Cancer Research UK. Cancer statistics reports for the UK, <http://www.cancerresearchuk.org/aboutcancer/statistics/cancerstatsreport/>; 2003 [accessed 13 March 2003].

Reference to a dataset:

[dataset] [6] Oguro M, Imahiro S, Saito S, Nakashizuka T. Mortality data for Japanese oak wilt disease and surrounding forest compositions, Mendeley Data, v1; 2015. <https://doi.org/10.17632/xwj98nb39r.1>.

Note shortened form for last page number. e.g., 51–9, and that for more than 6 authors the first 6 should be listed followed by 'et al.' For further details you are referred to 'Uniform Requirements for Manuscripts submitted to Biomedical Journals' (J Am Med Assoc 1997;277:927–34) (see also [Samples of Formatted References](#)).

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Appendix 1 - D CASP Checklist



CASP Checklist: 10 questions to help you make sense of a **Qualitative** research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

- ▶ Are the results of the study valid? (Section A)
- ▶ What are the results? (Section B)
- ▶ Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is “yes”, it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a “yes”, “no” or “can’t tell” to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

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Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- what was the goal of the research
 - why it was thought important
 - its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
 - Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

4. Was the recruitment strategy appropriate to the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the researcher has explained how the participants were selected
 - If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
 - If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the setting for the data collection was justified
 - If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
 - If the researcher has justified the methods chosen
 - If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
 - If methods were modified during the study. If so, has the researcher explained how and why
 - If the form of data is clear (e.g. tape recordings, video material, notes etc.)
 - If the researcher has discussed saturation of data

Comments:

6. Has the relationship between researcher and participants been adequately considered?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

8. Was the data analysis sufficiently rigorous?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
 - To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider whether

- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix 1 – E Full search details

PSYCINFO

	Search terms
	<p>FND terms (DE Somatization) OR (DE "Somatization Disorder") OR (DE "Somatoform Disorders") OR (DE "Conversion Disorder") OR (DE "Movement Disorders") OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) N3 dissociative) OR ((seizure* OR convulsion*) N3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) N3 seizure*) OR ((conversion OR psychogenic OR functional) N3 paralysis) OR ((conversion OR psychogenic OR functional) N3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional N3 paralysis"</p>
AND	<p>Qualitative research terms (DE "Qualitative Measures") OR (DE "Qualitative Methods") OR (DE "Focus Group") OR (DE "Focus Group Interview") OR (DE "Interview Schedules") OR (DE Interviewers) OR (DE Interviewing) OR (DE Interviews) OR (DE "Interpretative Phenomenological Analysis") OR (DE "Mixed Methods Research") OR DE ("Grounded Theory") OR ((personal OR patient OR subjective OR lived OR subjective) N3 experience) OR interview OR qualitative OR "focus group" OR phenomenological OR "phenomenolog*" OR "conversation analysis" OR "thematic analysis" OR "discourse analysis" OR "grounded theory" OR "narrative analysis" OR "mixed method*"</p>
Note: DE is a psycINFO subject heading search term	

Web of Science

	Search terms
	<p>FND terms Somatization OR "Somatization Disorder" OR "Somatoform Disorders" OR "Conversion Disorder" OR "Movement Disorders" OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) NEAR/3 dissociative) OR ((seizure* OR convulsion*) NEAR/3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) NEAR/3 seizure*) OR ((conversion OR psychogenic OR functional) NEAR/3 paralysis) OR ((conversion OR psychogenic OR functional) NEAR/3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional NEAR/3 paralysis"</p>
AND	<p>Qualitative research terms "qualitative measures" OR "qualitative methods" OR "focus group" OR "focus group interview" OR "interview schedules" OR "interviewers" OR</p>

	"interviewing" OR "interviews" OR "interpretative phenomenological analysis" OR "grounded theory" OR ((personal OR patient OR subjective OR lived OR subjective) N3 experience) OR interview OR qualitative OR "focus group" OR phenomenological OR "phenomenolog*" OR "conversation analysis" OR "thematic analysis" OR "discourse analysis" OR "grounded theory" OR "narrative analysis" OR "mixed method*"
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	Search terms
	<p>FND terms MH "Somatoform Disorders" OR MH "Movement Disorders" OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) N3 dissociative) OR ((seizure* OR convulsion*) N3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) N3 seizure*) OR ((conversion OR psychogenic OR functional) N3 paralysis) OR ((conversion OR psychogenic OR functional) N3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional N3 paralysis"</p>
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Note: MH is a CINAHL subject heading search term	

MEDLINE

	Search terms (boolean)
	<p>FND terms MH "Somatoform Disorders" OR MH "Movement Disorders" OR MH "Conversion Disorder" OR MH "Movement Disorders" OR "non-epileptic" OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non</p>

	epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) N3 dissociative) OR ((seizure* OR convulsion*) N3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) N3 seizure*) OR ((conversion OR psychogenic OR functional) N3 paralysis) OR ((conversion OR psychogenic OR functional) N3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional N3 paralysis"
AND	Qualitative research terms MH "Qualitative Research" OR MH "Focus Groups" OR MH "Interviews as topic" OR MH "Grounded Theory" OR MH Personal Narratives as Topic" OR ((personal OR patient OR subjective OR lived OR subjective) N3 experience) OR interview OR qualitative OR "focus group" OR phenomenological OR "phenomenolog*" OR "conversation analysis" OR "thematic analysis" OR "discourse analysis" OR "grounded theory" OR "narrative analysis" OR "mixed method*"
Note: MH is a MEDLINE subject heading search term	

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	Search terms
	FND terms Psychogenic nonepileptic seizure/ OR somatoform disorder/ OR conversion disorder/ OR somatization/ OR "functional neurological disorder" OR pseudoseizure OR pseudo-seizure OR "functional movement disorder" OR NEAD OR PNES OR NES OR "attack disorder" OR nonepileptic OR non-epileptic OR "non epileptic" OR pseudoseizure* OR ((seizure* OR convulsion*) NEAR/3 dissociative) OR ((seizure* OR convulsion*) NEAR/3 hysterical) OR "pseudoepilep*" OR hysteroepilep* OR ((conversion OR psychogenic OR functional) NEAR/3 seizure*) OR ((conversion OR psychogenic OR functional) NEAR/3 paralysis) OR ((conversion OR psychogenic OR functional) NEAR/3 movement) OR FND OR "functional neurologic*" OR "functional movement" OR "functional motor" OR "functional NEAR/3 paralysis"
AND	Qualitative research terms Qualitative analysis/ OR qualitative research/ OR interview/ OR semi structured interview/ OR structured interview/ OR telephone interview/ OR psychological interview/ OR unstructured interview/ OR phenomenology/ OR grounded theory/ OR ((personal OR patient OR subjective OR lived OR subjective) N3 experience) OR interview OR qualitative OR "focus group" OR phenomenological OR "phenomenolog*" OR "conversation analysis" OR "thematic analysis" OR "discourse analysis" OR "grounded theory" OR "narrative analysis" OR "mixed method*"
Note: / denotes a subject heading term used in EMBASE	

Section 2 – Empirical Paper

A case series investigating the benefits of a mindfulness training smartphone application for
Functional Seizures

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Abstract

This study used a single case experimental design incorporating both quantitative and qualitative data to assess the effect of mindfulness training delivered through a smartphone app for people experiencing functional seizures (FS). Four participants completed the study while one further participant completed it in part. Qualitative analysis identified that participants found an app-delivered intervention to be acceptable. Reliable and clinically significant changes were observed on outcome measures of seizure frequency, quality of life, psychological distress and a process measure of experiential avoidance for two participants and reliable and clinically significant improvements in mindfulness were observed in three participants. Similar benefits were not observed in the remaining participants indicating that this intervention may be suitable for some but not all people experiencing FS. Improvements in experiential avoidance appeared to be a key process for those who benefitted from this intervention. The implications of these findings for clinical practice and future research are discussed.

Keywords; Functional seizures, non-epileptic attack disorder, mindfulness, single-case experimental design.

1. Introduction

1.1 Functional Seizures

Functional Seizures (FS) are events that present in a similar fashion to seizures caused by epilepsy. However, FS do not have the same corresponding electrical brain activity seen in epileptic seizures. The difficulty in identifying an organic cause has led to some alternative names for FS – Non-Epileptic Attack Disorder (NEAD) or Psychogenic Nonepileptic Seizures (PNES). FS has been proposed as a term that does not confer the same level of stigma as other terms and is a term widely understood by professionals [1] and has been adopted by advocacy groups such as FND Hope [2]. Thus, the term FS has been used throughout this paper.

FS can be distressing for individuals who experience them as well as their families. They can be a source of significant burden [3] and impact upon people's ability to live their lives as intended, interfering with their ability to work [4], and placing a strain on social relationships [3]. Indeed, research has shown that FS have a greater impact on quality of life than epilepsy [5]. It is estimated that FS affect between two to 33 per 100,000 population [6]; another estimate measured an incidence rate of 4.9/100,000 per year [7]. By either measure, FS is a frequently occurring neurological condition and it is estimated that 20% of people presenting to epilepsy clinics later receive a diagnosis of FS [8].

In addition to the individual burden of FS, there is a societal impact in the form of healthcare costs as people with FS frequently present to health services seeking support [9, 10].

Diagnosis of this condition is often delayed, with one study identifying an average wait of 7.2 years [11]. This may lead to additional healthcare service utilization while awaiting a

diagnosis. The lack of appropriate treatment options for people with FS may play a role here, with people with FS returning to health services in times of crisis to seek further support. Many people with FS also report difficult interactions with healthcare services such as healthcare workers showing a lack of understanding or disbelief of their symptoms [3].

1.2 Present understandings of FS

At present, no single explanatory model exists for FS. While risk factors have been identified for FS, these are not present for all individuals that experience FS. For instance, psychological trauma [15] is central to FS for some but not all. Similarly, FS disproportionately affects females and people who live in areas characterised by high levels of socioeconomic deprivation [16] though neither play a causative role in the aetiology of FS. While risk factors and characteristics go some way to describing those who experience FS, no aetiology has been developed to describe all cases of FS.

For this reason, a biopsychosocial approach to FS has been proposed by some such as Baslet [17]. This model highlights predisposing factors such as trauma and adverse experiences, precipitating factors including stress combined with vulnerabilities such as avoidance tendencies and difficulty with emotional regulation. The perpetuating factors in this model include comorbid conditions further compounded by experiences such as chronic stress.

In a similar vein, an integrative cognitive model has recently been proposed by Brown and Reuber [15]. In this model, Brown and Reuber [15] identified four existing concepts that they

sought to integrate. These included FS as the activation of dissociated material, FS as a hard-wired behavioural response to events, FS as a physical manifestation of emotional distress and FS as learned behaviour. At its core, the model proposes a “seizure scaffold” described as a mental representation of seizures which, when activated by internal or external cues combined with a compromise in inhibitory functioning caused by chronic stress or other factors, results in a seizure. Additionally, the model recognises the varied pathways through which people develop FS. In doing so, it seeks to accommodate research on FS from a variety of perspectives and also recognise the variety of people that develop FS.

1.3 Psychological Treatments for FS

Despite the interest in this condition over such a long period of time, and models that indicate a psychological basis, research on effective psychological treatments is in its early stages. A recent meta-analysis [18] made the case for psychotherapy, highlighting that upon completion of therapy, 82% of participants showed a reduction in seizure frequency of over 50%. However, of the 13 studies included in the review, 11 were assessed as having a strong risk of bias. Moreover, the use of papers with this level of bias is an indication of the lack of available high quality research in this area which has been noted in a previous critical review [19].

However, research in this area has been progressing. For example, there has been increased interest in the application of cognitive behavioural therapy (CBT) to FS and a number of randomized controlled trials (RCTs) [20, 21] have shown promising results leading to significant reductions in seizure frequency and benefits such as improved quality of life. However, a recent larger scale RCT investigating CBT for FS [22] found that while this

treatment brought benefits to quality of life and psychological distress, it did not lead to a significant improvement in seizure frequency compared to standard medical care.

1.4 Third-wave approaches and mindfulness

Third-wave cognitive therapies such as mindfulness-based therapies and acceptance and commitment therapy (ACT) place an emphasis on individuals' relationships with their thoughts and emotions rather than on their content [23]. A position paper [24] highlighted the benefits that third-wave cognitive therapies could bring to people experiencing FS. In doing so it hypothesised that mindfulness may be useful in countering experiential avoidance – the avoidance of difficult thoughts and emotions - and in building psychological flexibility – the ability to remain fully present in the moment, allowing one to act in a way which reflects one's own values [25]. Further empirical research has highlighted a higher prevalence of psychological inflexibility and experiential avoidance in FS compared to other populations [26], suggesting that interventions focusing on this could be beneficial. Building on this, intervention studies based on third wave approaches are just beginning – for example a recent single case experimental design (SCED) study [27] showed the benefits of an ACT intervention for FS. Four out of six participants in this study reported that they found the mindfulness aspect of this intervention particularly influential in the change they experienced.

Mindfulness practice is an element of all third wave therapy models which involves developing the ability to pay attention to the present moment in a non-judgemental way. Mindfulness has a growing evidence base for a range of mental [28] and physical [29, 30] health conditions, including epilepsy [31]. However, less empirical research is available on

the processes through which mindfulness achieves these benefits. Nonetheless, a number of processes have been proposed such as increasing body awareness, improving people's ability to regulate attention, changing people's perspective of themselves and improving emotional regulation through exposure, reappraisal and reconsolidation [32]. Through these processes, it has been hypothesised that less useful strategies such as experiential avoidance become less automatic [33] giving people more choice around their reactions to internal and external events.

Considering mindfulness processes in light of the model proposed by Brown and Reuber [15], we may hypothesise that practising mindfulness could give people with FS greater control over the activation of the seizure scaffold schema. Similarly, linking with Baslet's model [34], the effect of mindfulness on precipitating factors including stress may provide benefits to those living with FS. Furthermore, as noted above, experiential avoidance plays a role in FS for many [26] and has been shown to improve with mindfulness practice [35]. In fact, a little empirical evidence exists indicating the possible benefits of mindfulness-based interventions for FS. For instance, a recent study [36] investigated the effects of individual mindfulness-based therapy delivered face-to-face. This study showed an improvement in seizure frequency and quality of life for participants who completed the intervention. However, the study noted that potential participants could not take part due to difficulties with transport arrangements, and a further 23 out of a total of 49 participants did not complete the intervention with a number of these participants citing difficulty attending appointments as a reason for dropping out of the study.

In addition to the above study, research has found high dropout rates from psychological and psychiatric interventions for people with FS. For example, a study examining adherence to psychiatric treatment found that only 17% of those referred attended the fourth of four appointments offered [37]. Additionally, high drop-out rates have been noted in an RCT study investigating CBT for FS where only 56% of participants attended all 12 sessions offered [22]. Despite these observations, little is known about the reasons why completion rates are so low. However, one reason as suggested above relates to physical barriers to travel to treatment centres. In this vein, a study investigated telehealth to support people with FS to access psychotherapy [38] and saw a completion rate of 84%.

Alongside recent developments in technology, numerous efforts have been made to deliver psychological treatments remotely through the use of smartphones. Mindfulness training programmes delivered in this fashion have been shown to improve wellbeing [39].

Furthermore, in the context of the covid-19 pandemic, the importance of remotely delivered healthcare has come to the fore.

Additionally, given the variety of presentations and maintaining factors proposed by the integrative cognitive model [15], it is important to identify further treatments to ensure that psychological treatments offered to individuals presenting with FS can be matched to their unique situation.

1.5 Aims

Consequently, the purpose of this study was to evaluate the effectiveness and acceptability of a mindfulness-based intervention for people experiencing FS delivered via smartphone app. A case series based on SCED was chosen to investigate this. The primary aims of the study were to investigate whether such an intervention would lead to reduction in the frequency of seizures and improvement in quality of life among this group. Furthermore, the study aimed to assess the impact of the intervention on the process measures of experiential avoidance and mindfulness. Finally, the research aimed to gain an insight into the experience of people who used this intervention to determine its acceptability among this group and to assess its strengths and weaknesses from the participants' perspective.

Therefore, the research questions in this study were; (i) what effect does a mindfulness training app intervention have on the quality of life of people experiencing FS? (ii) Does a mindfulness training app intervention reduce seizure frequency in people experiencing FS? (iii) Does a mindfulness training app intervention have an effect on levels of psychological distress for people experiencing FS? (iv) Does a mindfulness training app intervention lead to a reduction in experiential avoidance and increase in mindfulness in people experiencing FS? (v) How do people with FS experience a mindfulness training app intervention?

2. Method

2.1 Design

This study used a SCED methodology. The key advantage of this design is its ability to detect change on an individual level [40]. Through repeated measurement of target variables across distinct conditions, SCED allows participants to act as their own control rather than

comparing them to a group. Therefore, comparison is a central feature of this design in that it involves a systematic comparison of two or more experimental phases. This approach is particularly useful when assessing the impact of novel treatments with specific groups of interest [41]. Additionally, compared with larger scale group studies using pre and post measures only, SCED studies have the ability to give more information about the process of change [42].

Specifically, an AB multiple baseline design [43] was used in this study. This consisted of a baseline “A” phase and an intervention “B” phase. In SCED studies the baseline phase is an integral component which allows for experimental control. The baseline consists of a period where data are collected before the intervention is introduced and subsequently allows for a comparison between the intervention phase and the baseline for each participant. An underlying assumption in SCED research is that changes in the target variable that occur upon the introduction of an intervention and do not occur at other times are likely to be caused by that intervention [40]. Participants were recruited on a rolling basis which ensured that the phases were staggered for each participant which introduced randomization to the design, thus increasing the internal validity [44]. This specific SCED design was chosen as it allowed for numerous participants rather than just one, and its suitability to measure change over time compared to an introduction/withdrawal design such as ABAB. Participants completed a baseline phase (A). Following this, participants completed the intervention phase (B) where they engaged with the intervention.

2.2 Participants

Participants were recruited online with the support of a third sector organisation that provides advocacy, support and research into functional neurological disorders. Those who met the following inclusion criteria were invited to take part in the study: (1) had a current diagnosis of FS (or NEAD/PNES), (2) were over 18 years of age and able to provide consent to participate, (3) did not have a diagnosis of epilepsy, (4) were not currently experiencing addiction or psychosis, (5) did not have a current regular mindfulness practice, and (6) were not currently engaged in another psychological intervention.

The exclusion criteria around addiction and psychosis were chosen in order to maximise the safety of participants. In rare cases, mindfulness practice has resulted in difficulties for people who have experience of psychosis [45] and caution has been advised around its use with people experiencing addiction [46].

2.3 Intervention

The mindfulness training intervention used was the Smiling Mind smartphone app [47]. The app was developed by psychologists to provide mindfulness training in an accessible format. This particular app was chosen due to its quality rating [48] and its mirroring of meditations used in mindfulness programmes with an established evidence base such as mindfulness-based stress reduction [49]. Additionally, the app has been developing its own evidence base. For example, it has led to improvements in depressive symptoms, college adjustment and resilience in an RCT involving 208 undergraduate students [50] and a further study demonstrated a reduction in the concentration of salivary cortisol in a sample of athletes using the app during a competition [51].

2.4 Measures

All data in the study were collected via the Qualtrics online data collection platform.

Participants were sent a link each day of their active participation in the study to complete the relevant measures. See Table 2 for a summary of the measures used and their psychometric properties.

The study collected the following data on a daily basis:

Seizure frequency was a self-reported measure of the number of FS experienced in the previous 24 hours. For the purpose of this measure a non-epileptic seizure was self-defined by each participant.

Quality of life (QoL) was measured using the five-item work and social adjustment scale (WSAS) [52]. This measures functional impairment caused by FS in the domains of work, home management, social leisure activities, private leisure activities, family and relationships. The measure has been validated in a variety of healthcare populations [53] and has been used in an online study of FS [26].

Further measures used in this study were collected at baseline, weekly during the active phase of the intervention and at four-week follow-up:

The 7-item acceptance and action questionnaire (AAQ-II) [25] was used to measure the process of experiential avoidance. This measure has previously been used online in FS research [26].

The 15-item five factor mindfulness questionnaire (FFMQ-15) [54] which measures the mindfulness traits of observing, describing, acting with awareness, being non-judgemental and non-reactive. This was used to measure the process of mindfulness trait development.

The CORE-10 (10 items) [53] is a measure of broad psychological distress and was used to monitor participants' levels of distress throughout the study.

Participants were also asked to report the number of sessions of meditation that they completed each week using the app.

Finally, a brief change interview based on Elliot [55, 56] was administered by phone at one-month follow-up to assess the participants' experiences of the intervention, to develop an understanding of any qualitative benefits or difficulties experienced. See Table 1 for the change interview schedule.

Table 1 - Change Interview Schedule

	Theme	Question
1.	General Experience	What has using this intervention been like for you?
2.	Changes	What changes, if any, have you noticed in yourself since you began using this intervention?

- | | | |
|-----|---------------------------------|--|
| 3. | Likelihood without intervention | How likely do you think that these changes would have been without the intervention? |
| 4. | Expectedness | Did you expect any of those changes? |
| 5. | Importance | On a scale of 1 to 5, how important would you rate these changes? |
| 6. | Attribution | In general, what do you attribute these various changes to? |
| 7. | Resources | What personal strengths or aspects of your current life situation have helped you to use this intervention to deal with your problems? |
| 8. | Limitations | What things about you or your life situation have made it harder for you to use teaching from this intervention to deal with your problems related to NEAD or general wellbeing? |
| 9. | Helpful aspects | What have been the most helpful things about the intervention? |
| 10. | Problematic Aspects | What kinds of things about the intervention have been hindering, unhelpful, negative or disappointing for you? Was there anything that was difficult or missing? |
| 11. | Research Aspects | What has been like for you to be involved in this research? |
| 12. | Open | Is there anything else that you'd like to add? |

2.5 Ethics

Ethical approval was received from Lancaster University Faculty of Health and Medicine Research Ethics Committee.

Due to the nature of this study involving a psychological intervention the safety of participants was a central concern. While mindfulness practice is a low risk activity, in very rare instances it may lead to the experience of distress. The study took a number of steps to mediate this. Firstly, the risk was reduced through the use of a publicly available smartphone app which has been judged to be safe and using exclusion criteria to ensure those at greater risk of harm did not take part. Throughout the active intervention phase of the study, participants were offered a weekly phone or video call with the researcher. This sought to identify whether any participants were experiencing any harm. The researcher engaged in regular supervision during the intervention phase of the study and the field supervisor who is an expert in the field of FS provided oversight.

As participants were required to spend a significant amount of time completing tasks related to the study, they were offered a voucher for a maximum of £41.05 to compensate them for their time. Additionally, the payment was provided in line with NHS Health Research Authority guidance [57]. In adhering to this guidance, the payment was not excessive and was available to all participants who completed any part of the study on a pro-rata basis.

2.6 Procedure

Recruitment was conducted online, via a third sector organisation who shared details of it across their social media platforms.

Participants responded to the advertisement and all those interested were provided with a participant information sheet, had a further discussion with the lead researcher about the study and completed a screening and demographic questionnaire. Eligible participants who wished to take part then completed an online consent form.

Firstly, participants submitted baseline data, measuring seizure frequency and QoL on a daily basis. Where the baseline was stable, defined in line with guidance from Kratochwill et. al. [43] as level, not changing, or deteriorating, the intervention was started after seven days. Where data were trending in a therapeutic, improving direction the baseline phase was extended by three days at a time and reassessed. The baseline was not extended by more than six days for any participant due to the additional demand that this placed on participants.

After completing baseline measures, participants engaged in a mindfulness training intervention using the Smiling Mind app [47] over the course of seven weeks. Participants were asked to complete the “introduction to mindfulness” and “mindfulness foundations” programmes using the app. These two programmes provided teaching and guided meditation sessions that varied in length from one to 40 minutes with an average of nine minutes. A set of six to seven exercises from the app were recommended to each participant each week and they worked through them at their own pace. While engaging in the intervention, participants received a link daily to submit measures of seizure frequency and WSAS and weekly to complete the remainder of the measures. Throughout the intervention, the researcher offered a weekly phone-call to monitor participants’ safety and reflect on their experience of the intervention.

Four weeks after finishing the intervention, participants completed a follow-up set of daily and weekly measures along with a change interview with the researcher over the telephone which was recorded and transcribed verbatim.

Table 2 - Psychometric Properties and Characteristics of Measures

	Construct	No. of items	Example Item	Internal Consistency	Test-Retest Reliability	Scale Range and Direction
Daily measures						
Seizure frequency	Number of seizures daily	1	“How many seizures have you experienced over the past 24 hours?”	-	-	Higher score = more frequent seizures
WSAS[52]	Functional impairment	5	“Because of my seizures my social leisure activities (with other people e.g. parties, bars, clubs, outings, visits, dating, home entertaining) are impaired”	.87[26]	.73[52]	0-40 Higher score = greater impairment
Weekly measures						
CORE-10[53]	Psychological distress	10	“I have felt despairing or hopeless”	.9[53]	.83[58]	0-40 Higher score = greater psychological distress
AAQ-II[25]	Experiential Avoidance	7	“I worry about not being able to control my worries and feelings”	.84[25]	.81[25]	7-49 Lower scores = greater psychological flexibility, less experiential avoidance
FFMQ-15[59]	Observe	3	“I pay attention to sensations, such as the wind in my hair or sun on my face”	.64[54]	-	3-15
	Describe	3	“I’m good at finding words to describe my feelings”	.8		3-15
	Acting with awareness	3	“I find myself doing things without paying attention”	.68		3-15
	Non-judging	3	“I tell myself I shouldn’t be feeling the way I’m feeling”	.76		3-15
	Non-reactivity	3	“When I have distressing thoughts or images I just notice them and let them go”	.66		3-15 Higher scores = greater levels of mindfulness

2.7 Analysis

All data was transferred by the researcher from Qualtrics to a Microsoft Excel spreadsheet where further analysis was undertaken and graphs were produced.

Baseline data were assessed using SPSS, a line of best fit was drawn to assess the direction of the trend in the data. Once this was either stable, trending in a non-therapeutic direction or after the maximum baseline extension the intervention phase started.

A visual analysis [60] was conducted of the daily measures. Firstly, all quantitative data collected from each participant was plotted on a line graph. This was then assessed for trend, level and stability. Trend lines were drawn for daily measures using the “split-middle” method of trend estimation [60]. Percentage exceeding the median (PEM) [61] statistics were used to assess the effect size of the intervention. This method involves calculating the median for the baseline and assessing the percentage of data points that exceed this in a therapeutic direction. PEM was used on the measures for which sufficient data points were available – seizure frequency and WSAS.

Change was assessed between pre-intervention and post-intervention on the WSAS, CORE-10, AAQ-II and FFMQ-15. In order to assess whether changes observed were reliable and clinically significant, analysis of reliable change index (RCI) was conducted using guidance provided by Jacobson and Truax [62]. Where reliable change was found, clinically significant change CSC was assessed using the criteria outlined in Jacobson and Truax [62]. Under criterion c from this method, CSC is met when a participant’s post-intervention score is

closer to the mean score of the non-clinical group than that of the clinical group. As this is described as the least arbitrary of the three criteria [62] available and means and standard deviations were available from appropriate samples (see Table 3), criterion c was applied in this study.

Table 3 - Papers used for CSC calculations

Measure	Paper	Sample (number of participants)	Mean	SD
Non-clinical samples				
AAQ-II	Bond et. al. [25]	Community (568)	18.53	7.52
CORE-10	Bewick et. al. [63]	Students (1,129)	8.47	6.41
WSAS	Dawel et. al. [64]	Community (1,296)	20.5	9.3
FFMQ - O	Ortet et. al. [65]	Community (55)	9.91	2.23
FFMQ - D		Community (55)	10.31	2.62
FFMQ - A		Community (55)	8.82	1.98
FFMQ – NJ		Community (55)	11.02	2.78
FFMQ - NR		Community (55)	8.89	2.0
Clinical Samples				
AAQ-II	Baslet et. al. [66]	FS (31)	28.98	12.15
CORE-10	Goldstein et. al. [22]	FS (368)	18.2	6.5
WSAS	Mayor et. al. [67]	FS (36)	22.4	11.3
FFMQ - O	Gu et. al. [54]	Depression (238)	8.98	2.73
FFMQ - D			9.84	2.74
FFMQ - A			9.1	2.25
FFMQ – NJ			9.43	2.67
FFMQ - NR			8.58	2.3

AAQ-II = Acceptance and action questionnaire; WSAS = Work and Social Adjustment Scale; FFMQ = Five factor mindfulness questionnaire – 15 item; O = Observing; D = Describing; A = Awareness; NJ = Non-judging; NR = Non-reactivity. SD = standard deviation

Framework analysis [68] was used to analyse participants' responses to the change interview. It was chosen as it is often used to answer pragmatic questions and allows themes to be influenced a priori from research questions while also allowing for emergent themes from the data. Here, the analysis was guided by the questions in the change interview and the findings

were used to support or challenge those of the quantitative methods and to answer the final research question. This method represented a descriptive rather than an interpretative analysis. It involved coding the transcribed interview from each participant, developing a chart which represented the key themes covered by the change interview questions and adding each response to the chart alongside the most appropriate theme. Any outstanding data that did not fit with the chart could be developed into additional themes as appropriate.

3. Results

3.1 Sample demographics and adherence

Nine potential participants expressed an interest. However, three of these did not proceed due to additional life stresses (1) and not meeting the inclusion criteria (2). Six participants were recruited to the study. Over the course of the study, one participant dropped out after completing three weeks of the intervention and did not have any further contact with the researcher. Another participant was diagnosed with epilepsy during the study which meant that they no longer met the inclusion criteria, therefore this participant's data were not analysed. Full datasets from four participants who completed the study are reported here along with a partial dataset from the fifth participant who left the study early. Table 4 shows the demographics of the sample. There were four females and one male, ages ranged from 22-41 (mean age = 35.2). Three participants were engaged in university-level education, one of whom had paused their education due to difficulties associated with FS. The remaining two participants were not in employment at the time of the study, one of whom had ceased working due to difficulties with FS. Four participants described experiencing mental health difficulties alongside their FS.

In terms of adherence, P1, P2 and P4 showed high levels with each reporting that they completed in excess of five sessions of practice each week during the intervention phase. Mean reports of sessions completed each week were; 5.1 for P1, 7.2 for P2, 1.8 for P3 and 8.4 for P4. P5 reported completing a total of four sessions.

Table 4 - Demographics of sample

P	Age	Gender	M/H diagnosis	FS symptoms	FS diagnosis received	Previous Psychological Treatment for FS	Employment status	Data submitted
1	22	Female	Anxiety, Depression	1.5 years	Jan 2019	No	Trainee teacher (paused due to FS)	Full
2	28	Female	Anxiety Depression	3 months	March 2020	No	Student Nurse	Full
3	41	Male	None	4 years	March 2020	No	Postgraduate student	Full
4	44	Female	Mood difficulty	1 year	September 2019	No	Not currently working due to FS related disability	Full
5	41	Female	PTSD Depression Anxiety	10 years	2019	Emotional Freedom Technique	Not currently working	Partial

M/H; mental health diagnosis, PTSD; Post-traumatic stress disorder, FS symptoms; duration of FS symptoms at the time of the study

3.2 Visual Analysis

Graphs showing the quantitative measures for all participants are displayed in Figure 1. Formal visual analysis techniques [60] were used to analyse the measures collected daily i.e. seizure frequency and WSAS. A demonstration of all analysis methods used can be found in appendix 2 – A.

Assessment of the baseline measure of seizure frequency showed that it was stable or increasing for all participants with the exception of participant 2 whose baseline was

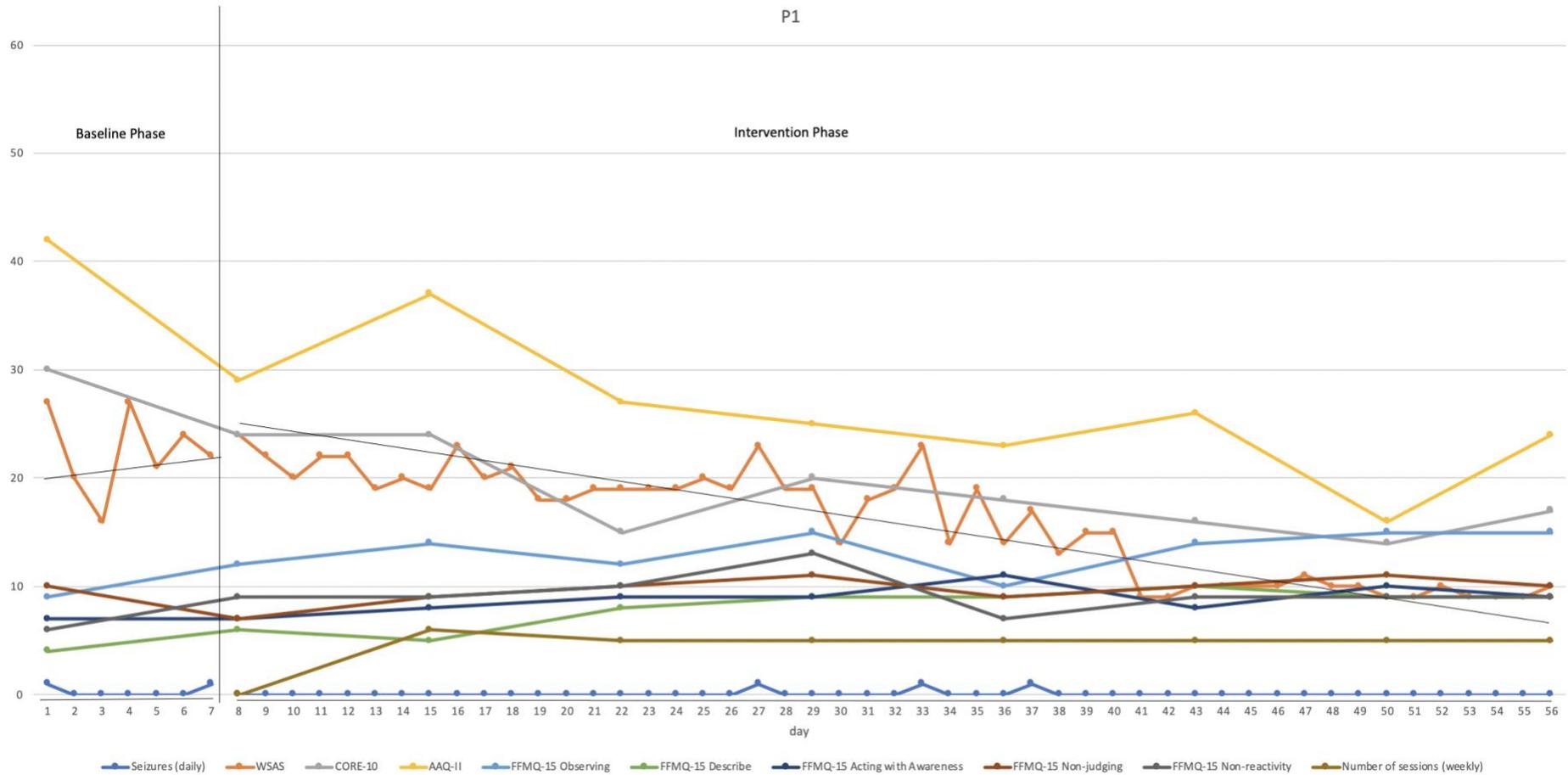
extended for 6 days in an effort to reach stability. However, as this was not reached by day 13 the intervention was started at this point due to the ethical implications of increasing the burden on the participant by delaying the start further.

Visual analysis of P1's seizure frequency showed a flat but unstable trend in the baseline phase. For P2 the trend at baseline was improving but unstable. The data for both P1 and P2 was flat and stable at 0 in the intervention phase. The trend for P3's seizure frequency data was flat in both the baseline and intervention stages though it was unstable in both conditions and there was a deterioration in the level with an increase of +2 in the median. In P4's case the trend changed from flat and stable at baseline to improving in the intervention phase. However, the data for P4's seizure frequency was variable with a stability rating of 19.15% and showed a deterioration in level with an increase in the median score of +5. P5's data showed a change from improving in the baseline to deteriorating in the intervention phase, the data for both of which were variable.

WSAS trends for P1 and P2 moved in a negative direction indicating a reduction in difficulty during the intervention. For P1 this changed from a deteriorating trend in the baseline while for P2 the improvement was from a less steeply improving trend in the baseline phase. In P3's case, the trend moved from an increasing (thus deteriorating) trend in the baseline phase to a reducing, improving trend in the intervention. However, there was a median level change of +3.5 meaning that despite the decreasing trend, P3's WSAS scores in the intervention phase were higher than those in the baseline. P4's WSAS score changed from a reducing, improving trend in the baseline phase to an increasing, deteriorating trend in the intervention phase as did that of P5.

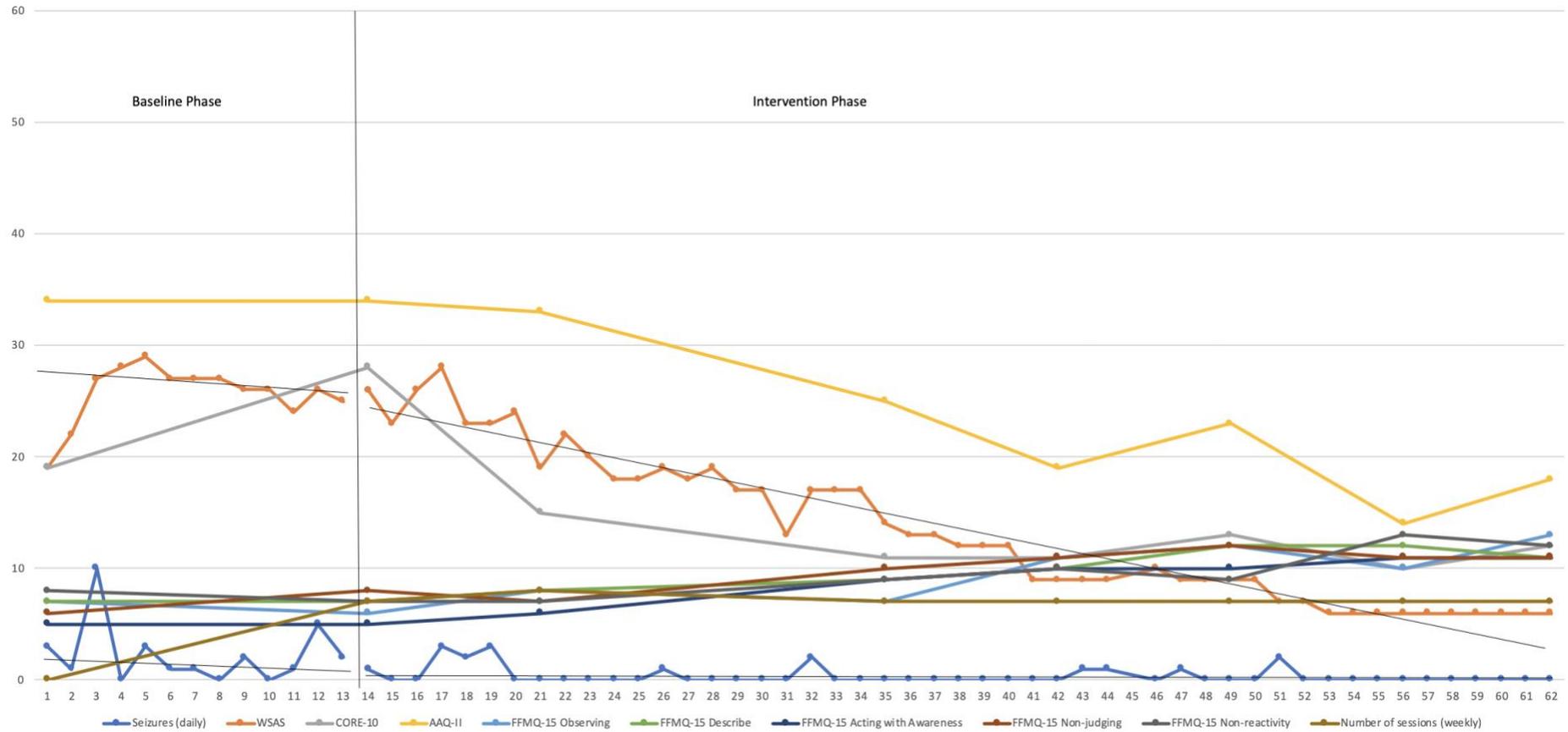
Alongside the formal visual analysis, representation of the data in graph format shows that as P1 and P2's scores on the outcome measures of WSAS and CORE-10 decreased, their scores on the AAQ-II process measure similarly reduced. Additionally, a marginal increase in the process measures of mindfulness was observed for both participants. Similar patterns were not observed for the remaining participants.

Figure 1 - Visual Presentation of data including trend lines for WSAS and seizure frequency for each participant

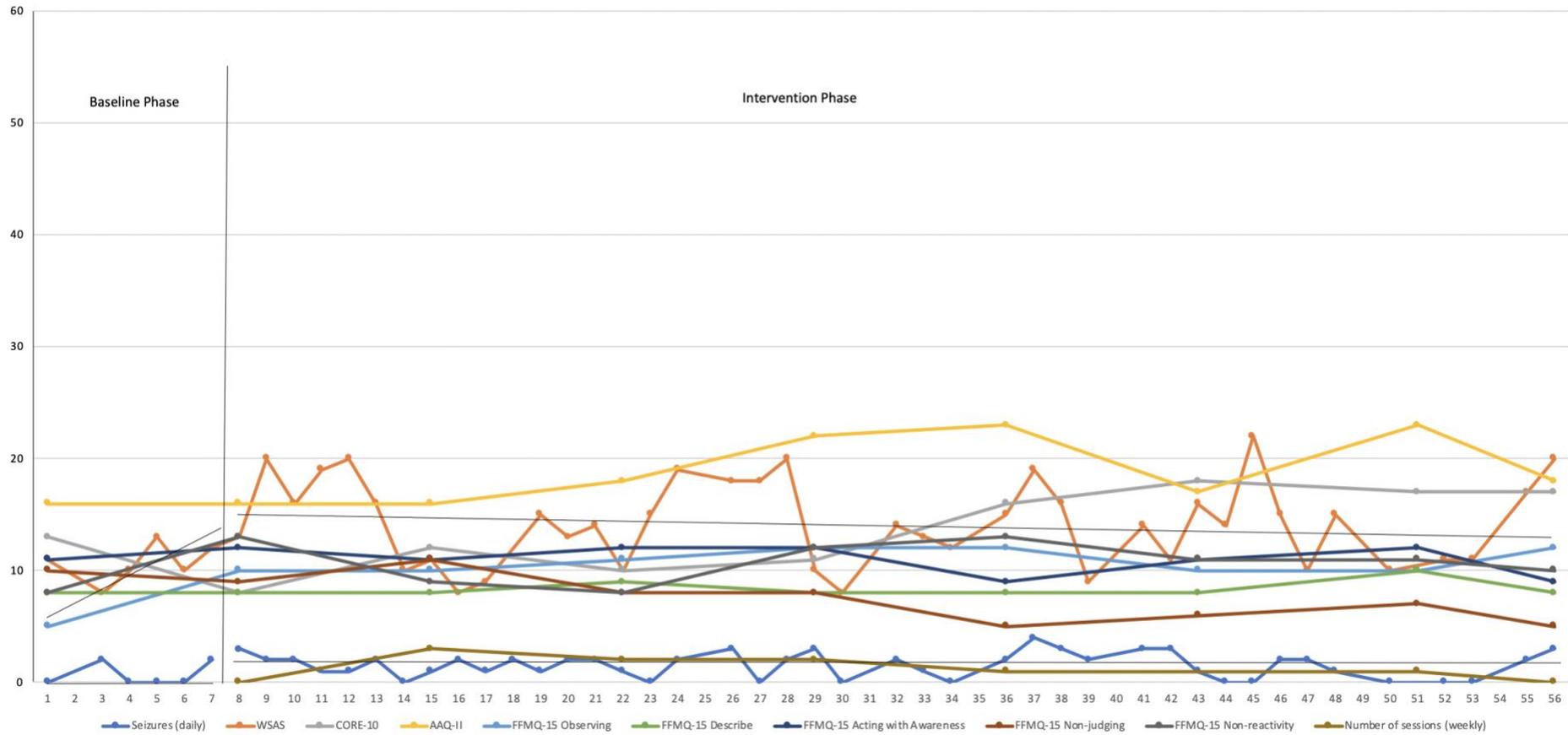


Note: WSAS = Work and social adjustment scale; FFMQ-15= Five factor mindfulness scale 15 item. Graphs are presented in the same scale to aid comparison. As a result, three data points on the daily seizure measure for P4 are missing. They are day 26 = 79, day 34 = 91, day 52 = 97. Black lines on WSAS and daily seizure frequency are trend lines calculated using the “split-middle” method

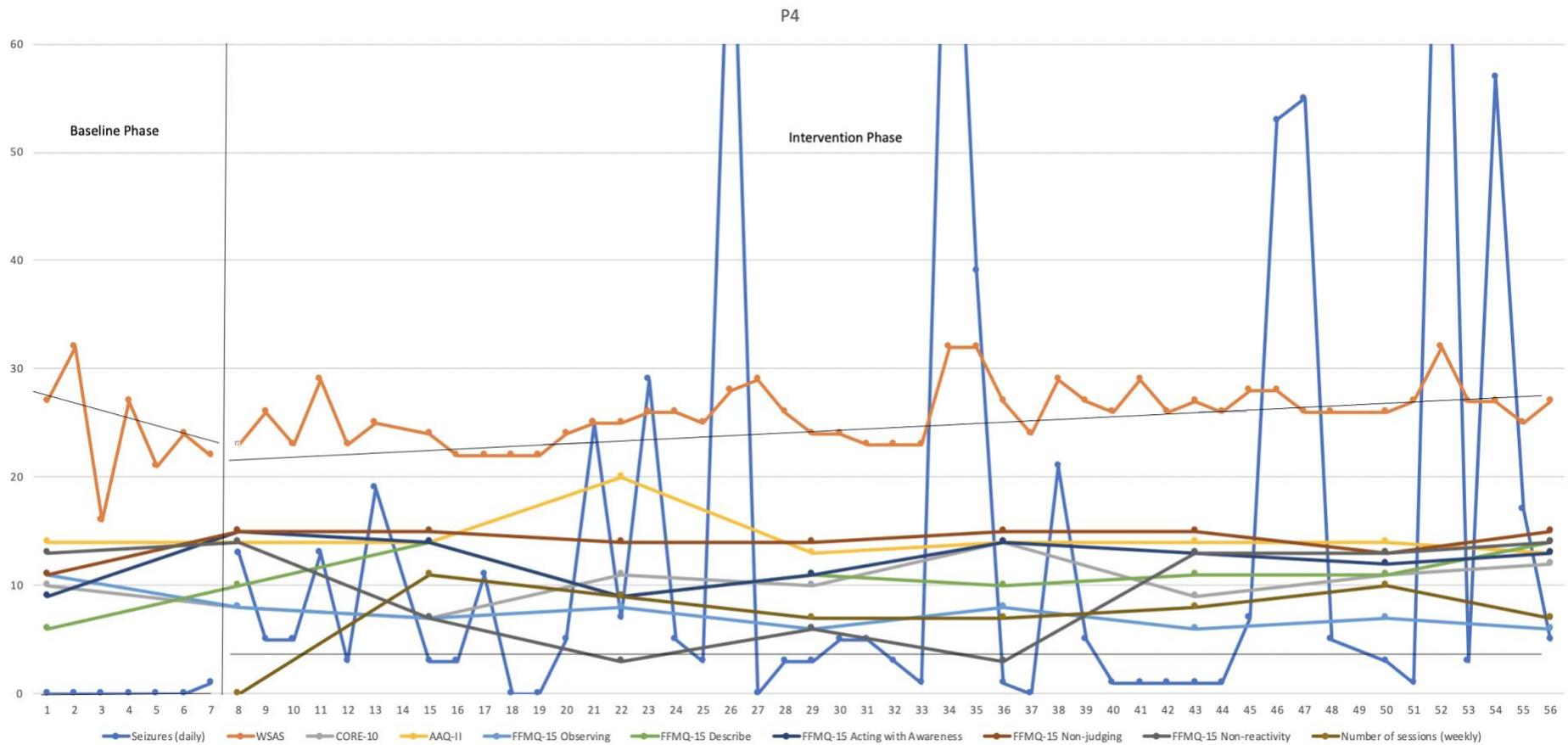
P2



Note: WSAS = Work and social adjustment scale; FFMQ-15= Five factor mindfulness scale 15 item. Graphs are presented in the same scale to aid comparison. As a result, three data points on the daily seizure measure for P4 are missing. They are day 26 = 79, day 34 = 91, day 52 = 97. Black lines on WSAS and daily seizure frequency are trend lines calculated using the “split-middle” method.

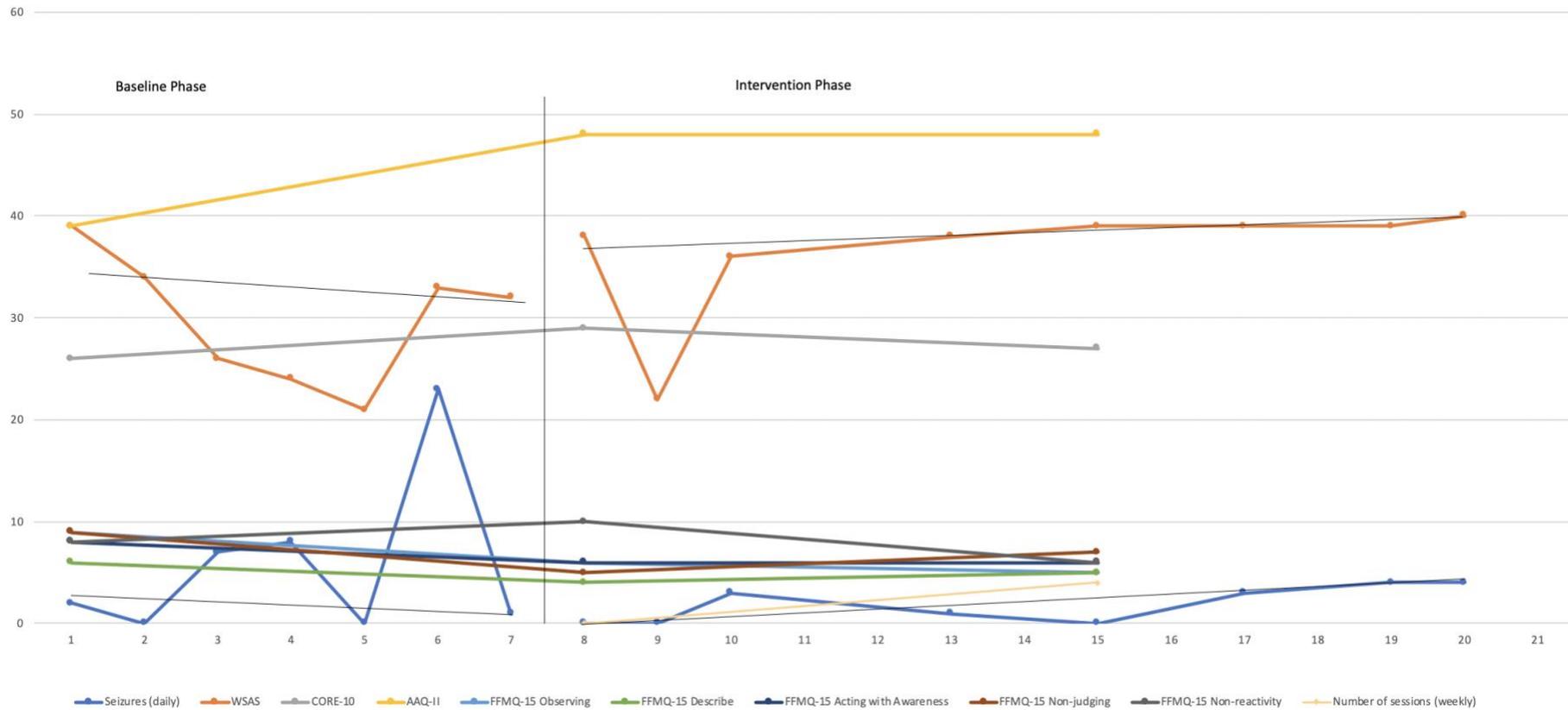


Note: WSAS = Work and social adjustment scale; FFMQ-15= Five factor mindfulness scale 15 item. Graphs are presented in the same scale to aid comparison. As a result, three data points on the daily seizure measure for P4 are missing. They are day 26 = 79, day 34 = 91, day 52 = 97. Black lines on WSAS and daily seizure frequency are trend lines calculated using the “split-middle” method.



Note: WSAS = Work and social adjustment scale; FFMQ-15= Five factor mindfulness scale 15 item. Graphs are presented in the same scale to aid comparison. As a result, three data points on the daily seizure measure for P4 are missing. They are day 26 = 79, day 34 = 91, day 52 = 97. Black lines on WSAS and daily seizure frequency are trend lines calculated using the “split-middle” method.

P5



Note: WSAS = Work and social adjustment scale; FFMQ-15= Five factor mindfulness scale 15 item. Graphs are presented in the same scale to aid comparison. As a result, three data points on the daily seizure measure for P4 are missing. They are day 26 = 79, day 34 = 91, day 52 = 97. Black lines on WSAS and daily seizure frequency are trend lines calculated using the “split-middle” method.

3.2.1 Effect size - Percentage Exceeding the Median

Ma's PEM [61] method was used to calculate effect size. This recommends the application of "highly effective" for scores over 90%, "moderate" between 70% and 90%, "mild effect" between 50% and 70% and considers scores below 50% to be "ineffective."

For seizure frequency this calculation found a moderate effect size of 79% for P2 and a mild effect size of 50% for P5. An effect size of zero was found for the remaining participants as the median seizure frequency score for each was 0 in the baseline phase.

Effect sizes on the QoL measure were in the moderate range for P1 and highly effective range for P2 with PEM scores of 86% and 94% respectively. The scores for the remaining participants fell in to the "ineffective" range with effect sizes of 22% for P3, 21% for P4 and 12.5% for P5.

3.3 Quantitative Results

3.3.1 Reliable and clinically significant change

See Table 5 for a summary of quantitative results for each participant showing RCI and CSC.

On the WSAS, scores from P1 and P2 showed reliable and clinically significant improvement at both post-intervention and follow up. Again, the scores from the remaining participants did not meet either RCI or CSC.

On the CORE-10 measure of distress, P1 and P2 showed a reliable improvement post-intervention. This was clinically significant for P2. This change remained reliable at follow up and scores for both P1 and P2 were clinically significant at this time. The scores for P3, P4 and P5 did not meet RCI or CSC on this measure.

On the AAQ-II process measure, P1 and P2 showed RCI in a therapeutic direction between baseline and post-intervention. This was maintained at follow-up. For P2 the change met CSC post-intervention and at follow up. However, for P1 this reached CSC only at follow up. No other participants met RCI or CSC criteria on this measure.

The FFMQ-15 subscale for observing measured reliable and CSC for three participants (P1, P2 & P3) post-intervention. At follow-up this was maintained by one of these participants (P2). A reliable deterioration on this measure was observed for P4.

For the describe subscale of the FFMQ-15, P1 P2 and P4 showed RCI post-intervention. This met CSC for P2 and for P4. At follow-up, RCI was maintained for P1 and P2 with both showing CSC. However, the score for P4 was no longer reliably improved at follow up. The scores for the remaining participants did not show RCI.

The FFMQ-15 acting with awareness subscale found reliable improvements post-intervention for P2 and P4, both of which met the criteria for clinical significance. At follow up, P4 no longer met either criteria while RCI and CSC for P2 was maintained. P1 also showed RCI and CSC at this time. No further RCI or CSC were observed.

On the non-judging subscale of the FFMQ-15, P2 and P4 showed reliable, clinically significant improvements post-intervention. At follow up P4 no longer met either criteria while RCI and CSC for P2 was maintained. P1 showed reliable and clinically significant change at this time. P3 showed a reliable deterioration post-intervention which was not sustained at follow-up.

On the non-reactivity subscale of the FFMQ-15, only P2 showed reliable and clinically significant improvement post-intervention. At follow-up, RCI and CSC was observed for both P1 and P2 on this measure.

Table 5 - Quantitative scores for each participant indicating RCI and CSC

Participant	Time	AAQ-II	WSAS	CORE-10	FFMQ - O	FFMQ – D	FFMQ – A	FFMQ - NJ	FFMQ - NR
1	Pre	42	27	30	9	4	7	10	6
	Post	24 ^{R+}	10 ^{R+, c}	17 ^{R+}	15 ^{R+, c}	9 ^{R+}	9	10	9
	Follow-up	15 ^{R+, c}	9 ^{R+, c}	13 ^{R+, c}	13	11 ^{R+, c}	11 ^{R+, c}	15 ^{R+, c}	11 ^{R+, c}
2	Pre	34	19	19	7	7	5	6	8
	Post	18 ^{R+, c}	6 ^{R+, c}	12 ^{R+, c}	13 ^{R+, c}	11 ^{R+, c}	11 ^{R+, c}	11 ^{R+, c}	12 ^{R+, c}
	Follow-up	17 ^{R+, c}	6 ^{R+, c}	10 ^{R+, c}	13 ^{R+, c}	13 ^{R+, c}	12 ^{R+, c}	15 ^{R+, c}	12 ^{R+, c}
3	Pre	16	11	13	5	8	11	10	8
	Post	18	20	17	12 ^{R+, c}	8	9	5 ^{R-}	10
	Follow-up	22	12	17	9	5	12	9	9
4	Pre	14	27	10	11	6	9	11	13
	Post	13	27	12	6 ^{R-}	14 ^{R+, c}	13 ^{R+, c}	15 ^{R+, c}	14
	Follow-up	10	-	11	6 ^{R-}	8	8	14	15
5	Pre	39	39	26	9	6	8	9	8
	Post	48	40	27	5	5	6	7	6
	Follow-up	-	-	-	-	-	-	-	-

Lower scores indicate improvements on the AAQ, WSAS and CORE-10. Higher scores indicate higher levels of mindfulness on the FFMQ. FFMQ-O: observing, FFMQ-D: describing, FFMQ-A: Awareness, FFMQ-NJ: Non-judgement, FFMQ-NR: Non-reactivity. R: Indicates reliable change. R+ indicates an improvement, R- indicates a deterioration. - indicates that data was missing for this calculation. c: indicates that criterion C for CSC was met. Post data for P5 were taken from the last date that they submitted data.

3.4 Qualitative Data

The four participants who completed the intervention in full also took part in a change interview about their experience. Using a framework analysis [68], participant responses were coded according to subthemes guided by the questions in the change interview. See Table 6 for a summary of responses. No additional themes emerged. Subsequently, these subthemes were grouped into broader themes of: general experience, helpful and problematic aspects; change; resources and limitations; and research participation.

3.4.1 Theme 1 – General experience, helpful & problematic aspects

Participants' general experience with the app was positive. Participants spoke about how the app facilitated their learning of mindfulness skills in a number of ways such as supporting their development of a routine (P4), clarifying mindfulness concepts (P2) and P1 described the general experience as allowing her to feel more tuned in to how her body and mind work.

Participants identified the fact that meditations were guided as a helpful aspect. Additionally, P2 noted the benefit of recording the frequency of her seizures while taking part in the research. P3 reported the benefits of contact with the researcher highlighting the importance of having somebody to communicate with about FS.

Considering problematic aspects of the intervention, P1 and P2 reported difficulty with the longer sessions which were introduced in the final weeks of the intervention. Additionally, P3 noted difficulties with the accent, he also highlighted that there are a lot of options within the app and that it may have been a struggle to choose an appropriate one without guidance.

Finally, a problematic aspect noted by P4 was that the practice did not have an effect on her seizures as she would have liked.

3.4.2 Theme 2 - Change

All participants reported some change as a result of using the app. P1 and P2 reported an effect on their seizures, highlighting that using the app resulted in “less seizures” and feeling less bothered by them” (P2). Additionally, both noted this reduction had led to improvements in their daily lives to the extent that made it possible for them to continue in their studies. P3 didn’t note any direct change in relation to seizures or well-being but did express that he experienced a greater awareness “an improvement with my conscious appreciation of what's causing problems” (P3). P4 reported feeling more relaxed and having greater control over situations that could otherwise lead to a state of panic. In relation to the likelihood of change, P1, P2 and P4 noted that they did not believe that they would have experienced the changes described without having used the app. Additionally, all participants reported that these changes were unexpected and important.

Participants attributed changes to a range of processes. P1 and P2 placed emphasis on slowing down “Some days I just need to have a day to myself to just relax. And it’s been very useful actually” (P1), “it’s kind of letting myself see things can be calm, things don’t have to go 100 miles an hour all the time and that makes all the difference” (P2). Similarly, P3 highlighted the importance of “timing, patience and temperance” (P3). Whereas P4 noted improvements in her sleeping pattern that led to further benefits in terms of managing stress.

3.4.3 Theme 3 – Resources and Limitations

Considering the personal resources that supported their engagement with the app, participants provided a variety of responses. P1 highlighted the benefits of a supportive partner and family, whereas P2 identified a level of personal determination that supported her to complete the programme. Somewhat similarly, P4 highlighted a level of determination developed through her belief that the app could be beneficial.

Turning to limitations originating in participants' lives that prevented them from engaging with the intervention, P2 highlighted difficulty continuing her practice while staying with friends as she did not feel comfortable to practise meditation in that environment. P3 highlighted that they have a range of other supports in place and noted that engaging fully with the app may displace some of these other supports. P4 reported distraction while trying to follow guided meditations as a difficulty.

3.4.4 Theme 4 – Research participation

Considering the research aspect of taking part in this study, participants highlighted the intensity of completing measures every day (P1). P2 and P4 highlighted the benefits of contributing to the knowledge base around FS. Finally, P3 noted the utility of collecting information as part of the study as this supported him to build an understanding of his own experience and he could share this with healthcare professionals.

Table 6 - Sample responses according to subtheme for each participant

Subtheme	P1	P2	P3	P4
General experience 1	A tool that helps me like, well. It helps me engage in myself more, more in tune with how my body works and how my minds works.	Yeah really good. The more that I practiced doing the mindfulness it just seemed an easier concept.	I don't normally have these mindfulness apps, that style, the smiling mind installed, so having it there and keeping it there is a very conscious reminder to use it.	I would say using the mindfulness, I now use it everyday. Whereas before I'd never used it and now I use it every single day as part of relaxation. Trying to cope with what I am trying to deal with on a day to day basis.
Changes 2	I'm a lot more confident and I'm less reserved now. And I think I'm a lot more calm. When I do have a spell of anxiety or something I do a meditation and calm myself down. And since doing the intervention I'm now able to start doing my teacher training again. Yeah so I started doing my teacher training again, I'm getting my life back really. More independently I've driven to one side of the country this month and pervious month a drive to the other side. I've gone through the whole of lockdown not having had one seizure. And I didn't have one passing out which is quite weird really because there was a lot of stress in the world.	Yeah I suppose less seizures. And also feeling less bothered by them. I've sort of maybe been having like one a week tops. And they hardly last and I'm kind of just not phased by them. And it's just not as scary and I kind of can tell when I'm going to have one now I've started to notice I can kind of say "I've had a really stressful day today, I need to take some time to unwind" because otherwise I know normally if I've had a really busy day I might have a seizure.	I would say that there is an improvement with my conscious appreciation of what's causing problems, in mindfulness, and I know that there are areas and better practice.	Yeah I'm more relaxed. I'm not overwhelmed with everything. I take it as it comes and then deal with it slowly, whatever I have to deal with first. I don't seem to panic as much.

Subtheme	P1	P2	P3	P4
Likelihood without intervention 2	Not very likely at all (laughs). Yeah I kind of hit a rock basically and I didn't know what to do.	So personally I think, I think I'd still be in the same boat if not worse if I hadn't of done it.	I did not expect that change.	Not very likely. In the sense of I think the panics the worries, trying to manage everything would have worsened. I think I'd probably have had to seek medical advice because you know, they wouldn't be able to help. But trying to deal with it myself it wasn't easy.
Expectation 2	I didn't expect any of them at all to be honest. I was like it's gonna be a thing that I have to do every single day and all that kind of stuff and no it's changed quite a lot and everyone's said I hold myself a lot better now I'm confident, I'm becoming more like my old self from before my NEAD and FND.	Honestly, I don't know for sure but I feel like it's massively helped.	I did not expect that change.	Not really. Because I've looked at mindfulness before but in books. I've got relatives, my daughter and my sister have both given me books to read and the reading part of it, not that I didn't understand it, I couldn't do what they asked just reading it.
Importance 2	Yeah definitely a five. Cos in order for me to start my teaching degree I needed to be a bit more confident in myself. Needed to be more relaxed, needed to be able to deal with stress because obviously it's a very stressful job. And it's given me the tools in order to so and you know I'm going in to a classroom next week and I'm so excited instead.	Five out of five, massively important.	When I speak to [the Neurologist], I was mentioning pain and how that was affecting me. That was important at the time, so it brought about significance to the conversation with the neurologist, that was important. So that was a change that was valued, I think.	I would say four, four to five.

Subtheme	P1	P2	P3	P4
Attribution 2	<p>I think with me being more in tune with myself. I know when things are too much and when things are just about right and when things are perfect. I know what to do when things are getting bad and what to do if they're good. I can't put too much stress on myself, I can't. Some days I just need to have a day to myself to just relax. And it's been very useful actually.</p>	<p>Definitely doing all of the mindfulness and I think time aswell. I think there was an element of getting used to things, getting my head around things you know but doing the mindfulness training its just kind of made me pause.</p> <p>The whole situation has just forced me to pause, kind of look at myself and go "I'm going 100 miles an hour here and that's just not manageable, how can I slow down and mindfulness has been one of those things that's given me that space and I feel like when I do it, it's kind of letting myself see things can be calm, things don't have to go 100 miles an hour all the time and that makes all the difference.</p>	<p>Timing, patience and temperance.</p>	<p>Mainly my sleep. Controlling my calmness before I go to sleep is the biggest change because then I wake up not worrying or thinking about what I've got to do with the next day. And that's the biggest cos otherwise if I stress or worry before I go to bed I obviously don't get enough sleep so then I am more stressed or more worries thinking about it, can't relax, trying to deal with it and it just gets worse. The before going to bed is the biggest change for me.</p>
Resources 3	<p>Yeah I mean it's helped a lot in my relationships like with my partner and with my family. Because they got into the habit ... And we've got a lot better of a relationship now and we've got a lot more communication. Then my partner was constantly reminding me everyday "have you done your mindfulness?"</p>	<p>I'm definitely the kind of person who if someone gives me like a challenge or a goal that I have to do it so in that sense having this OK like you have to do this every day for seven weeks. I was like "yep, challenge accepted" I don't like to do things by halves so I really wanted to give it my full.</p>	<p>I've tried to integrate what I'm using into other areas. So I'm trying to what you might say, marry or match the common fascinations of trending use in assessing states of mind and behaviour from online apps which are obviously very convenient and widely available.</p>	<p>I think for me it was knowing that it may make a difference with my whole life. The fact that I'm trying to deal with FND and the fact that it might be able to help with my everyday issues. That was the determination for me.</p>

Subtheme	P1	P2	P3	P4
Limitations 3	I'm not sure on that one actually.	We were staying at a friend's house for one of the nights for a couple of the nights, I didn't feel like I could do it there.	So the one thing that would interfere with this, it would be well, should I displace anything that is already there? This has been, I guess, another reason why I would not actively follow the strict you know, daily or twice a day routine, 'cause I'm doing other things.	Yes, I am easily distracted. Very easily distracted. So trying to remind myself, that's why at bedtime was the easiest time to do it. Whereas trying to get myself to focus on it through the day it was so much harder. So for me distraction was my challenge.
Helpful aspects 1	I think, the one I use the majority of the time is "mindful eating" so like instead of just eating a piece of fruit, I'll take my time eating a piece of fruit and notice where things are going and it's digestive track and where can I feel it in my tummy and the feelings that I feel when I eat such a thing. In fact at the weekend I decided I'm going to have a bar of chocolate and instead of eating it all at once you know, I savoured it, I basically looked at it explored it, ate it, explored it in my mouth, and then I did that and it feels really good to do that and be more mindful without even people knowing.	Definitely helped that it was guided. Recording my seizures everyday, ... so I think that because I was having to remember how many seizures have I had in the last 24 hours, writing it down and then it helped me to remember so then I could actually see like "oh, I haven't had a seizure in a week" and actually seeing that was like really motivating aswell and made me feel like positive that things were getting better cos I could actually see the improvement.	Having someone to communicate, record and create a, a segment of my life, I think is very important. As I said in my feedback in survey there's been zero help and support, and my view is that there is essentially something very close to that value in the NHS.	Yeah the most helpful part of it is knowing how to bring my distraction back to what I'm focusing on. That's the best thing for me. Before when it was talking about trying to stop your mind from wandering and then it talks to you about exploring what your mind is wandering to, what your mind is wandering at and you know what is it thinking of how did it get to that part. And then bringing it back.

Subtheme	P1	P2	P3	P4
Problematic aspects <i>1</i>	No really no, the only thing that can spring to mind is the longer sessions on Smiling mind. Like, sometimes I don't have the time to do the longer sessions. Though it was just the case of "no I've got to do this" and it could be the case if I have to rush to do it and I want to take my own time and, but that was the only thing really.	I think like the longer ones were quite hard. Especially because even though its half an hour or 45 minutes out of your day, I still felt like that is a lot of time. The five to ten-minute ones I could do and it felt realistic but there was just something about doing the half an hour or 45 minute ones that I just really struggled with.	I couldn't get on with the accent... I would have preferred an English female, but that's a matter of preference. I think that that's about the only thing that I could complain about. There was a very diverse list of options and available categories of approach, maybe technique, and you provided some guidance on that, but if I was not given that guidance, I think I would be very lost	I was hoping it would have an impact on the seizures. But saying that, I know things don't happen overnight.
Research <i>4</i>	I'm not going to lie it was very intensive with the questionnaires and stuff. But obviously it has benefitted me a lot. And I've gone from, I've gone through the whole of lockdown not having had one seizure. And I didn't have one passing out which is quite weird really because there was a lot of stress in the world.	Yeah really interesting actually. Especially from cos I'm a student nurse and we look at obviously research papers and stuff so it's been interesting to kind of see how it all comes together in the background. From a you know everyday person perspective. I like OK this is either going to be really helpful to me or, hopefully its going to help loads of people and I felt like contributing to something.	To collect this information as little bite-size surveys, which is very useful to look and reflect on.	It's been really important for me to be a part of it like because of, because I have FND and there isn't really much research regarding it, it's important for me to make a difference or know that I might be helping someone, help towards knowing how it works because obviously it affects everybody so differently and a small contribution that I can make to that, that's really important to me.

Note: Numbers in italics indicate the wider theme that each of these subthemes contributed to.

4. Discussion

4.1 Research questions

This project set out to address five research questions, these will now be considered in turn.

4.1.1 (i) *What effects does a mindfulness training app intervention have on the quality of life of people experiencing FS?*

Results from this study showed that for two participants (P1 and P2), there was an improvement in quality of life upon engaging with the mindfulness intervention used in this study as measured by the WSAS. The change was confirmed by visual analysis, RCI and CSC. Furthermore, it was maintained at 4-week follow up. Qualitative reports from both participants supported this finding.

Conversely, this effect was not detected in the quantitative data for the remaining participants in the study. However, P4 qualitatively reported improvements in her quality of life such as a greater ability to manage stressful situations and improved sleep.

As a result, this study was not able to satisfy the criterion proposed by Kratochwill et. al. [43], that a minimum of three demonstrations of effect should be observed in the visual analysis in order to confirm an effect in SCED designs. It is likely that the small sample size prevented this criterion from being met in full.

4.1.2 (ii) *Does a mindfulness training app intervention reduce seizure frequency in people experiencing FS?*

The quantitative measures in this study did not detect a reliable change in seizure frequency for any participant. While the visual analysis identified an improving trend in seizure

frequency during the intervention phase for P3 and P4, this was at a higher level than was recorded in the baseline phase for each of these participants. Additionally, P1 and P2 reported qualitatively that they were experiencing fewer seizures, however this was not captured by the quantitative seizure frequency measure. Reasons for this discrepancy include the floor effect observed on the daily seizure measure. As participants experienced seizure free days in the baseline phase, descriptive statistics such as the median were at zero pre-intervention and therefore any reduction was difficult to evidence. As the statistics used to detect an effect used in this study rely on this, they were unable to detect an effect in the intervention phase despite participants reporting feeling largely free from seizures in the change interview and neither reported a seizure in the final 11 days of the intervention.

This contrasts with the limited alternative research on mindfulness for FS. For example, Baslet et. al. [36] did find a reduction in seizure frequency following an individual mindfulness-based psychotherapy intervention. However, this only assessed those who completed the intervention and had a high drop-out rate indicating that perhaps those who did not benefit may have left the study early and did not form part of the final analysis.

Similarly, mixed results have been observed in large-scale trials of psychological treatments. For example, a recent large scale RCT investigating the benefits of CBT along with clinical care for FS [22] found benefits on measures of quality of life but no significant improvements in seizure frequency. While seizure frequency is the most common measure in FS research [69], it has not been standardised and is not statistically reliable. One proposal to address this has been the application of the clinical global impression scale [70]. However, this scale is based on the professional judgement of clinicians rather than the experience of people with

FS and may cause further difficulty in a population who often feel misunderstood by healthcare professionals [3] and would not be practicable in a design such as the present study.

4.1.3 (iii) Does a mindfulness training app intervention have an effect on levels of psychological distress for people experiencing FS?

Similar results to the quality of life measure were noted for this measure (CORE-10), with P1 and P2 showing CSC improvements on the distress measure at both post-intervention and follow-up. Conversely, these changes were not seen for P3-5. Again, this shows some benefits for psychological distress for these participants, but it does not meet the Kratochwill et. al replication criteria [43].

The variety in the results obtained connects to a wider issue in this field of research, that of a completely heterogenous group. As noted in the introduction, theories on the aetiology of FS are far from conclusive and though people with FS may present on the surface with the same difficulty i.e. seizures, the aetiology and wider experience of each individual can vary dramatically. This was reflected in the differing responses to the intervention and has been noted in previous intervention studies with people with FS [22].

4.1.4 (iv) Does a mindfulness training app intervention lead to a reduction in experiential avoidance and increase in mindfulness in people experiencing FS?

This study investigated process measures of experiential avoidance and mindfulness. Similar to the findings for other research questions, reliable change for the AAQ-II measure was

noted for P1 and P2. This was clinically significant at follow up for both participants. Similar changes were not observed for the remaining participants.

We can see that for the two participants who showed improvement on the quality of life and distress measures, there were also improvements on the AAQ-II. P1 and P2 scored above the clinical cut-off range of over 24 suggested for the AAQ-II [23] before taking part in the intervention and scored below this cut-off afterwards while other participants did not score above this cut off at any point. This improvement for P1 and P2 in occurred alongside the improvements in quality of life and distress. This suggests that a reduction in experiential avoidance may have been a key process that played a part in the improvements experienced by these participants. Indeed, this was hypothesised in the literature [24] on FS, noting that increased awareness and turning towards experience may support people experiencing FS to develop greater awareness in these situations and thus identify a supportive response. Additionally, qualitative reports from these participants reflect some of these ideas. For example, a comment from P2 shows a change in managing seizures that reflects a change in experiential avoidance; “I’m kind of just not fazed by them. Whereas before I’d be like “oh my gosh I’m going to have a seizure I’m going to die.” Whereas now I’m like “that’s annoying, go away please.” This finding adds to the emerging evidence around the role that experiential avoidance may play in FS [26]. Therefore, the result may be an indication that mindfulness or interventions focused on similar processes may be more beneficial for those experiencing FS who present with high levels of experiential avoidance.

However, this finding is somewhat limited by the measure used in this study. Whilst the AAQ-II was developed to measure the construct of psychological flexibility and experiential

avoidance [23], and has been used in a large number of studies investigating third-wave interventions [71], the discriminant validity of this measure has questioned by Tyndall et. al. [72] suggesting that the measure is more akin to a measure of both distress and experiential avoidance. Nonetheless, the measure captures processes that are central to third-wave therapies and has been used in this study in the absence of a well-established alternative.

Considering mindfulness, P1 and P2 showed reliable and CSC on a number of mindfulness facets measured by the FFMQ-15. Additionally, P4 displayed reliable and CSC on the awareness, describe and non-judging facet on the measure while showing a reliable deterioration on the observing facets. Furthermore, P1 experienced reliable and significant change on the observe facet. While the effects of the intervention appeared to vary across participants, RCI and CSC was observed in at least three participants on all of the facets in the FFMQ-15 with the exception of non-reactivity.

This indicates that practising mindfulness using this app leads to improvements in mindfulness traits. The benefits experienced by participants in this study may therefore be linked to improvements in these such as increased awareness and less reactivity [33], meaning that they have an improved awareness of and control over their seizure scaffold as defined by Brown and Reuber [15]. In large scale studies, changes in each of these factors have been associated with improvements in well-being [73]. While in the present study, two participants (P1 and P2) showed improvements in mindfulness facets, QoL and distress, other participants (P3 and P4) who showed reliable change on some mindfulness facets did not display similar outcomes. Indeed, they may have experienced some benefits that were not

quantitatively measured in the study. Both participants spoke about benefits such as improved sleep (P4) and awareness (P3).

4.1.5 (v) How do people with FS experience a mindfulness training app intervention?

This question is answered by drawing on the themes generated in the qualitative analysis.

Regarding the acceptability of the intervention, the results show that for all of these participants there was a degree of acceptability of support delivered in a way that embraces technology. Additionally, the completion rate was higher in the present study compared with traditional mindfulness interventions in this area [36]. Both of these factors show promise for the integration of technological solutions into healthcare treatment of those experiencing FS. Participants highlighted the guidance and structure that the app provided as a particular strength. Additionally, telephone support throughout the intervention was welcomed by participants.

However, participants identified some difficulties with the app. Addressing these may be useful for the development of similar interventions. Particular suggestions revolved around the length of sessions. Participants noted that ten minutes felt about right for them.

4.3 Limitations

This study took place in the context of the covid-19 pandemic. This was a time of change, stress and crisis for the world as a whole. While little is known about the impact of this on people with FS, large scale research has shown that it has resulted in increased mental health

difficulties in the general population [74]. Given the global nature of this pandemic, it is likely to have affected some of the outcomes for participants in the course of this study.

The AB design utilised in this study presented limits to internal and external validity. These include the number of replications, more of which would have increased the external validity. Additionally, while a degree of randomisation was achieved as participants started the baseline and intervention at different points, the study did not utilise a specific randomisation procedure which would have increased the internal validity of the study.

A further limitation in this study has been the role of the researcher. As the lead researcher handled all aspects of recruitment, weekly contact and follow up interviews with participants, there is potential for this to result in a degree of response bias [75]. It would have been preferable to involve an independent researcher to conduct the change interviews.

4.4 Future Work

4.4.1 Clinical implications

This type of intervention was acceptable to participants, as a result there may be scope to develop further interventions delivered in a similar manner for this population. Specifically, given the benefits to some participants and the lack of harm to others, along with the success of other supported, remotely delivered interventions for people with FS [27, 76, 77], there may be some justification for developing self-directed therapeutic supports such as apps or books based on third-wave approaches that are specifically designed for those with FS. It may be possible to use interventions such as these with people who are interested while on a

waiting list for further interventions, as part of a stepped care model, or as a model of intervention for people who find it difficult to attend appointments.

4.4.2 Future research.

Along with other studies of mindfulness in FS [17, 36], this study has shown that for some individuals experiencing FS, mindfulness practice may provide benefits to quality of life and psychological distress. However, given the mixed results from this and other studies, it is evident that this is not an intervention that will benefit all people with FS.

The variability in response to the intervention in this study and others shows the need for greater research into the profiles of people experiencing FS and a need to match these with appropriate treatment. Therefore, a key recommendation for the future is further investigation into specifically who will benefit from what intervention. Within FS there is considerable variation in individuals' presentations and aetiology [17]. Building this understanding may facilitate the matching of individuals with appropriate treatments. Indeed, a focus on processes such as experiential avoidance rather than on the diagnosis as a whole may be useful. Such research could be guided by theoretical constructs of FS such as the integrative cognitive model [15].

4.5 Conclusion

While this study cannot claim to have identified strong evidence for the treatment of FS using a specific approach, it has shown that for some individuals with experience of FS that mindfulness-based interventions may be useful. Additionally, this study highlights the role

that experiential avoidance may play in some individuals with FS suggesting that interventions that reduce high levels of experiential avoidance may be beneficial. Furthermore, the study shows that an intervention delivered using smartphone technology can enable people with FS to learn mindfulness skills and is broadly acceptable by those experiencing FS.

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Appendix 2 - A Demonstration of calculations

Example of working out trend, level and stability for participant one on the daily WSAS measure within phases

Description	<u>Within conditions</u>	
	Baseline	Intervention Phase
Total number of data points	7	49
Mean	22.43	15.9
Median	22	18
Range	16-27	9-24
Stability Envelope	22 x 0.25 = 5.5 Envelope = 22 ± 5.5 16.5 – 27.5	
Percentage of points in stability envelope	100%	65%
Relative level change (median second half – median first half)	+2 deterioration	-9 improvement
Absolute level change (first value – last value)	-5 improvement	-14 improvement
Mid date first half	2	19.5
Mid date second half	6	44.5
Mid rate first half	20	19
Mid rate second half	22	10
Stability	71.42%	95.12%
Direction	deteriorating	improving

Between conditions

Aspect	Description
Stability	The data was variable in the baseline phase and stable in the intervention phase
Trend	Baseline: accelerating – deteriorating Intervention: decelerating - improving
Absolute level change	First value of the intervention – last value of the baseline = +2 deterioration
Relative level change	Median of first half of the intervention – median of second half of the baseline = -3 improvement
Median level change	Median of the intervention phase – median of the baseline phase = -4 improvement
Mean level change	Mean of the intervention – mean of the baseline = -6.53 improvement
Percentage of non-overlapping data	Lowest value of baseline = 16 Number of values equal to or below 16 in the intervention phase = 22/49 Number of values above 21/number of sessions x 100 = 22/49 x 100 = 44.9%
Percentage of overlapping data	Lowest value of baseline = 16 Number of values equal to or above 16 in the intervention phase = 27/49 Number of values on or below 16/number of sessions x 100 = 27/49 x 100 = 55.1%
Effect size; percentage exceeding the mean	Median of baseline = 22 Number of intervention data points below 22 = 42 Value from earlier step divided by the number of data points in the intervention phase; 42/49 = 0.8571 Multiply value in earlier step by 100; 0.8571 x 100= 85.71% Therefore, the percentage of data points exceeding the median was 85.71%.

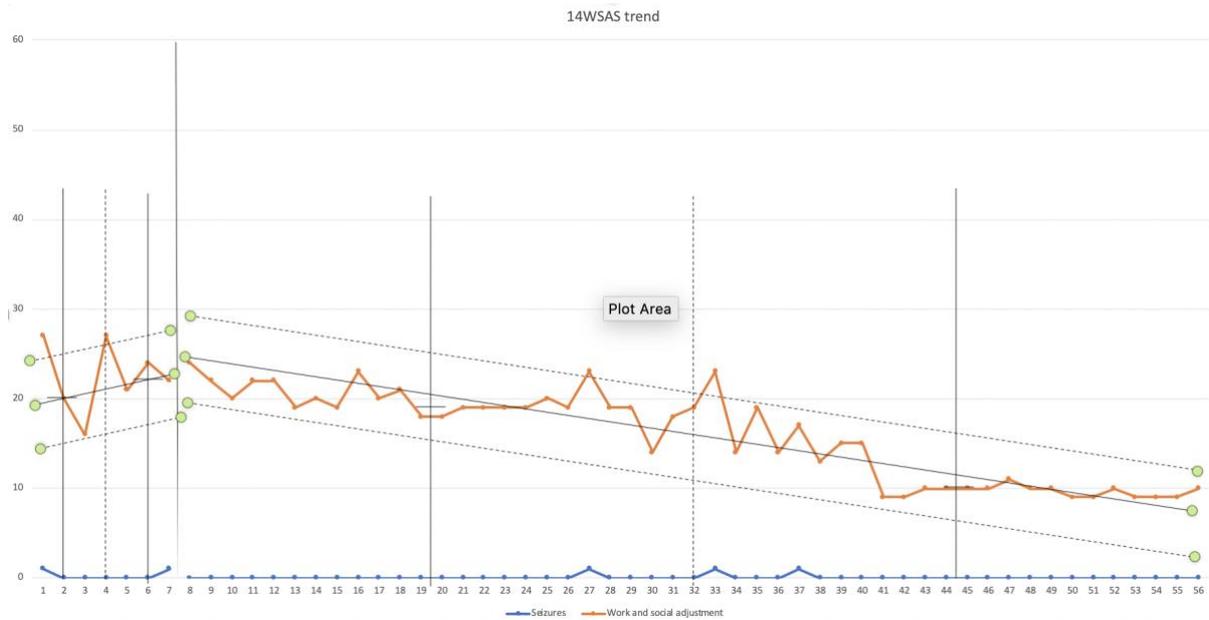


Figure showing trend lines applied using the “split middle” method – each phase was divided in half. The median of each half was marked on the mid-point of each half and a line connecting these points was the trend line. This was moved to a point where there were an equal number of data points above and below the line. The dotted lines represent the stability envelope applied to the trend line.

RCI calculation	
Standard error of the mean (SEM)	$12.15 \times \sqrt{0.16}$
Formula: $SD \times \sqrt{(1 - r)}$	12.15×0.4
r = reliability of measure	= 4.86
Standard error of difference (S_{diff})	$\sqrt{2 \times 4.86^2}$
Formula: $SE_{diff} = \sqrt{2 \times SEM^2}$	$\sqrt{2 \times 23.62}$
	$\sqrt{47.24}$
	= 6.87
RCI formula: (post treatment score – pre treatment score/ S_{diff})	$(24 - 42 / 6.87)$
	= -2.62
Is the score greater than ± 1.96 ?	Yes. Therefore, RCI criteria is met

CSC calculation	
Formula: (SD functional population x M dysfunctional population) + (SD dysfunctional population x M functional population) / (SD functional population + SD dysfunctional population)	$(7.52 \times 28.98) + (12.15 \times 18.53) / (7.52 + 12.15)$
	$443.07 / 19.67$
	= 22.5

Example of RCI and CSC calculations

Calculations used data from the AAQ-II measure for P1

Section 3 – Critical Appraisal

Critical appraisal of the thesis

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Formatted for submission to the Epilepsy & Behaviour (Author Guidelines attached in Appendix 1C)

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1. Introduction

This is a critical review of the project. Firstly, the key findings of the thesis will be presented. Following this, a background section reflects on my reasons for settling on this research project. In doing so, it will cover points relevant to the thesis as a whole. Following this, the review will proceed through sections focusing on the systematic review portion of the thesis. It will then consider the empirical paper before concluding with some potential future research and general remarks on the thesis.

2. Key results of the thesis

The systematic review was focused on the experience of stigma amongst people with a diagnosis of functional neurological disorder (FND). A systematic search retrieved 13 papers and a meta-ethnography identified four themes: delegitimization; excluded, isolated and abandoned – the social cost of stigma; the cost of attempts to manage stigma; and threats to identity and the meaning of mental health. The review highlighted the experience of stigma for individuals with an FND diagnosis and the effects that stigma has on their daily lives.

The empirical paper investigated the benefits of a mindfulness training app for functional seizures (FS). Five participants were recruited with support from a third-sector organisation. Results showed some variation, with two participants showing benefits from the intervention across measures of quality of life, distress, experiential avoidance and mindfulness in the study and a further two who completed the intervention not showing significant benefits on the measures used, the fifth participant did not complete the intervention and did not show improvements before departing the study. However, all participants who completed the

intervention described it in a positive light and also commented on aspects of it that could be improved.

3. Why this research?

I have always been attracted to integrating ideas, to looking at things from different, sometimes opposing perspectives and recognising the benefits that each position brings. This may have roots in my professional background having come from a Social Science informed background to one grounded in Psychology. I also enjoy the idea of being pragmatic and of research having some real-world impact. FND brought together a lot of these threads, the condition itself being one that connects ideas about the body, the brain and the mind in an often complex and overlapping nature. I was excited by the idea of getting involved in research in this area.

Again, my choice of method for the empirical paper reflected these threads – integrating both quantitative and qualitative methods through a single case experimental design respected both schools of thought and allowed for a much richer understanding in an intervention study compared with relying solely on psychometric measures. As well as this, focusing on the role of stigma in the review crossed a number of fields of interest for me including social and psychological factors that impact upon health and wellbeing.

4. An issue relating to both papers

As I looked into this area, it became clear that language was an important factor to consider. This was apparent for a number of reasons including the variety of language used to describe

FND conditions and also the history and stigma associated with the terms used to describe people experiencing FND [1]. While my initial reaction to this difficulty was to adopt the terminology used in some of the most recent, widely cited research – psychogenic non-epileptic seizures (PNES) [2, 3], it quickly became apparent in my discussions with FNDHope (a patient-led FND advocacy group) that this term was considered to be stigmatising and that it placed blame upon people experiencing FND for their condition. This was largely due to the fact that the term implied pure psychological cause for the condition in the “psychogenic” part of the title and also defined the condition by exclusion in the “non-epileptic” part of the title. In the development of the research project, I became aware that people who experience functional seizures often experience stigma [4] and that the name used can serve to promote this. As a result, the term functional seizures (FS) utilised by FND advocacy groups was adopted for the study. In managing this dilemma, I experienced first-hand the importance of language and how researchers have the ability to influence the wider narrative through the language adopted. I felt that this was a very important lesson learned in undertaking this research.

5. Issues relating to the systematic review

In order to undertake the systematic review a meta-ethnographic [5] approach was adopted. This is a well-known approach that has frequently been applied to healthcare topics [6]. It was adopted for this review as it has the ability to build a “whole” from the sum of the parts of qualitative papers that reported on stigma in FND to a greater or lesser degree. Syntheses of qualitative research have been both lauded and critiqued. One frequent critique of this type of review is the fact that the process involves the combination of studies that approach research from differing traditions with differing underlying philosophical assumptions [7]. However, in focusing on qualitative research this review brought together studies from a

similar philosophical orientation. A further critique of this method relates to the quality appraisal score. Some argue that imposing a quality appraisal score on qualitative research involves the imposition of positivist principles upon research that draws from the opposing philosophical standpoint [8] with its concerns for the unique experience of the individual at its core. However, in undertaking the systematic review it felt important to conduct a quality analysis in order to demonstrate a degree of consistency, highlighting the quality of the papers that made up the synthesis. In order to manage this dilemma, the ratings were displayed for the reader who may decide how they wish to apply them to their understanding of the review rather than adopting them to rule studies in or out of the review. Additionally, this approach facilitated a check that no theme relied solely on evidence from papers with lower ratings. This approach I feel served this purpose and did not fall in to the territory of uncritical adoption of such a technique [8]. While other methods may be appropriate, the review displays a clear strength in that it clearly outlines the search strategy and method that was used in order to arrive at the result. This approach contrasts with less rigorous narrative reviews that do not report this information [9].

As noted above, language and terminology are important issues when considering FND. This became apparent when conducting the literature search. To date, there is no consensus on terminology for FND or the conditions associated with it. Indeed, this is reflected in the evidence base that uses a variety of terms for FND conditions. This presented a challenge when undertaking the search. As can be seen in the strategy, this involved including a variety of potential search terms for FND, drawing on strategies used in other reviews in order to ensure that none were missed. While this led to a time-consuming phase of reviewing papers for eligibility for the review, I believe it was necessary in order to ensure the parameters of the search matched the aims of the review.

In conducting the review, at times I felt a tension between sociological and psychological constructs in developing a response to stigma. For example, when considering the wider implications of the review I was drawn to make pragmatic suggestions about what should be done to address stigma that clearly causes ongoing difficulty for a lot of people. In the report I focused on the societal changes or shifts that are required. However, I did not draw within myself to formulate these difficulties using psychological models. Indeed, psychological models exist that would actively support some of the experiences reported by participants as a result of the stigma they encountered. For example, at the core of compassion focused therapy [10] there is a focus on dealing with shame. While this may be useful clinically on an individual basis, it does not address the source of the problem. In recognising my position as a researcher on the topic of stigma it felt incredibly important not to perpetuate the idea that the problem and solution is located within the individual. Stigma is a societal issue and it felt greatly important to use the platform and position that research brings in a way that promotes positive social change rather than perpetuating the idea of personal responsibility in the face of social difficulty. I felt that this was an important lesson in my development as a researcher and the embedding of ethics within my research practice.

6. Issues relating to the empirical paper

The empirical paper used a single case experimental design (SCED) methodology. A clear benefit of using this method was the ability to integrate the qualitative experience of participants into the study. The lived experience of participants who take part in research investigating treatment methods and techniques tends to be lacking from the majority of research into psychological treatments in favour of psychometric measures. Indeed, this has

sometimes been considered an antithesis to quantitative evaluation [11]. While measures ensure a degree of standardisation, the individuals taking part in these studies are not “standardised” in the same way and will often have a diverse range of responses to the treatment. In taking this approach the study sought to bridge the gap between idiographic and nomothetic approaches to research, recognising the strengths of both quantitative and qualitative data, viewing both as complimentary rather than as dyadic, an approach that has been argued for elsewhere [12]. As a result, the strengths of this study revolve around the ability of the design to identify change on an individual level, this being something that can be obscured by larger group designs [13].

Considering the ethics of the study, safety was a key concern throughout. As can be seen in the proposal, a clear risk protocol was in place for the study. Thankfully, this served its purpose, and no participants reported any harm in the course of their participation. However, inspecting the data from P4, there are days where they experienced a large number of seizures. This was quite an alarming finding. However, P4 did not report any increased distress during the weekly check-in telephone calls throughout the study. I followed up with this participant upon discovering the number of seizures that she was experiencing, and she reported that they were not causing significant distress and included a large number of very short absence type seizures.

A further ethical issue raised by this study was the use of a payment for participants. This initially came about due to concerns in the design stage regarding the amount of time required of participants to take part in the study. The ethical debate around payment has been raised time and again [14] with the key concerns that this may coerce people in to taking part.

However, it has also been acknowledged that failure to provide remuneration for time spent on a project may raise ethical concerns [14]. The payment in this case was intended to compensate for the time that participants spent completing measures on a regular basis throughout the study. In developing the proposals for this, Health Research Authority (HRA) guidance was followed [15] to ensure that the payment was not coercive, was available to all participants including those who withdrew from the study and did not encourage risk-taking.

Given the timing of the study, the covid-19 pandemic had a significant impact. Firstly, recruitment plans were affected. The initial plan for recruitment had been to make links with local branches of third sector support groups and visit meetings of support groups in order to recruit to the study face-to-face. However, this was not possible due to the outbreak and restrictions that existed throughout and indeed beyond the recruitment and data collection phases of the study. As a result, the recruitment strategy was changed to recruit online. This proved quite difficult as at the time, many organisations who may have been interested in promoting the study appeared to have staff on furlough or were not operational in the usual manner. While one organisation was incredibly helpful in listing the study, recruitment proceeded quite slowly. There were a number of enquiries which did not result in potential participants taking up a place in the study. Quite understandably this was a time where the population as a whole experienced a degree of shock and change in their lives and taking part in a study which demanded a consistent degree of participation may have been more difficult than at other times. The aim was originally to have six participants as had been the norm in some similar studies [16]. While six participants were recruited, full datasets were only available for four as one participant withdrew, and another became ineligible. Indeed, despite six participants being frequently used in this type of study, several similar studies have been published with 2-4 participants [17-19]. Therefore, owing to the difficulties recruiting at this

time, it was decided to proceed with the four full datasets obtained. In doing so, the study met the criteria for methodological soundness advocated by the what works clearing house (WWC) guidance on SCED design [20] by making “at least three attempts to demonstrate an intervention effect at three different points in time or with three different phase repetitions [20 p. 15].”

A limitation in relation to recruitment was the possibility of sample bias. As recruitment was online, this limited the possibility of taking part to those who have sufficient IT skills and engage with the organisation that advertised the study. Additionally, the recruitment approach and time involved in the study meant that only those who had a high level of motivation and ability would take part. Furthermore, participants self-confirmed their diagnosis to the researcher through their application to take part in the study rather than seeking a video electroencephalogram confirmed diagnosis which has been advocated by some in FS research [3]. These issues could potentially be addressed in future research by recruiting via the NHS.

Adherence was an issue in the study. While recommendations were made to participants about the amount of mindfulness practice that they should complete, failure to adhere to the guidance was not used to exclude them from the study at any point. There may have been an argument to exclude data from P3 for example who showed low adherence completing only one session on some weeks. An alternative approach to managing this may have been to omit data from participants who did not complete a pre-determined number of sessions. However, including these data may actually add some information around what is realistic for participants to manage with this intervention. Indeed, the frequency of between-session practice in models such as cognitive behavioural therapy (CBT) is variable with research

showing that this is not always completed as planned [21] and that higher completion rates are associated with greater outcomes in therapy. Indeed, for the participants who benefitted most from this intervention, the rate of adherence was high.

If I were to run a similar study in future, I might reconsider the measures used. Firstly, considering the measure of seizure frequency (reported daily), despite two participants reporting qualitatively that they had experienced a reduction in seizure frequency, this was not detected using the visual methods used. This was likely due to a floor effect as some participants reported experiencing zero seizures in the baseline phase which meant that any improvement beyond that point could not be detected. Additionally, it was not possible to apply reliable change and clinically significant change to this measure as statistical data are not available for a self-report seizure measure. Indeed, it has been argued elsewhere that seizure frequency alone may not be the most useful outcome measure for FS when used in isolation [22]. Furthermore, recent reviews have identified that there is an absence of reliable outcome measures available specifically for FND conditions [23, 24]. A useful reliable and valid measure might integrate aspects of the other measures used here such as the work and social adjustment scale [25], taking into account both the frequency and potentially the duration of seizures along with the impact that they have on daily life. A further note on the measures was raised in the empirical paper in relation to the use of the acceptance and action questionnaire (AAQ-II) [26]. This instrument was developed to measure core concepts of experiential avoidance and psychological flexibility which are central to the practice of third-wave cognitive therapies such as acceptance and commitment therapy (ACT) and mindfulness. Underlying theories from these models note that practising mindfulness and ACT skills results in individuals developing greater freedom in choosing how they wish to respond to thoughts and experiences [27] and throughout the development of third-wave

therapies, the AAQ-II has been a central measure [28]. However, the measure has faced criticism in relation to its discriminant validity [29, 30].

Similarly, the methods of analysis can be critically analysed. It is worth noting that SCED and the established methods of data analysis were developed primarily to establish evidence for behavioural interventions. In particular, the methods such as those in the suite proposed by Lane and Gast [31] are perhaps more effective at detecting an effect in scenarios where an immediate change is hypothesised compared with an intervention-based study such as this one which involves participants who learn a skill or make a change over time. There is no specific point in time in a mindfulness or psychotherapeutic intervention where it can be said that the conditions are completely in place. This contrasts with behavioural interventions such as reinforcement schedules which may be implemented and removed instantly. Indeed, through analysing data early in the intervention phase, this may in fact weaken the power of these methods to detect a change. For example, Lane and Gast's [31] method seeks to confirm an instant effect. Conversely, it would be hypothesised that the impact of mindfulness training or psychotherapy would not be evident until after participants have completed the course of therapy or the intervention [32]. It may be useful to consider adapting the study design and measuring different phases in this case. For example, obtaining a baseline measurement, proceeding to deliver the intervention and measuring again once the skills have been learned and applied. However, this approach is not evident in the literature.

Alternative visual methods are available such as the Fisher et. al. [33] conservative dual criterion (CDC) method which is reported to have increased accuracy and is less likely to report a type 1 error. However, the decision was taken to proceed with the Lane and Gast [31]

method due to the comprehensive nature of this method incorporating both within and between condition analysis, the fact that it is widely used elsewhere (e.g. [16, 34, 35]) and finally, as the guidance for the CDC method only provides direction for datasets containing up to 23 points which the intervention phase in the present study exceeded. Additionally, the CDC method shares the traits described in the previous paragraph.

Turning to the results, this study broadly found two demonstrations of effect on the key outcome measures. As noted in the empirical paper, this does not meet the criterion set out in the what works clearing house document [20] which specify that for an effect to be shown in a SCED, three replications must be displayed. In the analysis of the study I recognised that it did not demonstrate an effect in accordance with these standards as there were two out of the four participants who completed the intervention that showed some effect. In doing so I wished to uphold quality standards. However, it should be recognised that other research has been published reporting an effect with fewer than three demonstrations [18]. Indeed, guidance on this is a little unclear, despite WWC [20] acknowledging that an effect may be detected without three replications, the paper does not describe how this may be achieved and notes that its' authors may decide whether or not a study constitutes evidence without defining how they would go about this. I felt that this raised some issues in relation to what we can consider evidence, particularly from a small-scale study such as this. As a consequence of the WWC guidance it felt that the study was in limbo on some level – that it did not show efficacy on one hand according to the guidance. Yet, it could not be said to be ineffective given the positive reports and data from two participants.

7. Future Directions

This thesis adds to two important areas in relation to FND – the evidence base for treatments and the social understanding of the condition. In doing so it opens up a variety of avenues in terms of future research.

Firstly, the systematic review identified stigma as an added difficulty that shapes the experience of those experiencing FND in a variety of ways. As identified in the review paper, further qualitative studies that explicitly focus on this would be beneficial in order to gain a deeper understanding of this experience. Grounded theory may be particularly useful to build a greater understanding of stigma with the aim of finding reliable methods to tackle the problem.

Considering the empirical paper, this adds to the literature building on the importance of third wave ideas of psychological flexibility and experiential avoidance in the make-up of functional seizures. Future studies may build on this through adding to the literature about what type of intervention works for whom. Considering the identification in this study of reducing experiential avoidance as a process that resulted in benefits for participants, ideas from process-based therapy [36] which changes the focus of intervention from the diagnosis to key processes that maintain difficulties, may be useful for FS given the variety of people who present with this difficulty. Indeed, using the integrative cognitive model as a guide to identify relevant cognitive processes and subsequently developing evidence for interventions targeting these may be a suitable approach to managing the problem that heterogeneity amongst this group brings to developing evidence-based interventions.

8. Conclusion

Overall, on a personal level this project has provided a steep learning curve in a variety of ways. It has challenged me to learn new research methods, to learn about a condition in-depth and to apply research to very real-world questions in a pragmatic fashion. I feel that in undertaking this project I have learned a set of skills that will support me in developing practice-based evidence throughout my career in clinical psychology.

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Section 4 – Ethics

Ethics application and information

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Doctorate in Clinical Psychology

Lancaster University

Formatted for submission to the Epilepsy & Behaviour (Author Guidelines attached in Appendix 1C)

Word count (including abstract but excluding references, appendices, figures and tables):

3,765

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1. Ethics Form

Faculty of Health and Medicine Research Ethics Committee (FHMREC)

Lancaster University

Application for Ethical Approval for Research

for additional advice on completing this form, hover cursor over 'guidance'.

Guidance on completing this form is also available as a word document

Title of Project: A case series investigating the benefits of a mindfulness training smartphone application for non-epileptic attack disorder (NEAD).

Name of applicant/researcher: Ciarán Foley

ACP ID number (if applicable)*:

Funding source (if applicable)

Grant code (if applicable):

***If your project has *not* been costed on ACP, you will also need to complete the Governance Checklist [\[link\]](#).**

Type of study

Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. **Complete sections one, *two* and four of this form**

Includes *direct* involvement by human subjects. **Complete sections one, *three* and four of this form**

SECTION ONE

1. Appointment/position held by applicant and Division within FHM Trainee Clinical Psychologist,
Faculty of Health and Medicine, Doctorate in Clinical Psychology

2. Contact information for applicant:

E-mail: c.foley@lancaster.ac.uk **Telephone:** 07378325400 (please give a number on
which you can be contacted at short notice)

Address: 639a Burnage Lane, Manchester, M19 1TF.

**3. Names and appointments of all members of the research team (including degree where
applicable)**

Research Supervisor

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Field Supervisor

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Royal Hospital, Salford, M6 8HD

Tel: +44 (0)161 206 4694, e-mail: antonia.kirkby@srft.nhs.uk

3. If this is a student project, please indicate what type of project by marking the relevant box/deleting as appropriate: (please note that UG and taught masters projects should complete **FHMREC form UG-tPG**, following the procedures set out on the [FHMREC website](#))

PG Diploma Masters by research PhD Thesis PhD Pall. Care

PhD Pub. Health PhD Org. Health & Well Being PhD Mental Health MD

DClInPsy SRP [if SRP Service Evaluation, please also indicate here:] DClInPsy Thesis

4. Project supervisor(s), if different from applicant:

Research Supervisor

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Field Supervisor

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5. Appointment held by supervisor(s) and institution(s) where based (if applicable): noted above.

SECTION TWO

Complete this section if your project involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants

1. Anticipated project dates (month and year)

Start date:

End date:

2. Please state the aims and objectives of the project (no more than 150 words, in lay-person's language):

Data Management

For additional guidance on data management, please go to [Research Data Management](#) webpage, or email the RDM support email: rdm@lancaster.ac.uk

3. Please describe briefly the data or records to be studied, or the evaluation to be undertaken.

4a. How will any data or records be obtained?

4b. Will you be gathering data from websites, discussion forums and on-line 'chat-rooms' n o

4c. If yes, where relevant has permission / agreement been secured from the website moderator? n o

4d. If you are only using those sites that are open access and do not require registration, have you made your intentions clear to other site users? n o

4e. If no, please give your reasons n/a

5. What plans are in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.

6a. Is the secondary data you will be using in the public domain? no

6b. If NO, please indicate the original purpose for which the data was collected, and comment on whether consent was gathered for additional later use of the data.

n/a

Please answer the following question *only* if you have not completed a Data Management Plan for an external funder

7a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

n/a

7b. Are there any restrictions on sharing your data?

n/a

8. Confidentiality and Anonymity

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications? yes

b. How will the confidentiality and anonymity of participants who provided the original data be maintained?

All data reported in the thesis and any journal articles based on the data will be anonymised

9. What are the plans for dissemination of findings from the research?

10. What other ethical considerations (if any), not previously noted on this application, do you think there are in the proposed study? How will these issues be addressed?

SECTION THREE

Complete this section if your project includes *direct* involvement by human subjects

1. Summary of research protocol in lay terms (indicative maximum length 150 words):

This study aims to investigate the use of an app-delivered mindfulness intervention with participants experiencing functional seizures (FS). With this condition, people experience seizures that do not have the same profile of electric activity as epileptic seizures. At present there is little evidence for any particular treatment for the condition and people experience it as extremely disabling; affecting work, family and social life. Additionally, some people with this condition find it difficult to engage with traditional healthcare.

Mindfulness involves paying attention to the present moment in a non-judgemental way and mindfulness-based treatments have been shown to be beneficial for a range of physical and psychological issues. The intervention in this study will involve people experiencing functional seizures completing mindfulness training guided by a smartphone app over an 8-week period. The study will assess a potentially helpful intervention delivered outside a traditional healthcare environment.

The study will measure the effect that the intervention has on seizure frequency and quality of life for participants. Further measures of distress, mood and psychological flexibility will investigate some of the processes that might lead to change in for people experiencing this condition. Participants will also be invited to share their experience of the intervention.

2. Anticipated project dates (month and year only)

Start date: February 2020

End date March 2021

Data Collection and Management

For additional guidance on data management, please go to [Research Data Management](#) webpage, or email the RDM support email: rdm@lancaster.ac.uk

3. Please describe the sample of participants to be studied (including maximum & minimum number, age, gender):

Between three and six participants aged 18 or over of any gender who self-report having a diagnosis of FS will be recruited to the study. Participants will be required to speak English to a level where they are comfortable completing mindfulness teaching through English. Participants will also need to have access to a smartphone or tablet in order to access the intervention. The study will aim to recruit between three and six participants. The minimum number of participants for this type of study to meet the standard set by Horner et al. (2005) is three. However, four is preferable (Kratochwill et al., 2010) and similar studies have set a precedence of six (Barrett-Naylor et al., 2018). Recruitment will continue on a rolling basis until a full dataset from six participants is obtained. Alternatively, recruitment will stop in October 2020 if the minimum number of three participants has been reached. A dataset will be considered full where participants have completed a minimum of 50% of the intervention and corresponding measures along with a change interview and follow-up measures. Participants will be recruited through FS support groups. Ciarán will approach relevant support organisations such as FNDHope and online support groups including; NEAD UK, NEAD caregivers and NEAD Awareness.

Exclusion criteria for this study will include addiction, psychosis and suicidality. Mindfulness is usually a safe activity but may be experienced as distressing by some in these groups (Kostanski & Hassed, 2008). People with an active mindfulness practice at present will also be excluded from the study, as will those who are currently engaging in psychotherapy. People with a comorbid diagnosis of epilepsy will also be excluded from this study. Participants will be screened by telephone or internet communication service using the demographics and screening questionnaire.

As this study involves a lot of time on the part of participants, a payment in the form of a voucher will be provided to them at the end of their participation in the study. This is discussed in further detail below.

4. How will participants be recruited and from where? Be as specific as possible. Ensure that you provide the *full versions* of all recruitment materials you intend to use with this application (eg adverts, flyers, posters).

Recruitment

Ciaran will contact a number of support organisations and groups that have an interest in NEAD/PNES Ciarán will make contact with groups supporting those with NEAD including FNDhope, FNDAction, NEAD UK, PNES caregivers and PNES awareness.

- Ciarán will create social media accounts based on the poster (e.g. <https://twitter.com/CiaranFoley19>). These will be used to communicate with groups concerned with NEAD.

- Ciarán will make contact with each group via their preferred contact method (e.g. phone, e-mail, online form, message via social media) to explain the study and gauge their interest in supporting recruitment.
- Ciarán will provide each group with a copy of the poster containing information about the study and contact details.
- Ciarán will request that they share with their members through any available channels including e-mail, newsletter, social media, at support groups or through pre-existing panels who are interested in research.
- These groups will be contacted one at a time (one per week) in order to manage potential demand and to ensure that the researcher has time to respond appropriately to all interested parties.
- If these groups meet in person, Ciarán will offer to attend a group meeting to describe the study to potential participants.
- Anybody interested in participating in the research can make contact with Ciarán via e-mail. Ciarán will provide an information to participants via e-mail and will discuss any further requests for information with participants by phone or e-mail if required.

When a participant makes contact by e-mail or through direct contact via social media, Ciarán will then contact the potential participant by e-mail or phone to explain more about the study and to answer any questions. If participants are still interested, they then will be provided with a participant information sheet via e-mail. Participants who wish to continue with the study at this point will complete the screening questionnaire with Ciarán by telephone. Participants will be informed whether or not they meet the study criteria over the telephone.

Participants who meet the inclusion criteria for the study will be invited to participate. They will be sent a link by text or e-mail to the Qualtrics page where they can consent to participate in the study.

Potential participants who do not meet the criteria for the study will be provided with a list of relevant support agencies. Any data gathered from these individuals will be deleted once this has been provided to them.

Participants will be asked whether they would prefer to receive reminders and links to questionnaires by text or by e-mail and this preference will be used throughout the study unless a participant changes their preference which they can do by informing the researcher during a weekly check in call or by e-mail at any point during the study.

5. Briefly describe your data collection and analysis methods, and the rationale for their use.

Materials and collection

A small battery of questionnaires will be collected daily for one week before the intervention and daily throughout the intervention. These will include a self-report measure of seizure frequency (number of seizures experienced in a day) and the 5-item work and social adjustment scale (Mundt, Marks, Shear, & Greist, 2002). This measure was chosen for its psychometric properties, ease of completion and its ability to capture changes in functioning.

Additional measures used in this study will be collected pre- and post-intervention, weekly throughout the intervention and at one-month follow-up.

The 7-item acceptance and action questionnaire version 2 (Bond et al., 2011) will be used to measure psychological inflexibility and experiential avoidance. Both have been shown to play a role in NEAD (Dimaro et al., 2014) and mindfulness has been shown to act on both processes (Hooper, Villatte, Neofotistou, & McHugh, 2010; Moore & Malinowski, 2009).

The 15 item five factor mindfulness questionnaire (Gu et al., 2016) measures different aspects of mindfulness and will be used to assess whether participants develop mindfulness traits through using the app.

The CORE-10 (10 items) (Barkham et al., 2013) will be used to monitor participants' levels of distress throughout the study. This measure will also collect data on mood. Both may affect the quality of life of people with NEAD.

A brief change interview (Elliott, 2010) administered by phone post-intervention will assess the participants' experiences of the intervention, qualitative benefits from the intervention and any difficulties with the intervention.

Demographic and clinical information for each participant will also be recorded: age, gender, ethnicity, occupational status, partnership status, time since diagnosis, other health conditions and medication.

All measures will be collected using Qualtrics online tools. Participants will be sent a link by text or e-mail requesting that they fill in the measures as required. See links below;

Qualtrics information:

Consent: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_3KoXv9TOKPC6skZ

Weekly measures: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_e99C35uuaRdFm05

Daily measures: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_25WPKn7Vr7tTe4J

Procedure

Baseline Phase

During the baseline phase, participants will complete measures of work and social adjustment (WSAS) and seizure frequency on a daily basis for one week. Participants will receive a prompt each day with a link to complete this questionnaire. Participants will also receive a prompt to complete weekly measures. Following this, Ciarán will check the trend of the participant's data to ensure that its trend is not already towards a reduction in symptoms and increase in functioning. This is an essential step in ensuring the internal validity of the study (Morley, 2017). If it is stable, they will begin the intervention phase. If this is not stable, the baseline phase will be extended in three-day increments until it reaches stability. Participants will complete the baseline phase at different times. It is anticipated that this will happen naturally due to the rolling nature of recruitment in this study. However, if a number of participants sign up at the same time, their baseline periods will be staggered in order to adhere to the design standards for the study.

Intervention

Participants will begin completing the exercises in the Smiling Mind application. They will complete these in the following order;

- Week 1: Complete 6 "introduction to mindfulness" guided sessions.
- Week 2: Complete the first set of 7 sessions of the "mindfulness foundations programme"
- Week 3: Complete the second set of 7 sessions of the "mindfulness foundations programme"
- Week 4: Complete the third set of 7 sessions of the "mindfulness foundations programme"
- Week 5: Complete the fourth set of 7 sessions of the "mindfulness foundations programme"
- Week 6: Complete the fifth set of 7 sessions of the "mindfulness foundations programme"
- Week 7: Complete the sixth set of 7 sessions of the "mindfulness foundations programme"

Participants will be asked not to go past the advised schedule for completing the intervention until after their participation in the study is complete. They will be advised that if they wish to do any additional practice that they can repeat any part of the program that they have completed already.

Participants will ideally complete one session each day during the intervention phase. In the event that a participant does not manage to complete it on one day, they can complete two the next day.

During the intervention phase, participants will complete daily measures throughout. Participants will receive a prompt each day with a link to complete this questionnaire. Participants will also receive a prompt to complete weekly measures once per week.

Follow up Phase

Four weeks after they have completed the intervention, the researcher (Ciarán) will contact each participant to complete a brief change interview (Elliott, 2010). These interviews will be recorded and transcribed. Participants will also be invited to complete one final set of both the daily and weekly measures at this time.

Analysis

Daily measures will be analysed through visual analysis according to guidance from Lane and Gast (2014). This analysis involves plotting all data points from the daily measures on to a graph. From this an analysis for each participant will be conducted to assess the change between the baseline and the intervention phase. Trend, and stability will also be analysed using this method. This will answer the research questions: What effects does a mindfulness training app intervention have on quality of life of people with NEAD? Does a mindfulness training intervention delivered through an app reduce seizure frequency in people with NEAD?

The remaining quantitative data will be analysed using a reliable change index analysis and measures showing reliable change will be further examined through clinically significant change statistics (Jacobson & Truax, 1991). A cut off for reliable change and clinically significant change will be calculated for each measure and each participants data will be examined to see if their scores exceed the cut-off. This approach has been shown to be useful in small sample research of this nature (de Souza Costa & De Paula, 2015). This analysis will answer the research questions; Does a mindfulness training app intervention lead to a reduction in experiential avoidance? Does a mindfulness training app intervention have an effect on levels of distress in people with NEAD?

Change interview responses will be transcribed and summarised in table format. A brief content analysis (Smith, 2000) will identify key themes from the interviews. These data will be used to support or to challenge the quantitative data collected in the study and will add the subjective experience of participants using the intervention answering the final research question; How do people with NEAD experience a mindfulness training app intervention?

At the end of their participation in the study, participants will be provided with a voucher for a maximum of £41.05. Where a voucher cannot be obtained in a round amount, the total due will be rounded to the nearest available sum. See below for further discussion on this proposal.

6. What plan is in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc.)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.

7. Will audio or video recording take place? no audio video

a. Please confirm that portable devices (laptop, USB drive etc) will be encrypted where they are used for identifiable data. If it is not possible to encrypt your portable devices, please comment on the steps you will take to protect the data.

The audio recording device will not be encrypted. However, directly after each interview, the audio file will be transferred to the secure University server and saved in an encrypted file.

b What arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?

The encrypted data will be stored on the secure University server until the thesis is examined. After this point, the files will be deleted by Ciarán.

Please answer the following questions *only* if you have not completed a Data Management Plan for an external funder

8a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

After the study is concluded, the research supervisor Fiona Eccles will manage the data in line with University policy for 10 years. Only the quantitative data and interview transcripts will be retained.

8b. Are there any restrictions on sharing your data?

This research project will involve a small sample. As a result there is a small risk that participants may be identifiable through their responses. As a result data will not be publicly available but any requests to share data will be considered on a case by case basis and this will only be granted to genuine researchers.

If any participant requests a copy of their data, this will be provided to them by the researcher at the end of their participation in the study

9. Consent

a. Will you take all necessary steps to obtain the voluntary and informed consent of the prospective participant(s) or, in the case of individual(s) not capable of giving informed consent, the permission of a legally authorised representative in accordance with applicable law? yes

b. Detail the procedure you will use for obtaining consent?

A participant information sheet will be provided to all participants outlining the requirements on their part for participation in the study. Additionally, the researcher will discuss this in further detail with each participant. Participants will be informed of their right to withdraw from the study.

Consent will be recorded via the completion of an online consent form that participants will complete through Qualtrics via the following link;

https://lancasteruni.eu.qualtrics.com/jfe/form/SV_3KoXv9TOKPC6skZ

10. What discomfort (including psychological eg distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.

This intervention is aimed at improving the psychological wellbeing of participants and is not anticipated to cause significant distress.

Mindfulness is generally considered a safe activity and this study uses a low-intensity mindfulness programme. Some people describe the risks involved in mindfulness as being similar to exercise (Baer & Kuyken, 2016). This comparison says that exercise is good for people's health but can be dangerous at times where we over-do it. Similarly, more intense mindfulness training may be more risky for people who have not practised it before. The intervention used here is a low-intensity mindfulness training app that is widely available to the public. This reduces the potential risk of participation.

Ciarán will have weekly contact with participants which will allow him to check in with participants around any adverse reactions that they may have to the intervention. If a participant experiences difficulty as a result of the intervention they will be signposted to the relevant support agency and the researcher will have a discussion with them about their continued participation in the study.

Participants will also be given advice on the Participant Information Sheet who to contact if they are experiencing distress as a result of their participation in the study.

Participants may withdraw from the study at any time. As noted elsewhere, participants who choose to withdraw from the study will remain eligible for a voucher in recognition of the time that they have spent completing the study. Participants will be able to withdraw their data up to one week after it has been submitted. Following this period, participant's data may be withdrawn if the researcher finds that it is possible to do so without compromising the study.

11. What potential risks may exist for the researcher(s)? Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).

It is not anticipated that any lone working will be required over the duration of this study with the exception of potentially attending a local support group run by a community organisation.

All contact with participants and organisations will be made through the researcher's University e-mail account, a University mobile phone for research purposes or via social media using an account created specifically for the study by the researcher linked with his University e-mail address.

12. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.

It has been well demonstrated that mindfulness can provide a range of health and wellbeing benefits. Participants may find that the intervention provides some benefits to their lives such lower levels of distress, an increase in day to day awareness, and an increased ability to engage with thoughts and feelings. There is a possibility that the intervention could have a positive effect on the frequency of participant's seizures but this is not guaranteed. Indeed, it may have no effect at all. We hope that the information gained from this study will help us to understand the nature of these seizures and approaches to treating them in the future.

13. Details of any incentives/payments (including out-of-pocket expenses) made to participants:

Due to the burden on the part of participants a reward is required to encourage participation. Participants will be required to complete a part of the intervention daily for 8 weeks, complete

measures every day for over 8 weeks, have a weekly phone call and a follow up phone call and complete weekly measures. It will not be possible to recruit adequately for the study without this funding. Precedence has been set in this area by other universities that have experience in supporting this type of study e.g. (Barrett-Naylor et al., 2018; Jinks, 2016; Roche, 2016). In providing this payment, the study will abide by the protocols outlined in the research ethics guidebook in the compensation of participants (Economic & Social Research Council, 2019).

The maximum payment of £41.05 to be provided has been calculated using the current living wage as a guide (living wage of £8.21 x a maximum time spent completing measures and interviews of 5 hours = £41.05). As a result the payment is not considered coercive or as placing undue coercion on participants. The provision of this payment also aims to avoid being paternalistic in nature through the provision of a choice of retailers from which participants may choose to receive a voucher.

The researcher will provide this voucher to each participant at the conclusion of their involvement in the study. In seeking to address this issue, cash would be the preferable option. However, having sought advice on this it appears that because of the University's finance department regulations this is not feasible in this study. A voucher will be provided to each participant. Participants may choose a voucher from; Amazon, Tesco, ASDA, Sainsburys or Lovetoshop. The voucher will be provided to each participant by the researcher at the end of their participation in the study. Participants who withdraw from the study will be provided with a voucher reflecting the time that they spent taking part in the study. For example, if a participant withdraws after completing 20% of the total study, they will be provided with a voucher for 20% of the maximum payment.

14. Confidentiality and Anonymity

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications? yes

b. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.

Anonymity of participants will be maintained throughout this study. Participants will be randomly allocated a participant identification number. An encrypted file containing participants' names and corresponding ID numbers will be stored in a separate file on the University's secure server.

o Once collected, all data will be moved from the Qualtrics platform to the University's encrypted server.

o Audio recordings will be deleted once the project has been examined.

o The typed version of the interview will be made anonymous by removing any identifying information including participants' names. Anonymised direct quotations from interviews may be used in the reports or publications from the study. As a result confidentiality cannot be guaranteed but anonymity will be as far as is possible.

o Participants will be informed of the limits of confidentiality through the following caveat: “if what is said during our contact makes me think that you, or someone else, is at significant risk of harm, I will have to break confidentiality and speak to a member of staff about this and potentially notify the relevant authorities if necessary. If possible, I will tell you if I have to do this.”

15. If relevant, describe the involvement of your target participant group in the *design and conduct* of your research.

A panel of people with a diagnosis of NEAD was consulted in the design of this research project. In doing this they stated that they valued this piece of research in to the area. Furthermore, highlighted that the participant materials provide a clear and concise guide to what is required of participants. A final point they highlighted related to ensuring that people who do not have any experience of NEAD gain some exposure of NEAD and the difficulties associated with it. This final point will be addressed through the dissemination of the research findings through publication, conferences, presentations at the doctorate in clinical psychology thesis presentation day and at appropriate conferences.

16. What are the plans for dissemination of findings from the research? If you are a student, include here your thesis.

The findings of this study will be submitted as a thesis for the Doctorate in Clinical Psychology. Findings will be presented to staff, students and the Lancaster University Public Involvement Network (LUPIN) at a thesis presentation day. It is hoped that findings will be submitted to a relevant journal, for example “Seizure.” Findings will be communicated to all groups that have had contact with the study through recruitment or promotion of the study and participants who are interested will be provided with a copy of the findings. Findings may also be presented at conferences, special interest groups and training events.

17. What particular ethical considerations, not previously noted on this application, do you think there are in the proposed study? Are there any matters about which you wish to seek guidance from the FHMREC?

We recognise that in the provision of an intervention, this study may be different in nature to others that have been conducted as part of the DClinPsy programme. We would appreciate any feedback about this that the panel feel is important. Additionally, we welcome feedback about the proposal to provide a payment to participants and whether they believe this has been managed in an ethical fashion.

SECTION FOUR: signature



Applicant electronic signature:

Date

Student applicants: please tick to confirm that your supervisor has reviewed your application, and that they are happy for the application to proceed to ethical review

Project Supervisor name (if applicable):

Date application discussed

Submission Guidance

1. **Submit your FHMREC application by email to Becky Case (fhmresearchsupport@lancaster.ac.uk) as two separate documents:**
 - i. **FHMREC application form.**

Before submitting, ensure all guidance comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line*.
 - ii. **Supporting materials.**

Collate the **following materials for your study, if relevant, into a single word document:**

 - a. **Your full research proposal (background, literature review, methodology/methods, ethical considerations).**
 - b. Advertising materials (posters, e-mails)
 - c. Letters/emails of invitation to participate
 - d. Participant information sheets
 - e. Consent forms
 - f. Questionnaires, surveys, demographic sheets
 - g. Interview schedules, interview question guides, focus group scripts
 - h. Debriefing sheets, resource lists

Please note that you DO NOT need to submit pre-existing measures or handbooks which support your work, but which cannot be amended following ethical review. These should simply be referred to in your application form.

2. Submission deadlines:
 - i. Projects including direct involvement of human subjects **[section 3 of the form was completed]**. The *electronic* version of your application should be submitted to **Becky Case by the committee deadline date**. Committee meeting dates and application submission dates are listed on the [FHMREC website](#). Prior to the FHMREC meeting you may be contacted by the lead reviewer for further clarification of your application. Please ensure you are available to attend the committee meeting (either in person or via telephone) on the day that your application is considered, if required to do so.

- ii. The following projects will normally be dealt with via chair's action, and may be submitted at any time. **[Section 3 of the form has *not* been completed, and is not required]**. Those involving:
 - a. existing documents/data only;
 - b. the evaluation of an existing project with no direct contact with human participants;
 - c. service evaluations.
- 3. **You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application**

Appendix A – Ethics approval letter



Applicant: Ciarán Foley
Supervisor: Fiona Eccles
Department: DHR
FHMREC Reference: FHMREC19142 (amendment to FHMREC19037)

18 August 2020

Re: FHMREC19142 (amendment to FHMREC19037)

A case series investigating the benefits of a mindfulness training smartphone application for non-epileptic attack disorder (NEAD).

Dear Ciarán,

Thank you for submitting your research ethics amendment application for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for the amendment to this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

A handwritten signature in blue ink that reads "E. Suri-Payer".

Dr. Elisabeth Suri-Payer,
Interim Research Ethics Officer, Secretary to FHMREC.

Appendix B – Participant information sheet

Participant Information Sheet

A case series investigating the benefits of a mindfulness training smartphone application for non-epileptic attack disorder (NEAD).

For further information about how Lancaster University processes personal data for research purposes and your data rights please visit our webpage:
www.lancaster.ac.uk/research/data-protection

My name is Ciarán Foley and I am conducting this research as a trainee clinical psychologist as part of the Doctorate in Clinical Psychology at Lancaster University, United Kingdom.

What is the study about?

The purpose of this study is to investigate whether a smartphone-based mindfulness training app provides any benefits for people with experience of non-epileptic attack disorder (NEAD).

Why have I been approached?

You have been approached because you are part of a group providing support to people experiencing NEAD/PNES. The study requires information from people who have a diagnosis of NEAD or PNES who are interested in taking part in research on a mindfulness app.

Do I have to take part?

No. It's completely up to you to decide whether or not you take part.

What will I be asked to do if I take part?

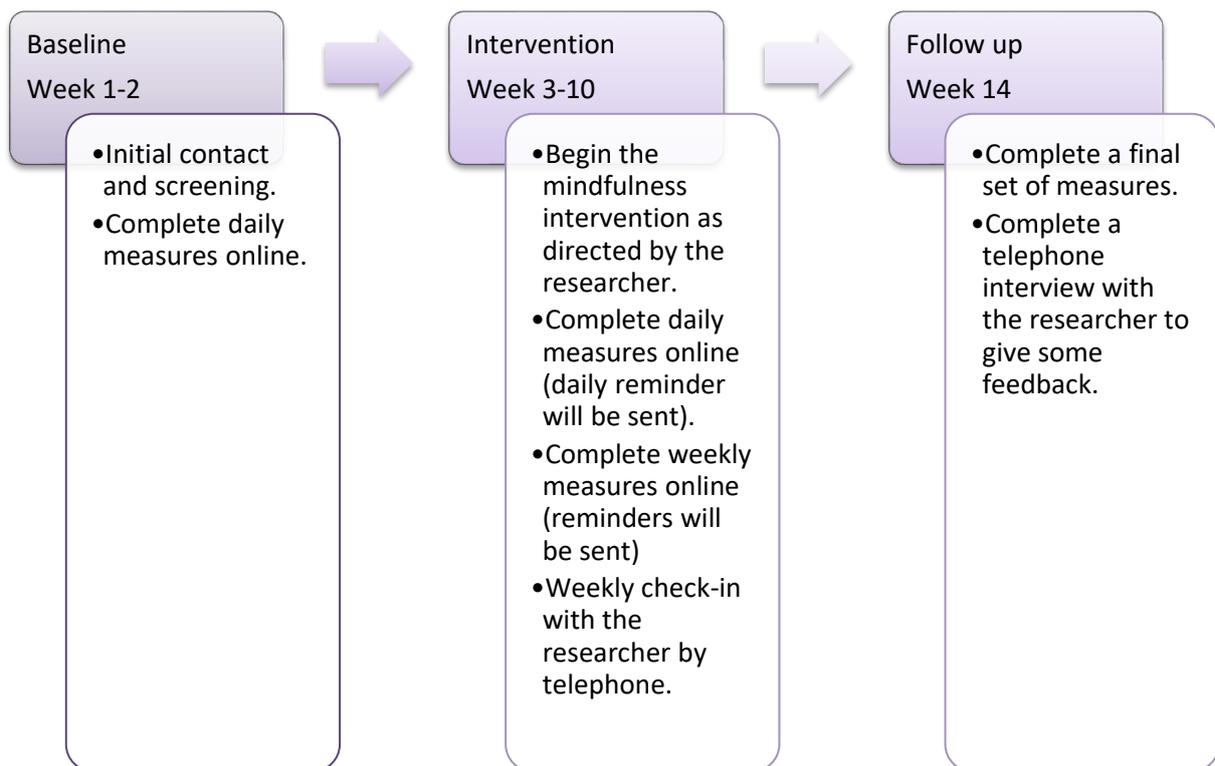
If you decide you would like to take part, your main involvement will be to complete a mindfulness training programme over an 8-week period and submit responses to questionnaires on a daily and weekly basis for a week before the intervention, during the intervention itself and 4 weeks after you have finished. The mindfulness app is called "Smiling Mind" and is widely available online and through the Apple "app store" and the Google "play store." The app provides a range of mindfulness training programmes and this study will focus on two of these. If you might want to take part, please do not engage with this app before the study begins as doing so could interfere with the results.

The study will involve completing exercises from the app each week. You will also have weekly phone contact with the researcher where you can discuss your experience and any issues that may arise from your engagement with the intervention. As part of the study, you will be asked to complete 2 brief questionnaires every day and also up to 3 questionnaires once each week. All questionnaires will be submitted online and can be completed using a smartphone, tablet or a computer. Daily questionnaires can be completed in approximately 1 minute and weekly questionnaires that will take approximately 5 minutes to complete.

Before you start the study, you will be asked to complete a screening interview by telephone. If you do not meet the screening criteria, then you will not be able to take part in the study and any information gathered about you will be deleted. If you do meet our eligibility criteria you will be invited to complete some more brief questionnaires. After 1 week of completing the daily and weekly measures, you will receive directions to begin using the app. From this point on you will continue to complete the daily and weekly measures and receive telephone support each week from the researcher.

Your active participation in the study is estimated to take 10 weeks and you will be contacted 4 weeks after this time to complete a follow up questionnaire online and a brief interview over the phone. The measures used in this study can be completed in a couple of minutes (less than 2 minutes for the daily questionnaire and 10 minutes for the weekly questionnaires). The time required to complete the mindfulness training will vary. The app provides a 6-session introduction to mindfulness lasting 15 minutes in total and a 42-session foundation course with exercises ranging from 1-40 minutes with an average duration of 9 minutes. We anticipate that completing the mindfulness training and completing the questionnaire will take an average of ten minutes each day.

Below is a flowchart outlining the main phases of the study;



The main procedure for taking part in the study is as follows in some more detail;

Contact the researcher by e-mail to express your interest in taking part in the study leaving a phone number and times that are suitable to contact you by phone



The researcher will provide you with a copy of this info sheet via e-mail



The researcher will be available by telephone or e-mail to discuss the study in more detail



If you wish to take part, you will be invited to complete a brief telephone interview with the researcher to see if you meet the screening requirements and to gain some background information about you.



If you meet the criteria for the study and wish to take part, you will be directed to a website where you can consent to participate in the study.



You will be sent a link to complete a weekly questionnaire online. You will be sent a link to complete this each week during your participation in the study.



For the next week you will be sent daily questionnaires to complete each day. You will receive your preference of an e-mail or a text message reminder to complete these along with a link to the website where they can be completed.



After one week, you will begin the mindfulness intervention. The researcher will direct you to complete a set number of exercises from the app each week. The researcher will contact you once per week to check in around your experience of the intervention.



This will be repeated for the next 7 weeks meaning that you will have completed 8 weeks in total of mindfulness practice along with daily and weekly questionnaires.



4 weeks after finishing the intervention you will be asked to complete the questionnaires once more and you will be invited to take part in a telephone interview with the researcher to discuss your experience of taking part in the study.

Expenses and payments

In recognition of the time commitment for participation in the study, you will be provided with a voucher worth a total of £40.75 for a retailer of your choice from a selection from Amazon, Tesco, ASDA, Sainsburys or Lovetoshop.

If you wish to withdraw from the study, you will still be entitled to a voucher. This will be calculated pro-rata for the time you have spent completing the study. For example a person who withdraws after completing half of the things required by the study will receive half of the total payment.

You should note that acceptance of this voucher may bring liability for tax or could count as income towards your means assessment for benefits. We strongly recommend that you check this before accepting the payment by contacting HM Revenue & Customs on 0300 200 3300 and/or your local job centre plus on 0800 169 0310 to discuss this before accepting the voucher. Additionally, you have the right to decline this voucher if you so wish and still participate in the study.

What will happen if I wish to withdraw from the study?

You have the right to withdraw from the study at any time after beginning to take part. If you do choose to withdraw from the study, the information already collected from you may not be erased and may be used in the analysis of the project.

Will my data be identifiable?

The information you provide is anonymous. The data collected for this study will be stored securely and only the researchers conducting this study will have access to this data:

- Audio recordings from the follow up interview will be deleted once the project has been examined.
- The files on the computer will be encrypted (that is no-one other than the researcher will be able to access them) and the computer itself password protected. These will be deleted 10 years after the project has been examined.
- The typed version of your interview will be made anonymous by removing any identifying information including your name. Anonymised direct quotations from your interview may be used in the reports or publications from the study, so your name will not be attached to them.
- All your personal data will be confidential and will be kept separately from your interview and survey responses.

There are some limits to confidentiality: if what is said during our contact makes me think that you, or someone else, is at significant risk of harm, I will have to break confidentiality and

Speak to a member of staff about this and potentially notify the relevant authorities if necessary. If possible, I will tell you if I have to do this.

What will happen to the results?

The results will be summarised and reported in a thesis and may be submitted for publication in an academic or professional journal or presented at conferences. I will also give you a copy of the results if you would like to see them.

Are there any risks?

There are minimal risks anticipated with participating in this study. Participation in this study does require that you give up some of your time. It is important that during the study you maintain a healthy life balance making time for the things that are important in your life. It is important to note that in rare cases people may find the mindfulness exercises difficult. If this comes up, the researcher will discuss possible support options for you and will have a discussion with you about your participation in the study going forward.

The study is an experimental study, and it is hoped that it will bring some positive changes for you. However, this is not guaranteed. Therefore, it is important for you to note that there may be no positive changes in the frequency of your seizures as a result of your participation.

Are there any benefits to taking part?

You may find that the intervention provides some benefits to your life such as your level of distress, your awareness and your ability to engage with thoughts and feelings. There is a possibility that the intervention could have a positive effect on the frequency of your seizures but this is not guaranteed. Indeed, it may have no effect at all. We hope that the information gained from this study will help us to understand the nature of these seizures and approaches to treating them in the future.

Who has reviewed the project?

This study has been reviewed and approved by the Faculty of Health and Medicine Research Ethics Committee at Lancaster University.

Where can I obtain further information about the study if I need it?

If you have any questions about the study, please contact the main researcher:

Researcher

Ciarán Foley

Trainee Clinical Psychologist, Doctorate in Clinical Psychology, Faculty of Health and Medicine, Lancaster University, Lancaster, LA1 4YT

email: c.foley@lancaster.ac.uk

Tel: [0750 840 6248](tel:07508406248)

Research Supervisor

Dr Fiona Eccles

Research Tutor, Doctorate in Clinical Psychology, Faculty of Health and Medicine, Lancaster University, Lancaster, LA1 4YT

Tel: +44 (0)1524 592807, email: f.eccles@lancaster.ac.uk

Field Supervisor

Dr Antonia Kirkby

Consultant Clinical Neuropsychologist, Neuropsychology Lead for Non-Epileptic Attack Disorder Service, Salford Royal Hospital, Salford, M6 8HD

Tel: +44 (0)161 206 4694, email: Antonia.kirkby@srft.nhs.uk

Complaints

If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:

Ian Smith Tel: (01524) 592282

Research Director, Doctorate in Clinical Psychology; Email: i.smith@lancaster.ac.uk

Faculty of Health and Medicine

Lancaster University

Lancaster

LA1 4YT

If you wish to speak to someone outside of the Clinical Psychology Doctorate Programme, you may also contact:

Professor Roger Pickup Tel: +44 (0)1524 593746

Associate Dean for Research Email: r.pickup@lancaster.ac.uk

Faculty of Health and Medicine

(Division of Biomedical and Life Sciences)

Lancaster University

Lancaster

LA1 4YG

Thank you for taking the time to read this information sheet.

Mindfulness and Functional Seizures



I am a trainee Clinical Psychologist currently enrolled on the Doctorate in Clinical Psychology in Lancaster University. I am seeking participants with experience of functional seizures also known as psychogenic non-epileptic seizures (PNES) or non-epileptic attack disorder (NEAD) who are interested in practising mindfulness using a smartphone app. My study is investigating whether this form of mindfulness training can provide any benefits for functional seizures.

If you decide to participate, you will be invited to complete a mindfulness training programme using a smartphone app over an 8-week period. You will be asked to complete a series of questionnaires for the duration of the study along with an interview over the phone 4 weeks after you've finished.

The study takes part over a long period of time and in recognition of the time spent participating, we can offer participants a voucher for **£41.05** in recognition of the time spent contributing to the research. Participants who meet the criteria for the study and are interested in taking part will be selected to do so on a first come first served basis.

To be eligible for the study, potential participants should be:



- Over 18 years old.
- Fluent in English.
- Have access to a smartphone or tablet.
- Diagnosed with FS/NEAD/PNES



- Not currently diagnosed with epilepsy.
- Not currently experiencing difficulty with addiction.
- Not already practicing mindfulness daily

If you would like more information please contact:

Ciarán Foley by e-mail at c.foley@lancaster.ac.uk

The project is supervised by Dr Fiona Eccles, who can be contacted by e-mail:
f.eccles@lancaster.ac.uk

Appendix D – Demographics and screening questionnaire

Demographics and screening questionnaire;

Name:

Date of birth:

Phone number:

Gender:

Ethnic background:

Occupation:

What stage of education did you reach?

Relationship status:

How long have you been experiencing symptoms of NEAD?

When were you diagnosed with NEAD?

What treatments have you had for NEAD to date?

Are you experiencing any other health difficulties?

Have you received any other mental health diagnosis?

Are you taking any medications at present?

Do you currently experience epilepsy?

Have you been diagnosed with epilepsy in the past?

Are you currently engaged in any form of psychotherapy?

Have you had any psychological treatment or psychotherapy in the past?

Do you practice mindfulness regularly?

Do you have access to a smartphone or laptop?

Are you confident in your ability to use a smartphone app and to complete online questionnaires with support?

Have you experienced any episodes of psychosis recently?

Have you recently experienced a dependence on any substance?

Have you had any thoughts recently about ending your life?

Appendix E – Consent form

I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

Yes

No

I confirm that I have read the information sheet and fully understand what is expected of me within this study

Yes

No

I confirm that I have had the opportunity to ask any questions and to have them answered.

Yes

No

I understand that my interview at the end will be audio recorded and then made into an anonymised written transcript.

Yes

No

I understand that data I submit through surveys will be anonymised and reported on.

Yes

No

I understand that audio recordings will be kept until the research project has been examined.

Yes

No

I understand that once my data has been submitted, I may withdraw it within one week. After this, data may be withdrawn if deemed possible by the researcher.

Yes

No

I understand that the information from my interview will be anonymised and may be published; all reasonable steps will be taken to protect the anonymity of the participants involved in this project.

Reasonable steps will be taken to protect the anonymity of the participants involved in this project.

Yes

No

I consent to my survey data and quotations from my interview being used in publications, reports, conferences and training events.

Yes

No

I understand that I will be provided with a voucher at the end of my participation in this study. If I withdraw from this study early, I understand that this voucher will reflect the proportion of the study that I have completed.

Yes

No

I understand that any information I give will remain confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the principal investigator will need to share this information with their research supervisor.

Yes

No

I consent to Lancaster University keeping the data from my questionnaires, consent forms and written transcriptions of the interview for 10 years after the study has finished.

Yes

No

I consent to take part in the above study.

Yes

No

Name

→

Powered by Qualtrics 



Appendix F – Resources for Participants

Resource List

Resources in the event of distress

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance;

If you need immediate help:

In an emergency:

- Call 999
- Go to your local A&E department

If you're in crisis and need to speak to someone:

- Call NHS 111 (for when you need help but are not in immediate danger)
- Contact your GP and ask for an emergency appointment
- Contact the [Samaritans](#) (details below)
- Use the '[Shout](#)' [crisis text line](#) - text SHOUT to 85258

Samaritans

Available 24 hours a day to provide confidential emotional support for people who are experiencing feelings of distress, despair or suicidal thoughts.

- www.samaritans.org
- **116 123** (free to call from within the UK and Ireland), 24 hours a day
- Email: jo@samaritans.org

NEAD support groups;

FND Dimensions

Facilitate support groups in partnership with NHS services throughout the UK.

www.fnddimensions.org

FNDHope

Provide advice and information and lobby on behalf of people with NEAD.

www.fndhope.org

FNDAction

Provide advice and information and lobby on behalf of people with NEAD.

www.nonepilepticattackdisorder.org.uk

Other sources of advice and support

Mind

Mind offers advice, support and information to people experiencing a mental health difficulty and their family and friends. Mind also has a [network of local associations in England and Wales](#) to which people can turn for help and assistance.

Lines are open Monday to Friday 9am to 6pm (except bank holidays).

- www.mind.org.uk
- **InfoLine: 0300 123 3393 to call, or text 86463**
- Email info@mind.org.uk

Campaign Against Living Miserably (CALM)

A helpline for men in the UK who are down or have hit a wall for any reason, who need to talk or find information and support.

- www.thecalmzone.net
- **Helpline for men: 0800 58 58 58**
- Webchat: www.thecalmzone.net/help/webchat/

5pm to midnight, every day of the year

Sane

SANE services provide practical help, emotional support and specialist information to individuals affected by mental health problems, their family, friends and carers.

- Support Forum: www.sane.org.uk
- **Saneline: 0300 304 7000** (local rate on BT landlines) Open 4:30-10:30pm every day

NHS mental health services

Find information, advice and local services on the [NHS website](#). You can also get [advice from the NHS 111 phone service](#).

Appendix G – Links to further information

Links to consent and questionnaires on Qualtrics

Consent: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_3KoXv9TOKPC6skZ

Weekly measures: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_e99C35uuaRdFm05

Daily measures: https://lancasteruni.eu.qualtrics.com/jfe/form/SV_25WPKn7Vr7tTe4J

Link to information about the mindfulness training programme

www.smilingmind.com

<https://apps.apple.com/gb/app/smiling-mind/id560442518>

https://play.google.com/store/apps/details?id=com.smilingmind.app&hl=en_GB