

## CASE STUDY



# Cognitive behavioural therapy for psychogenic nonepileptic seizures (PNES) in an adult with a learning disability: A case study

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## Funding information

Oxford Health NHS Foundation Trust

## Abstract

**Background:** Psychogenic nonepileptic seizures (PNES) are attacks that often look like epileptic seizures (ES). Unlike ES, PNES are not triggered by underlying neurological factors. PNES are relatively rare in the general population; however, their prevalence in people with learning disabilities is much higher—up to 10%. Individuals with PNES and learning disabilities often have psychiatric comorbidities, such as anxiety disorders, depression and posttraumatic stress disorder. There are currently no treatment guidelines for PNES in people with or without learning disabilities. However, talking therapies, such as cognitive-behavioural therapy (CBT), are recommended. This study explores the effectiveness of CBT for the treatment of PNES in a woman (Susan) with PNES and learning disabilities.

**Methods:** We used a single-case experimental design (SCED) to investigate the effectiveness of CBT intervention. Specifically, the withdrawal (also known as ABA) design was applied. The patient's average PNES frequency, as well as idiosyncratic mood measure and standardised outcome measures for people with learning disabilities (Glasgow Anxiety Scale for People with an Intellectual Disability and Mini-Maslow Assessment of Needs Scale-Learning Disabilities), were used to assess the effectiveness of treatment.

**Findings:** Results show that Susan's monthly PNES frequency reduced from phase A1 (assessment phase) to phase B (treatment phase), and this persisted to the follow-up phase (phase A2). However, the reduction was not statistically significant. The intervention was associated with improvements in Susan's mood, anxiety and quality of life. Reduction of Susan's anxiety was clinically significant.

**Conclusions:** This SCED study demonstrates that CBT might be associated with reduction of PNES frequency and improvements in general functioning in people with PNES and learning disabilities; however, further research, especially around confounding variables is needed.

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**KEYWORDS**

clinical psychology, epilepsy, intellectual disability, psychological therapy

**Accessible Summary**

- People sometimes experience episodes that look like epileptic seizures. Unlike epileptic seizures, these episodes are caused by psychological factors (such as stress). These are called 'psychogenic nonepileptic seizures' or PNES for short.
- We wanted to see if Cognitive Behaviour Therapy can help individuals with PNES and learning disabilities.
- We have written what we did with Susan (a woman with PNES and a learning disability) and how the therapy helped her.
- We met with Susan 15 times for therapy sessions.
- Susan reported fewer episodes of PNES after the therapy. The change in the number of episodes was not big. Following therapy, Susan also reported better mood and quality of life, and feeling less anxious.

## 1 | INTRODUCTION

Psychogenic nonepileptic seizures (PNES), also commonly known as functional seizures, dissociative seizures and nonepileptic attack disorder, are episodes of altered behavioural, sensory, motor or cognitive function that resemble epileptic seizures (ES), but without measurable changes in brain physiology typical to ES (Alsaadi & Marquez, 2005). PNES are a subcategory of functional neurological disorders (Hallett et al., 2016), described under the 'conversion disorder' category in the DSM-5 (American Psychiatric Association, 2013) and 'dissociative neurological symptom disorder, with non-ES in the ICD-11' (World Health Organisation, 2019, p. 11). The estimated prevalence of PNES in the general population is between 2 and 33 per 100,000 (Benbadis & Allen Hauser, 2000); however, people with PNES represent over 10% of referrals to outpatient specialist clinics (Angus-Leppan, 2008), making PNES a significant neurological condition, which warrants further research.

There are various models explaining the aetiology of PNES. For instance, psychodynamic models see PNES as a physical manifestation of emotional stress, whereas behavioural models interpret PNES as learned behaviour, activated via operant conditioning (see Anzellotti et al., 2020). Recently, an integrative model of PNES (Brown & Reuber, 2016) has been proposed. The model emphasises the role of various processes, in particular, the activation of a mental representation of a seizure, which develops as experiences and information about attacks accumulate and interact with behaviours. The model also emphasises PNES vulnerability factors (e.g., chronic stress, trauma), which play important role in the development and maintenance of PNES. Compared to previous models and theories, the integrative model

seems to capture a wide range of characteristics pertinent to a very heterogeneous group of people with PNES.

PNES commonly occur with other neurological and psychiatric conditions—for instance, up to 60% of people with PNES also have a confirmed diagnosis of epilepsy (Kutlubayev et al., 2018; Magaúda et al., 2011; Sigurdardóttir & Ólafsson, 1998). Rates of psychiatric comorbidities in individuals with PNES are even higher and range from 53% to 100%, with the highest rates for depression (up to 85%), anxiety disorders (up to 70%), post-traumatic stress disorder (PTSD) (up to 100%) and personality disorders (up to 74%) (Diprose et al., 2016; Jones & Rickards, 2021). Compared to individuals with ES, individuals with PNES and their caregivers experience higher levels of stigma and shame (Karakis et al., 2020; Reuber et al., 2022), which can further exacerbate mental health problems in the PNES population and lead to reduced quality of life of these individuals and their caregivers.

PNES are more common in individuals with learning disabilities than in the general population. The median prevalence rate of PNES in this population is 9.4%, and up to 36% of these individuals will also experience ES, which can make diagnosing PNES in people with learning disabilities even more difficult (Duncan & Oto, 2008; Rawlings et al., 2021). Similar to people without learning disabilities, high rates of psychiatric comorbidities (e.g., anxiety, depression, panic attacks) have also been reported in the PNES population with learning disabilities (Duncan & Oto, 2008; Kanemoto, Goji, et al., 2017; Magaúda et al., 2011).

An accurate and timely diagnosis of PNES is crucial for providing appropriate and effective treatment (LaFrance et al., 2013). However, overlapping clinical presentation features between PNES and ES often lead to diagnostic delays, which can

be particularly long for people with learning disabilities (Rawlings et al., 2021). For instance, Duncan and Oto (2008) reported a mean diagnostic delay of nearly 11 years in the learning disabilities population (compared to roughly 7 years in the general population). The 'gold standard' diagnostic tool for PNES is video electroencephalography (V-EEG) to exclude the diagnosis of ES by showing normal brain activity during the seizure episode. However, V-EEG is often not readily available and therefore clinicians may initiate treatment based on the expert's knowledge of the patient's history, seizure description and observation (Rawlings et al., 2021).

There are no treatment guidelines for people with PNES in the United Kingdom. In fact, treatment guidelines are sparse globally (Kanemoto, LaFrance, et al., 2017). A clear communication of the diagnosis of PNES represents a first and important step in treatment; however, less is known about effective communication of a PNES diagnosis to people with learning disabilities and their support networks (Rawlings et al., 2021). In addition, psychological treatments—focused on reducing PNES frequency, managing comorbid psychological difficulties and improving quality of life—are usually recommended for people with PNES (Kanemoto, Goji, et al., 2017), and meta-analyses suggest that psychotherapeutic interventions lead to a reduction of PNES frequency (Carlson & Nicholson Perry, 2017). Appropriate adaptations (e.g., additional sessions, repetitions) need to be put in place for people with learning disabilities (Beail, 2015), and the inclusion of the individual's environment and surrounding systems (e.g., caregivers, support workers) seems to be particularly important when working with people with PNES and learning disabilities (Kanemoto, LaFrance, et al., 2017).

Cognitive behavioural therapy (CBT) is the form of psychotherapy that has been most widely researched and applied in the context of treating individuals with PNES (Goldstein et al., 2010, 2020; LaFrance et al., 2014). One of the most widely researched and applied CBT protocols is based on the fear-avoidance model. In this model, a PNES is perceived as a dissociative response to arousal that allows a person to avoid unpleasant emotions/thoughts. Triggers to arousal can be internal (e.g., panic symptoms) or external (e.g., trauma) (Goldstein et al., 2010). A recent randomised controlled trial failed to demonstrate that CBT based on this model leads to a significant reduction of seizure frequency compared to standardised medical treatment in people with PNES (Goldstein et al., 2020); however, individuals in the CBT group reported significantly better psychological functioning, subjective clinical improvement and treatment satisfaction compared to the treatment as usual group, indicating that CBT could be beneficial for these individuals. However, this trial excluded people with learning disabilities and therefore the evidence base for CBT for PNES in people with learning disabilities is limited to case reports (Atnas & Lippold, 2013; Diamond & Delaney, 2020).

Single-case experimental design (SCED) is a methodological approach allowing the researcher or clinician to investigate the effectiveness of treatment for an individual client. Due to its strict design, SCED allows valid inferences about the effects of treatment and therefore it offers advantages over standard case study reports (Kazdin, 2019). SCEDs are particularly important where there is a need for development and evaluation of specific treatments (e.g., in the context of treating PNES in individuals with learning disabilities) (Morley, 2018). To our knowledge, no SCED on evaluating the effectiveness of CBT in the context of both PNES and learning disabilities has been published.

This study aims to address this gap in the literature by reporting a SCED study with a 38-year-old woman with a mild learning disability and the diagnosis of PNES. Our primary hypothesis was that the intervention would lead to a reduction in PNES frequency, which would be sustained after the therapy (follow-up phase). Our secondary hypotheses were that the intervention would lead to reduced anxiety and depressive symptoms and improved quality of life.

## 1.1 | INTRODUCTION TO THE CASE

Susan (pseudonym) is a lady in her late 30s with the diagnoses of a mild learning disability (General Ability Index between 60 and 70 and General Adaptive Composite between 52 and 60; assessed by a clinical psychologist in the same National Health Service—NHS team), epilepsy, PNES and two life-long physical health conditions—details of which will not be provided due to the risk of breaching patient confidentiality. Notably, one of Susan's life-long physical health conditions is associated with a slightly higher risk of epilepsy, but not PNES. Susan was referred to a learning disability crisis team in South-East England by her community learning disability nurse due to a substantial increase in the frequency of her PNES in addition to the sudden onset of experiencing voice-hearing. Notably, this was Susan's first episode of voice-hearing. Susan's worsening of symptoms was preceded by unsuccessful hospitalisation for treatment of PNES and the sudden withdrawal of diazepam (a medication that was previously prescribed by Susan's neurologist for when Susan was experiencing 3 PNES within a single day). The decision to stop with diazepam was made by Susan's neurologist after unsuccessful hospitalisation.

Susan was assessed by a clinical psychologist and a trainee clinical psychologist at the end of 2021. At the time of the initial assessment (10 days after the referral), Susan reported feeling significantly better—the frequency and intensity of hearing voices and the frequency of PNES were significantly reduced. Susan and her support workers said that the reduction of symptoms coincided with Susan starting to take risperidone (1 mg a day), which was prescribed to her by a psychiatrist after an urgent referral to the learning disability crisis team. This was the first time that Susan was

prescribed an antipsychotic medication and the medication was prescribed without diagnosing Susan with a psychotic disorder. Regardless of reduction of her symptoms, Susan still appeared anxious and distressed at the time of psychological assessment. She became very emotional when talking about the PNES and voice-hearing and said she would like psychological help to cope with these symptoms.

## 2 | ASSESSMENT OF THE PROBLEM SITUATION

### 2.1 | Relevant personal history

Susan was born at full term in the 1980s in England. She attended mainstream education until the age of 13 and was home-schooled for the remaining 3 years. Susan was not able to sit General Certificate of Secondary Education; however, she attended various vocational courses and had several part-time jobs from her mid-20s onwards. Susan lived with her parents and brother until her early 30s when she moved to supported living accommodation. At the time of the assessment, Susan lived with three other people with learning disabilities, and at least two members of support staff were always present in the supported living accommodation. Susan also had one-to-one support whenever leaving home (e.g., when going to the shop). In addition, Susan constantly wore a seizure detection alarm around her neck, which instantly notified her support staff whenever Susan experienced PNES.

Susan had several health problems at the time of the assessment. She had been diagnosed with epilepsy in her 20s, although had experienced ES from the age of 5. Susan's ES has been well managed with medication and Susan reported not experiencing them for years. Susan started experiencing PNES in 2018. Compared to her ES, which manifested as absence seizures, Susan's PNES manifested as motor seizures involving her whole body (i.e., whole body shaking and fully extended arms and legs). Susan always experienced PNES when sitting down, and therefore, never suffered any injuries as a result of a sudden whole-body seizure. Notably, Susan never experienced ES, which would manifest differently than absence seizures.

Susan reported a long history of traumatic experiences. She experienced a head injury with no loss of consciousness in her early 20s, was hit by a car roughly 10 years later and was physically and emotionally abused by her ex-partner with whom she had a decade-long relationship.

Susan was referred for CBT for PTSD in 2015 (primary care). However, we were unable to locate records of her attending and completing this treatment. Susan received CBT during her in-hospital treatment for PNES earlier in 2021. As Susan's hospitalisation resulted in a large increase in her distress levels, CBT was less accessible for her at the time and the treatment had to be terminated early.

### 2.2 | Presenting difficulties

At the point of the initial assessment, Susan's presenting difficulties were mainly related to anxiety. Susan reported worrying about her future, being ill and finding it hard to control her worries. Susan also reported strong physical sensations associated with worrying, such as her heart beating fast, feeling shaky in her legs and hands, fluttering sensations in her stomach and finding it hard to breathe. These sensations were particularly intense when she was in busy places (e.g., local supermarket) or in wide open places (e.g., work canteen). Although Susan has not been formally diagnosed with an anxiety disorder, her clinical presentation resembled many features of agoraphobia.

Susan also showed some symptoms of low mood. For instance, she became quickly upset and tearful when talking about her PNES. The staff members also reported that she had dropped some of her hobbies (e.g., playing clarinet and crocheting) due to low mood. Susan's risk to herself/others and from others was assessed during the psychological assessment. Based on the information from Susan and her support workers, it was concluded that Susan's risks were low. In general, Susan reported that her low mood was less interfering with her everyday life than her high anxiety.

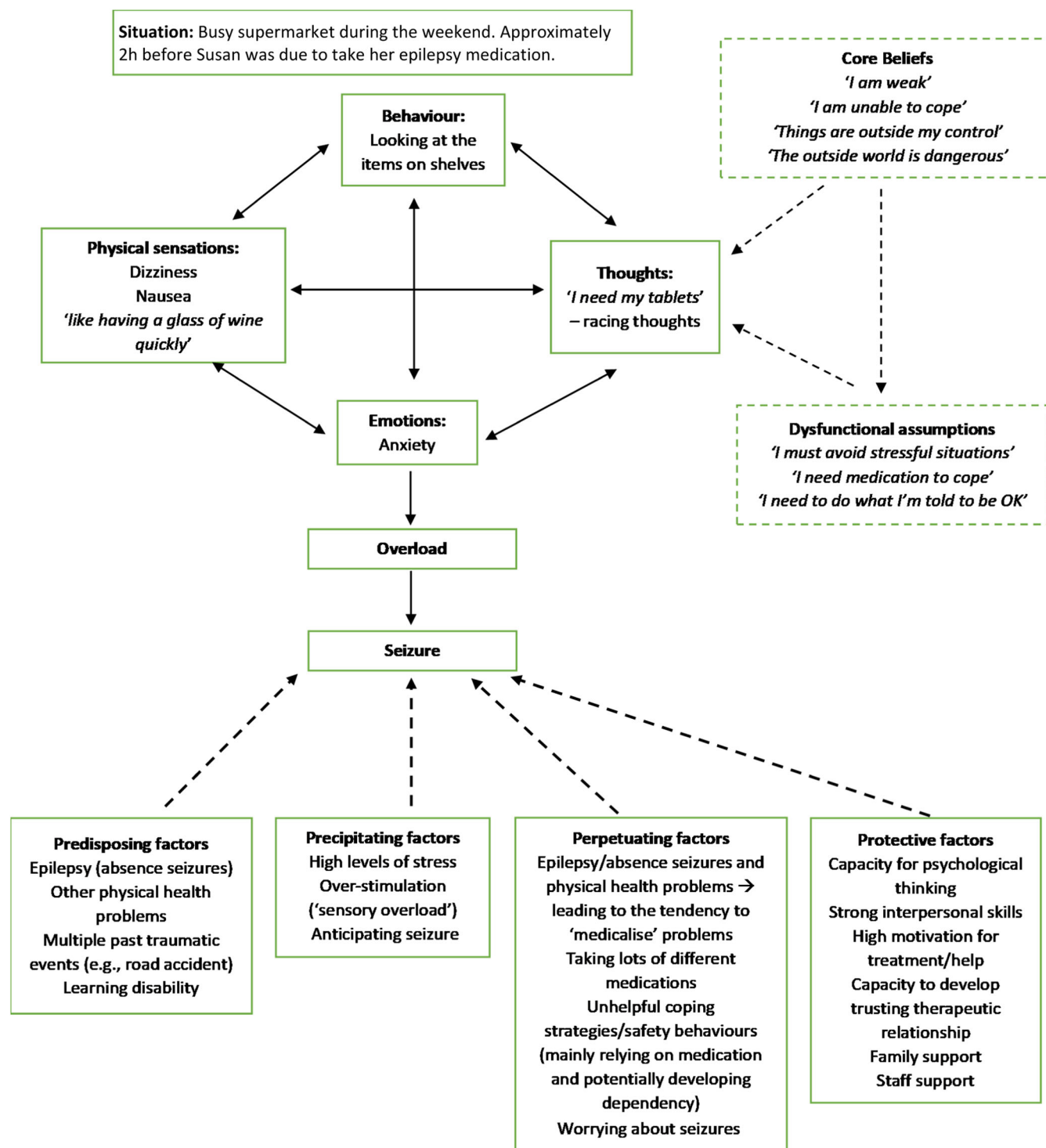
Susan agreed to a series of one-to-one weekly CBT sessions with a trainee clinical psychologist (referred to as the therapist). The therapist received regular supervision by a clinical psychologist (referred to as the supervisor). Following guidance from the Health Research Authority, this study did not constitute research that would require formal ethical approval. However, consent was obtained and the Trust's standard safeguarding procedures were implemented. Susan also provided consent for our work with her to be written up as a case study.

Susan was taking her regular antiepileptic medication (lamotrigine) twice a day and risperidone (1 mg) once a day throughout our work with her.

### 2.3 | CBT formulation

Susan's CBT formulation, including her predisposing, precipitating, perpetuating and protective factors, is shown in Figure 1.

Susan's CBT formulation shares many similarities with CBT anxiety models, in particular, the cognitive model of panic disorder (Clark, 1986) and health anxiety (Salkovskis & Warwick, 1986). We hypothesised that Susan's anxiety was triggered by interpreting internal (e.g., dizziness, nausea) and external (e.g., large open spaces, noisy environment) triggers as a threat. Susan's main coping strategy in these situations was to rely on medication (i.e., antiepileptic medication and paracetamol), indicating low perceived control over her symptoms (e.g., a core belief 'I am unable to cope'). When the levels of distress reached a certain threshold (described as 'overload' by her), Susan would experience a PNES.



**FIGURE 1** Susan's cognitive-behavioural therapy (CBT) formulation with predisposing, precipitating, perpetuating and protective factors.

There were many predisposing factors (e.g., having a learning disability, multiple past traumatic events) that made Susan more vulnerable for PNES. Experience of past traumatic events was a particularly strong predisposing factor, and Susan occasionally wanted to talk about these events during the therapy. However, she denied any PTSD symptoms, and therefore, our intervention focused on her

presenting symptoms— anxiety. Susan's expectations of PNES in certain environments (e.g., supermarkets) represented an important trigger, while her tendency to 'medicalise' problems and avoid anxiety-provoking situations represented significant perpetuating factors. Last, Susan had many protective factors, such as being highly motivated for psychotherapeutic work and having a strong support network.



### 3 | METHODS

#### 3.1 | Measures

##### 3.1.1 | Daily seizure charts

Susan's support workers used seizure diaries to record daily the frequency and duration of Susan's PNES. Daily seizure diaries were provided by Susan's neurologist and the support workers were instructed to fill in the charts immediately after Susan experienced a PNES. This was to ensure that the PNES were recorded consistently.

In line with previous studies on evaluating the effectiveness of CBT for PNES (Goldstein et al., 2020), PNES frequency was used as a main outcome variable. Daily seizure frequency was averaged on a monthly and weekly basis.

##### 3.1.2 | Idiosyncratic mood measure

We used a subjective mood rating scale ranging from 1 to 10 (1 meaning 'not happy' and 10 meaning 'very happy') to assess Susan's mood on a weekly basis.

##### 3.1.3 | Standardised questionnaire measures

Two validated self-report questionnaire measures for people with learning disabilities were completed by Susan at the start of therapy, mid-therapy/therapy review and end of therapy:

*Glasgow Anxiety Scale for People with an Intellectual Disability (GAS-ID)*

GAS-ID (Mindham & Espie, 2003) is a 27-item self-rating scale measuring anxiety symptoms in individuals with learning disabilities. Individuals answer the questions using a 3-point rating scale (0—'no', 1—'sometimes' and 2 'a lot'). The scale total is calculated by summing all responses, and the total score ranges from 0 to 45, with a higher score indicating higher anxiety. The cut-off score of 13 or more indicates the presence of an anxiety disorder. The GAS-ID has favourable test-retest reliability, internal consistency and concurrent validity (Mindham & Espie, 2003).

*Mini-Maslow Assessment of Needs Scale-Learning Disabilities (Mini-MANS-LD)*

The Mini-MANS-LD (Raczka et al., 2020) is a 9-item measure of the quality of life in people with learning disabilities. The measure was adapted from the original 19-item MANS-LD measure (Skirrow & Perry, 2009). The respondent answers the questions using a 5-point scale with associated pictures to facilitate understanding. The scale total is calculated by summing all responses, and the total ranges from 9 to 45, with lower scores indicating better quality of life.

#### 3.2 | Design

We used the ABA, also known as 'withdrawal/reversal design', which is a design widely used in clinical settings (Morley, 2018). Susan's support workers monitored daily the frequency of her PNES using the seizure charts, which established a baseline (phase A1) of symptoms 1 month before the first CBT session. The length of phase A1 was deliberately long to gain an insight into the stability of the frequency of Susan's PNES and consequently to improve the validity of the design (Morley, 2018). Furthermore, the long assessment phase allowed for measurement of clinical outcomes during and after stabilisation of Susan's response to the effect of psychotropic medication (risperidone), which was introduced at the point of Susan's referral to the service (before phase A1). The frequency of PNES was recorded daily by Susan's support staff throughout the intervention (phase B). Susan's daily seizure charts were analysed for another month after CBT (follow-up/phase A2) to establish whether the effects of the intervention were sustained.

#### 3.3 | Data analysis

First, we assessed within-phase stability using the stability criterion as defined by Neuman and McCormick (1995). For the phase to be stable, this criterion requires between 80% and 90% of data points falling within the 15% of the phase mean. Differences between the phases were then compared using the percentage of nonoverlapping data (PND) approach. This approach looks at the percentage of data points in phase B that are exceeding the highest data point in phase A (Scruggs et al., 1987). Following Lobo et al. (2017), a PND < 50% was interpreted as 'no observed effect', a PND between 50% and 70% as 'questionable effect' and a PND > 70% as 'the intervention was effective'. Finally, a graphical representation of the data was used to estimate the effectiveness of the intervention over time.

In terms of secondary outcome measures, we calculated Reliable Change Index (RCI; see Morley & Dowzer, 2014) for Susan's GAS-ID scores. For this purpose, we used normative data provided by the authors of GAS-ID (Mindham & Espie, 2003). Since there is no normative data available on Mini-MANS-LD, RCI or any other measure of reliable change could not be calculated for this measure.

#### 3.4 | Course of therapy

Susan attended 15 1-h CBT sessions over the course of 4 months. Eleven sessions were delivered face to face, and four sessions were delivered remotely due to coronavirus disease 2019 (COVID-19). As there is no established treatment protocol for PNES for adults with learning disabilities, our treatment process was informed by previously published case studies (Atnas & Lippold, 2013; Diamond & Delaney, 2020) and studies of people without learning disabilities (Goldstein et al., 2020). Based on Susan's formulation, session content was also informed by treatment protocols for panic

disorder and health anxiety (Clark, 1986; Salkovskis & Warwick, 1986). Following treatment recommendations for delivering CBT to people with learning disabilities (Beail, 2015; Kanemoto, Goji, et al., 2017), appropriate adaptations (e.g., additional sessions, multiple repetitions, inclusion of support workers) were made to ensure that the intervention was tailored to Susan's needs.

The first part of CBT (sessions 1–5) was focused on establishing good therapeutic rapport, socialising Susan to the cognitive model, which included drawing simple ('hot-cross bun') formulations and exploring relaxation and grounding techniques. Susan's support network (support workers and parents) helped Susan to practise these techniques between sessions by reminding her to do them in various situations. By the end of first phase of CBT, Susan had a wide range of tools to use to help manage her anxiety in stressful situations. These included simple breathing and grounding techniques, out of which Susan found the five-finger breathing technique and touch-based grounding techniques particularly helpful. With the assistance of her support network, Susan managed to practice these techniques daily.

In the second part of CBT (sessions 5–9), we focused on psychoeducation about 'fight or flight' response and PNES. We explored in which contexts Susan's PNES are most likely to occur and discussed why these contexts are particularly triggering. It was established that Susan's PNES most commonly occur in situations that she described as either 'overstimulating' (e.g., when in a busy place or in a large open place, when sitting on a

bench at the edge of a football field) or when she is doing something 'relaxing' (e.g., sitting on the sofa and reading). As there was no existing booklet for people with PNES and learning disabilities, the therapist adapted an information leaflet on PNES for the general population (CODES Trial Investigators, 2014) and shared it with Susan. The adapted booklet was subsequently approved by the CODES Trial Investigators to be used for other people with PNES treated within the same Trust. In addition, we continued with consolidating relaxation and grounding techniques throughout phase 2.

The third part of CBT (sessions 9–15) was focused on designing a hierarchy of anxiety-provoking situations and carrying out behavioural experiments (e.g., leaving the house and going for a walk in a busy street, sitting in a large open field) to challenge Susan's beliefs about her perceived lack of control of her anxiety. These situations provided opportunities for Susan to practise the techniques learned in the first two phases of CBT in real-life situations. Although Susan always reported behavioural experiments being very challenging and exhausting, she was also able to understand their importance in challenging her beliefs around the lack of control of her emotions. The final goal was for Susan to go to her local supermarket and be able to spend 10 min there (accompanied by her therapist). Unfortunately, this was not possible due to the final few sessions taking place remotely due to COVID-19. The last two sessions were focused on relapse prevention, which included a collaborative creation of a list of CBT techniques and tools that Susan found most

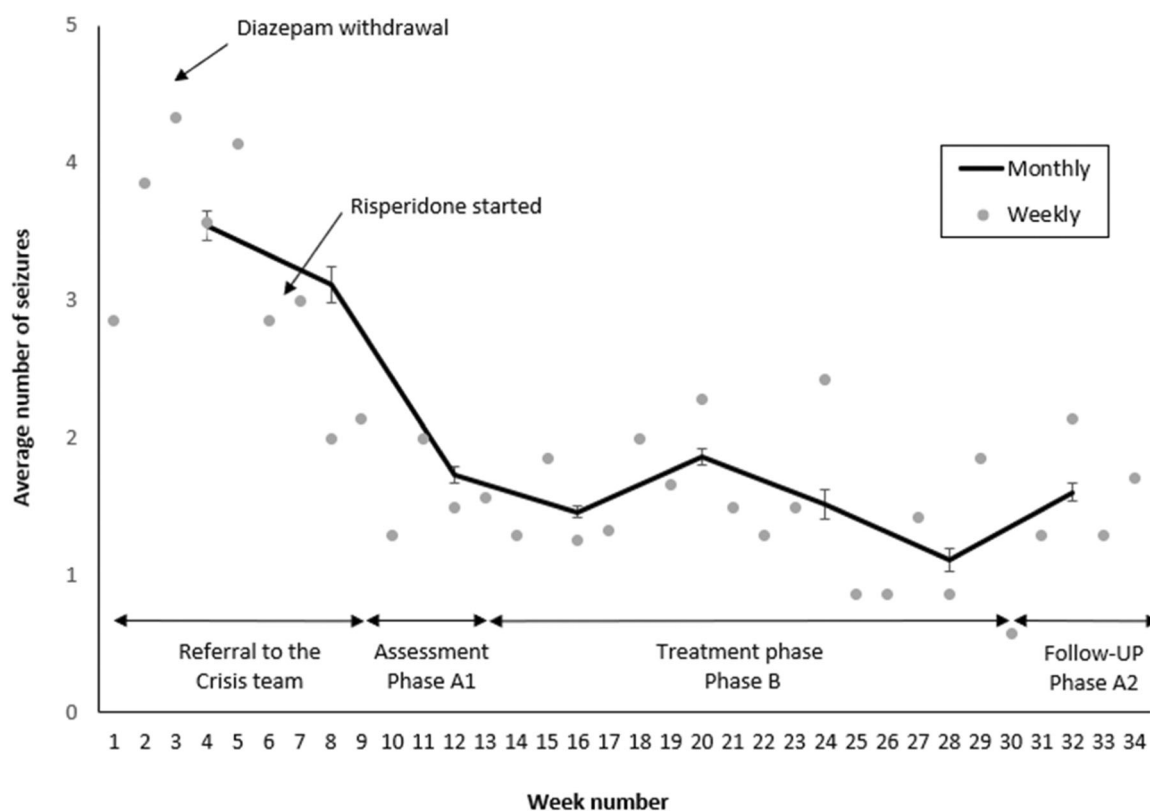
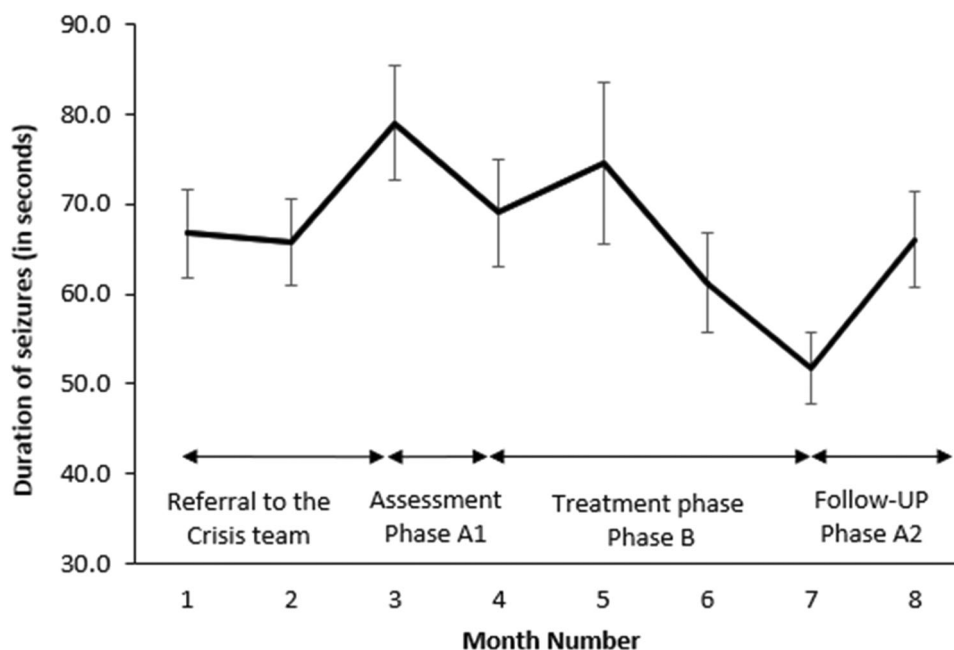


FIGURE 2 Susan's average number of seizures over time.



**FIGURE 3** Susan's average duration of seizures (in s) over time.

helpful. This list was subsequently shared with Susan and her support workers.

## 4 | OUTCOME

### 4.1 | Primary outcome—PNES frequency

**Within-phase stability:** Eighty percent of data points within the baseline phase (phase A1) were within the 15% interval of the phase mean ( $M_{A1} = 1.7$ ). Similarly, 75% of data points within the follow-up phase (phase A2) were in the desired phase mean ( $M_{A2} = 1.6$ ) interval. Within-phase stability of the treatment phase (phase B) was lower with 53% of data points falling within the 15% interval of the phase mean ( $M_B = 1.5$ ).

**Between-phase comparisons:** PND was calculated between the baseline phase (phase A1) and treatment phase (phase B). Sixty-five percent of data points in phase B met the PND criteria, indicating a questionable effect of intervention. PND criterion was also applied to compare phase B with the follow-up phase (phase A2). There was no indication of a significant increase in PNES from phase B to phase A2 (PND = 0%). However, PND between the baseline phase and follow-up phase was 50%, indicating that the follow-up effects of the intervention are questionable.

**Graphical representation:** Figure 2 outlines Susan's weekly and monthly average daily PNES frequency with 95% confidence intervals, over time. Overall, we can see that Susan's PNES frequency reduced from the point of referral to the Crisis team ( $M = 3.5$ ,  $SD = 1.2$ ) until the beginning of phase A1/assessment phase ( $M = 1.7$ ,  $SD = 1.0$ ), which coincided with the initiation of risperidone. Overall, we can see a trend of reducing Susan's monthly average PNES

frequency from phase A1 to phase B. The trend of slight PNES reduction is also present in phase A2 (follow-up).

### 4.2 | Secondary data from seizure charts

Figure 3 outlines Susan's monthly average PNES duration with 95% confidence intervals.

Overall, there is a trend of reduction in the duration of Susan's PNES from phase A1 to phase B. However, there is a trend of an increase in Susan's average PNES duration from phase B to phase A2.

### 4.3 | Secondary outcome measures

Susan's subjective mood rating scores improved from 6 to 8 out of 10 throughout the course of treatment. Similarly, Susan's anxiety scores (assessed with GAS-ID) significantly decreased from 32 (assessment phase) to 20 (mid-therapy) and 17 (end of treatment), demonstrating statistically reliable change ( $RCI = 8.84$ ;  $RCI \geq 1.96$ ) (Mindham & Espie, 2003). Susan also reported improved quality of life (assessed with Mini-MANS-LD) with her scores dropping from 14 (assessment phase) to 11 (mid-therapy/end of treatment). Due to the lack of normative data for Mini-MANS-LD, it was impossible to determine whether this change was reliable.

### 4.4 | Patient and support workers' feedback

Susan's engagement in CBT was excellent throughout. She regularly completed homework and practised techniques outside the sessions.



She reported that CBT had a positive impact on her mood and her ability to cope with the PNES. Similarly, Susan's support workers noticed positive changes in Susan's mood and anxiety. Susan also started to engage in her hobbies (e.g., crocheting), for the first time since her referral to the crisis team.

## 5 | DISCUSSION

This single-case experimental study aimed to assess the effectiveness of CBT for treatment of PNES in an adult with a mild learning disability. To our knowledge, this is the first study that aimed to address this question by adopting a SCED. The results partially confirm our primary hypothesis. Although the intervention led to the reduction of PNES frequency, the effect of intervention was not big enough to be statistically significant, and therefore, no firm conclusions about the effectiveness of CBT can be drawn. The results also partially support our secondary hypotheses. The intervention was associated with improvements in Susan's mood, anxiety and quality of life; however, only the change in Susan's anxiety was statistically reliable. Although the PNES duration was not one of our main outcome variables, the results indicate that the intervention was associated with a reduction in average PNES duration; however, these changes did not persist in the follow-up phase. Finally, the intervention was very positively perceived by the patient and their support network.

The results of this study are broadly consistent with the results of a large randomised-controlled study with people without learning disabilities (Goldstein et al., 2020) and case studies of people with learning disabilities (Atnas & Lippold, 2013; Diamond & Delaney, 2020). Previous research suggests that CBT is associated with improvements in various areas of psychological functioning in people with PNES (Goldstein et al., 2020), and with PNES reduction, but not complete remission of PNES (Atnas & Lippold, 2013; Diamond & Delaney, 2020). Based on the findings of the current study, we can tentatively conclude that CBT with adaptations for people with learning disabilities might have the potential to reduce PNES frequency and improve clinically relevant aspects of functioning in individuals with PNES. However, apart from the significant reduction of Susan's anxiety, other changes failed to achieve statistical significance, and therefore, results should be interpreted with caution. Significant reduction of Susan's anxiety could be due to her PNES being phenomenologically close to experiencing agoraphobia (i.e., occurring in very busy places and open spaces), and therefore, it is possible that the treatment was particularly effective for the parts of PNES that are similar to the symptom of this anxiety disorder. Notably, overlaps between subjective symptoms of PNES and panic attacks have been highlighted by previous research (e.g., Goldstein & Mellers, 2006), where PNES are sometimes described as 'panic attack without panic' (i.e., symptoms of autonomic arousal, somatic symptoms with little/without panic cognitions). It is possible that the reduction of anxiety symptoms in Susan presented a first step of her recovery, whereas a longer psychotherapeutic

intervention, focused on her thoughts (e.g., illness beliefs, beliefs about the controllability of her symptoms) might lead to a greater reduction of PNES.

There are many reasons why this CBT intervention did not lead to a significant reduction and a complete remission of Susan's PNES. First, Susan has a history of multiple past traumatic events, which represent a significant predisposing factor for developing PNES (Diprose et al., 2016; Jones & Rickards, 2021), and therefore, a long-term trauma-informed intervention might be more effective. In addition, PNES frequency and remission as an outcome measure has been questioned (Reuber et al., 2005) and researchers and clinicians suggest other variables, such as patient-reported quality of life, as a more meaningful outcome of CBT in PNES populations (Goldstein et al., 2020; Rawlings et al., 2017). Finally, it is important to acknowledge that Susan had a learning disability and a lifelong history of epilepsy—both of which are important predisposing and perpetuating factors in PNES (Rawlings et al., 2021).

This study has several strengths. First, both baseline phases (A1 and A2) were stable. Another strength is the inclusion of a follow-up phase to assess whether the effects of intervention persisted over time. In addition, we used a range of primary and secondary outcome measures. Appropriate adaptations to CBT protocols were made to fit the needs of Susan. For instance, sessions included plenty of repetitions, adaptation of existing therapy materials and were appropriately paced to meet Susan's needs. In addition, we included Susan's support network, which resulted in the consolidation of CBT techniques (e.g., relaxation and grounding). Finally, Susan's engagement and self-reported treatment satisfaction were high, indicating that the intervention was perceived as strongly positive.

This study also has limitations. First, within stability of phase B was lower than stability of phases A1 and A2, indicating that between-phase comparisons are less reliable. Although the intervention led to a reduction of Susan's PNES frequency, the intervention did not lead to a significant reduction and/or PNES remission. Furthermore, it is possible that the observed improvements occurred due to nonspecific factors (e.g., therapeutic alliance), which are significant predictors of treatment outcomes (Chatoor & Krupnick, 2001). It is possible that another treatment modality or a trauma-informed intervention could result in more favourable findings, in particular, given Susan's history of multiple traumatic events and her desire to discuss these in therapy. Finally, it is important to acknowledge that Susan's PNES reduction and an overall reduction of anxiety symptoms could have also been influenced by the long-term effect of psychotropic medication and Susan's better living circumstances—that is, staying at home rather than in hospital—compared with the circumstances at the time of the referral to the crisis team.

Our study suggests a few possibilities for future research. First, future SCED studies with a more complex research design and repeated intervention/follow-up phases are needed to gain a better insight into the stability of results. Furthermore, future SCEDs could also include longer baseline phases, in particular,

when participants also take psychotropic medication. A longer baseline phase could enable for better control of confounding variables (e.g., medication). Findings based on SCEDs could inform the design of larger, preferably randomised-controlled, studies with individuals with PNES and learning disabilities. In addition, all the existing case studies have explored the effectiveness of CBT in this client group, while no studies have explored other therapeutic approaches in the learning disabilities population. This is important as the nature of PNES in LD has traditionally been seen as behavioural (Rawlings et al., 2021), indicating that behavioural approaches might be more effective than CBT. Future studies should also explore the phenomenological aspects of PNES in people with learning disabilities. Research with individuals without learning disabilities suggests that PNES are phenomenologically very similar to panic attacks (Brown & Reuber, 2016); however, less is known about the experiences of people with learning disabilities. Understanding the phenomenology of PNES in this population is crucial to inform the development of treatments. Finally, future research should also focus on better understanding of the role of support systems (e.g., support workers and carers) in people with PNES and learning disabilities.

There are currently no treatment guidelines for adults with PNES and learning disabilities. This is the first SCED investigating the effectiveness of CBT in this client group. The results of this study contribute to the field by providing preliminary evidence that CBT might be effective in reducing PNES frequency and improving other areas of psychological functioning in this client group (e.g., anxiety). Further, single-case and group research is needed to establish whether CBT is significantly effective and should be offered as first-line treatment for this client group.

## ACKNOWLEDGEMENTS

Jerica Radez was funded by Oxford Health NHS Foundation Trust. Tom Crossland was supported by Berkshire Healthcare Foundation Trust and Park House School. The authors thank Dr Sarah Walden, Dr Ciorsdan Anderson and Dr Hannah Reville for their input on CBT and PNES, and Ms Miranda Dodd for her help with organising the information about the patient's medical history. Finally, the authors thank 'Susan' and her support workers for their collaboration and assistance with the data collection.

## CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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**How to cite this article:** Radez, J., Crossland, T., & Johns, L. (2023). Cognitive behavioural therapy for psychogenic nonepileptic seizures (PNES) in an adult with a learning disability: A case study. *British Journal of Learning Disabilities*, 51, 586–596. <https://doi.org/10.1111/bld.12531>