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Review

Factors associated with illness representations in adults with epileptic and functional seizures: A systematic review

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ABSTRACT

Illness representations refer to a person's beliefs about their health condition and are thought to influence clinical outcomes. By understanding factors related to illness representations, potentially modifiable targets for psychological intervention can be identified. The aim of this systematic review was to synthesise the literature on factors associated with illness representations in people with epilepsy and functional seizures. Three electronic databases (Psychinfo, EMBASE, and Proquest (Theses and dissertations)) were searched for studies that reported on associations between Illness Perception Questionnaire scores (or variations thereof) and biopsychosocial factors in people with epilepsy or people with functional seizures. Seventeen studies met inclusion criteria and were assessed with a bespoke quality appraisal tool. Overall, there was moderately strong evidence for an association between more threatening illness representations and poorer clinical outcomes relating to seizure characteristics, distress, coping, and quality of life; the evidence for these relationships was stronger for people with epilepsy than functional seizures. There was no clear difference between the illness representations of the two groups. The results of this review highlight the clinical importance of illness representations in people with seizure disorders, as well as opportunities for further research.

1. Introduction

Epilepsy and Functional Neurological Disorder (FND)[1] and are two causes of seizures. Epilepsy refers to a neurological disorder characterised by recurrent episodes of paroxysmal brain dysfunction due to sudden, abnormal neuronal discharge (epileptiform activity) [1]. Functional seizures are the seizure variant of FND, resembling epileptic seizures but thought to represent episodes of dissociation [2]. Both seizure disorders are common; epilepsy has an estimated international lifetime prevalence of 7.6 per 1000 people [3]. Difficulties with ascertainment mean that functional seizure prevalence figures likely underestimate the true number of people with the condition, however, a recent 10-year population-based study in Norway found a prevalence rate of 23.8 per 100,000 [4]. Both epilepsy and functional seizures may result in impairments of consciousness and/or convulsions, and both are therefore associated with significant disability and psychological distress [5,6].

1.1. Rationale

Illness representations refer to a person's beliefs and expectations about an illness or symptom(s) [7] and are therefore thought to be linked to how individuals respond to and manage their illness [8]. For example, the first-line of treatment and management for epilepsy is usually anti-convulsant medication, however, estimates of non-adherence range between 29% to 39% resulting in reduced seizure control [9]. Patient beliefs about medication, including their perceived necessity and concerns about side-effects, have been associated with non-adherence [10]. Adherence to many of the safety recommendations for people with epilepsy [11], such as increasing nocturnal supervision for those with night time seizures or taking showers rather than baths, is also arguably facilitated by an adequate understanding of the causes and potential consequences of seizures in epilepsy.

Psychological therapy is a further example of the clinical relevance of illness representations for people with epileptic and functional seizures. Psychological therapy can be employed as an adjunct to standard

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[1] FND = Functional Neurological Disorder

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treatment for people with epilepsy [12], helping to improve wellbeing and facilitate coping. It is also the main treatment of choice for people with functional seizures, aiming to reduce seizure frequency and/or help people to cope better with the condition [e.g., 13]. As psychological interventions like Cognitive Behavioural Therapy work explicitly with patients' beliefs about what caused and maintains their seizures as well as the consequences of their disorder, it is important for healthcare professionals to understand illness representations in both groups.

Furthermore, illness representations are theoretically important in the aetiology of functional seizures. Modern theories, such as the Integrative Cognitive Model [14] are based on predictive coding accounts; that is, the brain generates functional symptoms through predictions regarding the causes of sensory inputs [15]. These predictions are based on a combination of interoceptive data and prior beliefs about probable causes of interoceptive data; beliefs that can also be thought of as illness representations (i.e. assumptions about what sensory experiences constitute a 'symptom'). In the Integrative Cognitive Model, sensory experiences are interpreted as the onset of a seizure event, triggering the activation of a 'seizure scaffold' (the mental representation of a seizure) and resulting in functional seizures. Moreover, the seizure scaffold itself is thought to develop partly from exposure to models of seizures in the self or other people. Therefore, illness representations are considered relevant to both the development and experience of functional seizures. As such, there is both theoretical and clinical benefit to understanding illness representations in people with seizure disorders.

Illness representations can be assessed qualitatively (by means of interviews) and quantitatively, using self-report measures such as the Illness Perception Questionnaire (IPQ[2]) [16]. The IPQ was developed to assess five major cognitive components that theoretically constitute an individual's illness representations: 1. identity (symptoms the individual associates with the illness), 2. cause (ideas about aetiology), 3. timeline (perceived illness duration), 4. consequences (expected effects of the illness or outcomes), and 5. cure/control (how one recovers). Some subscales are scored such that higher scores reflect 'worse' illness representations, whereas others are reverse scored so higher scores mean the opposite. Therefore, the phrase "more threatening" illness representations indicates more negative perceptions. The IPQ has since been developed into two further iterations; the Illness Perception Questionnaire-Revised (IPQ-R[3]) [17] and the Brief Illness Perception Questionnaire (BIPQ[4]) [18]. The IPQ-R was developed to deal with psychometric problems with two of the subscales and to include new subscales measuring cyclical timeline representations (how unpredictable symptoms are), illness coherence (how much sense an individual can make of their illness), and emotional representation (how distressing symptoms are). The BIPQ was later developed to make assessment of illness representations quicker and therefore more accessible for patient groups as well as more convenient for practitioners in busy clinical settings.

If illness representations do indeed determine how patients respond to and manage their illness, then one would expect IPQ scores to be associated with a range of biopsychosocial characteristics or clinical outcome factors in patients with seizure disorders. To date, there has been a systematic review of qualitative studies [19] and a mixed qualitative and quantitative review of health care practitioners' perceptions [20], however, there has been no systematic review of factors associated with illness representations in either individuals with functional seizures or epilepsy, or reviews comparing illness representations between these groups. Synthesizing the quantitative literature may therefore generate new insights. For example, it would be useful to identify any modifiable factors associated with illness representations, as these may present opportunities for psychological intervention and therefore improved outcomes.

[2] IPQ = Illness Perception Questionnaire
[3] IPQ-R = Illness Perception Questionnaire Revised
[4] BIPQ = Brief Illness Perception Questionnaire

1.2. Objectives

The aim of this review is to synthesise existing findings regarding factors associated with the illness representations of people with functional seizures and people with epilepsy in studies that have used the IPQ (or variations thereof). The primary review question is, 'What factors are associated with IPQ scores in people with seizure disorders?' To explore for differences between seizure type and illness representations, a secondary review question is, 'How do IPQ scores compare between people with functional seizures and people with epilepsy?'

2. Methods

The conduct and reporting of this review adhere to the PRISMA guidelines [21].

2.1. Eligibility criteria

Studies were included in the review if they were published in English after 1995 (the year prior to the publication of the IPQ), included populations of people with epilepsy and/or functional seizures aged 16 years or over, and if they reported IPQ, IPQ-R, or BIPQ total or subscale scores. To be eligible for inclusion, studies also needed to report on an association between an IPQ/IPQ-R/BIPQ score and clinical outcome variables or characteristics (e.g., a correlation between BIPQ scores and psychological distress, or a comparison of IPQ-R scores between patient groups). Studies that included people with functional seizures and other presentations of FND in the same group (i.e. a mixed FND sample) but did not report IPQ scores for functional seizures separately were excluded. Intervention studies were included if baseline IPQ scores were compared with one or more clinical outcome variables or characteristics. To mitigate publication bias, unpublished studies (e.g., theses and dissertations) were eligible for inclusion. Review articles or case series and reports were excluded.

2.2. Information sources and search strategy

Following an initial scoping search, three bibliographic databases (Psychinfo, EMBASE, and Proquest – Theses and dissertations) were searched for relevant published and unpublished data from 1995 to June 2022. Searches were devised in collaboration with an information specialist and contained no methodological search filters that would limit results to specific study designs. Box 1 details the search syntax used. To identify additional relevant literature, the reference lists of included full-text studies and of relevant systematic reviews (e.g., Cochrane review of psychological treatments for epilepsy) were searched. Google Scholar was also used to identify any papers that had subsequently cited included studies.

Box 1
Search syntax
(Illness perception* or Illness representation* or Illness cognition* or common sense model or illness belief* or cause* or control* or cure* or identity or time line* or consequence*) AND (nonepileptic attack* or non-epileptic attack* or nonepileptic seizure* or non-epileptic seizure* or pseudoseizure* or dissociative seizure* or dissociative convulsion* or pseudoepilep* or hysterical seizure* or hysterical convulsion* or hysteroepilepsy* or conversion seizure* or psychogenic seizure* or functional seizure* or nonepileptic event* or non-epileptic event*) AND (epilep* or epileptic seizure* or seizure*)

2.3. Screening, selection, and data extraction

Screening, selection, and data extraction were all performed by one reviewer (IW) using an online systematic review production tool (Covidence). All titles and abstracts were screened against inclusion and exclusion criteria. Eligible full-text articles along with any relevant

supplemental information were downloaded for screening and data extraction using a bespoke data extraction tool.

2.4. Quality assessment

Study quality in relation to answering the review question was assessed using a quality appraisal tool adapted specifically for this review and intended to approximate quality based on factors relating to the categories of participants, measurement, analysis, and sample size. With respect to participant-related factors, the tool assessed i) the risk of bias introduced by the recruitment method and ii) the eligibility criteria, as well as iii) whether sufficient demographic data were collected to establish sample representativeness. With respect to measurement-related factors, the tool also assessed iv) if the authors had demonstrated fidelity to the administration instructions of the IPQ (i.e. adapting the measure as necessary and reporting on subscale or total scale reliability). Regarding the analysis, the tool assessed v) if appropriate statistical analyses accounting for confounding variables had been conducted and vi) if missing data had been reported and appropriately dealt with. Many studies did not provide a formal power calculation, therefore, sample size was rated according to vii) the power and effect size conventions reported by Cohen [22] and used in two previously published systematic reviews of functional seizures [23,24]. Assuming a two-tailed independent *t*-test with $\alpha = 0.05$, studies with a minimum sample size of <15 in each group (< 80% power to detect a very large effect size, Cohen's $d = 1.1$) were rated as 'insufficient', studies with sample sizes between 15 and 25 per group (i.e., < 80% power to detect a large effect size, $d = 0.8$) were rated as 'poor', studies with sample sizes between 26 and 63 per group ($\geq$ 80% power to detect a large effect size, $d = 0.8$) were rated as 'moderate', and studies with $\geq$ 64 participants per group were rated as 'good' (i.e. $\geq$ 80% power to detect a medium effect size, $d = 0.5$). Each criterion was scored on a scale of 0 – 3. To assign quality classifications to studies, citeria i - vi were summed, percentages of the total maximum score were calculated and entered into an algorithm (Table 1) along with criteria vii (sample size). Studies were rated as 'high' quality if they scored $\geq$ 70% on criteria i - vi and were rated as 'good' on sample size. 'Medium' quality studies scored 50 – 69% on criteria i - vi and were rated as 'medium' or 'good' on sample size, or if they scored $\geq$ 70% on criteria i - vi but were rated as 'medium' on sample size. Studies were categorised as 'low' quality if they achieved 30 – 49% on criteria i-vi or were rated as 'poor' on sample size and 'very low' quality if they scored < 29% on criteria i - vi and/or 'insufficient' on sample size.

All studies were quality assessed by two reviewers (MM and IW). Disagreements in ratings were resolved through discussion. An intra-class correlation coefficient was calculated in IBM SPSS statistics software [25] to establish inter-rater reliability.

2.5. Methods of synthesis/analysis

The heterogeneity of study designs and reporting of the key outcome measures (IPQ version, administration of IPQ, descriptive and inferential statistics) precluded a meta-analysis; findings are therefore synthesised narratively.

Table 1
Quality rating algorithm.

Quality rating (%)	Operation	Sample size rating	Overall rating
> 70	and	Good	High
> 70	and	Medium	Medium
50 - 69	and	Medium or Good	Medium
30 - 49	and/or	Poor	Low
< 29	and/or	Insufficient	Very Low

3. Results

In total, 17 studies (Fig. 1) using a version of the IPQ to measure illness representations in 1364 people with seizure disorders (epilepsy $n = 961$, functional seizures $n = 403$) were identified (Table 2). The weighted mean age of people with epilepsy was 35.1 years old and 61.6% were female. The weighted mean age of people with functional seizures was 35.5 years old and 80.1% were female. Study designs included: cross-sectional ($n = 8$), case-control ($n = 5$), intervention ($n = 2$), cohort ($n = 1$), and 2 × 2 factorial ($n = 1$) designs. The BIPQ was the most frequently used measure of illness representations ($n = 9$), followed by the IPQ-R ($n = 6$), and the original IPQ ($n = 2$) (Table 2).

3.1. Quality assessment of studies

Included papers were assessed for quality in relation to the review question (Table 3). The average Intra Class Correlation Coefficient for quality ratings was 0.89 (95% CI, LL = 0.85, UL = 0.93); $F(118,118) = 9.26$, $p < .001$, indicating good inter-rater reliability.

No studies were rated as 'very low' quality, five were rated as 'low' quality, twelve were rated as 'medium' quality, and none were rated as 'high' quality. To assess for strengths and limitations across the remaining quality criteria for the included studies, mean scores were calculated for each individual criterion (i - vi). A failure to report on missing data was the criteria most responsible for reducing study quality (Mean = 0.65). The strongest quality criterion was 'demographics' (Mean = 2.6), followed by 'statistical analysis' (Mean = 2.3), 'recruitment' (Mean = 2.2), 'eligibility' (Mean = 2.1), and fidelity to the measure (Mean = 1.7), the latter reflecting a tendency not to report total reliability or subscale scores and/or whether the measure was adapted to the population under study.

3.2. Factors associated with illness representations in people with epilepsy and functional seizures

Fourteen studies measured illness representations in people with epilepsy with a version of the IPQ. These studies reported on associations between clinical characteristics, psychological distress, coping style, and quality of life, as well as comparisons with other chronic conditions without seizures. Eight included people with epilepsy only and six included people with epilepsy and people with functional seizures.

Nine studies reported on factors associated with illness perceptions in people with functional seizures. These studies also reported on associations between clinical characteristics, psychological distress, and quality of life, as well as comparisons with other chronic conditions without seizures. Three of these studies included people with functional seizures as the only seizure disorder group.

3.3. Clinical characteristics

Illness representations were associated with several clinical characteristics of epilepsy across studies. Positive correlations between more threatening IPQ scores and other epilepsy-related variables included duration [40], the number of antiepileptic drugs taken [40,33], and the number of adverse events a person had experienced [33]. Weak to moderate strength positive correlations between seizure frequency and 'emotional representation' ($r = 0.275$, $p < .05$), 'timeline acute/chronic' ($r = 0.236$, $p < .05$), 'consequences' ($r = 0.255$, $p < .001$), and 'timeline cyclical' scores ($r = 0.268$, $p < .01$) when controlling for gender and age [40] were reported. These results indicated that people with a higher frequency of seizures experienced greater distress as a result of their epilepsy, perceived their condition to be more chronic and fluctuating, and have greater impact on themselves and their families. Negative associations with epilepsy-related variables included age at seizure onset, which was inversely correlated with 'illness identity' scores ($r = -$

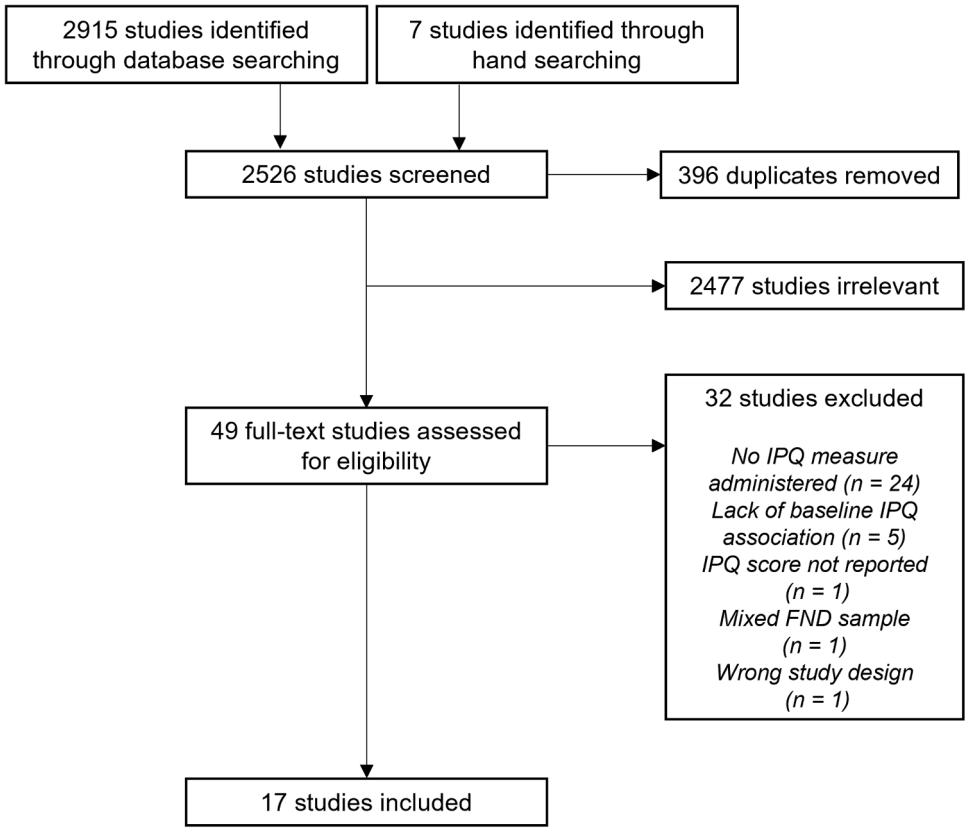

Fig. 1. PRISMA flow chart .

0.319, $p = .037$) [39], suggesting that patients whose seizures started earlier in life associated a greater number of symptoms with epilepsy. Seizure frequency was negatively correlated with 'controllability' scores, indicating that patients who experienced a greater number of seizures perceived their seizures to be less amenable to personal or treatment control [39,40]. People with epilepsy considered to have poor seizure control (defined as more than one seizure per month) scored more highly on the 'timeline' subscale ($p = .03$) than those with well-controlled epilepsy, suggesting that poor seizure control was associated with the belief that their epilepsy would last for longer [41]. Notably, 59% of the total sample in this study were considered non-adherent to their anti-epileptic medication, but adherence was not associated with seizure control. The above findings suggest that more threatening illness representations are associated with more severe epilepsy.

In contrast, only one study reported on associations between functional seizure clinical characteristics and IPQ scores. Tolchin et al. [34] found that lower baseline scores on the BIPQ were associated with nonadherence to psychiatric treatment, and higher scores on the BIPQ were significantly correlated with decreased odds of dropouts, with a hazard ratio of 0.77 (CI = 0.64 - 0.93) for every ten-point increment on the 80-point scale. The authors concluded that patients with more threatening perceptions of their illness are more likely to attend follow-up psychiatric appointments.

3.4. Psychological distress

Measures of psychological distress or psychopathology were also associated with IPQ scores in people with epilepsy. BIPQ total scores correlated positively with the somatisation, obsessive-compulsive, anxiety, and Global Severity Index subscales (all $p < .001$) of the SCL-90-R (a measure of psychopathology) in one study [33]. BIPQ total scores [32, 33] and the IPQ-R subscales of 'consequences' and 'emotional representation' [26] were positively correlated with more severe symptoms of depression (although the 'emotional representation' scale item asks about depression and so an association is perhaps to be expected). Conversely, the IPQ-R subscales of 'treatment control' and 'illness coherence' were negatively correlated with depression symptom severity [26]. However, Goldstein et al. [39] found that IPQ scores did not independently predict depression when controlling for coping style, and that when coping factors were controlled for, illness identity independently predicted anxiety scores [39]. Furthermore, Shallcross et al. [32] found no difference on BIPQ total or subscale scores between depressed and non-depressed people with epilepsy.

Three studies reported on the association between psychological distress/psychopathology and illness representations in people with functional seizures. BIPQ scores were positively associated with Emotional Processing Scale−25 scores ($r = 0.475$, $p < .01$), PHQ-15 scores ($r = 0.582$, $p < .05$), and CORE-10 scores ($r = 0.723$, $p < 0.01$) indicating that patients with more threatening illness representations experienced greater emotion dysregulation, a higher number of physical symptoms, and more severe psychological distress [28]. Cope et al. [37] found that prior to treatment with a Cognitive Behavioural Therapy psychoeducation group, people with functional seizures rated as 'high' in dissociative symptomology had higher BIPQ scores than those rated low in dissociative symptomology ($p = .009$). Finally, in people with functional seizures, perceived stigma correlated positively with 'timeline' only ($r_s = 0.38$, $p < 0.05$), meaning that people with functional seizures felt more stigmatised the longer they expected their functional seizures to last [30]. Taken together, these results suggest that more threatening illness representations tend to be associated with greater psychological distress in people with functional seizures as well as people with epilepsy.

Table 2
Summary characteristics from included studies.

Author and country	Design	Epilepsy N	Age (M)	Female (%)	Functional Seizures N	Age (M)	Female (%)	Other control Type (N)	Age (M)	Female (%)	IPQ	Other measures	Key finding
Cope et al. [26]UK	Intervention	–	–	–	25	NR	84	–	–	–	BIPQ	Seizure frequency, Seizure intensity, Attitudes to intervention[NV,] DES, Emotional Thermometer, PHQ-9, WSAS.	Participants higher in dissociation scored more highly on the BIPQ.
Evershed [27]UK	2 × 2 factorial	28	32	50	17	36	71	–	–	–	IPQ-R	IBQ, BSI, Mental health history questionnaire[NV.]	People with FS scored more highly on identity and timeline.
Goldstein et al. [28] UK	Cross-sectional	43	36	65	–	–	–	–	–	–	IPQ	Seizure frequency, WOC, HADS.	Illness identity scores independently predicted anxiety scores after controlling for coping. IPQ scores did not independently predict depression.
Gupta et al. [26] USA	Cross-sectional	55	41	62	–	–	–	–	–	–	IPQ-R	Seizure frequency, NDDI-E.	More threatening illness perceptions were associated with worse depression symptom severity.
Ji et al. [30] China	Case-control	117	27	52	–	–	–	87 (CLD)	39	17	CIPQ-R	SSRS, SCSQ.	People with ES had more limited understanding of their illness, poorer belief in control, and more negative emotional representations than CLD patients. IPQ scores were associated with social support, coping style, and ES characteristics.
Jones et al. [31]UK	Cross-sectional	54	38	54	–	–	–	–	–	–	IPQ	MSQ, BMQ, HADS.	Compared to patients with well-controlled ES, those with poorly-controlled ES thought their illness would last longer.
Lai et al. [27] Malaysia	Cross-sectional	154	37	53	–	–	–	–	–	–	BIPQ	HADS, Attitudes to intervention[NV] (e.g., willingness, barriers).	BIPQ scores positively correlated with willingness to participate in a positive psychological intervention.
Ludwig et al. [33] UK	Case-control	34	33	79	40	37	63	FW (n = 107) vs NDLW (n = 46)	39 39	79 83	IPQ-R	HADS	People with FS reported a low level of personal control, coherence, and a tendency to reject psychological explanations (less so than FW). People with FS had higher treatment control than people with FW. No differences between people with ES and NDLW.
Novakova et al. [28] UK	Cross-sectional	–	–	–	50	39[Md]	86	224 (HC)	32[Md]	86	BIPQ	Seizure frequency, CORE-10, SF-36, PHQ-15, EPS-25.	BIPQ significantly correlated with emotion processing difficulties, anxiety, psychological distress, and health-related quality of life.
Rawlings et al. [29] UK	Cross-sectional	62	38	69	45	40	91	–	–	–	BIPQ	GAD-7, NDDI-E, NEWQOL-6D, LSSS-3.	People with FS had more threatening illness perceptions. In both, illness perceptions were

(*continued on next page*)

Table 2 (*continued*)

Author and country	Design	Epilepsy N	Epilepsy Age (M)	Epilepsy Female (%)	Functional Seizures N	Functional Seizures Age (M)	Functional Seizures Female (%)	Other control Type (N)	Other control Age (M)	Other control Female (%)	IPQ	Other measures	Key finding
													negatively associated with quality of life.
Rawlings et al. [30] UK	Case-control	78	41[Md]	68	47	37[Md]	91	–	–	–	BIPQ	GAD-7, NDDI-E, NEWQOL-6D, LSSS-3.	In people with ES, perceived stigma correlated positively with consequences, timeline, symptoms, and emotional representation. In people with FS perceived stigma correlated positively with timeline.
Rawlings et al. [31] UK	RCT	27	44	76	16	38	88	–	–	–	BIPQ	Seizure characteristics, GAD-7, NIDDI-E, NEWQOL-6D, LSSS-3, Acceptability questionnaire (non-validated).	At baseline, people with FS scored more highly on BIPQ than people with ES.
Shallcross et al. [32] USA	Cross-sectional	70	38	49	–	–	–	–	–	–	BIPQ	NIDDI-E, QOLIE-31-P.	BIPQ scores mediate the relationship between depressive symptoms and quality of life.
Siarava et al. [33] Greece	Case-control	70	38	56	–	–	–	70	40	51	BIPQ	SCL-90R, clinical characteristics.	BIPQ was positively correlated with number of anti-epileptic drugs, Adverse Event Profile score, and the somatisation, obsessive compulsive, depression, anxiety, and Global Severity Index subscales of the SCL-90R.
Tolchin et al. [34] USA	Cohort	–	–	–	123	38	85	–	–	–	BIPQ	Semi-structured clinical interview to identify 'risk factors'.	Patients with more threatening illness perceptions were more likely to return for follow-up psychiatry appointments.
Tu et al. [35] China	Cross-sectional	135	27	79	–	–	–	–	–	–	CIPQ-R	QOLIE-31, SCSQ.	CIPQ-R correlated with quality of life. Coping style mediated association between CIPQ-R and quality of life.
Whitehead et al. [36] UK	Case-control	34	33	79	40	36	63	45 (Neur.)	45	36	IPQ-R	SAQ, LSSS-3, HADS, QOLIE-31.	Differences in illness representations between people with FS and ES < neurologists' perceptions of the disorder.

Note. BIPQ = Brief Illness Perception Questionnaire, BMQ = Beliefs about Medicines Questionnaire, CIPQ-R = Chinese Revised Illness Perception Questionnaire, CLD = Chronic Liver Disease, CORE-10 = Clinical Outcomes in Routine Evaluation – 10 (psychological distress), DES = Dissociative Experiences Scale, EPS-25 = Emotional Processing Scale –25, ES = Epilepsy, FS = Functional Seizures, FW = Functional Weakness, GAD-7 = Generalized Anxiety Disorder Scale – 7, HADS = Hospital Anxiety and Depression Scale, IBQ = Illness Beliefs Questionnaire, IPQ = Illness Perception Questionnaire, IPQ-*R* = Revised Illness Perception Questionnaire, LSSS-3 = Liverpool Seizure Severity Scale –3, [Md] = Median, MSQ = Morisky Scale Questionnaire, Neur. = Neurologists, NDLW = Non-disease causing Leg Weakness, NEWQOL-6D = Quality of Life Adjusted Years in Epilepsy measure, NIDDI-*E* = Neurological Disorders Depression Inventory for Epilepsy, NR = Measure of central tendency not reported, [NV] = Non-validated measure, PHQ-9 = Patient Health Questionnaire – 9 (depression), PHQ-15 = Patient Health Questionnaire – 15 (somatization), QOLIE-31 = Quality of Life in Epilepsy Inventory – 31, SAQ = Symptom Attribution Questionnaire, SCL-90 *R* = 90 item Revised Symptom Check List, SCSQ = Simplified Coping Scale Questionnaire, SF-36 = Short-Form 36 (Health-related quality of life), WOC = Ways of Coping Scale, WSAS = Work Social and Adjustment Scale.

3.5. Coping styles

Several studies demonstrated the close interplay between illness representations and coping styles in people with epilepsy. 'Active coping' (analogous to 'problem-focused' coping [43]) on the Simplified Coping Style Questionnaire [SCSQ; 44] was positively associated with 'personal control' [40], 'illness coherence' [35], and 'treatment control' [40,35] and negatively associated with 'emotional representation' [35] on the Chinese IPQ-R [CIPQ-R; 45]. Conversely, 'passive coping' (cf. 'emotion-focused' coping [43]) was positively associated with

Table 3
Quality assessment of included studies.

Author	Participants Recruitment	Eligibility	Demographics	Measurement Fidelity	Analysis Statistics	Missing data	Scoring Total score	Total score (%)	Sample size	Overall rating
Cope et al. [37]	1	1	3	2	2	2	11	52.4	Poor	Low
Evershed [38]	3	2	3	2	2	0	12	57.1	Poor	Low
Goldstein et al. [39]	1	2	3	3	3	0	12	57.1	Moderate	Medium
Gupta et al. [26]	2	2	3	3	3	0	13	61.9	Moderate	Medium
Ji et al. [40]	1	2	3	3	2	0	11	52.4	Good	Medium
Jones et al. [41]	3	2	2	2	2	0	11	52.4	Moderate	Medium
Lai et al. [27]	3	2	3	2	2	0	12	57.1	Good	Medium
Ludwig et al. [42]	3	3	2	2	2	2	17	81.0	Moderate	Medium
Novakova et al. [28]	3	2	2	1	2	3	14	66.7	Moderate	Medium
Rawlings et al. [29]	2	2	2	1	2	2	11	52.4	Moderate	Medium
Rawlings et al. [30]	2	2	2	0	2	0	8	38.1	Moderate	Low
Rawlings et al. [31]	3	2	2	0	2	2	10	47.6	Moderate	Low
Shallcross et al. [32]	1	2	3	1	3	0	10	47.6	Good	Low
Siarava et al. [33]	3	2	3	0	2	0	12	57.1	Good	Medium
Tolchin et al. [34]	3	3	3	0	3	0	12	57.1	Good	Medium
Tu et al. [35]	1	3	3	3	3	0	12	57.1	Good	Medium
Whitehead et al. [36]	3	3	2	3	2	0	13	61.9	Moderate	Medium

'consequences', 'timeline cyclical', [35], 'identity' [40], and 'emotional representation' [40,35] and negatively associated with 'illness coherence' [40,35]. Furthermore, Goldstein et al. [39] found that escape avoidant coping styles on the Ways of Coping Questionnaire [46] were positively associated with scores on the 'identity', and 'consequences' subscales of the IPQ, whereas planful problem-solving coping styles were associated with higher scores on 'controllability/cure subscale'. One study found that people with epilepsy who had more threatening illness perceptions were more willing to participate in a positive psychological intervention designed to help patients cope with their epilepsy ($r = 0.265$, $p < 0.01$) [27]. These findings suggest that more adaptive coping is related to less threatening illness representations in epilepsy.

No studies reported on associations between IPQ measures and coping styles in people with functional seizures.

3.6. Quality of life

Given the observed associations between IPQ scores, clinical characteristics, distress, and coping, it is perhaps unsurprising that IPQ scores have also been observed to be associated with quality of life in people with epilepsy. BIPQ total as well as 'timeline acute/chronic', 'timeline cyclical', 'consequences', and 'emotional representation' subscores on the CIPQ-R were negatively correlated with scores on the 31-item Quality of Life in Epilepsy Inventory (QOLIE-31) [QOLIE-31; 47] a self-report measure of health-related quality of life in epilepsy [32,35]. This finding suggests that patients who perceived their epilepsy to be more threatening, chronic, unpredictable, distressing, and to have greater impact on their lives, experienced poorer health-related quality of life. Some caution should be taken when interpreting significant correlations between the IPQ subscale of 'consequences' and quality of life scales; these measures are arguably capturing similar constructs and so an identified association may be unsurprising. Conversely, the 'illness coherence', 'personal control', and 'treatment control' subscales positively correlated with higher scores on a quality of life measure [35], indicating that patients who perceived themselves to have a better understanding of their epilepsy and considered their epilepsy to be more controllable, reported better health-related quality of life. Indeed, other researchers found that BIPQ total scores were significant predictors of health-related quality of life in people with epilepsy, accounting for 23.1% of variance ($p < .001$) [29]. Similarly, structural equation modelling results demonstrated that IPQ-R scores explained 77.5% of the variance in quality of life ($\beta = -0.775$, $p < .001$) and that IPQ-R scores had a direct impact on quality of life ($\beta = -0.6$, $p = .001$) [35]. Furthermore, BIPQ scores indirectly mediated the relationship between depressive symptoms and quality of life (CI $= -0.72, -0.22$, $p < .05$), and effects were robust when controlling for confounding variables such as age, sex, ethnicity, income, and seizure frequency [32]. This finding suggests that illness representations may influence the extent to which distress impacts upon quality of life in people with epilepsy. Indeed, perceived stigma correlated positively with 'consequences' ($r_s = 0.45$, $p < 0.001$),'timeline' ($r_s = 0.23$, $p < .05$), 'symptoms' ($r_s = 0.42$, $p = .05$), and 'emotional representation' ($r_s = 0.46$, $p < 0.001$) in people with epilepsy. This suggests that people with epilepsy felt more stigmatised the longer they had lived with the condition, the more symptoms they experienced, and the greater the perceived impact on their lives [30].

Health-related quality of life was associated with IPQ scores in two studies of people with functional seizures. BIPQ scores were negatively correlated with physical health- ($r = -0.442$, $p < .01$), and mental health-related quality of life ($r = -0.697$, $p < .1$) [28]. Furthermore, Rawlings et al. found that BIPQ total scores were significant predictors of health-related quality of life, accounting for 23.3% ($p = .02$) of variance [29]. 'Personal control' was the strongest predictor in people with functional seizures, meaning that of all the BIPQ subscales, the extent to which patients with people with functional seizures felt they could personally influence their disorder had the greatest impact on their quality of life. These studies indicated that, like people with epilepsy, people with functional seizures who have more threatening illness perceptions experience poorer health-related quality of life.

3.7. Comparison to chronic health conditions without seizures

To parse illness representations relating to epilepsy from those related to living with a chronic health condition, two studies compared illness representations in people with epilepsy or functional seizures to other patients with chronic health conditions without seizures. People with epilepsy scored lower on the 'identity' and 'consequences' subscales (both $p < .001$), but more highly on the 'emotional representation' subscale ($p = .046$) of the C-IPQR than patients with chronic liver disease [40]. Therefore, people with epilepsy associated fewer symptoms with their disorder and considered it to have a less severe effects on themselves and their family but had more negative emotional responses to their illness than patients with chronic liver disease. In a separate study, when compared to people with neurological disease causing leg weakness, people with epilepsy perceived themselves to have less personal control ($p < .001$) and for their condition to be more cyclical ($p = .048$) [42].

One study compared people with functional seizures to patients with

FND without seizures (functional limb weakness). People with functional seizures scored more highly on a single item indicating level of agreement with stress as causal ($p = .004$), 'consequences' ($p = .019$), treatment control ($p = .004$), and 'timeline (acute/chronic)' ($p = .041$), than patients with functional weakness [42]. This indicated that, compared to people with functional weakness, people with functional seizures perceived their condition to be more likely caused by stress, have a greater impact on the lives, be more controllable by treatment, and for it to last for longer (people with functional seizures did indeed have a significantly longer duration than those with functional weakness). However, the groups were similar in the extent to which they viewed their condition to be cyclical in nature, their degree of personal control, and on emotional impact. Overall, these findings do not clearly isolate illness representations to the presence of epilepsy or functional seizures in the context of comparable chronic conditions.

3.8. Comparisons of illness representations between people with epilepsy and functional seizures

Five studies were identified in which IPQ scores were measured and compared between people with epilepsy and people with functional seizures.

In two studies, people with functional seizures scored more highly on the BIPQ than people with epilepsy; this included the 'consequences' [29,30], 'treatment control' [29,30], 'identity/symptoms' [29,30], 'concern' [29,30], and 'emotional representation' subscales [29,30]. An exception to this pattern was the 'timeline' subscale – on which people with epilepsy scored more highly [30]. The two groups did not differ on levels of 'understanding' and 'personal control' [29,30]. The same research group later reported that at baseline of a pilot randomised controlled trial for a therapeutic writing intervention, people with functional seizures had more threatening illness representations on the total BIPQ score ($M = 55$, SD = 16.25) than people with epilepsy ($M = 47$, SD = 20.23) ($p < .001$) [31]. These results suggest that people with functional seizures have more threatening illness representations overall than people with epilepsy.

In contrast, Evershed [38] identified few differences in the IPQ-R scores of people with epilepsy and people with functional seizures pre- and post-diagnosis; both groups viewed their symptoms as beyond their personal control and endorsed psychological causes to a similar extent. However, people with functional seizures rated their expected duration as longer and associated a greater number of symptoms with their seizure disorder. Similarly, Whitehead et al., found no differences between people with epilepsy and people with functional seizures on IPQ-R total subscale scores [36]. Therefore, comparisons of IPQ scores between people with functional seizures and people with epilepsy yielded mixed results across studies.

4. Discussion

The findings of this review provide moderately strong evidence that more threatening illness representations are associated with poorer biopsychosocial outcomes for people with seizure disorders. These include worse seizure-related characteristics, responses to treatment, greater psychological distress, less adaptive coping styles (epilepsy only), poorer health-related quality of life, and greater perceived stigma. However, the relationship between illness representations and outcomes may not be direct; for example, coping style (active and passive) was found to mediate the relationship between illness representations and quality of life [35] as well as the relationship between illness representations and depression [39] in people with epilepsy. This highlights the potentially complex interplay between the way people think about their seizure disorders, their responses to these thoughts, and the impact their disorder has on their life.

At present, it is unclear whether people with functional seizures and people with epilepsy differ in terms of their illness representations. Although differences between the two groups have been identified on other relevant characteristics, including emotion dysregulation and dissociative symptomology [e.g., 23], the relatively small number of direct between-group comparisons make it difficult to draw firm conclusions comparing the two seizure disorders on their illness representations. There is also a lack of concordance between studies comparing functional seizures with epilepsy, which may be driven by methodological issues identified in the quality assessment, including failure to statistically control for between-group differences in demographic or clinical characteristics (e.g., gender, seizure frequency, and duration) as well as small sample sizes. Indeed, there are well-established challenges to recruitment and retention in neurological research including difficulties with access to patient populations, motivation for patients to engage with research at a difficult point in their lives, and strict eligibility criteria [48].

The cross-sectional design of these studies does not permit interpretation of causality. The relationship between illness representations and outcomes in seizure disorders is likely to be bi-directional in many instances; for example, increased seizure frequency may cause people to have more threatening perceptions of their disorder [e.g., 40], and feeling more threatened by an increased number of seizures may in turn heighten stress and trigger further seizures [49]. Longitudinal designs would help to establish how or why illness representations develop in people with seizure disorders. In addition, the relatively small number of studies investigating cross-sectional associations between IPQ scores and outcomes in people with functional seizures alone decreases the confidence with which it can be stated that illness representations are associated with biopsychosocial factors in this group of patients. Similarly, the relatively small number of studies comparing patients with seizure disorders to those with comparable chronic conditions without seizures, as well as a failure to control for important differences in demographic and clinical variables such as age and duration that may influence illness representations, raises the possibility that any between-groups differences were attributable to factors other than the experience of seizures. Therefore it is difficult to isolate illness representations to the presence or absence of seizures.

The findings of this review do however complement other systematic reviews of illness representations in people with seizure disorders. A systematic review of qualitative studies investigating the narratives of people with functional seizures identified five key themes relating to experiences of diagnosis, treatment and management, seizure and emotional events, and impact on daily life [19]. It was reported that people with functional seizures experience their seizures as threatening, confusing, and as having significant consequences for their daily lives. This contrasts somewhat with the findings of a systematic review of healthcare practitioners' perceptions of functional seizures [20], who seem to share patient perceptions of uncertainty (lack of coherence), lack of treatment control and severity of consequences, but perceive epilepsy to have more severe consequences than functional seizures - even though the findings of the present review suggest that there is insufficient evidence to conclude that people with epilepsy and people with functional seizures rate themselves as having significantly different illness representations. This mismatch between patient and healthcare practitioner illness representations about seizure disorders may reflect the stigma and mistreatment experienced by people with functional seizures from healthcare professionals [50].

The present finding of relationships between more threatening illness representations and poorer biopsychosocial outcomes such as distress in patients with seizure disorders is a pattern echoed across other chronic health condition populations. Illness representations have been shown to vary according to age and gender in patients with coronary heart disease [51], be related to survival/mortality in end-stage

renal patients [52], distress in stroke patients [53], and outcomes in long-term conditions including chronic pain and rheumatoid arthritis [54]. This consistency of this finding across health conditions supports the assertion that illness representations are indeed related to how patients respond to and manage their illness.

4.1. Limitations

Several limitations relating to the evidence base in this field are important to highlight. These include a lack of reporting on how missing data are dealt with and demonstrating fidelity to the measure (i.e. adapting the IPQ as necessary and reporting on subscale or total scale reliability). Authors also tended not to report total and subscale scores of their respective illness perception questionnaire measures, precluding a numerical synthesis of findings.

Factors relating to the IPQ itself also presented challenges to the review process. Firstly, there are no agreed cut-offs for what would constitute a 'threatening' versus a 'non-threatening' illness perception according to the IPQ. This creates a lack of clarity when interpreting IPQ scores. Cut-offs have been suggested by some researchers, for example, Kuiper et al. [55] recently proposed scores < 42 on the BIPQ to represent low experienced threat, 42–49 to represent moderate experienced threat and ≥ 50 to represent high experienced threat in a sample of patients with recently acquired spinal cord injuries, however, these cut-offs have not been universally agreed or validated in other samples. Secondly, although the IPQ was developed to measure illness representations according to the Common-Sense Model [7], there is no numerical subscale for causal representations, making statistical analyses on this domain more difficult. Some authors have attempted to address this issue by using additional self-report measures to assess causal beliefs (so called 'symptom attribution measures'), but these were non-validated [e.g., 56].

There are also limitations pertaining to the review itself. Firstly, it was only possible for one reviewer to initially screen and select titles, abstracts, and full texts. It was also decided that owing to the heterogeneity of study designs and reporting, a single generic quality assessment tool should be developed. Although this means that the quality assessment was as parsimonious as possible and tailored to the included papers as well as typical issues of quality in this research area (e.g., verification of seizure disorder diagnosis), it limits the validity of the quality assessment as well as comparability to tools used in other reviews. Finally, an additional limitation may be that a significant proportion (40%) of included studies were identified through hand searches of reference lists. This increases the risk that some relevant studies were not identified, however, a concerted attempt to mitigate this risk was made by thoroughly hand searching reference lists of included studies, subsequent studies that had cited identified studies, and other systematic reviews.

4.2. Implications of review

The findings of this review have clinical implications people with seizure disorders. The identified relationships between illness representations and biopsychosocial factors or outcomes in seizure disorders support the assertion that the way people think about their seizure disorder is related to how they respond to and manage their illness [7]. Therefore, therapy-associated changes in illness representations may bring about changes in important aspects of patients' experiences, such as symptomology or distress. Indeed, there is preliminary evidence from uncontrolled trials of psychoeducation and mindfulness-based interventions, that illness representations are amenable to change in people with functional seizures [37,57-59]. This means that, whilst acknowledging the likely bi-directional relationship between illness representations and outcomes, it is important for clinicians to ask patients not only what they believe about their seizure disorders, but also how that belief makes them feel and what impact that has on their life.

This review also highlights the importance of clear communication around diagnosis to facilitate the development of adaptive illness representations. This is increasingly recognised amongst clinicians working with FND [e.g., 60,61], who have developed diagnostic communication protocols. Based on the findings that people with seizure disorders who have more threatening illness perceptions are more likely to attend follow-up psychological and psychiatric care appointments [27,34], these protocols could help to develop balanced illness representations by emphasising the potentially serious impact of seizure disorders on quality of life as well as the possibility for improvement with psychological intervention.

Furthermore, potentially modifiable factors that mediate the relationship between illness representations and outcomes would appear to be suitable targets for psychological treatment; 'coping style' was one such factor identified in this review. For example, the observed relationship between more threatening illness representations and greater willingness to participate in a psychological intervention [27] may be partly explained by a self-identified need to improve coping skills in therapy. It would be clinically useful for researchers to establish if there are any other modifiable factors beyond active or passive coping strategies that mediate or moderate the relationship between illness representations and outcomes in patients with seizure disorders.

Future research could seek to address the identified gaps in the literature relating to cross-sectional associations between functional seizure-related characteristics (such as coping styles) and illness representations. Work aiming to establish the presence or absence of differences in illness representations between people with epilepsy and people with functional seizures or other comparable conditions without seizures would help to clarify the specificity of illness representations to patient populations and therefore develop more targeted psychological interventions. As an individual's management of their chronic illness is thought to be influenced by their social network [62], it would also be valuable to include carers or relatives in studies investigating the impact of psychological intervention on illness representations; none of the studies included in this review did so.

6 Conclusions

There is moderately strong evidence that more threatening or negative illness representations are associated with poorer biopsychosocial outcomes in people with seizure disorders, although the evidence is somewhat weaker for people with functional seizures than for people with epilepsy. There is insufficient evidence to conclude with confidence that differences exist between the illness representations of people with epilepsy and people with functional seizures. Further cross-sectional research is needed to improve understanding of factors associated with illness representations in people with functional seizures. Longitudinal designs could help to establish how or why certain illness representations develop in both seizure disorder groups.

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Registration

This review was not registered.

Declaration of Competing Interest

The authors have no conflicts of interest to declare.

References

[1] Moshé SLP, Perucca EP, Ryvlin PP, Tomson TP. Epilepsy: new advances. Lancet (British edition) 2015;385:884–98. https://doi.org/10.1016/S0140-6736(14)60456-6.

[2] LaFrance Jr WC, Baker GA, Duncan R, Goldstein LH, M Reuber. Minimum requirements for the diagnosis of psychogenic nonepileptic seizures: a staged approach. Epilepsia 2013;54:2005–18. https://doi.org/10.1111/epi.12356.

[3] Fiest KM, Sauro KM, Wiebe S, Patten SB, Kwon CS, Dykeman J, Pringsheim T, Lorenzetti DL, Jetté N. Prevalence and incidence of epilepsy: a systematic review and meta-analysis of international studies. Neurology 2017;88:296–303. https://doi.org/10.1212/wnl.0000000000003509.

[4] Villagrán A, Eldøen G, Duncan R, Aaberg KM, Hofoss D, Lossius MI. Incidence and prevalence of psychogenic nonepileptic seizures in a Norwegian county: a 10-year population-based study. Epilepsia 2021;62:1528–35. https://doi.org/10.1111/epi.16949.

[5] Terman SW, Hill CE, Burke JF. Disability in people with epilepsy: a nationally representative cross-sectional study. Epilepsy Behav 2020;112:107429. https://doi.org/10.1016/j.yebeh.2020.107429.

[6] Carson A, Stone J, Hibberd C, Murray G, Duncan R, Coleman R, Warlow C, Roberts R, Pelosi A, Cavanagh J, Matthews K, Goldbeck R, Hansen C, Sharpe M. Disability, distress and unemployment in neurology outpatients with symptoms 'unexplained by organic disease. J Neurol Neurosurg Psychiatry 2011;82:810–3. https://doi.org/10.1136/jnnp.2010.220640.

[7] Leventhal H, Phillips LA, Burns E. The Common-Sense Model of Self-Regulation (CSM): a dynamic framework for understanding illness self-management. J Behav Med 2016;39:935–46. https://doi.org/10.1007/s10865-016-9782-2.

[8] Hagger MS, Orbell S. A meta-analytic review of the common-sense model of illness representations. Psychol Health 2003;18:141–84. https://doi.org/10.1080/0887044031000813 21.

[9] O' Rourke G, O' Brien JJ. Identifying the barriers to antiepileptic drug adherence among adults with epilepsy. Seizure 2017;45:160–8. https://doi.org/10.1016/j.seizure.2016.12.006.

[10] Chapman SCE, Horne R, Chater A, Hukins D, Smithson WH. Patients' perspectives on antiepileptic medication: relationships between beliefs about medicines and adherence among patients with epilepsy in UK primary care. Epilepsy Behav 2014;31:312–20. https://doi.org/10.1016/j.yebeh.2013.10.016.

[11] National Institute for Health and Care Excellence. Epilepsies in children, young people and adults. NICE; 2022.

[12] Michaelis R, Tang V, Nevitt SJ, Wagner JL, Modi AC, LaFrance Jr WC, Goldstein LH, Gandy M, Bresnahan R, Valente K, Donald KA, Reuber M. Psychological treatments for people with epilepsy. Cochrane Database Syst Rev 2020;8:Cd012081. https://doi.org/10.1002/14651858.CD012081.pub3.

[13] Goldstein LH, Robinson EJ, Mellers JDC, Stone J, Carson A, Reuber M, Medford N, McCrone P, Murray J, Richardson MP, Pilecka I, Eastwood C, Moore M, Mosweu I, Perdue I, Landau S, Chalder T. Cognitive behavioural therapy for adults with dissociative seizures (CODES): a pragmatic, multicentre, randomised controlled trial. Lancet Psychiatry 2020;7:491–505. https://doi.org/10.1016/s2215-0366(20)30128-0.

[14] Brown RJ, Reuber M. Towards an integrative theory of psychogenic non-epileptic seizures (PNES). Clin Psychol Rev 2016;47:55–70. https://doi.org/10.1016/j.cpr.2016.06.003.

[15] Edwards MJ, Adams RA, Brown H, Pareés I, Friston KJ. A Bayesian account of 'hysteria. Brain 2012;135:3495–512. https://doi.org/10.1093/brain/aws129.

[16] Weinman J, Petrie KJ, Moss-morris R, Horne R. The illness perception questionnaire: a new method for assessing the cognitive representation of illness. Psychol Health 1996;11:431–45. https://doi.org/10.1080/08870449608400270.

[17] Moss-Morris R, Weinman J, Petrie K, Horne R, Cameron L, Buick D. The Revised Illness Perception Questionnaire (IPQ-R). Psychol Health 2002;17:1–16. https://doi.org/10.1080/08870440290001494.

[18] Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. J Psychosom Res 2006;60:631–7. https://doi.org/10.1016/j.jpsychores.2005.10.020.

[19] Rawlings GH, Reuber M. What patients say about living with psychogenic nonepileptic seizures: a systematic synthesis of qualitative studies. Seizure 2016;41:100–11. https://doi.org/10.1016/j.seizure.2016.07.014.

[20] Rawlings GH, Reuber M. Health care practitioners' perceptions of psychogenic nonepileptic seizures: a systematic review of qualitative and quantitative studies. Epilepsia 2018;59:1109–23. https://doi.org/10.1111/epi.14189.

[21] Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, Shamseer L, Tetzlaff JM, Akl EA, Brennan SE, Chou R, Glanville J, Grimshaw JM, Hróbjartsson A, Lalu MM, Li T, Loder EW, Mayo-Wilson E, McDonald S, McGuinness LA, Stewart LA, Thomas J, Tricco AC, Welch VA, Whiting P, Moher D. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. https://doi.org/10.1136/bmj.n71.

[22] Cohen J. Statistical power analysis for the behavioural sciences. Hillside: Erlbaum; 1988.

[23] Williams IA, Levita L, Reuber M. Emotion dysregulation in patients with psychogenic nonepileptic seizures: a systematic review based on the extended process model. Epilepsy Behav 2018;86:37–48. https://doi.org/10.1016/j.yebeh.2018.06.049.

[24] Brown RJ, Reuber M. Psychological and psychiatric aspects of psychogenic non-epileptic seizures (PNES): a systematic review. Clin Psychol Rev 2016;45:157–82. https://doi.org/10.1016/j.cpr.2016.01.003.

[25] IBM Corp. IBM SPSS statistics for windows. Armonk, NY: IBM Corp; 2017. 25.0 editor.

[26] Gupta SK, Margolis SA, Grant AC, Gonzalez JS, Nakhutina L. Relationships among illness representations and depressive symptom severity in predominantly African-American and Caribbean-American people with epilepsy. Clin Neuropsychol 2022;36:462–78. https://doi.org/10.1080/13854046.2021.1923802.

[27] Lai ST, Lim KS, Tang V, Low WY. Positive psychological interventions for people with epilepsy: an assessment on factors related to intervention participation. Epilepsy Behav 2018;80:90–7. https://doi.org/10.1016/j.yebeh.2017.12.019.

[28] Novakova B, Howlett S, Baker R, Reuber M. Emotion processing and psychogenic non-epileptic seizures: a cross-sectional comparison of patients and healthy controls. Seizure (London, England) 2015;29:4–10. https://doi.org/10.1016/j.seizure.2015.03.007.

[29] Rawlings GH, Brown I, Reuber M. Predictors of health-related quality of life in patients with epilepsy and psychogenic nonepileptic seizures. Epilepsy Behav 2016;68:153–8. https://doi.org/10.1016/j.yebeh.2016.10.035.

[30] Rawlings GH, Brown I, Reuber M. Deconstructing stigma in psychogenic nonepileptic seizures: an exploratory study. Epilepsy Behav 2017;74:167–72. https://doi.org/10.1016/j.yebeh.2017.06.014.

[31] Rawlings GH, Brown I, Stone B, Reuber M. A pilot randomised controlled trial of a home-based writing intervention for individuals with seizures. Psychol Health 2018;33:1151–71. https://doi.org/10.1080/08870446.2018.1478974.

[32] Shallcross AJ, Becker DA, Singh A, Friedman D, Montesdeoca J, French J, Devinsky O, Spruill TM. Illness perceptions mediate the relationship between depression and quality of life in patients with epilepsy. Epilepsia (Copenhagen) 2015;56:e186–90. https://doi.org/10.1111/epi.13194.

[33] Siarava E, Markoula S, Pelidou S-H, Kyritsis AP, Hyphantis T. Psychological distress symptoms and illness perception in patients with epilepsy in Northwest Greece. Epilepsy Behav 2020;102:106647. https://doi.org/10.1016/j.yebeh.2019.106647.

[34] Tolchin B, Dworetzky BA, Baslet G. Long-term adherence with psychiatric treatment among patients with psychogenic nonepileptic seizures. Epilepsia (Copenhagen) 2018;59:e18–22. https://doi.org/10.1111/epi.13969.

[35] Tu H, Gong G, Zhang S, Fu Y, Wang T, Chu Q, Hu S, Wang K, Zhu C, Fan Y. The association between illness perception and quality of life among Chinese adults with epilepsy: the mediating role of coping style. Epilepsy Behav 2022;130:108677. https://doi.org/10.1016/j.yebeh.2022.108677.

[36] Whitehead K, Kandler R, Reuber M. Patients' and neurologists' perception of epilepsy and psychogenic nonepileptic seizures. Epilepsia (Copenhagen) 2013;54:708–17. https://doi.org/10.1111/epi.12087.

[37] Cope SR, Smith JG, King T, Agrawal N. Evaluation of a pilot innovative cognitive-behavioral therapy-based psychoeducation group treatment for functional non-epileptic attacks. Epilepsy Behav 2017;70:238–44. https://doi.org/10.1016/j.yebeh.2017.02.014.

[38] Evershed, K. (2007). *Investigation of Illness Perceptions and Beliefs in People With Non-Epileptic Attack Disorder Pre- and Post-Diagnosis, in Comparison to Individuals With Epilepsy* (Order No. 13805168). Available from ProQuest Dissertations & Theses Global. (2169445090). https://www.proquest.com/dissertations-theses/investigation-illness-perceptions-beliefs-people/docview/2169445090/se-2.

[39] Goldstein LH, Holland L, Soteriou H, Mellers JDC. Illness representations, coping styles and mood in adults with epilepsy. Epilepsy Res. 2005;67:1–11. https://doi.org/10.1016/j.eplepsyres.2005.06.008.

[40] Ji H, Zhang L, Li L, Gong G, Cao Z, Zhang J, Zhou N, Wang Y, Tu H, Wang K. Illness perception in Chinese adults with epilepsy. Epilepsy Res. 2016;128:94–101. https://doi.org/10.1016/j.eplepsyres.2016.10.018.

[41] Jones RM, Butler JA, Thomas VA, Peveler RC, Prevett M. Adherence to treatment in patients with epilepsy: associations with seizure control and illness beliefs. Seizure (London, England) 2006;15:504–8. https://doi.org/10.1016/j.seizure.2006.06.003.

[42] Ludwig L, Whitehead K, Sharpe M, Reuber M, Stone J. Differences in illness perceptions between patients with non-epileptic seizures and functional limb weakness. J Psychosom Res 2015;79:246–9. https://doi.org/10.1016/j.jpsychores.2015.05.010.

[43] Folkman S, Lazarus RS. Coping as a Mediator of Emotion. J Pers Soc Psychol 1988;54:466–75. https://doi.org/10.1037/0022-3514.54.3.466.

[44] Xie YN. Preliminary study to the reliability and validity of the simple coping style scale. Chin J Clin Psychol 1998;6:114–5.

[45] Chen J, Zhang H, Suo R, Xiong C, Zhang X, Zhang Y, Yan J. Adaptation and psychometric testing of the Chinese version of the Revised Illness Perception Questionnaire for cervical cancer patients. Eur J Oncol Nurs 2020;48:101799. https://doi.org/10.1016/j.ejon.2020.101799.

[46] Folkman S, Lazarus RS. Ways of coping questionnaire manual. California, USA: MIND GARDEN; 1988.

[47] Cramer JA, Perrine K, Devinsky O, Bryant-Comstock L, Meador K, Hermann B. Development and cross-cultural translations of a 31-item quality of life in epilepsy inventory. Epilepsia 1998;39:81–8. https://doi.org/10.1111/j.1528-1157.1998.tb01278.x.

[48] Newberry A, Sherwood P, Hricik A, Bradley S, Kuo J, Crago E, et al. Understanding recruitment and retention in neurological research. J Neurosci Nurs 2010. https://doi.org/10.1097/jnn.0b013e3181c1fdd9.

[49] Balamurugan E, Aggarwal M, Lamba A, Dang N, Tripathi M. Perceived trigger factors of seizures in persons with epilepsy. Seizure 2013;22:743–7. https://doi.org/10.1016/j.seizure.2013.05.018.

[50] MacDuffie KE, Grubbs L, Best T, LaRoche S, Mildon B, Myers L, Stafford E, Rommelfanger KS. Stigma and functional neurological disorder: a research agenda targeting the clinical encounter. CNS Spectr 2021;26:587–92. https://doi.org/10.1017/S1092852920002084.

[51] Al-Smadi AM, Ashour A, Hweidi I, Gharaibeh B, Fitzsimons D. Illness perception in patients with coronary artery disease: a systematic review. Int J Nurs Pract 2016; 22:633–48. https://doi.org/10.1111/ijn.12494.
[52] Parfeni M, Nistor I, Covic A. A systematic review regarding the association of illness perception and survival among end-stage renal disease patients. Nephrol Dial Transplant 2013;28:2407–14. https://doi.org/10.1093/ndt/gft194.
[53] Pai HC, Li CC, Tsai SM, Pai YC. Association between illness representation and psychological distress in stroke patients: a systematic review and meta-analysis. Int J Nurs Stud 2019;94:42–50. https://doi.org/10.1016/j.ijnurstu.2019.01.015.
[54] Rivera E, Corte C, DeVon HA, Collins EG, Steffen A. A systematic review of illness representation clusters in chronic conditions. Res Nurs Health 2020;43:241–54. https://doi.org/10.1002/nur.22013.
[55] Kuiper H, van Leeuwen CMC, Stolwijk-Swüste JM, Post MWM. Reliability and validity of the Brief Illness Perception Questionnaire (B-IPQ) in individuals with a recently acquired spinal cord injury. Clin Rehabil 2022;36:550–7. https://doi.org/10.1177/02692155211061813.
[56] Chen DK, Majmudar S, Ram A, Rutherford HC, Fadipe M, Dunn CB, Collins RL. Change in illness perception is associated with short-term seizure burden outcome following video-EEG confirmation of psychogenic nonepileptic seizures. Epilepsy Behav 2018;83:186–91. https://doi.org/10.1016/j.yebeh.2018.03.007.
[57] Baslet G, Ehlert A, Oser M, Dworetzky BA. Mindfulness-based therapy for psychogenic nonepileptic seizures. Epilepsy Behav 2020;103:106534. https://doi.org/10.1016/j.yebeh.2019.106534.
[58] Sarudiansky M, Pablo Korman G, Lanzillotti AI, Areco Pico MM, Tenreyro C, Paolasini GV, Wolfzun C, Kochen S, D'Alessio L, Myers L. Report on a psychoeducational intervention for psychogenic non-epileptic seizures in Argentina. Seizure (London, England) 2020;80:270–7. https://doi.org/10.1016/j.seizure.2020.04.008.
[59] Wiseman H, Mousa S, Howlett S, Reuber M. A multicenter evaluation of a brief manualized psychoeducation intervention for psychogenic nonepileptic seizures delivered by health professionals with limited experience in psychological treatment. Epilepsy Behav 2016;63:50–6. https://doi.org/10.1016/j.yebeh.2016.07.033.
[60] Cope SR, Smith JG, Edwards MJ, Holt K, Agrawal N. Enhancing the communication of functional neurological disorder diagnosis: a multidisciplinary education session. Eur J Neurol 2021;28:40–7. https://doi.org/10.1111/ene.14525.
[61] Hall-Patch L, Brown R, House A, Howlett S, Kemp S, Lawton G, et al. Acceptability and effectiveness of a strategy for the communication of the diagnosis of psychogenic nonepileptic seizures. Epilepsia (Copenhagen) 2010;51:70–8. https://doi.org/10.1111/j.1528-1167.2009.02099.x.
[62] Vassilev I, Rogers A, Kennedy A, Koetsenruijter J. The influence of social networks on self-management support: a metasynthesis. BMC Public Health 2014;14:719. https://doi.org/10.1186/1471-2458-14-719.