

## Correlates of Stigma in Adults with Epilepsy: A Systematic Review of Quantitative Studies

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## **Highlights**

- The present study examined correlates of stigma in people with epilepsy
- Thirty-three articles reporting on 25 quantitative studies were identified
- Stigma was associated with demographic, illness-related, and psychosocial factors
- Predictors of stigma were highly culturally-specific
- Negative outcomes of stigma included poorer physical and psychological wellbeing

## **Abstract**

### **Objectives**

The aim of this review was to identify quantitative correlates, predictors, and outcomes of stigma in adults with epilepsy living in Western countries.

### **Methods**

To identify relevant literature, four academic databases (PsycINFO, CINAHL, PubMed, and Scopus) were systematically searched using key terms related to stigma and epilepsy.

### **Results**

Thirty-three research papers reporting findings from 25 quantitative studies of correlates of stigma in epilepsy were identified. The findings suggest that stigma can be predicted by demographic, illness-related, and psychosocial factors; although associations were found to be highly culturally-specific. Outcomes of stigma in people with epilepsy were replicated more consistently across cultures and its impact was significant. Detrimental effects included both worse physical health, including less effective management of the condition, and reduced psychological wellbeing, including difficulties such as depression and anxiety.

### **Implications**

Educational initiatives and therapeutic interventions that aim to address stigma in people with epilepsy are recommended; however, these need to be culturally-informed to ensure that they are valid and effective.

**Keywords:** Epilepsy; stigma; neurological conditions; chronic illness; mental health

## **1. Introduction**

### **1.1 Stigma**

Goffman defined stigma as a phenomenon in which a person is discredited or rejected by society because of a particular attribute, in a way that spoils their normal identity [1]. This may be due to “external deformations” such as physical disabilities and diseases, “deviations in personal traits”, such as being unemployed or addicted to drugs, and “tribal stigmas” based on, for example, ethnic group or nationality [1]. Jones et al. [2] developed Goffman’s description by defining stigma as a “mark” (attribute) that links a person to undesirable characteristics (stereotype). Crocker, Major, and Steele [3] similarly went on to describe stigma as the possession (or believed possession) of an attribute or characteristic that conveys a social identity that is devalued in a particular social context. More recently, Link and Phelan summarized that “stigma exists when elements of labelling, stereotyping, separating, status loss, and discrimination co-occur in a power situation that allows these processes to unfold” [4].

### **1.2 Stigma in epilepsy**

Informed by the work of Goffmann [1], Scambler and Hopkins [5] described how stigma can manifest in people with epilepsy (PWE) in their “Hidden Distress Model of Epilepsy”, which differentiated between “felt” stigma (e.g., PWE feeling embarrassed or ashamed about the condition) and “enacted” stigma [6] (e.g., experiencing discrimination or social exclusion from others). The model highlights the relative importance of felt stigma in comparison to enacted stigma, and can be broadly operationalized into three areas: the sense of felt stigma that people experience when being confronted by a diagnosis and as a result feeling the need to conceal their illness; the impact of this concealment in relation to others being unaware of their epilepsy; and the disruption that this felt stigma can cause, which can be even greater than when stigma is enacted externally [7].

### **1.3 Present context of epilepsy-related stigma**

This review will identify predictors and outcomes associated with stigma for adults in Western countries. Public myths and misconceptions of epilepsy endure [8] often reinforced by the use of derogatory language and negative or erroneous media representations [9], and PWE continue to face social and legal barriers even in Western countries. For example, in the UK, it was illegal for PWE to marry until as late as 1970 [10]. To protect the rights of PWE in England, Scotland, and Wales, epilepsy has been included in the Equality Act [11], and in Northern Ireland in the Disability Discrimination Act [12]. However, PWE continue to be discriminated against in the UK, for example in regard to employment and driving [13]. Thus, although they have diminished over time, stigmatizing negative attitudes towards epilepsy, underpinned by misconceptions of the condition and often enacted as discrimination, continue to impact on those living with the condition.

### **1.4 Justification for a review**

Whilst medical treatments for epilepsy have advanced, stigma around the condition has persisted over time [14]. Despite an increased awareness of the causes and effects of epilepsy, misconceptions that underpin stigma of the condition have not been eradicated, [15]. Previous reviews have described the frequency and nature of stigma towards epilepsy, examined misconceptions within the general population, and discussed issues related to stigma and quality of life [13,15,16,17,18,19]. The majority of published studies have investigated stigma in “Western” or “developed” populations (North America, South America, Europe, and Australia). It is hoped that the findings of this review will help to inform the future direction of interventions aimed at reducing the prevalence and impact of stigma in PWE in Western countries and encourage further investigation of stigma in other, non-Western, populations.

## **2. Methods**

## **2.1 Research aims**

The aim of this review was to identify quantitative correlations, predictors, and outcomes of stigma in adults with epilepsy.

## **2.2 Inclusion and exclusion criteria**

The following search parameters were chosen to provide a homogenous sample that would allow a clear picture to be obtained in relation to the current state of stigma in adults with epilepsy in a culturally specific context.

### **2.2.1 Inclusion criteria**

- Studies that have quantitatively measured correlates of stigma in adults with epilepsy using (a) validated measure(s) of stigma
- Studies focusing on adult populations (ages  $\geq$  16 years)
- Studies published in Western countries (North America, South America, Europe, and Australia)
- Studies published after 2000
- Studies available in English

### **2.2.2 Exclusion criteria**

- Studies using qualitative methods
- Studies examining misconceptions of epilepsy or perceptions of epilepsy stigma in the general population
- Studies including participants who have had seizures but do not have a diagnosis of epilepsy

## **2.3 Description of systematic search process**

Following consultation with an academic librarian, four databases were searched: PsycINFO, CINAHL, PubMed, and Scopus. Two key search terms were used: “epilepsy” and “stigma”.

Use of the truncation symbol in the context of “stigma\*”, to include suffixes such as

“stigmatizing” and “stigmatized”, was discounted as it was felt that this would likely result in a more cumbersome search which would not yield additional relevant papers. Keyword searches including the terms “stigma”, “social stigma”, “labelling”, “stereotyped attitudes”, “stereotyping”, combined with the term “epilepsy”, were completed in databases where this functionality was available (e.g. Thesaurus in PsychINFO, CINAHL Headings, and Medical Subject Headings [MeSH] in PubMed). This was then combined with a free text search of the “abstract” or “title and abstract” fields to identify additional articles missed by index searches. The articles identified across databases were entered into the referencing software, Endnote, and duplications were removed. Articles were then filtered and excluded by title, abstract, or full-text according to their relevance to the research question, methodology, date and location of publication, and sample population. Reference lists of included papers were also searched for additional relevant articles. An overview of the search strategy is provided in Figure 1.

*[Figure 1 here]*

Once all relevant articles had been identified, the findings were compared and contrasted using a narrative synthesis to allow for a meaningful integration and discussion of the available evidence. Due to the heterogeneity of research identified in the review, a meta-analysis was not undertaken.

#### **2.4 Appraisal of methodological quality**

To assess the methodological quality of studies included in the review, a quality appraisal tool for observational studies adapted from the Agency for Healthcare Research and Quality was used [20]. This comprised an eight-point checklist of key methodological considerations which researchers should take into account and report in studies of this type, including issues relating to sample selection, measures, data handling, and analysis. Studies were rated on each item and assigned an overall score to indicate an appraisal of the methodological

quality. To ensure the reliability and validity of appraisal ratings, a sub-sample of six papers was chosen at random and peer inter-rated; discrepancies were minor and final ratings were agreed by consensus.

### **3. Results**

#### **3.1 Synthesis of reviewed studies**

An overview of the studies identified for inclusion in the review is provided in Table 1.

Correlation coefficients (Pearson's  $r$ ) are also presented in Table 1, where available, as a measure of effect size [21].

*[Table 1 here]*

#### **3.2 Study characteristics**

Following the search procedure described above, 33 research papers were identified, reporting findings from 25 quantitative studies, with 16,942 adults with epilepsy. An additional 238 adults without a diagnosis of epilepsy were recruited as controls. Participant ages ranged from 16-98 years. Research was identified from countries in North and South America, Europe, and Australia. There were 12 papers from the US, five from Bulgaria, four from Turkey, three from the UK, three from countries across Europe, two from Australia, one from the Netherlands, one from Croatia, one from Mexico, and one from Canada. Of the studies identified, 30 were cross-sectional in design and three incorporated longitudinal methods. Two studies compared findings to controls without epilepsy. Eight papers used only correlational analyses and 25 included regression analyses.

#### **3.3 Measures**

The papers identified in the review used 10 different standardized measures of stigma.

Fifteen papers used the "Jacoby 3-Item Measure of Stigma" [22], which was the most widely used measure in the review. Twelve papers used the "Modified Parent Stigma Scale", also referred to as the "Epilepsy Stigma Scale (ESS)" [23]. Of the remaining studies, individual



papers used the “Felt Stigma Scale” [24], the “Perception of Stigma of Epilepsy Scale (PSE)” [25], the “Revised Stigma Scale” [26], the “Stigma Scale” [27], the “Stigma Scale for People with Intellectual Impairment” [28], and stigma items derived from the “Child Asthma Scale” [29].

### **3.4 Scope of the research**

The identified studies examined correlations, predictors, and outcomes of stigma in adults with epilepsy. Statistical data regarding epilepsy epidemiology or stigma prevalence was not addressed in this review. Whilst the majority of research was cross-sectional in design, and therefore directionality of effect or causation could not be determined, researchers typically framed their findings in relation to what they viewed as predictors or outcomes of stigma within the target population.

### **3.5 Summary of quality appraisal**

Overall, the methodological quality of studies in the review was satisfactory, with a mean score of 5.5 out of 8, although this ranged from 2.5 to 7 indicating variability of quality across studies (see Table 1). Most studies provided clear descriptions of participant samples, including details of inclusion/exclusion criteria and how participants were recruited. Details of statistical analyses were generally provided and appropriate for the type of study.

Consideration of confounding data was also widely taken into account, with the majority of studies using regression analyses to adjust for demographic or clinical factors likely to be correlated with outcomes. However, power calculations as a means of determining and justifying sample size were reported in only two studies. Validity of standardized measures was frequently referred to in relation to findings of previous studies; however reliability coefficients (e.g. Cronbach’s alpha) were rarely given, therefore validity and reliability could not be fully assumed [30]. Details of missing data were also rarely reported; again this limits confidence that data was obtained and presented in a way which minimizes bias.

## **3.6 Summary of main findings**

### **3.6.1 Demographic, illness, and psychosocial correlates and predictors of stigma**

Twenty studies examined correlations or predictors of stigma in PWE. Findings could be broadly categorized according to demographic, illness-related, and psychosocial variables found to be correlated with, or (for regressions) to predict, stigma.

#### **3.6.1.1 Demographic variables**

##### **3.6.1.1.1 Socioeconomic factors**

Several socioeconomic factors were identified as important. Yeni, Tulek, and Bebek identified a negative correlation between income and stigma [31]. Higher income was also found to predict lower stigma when other variables had been taken into account [32,33]. In a further regression study, Smith et al. found that people who were disabled or unemployed with greater seizure worry were more likely to report higher levels of stigma when adjusting for other variables (e.g. self-efficacy, social support, and race) [34]. Yeni, Tulek, and Bebek also identified a negative correlation between education and stigma [31]. In correlational studies comparing patients from clinics in “low and high sociodemographic communities”, participants from low socioeconomic status backgrounds were found to report higher felt stigma [35,36]; although when psychosocial variables including quality of life (QOL), depressive symptoms, and social support were entered into a regression model, these differences were found not to be significant [36]. These findings indicate that socioeconomic status may not in itself significantly affect stigma but that other related psychosocial variables may be of greater importance.

##### **3.6.1.1.2 Cultural factors**

The impact of cultural factors was also identified. In a large-scale continental study examining the relationship between stigma and health system performance across 10 European countries, including a sample of over 5,000 PWE, Baker et al. found country of

origin to significantly contribute to variance in reported levels of stigma in regression analyses [37]. For example, Spanish participants reported significantly lower levels of stigma than participants in France. The authors suggested that cultural differences may be due to a range of factors, including sociocultural bias against epilepsy, cultural norms, the structure of the health system, and the existence of high profile public figures with the condition who may act as role models, although they suggested that more research is needed. Brigo et al., reporting on the same data, identified a trend towards negative correlation between stigma and overall health system performance and health expenditure per capita; however, this association was non-significant [38].

#### **3.6.1.1.3 Personal factors**

Personal factors were also identified as potentially contributing to variance in stigma. When taking into account other clinical and demographic variables using regression analyses, Baker et al. identified that being married significantly predicted lower levels of stigma, alongside six other important illness-related and psychosocial variables [37]. Bautista, Shapovalov, and Shoraka replicated this finding [32]. Younger age was also found to be correlated with higher stigma in some studies [32,39,40]; this was found to independently predict lower levels of reported stigma when other variables had been taken into account in regression analyses [26]. In contrast, however, several other studies using regression analyses did not find age to significantly predict stigma [31,33,36,41,42]. Gender was also found to be uncorrelated with stigma [33,36,41,43]. It has been suggested that a lack of relationship between gender and stigma may be due in part to overarching negative social attitudes, which can cause other factors to “recede into the background” [43].

#### **3.6.1.2 Illness-related variables**

##### **3.6.1.2.1 Seizure type and severity**

Ni Eidhin and McLeavey found seizure type and severity to correlate significantly with stigma [44], although significant flaws were identified in their methodology. Baker also found seizure type (generalised seizures) to contribute to variance in stigma outcomes in regression analyses [45]; however, he stressed that the relevant contributions of these findings depended on the country of origin of those surveyed, highlighting the importance of cultural differences in determining the impact of illness-related variables on stigma. In contrast, in regression analyses Baker et al. found epilepsy-related injuries to significantly contribute to scores of stigma but not seizure type [37]. Viteva found no correlation between stigma and seizure severity [43].

#### **3.6.1.2.2 Seizure frequency**

Dilorio et al. found the number of seizures experienced during the past year to significantly predict stigma in regression analyses [33], and this was replicated in Croatian and UK studies using regression models which found number of seizures to date to significantly predict stigma [26,39]. Yeni et al. also identified positive correlations between seizure frequency and stigma [46]. Furthermore, Baker's large-scale study in European countries found greater seizure frequency to be the most consistent cross-cultural predictor of higher levels of reported stigma in regression analyses [45]. However, these findings were partially in contrast to those of a large-scale study by Baker et al., which found that whilst seizure frequency significantly correlated with measures of stigma, this variable did not predict significant variance in stigma when entered into a regression model alongside other variables including age at onset, marital status, worry about epilepsy, injury, feelings about life, general health, and duration of epilepsy [37]. Aydemir, Kaya, Yıldız, Öztura, and Baklan also found that number of seizures did not significantly predict stigma in regression analyses [41], and Viteva found no correlation at all between stigma and seizure frequency [43].

#### **3.6.1.2.3 Epilepsy onset**

The age of epilepsy onset (i.e. longer duration of epilepsy) was found to significantly correlate with stigma [46] and to contribute to higher scores of stigma in several regression studies [33,37,45]. However, cultural variations were again identified [37]. In another regression study, Smith et al. found that those with later seizure onset were more likely to report lower levels of stigma but only when they were experiencing a higher quality of care [34]. In contrast, Aydemir, Kaya, Yıldız, Öztura, and Baklan did not find duration of epilepsy to significantly predict stigma in regression analyses [41].

#### **3.6.1.2.4 Epilepsy treatment**

Aydemir, Kaya, Yıldız, Öztura, and Baklan found that taking a greater number of epilepsy medications was correlated with increased stigma [41]. Yeni et al. also identified positive associations between the use of epilepsy medication and stigma [46]. However, in contrast, Viteva found no correlation between stigma and prescribed treatment [43]. Observed associations may be due in part to iatrogenic effects of treatments. When taking into account other illness-related variables in regression analyses, adverse events and side effects relating to the use of anti-epileptic drugs were found to significantly predict stigma [26,47].

Aydemir, Özkara, Canbeyli, and Tekcan also examined the effects of epilepsy surgery by comparing participants who had already received surgery to those who were awaiting surgery using t-tests [48]. The authors found no significant differences in the pre- and post-surgery groups, which they argued might have been due to the long-term effect of being labelled as “epileptic”, even if epilepsy has gone into remission. It is also possible that for some people stigma related to refractory epilepsy (e.g. seizures) was replaced by stigma related to surgery (e.g. visible scarring), although this was not included in analyses.

#### **3.6.1.3 Psychosocial variables**

##### **3.6.1.3.1 Psychological factors**

Psychological and emotional factors which were found to predict higher levels of reported stigma in regression analyses included feelings about life and perceived impact of epilepsy [37], lower self-efficacy [33,34], lower patient satisfaction [33], feeling more socially restricted, and poor overall global QOL [26]. Social anxiety was also found to predict stigma in regression analyses, over and above depression and other types of anxiety [42]. Cognitive factors which were found to predict stigma variance in regression models included concerns related to social life and future occupation [41], negative outcome expectancies for seizures [33], and perception of the role of genetics in determining the condition [49]. Although previous research describes important differences between felt and enacted stigma [7], authors of the studies identified did not typically differentiate between the two; although in one study enacted stigma was found to predict felt stigma, with those experiencing discrimination, insults, threats or attacks reporting higher levels of the felt stigma [40]. Behavioral factors were also found to be important. After controlling for demographic and clinical variables including age, gender, duration of epilepsy, number of seizures, and number of medications using regression analyses, Aydemir, Kaya, Yıldız, Öztura, and Baklan found concealment of epilepsy to significantly predict felt stigma [41]. Similarly, the use of behavioral disengagement, a coping strategy whereby a person intentionally decreases the amount of effort needed to deal with a stressful situation, was also found in regression analyses to be independently associated with higher reported stigma [32].

#### **3.6.1.3.2 Relational factors**

Social support was found to be important. In a correlational study, participants with greater social support reported significantly lower stigma [31]. Furthermore, social support was found to significantly predict lower stigma even when other sociodemographic variables had been taken into account in regression analyses [36]. To ascertain whether participants' social cognitive skills and their ability to understand the thoughts, intentions, beliefs, and emotions

of others contributed to feelings of stigma, Noble, Robinson, and Marson compared “theory of mind” and stigma measures using regression analyses [50]; these were found to share little variance, regardless of participant seizure status, indicating that the model has little utility in understanding epilepsy stigma.

### **3.6.1.3.3 Knowledge and access to information**

Access to understandable information was also found to be important. Correlational studies identified negative associations between knowledge and attitudes towards epilepsy (increased knowledge and more positive attitudes) and stigma [31,46]. After taking into account demographic and clinical variables using regression analyses, Baker also found knowledge of epilepsy to negatively predict stigma [45]. Similarly, difficulties in understanding written information, which may limit access to epilepsy knowledge, were found to predict higher levels of stigma in regression analyses [32].

### **3.6.2 Stigma as a predictor and correlate of wellbeing**

Seventeen studies examined correlations between stigma and condition management, physical health, or psychological wellbeing, with 11 studies then going on to use more complex models (e.g.. regression or mediation) where stigma was a predictor of physical and psychological wellbeing.

#### **3.6.2.1 Physical wellbeing and condition management**

Chesaniuk, Choi, Wicks, and Stadler found that higher perceived stigma was correlated with lower medication adherence; mediation analyses revealed this association to be explained largely by information, motivation, and behavioral skills [51]. Similarly, using path analysis, Dilorio, Shafer, Letz, Henry, and Schomer found stigma to be indirectly related to medication self-management through its association with self-efficacy [52]. The association between stigma and lower self-efficacy was supported by a correlational study by Yeni et al., who found participants reporting higher levels of stigma to be more likely to hide their condition

from others and more likely to seek help from non-medical sources such as “mystics” [31]. In a regression study, Dilorio, Shafer, Letz, Henry, and Schomer found stigma to predict seizure severity [53], which they argued may be related to poor self-management or help-seeking behaviors; although it is possible that people who experience more seizures may be more likely to experience greater discrimination. Stigma was also found to be negatively correlated with social support [54] and epilepsy outcomes, including being identified as a significant predictor of “concerns about the social impact of epilepsy” alongside seizure severity in regression analyses [27]. These findings may help to explain those identifying positive correlations between seizure severity and social support and stigma discussed above [34,44], and brings into question the causal direction of these relationships. In contrast to other studies, Elliott, Jacobson, and Seals did not find stigma to predict self-efficacy or epilepsy self-management in regression analyses [55]. The authors of this study identified age and ethnicity as the only predictors of these variables, highlighting the potential importance of demographic and cultural factors in determining health outcomes alongside stigma.

### **3.6.2.2 Psychological wellbeing and QOL**

There was also evidence that stigma can affect psychological wellbeing and QOL. In several studies, stigma was positively correlated with depression and anxiety [31,35,36,43,52,53,54]. These findings were supported by a longitudinal study completed by Reisinger and Dilorio, in which stigma was found to be the third most important predictor of depression following employment status and social support, after controlling for demographic and seizure-related variables using regression analyses [56]. Similarly, in another regression study, stigma was found to predict depression and anxiety when gender, age, and epilepsy-related variables had been controlled for [57]. Viteva also found that stigma correlated with affective and obsessive compulsive disorders (defined by the authors as “mental status impairment”) [43].



In addition to depression and anxiety, Viteva found stigma to negatively correlate with QOL [27]. Regression studies also found stigma to predict poor health-related quality of life (HRQOL), reduced psychosocial function, and lower “emotional wellbeing” when other variables had been accounted for [58,59]. Similarly, in regression analyses Suurmeijer, Reuvekamp, and Aldenkamp found perception of stigma to be the fourth strongest predictor of low QOL after psychological distress, loneliness, and adjustment and coping; this association was significant regardless of participants’ physical status [60]. Eidhin and McLeavey also found stigma to be significantly correlated with lower perceived acceptance of the condition, with participants with higher stigma feeling less cared for and less valued by others [44].

## **4. Discussion**

### **4.1 Key findings**

The findings of the review suggest that stigma is a complicated construct to understand in the context of PWE and is associated with a range of important factors. A number of demographic variables were found to be associated with stigma, although these findings were not replicated across all studies. Being married, higher income, and higher age were found to be associated with lower levels of stigma. Being in a stable relationship may help to protect or mitigate against social rejection and the identification of an individual as “discredited” or having a “spoiled identity” [1], through the social support offered by partners/spouses [61]. In general, those with access to greater financial resources and social support may be better able to cope with adversity [62,63]. Financial resources may be particularly relevant to PWE if it helps them to overcome limitations, for example paying for taxis may help to mitigate against the impact of being unable to drive and lead to feeling more included. Older age has also been associated with increased resilience, which may be due to the development of

coping skills and emotional regulation abilities [64]; this may again help to protect against the negative impact of externally-enacted stigma associated with the condition.

The review also highlighted differences in relation to illness-related variables. Findings associated with seizure type and severity were mixed. Some studies found these factors to be associated with increased stigma whilst elsewhere the finding was not replicated. Regression analyses revealed that other illness-related variables such as age of epilepsy onset (lower age associated with higher stigma), number of seizures to date (greater number associated with higher stigma), and injuries associated with epilepsy, may be more important. Seizure frequency, whilst found to be associated with stigma, may also be less important in predicting stigma than the duration and impact of the condition, perhaps due to repeated exposure to negative health-related events, including experiences of discrimination by others. The cumulative number of seizures experienced may also increase the number of negative reactions from others (enacted stigma) and an increased perception of self as “externally deformed” (felt stigma), [1,6]. This longer-term exposure to seizures and negative reactions from others may also lead to an over-identification with the condition, exacerbated by negative language or labelling.

The findings of the review also suggested that the impact of illness-related variables on stigma can vary by country of origin, and therefore appeared to be, to a significant degree, culturally-specific. Stigma in epilepsy is highly culturally-dependent [37] and this has been highlighted in previous research; for example, a recent cross-continental comparative study of PWE found Swedish participants to report significantly lower levels of stigma than PWE in Iran; the researchers argued that this was likely due to differences in medical treatment and educational exposure [65]. These cultural differences informed the rationale to focus the review on countries of Western origin, however there was still considerable heterogeneity identified across studies of different geographical origin.

One possible explanation relates to the impact of overall health system performance and health expenditure; the hypothesis being that higher expenditure will result in lower stigma as a result of greater understanding of the condition and better support systems. However, Brigo et al. found that, whilst there was a trend towards negative associations between expenditure and stigma, findings related to these variables were non-significant [38]. This suggests that general investments in public health systems do not necessarily lead to improvements in stigma-related epilepsy. To achieve this, the authors argue, funds need to be directed specifically towards epilepsy awareness and stigma-reduction programs. Whilst public myths and misconceptions remain even in countries of higher socioeconomic status where educational campaigns have been launched [8,66], the negative impact of stigma on social identity in PWE can be greater in resource-poor countries [67]. Concealment of the condition in these countries is also likely to be higher [68], and issues of language and legality may increase the risks of stigma further [69]. It is therefore important that stigma reduction efforts are viewed as important and are culturally-informed [70].

Further variance in stigma can be explained by psychosocial factors. Knowledge of epilepsy, and the ability to access this, was universally found to be associated with lower stigma. Knowledge of epilepsy is also an important factor in optimizing control of seizures [71]; this may impact further on stigma and help to explain some of the geographical differences in stigma identified in different countries. Unsurprisingly, therefore, feelings of control and mastery over the condition were found to be negatively associated with perceptions of stigma. Where PWE reported lower feelings of self-efficacy or a deterministic view of the condition, or where they identified concerns about their ability to effectively manage their illness, to access support, or to cope in the future, stigma was higher. Such beliefs may also lead to maladaptive and avoidant coping strategies, such as concealment of the condition or behavioral disengagement with its management, which were found to increase stigma

[32,41]. This could furthermore serve to reinforce a lack of social support, condition management, and perceived ability to cope, completing a vicious cycle that provides a fertile ground for perceived stigma in PWE. In this case, stigma may be seen as self-perpetuating, and again fits in with Scambler's "Hidden Distress Model of Epilepsy", in which a person feels stigmatized, conceals their condition from others, and feels increasingly distressed [6]. Therefore, in addition to wider societal educational campaigns, therapeutic interventions at an individual level are also likely to be important.

The findings associated with outcomes of stigma were more straightforward and perhaps less surprising. Higher levels of stigma were associated with a reduced sense of self-efficacy, lower motivation, and compromised condition management, characterized by lower medication adherence and poor epilepsy outcomes, including increased seizure severity. As previously identified, however, it was not possible to determine causal directions and it is likely that these relationships are strengthened in both directions. Stigma was found universally to predict depression and anxiety, even when other variables had been taken into account, as well as being associated with lower "emotional wellbeing, lower perceived acceptance, and a greater incidence of obsessive compulsive disorders and low QOL.

#### **4.2 Implications and recommendations**

The findings of the review suggest that, in addition to demographic and illness-related variables, psychosocial factors are likely to be particularly important in determining stigma. These are likely underpinned by knowledge about the condition, social support, and a perception that the care system, and in turn society, takes an understanding view of epilepsy and its management. Public campaigns to address educational deficits have been advocated [72,73]. In the UK, this has been reflected by clinical guidelines that explicitly outline the responsibility of healthcare professionals to educate others about epilepsy as a means of reducing stigma [74], and awareness campaigns launched by charities [75,76]. Where such

campaigns have been introduced, there has been some evidence of effectiveness [77]. However, there is contradictory evidence that attitudes over the last 10 years may actually have worsened [78], perhaps due in part to online social networking platforms where derogatory communications about epilepsy and seizures are common, and in part to a negative economy and changes in the global political landscape, where people may have become less tolerant of diversity and immigration, fueling negative attitudes towards the condition [79].

Societal values which can lead people to feel stigmatized and to conceal health conditions such as epilepsy can also extend to the law [80], therefore further research is needed to ensure that legal structures serve to protect PWE. PWE who feel stigmatized by others are more likely to feel depressed and anxious [e.g. 56]; they may also feel less accepted and valued by others [44]. Everyone in society, including politicians, teachers, healthcare professionals, employers, community leaders, voluntary organizations, PWE and their friends and families should therefore help to give PWE a voice, and to promote the view that epilepsy is a manageable, socially acceptable, condition. There is some evidence that psychological approaches such as acceptance and commitment therapy (ACT [81]) and compassion focused therapy (CFT; [82]) can help to increase psychological flexibility and reduce internalized health-related stigma [83]. Narrative therapy may also be beneficial in shedding light on alternative perspectives and helping PWE to develop new narratives about themselves [84]. Hence psychologists working in health settings arguably have a key role to play in tackling stigma at a wider societal level as part of their widening influence in public health initiatives [85].

#### **4.3 Limitations and recommendations for further research**

One of the most significant limitations of this review was that it relied heavily on cross-sectional surveys gathering data via self-report measures, thus it was not possible to

determine causation [86] and relationships in this context may therefore be bidirectional with one factor reinforcing another. Cross-sectional designs have also been criticized for assuming that variables remain stable over time and for therefore failing to address chronological variability, leading to biased estimates and incorrect inferences [87]. Further research should aim to incorporate longitudinal methods to help determine causation and chronological variation. Another limitation is that findings derived from self-report measures are open to bias [88] and sensitive to culture [89], therefore the use of such measures in different countries requires careful consideration.

A large number of studies used a three-item measure of stigma originally used in a study of stroke patients [90], adapted for use in PWE by Jacoby [22]. Although this measure has been validated for use in this population [22,91], the measure is basic and may not detect subtle but important nuances such those associated with “felt” versus “enacted” stigma. This may be a significant omission as, for example, subtle differences in others’ language may be perceived as stigmatizing by a person with epilepsy even where this is not intentionally or objectively enacted [92]. Further research should aim to use more detailed measurement tools and consider this distinction, particularly as enacted stigma may point towards a need to direct change at public health level, whereas felt stigma may require support and interventions at an individual level.

A final limitation related to differentiating Western versus non-Western populations. Whilst the decision was pragmatic and informed by an aim to address a defined research question, it is important to acknowledge that the “othering” - and potential stigmatizing - of different social, cultural, and geographic groups may be perceived as in direct contrast to the spirit of this review. This is an entirely unintended consequence of the limited scope of the work, which reviews of other populations could address. The study of stigma in populations globally should be encouraged.

#### **4.4 Conclusions**

The findings of this review suggest that stigma in PWE may be predicted by demographic, illness-related, and psychosocial factors, with the latter explaining a large degree of variance. However, findings varied significantly by country of origin. This suggests that stigma is, to a significant degree, culturally determined and thus may present challenges to campaigners and legislators attempting to reduce stigma and its impact internationally. What appears to be important, however, is fostering education and understanding of the condition, both in PWE and in the general population. The outcomes of stigma appear significant and more universal; its impact relates to both physical health, including management of the condition, and psychological wellbeing, including difficulties such as depression and anxiety. It is therefore important that healthcare providers, legislators, policy-makers, and citizens take steps to try and address stigma to help improve outcomes for this often marginalized population.

#### **Conflict of interest**

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no financial support for this work that could have influenced its outcome.

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Table 1. Characteristics of the studies included in the review

Study	Design	Participants	Measures	Analysis	Effect size (Pearson's r)	Findings/Authors' Conclusions	Quality Appraisal Rating
Aydemir, Özkara, Canbeyli, and Tekcan (2004)	Cross-sectional survey	$n = 20$ patients awaiting epilepsy surgery and $n = 21$ who had already undergone surgery in Turkey ( $N = 41$ ; mean age = 25.9 years)	Jacoby 3-item measure of stigma, the Perceived Impact of Epilepsy Scale, the Medical Outcomes Study Short Form-36 (SF-36), Beck Depression Inventory (BDI), State-Trait Anxiety Inventory (STAI)	$T$ -test; Mann-Whitney $U$	Not reported	No significant difference was found relative to stigma levels between pre- and post-SAH groups ( $p=.82$ ). A high level of stigma was observed in only 6 (14.7%) of the patients, suggesting that stigmatization may be low among Turkish patients.	4.5
Aydemir, Kaya, Yıldız, Öztura, and Baklan (2016)	Cross-sectional survey	$N = 200$ adults with epilepsy in Turkey (age = 18-68 years, mean age = 31.68 years)	The Felt Stigma Scale, the Concealment of Epilepsy Scale, the Epilepsy Concern Scale, the Overprotection Scale	Correlation (r); hierarchical multiple regression	Stigma and overprotection ( $r=.34$ ). Stigma and concealment ( $r=.64$ ). Stigma and future occupation concerns ( $r=.62$ ). Stigma and social life concerns ( $r=.62$ ). Stigma and marriage/children concerns ( $r=.43$ ). Stigma and number of medications ( $r=.21$ ).	Concealment of epilepsy ( $\beta = .43$ , $p < .001$ ), concerns related to social life ( $\beta = .27$ , $p < .001$ ), and concerns related to future occupation ( $\beta = .26$ , $p < .001$ ) were found as the predictors of felt stigma after controlling for demographics (age and gender), and clinical variables (duration of epilepsy, number of seizures, and number of medications).	6

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Baker, Brooks, Buck, and Jacoby (2000)	Cross-sectional survey	<i>N</i> = 5211 adult epilepsy patients living in 15 European countries (69% from France, UK, Germany, and the Netherlands) (age = 16+ years, mean age = 35 years)	Jacoby 3-item measure of stigma, Perceived Impact of Epilepsy Scale, Extent of Worry over Epilepsy, the Medical Outcomes Study Short Form 36 (SF-36), Terrible-Delighted Faces Scale	Correlation (r); multiple regression analysis	Not reported	Impact of epilepsy ( $\beta = .43$ , $p < .0001$ ), age of onset ( $\beta = .09$ , $p < .0001$ ), country of origin, feelings about life ( $\beta = .05$ , $p < .001$ ), and injuries associated with epilepsy ( $\beta = .05$ , $p < .01$ ) were significant contributors to stigma. Whereas seizure type and frequency were significantly correlated with scores on the stigma scale, results of the multiple regression showed that neither seizure frequency nor seizure type accounted for a significant amount of the variance on scores on the stigma scale.	5
Brigo, Igwe, Ausserer, Tezzon, Nardone, and Otte (2015)  <i>*Used the same sample as Baker et al.(2000)</i>	Cross-sectional survey	<i>N</i> = 5211 adult epilepsy patients from 10 European countries including France, UK, Germany, and the Netherlands (age = 16-98 years, mean age = 37 years)	Percentages of people with epilepsy with epilepsy-related stigma obtained from Baker et al.'s (2000) study (which used the Jacoby 3-item measure of stigma), data on overall health system performance in 1997, data on health expenditure per capita in international dollars in 1997*	Correlation (r)	Stigma percentage and health system performance ( $r = -.16$ ). Stigma and health expenditure per capita ( $r = -.24$ ). Stigma and quality of life ( $r = -.33$ ).	Authors found a non-significant trend towards negative correlation between the epilepsy-related stigma percentage and the overall health system performance ( $r = -0.16$ ; $p = 0.57$ ), the health expenditure per capita in international dollars ( $r = -0.24$ ; $p = 0.4$ ), and the Economist Intelligence Unit's quality-of-life index ( $r = -0.33$ ; $p = 0.91$ ).	5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Baker (2002)	Cross-sectional survey	<i>N</i> = 6156 adult epilepsy patients from 10 European countries including France, UK, Germany, and the Netherlands (age = 16-98 years, mean age = 37 years)	Jacoby 3-item measure of stigma, the Epilepsy Knowledge Questionnaire, the Impact of Epilepsy Questionnaire, and the Acceptance of Illness Scale	ANOVA; stepwise multiple regression analysis	Not reported	After taking into account demographic and clinical variables, a number of factors were predictive of stigma, including seizure frequency, knowledge of epilepsy, duration of epilepsy, and seizure type. The relative contributions of these factors varied depending on the country of origin of those surveyed.	5
Bautista, Shapovalov, and Shoraka (2015)	Cross-sectional survey	<i>N</i> = 182 adults with epilepsy at epilepsy centers in the US (mean age = 43 years)	The Epilepsy Stigma Scale (ESS), the Quality of Life in Epilepsy-10 (QOLIE-10), the Beliefs about Medicine Questionnaire (BMQ), the Short Test of Functional Health Literacy in Adults (STOHFLA), the Brief-COPE	Correlation (r); ANOVA; multiple linear regression analysis	Stigma and age ( $r = -.164$ ). Stigma and QOL ( $r = .36$ ). Stigma and use of denial ( $r = .15$ ). Stigma and behavioural disengagement ( $r = .33$ ). Stigma and venting ( $r = .2$ ).	Using multiple linear regression, marital status (being single) ( $\beta = -4.027, p = .01$ ), being poorer, indicated by higher QOLIE-10 scores ( $\beta = .45, p < .01$ ), difficulties understanding written information ( $\beta = -2.19, p = .03$ ), and the use of behavioral disengagement ( $\beta = 2, p = .01$ ) were independently associated with poorer scores on the Epilepsy Stigma Scale.	6

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Begley, Shegog, Iyagba, Chen, Talluri, Dubinsky,... and Friedman (2010)	Cross-sectional survey	<i>n</i> = 167 patients from a "high socioeconomic status" epilepsy clinic and <i>n</i> = 71 from a "low socioeconomic status" clinic in the US ( <i>N</i> = 238 ; age = 18+ years, mean age = 40.9 years)	Modified Parent Stigma Scale, Epilepsy Self-Management Scale, Epilepsy Knowledge Scale, Epilepsy Self-Efficacy Scale, Treatment Outcome scale, Shared control portion of the Multidimensional Desire for Control Scale, Personal Resource Questionnaire 85, Part 2 (PRQ85-2), Center for Epidemiologic Studies Depression Scale (CES-D), Patient Satisfaction Questionnaire III	<i>T</i> -test; correlation ( <i>r</i> ); multivariate regression analysis	Stigma and self-management ( <i>r</i> =.077).	Stigma, along with self-efficacy, depression, social support, desire for control, and outcome expectations, was higher for those of high socio-economic status ( <i>P</i> < 0.01).	5
Leaffer, Hesdorffer, and Begley (2014)  <i>*Used the same sample as Begley et al. (2010)</i>	Cross-sectional survey	<i>n</i> = 167 patients from a "high socioeconomic status" epilepsy clinic and <i>n</i> = 71 from a "low socioeconomic status" clinic in the US ( <i>N</i> = 238 ; age = 18+ years, mean age = 40.9 years)	Modified Parent Stigma Scale, Epilepsy Self-Management Scale, Epilepsy Knowledge Scale, Epilepsy Self-Efficacy Scale, Treatment Outcome scale, Shared control portion of the Multidimensional Desire for Control Scale, Personal Resource Questionnaire 85, Part 2 (PRQ85-2), Center for Epidemiologic Studies Depression Scale (CES-D), Patient Satisfaction Questionnaire III	<i>T</i> -test; correlation ( <i>r</i> ); linear regression analysis	Stigma and QOL ( <i>r</i> =.41) Stigma and social support ( <i>r</i> =- .39). Stigma and self-efficacy ( <i>r</i> =-.21).	Reported levels of stigma were higher in low SES than in high SES ( <i>p</i> <0.0001), and all psychosocial variables were associated with stigma, including depression severity ( <i>p</i> <0.0001), knowledge of epilepsy ( <i>p</i> =0.006), quality of life ( <i>p</i> <0.0001), social support ( <i>p</i> <0.0001), and self-efficacy ( <i>p</i> =0.0009). Stigma was statistically significantly associated with quality of life in the low SES group and with depression severity and social support in the high SES group.	5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Bielen, Friedrich, Sruk, Prvan, Hajnšek, Petelin,... and Jacoby (2014)	Cross-sectional survey	<i>N</i> = 298 epilepsy outpatients in Croatia (age = 17-82 years, mean age = 45 years)	Revised version of the Jacoby 3-item measure of stigma, translated into Croatian.	ANOVA; Multiple stepwise regression (B)	Not reported	Feelings of stigma were significantly associated with age, younger age of epilepsy onset, more than 50 seizures to date, generalized tonic-clonic seizures, and a shorter seizure-free period. Multiple stepwise regression showed number of seizures to date as a significant variable ( $B=0.246$ ).	7
Chesaniuk, Choi, Wicks, and Stadler (2014)	Cross-sectional survey	<i>N</i> = 140 PWE in the US (age= 20-65 years, mean age = 38.51 years)	The Epilepsy Stigma Scale, Knobel Brief Adherence Questionnaire, adapted scale of adherence information, motivation and behavioral skills	Correlation (r); mediation analysis	Stigma and medication adherence ( $r=-.18$ ). Stigma and levels of information ( $r=-.28$ ). Stigma and motivation ( $r=-.55$ ). Stigma and behavioral skills ( $r=-.41$ ).	Higher stigma was associated with lower medication adherence ( $r = -0.18, p=.05$ ), lower levels of information ( $r = -0.28, p b .05$ ), motivation ( $r = -0.55, p b .05$ ), and behavioral skills ( $r = -0.41, p b .05$ ). Adherence information, motivation, and behavioral skills explained nearly all of the association between stigma and adherence.	7
Chong, Drake, Atkinson, Ouellette, and Labiner (2012)	Cross-sectional survey	<i>N</i> = 50 Hispanic epilepsy clinic patients of Mexican descent in the US (age = 18+ years, mean age = 38.6 years)	Edited version of the Parent Stigma Scale, the Epilepsy Self-Efficacy Scale, the Interpersonal Support Evaluation List (ISEL), the Family Emotional Involvement and Criticism Scale (FEICS), the Patient Health Questionnaire 9 (PHQ-9), the Acculturation Rating Scale for Mexican Americans II (ARSMA-II)	Correlation (r); Principal components analysis (PCA)	Stigma and depression ( $r=.39$ ). Stigma and social support ( $r=-.65$ ).	Stigma was positively correlated with depression ( $r=0.39, p<0.01$ ) and negatively associated with social support ( $r=-0.65, <0.001$ ). Stigma was not significantly correlated with perceived criticism, emotional involvement, self-efficacy, or national orientation.	5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Dilorio, Osborne Shafer, Letz, Henry, Schomer, and Yeager (2003)	Cross-sectional survey	<i>N</i> = 314 adult men and women with epilepsy recruited from "Project EASE" in the US (age = 19 to 75 years, mean age = 43 years)	The Parent Stigma Scale modified for use to measure stigma in adults, the Epilepsy Self-Efficacy Scale, the Epilepsy Self-Management Scale, the Self-Reported Medication-Taking Scale, the Patient Satisfaction Questionnaire—III, Multidimensional Desire for Control scale	ANOVA; Correlation (r); hierarchical regression analysis	Stigma and self-efficacy to manage epilepsy ( $r=-.431$ ). Stigma and outcome expectancies related to treatment ( $r=-.213$ ) and seizures ( $r=.652$ ). Stigma and medication management ( $r=-.200$ ). Stigma and medication adherence ( $r=.202$ ). Stigma and patient satisfaction ( $r=-.190$ to $-.350$ ). Stigma and expectancies related to information management ( $r=.159$ ).	Higher stigma was associated with lower self-efficacy to manage epilepsy ( $r=-0.431$ ); more negative outcome expectancies related to treatment ( $r=-0.213$ ) and seizures ( $r=0.652$ ); and lower levels of medication management ( $r=-0.200$ ), medication adherence ( $r=0.202$ ), and patient satisfaction ( $r=-0.190$ to $-0.350$ ). However, stigma was associated with more positive outcome expectancies related to information management ( $r=0.159$ ). In regression analysis, income, age at first seizure, seizures during the past year, lower self-efficacy, negative outcome expectancies for seizures, and less patient satisfaction explained 54% of the variance in perceived stigma.	7

Study	Design	Participants	Measures	Analysis	Effect size (Pearson's r)	Findings/Authors' Conclusions	Quality Appraisal Rating
Dilorio, Shafer, Letz, Henry, and Schomer (2004)  <i>*Used the same sample as Dilorio et al. (2003)</i>	Cross-sectional survey	N = 317 PWE recruited from "Project EASE" in the US (age = 19-75 years, mean age = 43.3 years)	Modified version of The Parent Stigma Scale (expanded to 10 items), Epilepsy Self-Efficacy Scale, Epilepsy Self-Efficacy Scale, Epilepsy Regimen-Specific Support Scale, Personal Resource Questionnaire 85 Part 2 (PRQ85-2), Center for Epidemiologic Studies Depression Scale (CES-D), Patient Satisfaction Questionnaire-III, Multidimensional Desire for Control Scale	Path analysis	Not reported	Stigma was directly related to self-efficacy and depressive symptoms. Stigma was indirectly related to medication self-management through its association with self-efficacy. These results suggest that those who feel highly stigmatized because of their epilepsy are less efficacious in taking their medications.	7
Dilorio, Shafer, Letz, Henry, and Schomer, (2006)  <i>*Used the same sample as Dilorio, Osborne Shafer, Letz, Henry, Schomer, &amp; Yeager (2003)</i>	Longitudinal survey	N = 272 PWE recruited from "Project EASE" in the US (age = 19-74 years, mean age = 43.7 years)	The Epilepsy Stigma Scale, The Epilepsy Self-Efficacy Scale, The Epilepsy Self-Management Scale, The Personal Resource Questionnaire 85 Part 2 (PRQ85-2), The Epilepsy Regimen Specific Support Scale, The Patient Satisfaction Questionnaire-III (PSQ), The Center for Epidemiologic Studies Depression Scale (CES-D)	Hierarchical regression	Not reported	Stigma was a potentially significant predictor of self-efficacy (F=3.643, p<0.057)but was less important than self-management, depressive symptoms and seizure severity. The inverse relationship found between perceived stigma and self-efficacy suggests that those who harbor negative thoughts about epilepsy also feel less confident in their ability to manage epilepsy.	7



Study	Design	Participants	Measures	Analysis	Effect size (Pearson's r)	Findings/Authors' Conclusions	Quality Appraisal Rating
Reisinger and Dilorio (2009)  <i>*Used the same sample as Dilorio et al. (2003)</i>	Longitudinal survey	N = 319 PWE recruited from "Project EASE" in the US (age = 19-75 years, mean age = 43.3 years)	Epilepsy Stigma Scale, Center for Epidemiologic Studies Depression Scale (CES-D), Epilepsy Self-Management Scale (ESMS), Epilepsy Self-Efficacy Scale (ESES), Self-Reported Medication-Taking Scale, Personal Resource Questionnaire 85 Part 2 (PRQ85-2), Patient Satisfaction Questionnaire	ANOVA; Correlation (r); stepwise multiple regression (B)	Stigma and depression at baseline, 3 and 6-month follow-up (r=.425, .343 and .371, respectively).	Stigma was correlated with depression at baseline, 3- and 6-month follow-up (r=.425, .343 and .371, respectively, p<.001). The third main predictor of depressive symptoms in the study was epilepsy-related stigma.	7
Whatley, Dilorio, and Yeager (2010)  <i>*Used a subset of the same sample as Dilorio et al. (2003)</i>	Longitudinal study	N = 147 adults with epilepsy recruited from "Project EASE" in the US (age = 19-75 years, mean age = 45 years)	10-item scale adapted from the Parent Stigma Scale, 31-item Quality of Life in Epilepsy (QOLIE-31) scale, adapted from the more comprehensive 89-item scale, Center for Epidemiologic Studies Depression Scale (CES-D), Personal Resource Questionnaire (PRQ85-part 2)	Correlation (r); multiple linear regression	Stigma and QOL (r=-.513).	Statistically significant negative correlations were found between depressive symptoms, stigma and sometimes regimen-specific support and QOL. In regression analyses, stigma was found to predict QOL at a later time.	7
Elliott, Jacobson, and Seals (2006)	Cross-sectional survey	N = 94 epilepsy patients in the US (age = 19-78 years, mean age = 45 years)	The Liverpool Stigma Scale (LSS), the Osteoporosis Knowledge Test (OKT), the Osteoporosis Health Belief Scale (OHBS), the Osteoporosis Self-Efficacy Scale (OSES), the Quality of Life in Epilepsy (QOLIE-31) scale, and the Epilepsy Self-Efficacy Scale (ESES)	ANOVA; Multivariate regression analysis (B)	Not reported	The Liverpool Stigma Scale did not predict any of the dependent variables (self-efficacy for calcium, exercise, and epilepsy self-management).	6.5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Heersink, Kocovski, MacKenzie, Denomme, and, Macrodimitris (2015)	Cross-sectional survey	<i>N</i> = 101 PWE in Canada (age = 18-65 years, mean age = 37.51 years)	Jacoby 3-item measure of stigma, the Social Phobia Inventory (SPIN), the Hospital Anxiety and Depression Scale (HADS), the Epilepsy Knowledge Questionnaire (EKQ), the Liverpool Seizure Severity Scale (LSSS), the Impact of Epilepsy scale, the Disclosure Management Scale, Brief Fear of Negative Evaluation (BFNE) scale, Acceptance and Action Epilepsy Questionnaire (AAEpQ)	Correlation (r); hierarchical regression analysis; ANCOVA	Stigma and social anxiety (r=.48).	Social anxiety positively correlated with felt stigma (r=.48, p<.001). This relationship remained significant after controlling for depression (p<.001). Social anxiety significantly predicted the variance in stigma above and beyond age, anxiety, impact of epilepsy, seizure frequency, and depression ( $\beta = .33, p < .001$ ).	4
McLaughlin, Pachana, and, McFarland (2008)	Cross-sectional study	Epilepsy group <i>N</i> = 64 older adults with epilepsy in Australia (age = 60+ years, mean age = 67.59 years). Control group <i>N</i> = 60 adults recruited from the general community in Australia (age 60+ years, mean age = 66.50 years)	3-Item Stigma scale, Mini mental state exam (MMSE), Washington Psychosocial Seizure Inventory (WPSI), Quality of life in epilepsy (QOLIE-31), Seizure frequency	MANOVA; multiple regression analysis	Not reported	Stigma contributed significantly to prediction of HRQOL ( $sr^2 = .21$ ). A greater perception of stigma was strongly related to poor quality of life and reduced psychosocial function. Less stigma and lower frequency of seizures uniquely contributed to the overall prediction of better HRQOL. Overall, the predictors of stigma and seizure frequency together accounted for 54% of the variability in HRQOL.	5.5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Ni Eidhin and McLeavey (2001)	Cross-sectional survey	<i>N</i> = 52 people with a diagnosis of epilepsy attending an outpatient clinic in Northern Ireland	Jacoby 3-item measure of stigma; Perceived Severity of Epilepsy Scale; Perceived Acceptance Scale; Questions relating to epilepsy and seizure type and frequency	Correlation (r)	Stigma and seizure severity ( $r=.37$ ). Stigma and perceived acceptance ( $r=-.35$ ).	Seizure severity was significantly correlated with perception of stigma ( $r=.37$ , $p<.01$ ). A significant negative correlation were found between perceived stigma and perceived acceptance ( $r=-.35$ , $p<.05$ ).	2.5
Noble, Robinson, and Marson (2016)	Cross-sectional study	<i>N</i> = 503 PWE in the UK and Republic of Ireland (age = 18-79 years, median age = 37 years)	Jacoby 3-item measure of stigma, the Faux Pas Task-Short Version (FPT), the Reading the Mind in the Eyes Test (RMET)	Correlation (r); multiple regression analysis	Stigma and theory of mind performance ( $r=-.02$ on the RMET and $r=-.05$ on the FPT).	Feelings of stigma held a negligible, negative, and nonsignificant association with ToM performance ( $r=-.02$ and $-.05$ ). The ToM model for understanding epilepsy stigma has limited utility.	7
Peterson, Walker, and Shears (2014)	Cross-sectional survey	<i>N</i> = 300 PWE in Australia completing the 2010 Australian Epilepsy Longitudinal Survey to register participants on the Australian Epilepsy Research Register (AERR) (age = 18+ years)	Stigma scale emerging from factor analysis of items principally derived from the Child Asthma scale (including social scale and personal scale subscales), the Hospital Anxiety and Depression Scale (HADS)	Correlation (r); Multiple regression analysis (B)	Not reported	Significant correlations were found between anxiety and depression and stigma. Social aspects of stigma significantly predicted depression and anxiety ( $B=.34$ and $.32$ , respectively, $p<.01$ ) when gender, age and epilepsy-related variables had been controlled for. Social aspects of stigma had the strongest effect on anxiety, followed by the effectiveness of current control on seizures. Those who take more epilepsy drugs experienced greater stigma as a result and, therefore, had higher rates of depression and anxiety.	4.5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Sabatello, Phelan, Hesdorffer, Shostak, Goldsmith, Sorge,... and Ottman (2015)	Cross-sectional survey	$n = 181$ PWE and $n = 178$ biologic relatives without epilepsy in the US ( $N = 359$ ; mean age = 52 years)	Epilepsy Stigma Scale (ESS), Family Epilepsy Stigma Scale (FESS), three questions related to genetic causal attribution	<i>T</i> -test; Correlation ( <i>r</i> ); multivariate analyses using generalised estimating equations (GEE) models	Not reported	Felt stigma was higher among individuals who were aged $\geq 60$ years, were unemployed, reported epilepsy-related discrimination, or had seizures within the last year or $>100$ seizures in their lifetime. Adjusting for other variables, ESS scores in people with epilepsy were significantly higher among those who perceived genetics played a "medium" or "big" role in causing epilepsy in the family than in others (3.4 vs. 2.7, $p = 0.025$ ).	6
Smith, Ferguson, Saunders, Wagner, Wannamaker, and Selassie (2009)	Cross-sectional survey	$N = 244$ adults with epilepsy in the US (age = 18+ years)	The Stigma Scale (8 questions modified from the scale developed by Dilorio), the Epilepsy Self-Efficacy Scale (ESES),	Kruskal–Wallis test; multiple linear regression	Not reported	Reported levels of stigma were associated with interactions of seizure worry and employment status (disabled or unemployed with higher seizure worry=higher stigma), self-efficacy and social support (higher scores=lower stigma), and quality of care and age at seizure onset (higher quality of care and over 40=lower stigma).	6

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Suurmeijer, Reuvekamp, and Aldenkamp (2001)	Cross-sectional survey	<i>N</i> = 210 PWE attending outpatient clinics in The Netherlands (age = 18-65 years, mean age = 38 years)	Perception of stigma of epilepsy (PSE), Perception of epilepsy seizures (PES), Health perceptions (HP), Life-fulfillment questionnaire (LFQ), Loneliness scale (LS), General adjustment to epilepsy (GATE), Self-esteem (RSE), Mastery (MAS), Mental health (MH), Psychological distress (GHQ), Visual Analogue Scale (VAS-DT)	Correlation (r); Hierarchical multiple regression analysis (B)	Stigma and QOL (r=0.17).	Perception of stigma in epilepsy was negatively correlated with QOL (r=0.17, p<.01). In decreasing order of importance, psychological distress, loneliness, adjustment and coping, and stigma perception (B=.17, p=.4) contributed most significantly to QoL.	7
Taylor, Baker, and Jacoby (2011)	Cross-sectional survey	<i>N</i> = 1566 adults with epilepsy in the UK (mean age = 40 years)	The Revised Stigma Scale, the Newly Diagnosed Epilepsy Quality of Life (NEWQOL) battery	Correlation (r); Kruskal-Wallis test; x <sup>2</sup> test; stepwise multiple regression	Stigma and anxiety (r=.41). Stigma and depression (r=.41). Stigma and mastery (r=-.41). Stigma and cognitive effects of anti-epileptic drugs (r=.43). Stigma and adverse events (r=.45).	Felt stigma was associated with younger age, a previous or current neurological disorder, being unmarried, experiencing more seizures, having no formal educational qualifications on leaving school, and being unemployed. Gender, seizure type, presence of a neurological deficit, and social class were not associated with degree of felt stigma. A multivariate linear regression demonstrated that scores on the AEP, mastery scale, and ABNAS, poor overall global QOL, age < 50 years, more than four seizures at baseline, and feeling more socially restricted were significant predictors of stigma.	6.5

Study	Design	Participants	Measures	Analysis	Effect size (Pearson's r)	Findings/Authors' Conclusions	Quality Appraisal Rating
Viteva (2012)	Cross-sectional survey	<i>n</i> = 94 patients with refractory epilepsy (RE) and <i>n</i> = 70 patients with pharmacosensitive epilepsy (PSE) in Bulgaria ( <i>N</i> = 164; age = 18-65 years, mean age = 41.72 years)	Jacoby 3-item measure of stigma, the Beck Depression Inventory (BDI-II), the Hamilton Anxiety Scale (HAS), the Liverpool Seizure Severity Scale (LSSS), and	Correlation (r)	Stigma and depression ( $r=-.40$ ). Stigma and mental status impairment ( $r=.19$ ).	No correlation was found between stigma and age and gender, education, marital status, employment, seizure frequency and severity, prescribed treatment, or anxiety ( $P > 0.05$ ). A moderate correlation was found between depression and stigmatization frequency and severity ( $r=.40$ , $P < .01$ ). A mild correlation was found between mental status impairment and stigmatization. Mental status impairment was associated with a more frequent and more severe stigmatization ( $r=.19$ , $P < .05$ ).	5
Viteva (2013)  <i>*Used the same sample as Viteva (2012)</i>	Cross-sectional survey	<i>N</i> = 140 PWE (70 patients with refractory epilepsy and 70 patients with pharmacosensitive epilepsy) in Bulgaria (age = 18-65 years, mean age = 41.7 years)	Jacoby 3-item measure of stigma, the Health Related Quality of Life measure (QOLIE-89)	Correlation (r)	Stigma and QOL ( $r=-.6$ ).	Stigma had a negative impact on QOL ( <i>T</i> -score 47.8), including all sub-scales of QOLIE-89, with the exception of "change in health" and "sexual relations". There was a negative correlation of all QOLIE-89 sub-scales with perceived stigma severity.	5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Viteva (2014)	Cross-sectional survey	<i>N</i> = 64 patients with refractory epilepsy and intellectual impairment in Bulgaria (age = 18-65 years, mean age = 44.88 years)	The stigma scale, the Glasgow Depression Scale for people with a Learning Disability (GDS-LD), <sup>4</sup> the Glasgow Anxiety Scale for people with Intellectual Disability (GAS-ID), The Liverpool Seizure Severity Scale (LSSS), The Glasgow Epilepsy Outcome Scale (GEOS-35), the carer supplement of the GDS-LD (GDS-CD)	Correlation (r); multivariate regression analysis	Stigma and health-related QOL (r=.43).	GEOS-35 total scores were associated with seizure frequency and severity, stigma, depression, and anxiety. On multivariate regression analysis predictors of the GEOS-35 total score were anxiety, seizure severity, and stigma <i>P</i> < 0.001 ( <i>F</i> = 14.66). Regarding GEOS-35 sub-scales, on multivariate regression analysis seizure severity and stigma were predictors of "concerns about social impact" <i>P</i> < 0.001 ( <i>F</i> = 18.31).	5
Viteva and Semerdjieva (2015)  <i>*Used the same sample as Viteva (2014)</i>	Cross-sectional survey	<i>N</i> = 64 patients with refractory epilepsy and intellectual impairment in Bulgaria (age = 18-65 years, mean age = 44.88 years)	The stigma scale for people with intellectual impairment (10-item), Evaluation rapide des fonctions cognitives (ERFC), interview about enacted stigma comprising four statements about a real experience of discrimination	Correlation (r); multiple regression analysis	Stigma and discrimination (r=.71). Stigma and experienced insults and threats and/or attacks (r=.43)	Participants who gave a greater number of positive answers about experienced discrimination or insults and/or threats and attacks reported a more pronounced perceived stigma (F=19.30, P<0.001 and F=12.91, P<0.001, respectively). Perceived stigma and the experience of insults and/or threats and attacks proved to be predictors of discrimination on multivariate regression analysis (F=40.54, P<0.001).	5

<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Viteva (2016)	Cross-sectional survey	<i>N</i> = 153 patients with epilepsy in Bulgaria (age = 18-65 years, mean age = 39.34 years)	Jacoby 3-item measure of stigma, the Liverpool Adverse Events Profile (LAEP)	Correlation (r); multiple regression analysis	Stigma and the presence of neurological and psychiatric adverse events (r=.60).  Stigma and the presence of non-neurological adverse events (r=.20).	A significant correlation between perceived stigma and the presence of neurological and psychiatric AEs ( $p < 0.001$ , $r = +0.60$ ) and a mild correlation between perceived stigma and the presence of non-neurological AEs ( $p < 0.01$ , $r = +0.20$ ) was identified. In a multivariate regression analysis the only predictors of perceived stigma were AED polytherapy and the presence of neurological and psychiatric AEs.	5.5
Yeni, Tulek, Bebek, Dede, Gurses, Baykan, and Gokyigit (2016)	Cross-sectional survey	<i>N</i> = 70 patients with epilepsy in Turkey (age = 18-70 years, mean age = 31.7 years)	Jacoby 3-item measure of stigma, The Epilepsy Attitude Scale, the Epilepsy Knowledge Scale, Rotter's Locus of Control Scale, the Hospital Anxiety and Depression Scale (HADS), the Quality of Life in Epilepsy Inventory-10 (QOLIE-10-P)	Mann-Whitney U test; Kruskal-Wallis test; correlation (r)	Stigma and attitude towards epilepsy (r=-.267). Stigma and anxiety (r=.283). Stigma and depression, r=.282). Stigma and QOL epilepsy effects (r=-.255). Stigma and QOL role functioning (r=-.336).	Significant correlations were obtained between stigma and attitude towards epilepsy (r=-.267, $p=.026$ ), anxiety and depression (r=.283, $p=.018$ , r=.282, $p=.018$ ), QOL epilepsy effects (r=-.255, $p=.033$ ), and QOL role functioning (r=-.336, $p=.004$ ).	4



<i>Study</i>	<i>Design</i>	<i>Participants</i>	<i>Measures</i>	<i>Analysis</i>	<i>Effect size (Pearson's r)</i>	<i>Findings/Authors' Conclusions</i>	<i>Quality Appraisal Rating</i>
Yeni, Tulek, and Bebek (2016)	Cross-sectional survey	<i>N</i> = 194 patients with epilepsy in Turkey (age = 18-80 years, mean age = 31.4 years)	Jacoby 3-item measure of stigma, the Multidimensional Scale of Perceived Social Support (MSPSS), the Social Support Scale, the General Self-Efficacy Scale, the Epilepsy Knowledge Questionnaire, and the Epilepsy Attitude Scale	Mann–Whitney U test; Kruskal–Wallis test; correlation (r)	Stigma and social support ( $r=-.3$ ) Stigma and knowledge of epilepsy ( $r=-.18$ ). Stigma and attitudes towards epilepsy ( $r=-.152$ ). Stigma and self-efficacy ( $r=-.185$ ).	Education ( $\chi^2=8.23$ , $p=.016$ ), income ( $\chi^2=9.735$ , $p=.008$ ), age at onset ( $r=-0.183$ , $p=.01$ ), seizure frequency in previous year ( $\chi^2=9.26$ , $p=.01$ ), social support ( $r=-.3$ , $p=.001$ ), and knowledge and attitudes towards epilepsy ( $r=-.18$ , $p=.012$ , $r=-.152$ , $p=.034$ ) were significant factors determining stigma. It was also determined that stigma was associated with seeking non-medical help ( $Z=3.60$ , $p=.001$ ), disclosure of the diagnosis ( $Z=2.59$ , $p=.01$ ), and self-efficacy ( $r=-.185$ , $p=.01$ ).	4

## Figure caption

Figure 1. Flow chart of search and inclusion/exclusion process

