Special Report
Epilepsy-Related Stigma and Attitudes: Systematic Review of Screening Instruments and Interventions Report by the International League Against Epilepsy Task Force on Stigma in Epilepsy

Running title: Epilepsy Stigma Instruments and Interventions

Joan K. Austin[1], Gretchen Birbeck[2], Karen Parko[3], Churl-Su Kwon[4], Paula T. Fernandes[5], Patricia Braga[6], Kirsten M. Fiest[7], Amza Ali[8]*, J. Helen Cross[9]*, Hanneke de Boer[10]*, Tarun Dua[11]*, Sheryl R. Haut[12], Ann Jacoby[13]*, Diane L. Lorenzetti[14], Janet Mifsud[15]*, Solomon L. Moshé[16]*, Manjari Tripathi[17]*, Samuel Wiebe[18]*, Nathalie Jette[19]

* Listed alphabetically

1. Indiana University School of Nursing, Indianapolis, IN, USA
2. Epilepsy Division, University of Rochester, Rochester, NY, USA and Epilepsy Care Team, Chikankata Hospital, Mazabuka, Zambia
3. Department of Neurology, University of California at San Francisco, San Francisco, CA, USA and Epilepsy Center, San Francisco VA Medical Center, San Francisco, CA, USA
4. Departments of Neurosurgery and Neurology, Icahn School of Medicine at Mount Sinai, New York, NY, USA
5. Department of Sport Science, Faculty of Physical Education, UNICAMP, Campinas, SP, Brazil
6. Institute of Neurology, Facultad de Medicina, Universidad de la República, Uruguay
7. Department of Critical Care Medicine and Department of Community Health Sciences, Hotchkiss Brain Institute, O'Brien Institute for Public Health, University of Calgary, Calgary, Canada
8. Kingston Public Hospital and University of the West Indies, Jamaica
9. Developmental Neurosciences Programme, UCL-Great Ormond Street Institute of Child Health, London
10. Deceased
11. Department of Mental Health and Substance Abuse, World Health Organization, Geneva, Switzerland
12. Saul R. Korey Department of Neurology, Albert Einstein College of Medicine and Montefiore Medical Center, Bronx, NY, USA
13. Department of Public Health and Policy, University of Liverpool, UK
14. Department of Community Health Sciences, University of Calgary and Heath Sciences Library, University of Calgary, Canada
15. Department of Clinical Pharmacology and Therapeutics, Faculty of Medicine and Surgery, University of Malta, Msida, Malta
16. Department of Pediatrics and Dominick P. Purpura Department of Neuroscience, Albert Einstein College of Medicine, Bronx, NY, USA
17. Department of Neurology, All India Institute of Medical Sciences, New Delhi, India
18. Department of Clinical Neurosciences and Department of Community Health Sciences, Hotchkiss Brain Institute, O'Brien Institute for Public Health, University of Calgary, Calgary, Canada
19. Departments of Neurology and Population Health Sciences & Policy, Icahn School of Medicine at Mount Sinai, New York, NY, USA

Corresponding Author:
Dr. Nathalie Jette
Department of Neurology
Icahn School of Medicine at Mount Sinai
New York, NY 10029
(T) 212-241-5050
(E) nathalie.jette@mssm.edu

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Key Point Box (max 140 characters per point including space)

- Most screening instruments were developed using established methods; the tools with the strongest psychometric properties are identified.
- Investigators should further develop, refine, and translate existing tools, except in children and the elderly where new tools are needed.
- Most intervention studies lacked design rigor, were underpowered, and were of short duration.
- No intervention studies used participatory praxis to engage people with epilepsy in study design, conduct, analysis, and dissemination.
- Many ILAE regions lack instruments and interventions that are validated in the language and culture.
- To accelerate development of epilepsy stigma reduction interventions, unified theories that transcend individual conditions are needed.

Summary

Objective: This is a systematic review aimed at summarizing the evidence related to instruments that have been developed to measure stigma or attitudes towards epilepsy and on stigma-reducing interventions.

Methods: This review followed the PRISMA standards. A broad literature search (1985-2019) was performed in 13 databases. Articles were included if they described the development and testing of psychometric properties of an epilepsy-related stigma or attitude scale or stigma-reducing interventions. Two reviewers independently screened abstracts, reviewed full text articles, and extracted data. Basic descriptive statistics are reported.

Results: We identified 4,234 abstracts, of which 893 were reviewed as full text articles. Of these, 38 met inclusion criteria for an instrument development study and 30 as a stigma reduction intervention study. Most instruments were initially developed using well established methods and were tested in relatively large samples. Most intervention studies involved educational programs for adults with pre- and post-evaluations of attitudes towards people with epilepsy. Intervention studies often failed to use standardized instruments to quantify stigmatizing attitudes, were generally under-powered, and often found no evidence of benefit or the benefit was not sustained. Six intervention studies with stigma as the primary outcome had fewer design flaws and showed benefit. Very few or no instruments were validated for regional languages or culture and there were very few interventions tested in some regions.

Significance: Investigators in regions without instruments should consider translating and further developing existing instruments rather than initiating the development of new instruments. Very few stigma reduction intervention studies have been conducted, study methodology in general was poor, and standardized instruments were rarely used to measure outcomes. To accelerate the development of effective epilepsy stigma reduction interventions, a paradigm shift from disease-specific, siloed trials to collaborative, cross-disciplinary platforms based upon unified theories of stigma transcending individual conditions will be needed.

Key words: KAP, instruments, scales, questionnaires, psychometric properties

Introduction

Stigma, described by Goffman as "an attribute that is deeply discrediting" and the perception that someone is different than expected in society, is a major burden for many people with epilepsy.[1] Effects of stigma can negatively impact the quality of life of people with epilepsy by reducing their opportunities related to education, employment, and social relationships.[2] An important step in better understanding epilepsy-related stigma is to accurately measure perceptions of stigma in people with epilepsy (self-stigma) as well as in people with whom individuals with epilepsy interact including healthcare professionals, educators, and the general population (enacted stigma). A key approach to measuring epilepsy-related stigma has been the use of instruments or questionnaires in which people self-report their beliefs and attitudes related to epilepsy. If we are to improve the quality of life of people with epilepsy by reducing stigma, it is important that interventions be developed to reduce perceptions of epilepsy-related stigma and be tested to determine if they are effective.

The purpose of this paper is twofold: (a) to review instruments that were developed to measure stigma or attitudes associated with epilepsy and (b) to review epilepsy stigma-reducing interventions that were developed and tested. These reviews were carried out by a task force of the International League Against Epilepsy (ILAE).

Methods

This systematic review was done following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (unless otherwise specified). The protocol was not registered in PROSPERO.[3]

Search Strategy

The search strategy, including the 13 databases (Medline, PubMED, Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, EMBASE, PsychINFO, Health and PsychoSocial Instruments Database, CINAHL, Social Services Abstracts, Sociological Abstracts, SocINDEX, LILACS, Web of Science) used in this study, is described in detail in Appendix 1.[1] Studies had to include an abstract in French or English but once selected, were included regardless of region or language of publication, within the time

frame 1985 to November 5, 2019. *No restrictions were placed on the region or language of publication except that articles without an abstract that were not in English or French were excluded.* Thus, non-English articles were included if they had an English abstract and otherwise met our eligibility criteria. The reference lists of included articles were also hand searched in duplicate to identify additional studies of relevance to this review.

Study Selection

For this manuscript, only papers that reported on the use of scale/questionnaire tools to measure stigma or attitudes towards epilepsy as outcomes as well as those that developed, conducted, and or implemented an intervention aimed at decreasing negative attitudes towards epilepsy or epilepsy-related stigma were included. There were no restrictions on study design (e.g., controlled and uncontrolled studies) or age of participants, and the presence of a comparison group was not required. For the tool studies, psychometric properties of each scale were the outcomes of interest. Tools had to address felt stigma, enacted stigma, attitudes towards having epilepsy, or attitudes toward persons with epilepsy. In addition, only articles that provided information on at least one psychometric property of an instrument were eligible since we were interested in reporting the published psychometric properties of existing tools. When instruments were developed to measure stigma in a more inclusive group (e.g., neurological disorders), a final inclusion criterion was that the testing of the instrument needed to include people with epilepsy and provide information on psychometric properties for the epilepsy sample. For the intervention studies, we excluded studies that only addressed knowledge about epilepsy without attitude or stigma assessments, or were attitudes about treatment rather than about people with epilepsy was evaluated. Stigma reduction activities reported without any associated outcome assessments were also excluded.

All studies were independently reviewed in duplicate to ensure they met all eligibility criteria. Disagreements were resolved by consensus or through the involvement of a third author as necessary.

Study Quality

Appraisal of study quality for studies addressing tool development was done using a quality and validity questionnaire that evaluated the following criteria: content validity, internal consistency, criterion validity, construct validity, reproducibility, responsiveness, floor and ceiling effects, and interpretability.[4] An overall

quality score is not calculated as this would assume that every item is weighted equally.[4] Also, not all criteria are relevant for every instrument, for example, internal consistency reliability is not relevant for a one-item scale. Study quality for interventions was evaluated using the National Institute of Health National Heart, Lung, and Blood Institute Quality Assessment of Controlled Intervention Studies tool.[5] This is a 14-item tool that captures information about a wide range of variables that could influence study bias (e.g., randomization, blinding, drop out rate, validity of outcome measures). A rating of good, fair, or poor is given by the reviewer based on an overall rating of the internal validity of each study (i.e., the ability to make causal conclusions about the effects of the intervention) and the risk for potential bias in the study. The higher the risk of bias, the lower the rating. Although this tool is for controlled studies, we selected it for uncontrolled studies as by definitions those studies are of lower quality and the tool was still felt to be applicable to these studies.

Data Extraction

All study authors were involved in abstract review, full text review and data extraction, following training sessions. Data were extracted by two reviewers with detailed structured data abstraction forms, one for tools and one for interventions (Appendix 2) including socio-demographic variables, study characteristics, type (i.e., persons with epilepsy, persons without epilepsy) and characteristics of the target population, intervention type, tool characteristics (i.e., tool name, number of items, cut-off score), the psychometric properties of the tool, and the type of validation conducted (i.e., content validity, construct validity), where applicable. Free text descriptions within the abstraction tool allowed for capture of a description of the tool validated/developed, intervention (where applicable), main outcome, and other relevant findings.

Results

Of the 4,234 abstracts reviewed, 893 were selected for full text review (Figure 1, PRISMA flow diagram). Overall agreement between the two reviewers at this stage was excellent at 86.15%. Of these, 38 met all eligibility criteria for tool development and 30 met criteria as a stigma or negative attitude reduction intervention study. Results for measurement instruments and intervention studies are presented separately below.

1. Overview of Studies on Measurement Tools

Of these 38 articles, 37 described measurement scales that were specific to epilepsy. One instrument described a tool that was developed for neurological conditions.[6] The 38 articles provided information on a total of 32 instruments (Tables 1-5). One instrument described a tool that was developed for neurological conditions, the other 31 measurement scales were specific to epilepsy. Only six instruments were the focus of more than one study in which information was provided about development or testing of psychometric properties. These instruments are identified with asterisks in the Tables. During the period of this review, there were 5 instruments developed from 1990 to 1999, 18 from 2000 to 2009, and 9 from 2010 to 2019. All studies reported on the number of participants included, and the sample sizes ranged from 34 to 19,441 with a median sample size of 253. Authors reported non-population-based sampling in 28, population-based sampling in four, and incomplete information on sampling in six studies. Response rates were reported in 15 papers, ranging between 17% and 98%, with a median of 85% and a mean of 73%.

Of the 32 instruments, 15 measure perceptions of epilepsy-related stigma in people who do not have epilepsy (e.g., general public, family members, and professionals), 14 measure perceptions of people who have epilepsy, and three measure perceptions in people with and without epilepsy [7-10] For ease of presentation, the reviewed studies are divided into three groups based on the target audience: people without epilepsy, people with epilepsy, and people with or without epilepsy. Within each table, instruments are in order of year of the article that reported on instrument development, and information on each instrument includes number of citations, age of participants, number of items that specifically measure attitudes/stigma, psychometric properties, method of data collection, region of sample and language of questionnaire. Citations are included to provide a rough estimate of instrument update and use in future studies.

2. Instruments for People without Epilepsy

A total of 19 articles reported on 15 instruments developed to measure perceptions of people without epilepsy (Tables 1 & 2). Most studies were conducted in either North America (n=9) or Africa (n=4). Of the 15

instruments, 9 were multidimensional in nature and measured attitudes toward people with epilepsy and at least one other concept including epilepsy awareness, knowledge, familiarity, practices, or beliefs and 6 of the instruments focused solely on measuring attitudes toward people with epilepsy. Of the 15 instruments, 12 were developed for adults (Table 1) and three were developed for children and adolescents to complete (Table 2).

a. Instruments for Adults without Epilepsy

As a group, the instruments for adults without epilepsy were relatively strong methodologically in the initial creation of the scale, with authors presenting information on both reliability and validity on ten of the twelve instruments (Table 1). Half (n=6) of the instruments were developed for and tested on samples from the general population; half (n=6) were tested on other samples including health care personnel, teachers, service providers, adult students, parents of children with epilepsy, or adult relatives of people with epilepsy. Of the 12 adult instruments, 8 were multidimensional, with 3 of the 8 focusing on knowledge, attitudes, and behavior or practices. [11-15] One group of authors developed scales specifically targeting teachers, clerics, and health care workers in Africa. [11-13] health care personnel, teachers, service providers, adult students, parents of children with epilepsy, and adult relatives of people with epilepsy. Two other groups tested the instruments with similar dimensions on adults in the general population in North America[14] and in Malaysia[15].

Although no one instrument appears to be the gold standard in the area, most have strong internal consistency reliability and validity (i.e., content, construct) and 8 have over 30 citations, which suggests that instruments are being used in other studies. However, very few provided information on reproducibility, and none provided information on responsiveness, floor and ceiling effects, or interpretation. One of the better developed instruments is the *Attitudes and Beliefs about Living with Epilepsy (ABLE)*.[16, 17] This 29-item instrument was developed by the Centers for Disease Control and Prevention (CDC) in the United States with the support of an international team of epilepsy experts. This instrument was found to have excellent psychometric properties in a large population-based sample.[16-18] It is one of only two instruments for adults without epilepsy that provided information on test-retest reliability. Moreover, four different subscales emerged: Negative Stereotypes; Risk and Safety Concerns; Work and Role Expectations; and Personal Fear and

Social Avoidance. Items from this scale are being used by the CDC in ongoing national surveys to monitor changes in attitudes toward epilepsy in the United States.[18]

Another instrument that had extensive testing of reliability and validity at initial development is the *Attitudes Toward Persons with Epilepsy* (Table 1).[19] Of the 28 items included in this instrument, 21 measure attitudes toward persons with epilepsy. The instrument was tested on a large, diverse sample of adults including graduate and undergraduate college students, professionals at health and human service agencies, teachers and school administrators, and participants at professional conferences and workshops. It was found to have three factors: Destructive Personal Stereotypes, Integration in Society, and Behavioral Optimism.

b. Instruments for Children without Epilepsy

All three instruments developed for administration to children without epilepsy were multidimensional (Table 2). The instruments were developed and tested in North America. One questionnaire, *Teen Survey on Epilepsy,* was tested in a large Epilepsy Foundation survey of 19,441 adolescents attending high school across a large region of the United States.[20] However, a major limitation of the instrument is that the only psychometric property described was content validity. The other two instruments were developed in Canada, and both incorporated knowledge and attitude subscales.[21, 22] These instruments had better initial development, and the articles provided information that both reliability and validity were strong. One instrument, *Thinking about Epilepsy,* was developed for children in the 5th grade[21] and adminstered to a large sample of 783 school children. The other instrument, *Elementary School Epilepsy Survey,* was developed for children in grades 4, 5, and 6[22] and tested on a sample of 155 children. Both used content evaluation by experts and factor analysis to support validity, and both found good internal consistency reliability. For the latter scale, test-retest reliability for the entire scale was found to be good.[22] None of the scales provided information on responsiveness, flooring or ceiling effects, or interpretability.

3. Instruments for People with Epilepsy

Sixteen studies described the development of 14 instruments to measure stigma perceptions in people with epilepsy. Studies were conducted in North America (n=6), Europe (n=6), Asia (n=2), and Africa (n=2). Only one study was conducted on a population-based sample.[6] All of the scales measured felt stigma with the

exception of one, which measured attitudes toward having epilepsy.[23] Three instruments measured both felt and enacted stigma,[6, 24, 25] and one measured both felt stigma and attitudes toward self.[26] Only two scales measuring stigma in people with epilepsy were tested in more than one study. The 3-item stigma scale developed by Jacoby and colleagues [27, 28] and the child attitude toward illness scale[23, 29] developed by Austin were tested in two studies each during the time frame of this literature review.

a. Instruments for Adults with Epilepsy

Among the 10 instruments that were developed for adults with epilepsy, three studies stated that older adolescents (defined as 17 year olds) would be included;[30-32] however, only two studies included participants who were under 18 years in their sample.[31, 32] The number of items on the scales ranged from 3 to 34. Information was provided on reliability for six instruments, on validity for six instruments, on both validity and reliability for six instruments (Table 3). On the other hand, only two instruments were tested for reproducibity (i.e., test-retest reliability)[26, 32] and none were tested for responsiveness and interpretation (Sopplemental Table 1).

One of the scales with the most extensive testing and almost 600 citations is the *Stigma Scale*, which was developed by Jacoby and colleagues in 1994 in Western Europe.[27] The *Stigma Scale* is a 3-item instrument that measures perceptions of people with epilepsy. Content for this scale was based on items adapted from an instrument that measured stigma related to having had a stroke.[33] This scale was the only one that was specifically tested for flooring and ceiling effects. Because the original version of the *Stigma Scale* was found to have both flooring and ceiling effects, the response scale was expanded from a 3-point to a 4-point Likert-type scale that ranged from 0 to 3. When the revised version was tested on a sample of 1566 people in a larger study, both the flooring and ceiling effects were reduced.[28] In addition, the revised scale was found to have excellent psychometric properties.

Another scale that was developed to measure stigma in adults with epilepsy is the *Epilepsy Stigma Scale (ESS)* developed by DiIorio and colleagues.[16, 17] Items from the *Parent Stigma Scale* by Austin were modified to make them relevant for self-report of felt stigma by adults with epilepsy.[34] Respondents rate each

of the 10 items on a 7-point scale from *Strongly Agree* to *Strongly Disagree* with higher scores reflecting greater stigma. Internal consistency reliability for this scale is excellent (α=.91).

A final well-developed scale is the *Stigma Scale for Chronic Illness (SSCI),* which was developed as part of a larger project, and funded by the National Institutes of Health in the United States of America.[6] The goal of the *SSCI* is to measure stigma in a range of chronic neurological disorders, including epilepsy. Selection of content for the development of items involved review of the literature, focus groups (one was composed of people with epilepsy), interviews with patients, and review of content by experts. The SSCI was tested in a large sample of 511 adults with chronic illness including 183 people with epilepsy.[6] Although testing of psychometric properties was extensive and results indicated excellent support for reliability and validity, results were not provided for responsiveness, floor and ceiling effects, and interpretation (Supplemental Table 1).

b. Instruments for Children with Epilepsy

The four instruments for children (Table 4) were all relatively brief with the number of items ranging from 8 to 15. Two of the four scales, which were both developed in Africa, initially included adults in the development of the instruments.[25, 35] For example, most of the sample for the testing of the *Kilifi Stigma Scale* were children and adolescents (58%), and among the adults tested, almost half had no formal education.[35] The oldest and best developed instrument for children is the *Child Attitude Toward Illness Scale (CATIS)*, which was developed to measure attitudes in children with either epilepsy or asthma.[23] Each item has a blank space in which the specific condition (i.e., epilepsy) can be inserted. Although the CATIS was originally developed for children of ages 8 to 12 years, it has also been found to be reliable and valid in adolescents.[29] It was tested in relatively large samples of children (n=269) and adolescents (n=197) and found to have excellent psychometric properties. The CATIS has been used with other conditions [35] and has over 200 citations. The other scales that are well developed are the *Child Stigma Scale* [34] and the *Kilifi Stigma Scale for Epilepsy.* [35] Both measure felt stigma, were tested on relatively large simples, provide information on both reliability and validity and have relatively large numbers of citations. The *CATIS* and *Kilifi Stigma Scale for Epilepsy* scales were the only ones that were tested for reproducibility and none of the four scales were tested for responsiveness, floor and ceiling effects, and interpretation (Supplemental Table 1).

4. Instruments for Adults With and Without Epilepsy

Three scales were targeted at people with or without epilepsy and all were for adults (see Table 5). The oldest is the *Epilepsy Beliefs and Attitudes Scale,* which was developed in a study conducted in North America in 2000.[10] The scale includes a vignette describing a child with epilepsy that provides a common reference for the response to the 51 items on 4-point scales measuring the strength of the respondents' beliefs about the statements from "Not at all Believe" to "Totally Believe." Factor analysis indicated three domains each having satisfactory internal consistency reliability: metaphysical (14 items), enviro-psycho-physical (12 items), and neurological (9 items). The scale was adapted for administration in Brazil; concepts were found to be relevant for that culture and items appropriately reflected the concepts. The authors concluded that the instrument was suitable to use with the general population. [36]

In three different studies, Fernandes and colleagues described the development of the *Stigma Scale of Epilepsy*.[7-9] The studies were conducted in Latin America's tropical region, specifically Brazil. The development of the scale included a review of literature to identify the main domains of stigma, open-ended questions on medical, social, and personal aspects of stigma, and a review by experts for content validity. The scale was found to have satisfactory internal consistency and reliability for both people with and without epilepsy. The reliability was slightly higher for people with epilepsy than for people without epilepsy. The final scale, *Prejudice toward Chronic Diseases,* was also developed by Fernandes and colleagues. [8] Support for validity was found when as hypothesized, AIDS was found to have the highest level of stigma, epilepsy was a close second to AIDS, and diabetes had the lowest level of perceived stigma. None of the scales provided information on reproducibility, responsiveness, floor and ceiling effects, or interpretation (Supplemental Table 1).

5. Overview of Intervention Studies

We identified 52 published reports from interventions studies for epilepsy-associated stigma reduction. Of these, 22 of the English articles were excluded. Reasons for exclusion included no intervention was implemented (n=13), no stigma measurement post-intervention (n=4), only descriptive assessments of beliefs were measured (n=2), no outcome results in the paper (n=1), stigma was treated as a covariate rather than as an outcome in the intervention study (n=1), and duplicate publication of the same intervention in a different language under a different first author (n=1).

Table 6 provides demographic details on the 30 stigma and negative attitude reduction intervention studies included in this review. The largest number of studies were conducted in the Americas (n=12) and Europe (10); fewer were conducted in the Western Pacific (n=3), Southeast Asia (n=2), Sub-Saharan Africa (n=2), and North Africa (1). Most (16/30) involved interventions with adults that were aimed at improving attitudes toward people with epilepsy. Only one intervention was clearly population based. [37] Most studies used a pre-and post-intervention study design.

a. Interventions

All intervention studies were disease-specific and focused exclusively on epilepsy. Most interventions were educational in nature (n=24) with the remaining six focused on establishing medical care, [38] having epilepsy surgery, [39, 40] being exposed to a door-to-door epilepsy prevalence study that included giving a questionnaire designed to elicit the degree of stigma associated with epilepsy, [37] attending peer support groups, [41] and a group self-management educational course. [42] The majority of the educational interventions were targeted at teachers (n=6) or students (n=12). Three targeted the general public [37, 43, 44] and one each was directed to children with epilepsy and their parents, [45] parents of children with epilepsy, [46] and to medical providers. [47]. Further details on the educational interventions, of which three were controlled, are provided in Table 7.

Most interventions were directed toward people without epilepsy, the majority of which (n=18) were in the educational system as students or teachers. Eight studies targeted people with epilepsy, [37-42, 45, 46] four of which focused on adults, [38, 39, 42, 46] one on both adults and youths, [41] and four on families.[37, 40, 45, 46]

b. Outcomes

Studies evaluating stigmatizing attitudes rarely used validated measures. Instead, most attitude studies developed a compilation of questions adapted from knowledge, attitude, and practice (KAP) items commonly employed across a broad range of previous studies (of epilepsy as well as other common chronic conditions). Of the 24 attitude-only outcome papers, only three used validated instruments. [48-50] Two studies [48, 50] used the *Stigma Scale of Epilepsy* [51] and one study [49] used the *Epilepsy Stigma Scale*. [16]

Six studies had stigma as the primary outcome. [38-42, 52] Five of these studies used validated instruments. One [38] used the *Stigma Scale of Epilepsy,* [51] one [40] used Austin and collagues's *Parent and Child Stigma Scales*, [34] and three [39, 41, 42] used the 3-item stigma scale developed and validated by Jacoby and colleagues specifically for epilepsy.[53]

Among the 24 educational intervention studies, three provided primarily qualitative descriptive details of the outcome,[42, 45, 53] one used the Attitudes and Beliefs about Living with Epilepsy (ABLE) scale[15] and two used the Stigma Scale of Epilepsy as an outcome measure,[47, 49] with one of these having follow-up limitations in post-intervention assessments, making the impact of the intervention difficult to determine.[47] The remaining 18 captured attitude changes in a KAP assessment, but ten[43, 46, 51, 54-60] assessed changes in individual ítems rather than a summary score; none of these studies adjusted their analyses for multiple comparisons and thus might have overstated the benefit of the intervention. KAP summary scores were used in 8 studies,[44, 48, 61-66] and all but one of these[62] showed improvements immediately after the intervention. Four studies explicitly looked at whether the effect of the intervention was sustained over time.[49, 59, 60, 63] Three showed no sustained effects,[59, 60, 63] and one utilizing an educational video showed sustained improvements from baseline to 6 months post intervention, but the stigma reduction magnitude was greater at 6 months after the intervention than immediately post intervention, which suggests that some other temporal factors aside from the intervention of interest might have been at play.[49]

People undergoing epilepsy surgery compared to non-surgical patients showed decreases in stigma over time after the surgery.[38, 39] A similar improvement in stigma was found in people establishing epilepsy care.[37] All three of the controlled studies that used KAP as an outcome demonstrated improvements.[44, 48, 73]

6. Instruments and Interventions by ILAE Region

ILAE regions were unevenly represented in relation to the location where instruments and interventions were developed and/or tested. Of the 32 instruments, 15 were developed and tested in America, 8 in Africa, 5 in Asia, and 4 in Europe. Of the 30 interventions, 12 were developed and tested in America, 7 in Europe, 6 in Africa, and 4 in Asia. The specific ILAE region is listed for each of the instruments and interventions in Tables 1-6. It is interesting to note that most (5/7) of the instruments for children were developed and tested in North America. The two most recently published child instruments were developed in Africa; unfortunately, little information was available for one scale.[23] Another observation is that although there were only two instruments developed in Latin America, the largest number of interventions (n=7) were conducted in that area. The two scales developed in Latin America were for adults both with and without epilepsy. [5-7] It is notable that in the latter study, in addition to Caucasians (48.6%) two different cultural groups were specifically included, South Asians (29%) and East Asians (15.2%). There are many ILAE regions where there are very few instruments or interventions that have been published in the past 27 years.

7. Study Quality

The quality assessment of the 32 instruments indicated that 18 had relatively good initial development (i.e., assessment of internal consistency reliability, content validity, or construct validity) and 14 were poorly developed in that they met either none or only one of the quality criteria. Even among the instruments that were initially developed by established methods, none have been tested for all eight criteria; the highest number of criteria met was five. Moreover, four criteria were only met by either none (i.e., responsivity) or one instrument (i.e., criterion validity, floor and ceiling effects, and interpretability). The assessment of study quality suggests that all instruments could benefit from at least some further development. (See Supplemental Table 1).

The overall quality of the intervention studies was generally poor. Of the 30 intervention studies, 21 fell into the poor category, six into the fair category, and three in the good category. Areas of greatest

weakness across the studies were in the use of blinding, concealment of treatment, randomization, and adequate sample size. None of the studies rated as poor used randomization, concealed treatment allocation, used blinding of participants or raters, or had large enough samples to detect a difference between groups, which significantly increased the opportunity for study bias. Areas where at least half of the studies met criteria were in the analysis of subgroups and the use of reliable and valid outcome measures. The studies rated as fair were relatively more likely than those rated as poor to explore dropout, to adhere to the intervention protocol, and to avoid other interventions or keep similar background treatments. It was only the three studies rated as good that one could be reasonably sure that the intervention was adequately tested and the risk for bias was low. All of the ones rated as good met at least 11 of the 14 criteria including randomization that was rated as adequate and use of an intention-to-treat analysis. (See Supplemental Table 2).

Discussion

A large number of instruments have been developed to measure epilepsy-related stigma and atittudes in both people with and without epilepsy from childhood through adulthood. Many of the authors employed well-established methods for item development, such as reviews of the literature, interviews with the populations of interest, and modifications of items from established instruments for other conditions. Many instruments were also evaluated by experts for content validity, analyzed using factor analysis to evaluate construct validity, and tested for internal consistency reliability. Moreover, many instruments were pilot tested in early phases of the development process. Finally, most instruments were tested on relatively large samples that were of sufficient size to adequately test psychometric properties. Although fewer instruments were developed for people with epilepsy than for those without, there are still many scales to choose from for use in future studies. Finally, the large number of citations for many of the instruments suggest that these instruments are being used by others.

Limitations were found in the review of instruments. The quality assessment indicated that all instruments could use at least some further testing and development and that many instruments a lot of further development. Only a few instruments had information on psychometric properties published in more than one

study, were tested for test-retest reliability or reproducibility, tested for floor and ceiling effects, or provided information on interpretation. Another limitation was that most of the authors of the articles failed to provide a rationale for the need to develop a new scale rather than modifying an existing instrument and none provided information on responsiveness. Finally, there are no scales that were specifically developed to measure attitudes toward epilepsy in children with epilepsy under 8 years or in adults over age 65 years. Investigators should adapt, translate, and further develop existing instruments rather than develop new ones, except in children and the elderly where gaps exist.

Despite the large number of articles reviewed, only 30 studies of epilepsy-associated stigma reduction interventions were found that met our eligibility criteria. Intervention studies aimed at addressing negative attitudes were especially prone to critical design flaws. Those evaluating felt and/or enacted stigma were less flawed and more likely to show benefit. Although "stakeholders" were sometimes involved in intervention development, none of the 30 studies employed participatory praxis to fully engage a community of people with epilepsy in study design, implementation, analysis, and dissemination. Moreover, no studies were registered with US ClinicalTrials.gov.

Other limitations focus on the use of KAP surveys as outcomes across multiple intervention studies. First, there was not a standardized KAP instrument that was used, which limited the ability to compare findings across studies. Second, authors reported change across individual items instead of summary scores, and when summary scores were used, information was not provided as to which items specifically measured "attitude" vs. "knowledge" or "practice." Finally, there was an absence of analysis on captured attitudes and practices data. KAP studies were also frequently flawed by biased participation and post-intervention assessments being limited to a subset of self-selected people rather than everyone who underwent the intervention. Power calculation estimates were rarely provided and when available, they indicated significantly underpowered studies.

The ubiquitous nature of epilepsy-associated stigma and ample data illustrating its sinister consequences on mental and physical health, wealth, well-being, and quality of life is well documented. The development, implementation, and evaluation of stigma reduction interventions should be a high priority in

epilepsy research and health-related stigma in general, and has recently been identified by the NIH as an ongoing priority area.[54]

There was little overlap between the instruments developed to measure epilepsy-related stigma and negative attitudes and their use as outcome measures for the interventions reviewed. Most of the authors of the intervention studies developed their own instruments to measure outcomes rather than selecting those available in the literature. One possible reason might be that most of the interventions were educational in nature and that many of the instruments reviewed did not measure knowledge. Future studies and instrument development activities should occur primarily within the framework of intervention development rather than in isolation. Intervention studies would benefit from the use of established guidelines for clinical trial conduct and reporting[55-57] and the use of validated instruments to measure stigma and long-term outcomes assessment to measure the true impact of interventions. Expert review of the existing data on epilepsy stigma with the aim of synthesizing general principles and intervention approaches that could be applied across geographic regions and cultures would significantly advance our capacity to begin to potentially reduce the burden of stigma.

Advances in the discipline of stigma studies over the past 5 years have moved toward unified theories of stigma that transcend individual conditions. There is good evidence that health-related stigma has similar drivers, manifestations, and outcomes across most conditions[58, 59] leading to a call for a framework shift to explore methods and interventions for stigma reduction across a range of disciplines. Intervention studies are costly both in terms of finance and human resources. Well-designed epilepsy-associated stigma reduction interventions will require collaborative efforts involving social scientists, clinical trialists, and epilepsy specialists. Stigma reduction interventions that transcend individual conditions may be able to leverage sufficient resources to incorporate multi-level interventions,[60] engage participatory praxis,[61] tackle the added complexity of being affected by multiple stigmatizing conditions,[62] and address intersectional stigma.[63, 64]

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We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Disclaimer

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Box 1

Recommendations for Future Research

- Investigators who want to measure attitudes or stigma in future studies are encouraged to:
  - use well developed established instruments[#] rather than develop new ones except in children and older adults where gaps exist
  - report on psychometric properties of existing instruments in order to build the body of literature on available instruments
- Studies are needed to develop and test interventions to address negative attitudes and epilepsy-associated stigma.
- Investigators who are planning interventions are strongly encouraged to:
  - explore use of unified theories of stigma that transcend individual conditions
  - engage a community of people living with epilepsy in the development and implementation of the study
  - use existing, well-developed instruments to measure changes in attitudes and stigma
  - conduct randomized clinical trials
  - register the studies in ClinicalTrial.gov

[#]see Tables 1, 2 and 3 for best instruments identified with a *****