CRITICAL REVIEW AND INVITED COMMENTARY

Self-management interventions in pediatric epilepsy: What is the level of evidence?

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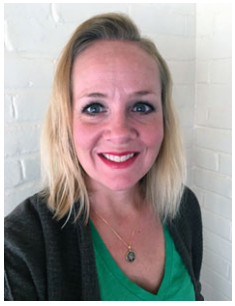

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Summary

Objective: To respond to recommendations put forth by the Institute of Medicine to improve self-management resources for youth with epilepsy by conducting a systematic review of the self-management literature in pediatric epilepsy.

Methods: Inclusion criteria: youth birth to 18 years with a seizure disorder or an epilepsy diagnosis and/or their caregivers, published 1985–2014 in English, and conducted in countries with a very high human development index. Abstract and keywords had to explicitly refer to "self-care" (pre-1996) and/or self-management (post-1996). The review was conducted in seven phases: (1) identification of bibliographical search criteria and databases; (2) abstract assessment; (3) full article review; (4) organization of final citations into instrument development, intervention, factors associated with self-management categories; (5) American Academy of Neurology level of evidence (LOE) assessment for intervention studies; (6) CONsolidated Standards of Reporting Trials (CONSORT) evaluation of LOE level III articles utilizing a control group; and (7) categorization of intervention outcomes across four self-management domains.

Results: Of the 87 articles that met eligibility criteria, 24 were interventions and received LOE scores of level III or IV. Most studies (n = 20, 80%) were scored at level III; however, only eight had a control group and adhered to CONSORT guidelines. They largely neglected information on intervention components (e.g., implementation, treatment fidelity), randomization, participant flow, missing data, and effect size or confidence intervals. The 24 intervention studies reported significant impact in four domains: individual (n = 13), family (n = 6), health care system (n = 3), and community (n = 2).

Significance: There are no level I or II studies. No study met full CONSORT guidelines. Outcomes were well described; however, the nature of self-management interventions (e.g., multiple foci, skills targeted) and the observed heterogeneity in outcomes complicates comparisons across studies. Randomized controlled trials (RCTs) that include large sample sizes, impact of the intervention, treatment fidelity, and power analyses are necessary to further this evidence base.

KEY WORDS: Family management, Behavioral health intervention, Psychosocial intervention, Psychobehavioral intervention.

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Key Points

- Eighty percent of self-management intervention studies reviewed were at level III; only half of these had a control group
- Self-management interventions had a significant impact in individual, family, health care system, and community domains
- No studies achieved level I or II status or adhered to all components of CONSORT. Multisite RCTs are necessary to examine the impact of self-management interventions

Approximately 1% of youth have had a seizure in the past year, and up to 50% of these youth experience psychiatric and/or cognitive comorbidities.[1,2] In youth without comorbidities, epilepsy still impacts daily functioning including social difficulties, worries about seizures, and treatment adherence. Self-management programs are needed to help youth with epilepsy and their families overcome such challenges. According to a 2012 Institute of Medicine (IOM) report, self- and family-management interventions should provide knowledge and skills to manage epilepsy and comorbid conditions, maintain a healthy lifestyle, effectively partner with health care providers, and live independently.[3] Studies have tested self-management interventions for youth with epilepsy and their families, and prior reviews indicate some benefit.[4–6] However, these reviews employ various and ambiguous definitions of self-management, with adult and pediatric studies grouped together, making it difficult to systematically evaluate this literature.

To address the IOM's recommendations for improved pediatric epilepsy self-management resources,[3] the current systematic review utilizes the Modi et al.[7] pediatric self-management model to organize interventions aimed at managing epilepsy or its comorbidities in youth with epilepsy (0–18 years) and their caregivers. Goals of this systematic review are the following: (1) to categorize eligible citations into one of three categories: instrument development, intervention, or factors associated with self-management; (2) to describe self-management interventions appraised through the strength of evidence using the American Academy of Neurology (AAN) level of evidence (LOE) classification system[8]; (3) to evaluate the design of and reporting of results for self-management intervention studies according to CONsolidated Standards of Reporting Trials (CONSORT) guidelines for nonpharmacologic treatment[9]; (4) to describe outcomes of self-management interventions; to (5) identify gaps in the literature; and (6) to make recommendations for future research and clinical practice.

Methods

The Pediatric Epilepsy Self-Management Workgroup of the Managing Epilepsy Well Network[10,11] conducted a systematic review of the extant literature on pediatric epilepsy self-management.

Definitions

Modi et al.[7] define self-management as "the interaction of health behaviors and related processes that patients and families engage in to care for a chronic condition." Their model articulates individual, family, community, and health care system level influences that impact self-management behavior through cognitive, emotional, and social processes. In line with the Pediatric Self-Management Model, the Workgroup employed a comprehensive definition of "self-management" that included the following: (1) factors that may affect seizure control and well-being (e.g., medication/therapy use [adherence], stress, self-efficacy, coping skills [only when mentioned in the context of self-management]); (2) behaviors or steps taken to prevent or cope with the consequences of epilepsy and its comorbidities on physical and mental health in the context of daily life (e.g., psychosocial interventions, adherence, education only interventions, lifestyle modifications); (3) complex role(s) and behaviors of the caregiver(s) in epilepsy management and the dynamic processes that frame caregivers' responses to epilepsy and link caregiver and child well-being; (4) promotion/transition to increasingly independent self-management when developmentally appropriate for the child; (5) navigation of social, health, and community systems; and (6) partnering with the child's health care team and school.[3] An "intervention" was defined to include any psychosocial/psychological/psychiatric/educational treatment. This definition allowed any steps taken to cope with, or behavior changes made to manage, epilepsy or its comorbidities to be considered a self-/family- management intervention (e.g., cognitive-behavioral intervention, adherence intervention, coping skills intervention).

The review was conducted in seven phases: (1) identification of bibliographical search criteria and databases, with initial search results; (2) abstract assessment based on relevance to review focus; (3) full review of articles for eligibility assessment; (4) organization of final set of citations into one of three categories (instrument development, intervention, or factors associated with self-management); (5) AAN LOE assessment for intervention studies (Fig. 1)[8,12]; (6) evaluation of level III intervention studies with a control group according to the CONSORT guidelines for reporting nonpharmacologic treatments[9]; and (7) categorization of all intervention study outcomes across four self-management influence domains: individual, family, health care system, and community,[7] and process (e.g., satisfaction, feasibility) outcomes.

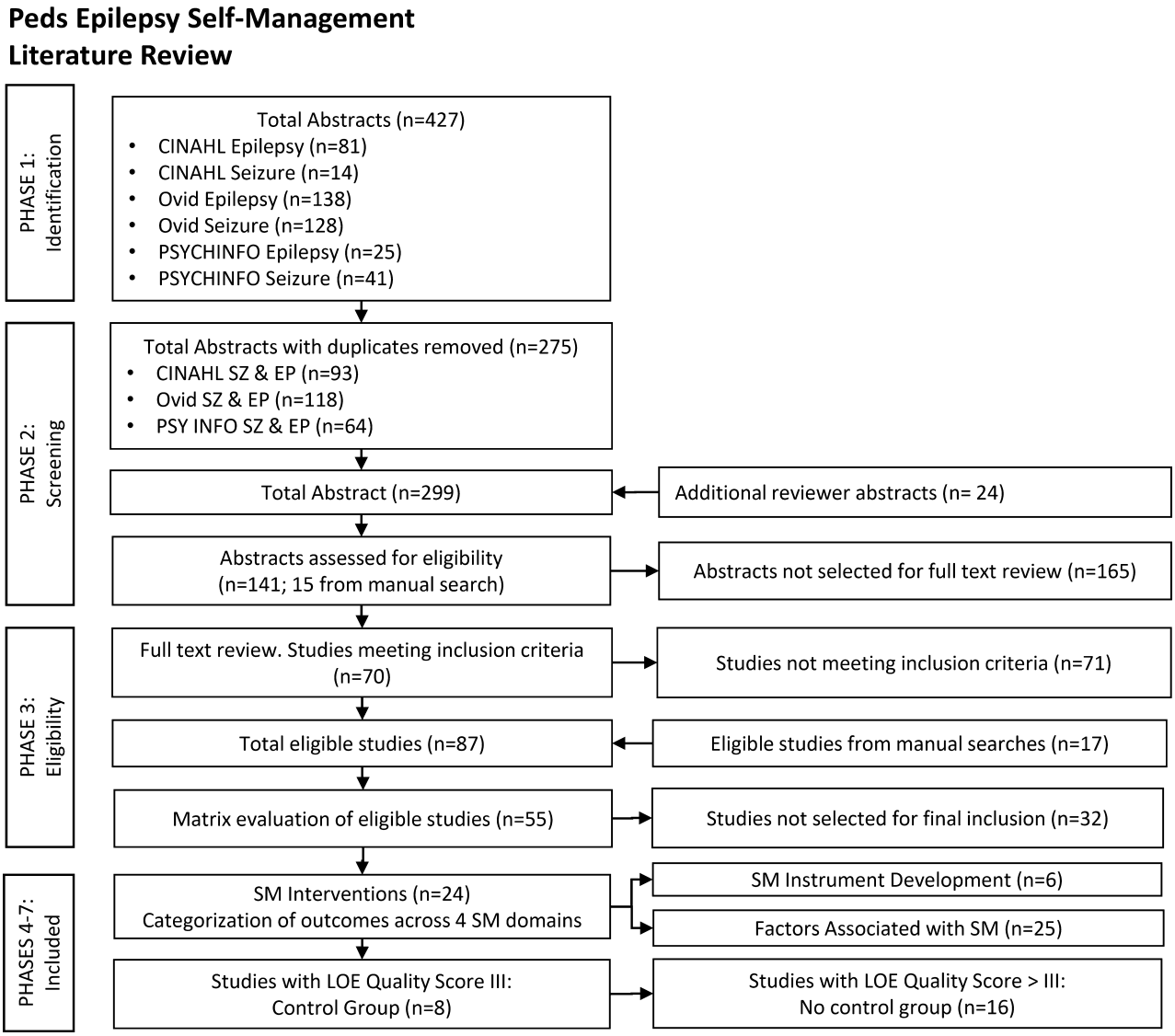

Figure 1.
PRISMA flow chart.
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In the initial phase, a university reference librarian was consulted to review the search process and to ensure the inclusion of all possible search terms, including 53 terms that were identified (Table 1 Search terms) and combined in various manners. Using the selected subject terms, bibliographical searches were conducted in the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and OVID MEDLINE databases.

Inclusion and exclusion criteria

Final inclusion criteria for articles included studies focused on youth from birth to 18 years with a seizure disorder or an epilepsy diagnosis published between 1985 and 2014 written in English, and conducted in the United States or in countries with a very high human development index, as rated by the United Nations Development Program.[13] Studies that focused on caregivers (parents, guardians) of youth ages 0–18 were also included. The study abstract and keywords had to explicitly refer to "self-care" (studies predating 1996) and/or self-management (studies postdating 1996). Study categories included literature reviews, meta-analyses, and original empirical studies. Intervention studies that included specific behavioral health symptoms (e.g., anxiety or depression) as the primary outcome had to target an aspect of self-management (e.g., coping, behavioral change to manage epilepsy, etc.). Unpublished manuscripts, dissertations, and non-English publications were excluded.

Data collection and coding

In phase 1, the comprehensive search strategy yielded 427 citations, with 275 nonduplicated studies. Because subject terms were inconsistently used and indexed in

Table 1. Search terms

Pediatric
Epilepsy-required
Seizure-required
Seizure disorder-required
Chronic illness
Self-determination
Self-management
Self-revelation
Medication management
Family management
Adherence
Psychosocial
Psychosocial issues
Psychosocial problems
Mental health
Behavioral health
Education
Health care
Transition
Life course
Development
Self-efficacy
Coping
Quality of life*
Infant
Infancy
Toddler
Preschool
Child
Youth
Tween
Adolescence
Adolescents
Teen
Teenager
Caregiver
Parenting*
Parent
Family
Family systems
Family problems
Psychological intervention
Program
Group intervention/therapy
Qualitative evaluation*
Quantitative evaluation*
Interventions
Intervention trials
Health outcomes*
Medical home
Generation plurals
Millennium generation

*Indicates search terms with the highest yield.

computerized databases, reviewers were able to identify 24 additional articles by cross-referencing the reference lists with the initial search results. All descriptive and experimental studies that measured specific factors in pediatric self-management initially were included for consideration in phase 1. The subject terms with the highest yield in various combinations were seizure disorder, epilepsy, quality of life, parenting, qualitative evaluation, quantitative evaluation, and health outcomes (Table 1: Search terms marked with *).

In phase 2, the 299 abstracts were divided in subsets of equal or comparable numbers, and assigned to a pair of independent reviewers to be assessed for relevance to pediatric epilepsy self-management. In phase 3, pairs of independent reviewers conducted a full review of the 141 retained articles. Reviewers then selected articles based on the inclusion and exclusion criteria listed earlier.

In phase 4, reviewer pairs conducted a matrix evaluation of the studies, identifying 55 eligible studies. We used the final matrix to review and evaluate studies, which included the reference, quality score, intervention tier (if applicable), sample and setting, study design/methods, survey/instrument(s) used, primary and secondary study outcomes, and reviewer comments. The selected articles were categorized into three groups: (1) self-management instrument development, (2) factors associated with self-management, and (3) self-management interventions.

A final set of self-management intervention studies was identified and included in the review for the current study. In phase 5, independent reviewer pairs evaluated the merit of these studies according to the four AAN LOEs.[8] Because the LOE classification is designed primarily to evaluate pharmacologic or medical interventions (and not for psychosocially focused self-management interventions), the Workgroup added a "tier" to the proposed level III to distinguish those studies employing a control group compared from those without. Due to variability in reviewer scoring, the Workgroup chairs then conducted a final evaluation of the studies and discussed any discrepancies with the committee in order to achieve agreement on a LOE rating for each item (See Fig. 1 for Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart). The risk of bias in study appraisal was addressed through use of the AAN LOE, as well as separate reviews by each committee pair group, independent review within pairs, and full committee review and discussion. The full matrix was reviewed and approved by all members of the Workgroup.

In phase 6, the studies achieving a level III score for AAN LOE, which included a control group, were evaluated for scientific rigor of methodology against the CONSORT guidelines for reporting trials of nonpharmacologic treatments.[9] Finally, in phase 7, outcomes in all intervention studies were categorized into self-/family-management domains.[7]

Data analyses

The team summarized key study characteristics, and LOE quality scores (Table 2) and CONSORT elements (Table 3) were abstracted in tables. In addition, intervention outcomes

across four self-management influence domains: individual, family, health care system, and community,[7] as well as process (e.g., satisfaction, feasibility) outcomes, are presented in Table 2. A supplementary summary of detailed study characteristics is available (Table S1). Key descriptions of outcomes and scientific rigor of study designs are described in a narrative form, and it is noted if the outcomes increased or decreased significantly or had no change.

Results

Overall search results

Overall, the search identified 427 articles related to self-management for pediatric epilepsy and 141 abstracts were included for further review. A total of 87 articles met the inclusion criteria for the larger review of self-management determinants, instrument development, and interventions. Of those, 24 self-management interventions for youth with epilepsy ages 0–18 and/or their caregivers, health care providers, and educators are included in this review (Fig. 1). Table S1 provides a detailed summary of the characteristics of each intervention study evaluated. These 24 intervention studies are reviewed in detail below.

Systematic review characteristics for intervention studies

The 24 studies that met the inclusion criteria represented publications from 1991 through 2014 (see Table 2). The majority of publications have appeared since 2008 (n = 14) with a mode in 2013 (n = 4). The 24 studies that met inclusion criteria for review had LOE scores of level III or IV.[8] Most studies (n = 20, 80%) were at level III; however, only 8 of these had a control group comparison.

The eight level III studies with control group comparison were further evaluated against CONSORT guidelines[9] (Table 3). In all eight studies, outcomes improved following the intervention. Most studies adhered to CONSORT guidelines for description of background information, participants, outcomes, statistical methods and inclusion of objectives (hypotheses), baseline data, and ancillary data analyses. However, studies largely neglected intervention components (e.g., implementation, treatment fidelity), participant flow, sample size determination (e.g., power analyses), how missing data were handled (intent to treat analyses), and effect size or confidence intervals. Six of the studies utilized randomization, but specific information regarding who completed randomization procedures, how randomization was done, and whether assessors were blind to group assignment was not included in the article. No study reported presence or absence of adverse events. Interpretation of findings varied in scope and depth, with some articles[14–16] including a comprehensive discussion of study limitations and evaluation of the study's contribution to the evidence base.

Self-management outcomes

Collectively, the 24 intervention studies reported significant impact on individual (n = 13) and family (n = 6) domains and, to a lesser extent, health care system (n = 3) and community (n = 2) domains. Each study examined between one and 11 outcomes associated with self-management, and 16 studies (67%) reported a significant impact on at least one measured outcome. Thirteen studies (58%) reported a positive impact on process-related factors of satisfaction, feasibility, accuracy, and acceptability of the self-management programs.

Individual outcomes

The impact on individual outcomes was predominantly for the categories of child mental health and behavioral problems (n = 7), self-concept and self-efficacy (n = 6), child knowledge of epilepsy (n = 6), quality of life (n = 5), and social skills, support, and disclosure (n = 5). Of the eight level III (control group) studies, 6 (75%) reported a positive impact on individual outcomes. Of these, the majority of findings were for child mental health and behavioral problems (n = 4) and social skills, support, and condition disclosure (n = 4). Only two studies reported a positive significant effect on adherence and seizures, respectively. No level IV studies reported impact on individual level outcomes.

Family outcomes

The impact on family outcomes was predominantly for the categories of parent knowledge (n = 5), family functioning (n = 3), and parent worries and fears (n = 3). Of the eight level III (control group) studies, only four (50%) reported a positive impact on family outcomes. Of these, the majority of findings were for parent knowledge (n = 3) and parent worries and fears (n = 3). No level IV studies reported a positive impact on family outcomes.

Health system outcomes

The significant impact on health system outcomes was for the categories of therapeutic alliance/communication (n = 1), health care utilization (n = 1), and nursing confidence (n = 1). Of the three level III (control group) studies, there were variable results for both health care utilization and communication/therapeutic alliance.

Community outcomes

The impact on community outcomes was measured only in a minority of studies (n = 3), and the only level III (control group) study found a positive impact on school attendance.

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Table 2. Pediatric self-management outcomes

	Individual outcomes							Family outcomes							Process-related outcomes
Article/quality score	Child mental health and behavior	Quality of life	Adherence/ self-management skills	Self-concept, self-efficacy, confidence, independence	Child knowledge	Social skills, support and disclosure	Seizures	Family function	Parent Stress	Parent worries and fears	Parent knowledge	Confidence/ efficacy	Health care system outcomes	Community	Acceptability, feasibility, satisfaction
Quality score-III (control group)															
Glueckauf et al. (2002)[22]	= Beh	+	= Missed treatment appointments and homework	NR	NR	+ Parent report	NR	+ Teen and parent report	NR	NR	NR	NR	+ Therapeutic alliance mediated by condition	NR	NR
Jantzen et al. (2009)[23]	NR	+	+ Self-management skills	+ Independence	+	+ Disclosure	=	NR	+	+ Parent worries	+	NR	= Health care utilization	NR	+
Lewis et al. (1991)[24]	NR	NR	NR	NR	NR	NR	NR	= Protect/ decision-making	NR	+ Anxiety	+	NR	NR	NR	+ Program benefits
Lewis et al. (1990)[25]	+ Beh	NR	NR	+	+	+ Social interaction and skills; = disclosure	NR	NR	NR	NR	NR	NR	NR	NR	NR
Martinovic et al. (2006)[16]	+ Dep	+	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR
Modi et al. (2013)[15]	NR	NR	=	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Acceptability/ feasibility
Pfafflin et al. (2012)[14]	+ Anx	NR	+ Self-management	= Coping/ adaptation	+	+ Rules and limitations	+	NR	NR	NR	NR	NR	NR	NR	+ Satisfaction
Tieffenberg et al. (2000)[26]	+ Anx	NR	NR	+ Locus of control	NR	NR	+ Seizure reduction	+ Allow sleepover	NR	+ Anxiety/ fear	+	NR	+ ER visits; = Communication with physician	+ School attendance	NR

Continued

Table 2. Continued.

Article/quality score	Individual outcomes							Family outcomes						Health care system outcomes	Community	Process-related outcomes
	Child mental health and behavior	Quality of life	Adherence/ self-management skills	Self-concept, self-efficacy, confidence, independence	Child knowledge	Social skills, support and disclosure	Seizures	Family function	Parent Stress	Parent worries and fears	Parent knowledge	Confidence/ efficacy			Acceptability, feasibility, satisfaction
Quality score-III (no control group)																
Austin et al. (2002)[27]	NR	NR	NR	NR	+	NR	NR	+	NR	NR	+	NR	NR	NR	+ Satisfaction	
Blocher et al. (2013)[28]	+	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Satisfaction	
Buelow et al. (2013)[29]	=	=	NR	NR	NR	NR	NR	=	NR	NR	NR	NR	NR	NR	+ Usability, Feasibility, Satisfaction	
Carbone et al. (2014)[30]	+	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	
Conant et al. (2008)[31]	NR	+	NR	+	NR	NR	NR	NR	=	NR	NR	NR	NR	NR	NR	
Frizzell et al. (2011)[32]	=	NR	NR	+	+	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Satisfaction	
Shore et al. (2008)[33]	+ Dep (child)	+	NR	=	=	NR	NR	NR	NR	=	+	+	=	=	NR	
Snead et al. (2004)[34]	= Dep; = Anx	=	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+	
Wagner et al. (2010)[35]	= Dep; = hopelessness	NR	NR	+ Seizure efficacy: = attitude toward illness	+	+ Seeks support	NR	NR	=	NR	NR	= Parenting SM efficacy	NR	NR	NR	
Wagner et al. (2011)[36]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Satisfaction +/− feasibility + Accuracy	

Continued

Table 2. Continued.

	Individual outcomes							Family outcomes							Process-related outcomes
Article/quality score	Child mental health and behavior	Quality of life	Adherence/ self-management skills	Self-concept, self-efficacy, confidence, independence	Child knowledge	Social skills, support and disclosure	Seizures	Family function	Parent Stress	Parent worries and fears	Parent knowledge	Confidence/ efficacy	Health care system outcomes	Community	Acceptability, feasibility, satisfaction
Quality score-IV															
Austin et al. (2010)[37]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Nurse confidence	NR	+ Satisfaction
Guilfoyle et al. (2013)[38]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Acceptability, feasibility, uptake
Hoare and Kerley (1992)[39]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	− Feasibility/ acceptability
Hufford et al. (1999)[40]	NR	NR	NR	NR	NR	NR	NR	=	NR	NR	NR	NR	= Therapeutic alliance	NR	+ Acceptability, + Comfort with technology by condition
Jurasek et al. (2010)[41]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Satisfaction
Price et al. (2004)[42]	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	+ Special needs educators knowledge; + self-efficacy in seizure management and working effectively with families	NR

NR, not reported or measured; Anx, anxiety; Dep, depression; Beh, behavior; Prob, problems.
+ Symbol indicates the outcome increased significantly; = no change in the outcome; − indicates the outcome decreased significantly

Process outcomes

Acceptability, feasibility, and satisfaction data were reported in a majority of the studies (n = 15), with positive findings for most of these, including n = 4 of the eight level III studies. Satisfaction was the most common patient/caregiver reported outcome. Notably, most level IV studies reported only on process variables.

Conclusions

This systematic review describes and critiques self-management (psychosocial, psychological, psychiatric, or educational) interventions to manage epilepsy or its comorbidities in youth with epilepsy and their caregivers. This is the only known review that provides such a comprehensive, yet well-defined, conceptualization of self-management, and that was not limited to randomized, controlled trials,[17] evaluates intervention studies per CONSORT guidelines, and systematically organizes study outcomes across self-management influence domains (e.g., individual, family, health care, and community[7]). Given the paucity of self-management intervention studies, it is important to include pilot work in the evaluation of this literature.

The identified 24 self-management intervention studies included pilot projects, randomized controlled trials, and within-subject designs. When studies were evaluated with the AAN's levels of evidence,[8] only one third (n = 8) of the interventions met criteria for level III and employed a control group. No studies received a score above level III. Two thirds of studies were pilot studies with smaller sample sizes and no control group and/or had employed less rigorous designs than randomized controlled trials (RCTs). The eight level III studies with control groups adhered to CONSORT guidelines for descriptive information (e.g., background, inclusion/exclusion criteria, objectives [hypotheses], outcomes), and general statistical analytic plans (comparison of groups' baseline data, ancillary data analyses). However, the studies largely neglected to include participant flow charts and salient analytic methods (e.g., intent to treat analyses, power analyses, effect size, or confidence intervals). Therefore, we are unable to evaluate whether a study was adequately powered to detect true differences between groups.

Notably, some studies were pilot studies and contained very small sample sizes, limiting analytic plan choices.[15] Although details on randomization procedures were limited, it should be noted that blinding to group is impossible for those providing the treatment in psychological intervention studies. Study design may allow for those persons conducting pre-post assessments to be blind to group assignment; however, the articles reviewed did not include this information. Studies also did not include important components of the interventions (e.g., how interventions were implemented, who disseminated the intervention, or whether therapists followed the intervention protocol [i.e., treatment fidelity]). It should be noted that six of the eight studies were published prior to the current CONSORT statement and therefore did not have guidelines for publication of their nonpharmacologic intervention trials. Indeed, the two studies published following the CONSORT statement were stronger in scientific rigor and more thorough in their presentation of methods and design compared to the other six. Across the six studies prior to CONSORT, there does not appear to be a trend for improved reporting over time in any specific CONSORT domain.

Types of interventions (N = 24) and their outcomes were organized within the framework of the Modi et al.[7] model of pediatric self-management. Not surprisingly, individual outcomes were most commonly reported. However, few studies included the same individual outcomes. Half of the studies reported child behavioral health problem outcomes, with the most common being depressive and anxiety symptoms. Across the studies reporting behavioral health outcomes, various assessment measures (e.g., diagnostic interview, self-report behavioral rating scale) were used to capture these outcomes. Health systems and community level outcomes were reported in only a minority of studies, and only 2 of the 24 intervention studies reported adherence outcomes. There is a trend over time for intervention studies to include process-related measures such as satisfaction, feasibility, and acceptability.

Indeed, investigators have not utilized common outcome measures within these four self-management domains. The nature of self-management interventions (e.g., multiple foci, skills targeted) and the heterogeneity in outcomes observed in this review illustrate a dilemma borne of the need for a common set of outcomes and data elements, and the need to measure specific outcomes targeted by the intervention. In other words, an intervention aimed at improving antiepileptic drug adherence might include an electronic monitoring device to assess adherence as a primary outcome, and an intervention focused on enhancing coping skills would include self-report coping skill outcomes, but not necessarily a measure of AED adherence. Therefore, it is challenging to compare outcomes across interventions. The National Institute of Neurological Disorders and Stroke (NINDS) has created common data element (CDE) recommendations based on the available evidence for seizure, cognitive, and behavioral outcomes.[18] As we move forward and design multisite RCTs, use of these CDEs, which include depression and quality of life, will be imperative. The Pediatric Epilepsy Research Consortium (PERC; www.pediatricerc.com) is currently engaged in efforts to discern common behavioral health outcomes across epilepsy clinics and encourage clinical research sharing opportunities for clinics across the United States.

Table 3. CONSORT guidelines for reporting nonpharmacologic treatment studies

	Intro	Methods								
Article	Background	Participants	Interventions	Objectives	Outcomes	Sample size	Randomization-sequence generation	Allocation concealment	Implementation	Blinding
Glueckauf et al. (2002)[22]	+	+	*	+	+	–	*	–	–	–
Jantzen et al. (2009)[23]	+	+	*	*	+	–	NA	NA	NA	–
Lewis et al. (1991)[24]	+	+	*	–	+	–	*	–	–	–
Lewis et al. (1990)[25]	*	*	*	–	+	–	*	–	–	–
Martinovic et al. (2006)[16]	+	+	*	+	+	*	+	–	+	–
Modi et al. (2013)[15]	+	+	+	+	+	–	*	–	–	–
Pfafflin et al. (2012)[14]	+	+	*	*	+	–	NA	NA	NA	–
Tieffenberg et al. (2000)[26]	+	+	+	+	+	*	*	–	–	–

NA, not applicable.
+ Symbol indicates the article sufficiently reported the CONSORT checklist element; * symbol indicates the article reported partial information for the CONSORT checklist element; – symbol indicates the article did not report the CONSORT checklist element.

In a number of studies (25%; n = 6), there were no improvements across the four self-management domain outcomes postintervention. Possible explanations include lack of power due to small sample sizes, shorter time frames for evaluation, choice of outcome, or ineffective/less impactful intervention strategies. To further illustrate, outcomes such as quality of life may require a longer time postintervention to improve; therefore, careful consideration of time frame between postintervention and follow-up assessments is necessary. The choice of outcome is also critical. If investigators choose an outcome that is not directly addressed in the intervention (e.g., is not a target of the intervention) or does not have a strong indirect relationship with the content or skills taught as part of the intervention, such outcome will likely not improve with treatment. For example, investigators may have chosen measures of anxiety or depression outcomes for self-management focused interventions because there was no available psychometrically sound measure of pediatric epilepsy self-management. Some self-management interventions may be developed solely to enhance behaviors related to daily epilepsy management; therefore, we would not expect them to improve depressive symptoms, particularly, if the sample of participants targeted was not necessarily depressed or at risk for depression. Investigators are encouraged to carefully select outcomes related to intervention aims and the time points at which to assess them.

In summary, these 24 intervention studies provide a preliminary evidence base for self-/family-management interventions in pediatric epilepsy. Generalization of findings is somewhat limited given the smaller sample sizes, and geographical (e.g., international locations) and variety of outcomes assessed. However, pilot studies and feasibility and satisfaction data offer pertinent "lessons learned" for the future development and dissemination of such interventions. Improved outcomes postintervention reported in the more rigorously designed RCTs are promising for the efficacy of self-/family-management interventions. However, to improve the level of evidence for such interventions, improvements in the rigor of study design, implementation, and reporting of findings are necessary.

Limitations to the current review

The current study is not without limitation. Bias could have been introduced into the scoring of articles and assignment of LOE; however, independent reviewer pairs were employed to reduce individual bias. When reviewers disagreed, the Workgroup chairs evaluated the studies and made the final decision on scores. Meta-analyses are the gold standard for evaluating literature; however, we were unable to perform a meta-analysis due to the high level of variability in outcomes reported across the studies.

Future directions

Moving forward, it will be important that clear, concise definitions of self-management and theoretical models are utilized to develop self-management interventions. Investigators are encouraged to collaborate with other sites to provide the resources necessary to conduct RCTs to evaluate the impact of self-management interventions for youth with epilepsy and their caregivers. Only then will such intervention studies contain the rigorous scientific design

Results									Discussion		
Statistical methods	Participant flow	Implement intervention	Recruitment	Baseline data	Numbers analyzed	Outcomes and estimation	Ancillary analyses	Adverse events	Interpretation	Generalizability	Overall evidence
+	–	*	*	+	*	*	+	–	*	+	+
+	–	*	+	+	–	+	+	–	*	+	–
+	–	–	+	+	–	*	+	–	*	*	*
+	–	–	*	+	–	*	+	–	*	*	*
+	*	–	–	+	*	*	+	–	+	*	+
+	+	+	*	*	*	*	+	–	+	*	+
+	+	+	+	+	*	*	+	–	+	+	+
+	*	+	*	*	*	+	+	–	*	+	+

components required to be scored as level 1 or II. For example, given the high rate of intellectual disability in youth with epilepsy[1] and the neurocognitive skills required for some behavioral health interventions (cognitive-behavioral interventions), multisite design is necessary to recruit a sufficient number of participants to power comparison analyses. In addition, it will be necessary to consider use of CDEs and their applicability to the target skills/behaviors of the intervention. For studies targeting behaviors/skills other than medication adherence, it will be necessary to use psychometrically sound outcomes that measure behavior change in the particular domains of self-management in which the intervention targets. Given the absence of pediatric epilepsy self-management surveys or tools, this will require development and validation of such measures prior to execution of an RCT.

Very few studies have examined the benefit of behavioral health interventions on health care system and community domains, and inclusion of these salient influences on self-management behaviors in intervention development is likely valuable. Only two studies to date have evaluated the impact of intervention on adherence to antiepileptic drugs. Given the high rates of nonadherence[19] and significant negative outcomes associated with nonadherence to antiepileptic drugs,[20,21] it will be crucial to further develop and implement interventions to address medication adherence. In conclusion, the development of high-quality intervention studies for pediatric epilepsy self-management hinges on a concise and clear definition of self-management; multisite recruitment for RCT designs; well-defined intervention components with targeted skill development in particular self-management domains; use of psychometrically sound self-management behavior/skill outcomes that map onto the skills targeted in the intervention (use of existing CDEs when applicable); and reporting of methods, analyses, and findings in accordance with CONSORT (e.g., randomization and intent to treat analyses).

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Disclosure

The authors declare no conflicts of interest. 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.

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Supporting Information

Additional Supporting Information may be found in the online version of this article:

Table S1. Summary table of articles.