Epilepsia®

Measurement in pediatric epilepsy self-management: A critical review

Gigi Smith[1] | Avani C. Modi[2] | Erica K. Johnson[3] | Ross Shegog[4] | Joan K. Austin[5] | Janelle L. Wagner[1]

[1]College of Nursing, Department of Pediatrics, Comprehensive Epilepsy Center, Medical University of South Carolina, Charleston, SC, USA

[2]Center for Treatment Adherence and Self-Management, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA

[3]School of Public Health, Health Promotion Research Center, University of Washington, Seattle, WA, USA

[4]Center for Health Promotion and Prevention Research, UT Health School of Public Health, Houston, TX, USA

[5]School of Nursing, Indiana University-Purdue University Indianapolis, Bloomington, IN, USA

Correspondence
Gigi Smith, Medical University of South Carolina, College of Nursing, Charleston, SC, USA.
Email: smithgi@musc.edu

Summary

Given the paucity of information available regarding self-management, the aims of this paper are to synthesize the literature on factors associated with and measures to assess self-management in pediatric epilepsy. 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. The review was conducted in 6 phases: (1) identification of bibliographical search criteria and databases; (2) abstract assessment; (3) full article review; (4) organization of final citations into categories; (5) identification of predictors, potential mediators/moderators, and outcomes associated with self-management factors and categorization of factors as influences, processes, or behaviors across individual, family, community, and health care domains; and (6) critique of self-management instrument studies. Twenty-five studies that evaluated factors associated with self-management were identified. Individual and family-focused factors were the most commonly studied predictors of self-management, with psychosocial care needs and self-efficacy for seizure management identified as key factors associated with pediatric epilepsy self-management. Few studies have included mediator and moderator analyses. Measures of adherence were the most commonly used outcome. There has been a predominant focus on pediatric epilepsy influences and processes that are modifiable in nature, potentially at the expense of evidence for the role of community and health systems in pediatric epilepsy self-management. The 6 self-management instrument tools reported scientific rationale and good psychometric properties. Results highlight several key modifiable cognitive and behavioral targets for skills development: adherence, self-efficacy for seizure management, attitudes toward epilepsy, and family variables. Moving forward, a comprehensive pediatric epilepsy self-management model, well-validated measures of self-management behaviors, mediator/moderator designs to examine the complex relationships between predictors and pediatric epilepsy self-management outcomes, and studies examining the community and health care domains of self-management are necessary.

KEYWORDS

behavioral health assessment, pediatric epilepsy, psychosocial assessment, self-management factors, self-management instruments

1 | INTRODUCTION

The impact of epilepsy on a child and family includes several challenges beyond the unpredictability of seizures, such as complicated treatment regimens (eg, diet, medication) and psychosocial comorbidities. These challenges affect daily life, place significant economic and social burden on families, and are associated with poor quality of life.[1-3] The Institute of Medicine[4] has identified self-management assessment and intervention as salient methods to improve epilepsy outcomes. A growing body of literature, primarily in adult epilepsy, has shown promise for the effectiveness of self-management interventions.[5-11] However, a recent systematic review of 24 pediatric epilepsy self-management intervention studies revealed that none of the pediatric epilepsy interventions met criteria for Level I or II evidence.[12,13] Furthermore, in many cases, self-management outcomes were not well described, and there was a wide diversity of intervention targets and outcomes.[13] There are only a few pediatric epilepsy self-management scales and they assess only a single domain (eg, Modi's medication adherence),[14] and there are a few pediatric generic self-management scales (eg, Medication Adherence Measure [MAM])[15]; however, these scales do not assess domains or tasks specific to pediatric epilepsy. To begin to address this research gap in pediatric epilepsy, we must (1) ascertain the factors that contribute to self-management outcomes and consider them as interventions targets and (2) evaluate the existing measures of self-management in pediatric epilepsy.

As a follow-up to the Wagner and colleagues[13] critique of interventions, aims of the current paper are 3-fold: (1) to identify self-management factors as predictors, mediators/moderators, and outcomes and categorize them according to Modi and colleagues'[16] pediatric self-management model; (2) to evaluate existing self-management instruments according to psychosocial measurement guidelines[17]; and (3) to make recommendations for future research and clinical practice.

2 | METHODS

The Pediatric Epilepsy Self-Management Workgroup of the Managing Epilepsy Well Network[18,19] conducted a systematic review following the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines[20] of the extant literature on pediatric epilepsy self-management. The review and systematic evaluation of self-management interventions has been published recently.[13] Therefore, the current study reviews the factors associated with self-management factors and instruments developed to measure pediatric epilepsy self-management.

Key Points

- Necessity of a comprehensive conceptual model of pediatric self-management in epilepsy
- Development of pediatric self-management instruments specific to epilepsy

Knowledge regarding:

- ○ Role of family (beyond child and parent), health care, and community domains to assist with intervention development and evidence-based guidelines
- ○ Inherent complexities of self-management instruments and how to translate this knowledge into practice
- Development of evidence-based pediatric epilepsy self-management interventions

---

2.1 | Conceptual model

Definitions and models utilized in this comprehensive pediatric epilepsy self-management review have been extensively highlighted in our previous paper.[13] Similarly, the aims of the current study use as a framework the Modi and colleagues[16] pediatric self-management model. In this model, self-management is defined as "the interaction of health behaviors and related processes that patients and families engage in to care for a chronic condition.[16]" Central to the model are system level influences, processes, and behaviors, which interact across individual, family, community, and health care domains to impact health outcomes (eg, quality of life, adherence). Influences (eg, health beliefs, treatment knowledge, peer support) are either modifiable or non-modifiable contextual variables that promote or detract from the conduct of health behaviors and impact self-management behaviors through cognitive, emotional, and social processes. Processes can also be modifiable or non-modifiable and are critical cognitive, emotional, social, and perception variables (eg, coping, psychosocial care needs) that link self-management behaviors and the variables that influence them. Self-management behaviors are actions performed in the context of care for epilepsy. Influences, processes, and behaviors interact. It is important to identify such potentially modifiable influences and processes for a particular condition because they may be intervention targets to enhance self-management behaviors and quality of life.[16]

2.2 | Inclusion and exclusion criteria

Inclusion criteria for articles reviewed were: 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 Programme.[21] Studies that focused on caregivers (eg, 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) to be included. Study categories included literature reviews, meta-analyses, and original empirical studies. Unpublished manuscripts, dissertations, and non-English publications were excluded.

2.3 | Data collection and coding

Data collection and coding involved 6 phases. The first 4 phases of the review are detailed in Wagner et al.[13] and included the following: (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; and (4) organization of final set of citations into 1 of 3 categories (instrument development, intervention, or factors associated with self-management). In phase 2, we also identified potential studies from the reference lists of the abstracts selected for inclusion per the literature search. Phases 5-6 are specific to the current study and are explained in the following paragraphs. See Figure 1 for the PRISMA flow chart, abbreviated from Wagner and colleagues,[13] to include only the information relevant to the current study.

2.3.1 | Phase 5

The research team summarized the studies by first identifying and examining predictors, potential mediators/moderators, and outcomes associated with self-management factors. Notably, some factors (eg, self-efficacy) were identified as predictors in one study and mediators or outcomes in another study; more detail on this is below. The factors (ie, predictors, mediator/moderator, and outcomes) in these studies were then categorized as influences, processes, or behaviors according to the Modi et al.[16] model. The results of these 2 steps were abstracted into the data presented in Table 1.

2.3.2 | Phase 6

Studies that focused on self-management instrument development were critiqued according to Holmbeck and Devine's[17] criteria for psychosocial measure development and are presented in Table 2. Key descriptions of self-management factors, their frequency of study and associated measurement, as well as a critique of the scientific rigor of instrument development studies are presented below.

3 | RESULTS

3.1 | Self-management factors

In Aim 1, 25 studies that evaluated factors associated with self-management were identified per the search inclusion criteria. As mentioned earlier, depending on the study and its design, any variable (eg, self-efficacy) could potentially be conceptualized by the original study authors as a predictor, a mediator/moderator, or an outcome variable.

3.2 | Identifying self-management factors as predictors, mediators/moderators, and outcomes

3.2.1 | Predictors

Studies that examined predictors of self-management outcomes most frequently evaluated parent variables (family mastery, psychosocial care needs) (n = 5), child/parent self-efficacy (n = 4), and attitude toward epilepsy (n = 3). For example, attitudes were identified as a significant predictor of medication-giving behavior[22] and child depressive symptoms.[23] Self-efficacy and family mastery were significant predictors of attitude toward epilepsy.[24] Seizure control trajectory group status was predicted by adherence group status.[25,26] Other predictors of adherence included number of previous seizures, number of behavioral restrictions, parent/child satisfaction with care, and parent anxiety about child's health,[27] as well as socioeconomic status.[28,29] Predictors of self-management included children's educational qualifications/achievement, cognitive and physical effects, and seizure effects.[30]

3.2.2 | Mediators/moderators

Most studies did not identify mediators or moderators of self-management via statistical analyses. Notably, research into these factors is mostly limited to regression analyses with only limited attempts at structural equation modeling. Of those that did include mediator and moderator analyses, (n = 3), children's belief in personal control over health increased with age.[31] In one study, children's worries and self-efficacy for seizure management mediated the relationship between psychosocial care needs and attitude toward epilepsy.[24] Finally, hopelessness was identified as a mediator between the attitude toward illness and depressive symptom relationship after controlling for self-efficacy.[23]

3.2.3 | Outcomes

Measures of adherence (n = 8), followed by behavioral/mental health functioning (n = 5) and health beliefs and
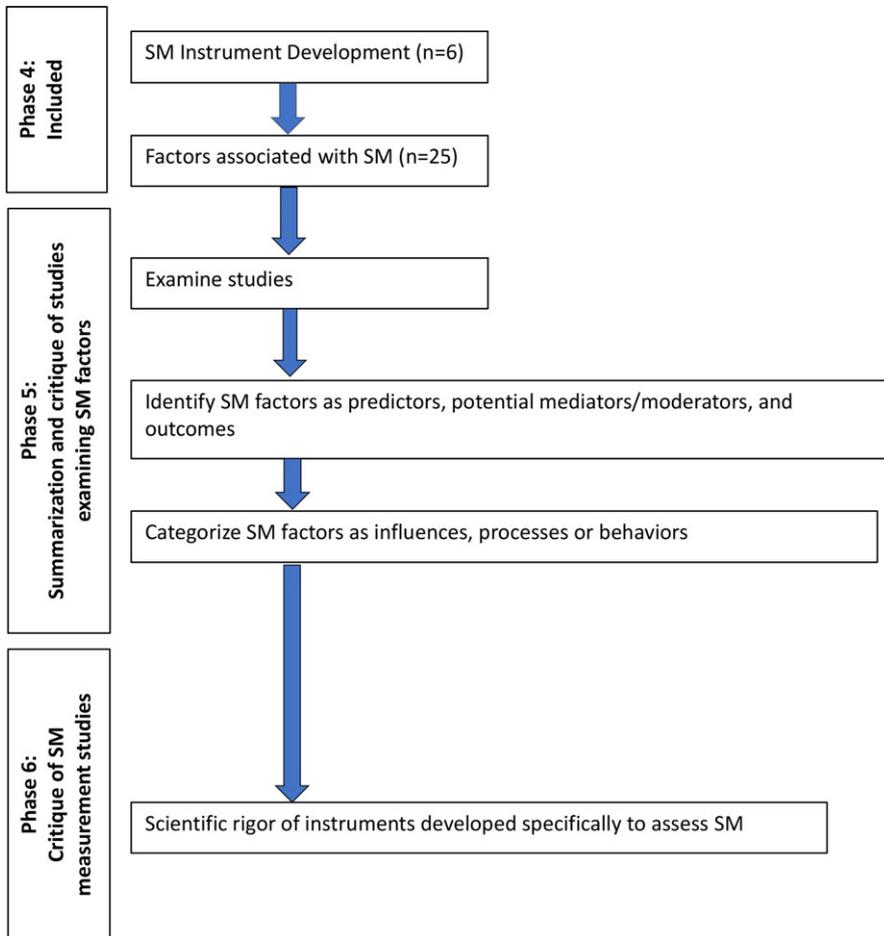
FIGURE 1 Abbreviated PRISMA flow chart for pediatric self-management (SM) studies[13]

behaviors (ie, attitudes, condition management) (n = 4) were the most frequently reported outcomes. Of the adherence studies, 5 used the same measurement tool (ie, MEMS TrackCap electronic device; Aradex Corporation Union City, CA, USA),[25,26,28,29] and one study[32] also used the Pediatric Epilepsy Side Effects Questionnaire.[33] Notably, these studies originated from the same lab. None of the other studies (n = 3) used the same measurement tool.[33–35] The Children's Depression Inventory[37] (CDI) was used in 3 of the 5 studies with behavioral/mental health outcomes,[23,38,39] and the Wagner study[39] also used the Behavior Assessment System for Children-II, Depression Clinical Subscale.[40] The Child Attitude Towards Illness Scale[41] (CATIS) was used in 2 studies,[23,42] and the Parent Response to Child Illness Scale[1] (PRCI) was also used in 2 studies.[42,43]

3.3 | Categorizing factors as influences, processes, and behaviors

3.3.1 | Influences

Of the 25 studies reviewed, 18 (72%) included discernable contextual influences. Most identified influences focused on the individual level of the child and were predominantly modifiable in nature. For example, the most commonly identified modifiable influences included self-efficacy, attitude toward epilepsy, and behavioral health (eg, overall psychological functioning, depression).[22–24,30,35,38,39,43–47] Three of the 4 studies that measured self-efficacy used the Seizure Self-Efficacy Scale for Children[48] (SSES-C), and 1 study[35] used the General Self-Efficacy Scale.[49] Attitudes were almost uniformly measured using the CATIS,[41] and behavioral health was measured with a variety of instruments, such as the Child Behavior Checklist[50] (CBCL), the Positive and Negative Affect Schedule[51] (PANAS), the CDI,[37] and the Hopelessness Scale for Children[52] (HSC). Other, less frequent, modifiable influences included stigma,[30,43] locus of control,[31,35] independent living and social skills,[45,47,53] fatigue,[45] and satisfaction with and understanding of care.[27,31,42] The only nonmodifiable influence identified was cognitive ability, which was explicated in 2 studies.[34,44]

The second most frequent domain of influences was reported in the family system. All reported influences were modifiable and included variables such as family mastery and communication,[24,43] parental attitudes, knowledge, and expectations, and anxiety or concern about child health. Only one study[54] focused on both community-level and health care system modifiable influences. Specifically,

TABLE 1 Pediatric self-management factors

Author(s)	Outcome	Predictors	Mediators/moderators	Influences	Processes	Behaviors
Asato et al.[33]	Adherence			Cognitive ability	Productivity at school	Medication adherence
Austin et al.[21]	Giving medications	• Behavioral intention • Parent attitude		Health behavior attitude	Child psychosocial care and health care needs	Medication giving by parents
Austin et al.[23]	Attitude toward epilepsy	• Family mastery • Self-efficacy	• Child worry • Self-efficacy	• Child worry • Self-efficacy • Family mastery and communication • Attitude toward epilepsy	Parent and child psychosocial care and health care needs	Family communication
Austin et al.[42]	Behavior problems at baseline and 24 mo	• Family mastery • Confidence in discipline • Parent worry • Encouragement of autonomy • Stigma • Parent information and support needs • Provision of child support		• Parent affect • Parent response to illness • Family mastery and communication • Stigma • Self-efficacy	Parent and child psychosocial care and health care needs	• Child behavior problems/issues • Parent epilepsy management • Family communication
Aylward et al.[27]	Persistence in medication taking behaviors	Socioeconomic status				Medication adherence
Bellon et al.[43a]	Parent epilepsy treatment involvement			• Child mental/physical health • Self-efficacy • Cognitive ability		Parent epilepsy treatment involvement
Buelow et al.[54a]	Sources of stress for parents			• Concerns about child • Communication with health care and school • Community support		Parent communication with epilepsy team

(Continues)

TABLE 1 (Continued)

Author(s)	Outcome	Predictors	Mediators/moderators	Influences	Processes	Behaviors
Carbone et al.[35]	Self-reported treatment adherence	• Epilepsy treatment knowledge and expectations (adolescent) • Number of siblings • Number of antiepileptic medications		• Locus of control • Self-efficacy • Treatment knowledge • Treatment expectations		Medication adherence
Dahl[53a]	Seizure behaviors			• Attitude • Independent living skills • Social skills		Self-monitoring
Elliott et al.[45a]	Quality of life			• Unpredictability of seizures • Medication side effects • Exclusion by friends • Parent limits • Self-efficacy • Independent living • Social skills	Social isolation	• Fatigue • Emotional distress • Lack of confidence
Hazzard et al.[27]	Adherence	• Number of previous seizures • Behavioral restrictions • Parent/child satisfaction with care • Parent anxiety about child health		• Behavioral restrictions • Parent anxiety about health • Parent/child satisfaction and understanding of care		• Medication adherence • Parent behavioral restrictions
Kerr et al.[30a]	Impact of epilepsy			• Parent and family functioning • Hope • Self-esteem • Stigma		
Kirton et al.[47]	Quality of life	Living with a seizure-sensing dog		Living with seizure-sensing dog		
Modi et al.[29]	Electronically monitored adherence	Socioeconomic status				Medication adherence

(Continues)

TABLE 1 (Continued)

Author(s)	Outcome	Predictors	Mediators/moderators	Influences	Processes	Behaviors
Modi et al.[32]	Electronically monitored adherence	Clinic appointments				Medication adherence
Modi et al.[25]	Seizures	• Presence of epileptiform discharges on EEG • Recognizable epilepsy syndrome • Adherence status				Medication adherence
Modi et al.[26]	Seizures	• Electronically monitored adherence • Pediatric epilepsy side effects Questionnaire				Medication adherence
Nolan et al.[55a]	Coping responses					Coping responses
Painter et al.[36]	Recreation time	Child having epilepsy				• Treatment adherence • Recreational activities
Reeve et al.[46]	Coping responses	Child having epilepsy		• Self-esteem • Self-efficacy	Coping responses	
Shagena et al.[31]	Understanding of disease	Internal control	Mediator: Age	• Understanding of illness • Health locus of control • Satisfaction with and understanding of care		
Shore et al.[42]	Coping responses			Parent information and support needs	Parent and child psychosocial and health care needs	• Parent need for epilepsy information and support • Parent epilepsy management
Wagner et al.[39]	Depressive symptoms	Coping style		Self-efficacy	Coping responses	• Depression comorbidity effects • Coping behaviors
Wagner et al.[38]	Depressive symptoms	• Caregiver self-efficacy • Caregiver report of child's functioning (BASC-II)		• Self-efficacy • Child behavioral health • Depressive symptoms	Coping responses	

(Continues)

TABLE 1 (Continued)

Author(s)	Outcome	Predictors	Mediators/moderators	Influences	Processes	Behaviors
Wagner et al.[23]	Depressive symptoms	• Child attitude toward illness • Hopelessness • Self-efficacy	Moderator: Hopelessness	• Self-efficacy • Child depression • Hopelessness • Attitude toward illness	Coping responses	

aDenotes qualitative, descriptive, or review studies; therefore, "outcomes" are interpreted with caution as the main variable of interest.

community support and communication, and communication between the health care and school systems were examined.

3.3.2 | Processes

A few studies examined specific processes that are needed to optimize self-management. Nine studies reported 3 different key process level variables, including parental and child psychosocial and health care needs[22,24,42,43] and coping.[23,38,39,46,55] Data from these studies suggested that parents and children have support and information needs, as measured by the Parent and Child Report of Psychosocial Care,[56,57] related to their epilepsy that should serve to enhance self-management behaviors if this level of information was provided.

Difficulty in coping with epilepsy was conceptualized as a barrier to effective self-management, which could also be considered a target for interventions to improve self-management. Children's coping behavior in relationship to the diagnosis of epilepsy has been assessed using the KID-COPE survey,[58] which assesses 10 coping strategies for chronic disease (distraction, social withdrawal, wishful thinking, self-criticism, blaming others, problem solving, emotional regulation, cognitive restructuring, social support, and resignation).[38] A child version (7 to 12 years old) consists of 10 questions with 5-point Likert scale response choices. An adolescent version (ages 13 to 17 years old) consists of 15 questions corresponding to 10 coping strategies with 3-point Likert scale response choices. Responses include both how often a particular coping strategy is used (ie, frequency), and how much it helped (ie, efficacy).[58] A 45-item scale, the Coping Health Inventory for Children (CHIC), has been used to obtain parental assessment of the child's coping behaviors.[38,59]

Notably, coping is a complicated phenomenon and involves both cognitive processes and behavioral responses. Therefore, researchers have conceptualized coping both as a process and a behavior, and coping instruments have been designed accordingly.[37] For example, the KIDCOPE focuses on coping processes, which are largely cognitive in nature (eg, distraction, cognitive restructuring, self-criticism, etc.) and includes only a few behaviors (social support, problem solving).[58] Conversely, the CHIC is more heavily focused on coping behaviors (compliance with treatment, seeking support, acting out).[59] We have included coping as a process level variable, given that coping behaviors are typically exhibited as a result of underlying cognitive process.

3.3.3 | Behaviors

Of the 25 studies reviewed, 17 (68%) included specific measures of self-management behaviors as described in the

TABLE 2 Criteria for measure development[17]

Instrument	Scientific need	Content validity	Reliability	Quantitative item analysis	Factor analysis	Validity	Clinical utility	Cost-effectiveness	Translated to other languages
Psychosocial Care Needs of Children[56]	+	+	+	−	−	+	−	−	+
Parent Response to Child Illness Scale[1]	+	+	+	+	+	+	−	−	−
Seizure Self-Efficacy Scale for Children[48]	+	+	+	+	+	+	−	−	+
Denye's self-care agency for adolescents[63]	+	+	+	−	−	+	−	−	−
Pediatric Epilepsy Medication Self-Management Questionnaire[14]	+	+	+	+	+	+	+	−	−
Allocation of Treatment Responsibility Scale[64]	+	+	+	−	−	+	−	−	−

Modi et al.[16] model. Behaviors were focused on the individual and family levels and included assessment of youth self-management behaviors and parent support behaviors. In these studies, behavioral assessments were gathered either from youth (n = 7), parents (n = 5), or both youth and parents (n = 5).

Individual behaviors

The predominant behavior assessed was the child's adherence to antiepileptic medication (AED) (n = 8). Medication adherence was mainly assessed using an electronic monitoring system (MEMS TrackCap; n = 5).[25–27,29,32] Other studies used survey methods. Carbone et al.[35] administered the Pediatric Epilepsy Medication Self-Management Questionnaire to parents and the Adolescent Medication Self-Management Questionnaire to youth. Both surveys have a medication adherence subscale. Other studies used survey items developed specifically for the study that queried missed doses or stopped medications[27,34] (n = 2). Hazzard et al.[27] used a biological marker, blood serum assays, to additionally validate adherence.

Disease-specific treatment regimens (eg, taking AEDs, going to clinic appointments, calling the doctor/nurse, filling prescriptions) were assessed using daily phone diary (DPD) protocols with parents of children with epilepsy.[36] The DPD is a computerized system with which interviewers utilize a cued-recall procedure to record all activities that lasted 5 minutes or longer over a 24-hour period.[60,61] Finally, Dahl[53] assessed self-management of seizure episodes using written seizure logs to describe self-monitoring (recognizing seizure onset), behavioral response to seizures (eg, relaxation techniques), and self-reinforcement practices.

Parent/caregiver/family behaviors

Parent management behaviors were measured using differing assessment methods. Austin[22] assessed parent AED-giving behavior using a 6-item survey created by the study authors. Parent management of the child's epilepsy was assessed through the condition management subscale of the Parent Response to Child Illness Scale.[42,43] Parents' restriction of their child's activities due to epilepsy was assessed with a survey by measuring the frequency of the number of behavioral activities restricted based.[27]

Parental involvement with the epilepsy care team in developing the management plan was assessed using a single item querying direct involvement in writing the management plan with a yes/no response format.[44] Parent communication with health care providers, interaction with the school, and degree of community support was assessed using semistructured interviews.[55] An indirect assessment of behaviors to seek information and social support was measured using the Parent Report of Psychosocial Care Scale,[56] in which parents responded to questions querying their needs for information on epilepsy and its management (n = 6 items) and their needs for support in relation to their child's seizures (n = 8 items).[24,41] The assessment was also administered with youth using the Child Report of Psychosocial Care Scale.[22,57]

Assessment of family mastery accounts for a family's capacity to manage a chronic illness episode and includes assessment of behaviors including the level of cooperation among family members and degree of communication of concerns among family members.[24,43] This has been measured using the Family Inventory of Resources for Management (FIRM).[62]

3.4 | Review of instrument development and validation

In Aim 2, 6 studies were identified as instrument development studies designed to examine the psychometric properties of self-management instruments in children with epilepsy. Of these, 4[47,56,63,64] reported on instruments administered to children and 2[1,14] to parents or caregivers. The focus of most of the instruments (n = 4) was on adherence and self-management skills.[1,14,63,64]

Three instruments assessed knowledge, with 2 focusing on child knowledge[56,63] and 1 focusing on caregiver knowledge.[14] Two instruments measured child social support, with 1 focusing on the parent providing support to the child[1] and 1 measuring child need for support.[56] One scale each focused on child mental health,[56] on child self-efficacy for managing their seizures,[48] and on family.[1]

In general, instruments passed minimal psychometric property standards (see Table 2). All authors provided a rationale for the scientific need for the instrument and evidence of reliability and validity. Factor analysis and information on quantitative item analysis were reported for 3 instruments.[1,14,48] Reliability was most often evaluated via internal consistency, and evidence of construct and criterion-related validity was reported on all 6 instruments. Only 1 instrument provided empirical support for clinical utility.[14] In contrast, no authors provided information on reading level or cost requirements, and none of the instruments have been normed.

A review of citations using Google Scholar indicated that all except 1 instrument[64] have been cited in the literature, and at least 2[48,56] have been translated into other languages. Citations were as follows: Seizure Self-Efficacy Scale for Children (n = 38)[48]; Psychosocial Care Needs of Children (n = 34)[56]; Denye's Self-Care Agency for Adolescents (n = 25)[63]; Pediatric Epilepsy Medication Self-Management (n = 22)[14]; and Parent Response to Child Illness Scale (n = 15).[1] In conclusion, these 6 well-developed instruments have potential for use in self-management research for children.

4 | DISCUSSION

This systematic review provides an overview of measurement of self-management in pediatric epilepsy. The critique of existing self-management measures occurred through examining predictors and outcomes, further categorizing factors of self-management into modifiable influences and processes. Such a synthesis of the literature can inform the development of intervention targets for self-management behavior modification. In general, there is a relatively small number of studies that have investigated these complex clinical issues. Furthermore, there is variability in the conceptualization of and methodologies utilized to investigate pediatric epilepsy self-management. This is not surprising given that even the well-recognized self-management models, such as the pediatric model nonspecific to epilepsy[16] and adult epilepsy specific model,[18,65] may include the same factors (eg, information and support seeking) but conceptualize their relationships to self-management behaviors differently.

Per Aim 1, individual and family-focused factors were the most commonly studied predictors of self-management, with psychosocial care needs and self-efficacy for seizure management identified as the key factors associated with pediatric epilepsy self-management. Few study designs have included mediator and moderator analyses, and this finding suggests that simplistic research designs have predominated, with a resulting focus on direct relationships between predictors and outcomes. In terms of outcome measures research thus far has mostly emphasized adherence, demonstrating a very narrow focus for self-management outcomes.

Because of the early state of the literature in the area, there has not been a dominant theoretical model to guide the design of self-management research in pediatric epilepsy. As a result, there has been considerable variation in predictors and outcomes investigated, as well as in study designs and measurement of variables. Even when a few studies investigated the same variable, that same variable may have been treated as a predictor in one study and an outcome in another. For example, attitudes toward epilepsy was treated as a predictor by Austin et al.,[22] and an outcome by Wagner et al.[23] The same is true regarding outcome variables, with varying measurement approaches (eg, electronic monitors vs self-report) and conceptualizations. As a result, most findings have been supported by very few studies and often by only one research group.

In a second step of Aim 1, the analysis of self-management factors occurred across 3 areas (ie, influences, processes and self-management behaviors) in 4 domains (ie, individual, family, community, and health care). Results are notable for 2 patterns: First, there has been a predominant focus on pediatric epilepsy influences and processes that are modifiable in nature. Second, there is a strong focus on individual child and family influences and processes, potentially at the expense of evidence for the role of community and health systems in pediatric epilepsy self-management. Influences are those aspects that promote or detract from self-management behaviors[16]; therefore, the modifiable child, family, and environmental influences (eg, self-efficacy, family mastery and communication, and attitudes towards epilepsy),[24,35,38] which had a demonstrated association to pediatric epilepsy self-management

behaviors, should be considered targets for self-management intervention.

As mentioned previously, the manner in which variables are conceptualized in studies influences our understanding about them and their relationship to self-management; however, variability among study conceptualization of self-management factors is not an inherent problem in study designs, but is instead reflective of the complexity of self-management. Furthermore, a domination of medication adherence over other individual and family self-management behaviors indicates a need to adopt a broader understanding of self-management behaviors to include coping, sleep, quality of life, and stress and mood management, etc., which are identified in more contemporary models of self-management.[16,66] As an illustration, the efficacy of an intervention targeting coping skill enhancement should be measured via coping responses as the primary outcome, suggesting a need for an epilepsy-specific measure of self-management behaviors that includes cognitive and behavioral coping responses. Indeed, behavioral assessment has not been attentive to epilepsy-specific self-management but instead, employed general chronic disease self-management and coping and/or generic instruments of parenting, general care, and family mastery (Table 1). These results indicate a need and priority for common data elements for pediatric epilepsy specific self-management behaviors (eg, Centers for Disease Control and Prevention–Managing Epilepsy Well Network (MEW) data-based rationale).[67]

With regards to Aim 2, the 6 instruments evaluated had a strong scientific rationale and psychometric properties. Although these measures cover several specific clinical aspects of pediatric self-management (ie. adherence), development of measures that more thoroughly address the totality of pediatric self-management is necessary. Furthermore, these instruments have not been widely applied in diverse settings, comprehensive in their assessment of epilepsy self-management, or subject to more complex statistical instrument development analysis compared to the established adult instruments.[18,65]

4.1 | Limitations to the current review

The current study has limitations. Each reviewer (n = 5) was assigned a number of articles to review for pediatric epilepsy self-management factors, and 1 additional reviewer evaluated the development of self-management instruments. Bias could have been introduced; however, the workgroup reviewed methods on multiple phone conference calls to abstract the findings. When reviewers were uncertain of how to categorize a study, the workgroup discussed the findings and made a group decision. 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. It is also possible that very recent publications, or those not using our defined key terms but the same constructs, may have been omitted from this review.

5 | CONCLUSION

In conclusion, these findings of a review of 25 self-management studies in pediatric epilepsy highlight several key cognitive and behavioral targets for skills development in self-management intervention: adherence, self-efficacy for seizure management, attitudes toward epilepsy, and family variables (family mastery, communication). Results also identify several gaps in the literature: a paucity of well-validated measures of self-management behaviors, a lack of mediator/moderator designs to examine the complex relationships between predictors and pediatric epilepsy self-management outcomes, and few studies examining the community and health care domains of self-management. Prior to the development of future evidence-based interventions, a pediatric epilepsy self-management definition, research about community and health care domains, and development of pediatric self-management instruments to measure self-management behavior change are necessary.

5.1 | Future directions

A strong and well-defined conceptual model of pediatric epilepsy self-management would provide many research opportunities and precise targets for intervention. For example, attention to other domains such as family (beyond the child-parent dyad), health care, and community systems would develop knowledge and effect comprehensive self-management. Indeed, community and health care variables play a role in intervention success[68] and identification of desired self-management results beyond medication adherence are paramount, given what is known about the relationship between psychosocial variables and seizure effects. Considering desired outcomes beyond treatment adherence to include improved problem-solving efficacy, coping, quality of life, school and social adjustment, patient/self-advocacy, and effective clinic visits has the potential to change the role and function of pediatric self-management interventions.

Focus group and survey methodologies have proven to be valid and informative in advancing the state of adult epilepsy self-management, and promote identification of issues from a patient-centered standpoint, which can differ from the clinician perspective.[69–72] These methodologies highlight the difference between top-down ans bottom-up approaches to intervention development, with the latter resulting in excellent participant retention

in adult intervention trials.[8] In addition, studies that focus on the day-to-day problem solving and information needs of the patient and family, along with the modifiable influences and processes (eg, coping) will lead to more complex designs that include mediating and moderating variables, longitudinal design, and the development of specific intervention components. Multivariate studies can facilitate identification of key variables that both influence and determine outcomes. For example, use of structured equation modeling allows for better understanding of measured variables, including latent variables, by revealing the direction and influence of hypothesized relationship.

Finally, the knowledge gained of the relationships between such variables and self-management behaviors will contribute to the development of instruments that measure self-management behaviors specific to pediatric epilepsy and take into consideration the roles or contributions of family and caregivers and their relevance (ie, developmental transition to independent care), the community, and health care systems to pediatric epilepsy management. Such instruments should be validated using more complex analyses such as Item Response Theory and Rasch Analysis, which may inform the development of item banks from which specific items or domains of interest could be selected for research or health care services.

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.

ACKNOWLEDGMENTS

AUTHOR CONTRIBUTIONS

Gigi Smith and Janelle L Wagner conceptualized and designed the study, were involved in the organization and writing of all sections of the manuscript, and approved the final manuscript as submitted. Avani C. Modi and Erica Johnson conceptualized and designed the study, were involved in the organization and writing of the results section and writing of the discussion section of the manuscript, and approved the final manuscript as submitted. Ross Shegog conceptualized and designed the study, was involved in the organization and writing of the results and discussion sections of the manuscript, and approved the final manuscript as submitted. Joan K. Austin conceptualized and designed the study, was involved in the writing of the results and discussion sections of the manuscript, and approved the final manuscript as submitted.

DISCLOSURE

None of the authors report 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.

REFERENCES

1. Austin JK, Shore CP, Dunn DW, et al. Development of the parent response to child illness (PRCI) scale. Epilepsy Behav. 2008;13:662–9.
2. Ferro MA, Avery L, Fayed N, et al. Child- and parent-reported quality of life trajectories in children with epilepsy: a prospective cohort study. Epilepsia. 2017;58:1277–86.
3. Guilfoyle SM, Monahan S, Wesolowski C, et al. Depression screening in pediatric epilepsy: evidence for the benefit of behavioral medicine service in early detection. Epilepsy Behav. 2015;44:5–10.
4. IOM (Institute of Medicine). Epilepsy Across the Spectrum: Promoting Health and Understanding. Washington, DC: The National Academies Press; 2012.
5. Bradley PM, Lindsay B, Fleeman N. Care delivery and self-management strategies for adults with epilepsy. Cochrane Database Syst Rev. 2016;2:CD006244.
6. Buelow JM, Johnson J. Self-management of epilepsy: a review of the concept and its outcomes. Dis Manag Health Out. 2000;8:327–36.
7. Edward K, Cook M, Giandinoto J. An integrative review of the benefits of self-management interventions for adults with epilepsy. Epilepsy Behav. 2015;45:195–204.
8. Fraser R, Johnson EK, Lashley S, et al. PACES in epilepsy: results of a self-management randomized controlled trial. Epilepsia. 2015;56:1264–74.
9. Lewis S, Noyes J, Hastings RP. Systematic review of epilepsy self-management interventions integrated with a synthesis of children and young peoples views and experiences. J Adv Nurs. 2014;73:478–97.
10. Lindsay B, Bradley PM. Care delivery and self-management strategies for children with epilepsy. Cochrane Database Syst Rev 2010;8:CD006245.
11. Tang V, Michaelis R, Kwan P. Psychobehavioral therapy for epilepsy. Epilepsy Behav. 2014;32:147–55.
12. Gross R, Johnston K. Levels of evidence: taking Neurology to the next level. Neurology. 2009;72:8–10.

13. Wagner JL, Modi AC, Johnson EK, et al. Self-management interventions in pediatric epilepsy: what is the level of evidence? Epilepsia. 2017;58:743–54.

14. Modi AC, Monahan S, Daniels D, et al. Development and validation of the pediatric epilepsy medication self-management questionnaire. Epilepsy Behav. 2010;18:94–9.

15. Zelikovsky N, Schast AP. Eliciting accurate reports of adherence in a clinical interview: development of the Medical Adherence Measure. Pediatr Nurs. 2008;34:141–6.

16. Modi AC, Pai AL, Hommel KA, et al. Pediatric self-management: a framework for research, practice, and policy. Pediatrics. 2012;129:e473–86.

17. Holmbeck GN, Devine KA. Editorial: an author's checklist for measure development and validation manuscripts. J Pediatr Psychol. 2009;34:691–6.

18. DiIorio CK, Bamps YA, Edwards AL, et al. The prevention research centers' managing epilepsy well network. Epilepsy Behav. 2010;19:218–24.

19. Managing Epilepsy Well. Managing Epilepsy Well. [cited 2017 Sept1]. Available from http://www.managingepilepsywell. org/

20. Liberati A, Altman DG, Tetzlaff J, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration. BMJ. 2009;339:b2700.

21. United Nations Development Programme (UNDP). Human Development Report 2015. New York, NY: UNDP; 2015.

22. Austin JK. Predicting parental anticonvulsant medication compliance using the theory of reasoned action. J Pediatr Nurs. 1989;4:88–95.

23. Wagner JL, Smith G, Ferguson PL, Horton S, Wilson E. A hopelessness model of depressive symptoms in youth with epilepsy. J Pediatr Psychol. 2009;34:89–96.

24. Austin JK, Dunn DW, Perkins SM, Shen J. Youth with epilepsy: development of a model of children's attitudes toward their condition. Child Health Care. 2006;35:123–40.

25. Modi AC, Rausch JR, Glauser TA. Early pediatric antiepileptic drug nonadherence is related to lower long term seizure freedom. Neurology. 2014;82:671–3.

26. Modi A, Wu Y, Rausch J, Peugh JL, Glauser TA. Antiepileptic drug non-adherence predicts pediatric epilepsy seizure outcomes. Neurology. 2014;83:2085–90.

27. Hazzard A, Hutchinson SJ, Krawiecki N. Factors related to adherence to medication regimens in pediatric seizure patients. J Pediatr Psychol. 1990;15:53–5.

28. Aylward BS, Rausch JR, Modi AC. An examination of 1-year adherence and persistence rates to antiepileptic medication in children with newly diagnosed epilepsy. J Pediatr Psychol. 2015;40:66–74.

29. Modi AC, Rausch JR, Glauser TA. Patterns of non-adherence to antiepileptic drug therapy in children with newly diagnosed epilepsy. J Am Med Assoc. 2011;305:1669–76.

30. Kerr C, Nixon A, Angalakuditi M. The impact of epilepsy on children and adult patients' lives: development of a conceptual model from qualitative literature. Seizure. 2011;20: 764–74.

31. Shagena MM, Sandler HK, Perrin EC. Concepts of illness and perception of control in healthy children and in children with chronic illnesses. J Dev Behav Pediatr. 1988;9:252–6.

32. Modi AC, Ingerski LM, Rausch JR, Glauser TA, Drotar D. White coat adherence over the first year of therapy in pediatric epilepsy. J Pediatr. 2012;161:695–9.

33. Morita DA, Glauser TA, Modi AC. Development and validation of the pediatric epilepsy side effects questionnaire. Neurology. 2012;79:1252–8.

34. Asato MR, Manjunath R, Sheth RD, et al. Adolescent and caregiver experiences with epilepsy. J Child Neurol. 2009;24:562–71.

35. Carbone L, Zebrack B, Plegue M, Joshi S, Shellhaas R. Treatment adherence among adolescents with epilepsy: what really matters? Epilepsy Behav. 2013;27:59–63.

36. Painter E, Rausch JR, Modi AC. Changes in daily activity patterns of caregivers of children with newly diagnosed epilepsy: a case controlled design. Epilepsy Behav. 2014;31:1–6.

37. Kovacs M. The children's depression inventory (CDI) and a structured interview. J Clin Child Adolesc Psychol. 2004;33: 149–57.

38. Wagner JL, Ferguson PL, Smith G. The relationship of coping behaviors to depressive symptoms in youth with epilepsy: an examination of caregiver and youth proxy report. Epilepsy Behav. 2012;24:86–92.

39. Wagner JL, Smith G, Ferguson PL. Self-efficacy for seizure management and youth depressive symptoms: caregiver and youth perspectives. Seizure. 2012;21:334–9.

40. Reynolds C, Kamphaus R. Behavior Assessment System for Children, Second Edition (BASC-2). Manual. Circle Pines, MN: AGS Publishing Inc; 2004.

41. Austin J, Huberty TJ. Development of the child attitude toward illness scale. J Pediatr Psychol. 1993;18:467–80.

42. Shore CP, Buelow JM, Austin JK, Johnson CS. Continuing psychosocial care needs in children with new-onset epilepsy and their parents. J Neurosci Nurs. 2009;41:244–50.

43. Austin JK, Dunn DW, Johnson CS, Perkins SM. Behavioral issues involving children and adolescents with epilepsy and the impact of their families: recent research data. Epilepsy Behav. 2004;5(Suppl 3):S33–41.

44. Bellon M, Pfeiffer W, Maurici V. Choice and control: how involved are people with epilepsy and their families in the management of their epilepsy? Results from an Australian survey in the disability sector. Epilepsy Behav. 2014;37:227–32.

45. Elliott IM, Lach L, Smith ML. I just want to be normal: a qualitative study exploring how children and adolescents view the impact of intractable epilepsy on their quality of life. Epilepsy Behav. 2005;7:664–78.

46. Reeve DK, Lincoln NB. Coping with the challenge of transition in older adolescents with epilepsy. Seizure. 2002;11:33–9.

47. Kirton A, Wirrell E, Zhang J, Hamiwka L. Seizure-alerting and -response behaviors in dogs living with epileptic children. Neurology. 2004;62:2303–5.

48. Caplin D, Austin JK, Dunn DW, et al. Development of a self-efficacy scale for children and adolescents with epilepsy. Child Health Care. 2002;31:295–309.

49. Schwartzer R, Jerusalem M. Generalized self-efficacy scale. In: Weinman J, Wright S, Johnston M, editors. Measures in Health Psychology: A User's Portfolio. Causal and Control Beliefs. Windsor, UK: NFER-NELSON, 1995; p. 35–7.

50. Achenbach T. Manual for the Child Behavior Checklist/4-18 and 1991 Profile. Burlington, VT: University of Vermont Department of Psychiatry; 1991.

51. Watson D, Clark LA, Tellegen A. Development and validation of brief measures of positive and negative affect: the PANAS Scales. J Pers Soc Psychol. 1988;54:1063–70.

52. Kazdin AE, Rodgers A, Colbus D. The hopelessness scale for children: psychometric characteristics and concurrent validity. J Consult Clin Psychol. 1986;54:241–5.

53. Dahl JC. A behavior medicine approach to epilepsy – Time for a paradigm shift? Scand J Behav Therapy. 1999;28:97–114.

54. Buelow JM, McNelis A, Shore CP, Austin JK. Stressors of parents and children with epilepsy and intellectual disability. J Neurosci Nurs. 2006;38:147–54.

55. Nolan KJ, Camfleld CS, Camfield PR. Coping with Dravet syndrome: parental experiences with a catastrophic epilepsy. Dev Med Child Neurol. 2006;48:761–5.

56. Austin JK, Dunn DW, Huster G, Rose D. Development of scales to measure psychosocial care needs of children with seizures and their parents. J Neurosci Nurs. 1998;30:155–60.

57. McNelis A, Musick B, Austin J, Dunn D, Creasy K. Psychosocial care needs of children with new-onset seizures. J Neurosci Nurs. 1998;30:161–5.

58. Spirito A, Stark LJ, Williams C. Development of a brief checklist to assess coping in pediatric patients. J Pediatr Psychol. 1988;13:555–74.

59. Austin JK, Patterson JM, Huberty TJ. Development of the coping health inventory for children. J Pediatr Nurs. 1991;6:166–74.

60. Quittner AL, DiGirolamo AM, Eigen MM. Parental response to cystic fibrosis: a contextual analysis of the diagnosis phase. J Pediatr Psychol. 1992;17:683–704.

61. Quittner AL, Espelage DL, Levers-Landis C, Drotar D. Measuring adherence to medical treatments in childhood chronic illness: considering multiple methods and sources of information. J Clin Psychol Med Settings. 2000;7:41–54.

62. McCubbin HI, Comeau JK, Harkins J, FIRM: Family inventory of resources for management. Family Assessment: Resiliency, Coping and adaptation. Inventories for Research and Practice. Madison, WI: University of Wisconsin Publishers; 1996.

63. Gout DA, Kieckhefer GM. Assessment of self-care agency in chronically ill adolescents. J Adolesc Health Care. 1988;9: 55–60.

64. Ryan JL, Arnett AD, Pai ALH, Modi AC. An examination of the allocation of treatment responsibility scale in adolescents with epilepsy. Epilepsy Behav. 2014;41:1–5.

65. Escoffery C, DiIorio C, Bamps Y, et al. Development of the adult epilepsy self-management instrument. Epilepsy Behav. 2015;50:172–83.

66. Grey M, Schulman-Green D, Knafl K, Reynolds NR. A revised self- and family management framework. Nurs Outlook. 2015;63:162–70.

67. England MJ, Liverman CT, Schultz AM, Strawbridge LM. Summary: a reprint from epilepsy across the spectrum: promoting health and understanding. Epilepsy Curr. 2012;12:245–53.

68. Wu YP, Pai AL. Health care provider-delivered adherence promotion interventions: a meta-analysis. Pediatrics. 2014;133: e1698–707.

69. Fraser R, Johnson E, Miller J, et al. Managing epilepsy well: self-management needs assessment. Epilepsy Behav. 2011;20:291–8.

70. Johnson EK, Fraser R, Miller JW, et al. A comparison of epilepsy self-management needs: provider and patient perspectives. Epilepsy Behav. 2012;25:150–5.

71. McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure. 2004;13:15–31.

72. Wagner JL, Smith G, Ferguson PL, van Bakergem K, Hrisko S. Pilot study of an integrated cognitive-behavioral and self-management intervention for youth with epilepsy and caregivers: coping openly and personally with epilepsy (COPE). Epilepsy Behav. 2010;18:280–5.