Adherence and Self-Management in Pediatric Populations

Edited by Avani C. Modi and Kimberly A. Driscoll

ADHERENCE AND SELF-MANAGEMENT IN PEDIATRIC POPULATIONS

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Preface

PubMed indexes more than 40,000 articles on adherence. Several texts are also dedicated to pediatric adherence including *Adherence to Pediatric Medical Regimens* (Rapoff, 1999, 2009) and *Promoting Adherence to Medical Treatment in Chronic Childhood Illness: Concepts, Methods, and Interventions* (Drotar, 2000). This book, *Adherence and Self-Management in Pediatric Populations,* is a much needed update on the state of the adherence field with chapters dedicated to specific diseases, many of which have not been written about in other texts (e.g., sleep, obesity, dermatological diseases). The introduction provides a review of adherence and self-management theories, measurement approaches, and intervention strategies used across pediatric chronic conditions. The final chapter focuses on emerging areas in the field that provide guidance and recommendations for future directions in this important field. Adherence experts in their respective fields wrote the chapters included in this text.

This book is unique because disease-specific chapters include the following: (1) basic background about the disease—definition, etiology, diagnosis, prevalence; (2) guiding adherence theories; (3) evidence-based assessment of adherence; (4) evidence-based interventions and promising interventions to improve adherence; (5) case study; and (6) emerging areas and conclusions. Clinicians, researchers, instructors, and trainees of all levels and from many fields including medicine, psychology, social work, and nursing will find this to be a beneficial introductory text summarizing the state of adherence in each disease, which we hope will stimulate highly significant and innovative research and clinical care.

Fifty years ago, R. Brian Haynes defined the term medical compliance as "the extent to which the patient's behavior (in terms of taking medications, following diets, or executing other lifestyle changes) coincides with medical or health advice." Since that time, much debate has taken place about the appropriate term that should accompany this definition. Compliance eventually fell out of favor as it implies a "paternalistic attitude toward the patient on the prescriber's part" (Aronson, 2007). Concordance followed and also fell out of favor as an acceptable term because it is a synonym for compliance (Aronson, 2007). For the past 20 years, the term adherence has dominated the extant literature, but the debate about its appropriateness has recently surfaced with arguments that it also focuses too much—like the other terms—on what someone

else wants and what an individual is doing wrong (Dickinson et al., 2017). Thus, Dickinson and colleagues suggest replacement language such as engagement, participation, involvement, and medication taking. We do not intend to engage in a debate about the accuracy or acceptability of the "most appropriate term" but rather acknowledge that this debate will likely continue for decades to come. With this in mind, we have elected to continue to use adherence and self-management given they are accepted terms in the field of pediatric psychology.

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CHAPTER

1

Introduction

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Adherence to medical treatment regimens has been a topic of research and clinical interest in the field of pediatric psychology since its inception. Virtually all children with chronic conditions are asked to engage in treatment or self-management tasks. These treatment tasks include taking oral medications, performing various therapies (e.g., nebulizer, physical and occupation therapy, airway clearance), following dietary and physical activity recommendations (e.g., increased fluid intake, healthy food choices, increased physical activity), and avoidance of triggers of disease symptoms (e.g., avoid asthma or seizure triggers, avoidance of particular foods). Adhering to these treatment regimens on a daily basis is difficult for adults, much less children who simply want to do what their peers and siblings are doing. Thus, it is not surprising that adherence rates average 50% and that these rates have not changed in several decades (Rapoff, 2010). Unfortunately, the consequences of nonadherence are often dire for youth with chronic conditions. Treatment nonadherence leads to greater morbidity and mortality, resulting in higher healthcare utilization and healthcare charges and costs (Hommel et al., 2017). For example, youth with leukemia with suboptimal adherence are 2.5 times more likely to relapse than those with optimal adherence (Bhatia et al., 2012). Similarly, youth with epilepsy are 3.24 times more likely to have seizures when they are nonadherent versus adherent (Modi, Rausch, & Glauser, 2014; Modi, Wu, Rausch, Peugh, & Glauser, 2014). In addition to these deleterious physical health outcomes, treatment nonadherence results in greater family distress and lower quality of life, which may lead to unnecessary changes in the treatment regimen (e.g., dose escalation or discontinuation

of medication) in clinical practice (Goodhand et al., 2013; Modi, Wu, Guilfoyle, & Glauser, 2012). These data highlight the tremendous impact of nonadherence across individual, family, and healthcare system factors.

The pediatric psychology field continues to grow, as evidenced by a cursory search on PubMed of the search terms "treatment adherence" and "pediatric," with 119 publications in 1998, 322 publications in 2008, and 957 in 2018. With advances in adherence and self-management research and clinical practice, the ultimate goal is to continue to improve the physical health and psychosocial outcomes of youth with chronic conditions and their families. The state of the field of pediatric adherence and self-management is described in this book with individual chapters focusing on specific chronic conditions. The introduction chapter serves as a primer to subsequent chapters to understand the field of pediatric adherence science, including the terminology and taxonomy of adherence, general measurement approaches, application of theories, and examination of adherence interventions.

Adherence definitions and terminology

As mentioned in the preface of this book, we do not intend to debate whether terminology is appropriate or not; we retained the use of the word adherence as it is currently commonly accepted. It is well recognized that no matter the topic or field, terminology frequently goes through a metamorphosis—in that decades ago, a word was appropriate given the culture and context of that time, but as time passes and new education, insight, and reflection are gained, a word or words become inappropriate. Such was the case with compliance, which was introduced in the 1970s by Sackett (Sackett & Snow, 1979), but fell out of favor in the 1990s because compliance was interpreted as the need for an individual (previously a patient as the term patient is currently falling out of favor) to obey their medical provider's treatment recommendations, which was perceived as paternalistic (Vrijens et al., 2012). Although adherence has been the preferred term for more than two decades, health providers still use the term compliance in conversations and the term continues to be used in many recently published studies. The European Society for Patient Adherence, COMpliance, and Persistence published the ABC taxonomy for medication adherence in 2012 (Vrijens et al., 2012), which divides adherence behaviors into four distinct but interrelated phases: initiation (the start of the first dose of a prescribed medicine), implementation (e.g., how an individual's actual dosing of a medication matches what is prescribed), discontinuation (e.g., the point at which the last dose is taken and no subsequent doses are taken), and persistence (e.g., length between initiation and the last dose taken). Use of these terms

is rarely seen in pediatric adherence studies, but use of systematic nomenclature could benefit the operationalization and consistency of reporting adherence and self-management research. Importantly, it is unclear how individuals with chronic conditions and their families truly perceive these terms and whether the actual words of "compliance," "adherence," or "persistence" truly matter to them. As the chapters in this book highlight, behaviors related to medical treatment are challenging—regardless of which terms or words are used.

Individuals in the field of diabetes recently contested the use of the word adherence, and other words, proposing another shift in how we think about how we communicate about and with individuals with diabetes and even individuals with other chronic conditions. Dickinson and colleagues note that, "For decades, a substantial amount of the language around diabetes has been focused on negative outcomes and laden with judgment and blame, and it has not adequately considered individual needs, beliefs, and choices" (Dickinson et al., 2017). The authors make an excellent point—ask any psychologist who is about to see or has seen a child or adolescent with type 1 diabetes about the judgment they feel about their A1C result (i.e., a numerical representation of the individual's average blood glucose in the previous 2–3 months). Clinical observations suggest that many youth are nervous before appointments or upset and deflated afterward because of the way they were made to feel about "the numbers" that define their health, in this case A1C benchmarks made by the American Diabetes Association. Relatedly, many providers, caregivers, and youth are relieved and overjoyed when the A1C goal is attained. Indeed, that A1C number or other levels or numbers (e.g., FEV1% predicted, renal or liver lab values) place a burden and weight on the shoulders of many individuals with chronic conditions. Similarly, providers are also evaluated based on how the children and adolescents they care for are doing—or at least the clinics in which they work (e.g., U.S. News and World Report rankings).

While it remains unclear what terminology is optimal for use when engaging in conversations about medical treatments, Dickinson and colleagues made the following recommendations about how to communicate about and with individuals with chronic illness (Dickinson et al., 2017). Use language that is (1) neutral, nonjudgmental, and based on facts, actions, or physiology/biology; (2) free from stigma; (3) strengths based, respectful, inclusive, and imparts hope; (4) collaborative between individuals and providers; and (5) person centered. Not only is choice of word(s) important, but tone and expressions of empathy and compassion are equally, if not, more important. Moreover, the majority of adherence research and clinical work focuses on individuals with chronic illness and their families and often neglects important provider level factors, including patient-provider communication and provider adherence to medical guidelines. More research is needed in these areas as adherence

and self-management really is a partnership between individuals with chronic illness and their medical teams.

Methods for assessing adherence and self-management behaviors

There are many subjective and objective methods for assessing adherence and self-management behaviors. Despite the wealth of data available from objective methods, as evidenced in many chapters of this book, self-report remains the most common adherence and self-management assessment method regardless of whether the setting is research or clinical (Plevinsky et al., in press; Quittner, Modi, Lemanek, Ievers-Landis, & Rapoff, 2008). However, objective methods are almost exclusively used in research settings and are only recently used in clinical settings (Herzer, Ramey, Rohan, & Cortina, 2011; Hilliard, Ramey, Rohan, Drotar, & Cortina, 2011).

Subjective methods

Patient-reported outcomes

A recent review of patient-reported outcomes for adherence and self-management behaviors in pediatrics identified 50 validated measures within the field (Plevinsky et al., in press). Assessing adherence and self-management behaviors through patient-reported outcomes (i.e., self-report) is common and offers several advantages including minimal or no cost, ease and quick administration in busy clinical settings (e.g., paper/pencil, online databases such as RedCap), and literacy and cultural adaptation (Stirratt et al., 2015). However, the primary criticisms of these methods include inaccurate recall and social desirability (Stirratt et al., 2015). Indeed, several studies demonstrate that individuals with chronic illness self-report better adherence and higher engagement in self-management tasks compared to other methods. (Craker, Tarantino, Whiteley, & Brown, 2019; Mazze et al., 1984; Modi, Guilfoyle, Morita, & Glauser, 2011; Williams et al., 1988). For example, individuals with type 1 diabetes recorded higher blood glucose checking frequencies in written logbooks than were actually occurring when they were not aware that blood glucose monitors had recording devices (Mazze et al., 1984; Williams et al., 1988). Similarly, self- and parent-reported adherence of several cystic fibrosis treatments were almost double that of objective measures (Modi et al., 2006).

Diary methods

Written and digital logbooks and diet records are diary methods that may be used to assess adherence and self-management behaviors (Stumbo,

2013; Vuckovic, Ritenbaugh, Taren, & Tobar, 2000); however, they are vulnerable to the same weaknesses as self-report questionnaires—lack of accurate recall (if not completed in real time) and social desirability (Quittner et al., 2008; Yang et al., 2016). In addition, individuals may forget to record self-management behaviors, which artificially deflate reporting accuracy. In an attempt to remedy the inherent challenges with self-report questionnaires, logbooks, and diet records, the 24-hour recall method for assessing adherence and self-management behaviors was developed (Freund, Johnson, Silverstein, & Thomas, 1991; Quittner & Espelage, 1999; Quittner & Opipari, 1994; Reynolds, Johnson, & Silverstein, 1989). Contemporary diaries consist of similar methodology but use more robust approaches, including ecological momentary assessment (we use this term throughout), the experience sampling method, and ambulatory assessment (Heron, Everhart, McHale, & Smyth, 2017; Kahneman, Krueger, Schkade, Schwarz, & Stone, 2004; Stone & Shiffman, 1994). Ecological momentary assessment consists of repeated, and sometimes random, sampling of an individual's characteristics including moods, behaviors, and experiences in natural environments, with the goal of minimizing recall problems or biases. Other advantages of ecological momentary assessment include better ecological validity and the ability to examine data across minutes, hours, days, weeks, months, and years. Data collection is easier than ever and can be collected via apps on smart phones. For the field of adherence and self-management to continue to advance, objective methods that are easy to use, cost-effective, and efficient must be developed for use in both clinical and research settings.

Objective methods

Technology

The type 1 diabetes field leads the way regarding use of technology to objectively measure and record adherence and self-management behaviors, in both research and clinical settings. Continuous glucose monitors and insulin pumps can be uploaded to cloud-based software for analysis and clinical care, and with just a few exceptions so can blood glucose monitors. Several insulin pens now store data, and one (InPen) can also be uploaded to cloud-based software. These devices are reimbursed by insurance providers, which is particularly advantageous when compared with other electronic monitors and devices that are not, including sensors attached to meter dose inhalers and electronic pillboxes and caps. For example, the SmartTouch Ventolin monitor (Adherium, Auckland, New Zealand) is a small sensor positioned under the base of a meter dose inhaler (Patel et al., 2013). Imagine how much more asthma clinicians would know about asthma self-management tasks if a similar sensor was attached to all inhalers distributed by pharmacies and used in daily life. Similarly, several

electronic pill bottles and caps and pillboxes exist, with a recent study evaluating the accuracy and usability of these devices (McGrady et al., 2018). This lab-based evaluation examined 10 devices (e.g., MEMS Caps, eCAP, SimpleMed+, WisePill, iCAP) and found accuracy ranged from 64% to 100% with capabilities of the devices varying widely (e.g., push notification, alarms, lights, capacity). Although these devices record the dates and times of opening and not actual ingestion of the medication, electronic monitors are still considered the optimal measure of adherence behaviors (McGrady et al., 2018; Quittner et al., 2008).

Biomarkers

Biomarkers collected via blood, urine, saliva, and hair samples may provide a direct and accurate way of measuring adherence (Lam & Fresco, 2015). However, biomarkers also have weaknesses. For example, blood serum levels are affected by genetic, demographic, and dosing factors. When serum levels are zero, it can be assumed that the individual is completely nonadherent to their medication for the few days prior to having their blood taken. However, when an individual has subtherapeutic drug serum levels, it is unclear how this information translates to daily adherence behaviors (e.g., did he/she miss one, two, or three doses of medicine?). Blood serum levels are further complicated by pharmacokinetic variability, which involves the rate that a drug is absorbed, distributed, metabolized, and eliminated by the body (Perucca, 2002). Factors, such as demographics (e.g., body weight), pathophysiological (e.g., metabolic function), food intake, and therapeutic features (e.g., comorbid medications), can regularly alter dose-concentration relationships (Lam & Fresco, 2015).

Another adherence biomarker is examination of the standard deviations of consecutive blood levels of medication across time, known as the Medication Level Variability Index, which has been used in pediatric transplant (Shemesh et al., 2017). Although this method cannot be used for all medications across chronic conditions, it is an important adherence measure in pediatric transplant populations and can be used to identify individuals at risk for suboptimal health outcomes (Rich et al., 2018).

Hair and saliva methods are relatively painless compared with blood levels, and there is growing interest in these bioassays. For example, antiepileptic drug levels have been detected in human hair samples (Williams et al., 2002) and saliva concentrations (Herkes & Eadie, 1990). Hair samples have also been successfully used for adherence monitoring in HIV (Saberi et al., 2017). With more research, these noninvasive methods for measuring adherence in clinical practice may be beneficial.

Pharmacy refills and pill counts

Other objective measures of adherence include pharmacy refills (Blais et al., 2014) and pill counts. Pill counts and prescription refills are often used to infer adherence by recording the amount of medicine that remains in a pill bottle or the number of refills that have been obtained since the prior clinic visit. Although these methods are noninvasive, they are plagued by several problems, including (1) the need for individuals to bring their pharmacy bottles back to clinic; (2) they do not account for samples that may have been provided; (3) there is potential for human error/deception (e.g., pill dumping) (Lam & Fresco, 2015); (4) HIPPA makes capturing pharmacy refill data more difficult; and (5) automatic refills completed by pharmacies may overestimate adherence behaviors if medications are directly sent to individuals via mail (Modi & Guilfoyle, 2011). Furthermore, liquid medications, which are often used with young children due to challenges with swallowing pills, are difficult to quantify from a "pill count" perspective. Finally, pill counts and prescription refills do not provide the level of detail necessary to ascertain dosing-specific adherence, which is necessary for targeted intervention. However, it is notable that many electronic medical records are now integrating pharmacy refill information directly into their reporting, which may make this a potential area to explore in the future.

Theories and models of adherence and self-management

The field of psychology is founded on theory because it provides a systematic way to understand behaviors. Theory is a set of ideas and interrelated concepts used to explain phenomena, which guide our understanding of adherence and self-management behaviors in children and adolescents. Theory allows researchers to generate testable hypotheses about the constructs that may influence adherence behaviors. In other words, they provide a guide and framework to understand factors that predict, moderate, and mediate adherence and self-management behaviors. Many theories in the field of psychology and health have been used to examine early work in the area of adherence and self-management, including the Health Belief Model, Transtheoretical Model of Change, Social Cognitive Theory, and Theory of Planned Behavior/Reasoned Action. Newer and more contemporary theories and models have since been proposed and are increasingly used to guide adherence and self-management research, including the Pediatric Self-Management Model, the Self- and Family Management Model, and the Individual and Family Self-management Theory.

Health Belief Model

In 1975, Becker and Maiman applied the Health Belief Model (Rosenstock, 1974) to the examination of adherence behaviors for the first time in an effort to identify predictors of adherence behaviors (Becker & Maiman, 1975). The Health Belief Model has four core components: (1) perceived susceptibility; (2) perceived severity; (3) perceived benefits; and (4) perceived barriers. Perceived susceptibility is an individual's assessment of risk related to developing a health issue/illness. Perceived severity is an individual's assessment of the severity of their illness and its potential consequences. Perceived benefits are the values that an individual places on engaging in a particular health-related behavior. Finally, perceived barriers are the things that interfere with engaging in health-related behaviors. The Health Belief Model also purports that a cue action or trigger is needed for individuals to engage in health-promoting behaviors. The Health Belief Model has been used extensively to examine a multitude of health behaviors, including smoking cessation, physical activity, seatbelt use, and adherence. One meta-analysis examining the Health Belief Model across several health behaviors found strong support for components of the model. Specifically, strongest support was found for perceived benefits and perceived barriers, weak support for perceived severity, and no support for perceive susceptibility (Carpenter, 2010). Furthermore, the meta-analysis indicated a moderator effect for adherence compared with other health behaviors, suggesting the need to further examine Health Belief Model constructs in the context of medication adherence. Notably, the studies included in this meta-analysis did not include pediatric samples; however, various components of the Health Belief Model in pediatric adherence studies have been tested, many of which are cited in subsequent chapters. One recent study found a significant relationship between the Health Belief Model constructs of barriers and cues to action for overall adherence in youth with cystic fibrosis (Dempster, Wildman, Masterson, & Omlor, 2018).

In an effort to expand the Health Belief Model to children's health behaviors, Bush and Iannotti (1990) developed the Children's Health Belief Model. This model recognized that child adherence behaviors occur within the context of families. It incorporates the influences and beliefs of caregivers, as well as the cognitive and emotional factors that can change as children develop and expected health behaviors, such as medication adherence, as an outcome (Bush & Iannotti, 1990). However, few studies have applied this model in pediatric adherence and self-management. Overall, compared with other theories, components of the Health Belief Model have the largest evidence base supporting its

application for pediatric adherence research; however, the full Health Belief Model is not well supported.

Transtheoretical Model of Change

The transtheoretical, or stages of change, model was developed by Prochaska and DiClemente in 1983. This model has six possible stages that represent a process of behavior change (Prochaska & Diclemente, 1983). These stages are not meant to be linear but instead can be circular or nondirectional in nature. The six stages are precontemplation, contemplation, determination, action, maintenance, and relapse. Examples of these phases as they apply to pediatric adherence behaviors are described next.

Precontemplation is a phase in which the individual with a chronic illness/family does not recognize any problems with their adherence behaviors, but the healthcare provider or other family members may recognize issues with adherence. Contemplation is the stage when the individual with a chronic illness/family begins to weigh the pros and cons of changing their adherence behaviors, but no changes in behavior have occurred yet. The determination or preparation phase is when the individual/family decide they are going to make a change or adopt ways to improve their adherence behaviors. The action phase is the act of putting the decision to change into motion; the individual/family make an initial change in their adherence behaviors. This phase typically lasts 6 months. After 6 months of executing the behaviors, the individual/family move into the maintenance phase, which signifies that they are actively maintaining changes in adherence behaviors. As expected, relapse occurs when the individual with a chronic illness/family return to previous behaviors, such as nonadherence. A recently published randomized controlled clinical trial in pediatric asthma provided caregivers with education and support for their child's asthma care based on the caregiver's readiness to change (Garbutt, Highstein, Yan, & Strunk, 2012). Results indicated increased outpatient asthma monitoring visits but no changes in emergency room visits, suggesting the intervention was only partially efficacious (Nelson et al., 2012).

Stages of change theories have also been used to develop motivational interviewing interventions for pediatric adherence. Motivational interviewing is defined as a collaborative style between an individual with a chronic illness or caregivers and providers to elicit intrinsic motivation and strengthen the desire to change behaviors (Miller & Rollnick, 2012a, 2012b). Motivational interviewing strategies have been tested across several pediatric conditions (e.g., HIV, asthma, diabetes) to improve adherence behaviors with mixed results. For example, Powell and colleagues concluded

that motivational interviewing interventions for pediatric type 1 diabetes adherence yield greater effects compared with other pediatric conditions, with more support when parents are involved versus the intervention focusing on the child/teen alone (Powell, Hilliard, & Anderson, 2014). Although the evidence for motivational interviewing-focused interventions in pediatric conditions is growing, more research examining for whom motivational interviewing is most effective is necessary.

Social Cognitive Theory

Social Cognitive Theory, proposed by Bandura, focuses on an individual's learning through dynamic, reciprocal, and continuous interactions between the environment and themselves (Bandura, 1998). Essentially, behaviors are learned, and two cognitive processes are involved that influence adherence behaviors: self-efficacy and outcome expectation. Self-efficacy is an individual's belief in their ability to master a particular task or skill, whereas outcome expectancies are beliefs that a certain behavior will result in a particular outcome, which can be negative or positive. Aspects of Social Cognitive Theory have been examined extensively in the pediatric adherence literature, with many studies assessing self-efficacy. For example, in the context of pediatric obesity, a 2016 review of 16 studies using a Social Cognitive Theory framework found that higher self-efficacy was related to increased fruit and vegetable intake and lower fat, sugar, and sodium intake in youth who were obese (Rolling & Hong, 2016). Furthermore, cognitive and environmental factors also played a key role in adherence to dietary behaviors. This review highlights the application of Social Cognitive Theory to adherence in a specific pediatric population, but this theory can also be tested and applied to other chronic conditions, in which dietary adherence may be critical (e.g., inflammatory bowel disease, cystic fibrosis, food allergies).

Theory of Planned Behavior and Theory of Reasoned Action

The Theory of Planned Behavior and Theory of Reasoned Action (Fishbein & Ajzen, 1975) both stipulate that an individual's behavior is determined by their intention to engage in that behavior. The original theory of Reasoned Action included four constructs: beliefs, attitudes, intentions, and behaviors. Building upon this theory, the Theory of Planned Behavior, added one additional construct of perceived behavioral control, which is defined as the perceived ability to control one's specific behaviors. The construct of intention, which is the most salient aspect of the models, is shaped by three primary elements: attitude (e.g., the way an individual feels about something or their opinion regarding a behavior), subjective norms (e.g., belief that others of importance to the

individual approve or disapprove of their behaviors), and self-efficacy (e.g., the confidence one has that a behavior can be performed or completed). In the context of pediatric adherence, Grossoehme et al. (2016) examined the Theory of Reasoned Action for adherence in cystic fibrosis. Engaged spirituality was related to greater perceive utility of airway clearance (one primary treatment for cystic fibrosis) and more supportive norms for doing the treatment from close friends. This in turn predicted stronger intentions to perform airway clearance treatment, which predicted greater actual adherence to airway clearance. This study is an exemplar for examining the Theory of Reasoned Action/Planned Behavior in pediatric adherence.

Contemporary theories and models

The theories and models noted previously were developed decades ago and provided an important framework for early efforts in adherence research. Although application of these models in pediatric adherence has been studied extensively, no one model has proven better than the rest. This is likely due to evidence supporting particular aspects of each model but not the model fully, as well as the overlap of certain constructs across models. Furthermore, outside of the Children's Health Belief Model, none of these models were developed specifically for pediatrics or within the context of pediatric adherence and self-management. More contemporary models have been developed in the past 10 years based on the contexts in which children live, including the family unit, community, and healthcare system. These models were developed with a specific focus on pediatric adherence and self-management and are described next.

Self- and Family Management Model

Grey, Knafl, and McCorkle (2006) recognized the important role of risk and protective factors that influence self- and family management across chronic diseases. Their conceptual model was built from several core features. First, individual self-management behaviors for children occur in the context of their families, communities, and broader environments (Grey et al., 2006). Second, self-management is a dynamic and multifaceted process with risk and protective factors that promote self- and family management. Third, both the family and individual context are important, and interventions must address risk and protective factors within both contexts. This model identified several risk and protective factors in the following areas: condition (e.g., severity, regimen, genetics), individual (e.g., age, sex), psychosocial (e.g., psychological comorbidities, self-efficacy, diversity), family (e.g., socioeconomic status, structure, functioning), and environment (e.g., social networks, community). Research across these areas has shown that these factors impact key

outcomes, including morbidity, mortality, quality of life, adherence behaviors, healthcare access and utilization, and family lifestyle (Grey et al., 2006). Their model provides an initial approach to understanding self- and family management across a variety of pediatric conditions, with the goal of delineating targets for intervention, which has been updated and revised in recent years (Grey, Schulman-Green, Knafl, & Reynolds, 2015). In the revised model, four key components exist, including facilitators and barriers (previously risk and protective factors), processes, proximal outcomes, and distal outcomes. The revised model also incorporates aspects of the Individual and Family Self-Management Theory (Ryan & Sawin, 2009) described next. Both of these models have been developed and used in the field of nursing.

Individual and Family Self-Management Theory

Based on an extensive review of the pediatric self-management literature and lack of consensus on the theoretical constructs of interest necessary to improve self-management, the Individual and Family Self-management Theory was published in 2009 (Ryan & Sawin, 2009). Their theory focuses on defining concepts of self-management and focusing on the child/teen, dyads within the family, and the family unit as a whole when examining pediatric self-management. Specifically, Ryan and Sawin (2009) identify risk and protective factors that provide a context for self-management focused in several areas, including condition (e.g., condition trajectory, treatment complexity), physical and social environment (e.g., access to care, culture, transportation), and individual and family (e.g., developmental stages, capabilities). These context-specific factors influence self-management processes, such as knowledge and beliefs, self-regulation skills and abilities, and social facilitators. The process then influences both proximal and distal outcomes. Proximal outcomes are condition-specific self-management behaviors, such as blood glucose monitoring, airway clearance therapy, or oral medication taking, or the cost of healthcare services. Distal outcomes include the child's health status, quality of life, and the cost of health itself. As noted previously, aspects of this model have been integrated into a revised model (Grey et al., 2015). Application of these models outside of the nursing field was difficult to find, but these key concepts shed light on key areas of adherence interventions, as well as moderators and mediators of treatment adherence and self-management.

Pediatric Self-Management Model

The Pediatric Self-Management Model is a comprehensive conceptual model that focuses on individual, family, community, and healthcare system processes that lead to adherence behaviors, which in turn impact health outcomes in youth with chronic conditions (Modi, Pai, et al., 2012). The

individual, family, community, and healthcare self-management behaviors each have modifiable and nonmodifiable aspects that influence cognitive, emotional, and social processes. An example of a family-level modifiable factor is family conflict, whereas an example of a nonmodifiable factor is family structure. Processes involved for the family influences may include how treatment is allocated between caregivers and adolescents, and the adherence behavior realized by these processes may be taking medications. The Pediatric Self-Management Model has guided understanding of the factors that play key roles in adherence and health outcomes in pediatric chronic conditions. Kichler and colleagues applied the Pediatric Self-Management Model to type 1 diabetes to review the existing literature and examine how self-management processes at the individual, family, community, and healthcare system level influence a multitude of type 1 diabetes adherence behaviors (Kichler, Moss, & Kaugars, 2012). Similar applications of the Pediatric Self-Management Model are found in Chapter 9 in Epilepsy or in the larger literature (cancer (Hullmann, Brumley, & Schwartz, 2015); sickle-cell disease (Loiselle et al., 2016); asthma (Fedele et al., 2018)). Together, these studies highlight the need for adherence and self-management interventions to address modifiable factors and to use nonmodifiable factors to identify high-risk populations. Recognition that involvement across all systems in which the child lives (e.g., family, school, peers, hospital/clinics) is integral to improving health outcomes.

Contemporary models of self-management and adherence provide a framework to guide clinicians and researchers in identifying subgroups at highest risk for suboptimal health outcomes, targeting modifiable factors in adherence and self-management interventions, and recognizing that interventions will likely need to occur across multiple systems to be effective. Unlike prior theories, these contemporary models are not meant to be proven but instead are to be used to steer the field of pediatric adherence and self-management.

Adherence interventions

A plethora of interventions have been developed and tested to address adherence difficulties in youth across chronic conditions. In general, adherence interventions comprise a wide variety of strategies, including psychoeducation, cognitive, behavioral, or a combination of these (Graves, Roberts, Rapoff, & Boyer, 2010; Kahana, Drotar, & Frazier, 2008). Since the advent of new technologies and digital health platforms, adherence interventions are increasingly delivered using these technology-based approaches. In this section, we discuss adherence intervention strategies and the empirical evidence for their efficacy across pediatric conditions.

Educational strategies were some of the first adherence promotion efforts in the field. The premise behind education is to ensure that children and their families have factual information regarding the nature of the illness, the treatment regimen and how it should be followed, and possible side effects of treatment. Education on the prescribed regimen, including anticipatory guidance about treatments, is critical because lack of adequate knowledge can potentially compromise adherence. For example, some medications taken on an empty stomach can lead to stomach irritation and nausea. Discussion of this side effect and guidance for individuals with chronic illnesses and their caregivers about what to eat prior to medication dosing may prevent this side effect, thereby increasing adherence. Educational efforts should not occur at a single point because as children age or family dynamics change, reeducation is often necessary. For example, during adolescence, if appropriate, healthcare providers should ensure the adolescent has knowledge about the disease and its treatments since responsibility for disease management often begins to shift during this developmental period. Although educational efforts are clearly necessary to ensure optimal adherence, they are typically insufficient in changing adherence behaviors (Kahana et al., 2008).

Behavioral approaches designed to optimize medication adherence often include behavioral contracting, reinforcement, self-monitoring (e.g., tracking medication dosing through an app), problem-solving, and using visual reminders (e.g., calendars, charts) (Drotar, 2013). Chaining of behaviors, which is associating or pairing new behaviors with established behaviors (i.e., daily routines), is also a helpful strategy to promote adherence. For example, individuals often benefit from chaining medication dosing to activities, such as eating breakfast or brushing teeth. Contingency management strategies, which use basic behavioral principles, are helpful to motivate behavior change in young children. Specifically, caregivers can use charts or calendars to encourage behaviors that promote better adherence. By using behavioral charts/calendars, youth or their caregivers can mark off the days they perform a treatment (e.g., nebulizer) on a calendar or chart with the goal of working toward a reward, such as time on a tablet or picking out a family movie. These more intensive interventions often include caregivers to implement necessary behavior change strategies to optimize adherence. Children and adolescents may experience normative developmental challenges (i.e., behavioral noncompliance), which may interfere with medication adherence. Strategies such as contingency management and token economy systems are effective in addressing behavioral issues that compromise adherence (Kahana et al., 2008). For example, a young child may have temper tantrums prior to medication dosing due to the medication taste or difficulties with pill swallowing. The implementation of a token economy

system by caregivers can ensure the necessary reinforcement to minimize the frequency of temper tantrums and increase adherence behaviors.

Multicomponent interventions integrate components in the aforementioned strategies to provide a more comprehensive and individualized intervention to target adherence behaviors. For example, multicomponent interventions that combine education, problem-solving, and family communication have been used in several pediatric conditions to address adherence barriers, including type 1 diabetes (Driscoll, Killian, Johnson, Silverstein, & Deeb, 2009; Wysocki et al., 2000, 2006, 2007; Wysocki, Harris, et al., 2008; Wysocki, Iannotti, et al., 2008), cystic fibrosis (Quittner et al., 2019), and epilepsy (Modi, Guilfoyle, Mann, & Rausch, 2015; Modi, Guilfoyle, & Rausch, 2013). Multisystemic therapy, which is an intensive family and community-focused therapy, has also successfully been used across a number of pediatric conditions, such as type 1 diabetes (Ellis et al., 2005; Ellis, Naar-King, Cunningham, & Secord, 2006) and HIV (Ellis et al., 2005, 2006). These interventions generally have the most robust effects as they address several adherence barriers that may exist for children and their families (Kahana et al., 2008).

Technology-focused delivery methods have increased tremendously in the past decade, as noted in several subsequent chapters. Mobile health (mHealth), electronic health (e-Health), and telehealth are used in medical and mental health because they are practical, accessible, culturally and developmentally acceptable, and cost-effective. An increasingly large number of adherence interventions capitalize on the use of mobile health platforms, with a 2017 review identifying 824 commercial adherence apps (Dayer et al., 2017). These types of tools are well matched to the information consumption patterns of adolescents and adults, as approximately 92% have cell phones (~70% smartphones), 90%–97% of those with cell phones use text messaging, and ~90% access the Internet on their mobile device (Pew Research Center, 2015). Individuals with lower income or of minority status are increasingly depending on smartphones for Internet access (Pew Research Center, 2015). A recent meta-analysis of text-messaging interventions to improve adherence revealed significant benefits in 18 of 29 studies; however, only 3 of the 18 studies were pediatric focused (Park, Howie-Esquivel, & Dracup, 2014). Another review of electronic medication packaging and digital reminders found variable improvements in adherence, but studies lacked scientific rigor (e.g., small sample, nonrandomization) and were primarily adult focused (Checchi, Huybrechts, Avorn, & Kesselheim, 2014). Furthermore, these technologies work best when the content is tailored to the individual needs of the child/adolescent and his/her family (Park et al., 2014). One example of this is the provision of adherence feedback via technology back to the individual on a routine basis. For example, with the use of electronic adherence monitors that allow for real-time data collection (e.g., AdhereTech or SimpleMed