IMPROVING ADHERENCE IN ADOLESCENTS WITH HEMODIALYSIS-DEPENDENT END-STAGE RENAL DISEASE: A FEASIBILITY STUDY

by

MARY E. PAYNE

(Under the Direction of Ronald Blount)

ABSTRACT

The goal of the current study was to examine the feasibility of implementing a six-module, multi-component, adherence-enhancing intervention with two adolescent hemodialysis patients. Single-subject, case study design was utilized, and baseline and treatment data are presented for the participants. Patients’ responses to the intervention were mixed. During the treatment phase, one participant showed marked improvements in adherence to medications after training on overcoming cognitive barriers to adherence. She and her mother both reported high levels of responsibility for her medical care throughout the study. The other participant had high baseline levels of adherence, but this was due almost exclusively to his mother’s responsibility for the medical regimen. He and his mother reported that he assumed almost no responsibility for adherence behavior. He demonstrated worse adherence in the latter half of baseline which continued during the majority of the treatment sessions. These reductions in adherence coincided with his mother’s increased frustration with his not assuming responsibility for his care and his high oppositional behavior. She decreased her responsibility for his care and withdrew from the study after the third treatment module. However, she allowed her son to continue to participate. The role of adolescent and parent regimen responsibility in response to treatment are explored. Additional possible explanations for differential responses to the
treatment are discussed. Limitations, future directions, and clinical implications of the research are addressed.

INDEX WORDS: adherence, adolescent, hemodialysis, regimen responsibility, renal disease
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DEDICATION

This research is dedicated to all of the staff affiliated with the pediatric hemodialysis unit where this study took place. Their continued support of this patient population is inspiring, and I have learned much from working with them. I hope they recognize daily what a change they make in the lives of these children. Further, I must acknowledge the role of the participants in helping me grow as a researcher and a clinician.
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On a more personal level, thanks to my future husband for putting up with years of graduate school and lots of driving. And I cannot forget my family who are always in the background as my lifeline, especially, my sister who is always there to remind me that what I am doing matters!
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CHAPTER 1
INTRODUCTION

End-stage renal disease (ESRD) is an incurable, chronic medical condition that is typically defined as the final stage of kidney failure in which both kidneys are functioning at less than 15%. ESRD has a prevalence of more than 450,000 people in the United Stated. This illness is often associated more with elderly patients; however, approximately 7,000 pediatric patients, ages 0-19 years, are affected (U.S. Renal Data System [USRDS], 2007). This represents an increase of approximately 2,000 patients from eight years prior (USRDS, 1999).

Treatment for ESRD is multifaceted. Primary treatment methods include home- or clinic-based dialysis and kidney transplantation. Transplantation often is seen as the best treatment option. However, due to a limited supply of donor organs, this is not always feasible making dialysis necessary for many patients with ESRD. Home-based dialysis, known as peritoneal dialysis, is completed nightly by patients in their own homes. Hemodialysis (HD) involves attending four-hour sessions at a clinic setting, three times a week. In addition to dialysis procedures, patients are prescribed changes in their diet (e.g., decreased intake of phosphorus, sodium, and potassium, increased caloric intake) and restrictions to their fluid intake (e.g., typically limited to 1 L to 1.5 L per day). They also are required to take multiple daily medications. Several of these prescribed changes, especially fluid reduction, are most applicable to patients on hemodialysis.

More than 1,000 children in the United States currently receive hemodialysis, with a yearly incidence rate of approximately 600 children (USRDS, 2007). These children have very complex medical regimens and often exhibit difficulties following their medical recommendations. Non-adherence is associated with increased burden on the health care system through medical complications, increased hospitalizations, and even increased mortality.
(Christensen & Ehlers, 2002; Morgan, 2000). Also, because the supply of donor kidneys is limited, patients who are found to be non-adherent may be less likely to receive a kidney transplant, possibly resulting in death. Additionally, if given a transplant, pre-transplant non-adherence is associated with a greater likelihood of post-transplant non-adherence to anti-rejection medications, which can lead to organ loss, a return to dialysis, or even death (Chisholm, Williamson, Lance, & Mulloy, 2007; Rianthavorn & Ettenger, 2005).

Despite the severe consequences of non-adherence, a study by Simoni et al. (1997) found that average blood levels were indicative of adherence to medications and diet only 39% to 43% of the time in pediatric populations on HD for ESRD. Further, 17-26% of these children never had normal blood levels for phosphorus or calcium. More generally, non-adherence to medical regimens in a variety of pediatric populations is estimated to be between 50%-55% with estimates typically highest in adolescent populations (Rapoff, 1999).

The Problem of Non-Adherence

Definition of adherence. A report by the World Health Organization (WHO) defined adherence as “the extent to which a person’s behaviour – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed [upon] recommendations from a healthcare provider” (Sabate, 2003, p. 3). This definition has a high level of clinical utility in that it emphasizes the role of behavior, which is malleable. Also, by highlighting adherence as the extent of correspondence between behavior and recommendations, this definition takes a dimensional view of adherence. This allows for a patient’s adherence classification to move along a continuum throughout the course of his/her treatment. It also individualizes adherence behavior to one’s own prescribed regimen and not an ideal regimen that may not be consistent with an individual’s prescription (La Greca, 1990). Compliance is a term that is commonly used interchangeably with adherence. However, adherence is the more accepted term as compliance can suggest that patients are to follow doctor recommendations without question, making them
passive players in their own healthcare, while the term adherence better implies and promotes collaboration, highlighting patients’ active roles in their own care (Rikert & Drotar, 2000).

Several different types of non-adherent behavior have been identified, including volitional and inadvertent non-adherence (Bauman, 2000). Volitional non-adherence occurs when the patient’s treatment goals are not in line with those of the healthcare provider, especially if the recommended treatment is in opposition with the patient’s lifestyle or health beliefs. Volitional non-adherence often occurs with complex and intrusive regimens that do not guarantee either symptom relief or illness recovery (Deaton, 1985; Donovan & Blake, 1992; Johnson, 1994; Mattar, Markello, & Yaffe, 1975). Conversely, inadvertent non-adherence is unintentional and may result from several different processes. For example, patients may believe that the extent of their following through on recommendations, despite occasional non-adherence, is “good enough” resulting in low motivation to improve adherence behavior. Another process occurs when highly motivated families encounter barriers to adherence such as high medication costs, forgetting, or difficulty planning around the treatment regimen. A third process leading to inadvertent non-adherence occurs when the recommended regimen is misunderstood or inadequately communicated, and the patient adheres to an incorrect regimen.

The distinctions between non-adherent behavior are important to acknowledge as they may modify the components of adherence-enhancing interventions. Volitional non-adherence would likely benefit from improved communication and strategies to effectively address conflicting goals between the patient and the prescribed regimen. For inadvertent non-adherence, education and knowledge are necessary factors. Also, depending on the process that is contributing to non-adherence, interventions may include strategies that range from addressing health beliefs to problem-solving as ways to identify and overcome barriers (Greenstein & Siegal, 1998; Rianthavorn & Ettenger, 2005).
More recently, a multidimensional adherence classification system (MACS) has been developed from work with transplant recipients (Simons & Blount, 2007; Simons, McCormick, Mee, & Blount, 2009). The MACS combines serum drug assays and self-reported adherence levels to yield four adherence classification categories. The first category accounts for patients who report being adherent and who have acceptable serum drug levels. These patients are classified as *genuinely adherent*. Patients who report being adherent, but have drug levels that are unstable or outside of a recommended range are categorized as *deniers or medically complicated*. This group could be engaging in non-adherent behavior and denying it, or they could have biological factors that make their disease more difficult to manage even with adherent behavior. The third category reports being non-adherent but have stable and acceptable drug levels. This group is classified as *at-risk* as they could develop unstable drug levels in the future if their report of non-adherent behavior is accurate. The final group accounts for those patients who report non-adherent behavior and have concerning or unstable drug levels. This group is classified as *genuinely non-adherent*. This type of classification system helps account for results of studies, such as Johnson (1994), that found approximately one-third of patients with diabetes who reported good adherence had poor metabolic control (*deniers or medically complicated*), while another third with poor adherence had good metabolic control (*at-risk*).

**Rates of non-adherence.** Estimated rates of non-adherence in pediatric populations are about 50-55% (Litt & Cuskey, 1980; Rapoff, 1999). Notably, this high rate of non-adherence is more applicable to chronic than acute medical conditions (Rapoff & Barnard, 1991). Rapoff (1999) reviewed the prevalence of non-adherence found in several empirical studies and found rates of reported non-adherence ranging from 4-98%. This extremely wide range highlights inconsistencies in defining and measuring adherence throughout the literature. A weakness of past research has been in the varying methods used to determine adherence rates. Research
often arbitrarily defines non-adherence as less than 80% adherent. Shockingly, Burkhart and Dunbar-Jacob (2002) found that in the pediatric adherence literature between 1987 and 1996, only 83% of studies reported adherence rates. The other 17% of studies only presented data on factors correlated with adherence without mention of adherence rates for the sample studied. This is especially concerning given a call in 1983 by Dunbar for researchers to report the percentage of regimen with which the patient adheres so that comparison of adherence assessments across studies can be more easily made.

How adherence is measured also can impact results. Commonly used assessments for adherence include assays, observation, technological monitoring (e.g., electronic caps for medication bottles), pill counts, provider estimates, and patient reports. Each method has associated assets and liabilities, and none is a gold standard assessment. Research has generally indicated that self- and parental-report are the most convenient ways to assess adherence behavior but that these reports are prone to social desirability and can overestimate adherence behavior (Bond & Hussar, 1991; Rand & Wise, 1994; Rapoff & Barnard, 1991; Rudd, 1993). A strength of self-report is that it can be used to assess adherence to a variety of treatment regimen components such as diet and exercise programs in addition to medication taking. Research has found that the use of assays, electronic monitors, or pill counts may provide more objective adherence estimates, but these are not always feasible and are typically only useful to assess adherence to medication regimens (Sluijs et al., 2006). Additionally, serum assays can be influenced by a variety of factors such as individual metabolism. Adherence rates also vary depending on which components of medical recommendations are being measured (e.g., medication versus dietary adherence in ESRD). Reported non-adherence rates for adult dialysis patients have ranged from 70-80% for fluid restrictions and as low as 10-20% for dietary potassium restrictions (White, 2004). Generally, non-adherence appears to be most common to fluid recommendations compared to dietary and medication guidelines (Christensen & Ehlers,
In a study looking at pediatric ESRD patients, 69% were found to be non-adherent with medication taking; 39% were non-adherent concerning their blood pressures, urine, and/or weights; and 19% were non-adherent in their appointment keeping (Wolff, Strecker, Vester, Latta, & Ehrich, 1998).

**Consequences of non-adherence.** Such high rates of non-adherence are alarming given the significant consequences of non-adherence for both health and the economy. Across pediatric chronic illnesses, non-adherence has been associated with treatment failure, increases in mortality, and a re-emergence of infectious diseases that are increasingly antibiotic resistant (Rapoff, 1999). Additionally, poor adherence negatively impacts quality of life through more frequent experiencing of symptoms, more hospitalizations, and lower daily functioning. For pediatric patients on dialysis due to ESRD, non-adherence to fluid restrictions can lead to congestive heart failure, hypertension, pulmonary edema, and decreased survival rates. Non-adherence to phosphate binding medication and recommendations to decrease phosphorus intake has been associated with renal osteodystrophy and significant calcium deficiencies resulting in bone demineralization that may be irreversible. Non-adherence to restrictions of potassium-rich foods has been associated with increased serum potassium levels that can result in fatal cardiac arrhythmias. Research has shown that when non-adherence leads to low hemoglobin or serum albumin levels, patients have significantly higher risk for both hospitalization and mortality (Amaral et al., 2006; Wong, Hingorani, & Gillen, 2002).

Non-adherence also can impact the quality of healthcare received by a patient. If patients are not honest about or forget to report their non-adherent behavior, their doctors may interpret a lack of progress as due to ineffective medication dosage. This can lead doctors to make uninformed medical decisions that could, in turn, worsen patient outcome. Also, if doctors know or suspect chronic non-adherence, they may be less open to making necessary regimen changes. For ESRD patients, significant non-adherence may lead a medical team to delay or
deny renal transplant (Rianthavorn & Ettinger, 2005) since transplantation requires life-long adherence to immunosuppressant medication regimens to maintain and protect the organ. Non-adherence to this medication could result in organ loss and potentially death. This is an especially important point as past adherence is a predictor of future adherence behavior.

Non-adherence also has significant costs to the healthcare system and broader general economic consequences. Notably, medication non-adherence can mean that insurance companies or government health care plans are paying for unused medication. The healthcare system also becomes burdened by potentially avoidable emergency room visits and hospitalizations. Berg and colleagues (1993) estimated the direct U.S. costs of non-adherence across children and adults to be about $100 billion a year, and this number was expected to rise. Yearly indirect costs associated with non-adherence include lost earnings by the patient or family, estimated at $1.5 billion, and loss in productivity which accounts for another $50 billion (Peterson, Takiya, & Finley, 2003). This unnecessary strain on the economy caused by non-adherence potentially can lead to increased health care and insurance costs as well as higher tax rates. This point is especially important to consider with the ESRD population as all children on dialysis for ESRD are Medicare eligible.

Factors Associated with Adherence

Research on adherence in pediatric ESRD patients is limited and largely correlational. However, the literature on adherence for other pediatric medical conditions (e.g., asthma, diabetes, and cystic fibrosis) and for adults with ESRD provides insight into factors associated with non-adherence and ways to improve adherence in adolescents with HD-dependent ESRD. This body of research takes into account nonmalleable factors, such as demographics and disease factors, which, even though they are not modifiable using behavioral interventions, may help identify those who are in need of treatment. The literature also has examined the association between adherence and modifiable cognitive and behavioral skills. The combination
of these factors is generally divided into three broad categories: disease factors, regimen factors, and patient/family factors (Rapoff, 1999). Additionally, there is literature that emphasizes the role of developmental issues in adolescence that can influence adherence rates. Each of these areas is discussed in detail, and within each area, nonmalleable and modifiable factors are identified.

**Disease factors.** The associations between adherence and disease-related factors, including duration, course, symptoms, and perceived severity have been examined. Diseases with a longer duration typically have poorer adherence levels. This is especially important given that most chronic illnesses, including ESRD, are life-long conditions without cures (Bond, Aiken, & Somerville, 1992; Brownbridge & Fielding, 1994; Jacobson et al., 1990; Kovacs, Goldston, Obrosky, & Iyengar, 1992; Rapoff, 1999). Even if a child with HD-dependent ESRD receives a kidney transplant, he/she is faced with a life-long medical condition for which immunosuppressant medications are required to maintain the new organ. The association between adherence and duration of the disease suggests that it is important to periodically monitor adherence for families who initially appear adherent soon after diagnosis. Given that adherence generally decreases over time, La Greca and Bearmen (2003) highlight the need to focus on supporting adherence before it becomes a problem.

The effects of disease course on adherence have been less researched in chronic medical conditions; however, some studies suggest that adherence rates correlate with the presence of overt symptomatology, with lower adherence occurring during asymptomatic periods (Lemank, Kamps, & Chung, 2001; Rapoff, 1999). Often patients with acute medical conditions take medication for symptom relief rather than to avoid being symptomatic. This pattern becomes especially problematic when it occurs in patients with chronic medical conditions. These patients likely do not understand the connection between non-adherence and the manifestation of physical symptoms. Therefore, it is important to manage patients’ symptom
perception throughout the course of their illness so that they can recognize the relationship between adherent behavior and the prevention of overt symptoms.

Associations between adherence and both objective disease severity and a patient’s perceived severity also have been examined. Generally, the literature does not show significant associations between objective disease severity and adherence (Rapoff & Barnard, 1991). In contrast to objective disease severity, an individual’s perception of illness severity has been significantly associated with adherence (Gudas, Koocher, & Wypig, 1991; Rapoff & Barnard, 1991). Research indicates that for pediatric patients, high levels of perceived severity are associated with more adherence problems. Conversely, high parental perceived severity regarding their child’s condition is associated with better adherence, likely through increased levels of monitoring by the parents (Bond et al., 1992; Gudas et al., 1991; Radius et al., 1978). This is important in that perceptions can be modified using cognitive-behavioral therapy techniques. Rather than potentially overwhelming adolescents by strongly emphasizing the severity of the condition, the focus should be on what they can do to control their symptoms.

*Regimen factors.* Aspects of the medical treatment also are correlated with adherence rates. The literature consistently shows that the more complex a treatment regimen, the less adherent the patient (La Greca & Bearman, 2003; Rapoff, 1999). This finding is especially notable for patients with HD-dependent ESRD. These patients have multiple medications, very specific and complex dietary needs, significant fluid intake restrictions, three weekly four-hour clinic-based dialysis sessions, and other routine clinic visits that must be maintained for optimal health. This treatment regimen is burdensome and interferes with normal daily life activities, two factors that are associated with lower adherence rates (Fotheringham & Sawyer, 1995; Matsui, 2000). This regimen also can be intrusive for parents who must provide transportation to and from clinic for their adolescent multiple times a week.
The presence of treatment side effects repeatedly has been associated with lower rates of adherent behavior, even when faced with life-threatening consequences (Blowey et al., 1997; Korsch, Fine, & Negrete 1977; Radius et al., 1978; Simons & Blount, 2007). In some cases, an alternative medical treatment may be prescribed. However, that is not always possible. In those situations, it may be possible to work with patients on strategies to minimize negative effects or develop plans to cope with the negative effects of treatment.

Another regimen factor is the efficacy of the treatment. Healthcare providers view treatments as efficacious when symptoms decrease and related measures such as laboratory values indicate improvements. Unfortunately, many medical treatments, especially those for chronic conditions, are only partially efficacious in managing symptoms (Johnson, 1994). From a patient’s perspective, treatment efficacy is comprised of both a patient’s perception that the treatment has immediate effect and the perceived benefit of the treatment. Both of these factors have been shown to influence adherence (Horne & Weinman, 2002; Rapoff, 1999). For chronic illness, treatment often serves a maintenance role or may slow disease progression, rather than a curative role with observable improvements in health. It is often the case that, for many maintenance treatments, there are few or no observable or immediate consequences for non-adherence (Dunbar, 1983; Litt & Cuskey, 1980). However, research indicates that patients who have a higher level of perceived treatment benefit, in spite of the presence of adverse effects, have higher adherence levels (Bond et al., 1992; Greenstein & Siegel, 1998; McCaul, Glasgow, & Schafer, 1987). Given this, it is important to help patients understand how their treatment works and to adapt their cognitions and behavior to maintain health and prevent future medical complications.

Many treatments for chronic medical conditions are very expensive. Further, insurance coverage varies between families. Fortunately, all ESRD patients on dialysis are Medicare-eligible, including pediatric patients. Medicare generally pays for clinic visits, dialysis, and
medications. However, the costs for following strict dietary recommendations also must be considered. In addition, the costs of seeking mental health care to work on quality of life or adherence difficulties often are not covered as extensively as are medical issues. Often, there are additional costs that accompany adherence. For example, parents miss work or are unable to maintain a job in order to care for their children adequately. This can lead to decreased income and increased stress for families, potentially further negatively impacting adherence.

While not all parts of a medical treatment regimen can be modified, aspects of the doctor-patient relationship can be improved. The literature has shown that specific aspects of this relationship are associated with adherence behavior. La Greca and Bearman (2003) identified the following factors as positively associated with adherence: parental satisfaction with medical care (Cromer & Tarnowski, 1989; Litt & Cuskey, 1984); established doctor-patient rapport including a friendly, warm, empathetic, and supportive perception of the doctor (Litt & Cuskey, 1980); and continuous service with a provider (Litt & Cuskey, 1984). In addition, good doctor-patient communication is important for ensuring that regimens are recalled and followed appropriately by patients and families (Alexander, 1983; Ievers-Landis & Drotar, 2000; Page, Verstraete, Robb, & Etzwiler, 1981; Schraa & Dirks, 1992; Staples & Bravender, 2002). The healthcare setting also can impact adherence. Worse adherence is associated with inconveniently located medical clinics, lack of transportation, and lack of availability of childcare for siblings (Hazzard, Hutchinson, & Krawiechi, 1990; Saylor, Elksnin, Farah, & Pope, 1990). Pediatric dialysis centers are not always easily accessible and often are located in cities, making them less accessible to rural populations. There rarely is access to childcare for non-patients during these visits, making it difficult for parents with multiple children to be present during a patient’s four-hour dialysis session.

Patient and family factors. Patient factors include demographic data, psychiatric and behavioral problems, knowledge about illness and treatment, cognitive factors such as health
beliefs, coping strategies for managing illness and treatment, and family functioning (Anderson, Auslander, Jung, Miller, & Santiago, 1990; Redding, Rossi, Rossi, Velicer, & Prochaska, 2000; Shemesh et al., 2004; Treadwell et al., 2005). Demographic factors (e.g., race, gender, religion, and education level) have not consistently been associated with adherence (Burkhart & Dunbar-Jacob, 2002; Cromer & Tarnowski, 1989). A quantitative review by DiMatteo (2004) suggested that in pediatric populations, females have higher adherence levels than males but that these gender differences may be a result of differences in risk-taking behavior and do not continue into adulthood. This same review suggested that for chronic illnesses, but not acute ones, higher education levels may be associated with better adherence. Results from research exploring the effects of socioeconomic status on adherence have been mixed with some studies showing no correlation (Kovacs et al., 1992; Wysocki et al., 1996) while others identify a positive association (Brownbridge & Fielding, 1994; Charron-Prochownik, Becker, Brown, Liang, & Bennett, 1993; DiMatteo, 2004; Manne, Jacobsen, Gorfinkle, Gersten, & Redd, 1993; Miller-Johnson et al., 1994). These factors are not modifiable, but it is important to understand them as they could have moderating influences on both adherence levels and intervention effectiveness.

Age and developmental factors have more consistently been shown to be associated with adherence in that adolescents show the worst adherence rates (La Greca & Berman, 2003). The OPTN/SRTR annual report (2004) found that adolescents who have received a transplanted organ have the worst 5-year survival rates of any age group. Additionally, research on patients with diabetes has found that not only do adolescents have significant difficulties with treatment adherence (Schmidt, Klover, Arfken, Delamater, & Hobson, 1992), but also they exhibit declines in adherence throughout adolescence (Wysocki, Greco, & Buckloh, 2003). A review by Burkhart and Dunbar-Jacob (2002) of pediatric adherence research found similar patterns across several studies. To date, research suggests that adolescents may have
difficulty internalizing the significance of being adherent which requires cognitive development, abstract thinking, and the ability to recognize short- and long-term consequences of their behavior (Ringewald et al., 2001; Shaw, Palmer, Blasey, & Sarwal, 2003). It is thought that adolescents commonly see themselves as invulnerable and have not fully developed the complex abstract reasoning necessary to understand the future consequences of non-adherent behavior that has no immediate effect (Rianthavorn & Ettenger, 2005).

Adolescents also are at high risk for non-adherence as they struggle to achieve independence from parents (Bauman, 2000). Adolescence is a time of separation and individuation from parents and families; however, in the case of a teen with a chronic medical condition, the level of parental monitoring and supervision is higher than would be necessary in a healthy teen (La Greca, Follansbee, & Skyler, 1990; Shaw et al., 2003). It is important to understand how regimen responsibility is successfully transferred from parent to teen including who assumes which responsibilities and how responsibilities are shared (Anderson et al., 1990; La Greca et al., 1995). It has been suggested that with an increasing prevalence of single parent homes and dual career households, there is an increasing responsibility placed on children and teens to self-manage their treatment regimen, often without appropriate levels of monitoring (Burkhart & Dunbar-Jacob, 2002). Research suggests that premature transition of responsibility, especially in the absence of maternal monitoring, is associated with higher levels of non-adherence (Mellins, Brackis-Cott, Dolezal, & Abrams, 2004; Simoni et al., 2007). Research strongly supports that navigating the transition of responsibility while maintaining good communication, clear responsibility expectations, and a collaborative approach to illness management is associated with higher adherence levels (Anderson et al., 1990; Fiese & Everhart, 2006; Mellins et al., 2004; Staci et al., 2007; Wiebe et al., 2005).

Additionally, there can be difficulties adjusting to being different from peers and a struggle to prioritize time for regimen management when it seems to interfere with peer
relationships (Gross, Johnson, Wildman, & Mullett, 1981; La Greca & Berman, 2003). Some non-adherent behavior may result from testing the boundaries of social limitations that an illness or treatment regimen places on the teen (La Greca et al., 1995). Additionally, common adolescent issues, including struggles with self esteem, defining social roles, and increasing peer-related and body image issues, often are made more difficult to navigate by the presence of a chronic medical condition (Burkhart & Dunbar-Jacob, 2002). This research emphasizes that developmental factors must be considered when extrapolating adult data and utilizing or developing theoretical models or interventions to child and adolescent populations.

Non-adherence to treatment regimen may co-occur with psychiatric or family problems (Bauman, Drotar, Leventhal, Perrin, & Pless, 1997; Rapoff & Barnard, 1991; Shaw, 2001). Significant psychosocial problems have been associated with worse adherence to treatment regimens (Griffin & Elkin, 2001; La Greca & Bearman, 2003). Low self-esteem, substance abuse, oppositional defiant disorder, and the presence of other family psychopathology have been associated with non-adherence behavior (Rianthavorn & Ettenger, 2005). In a study with pediatric renal dialysis patients, more depressive symptoms including increased hopelessness, negative self-perceptions, and a depressogenic attributional style were associated with lower adherence rates (Simoni et al., 1997). Conversely, positive psychosocial adaptation, including better self-esteem and less anxiety or depressive symptomatology, is associated with increased adherence (Brownbridge & Fielding, 1994; La Greca & Bearman, 2003; Littlefield et al., 1992).

Several researchers have indicated a need for adherence research to account for quality of life outcomes and general adjustment (Johnson, 1994; Roter et al., 1998). Currently, the association between adherence and quality of life is equivocal. There is some evidence that strict adherence to a complex prescribed regimen may have an adverse effect on quality of life (Varni, Jacobs, & Seid, 2000). This may be due to complex regimens interfering with normal developmental activities with family and peers at home and school. However, there also is
research that suggests that lower adherence levels are associated with poor adjustment and quality of life (Kurtin, Landgraf, & Abetz, 1994). A study with pediatric patients receiving dialysis found that lower reported adherence was correlated with poorer adjustment to both the diagnosis and the treatment regimen for both parents and children (Johnson, 1994). Regardless of the direction of the association, a study examining differences in quality of life in pediatric ESRD populations showed significantly lower scores when compared to healthy controls (Goldstein et al., 2006).

In a 2003 review, La Greca and Berman discussed the predictive power of both knowledge and problem-solving skills on adherence. Their review of the literature found that for younger children, parent’s problem-solving skills (Fehrenbach & Peterson, 1989) and knowledge (La Greca et al., 1990) were predictive of adherence, while for adolescents, it was their own knowledge that was predictive (La Greca et al., 1990; Lorenz, Christensen, & Pichert, 1985). However, it also is important to clarify that while knowledge is necessary for adherence behavior to occur, it is not sufficient. This is especially true for complex, repeated, or aversive regimens, as other factors can impede adherence behavior (La Greca & Skylar, 1991; Mackner, McGrath, & Stark, 2001; Milas et al., 1995; Sluijs et al., 2006). Still, knowledge of the treatment regimen should be considered as a prerequisite for adherence. The importance of accurate knowledge and understanding of the treatment regimen is made clear by a study of 15-31 year old patients receiving hemodialysis (Chan & Greene, 1994). Results indicated that only 27% of the sample adequately recalled their potassium recommendations, only 23% recalled their prescribed sodium restrictions, and only 47% adequately remembered their fluid restrictions.

Cognitive factors including illness perceptions and beliefs about the medical regimen have been shown to be associated with adherence and often are viewed as modifiable with cognitive therapy techniques. Although much of this research has been done with adult populations (Bandura, 2004; Redding et al., 2000; Strecher, Devellis, Becker, & Rosenstock,
1986), there have been some studies to examine this further with pediatric populations (Bush & Iannotti, 1990; Reikert & Drotar, 2000; Zebracki & Drotar, 2004). A prospective study indicated that, over time, adjustment to dialysis improved, and the improvement was greater in those patients with a high sense of self-efficacy (Lev & Owen, 1998). The presence of self-efficacy, perceived consequences of illness, and perceived symptoms were identified as positive predictors of adherence behavior in a study of patients with Type-1 diabetes (Newman, Steed, & Mulligan, 2004). Together, these three cognitive factors accounted for 31% of the variance in metabolic control for the sample. Studies with adult ESRD populations suggest that expectations of self-efficacy for treatment management are positively related to fluid intake and medication adherence among patients receiving dialysis (Brady, Tucker, Alfino, Tarrent, & Finlaysos, 1997; Christensen, Wiebe, Benotsch, & Lawton, 1996). Other research with this population also shows that the presence of an internal locus of control is associated with higher adherence levels (Christensen & Ehlers, 2002). A large-scale study of adult patients with ESRD found that those who perceive themselves as successful at being adherent and those who report greater satisfaction with recommended eating habits were more adherent to dietary restrictions (Milas et al., 1995). Higher perceived control over the illness and its treatment also has been associated with increased adherence to dietary recommendations (Skinner & Hampson, 2001). The perception of consequences for non-adherent behavior also is an important correlate. Adult research suggests that without the perception of severe consequences (e.g. death, worsening condition), non-adherent behavior will be more prevalent (Brewer, Chapman, Brownlee, & Leventhal, 2002; Byer & Myers, 2000). However, this is contrary to research with adolescents finding that higher perceived severity is associated with less adherence. More data on these factors are needed, especially with pediatric populations.

Researchers have investigated the impact of coping strategies on adherence. Results indicate that avoidance strategies often are used in attempts to manage unpleasant emotional
aspects of adherence but that this strategy is associated with lower levels of adherence (Shemesh, Lurie, & Stuberm, 2000). This suggests a need for interventions that promote the use of more appropriate coping skills such as seeking social support, distraction, or relaxation skills.

For patients with ESRD, there are a number of differences between the two dialysis treatment options that involve factors associated with adherence. Patients undergoing peritoneal dialysis receive their treatments at home, giving them higher levels of control over treatment than patients receiving clinic-based hemodialysis. Christensen (2000) proposed a model suggesting that when the patient’s coping behavior with illness and treatment-related experiences is congruent with the demands of the medical intervention, adherence is improved. Results from this research indicate that patients with ESRD undergoing peritoneal dialysis at home have better adherence when they use more active coping behavior while patients receiving hemodialysis, a provider-controlled, hospital-based treatment, show better adherence when employing more passive coping behavior (Christensen, 2000).

Studies have validated this model, finding that both adherence and adjustment are maximized when coping behavior matches the context of the treatment. More specifically, this research found that patients with active and/or vigilant coping styles had higher rates of adherence when in a primarily patient-controlled treatment situation (e.g., peritoneal dialysis), while patients with less active coping styles had better adherence when in clinic-based treatment settings (e.g., hemodialysis; Christensen et al., 1995, 1996, 2002). These data suggest that patients undergoing HD would likely benefit from interventions that highlight controllable aspects of treatment and active coping skills to deal with these. Additionally, teaching appropriate emotion coping techniques (e.g., distraction and acceptance) for non-controllable aspects would likely be a necessary additional component for maximizing adjustment.
The role of family support and encouragement has been described throughout the adherence literature (Gerson, Furth, Meu, & Fivush, 2004; Kovacs et al., 1992; La Greca & Bearman, 2003; Staples & Bravender, 2002). These family functioning factors also have been shown to have a significant impact on adherence, so much so that La Greca (1990) has emphasized that interventions to improve adherence should necessarily include a family component. Increased parental support has been associated with better adherence by adolescents (La Greca & Bearman, 2003). On the other hand, this support may be difficult for parents to provide as complex treatment regimens can disrupt not only patient routines and lifestyle but also those of the family. A study of pediatric patients with ESRD showed that family characteristics, such as high family expressiveness, are positively correlated with both medication and dietary adherence (Davis, Tucker, & Fennell, 1996). Conversely, high family stress and conflict are inversely associated with adherence (Griffin & Elkin, 2001; Hauser et al., 1990; La Greca & Bearman, 2003).

Drotar et al. (2000) also expressed concern about effects of non-adherence on families, speculating that non-adherence can increase family conflict. Non-adherence also can increase parental anxiety about the child’s health status. For this reason, parents may become overprotective, controlling, or unable to provide appropriate emotional support for their children (Dobbels, Van Damme-Lobart, Vanhaecke, & DeGest, 2005; Korsch et al., 1977; Kulik & Mahler, 1993). Anderson and Coyne (1991) proposed that children come to resent efforts perceived as interference by parents and become increasingly non-adherent to treatment, subsequently increasing parental guilt or worry about their children’s future health. More generally, social support is an important resiliency factor associated with adherence. A review by DiMatteo (2004) shows that higher levels of social and family support, especially practical support (e.g., choosing and preparing healthy foods and going to doctor’s appointments), are
associated with increased adherence. This suggests that there is a need for interventions to provide practical help to patients with regard to adhering to their medical regimens.

**Barriers to Adherence**

The adherence literature has moved beyond simply identifying factors associated with adherence to examining more proximal barriers to adherent behavior. This literature is discussed separately to highlight the variety of factors that patients and families perceive as causal to their non-adherent behavior. This research consistently shows increased barrier perception to be associated with decreased adherence (Chisholm, Lance, & Mulloy, 2005; Chisholm, Lance, Williamson, & Mulloy, 2005; Marhefka et al., 2004; Reddington et al., 2000; Simons, McCormick, Mee, & Blount, 2008) and increased health problems (Logan, Zelikovsky, Labay, & Spergel, 2003; Riekert & Drotar, 2000). Simons and Blount (2007) assessed barriers to immunosuppressant medication after transplant. They recommended that assessing barriers to adherence is important not only after transplant but also before, as pre-transplant non-adherence is a risk factor for non-adherence post-transplant. They suggested that early intervention could lead to better medical outcomes. For adolescent renal patients, the top three barriers to medication adherence were forgetting, being away from home, and being engaged in another activity (Zelikovsky & Schast, 2008).

Several modifiable barriers to medication adherence in adolescents were identified by Simons and colleagues (2007, 2009). These include disease frustration, a lack of adaptation to the medical regimen, difficulty swallowing medication, and lack of appropriate parental reminders (Simons & Blount, 2007). Using open-ended questions, themes identified included forgot/distracted, poor planning/scheduling issues, physical barriers/medication issues, and voluntary resistance/attempts to be normal (Simons et al., 2009). Parental reminders were found to be protective when there was a lack of appropriate adolescent knowledge, a large number of prescribed medications, or recommendations emphasizing the need for parental
involvement. Notably, much of the barriers research is focused on medication adherence. This field would benefit from identifying barriers to other regimen factors (e.g., dietary restrictions) for chronic illness populations. A study by Logan et al. (2003) examined barriers to asthma management and developed the Illness Management Survey which asks broadly about adherence to the treatment regimen and is not medication specific. A study by Modi and Quittner (2006) examined barriers to various aspects of treatment adherence for children with cystic fibrosis and asthma and found that similar barriers were reported by the two patient groups for general treatment aspects such as medication taking. However, they additionally identified unique barriers for disease-specific aspects of the treatments (e.g., airway clearance for patients with cystic fibrosis).

A study by Wolff and colleagues (1998) examined barriers that may exist in the relationship with healthcare providers. The results show that 40% or more of patients identified communication barriers, such as being given insufficient information about their disease and treatment, having limited time with the healthcare provider, and feelings of dependency on the provider. Other common barriers reported were feeling that the healthcare provider had “given up” and patients feeling powerless. Relational issues such as feeling disappointed with the doctor, thinking that they were not taken seriously, and not being praised by the doctor for positive behavior also were listed as barriers. Another important factor highlighted in this study was that the medical staff did not take steps to understand the reason for non-adherent behavior.

Theory and Conceptual Models

Theoretical models utilize hypothesized or experimentally established relationships to develop general principles that may explain a phenomenon (Johnston & Pennypacker, 1993). Several health-related theoretical models have been developed that partially explain the occurrence of non-adherent behavior. However, a meta-review (i.e., a review of reviews) on
patient adherence to medical treatment argues that there is a knowledge gap between the literature examining correlated factors and the intervention literature (Sluijs et al., 2006), suggesting that a comprehensive theoretical framework would enhance understanding of the successes and failures of intervention research. Other literature also has highlighted the lack of theory in guiding both correlational research and intervention development (Burkhart & Dunbar-Jacob, 2002; Drotar et al., 2000; Dunbar-Jacob, Dunning, & Dwyer, 1993; Rapoff, 1999). These reviewers suggest that current theories do not adequately describe the problem of non-adherence or give sufficient insight into methodologies for improving adherence. In part, this has lead to approximately 80% of the adherence literature being described as atheoretical (Burkhart & Dunbar-Jacob, 2002). Additionally, these reviewers highlight difficulties in testing the validity of current theoretical models given their limited utilization in research.

A focus group of psychologists identified several factors that should be considered in the development and refining of theoretical models of pediatric adherence (Drotar et al., 2000). These include ideas such as incorporating multiple systems including the child, family, and medical staff as well as considering cognitive, social, and emotional development. This same group also recommended that researchers utilize the risk and protective factors identified in the data on adherence and design interventions to target adherence issues (Drotar et al., 2000). An additional consideration in this study is that many theoretical models were developed from the adult literature that are not fully relevant to pediatric medical adherence, which must consider developmental issues, cognitive ability, and the role of family and peers (Drotar et al., 2000; Rapoff, 1999).

The limited theoretically-based literature in the field of adherence utilizes several different models. The primary models used in pediatric adherence studies are the Health Belief Model and Social Learning Theory. Additionally, theories about health locus of control, approach coping, family systems, stress-coping, and social support models can be found loosely tied to
pediatric adherence literature (Burkhart & Dunbar-Jacob, 2002). While the Theory of Planned Behavior is not often seen in the pediatric adherence literature, it has been used in the adult literature and shows promise of being an effective model; however, like many models, it needs modifications to include the influences of parental, familial, and adolescent transitional factors to better explain non-adherence in this population. While all theories uniquely address the problem of non-adherence, none fully explain the myriad factors and systems that impact adherence-related behavior. Rapoff (1999) provided a review of adherence-related theoretical models and discussed the clinical implications of each theory. His conclusion was that despite the differences in models, how clinicians activate behavior change processes in each of the models would be similar. Specific treatment recommendations included: helping families believe in the value of the prescribed regimen, teaching the necessary skills for carrying out regimen tasks, providing role models who demonstrate successful management of prescribed regimens, assisting patients and families with goal setting and monitoring, and helping patients and families establish reinforcing consequences for adherence (Rapoff, 1999).

The Health Belief Model, developed by Becker and colleagues (1972, 1974), is one of the most commonly used models. This model consists of four key elements: perceived susceptibility to illness or complications, perceived severity of illness, perceived benefits of health-related behavior, and perceived barriers to health care. All of these are seen to predict good adherence with the exception of perceived barriers. This factor, when present to a high degree, is negatively associated with adherence. According to this theory, extreme non-adherent behavior would most likely be found in patients who perceive themselves as safe from disease or complications, not accurately perceive the seriousness of their illness, not believe in the prescribed treatment, and identified significant barriers to treatment. Variations of this model additionally consider other moderating factors including age, personality, the doctor-patient relationship, social support, self-efficacy, and cues to action (e.g., triggers that increase
motivation). While this model has been shown to have utility (Becker et al., 1972; Becker, 1974; Reese & Smith, 1997), it lacks adequate consideration of developmental issues, and research with adolescents has shown that contrary to theory, high adolescent perceived threat is associated with decreases, rather than increases, in adherence behavior (Bond et al., 1992; Brownlee-Duffeck et al., 1987). A series of studies by Gochman (1970, 1971, 1972, 1977a, 1977b) evaluated perceived vulnerability to illness, specifically dental health problems, in children and found weak correlations with dental health care behavior. It also has been noted that in children, many of the other factors in the model have been relatively unexplored (Bush & Iannotti, 1990).

The Health Belief Model has been used to guide pediatric studies on adherence with mixed, but promising, results (Anthony, Gil, & Schanberg, 2003; Bond et al., 1992; Logan et al., 2003; Steele et al., 2001). The current study primarily utilized the Health Belief Model along with adaptations proposed by Bush and Iannotti (1990) to make the model more applicable to pediatric patients. The Child Health Belief Model integrates the primary elements of the Health Belief Model presented above with developmentally appropriate elements from other theories such as emphasizing consequences of behavior, recognizing the level of understanding of social and physical events, and emphasizing specific, not abstract, behavior. Additionally, this model highlights the contexts in which child health behavior occurs including family, peer, and social influences. The current intervention was developed consistent with factors emphasized in this theory, especially through the focus on acknowledging and problem-solving around perceived barriers including practical, behavioral, cognitive, and affective barriers.

**Adherence Enhancing Interventions**

In comparison to correlational research, intervention studies to improve adherence to pediatric treatment regimens are scarce. The WHO study highlighted the need for action to be taken to improve adherence to long-term therapies, which would included hemodialysis
treatment for ESRD (Sabate, 2003). Given the identification of adherence-related factors and attempts to develop theoretical models to explain non-adherent behavior, several researchers have developed adherence-enhancing interventions. However, interventions have not been designed for adolescents with HD-dependent ESRD. This population has special needs in that the medical recommendations cover a range of areas including prescribed medications, fluid restrictions, and strict dietary guidelines. The addition of dietary and fluid restrictions makes generalizing from treatments developed for other pediatric conditions problematic (Burkhart & Dunbar-Jacob, 2002).

Regardless of theoretical frameworks, adherence intervention strategies can be classified into three broad categories: educational, organizational, and behavioral (Rapoff, 1999). Educational strategies utilize written and/or verbal instruction to inform patients and families about the disease, treatment regimen, potential side effects, and threats of non-adherence. Education is necessary for adequate adherence, but it is not sufficient (Dobbels et al., 2005). Organizational strategies work to simplify regimens, increase supervision, increase access to healthcare services, and target other ways in which health care is delivered to the patient. Behavioral strategies target problems in specific adherence-related behavior such as monitoring, problem-solving, and goal setting. In addition to the three factors listed above, cognitive-behavioral strategies have been found to be especially effective in improving adherence for patients with chronic conditions (Rapoff & Barnard, 1991).

Despite of a lack of adequate conceptual models, a meta review of the intervention literature suggests that to change adherence levels, focusing on changing situational or technical factors (e.g., removing barriers by developing systems to remember medications or plan meals) may be more effective than programs solely focused on changing the patient (Sluijs et al., 2006). However, this literature additionally indicates that changing patient characteristics (e.g., knowledge, motivation, and health beliefs) is important for ensuring change and
maintaining behavioral changes. A review of adult adherence intervention research by Roter and colleagues (1998) found that comprehensive interventions which included cognitive, behavioral, and affective modules were more effective for improving adherence than were interventions with only one area of focus. However, it is important to note that the meta-review generally found weak to moderate effects for all interventions and did not find any “gold standard” intervention method (Sluijs et al., 2006). Several categories of interventions were identified as potentially useful including technical strategies (e.g., pill organizers), behavioral strategies (e.g., external reminders and skills training), education, and multifaceted programs. Results from the intervention literature are discussed for general pediatric interventions and for interventions designed for adults with ESRD.

**General pediatric interventions.** In pediatric illness populations, diabetes and asthma are the most prevalent conditions, and they also are the most commonly studied. A review by Lemanek and colleagues (2001) summarized the literature on regimen adherence with asthma, type-1 diabetes, and juvenile rheumatoid arthritis patients and found that there are currently no well-established interventions. However, several candidates for potentially effective interventions were identified including those with multi-component packages consisting of organizational, behavioral, educational, and CBT strategies. An additional review by Rapoff (1999) found that few of the studies assessed clinical outcomes, and of those that did, fewer demonstrated clear improvements. Improvements in regimen adherence have been shown in children and adolescents with Type 1 diabetes in response to family-based behavioral intervention procedures including goal setting, behavioral contracts, and appropriate sharing of management responsibilities (Anderson, Wolf, Burkhart, Cornell, & Bacon, 1989; Satin, La Greca, Zigo, & Skyler, 1989). For the area of pediatric diabetes, glycemic control, often a result of adequate adherence, also has been improved by coping skills training (Grey et al., 1998). Behavioral change interventions consisting of contracting and education have been shown to
improve adherence in pediatric patients with sickle cell disease (Berkovitch, Papadouris, Shaw, Onuaha, & Dias, 1998) and to improve nutrition in pediatric patients with cystic fibrosis (Stark et al., 2003). Additionally, monitoring and reinforcement are associated with improvements in adherence in children with juvenile rheumatoid arthritis (Pieper, Rapoff, Purviance, & Lindsley, 1989; Rapoff et al., 1984, 1988a, 1988b).

Educational techniques have been found to be most important for children and families at initial diagnosis (Delamater et al., 1990) and during the transition of regimen responsibility from parent to teen (La Greca & Schuman, 1995). However, a review of adherence interventions by Rapoff (1999) found that for chronic diseases, behavioral techniques are a necessary addition to educational and/or organizational strategies. Multi-component interventions with pediatric populations, including self-management training (Baum & Creer, 1986; Delamater et al., 1990) and group interventions (Anderson et al., 1989) which combine factors such as self-monitoring, incentives, and education have been found to improve management of illness. Overall, multi-component treatments appear to be more effective than individual component treatments. With regard to adherence-enhancing interventions for pediatric ESRD, there are no published studies (Mackner et al., 2001). However, research with pediatric renal transplant patients highlights the potential benefit of and need for pre-transplant interventions (Riantavorn & Ettenger, 2005).

**ESRD adult interventions.** Intervention research on adult populations with ESRD largely has been designed to address one specific area of non-adherence (e.g., fluid restriction or medication adherence). For example, Christensen and colleagues (2002) developed a 9-module group protocol based on the self-regulatory framework to increase adherence to fluid intake restrictions and found promising trends toward improving adherence. Sharp, Wild, Gumley, and Deighan (2005) developed a 4-week group intervention utilizing education, behavioral, and cognitive components, relaxation techniques, and building social support networks to increase
fluid adherence. This study did not find significant post-treatment improvements, but there were notable improvements compared to controls at follow-up. Overall, the literature suggests that treatment components designed to improve self-efficacy, enhance self-management of the treatment regimen, overcome barriers to health behavior, and develop skills and cognitive and coping strategies to meet the demands of the medical treatment regimen will improve overall adherence. Thus far, there have not been any adherence-enhancing investigations conducted with pediatric ESRD patients.

In summary, intervention research has demonstrated that technical, behavioral, educational, and social support strategies have some level of efficacy, and interventions involving multiple components are typically most efficacious. There remains a need for interventions which are simple and feasible that can be incorporated into clinical practice (Sluijs et al., 2006). Also, Drotar et al. (2000) suggested that interventions for improving adherence ideally should be easily implemented, and they should reduce conflict and promote positive communication concerning illness management among families. In addition, they caution that strict adherence may reduce quality of life because it can become burdensome and restrictive of many normal childhood activities.

**Summary of the Literature**

This study attempts to establish initial efficacy for an intervention designed to improve adherence in adolescents with HD-dependent ESRD. The literature identifies several malleable factors associated with adherence that may serve as a guide for intervention development. These include knowledge about the treatment regimen, perceived barriers, cognitive factors including health beliefs, and coping strategies to manage the treatment regimen and deal with the challenges of having a chronic medical condition. However, there is little evidence of well-established interventions to improve regimen adherence in pediatric populations in general (Lemanek et al., 2001) and no interventions that have been developed for adolescents with
ESRD. This pilot feasibility study, which utilizes single-subject methodology, is a necessary and appropriate step for the field. Drotar, LaGreca, Lemanek, and Kazak (1995) have supported the use of well-executed case studies and case series designs in intervention research. Additionally, Drotar (2006) discusses strategies for effectively implementing psychological research into pediatric settings. He recommends that the utilization of single subject design and smaller scale studies can serve to establish intervention feasibility as well as acceptability to the families and hospital staff.

**Rationale and Hypotheses**

This research represents the next step in translating correlational research into the design of treatment intervention studies to address adherence in adolescents with ESRD. The intervention was developed using past research combined with the theoretical basis of the Health Belief Model. The six-module, multi-component educational, problem-solving, and cognitive/coping skills intervention was designed to increase adherence in adolescents with ESRD from baseline levels by the end of the treatment condition. The intervention targeted difficulties adhering to medication taking and/or fluid restrictions. These changes were indicated by patient and parent reports of adherence. Medical markers associated with fluid adherence (i.e., interdialytic weight) were assessed to supplement this report.
CHAPTER 2

METHOD

Participants

Adolescent patients (ages 12 to 17 years) with ESRD who were undergoing HD at a children’s hospital in the Southeast were recruited to participate in the study. Inclusion criteria consisted of a diagnosis of renal disease for which the patient was actively receiving hemodialysis treatment. Exclusion criteria included low cognitive ability as reported by the parents, having a primary language other than English, and parental unavailability or unwillingness to be involved at any level (in person or via phone) with the intervention.

At the beginning of recruitment, there were seven eligible adolescent participants on the unit. Of those, two were ineligible because a parent or guardian was never present on the unit, one declined to participate feeling he did not have a need at this time, one consented to participate but dropped out before completing any study measures or baseline assessments, and one consented to participate and dropped out after completing six baseline sessions. This left two participants in the study, who are described below.

At enrollment, Jane was a 16-year, 6-month old African American female. She was diagnosed with systemic lupus erythematosus which led to lupus nephritis approximately eight years prior to study enrollment. As a result of complications from lupus, she had both kidneys removed. Additionally, she had been diagnosed with a seizure disorder and high blood pressure. Jane began her current HD regimen approximately three months prior to enrollment. Previously, Jane lived in a different state where she was on peritoneal dialysis for 3.5 years followed by approximately one year of HD. Jane is home-schooled and completing 9th grade level work. Jane participated in the study with her 36-year-old biological mother who was separated from Jane’s father, had some college education, and was unable to work due to
Jane’s health. Jane had two siblings, an aunt, and an uncle also living in the household. Jane had Medicare and private health insurance, and her prescription drug coverage consisted of a small (<25%) co-pay.

At enrollment, Jack was a 14-year, 11-month old African American male. He was diagnosed with focal segmental glomerulosclerosis (FSGS) approximately eight years prior to enrollment in the study. He began his current dialysis regimen one and a half months prior to study enrollment. He had no previous history of dialysis or renal transplantation. Additionally, Jack had been diagnosed with hypertension and ADHD. He is in the 9th grade at a public school. Due to his dialysis schedule, he only attends school on Tuesdays and Thursdays. Jack was failing school prior to beginning dialysis, and his grades continued to suffer. In the months prior to being diagnosed with FSGS, he was in an automobile accident in which a peer was killed. It is unclear who was driving. He was enrolled in psychological counseling prior to and throughout beginning this investigation. His mother reported that he was seemingly unresponsive to counseling and did not talk during his sessions. Jack’s mother was 35 years old and single. She worked part time and has a professional degree. His mother also receives dialysis for FSGS. Jack is an only child but does have a grandparent that lives in the house. He reported that the causes for his kidney disease and need for dialysis were his mother, grandmother, and grandfather, each of whom he reported as having FSGS. His mother also reported that she believed Jack had his disease because of hereditary factors. Jack had health insurance that provides full coverage for prescription medication.

Measures

**Demographic and medical information.** Demographic information collected about children included: (a) child’s age, (b) gender, (c) education level, and (d) ethnicity/race. Demographic information collected about parents included: (a) marital status, (b) educational attainment, (c) current household composition, and (d) health coverage. Information on
historical medical and psychiatric diagnoses was collected, along with adherence-related lab values since beginning their current HD regimen and throughout their participation in the study.

Medical Adherence Measure (MAM; Zelikovsky & Schast, 2008). The MAM is a semi-structured interview to assess three domains of medication adherence behavior: medication knowledge, regimen responsibility, and medication adherence. The full measure was completed separately by adolescents and parents at baseline and at the end of treatment. Additionally, the medication adherence domain was assessed weekly throughout baseline and treatment. The MAM recorded participant knowledge of medication name, purpose, dosage frequency, and amount to be taken. This yielded a score of zero to four for each prescribed medication. Scores for each medication were summed and divided by the total possible score to obtain a percentage, with higher percentages being associated with greater medication knowledge. Regimen responsibility examined the agreement between parent and patient regarding who manages various aspects of the treatment regimen. This was then classified as either agreement- parent responsible, agreement – child responsible, or disagreement for each aspect. The medication adherence module assessed non-adherence behavior via the number of missed doses for medication. Medication non-adherence was computed into a percentage by summing the number of missed doses and dividing this by the total number of prescribed doses.

A previous study using the MAM to assess medication adherence found internal reliability to be good for both parent and child reports ($\alpha = .88$ and .84, respectively; Simons & Blount, 2007).

Regimen responsibility. A measure was developed for this study to assess parent and teen responsibility for medication taking. The measure consisted of eight questions and was adapted from an unpublished measure assessing regimen responsibility in inflammatory bowel disease (Raboin, Greenley, Liace, & Lamb, 2008). Both parent and teen reported on their own perceived responsibility and the responsibility taken on by the other (e.g., How involved was each person in knowing the names and dosages of [child’s] medications? and How involved
was each person in organizing daily/weekly medications?). Each question was rated on a Likert-type scale from 1 (not involved at all) to 9 (involved all the time). This allowed for multiple people to be simultaneously highly involved in medication management. A mean score for the measure was computed.

Medical record indicators of fluid adherence. Interdialytic weight gains (IWG) between dialysis sessions were obtained and weekly means calculated. This variable has been shown to be an accurate and valid reflection of fluid adherence (Manley & Sweeney, 1986). It was recommended for both participants that IWG should be less than 3 kg between sessions.

**Procedures**

After receipt of parental informed consent and adolescent assent, measures designated for completion at baseline were collected during the adolescent’s dialysis appointment. Baseline medication adherence was established using self- and parent-reported levels of adherence. IWG was used to establish a fluid adherence baseline and monitor progress in this area. After establishing baseline, participants began the six-module intervention. Data collection and each of the intervention components was administered while the patient was undergoing HD. Baseline data collection took approximately 15 minutes while each module, including data collection, took approximately 60 to 90 minutes. This occurred during the participant’s 3-hour HD session.

**Intervention components.** The intervention was a six-module, multi-component, manualized intervention. The goal of all modules was to identify and address barriers to adherent behavior including inadequate knowledge, behavioral barriers (e.g., inability to plan, problem solve, or use effective active coping skills), cognitive barriers (e.g., negative illness perceptions), and affective barriers (e.g., lack of appropriate coping skills to manage negative emotion). The treatment was designed to concurrently address difficulties with fluid and medication recommendations as relevant for the participant. The number of modules was
limited to six with the goal of developing an effective yet clinically practical treatment. However, based upon the patient needs, the intervention was designed to provide additional sessions for components in which the patient struggled to demonstrate mastery. Mastery was assessed via a brief quiz at the end of each session to assess knowledge of skills introduced. In general, sessions were completed individually with the adolescent during dialysis treatment. Parents were concurrently involved in the treatment with the adolescent in Modules 2 and 3, focusing on the development of systems at home and elsewhere to improve adherence. For the other modules, parents received a brief presentation of the skills taught in order to improve parental support, encouragement, and monitoring of the adolescents’ adherence behavior.

Although the overall goals and general procedures for any given module were the same for all participants, sections of the treatment were individualized based on questionnaire results and participant input. A description of measures used to help individualize treatment components can be found in Appendix A. Prior to each session, the participant and parent completed the weekly adherence ratings. All modules began with a review of skills learned in the prior module and allowed for discussion of difficulties incorporating the skill into daily life. Additionally, medical recommendations were reviewed to ensure that the participant was aware of any changes in his/her prescription.

Module 1 provided an introduction to the intervention and highlighted knowledge and education about the treatment regimen. This module was completed separately with patient and parent. Knowledge was assessed, and the patient's current medical prescription was reviewed with both patient and parent. The patient's current adherence and difficulties following medical recommendations were discussed. Information obtained from the MAM and a review of medical records guided this section. Barriers to adherence were discussed, as was the impact of these barriers on health behavior and motivation for change. Additionally, parents were asked to
discuss how difficulties with limit setting (e.g., establishing appropriate rules and consequences) and consequence enforcement may influence their child’s non-adherent behavior.

Module 2 was completed jointly with teen and parent. In this module, the parent’s and child’s responsibilities for different behavior related to adherence were reviewed and ideal levels of responsibility were discussed. Additionally, problem-solving skills were taught to help the family identify non-adherent behavior as a problem, learn problem-solving skills, and develop solutions to address technical and behavioral adherence barriers. The barriers included those identified in Module 1 and others identified by participants.

The skills taught in Module 2 were revisited in Module 3. In Module 3, participants reviewed the successfulness of problem solutions implemented after Module 2. Successful use of strategies was praised, while those strategies that were unsuccessful or untested were re-evaluated. Revised plans were developed as needed with the family after re-navigating the problem-solving skills learned in Module 2.

Module 4 focused on the role of negative cognitions about the illness or treatment as a barrier to adherence. This module was completed separately with parents and teens. The thoughts-feelings-behavior triangle, of the cognitive model, was presented to explain how thoughts can impact mood and behavior, including adherence. Examples of negative health beliefs were introduced and participants were encouraged to discuss the role of their cognitions (e.g., health beliefs and illness perceptions) in adherence. Strategies to change negative thoughts by identifying alternate ways to think about situations that would lead to more adherent behavior were discussed. For parents, this module also focused on challenging thoughts that make it more difficult to set appropriate limits and establish consequences for their child’s non-adherent behavior.

Module 5 discussed the role of emotion regulation and coping skills in overcoming adherence barriers. This module was completed separately with parents and teens. This section
worked to help teens and parents identify sources of frustration and other negative emotions that promote non-adherence and develop strategies for coping (e.g., relaxation skills, distraction) that are healthier than avoidance. Matching coping skills to problem areas (e.g., active coping skills for controllable stressors and passive/emotion coping skills for uncontrollable illness and treatment aspects) also was highlighted.

Module 6 consisted of a review of skills learned throughout the intervention. The goal was to refine and integrate the use of all skills so that they can be used consistently. Additionally, changes in adherence since baseline were discussed. Participants also were asked to discuss areas of concerns and future problems that may lead to non-adherent behavior and to problem-solve around these. Additionally, future goals were set by the participant.

Data Analysis

This research employed a case study, AB design to provide an initial assessment of the feasibility and effectiveness of the proposed intervention. Treatment was initiated after 5 or 6 baseline sessions. Consistent with single subject research design methodology (Barlow et al., 2009), the efficacy of the intervention was analyzed visually within subjects. Given the weekly assessment of self- and parent-reported medication adherence, mean IWG, and regimen responsibility, a graph of each participant’s data is used to show changes.
CHAPTER 3

RESULTS

Results are reported separately for Jane and then for Jack.

Case 1 - Jane

Baseline. At the initial assessment, medication adherence was identified as an area of concern for Jane. Jane’s self-reported medication knowledge was 87.5% while her mother’s self-reported medication knowledge was 88.8%. Jane participated in five baseline sessions over seven weeks. The family missed two non-consecutive weeks due to difficulties with her fistula (her access point to the dialysis machine) which precluded dialysis treatment on several days. The average non-adherence rate to medication during baseline according to self-report was 38.9%, with a range of 30.5% to 48.6%. For parent report, the average non-adherence rate during baseline was 35.8% with a range of 26.7% to 52.8%. Both parent and child reports indicated somewhat erratic levels, but generally stable to slightly increasing rates of non-adherence to medication across the baseline phase (Figure 1). Non-adherence for Jane reflects selective non-adherence. She was taking ten different medications. During baseline, she and her mother reported near perfect adherence to six medications including an anti-epileptic, medications for hyperthyroidism, and immunosuppressants (non-adherence $M = 9.0\%$, range $= 2.0\%$ to $24.5\%$ for teen report, and $M = 9.7\%$, range $= 0\%$ to $24.5\%$ for parent report). In contrast, she was almost always non-adherent to four other medications, including vitamins and binders, which help remove phosphorous from her body (non-adherence $M = 65.7\%$, range $= 50\%$ to $76.5\%$ for teen report, and $M = 61.2\%$, range $= 46.4\%$ to $87.5\%$ for parent report). This differential adherence can be seen in Figure 2. Even though Jane was able to accurately report the purpose of each medication she was prescribed, she reported viewing the medications for
which she was non-adherent as being less vital to her immediate health. Jane did not exhibit any difficulties with fluid adherence with all mean IWG scores less than 3 kg. Therefore this was not directly targeted during the intervention.

In the initial session, both Jane and her mother agreed that the mother was primarily responsible for her adherence. Weekly measurement of regimen responsibility began in the second baseline session. Figure 3 shows that during baseline assessment, Jane and her mother consistently reported that Jane was involved “a lot of the time” (\( M = 6.60, \) range = 5.63 to 7.38; \( M = 7.50, \) range = 7.29 to 7.63, respectively). Jane’s mother also was rated as involved “a lot” to “all of the time” by both her own report (\( M = 8.63, \) range = 8 to 9) and Jane’s report (\( M = 7.94, \) range = 6.88 to 8.5) during the baseline period, as can be seen in Figure 4.

**Treatment: Response to individual modules.** Jane began the intervention immediately after collection of the fifth baseline data point. In Module 1, she displayed knowledge of all her medical recommendations. She also was able to identify personal strengths and weaknesses related to adherence as well as personal barriers including “slacking when labs are good” and “feeling anxious.” Jane’s mother also showed perfect knowledge of the medical regimen and was able to identify barriers for Jane of being overwhelmed by the time and effort required by her medical regimen. Additionally, she identified her own guilt about making Jane adhere and the distraction of caring for other children in the home as barriers for her facilitating her daughter’s adherence.

In Modules 2 and 3, Jane and her mother worked well together. Each completed a worksheet identifying who was responsible for different adherence-related tasks and worked together on areas of discrepancy to clarify who was responsible for different tasks. Additionally, they were able to identify ideal regimen responsibility levels specifying who should take primary responsibility for various tasks. Notably, Jane’s mother identified the need to maintain some level of responsibility, identifying the importance of monitoring Jane’s adherence behavior while
encouraging her to take more responsibility. Through problem solving, they also were able to effectively generate options for improvements in medication taking and chose several behaviors to target in the next week. Jane and her mother were able to successfully develop an organizational system for managing medication.

Module 4 targeted Jane’s mother’s self-identified guilt as a primary barrier to enforcing full adherence. She became tearful while discussing the topic, acknowledging that she had allowed medication non-adherence despite her understanding of the importance of the medication. Jane’s mother was able to develop cognitive challenges to the guilt-inducing thoughts and identified a goal of balancing emotional understanding with adherence enforcement. Examples include, “She has to have it” versus “Jane is so frustrated, and I feel guilty.” In Module 4, Jane largely identified not understanding the purpose of the medical recommendations for those drugs for which she was non-adherent. However, this statement was inconsistent with her knowledge scores, and further exploration of this statement found that Jane had a surface understanding but did not appear to recognize the long-term health consequences of her non-adherent behavior. She also identified having so many recommendations as a primary barrier and desired to exert personal control and not do things that she believed to be medically nonessential. She was able to develop effective alternate thoughts that were more likely to lead to adherent behavior, such as “I have control over my life, my kidney disease doesn’t have control.”

Both Jane and her mother were receptive to learning coping skills in Module 5. Both were able to differentiate controllable (e.g., adherence behavior) from uncontrollable (e.g., being sick, needing dialysis) aspects of the disease and treatment and identify appropriate coping strategies for both. Additionally, Jane’s mother was able to identify a need for self-care as a cognitive strategy to help her be better prepared to set reasonable expectations for Jane’s adherence behavior.
In Module 6, Jane was able to adequately describe each of the skills learned throughout the intervention and describe how they were applicable to her adherence. When shown a graph of her adherence improvement, she reported being shocked that her non-adherence was so high initially, yet she was very happy that her adherence was improving. A notable part of this session was to discuss potential future barriers and strategies for overcoming them. Jane identified negative emotions and attitudes around her treatment as a primary barrier, and she described appropriate cognitive and behavioral techniques for overcoming these. Given Jane’s recent hospitalization, strategies for maintaining adherence behavior once home were discussed. Jane’s mother was not available at Module 6. However the importance of maintaining strategies for improving adherence once released from the hospital was emphasized over the phone.

**Treatment: Changes in adherence.** At the end of treatment, both Jane and her mother showed small increases in medication regimen knowledge with Jane at 93.2% and her mother at 97.7%. The average non-adherence rate to all medication during treatment according to self-report was 20.1%, with a range of 0% to 41.4%. For parent report, the average non-adherence rate during treatment was 20.7% with a range of 3.8% to 35.2%. Data can be seen in Figure 1. Figure 2 shows that the decrease in non-adherence appears to be primarily a result of improvements in adhering to vitamins and binders, those medications Jane initially perceived as less vital to her health (intervention non-adherence, $M = 36.8\%$, range = 0% to 72.7% for teen report, and $M = 44.1\%$, range = 4.8% to 79.5% for parent report). Jane’s adherence to those mediations she perceived as “more vital” to her health remained relatively stable and high, with some possible improvement from baseline (intervention non-adherence, $M = 4.8\%$, range = 0% to 12.2% for teen report and $M = 0\%$, for parent report with no range). Initially, both Jane’s and her mother’s reports showed little to no improvements in adherence. Through the material covered in Module 3, Jane’s rate of non-adherence evidenced levels similar to those obtained
during baseline, with a possible increasing trend. Jane’s mother reported levels of non-adherence that were generally within the lower ranges found during baseline, with a possible decreasing trend. However, after completion of Module 4, there was a significant decline in medication non-adherence reported by both Jane and her mother. This improvement in adherence was maintained through the end of treatment.

Figure 3 shows that during the intervention, Jane and her mother consistently reported that Jane was involved “a lot of the time” ($M = 7.4$, range $= 6.8$ to $8.4$; $M = 6.9$, range $= 6.1$ to $7.5$, respectively). Jane’s mother also was rated as involved “a lot” to “all of the time” by both her own report ($M = 8.2$, range $= 7.3$ to $8.7$) and Jane’s report ($M = 8.5$, range $= 8.3$ to $8.8$) during the baseline period, as can be seen in Figure 4. Notably, these scores are consistent with baseline levels. Jane did experience a few hospitalizations for complications not directly associated with her ESRD during the intervention. These time periods are noted in Figure 1.

Case 2 - Jack

Baseline. Jack participated in six baseline sessions. During the first three sessions, Jack and his mother reported perfect adherence (Figure 5). However, Jack’s mother reported that Jack had taken no responsibility for his medical care and that his adherence was totally due to her efforts, which were becoming increasingly frustrating to have to perform. This perceived lack of responsibility was supported by Jack’s low medication regimen knowledge score of 50%. His mother’s knowledge was notably higher at 82.1%. During the third baseline session, weekly assessment of responsibility for medication adherence tasks began. Throughout the baseline period, Jack’s mother’s reported frustrations increased, and her regimen responsibility for his adherence decreased. Jack and his mother reported that Jack assumed little to no responsibility for his own medical care ($M = 2.3$, range $= 1$ to $4.75$ for self-report and $M = 2.8$, range $= 2.38$ to $3.25$ for parent report, Figure 6). Jack reported that his mother was responsible “some of the time” while Jack’s mother reported that she was responsible “a lot to all of the time,” both
reports indicated a decreasing trend in maternal responsibility \( (M = 5.5, \text{ range } = 4 \text{ to } 8.38 \) for self-report and \( M = 8.3, \text{ range } = 7 \text{ to } 9 \) for parent report Figure 7). After the three sessions of self- and parent-reports of perfect adherence, and generally coinciding with decreasing reports by Jack’s mother of her responsibility for his care, there was a steady and consistent increase in both self-reported and parent-reported non-adherence (Figure 5). The average medication non-adherence rate during baseline for self-report was 20.0% with a range of 0% to 42.9%. For parent report, the average medication non-adherence rate during baseline was 17.1% with a range of 0% to 57.1%. There was a steep increase in Jack’s nonadherence over the last three baseline sessions as reported by Jack’s mother, and over two of the last three sessions as reported by Jack. During baseline, Jack’s weekly mean IWGs implied increasing non-adherence to fluid restrictions with a average baseline weekly mean IWG of 2.95 kg with a range from 1.47 kg to 4.73 kg (Figure 8).

_Treatment: Response to individual modules._ Jack was resistant from the beginning to the intervention and exhibited some defiant behavior. Throughout the training period, he required high levels of coaxing and persuasion by the trainer. Jack demonstrated incomplete knowledge of his medication regimen in Module 1, which was consistent with his initial assessment. Also, in identifying strengths, Jack identified his mother’s helping him to remember and prompt him to be adherent. He reported his two biggest barriers as “I just don’t want to” and “being away from home (and mom).” Jack’s mother showed good knowledge of Jack’s medical recommendations, and she seemed to have an accurate knowledge of his strengths and barriers. Similar to Jack, his mother identified his barriers as being “his attitude” and his “being away from home.” She identified her own barriers to his adherence as being her anger and frustration and her difficulty monitoring his behavior when they are not together.

In Module 2, working with both Jack and his mother together highlighted significant conflict in the family. Notably, when working on clarification of regimen responsibility, Jack
identified that he does not take responsibility over any parts of his regimen other than attending dialysis and that he did not plan to do so in the future. This was frustrating for Jack’s mother who ideally would like for Jack to become between 50% to 100% responsible for varying aspects of his medical regimen.

Module 3 was completed separately on different days within the same week with Jack and his mother, given her unavailability on the first day. Jack indicated that he had not tried any of the strategies identified in Module 2 and showed little motivation to make changes. Jack’s mother stated that she attempted to help Jack make changes, but that these changes were unsuccessful due to his attitude. Given Jack’s lack of motivation to make environmental changes and his lack of success in making changes in any areas, the decision was made to continue to Module 4.

In Module 4, Jack was able to identify unhelpful cognitions but was resistant to challenging them. He reported understanding of the cognitive model, but reported that he did not believe it would be effective. With prompting, he did complete an example of how challenging thoughts could influence adherence behavior. Despite his lack of engagement, he was able to successfully complete the module quiz and progress to Module 5. Jack’s resistance continued in Module 5. When discussing appropriate coping strategies, Jack could not identify helpful strategies. He often chose “distract myself” as a coping strategy for dealing with his illness and treatment-related frustrations but would not identify any activities he could use as enjoyable distracters. This was indicative of an avoidant coping style. He demonstrated a very rudimentary understanding of controllable versus uncontrollable stressors. In this session, he re-stated that he blamed his mom for his illness and that he did not think that his medications were helpful, yet he refused to discuss possible strategies for coping with these thoughts.

Given Jack’s worsening adherence, before completing the termination session, Jack was offered the opportunity to repeat any or all of Modules 3 through 5. He identified that he was
most interested in repeating Module 4. Jack was still somewhat resistant during this second presentation, but was better able to challenge negative thoughts (primarily by choosing the opposite thought). He struggled to identify emotion words and reported that he usually doesn’t feel or care. Jack was able to show a better understanding of the cognitive model and challenging thoughts after this session. However, his affect was notably blunted. Given his presentation, increasing non-adherence, and the researcher’s difficulty contacting his mother (who was not present for Module 4 or 5), the hospital social worker contacted Jack’s mother. At this time, Jack’s mother explained that she was “tired of discussing Jack’s issues” given his increase in oppositionality and defiance. She verified that Jack was completely non-adherent despite setting medications out for him to take. She asked to be withdrawn from the study but stated that Jack could finish the intervention if he wished.

Jack was approached after this and, despite his generally oppositional and withdrawn presentation, stated that he was willing to complete the termination session and continue to complete study measures but did not want to repeat any other modules. At this meeting, he was briefly introduced to Module 6. He identified the cognitive model (“the triangle”) as one thing he had learned but was not using. Jack completed Module 6 the next week. Jack was able to briefly describe the skills discussed throughout the intervention. He identified “the triangle” as the most useful skill. He did not identify any specific future goals beyond “just doing it” for his adherence-related behavior. Notably, his medication and fluid adherence had shown some improvement in this last assessment. Jack attributed these changes to “just doing it” as discussed in Module 4. He also reported that his mother had been more involved in the past week with some improvements in communication, yet he did not attribute this to his improvements and greater participation in his own care.

*Treatment: Changes in adherence.* Given that Jack’s mother did not complete any measures after Module 2, results from her report are not presented here but can be seen in
Figures 5 through 7. Jack’s medication knowledge at the end of treatment was somewhat lower at 42.9%. However, it is notable that he claimed not to know the names of any of his medications which he has independently reported in the past. The average non-adherence rate to medication during treatment showed an increase according to self-report \((M = 63.9\%, \text{ range } = 28.6\% \text{ to } 100\%; \text{ Figure 4})\). This reflects a continuation of the steep increase in nonadherence that started during the baseline phase. Yet, some improvement in non-adherence was noted at the very end of treatment. Jack’s responsibility for medication adherence remained at almost zero throughout the treatment \((M = 1.5, \text{ range } = 1.0 – 3.7; 1 = “\text{none at all;}” \text{ Figure 5})\). There was a slight increase to being “a little” involved \((M = 3.71)\) coinciding with adherence improvements toward the end of treatment. Additionally, his report of his mother’s regimen responsibility also showed continued decreases to the level of no involvement \((M = 3.8, \text{ range } = 1.0 – 5.43; \text{ Figure 6})\) with increases in his report of her involvement on the last day of treatment. Jack’s weekly mean IWG initially increased during the treatment phase as well \((M = 2.7 \text{ kg, range } = 1.4 \text{ kg to } 4.8 \text{ kg})\) yet showed improvements toward the end of intervention. However, this showed an unstable pattern which generally remained in the range of the baseline assessment (Figure 8).
CHAPTER 4
DISCUSSION

The current study provides two case examples of responses to a new intervention designed to improve adherence in adolescents with HD-dependent ESRD. The study found that it is feasible to conduct an adherence-enhancing intervention during HD treatments. Participants were able to complete all modules and questionnaires without difficulty while connected to the dialysis machine. Nonetheless, treatment implementation did require flexibility as there were situations in which the trainer had to return on a different day to implement a module or collect data because a participant did not feel well or had complications (e.g., difficulties connecting to machine) that prevented dialysis treatment that day. Additionally, completing the parent components of the intervention was, at times, more difficult as the parents did not always attend their children’s dialysis session.

Results examining improvements in adherence were mixed. One participant had improvements in medication adherence while the other participant actually had declines in adherence. The use of single-subject methodology allows for a visual examination of individual responses. Observations made during treatment administration allow for a close examination of probable explanations for differential responses to the intervention and the potential impact of the various treatment components.

An examination of the participants’ demographic and medical factors indicates that both patients were African-American and of similar age. There were differences in gender, time since diagnosis, and time on dialysis; however, current research does not suggest that these variables should explain the differential responses to treatment. Several other differences between the two participants more likely explain the mixed findings, including differences in
parental involvement and family conflict, level of regimen responsibility, and differences in child factors such as presence of psychological difficulties. Each of these will be addressed. 

Research indicates that high family conflict and low levels of parental monitoring are associated with decreased adherence rates (Anderson et al., 1990; Wiebe et al., 2005). Although this study did not utilize an objective measure of family cohesion, subjective reports throughout the study indicated that Jack and his mother struggled with communication, and conflict was high. This was further evidenced by decreased scores for maternal regimen responsibility and by Jack’s mother withdrawing from the study in the middle of the intervention. Jack’s mother was frustrated by his lack of assumption of any responsibilities for his care and his increase in oppositional behavior. This decrease in maternal monitoring and responsibility occurred while Jack was consistently demonstrating almost no responsibility for his treatment regimen. Jack also blamed his mother and grandparents for his kidney disease, noting that they also suffered from the same condition. This could have added to family conflict. On the other hand, Jane’s mother was responsive throughout the intervention. Both Jane and her mother consistently reported a high level of maternal and child responsibility for medication management which was indicative of a collaborative approach to illness management. Collaborative approaches have been associated with better adherence outcomes (Fiese & Everhart, 2006; Wiebe et al., 2005).

Another notable difference between the two participants was the prevalence of preexisting psychological conditions. Jack had a preexisting ADHD diagnosis with reported and observed oppositional behavior. Research has identified such factors to be associated with lower adherence levels (Bauman et al., 1997; Griffin & Elkin, 2001; La Greca & Bearman, 2003; Shaw, 2001). Additionally, Jack’s presentation during the intervention showed symptoms of depression (e.g., negative affect with a restricted range, no motivation, unable to identify future-oriented thoughts). Depression has been shown to negatively impact motivation, which in turn
can negatively influence adherence (Simoni et al., 1997). Jack also had existing academic difficulties prior to treatment. Further, Jack had been present in a fatal car wreck during the past year in which a friend died. This occurred prior to beginning his dialysis regimen and raises the possibility of post-traumatic symptomology that may be influencing Jack’s current behavior, including adherence. Unlike Jack, Jane had no reported significant psychiatric history. Jane’s mother did indicate that Jane had a history of anxiety difficulties. However, this was noted as a potential barrier early on, and both Jane and her mother were open to discussing how anxious thoughts can interfere with adherence.

It is also interesting to examine the current results through the lens of the Health Belief Model. The main aspects of this model include perceived susceptibility to illness or complications, perceived benefits of health-related behavior, and perceived barriers to health care. Regarding perceived susceptibility, Jack reported on several occasions that he felt there were no negative consequences to his non-adherent behavior. Additionally, Jack’s mother reported that after being in a car accident with a peer, Jack indicated that he believed himself to be invincible. Jane, on the other hand, was able to acknowledge how her adherence to treatment reduced her susceptibility to complications. Jane also was better able to identify how her medications were helpful for maintaining health, an area in which Jack showed significant difficulties. Her adherence behavior was associated with her perception of health-related benefits, which Jack denied. Further, while both Jack and Jane identified barriers to health behavior, Jane was more open to addressing strategies for overcoming barriers while Jack reported feelings of powerlessness and hopelessness regarding his medical condition. The participants’ differential responses to treatment are consistent with the theoretical underpinnings of the Health Belief Model. However, this model still fails to account for potential environmental and family factors that affect adolescent adherence such as parental monitoring.
The six-session, multi-component intervention was not expected to impact adherence rates immediately. An examination of the data in combination with participant observations indicated that Jane made the most notable declines in non-adherence after Module 4, which focused on cognitive skills. This was the session in which Jane’s mother was able to identify strategies for challenging her own guilt-inducing thoughts, and Jane could challenge her thoughts that certain medications were “less important.” It may be that these cognitive skills were necessary to enhance motivation to use the behavioral strategies that were previously discussed. Notably, while Jack did not show any improvements after this module, this was the skill he described when asked what he had learned during the intervention. It is possible that although he thus far had not consistently implemented the skills to change adherence, he had built a knowledge base that he might utilize in the future. This notion is tentatively supported by his late-improving trend in adherence.

Another important point for discussion is to consider adherence at enrollment. Reportedly at enrollment, Jack was completely adherent to his medical regimen, in that he was ingesting the medications, yet he remained in the study given his mother’s frustrations over his lack of responsibility and his lack of regimen knowledge. While some may argue that it is best not to intervene when a family is adherent, regardless of how adherence is achieved, it also could be argued that Jack had significant risk factors and that a prevention approach would be warranted. While it will be important for continued research to assess treatment effectiveness in improving adherence problems, clinical researchers also should consider the need for development, implementation, and assessment of prevention interventions. A component of these interventions should be to increase adolescents’ responsibility for adherence, while maintaining parental responsibility for supervision.

There are a number of limitations to this investigation that should be noted. Among these is a lack of experimental control. The study originally intended to incorporate a multiple-baseline
design. However, this had to be abandoned after two participants withdrew from participation and Jack exhibited no treatment response. Additionally, thus far, the current study does not present follow-up data examining maintenance of treatment effects for Jane. Also, the treatment effects for Jane are made more difficult to interpret given her hospitalization toward the end of the intervention. Yet, it is notable that during a baseline week with a four-day hospitalization, the family continued to report high non-adherence rates. An additional issue is that the small sample does not allow for easy generalization of the results to other adolescent HD patients. This is an especially important point given there was differential treatment response between the two participants. Additionally, the current intervention does not require a competency in skill use to progress through the modules; rather, it requires a knowledge-based mastery. As seen in the case of Jack, one can acquire a rudimentary knowledge of skills without demonstrating behavioral competency. Clinically, therapists would aim for behavioral mastery in their patients.

In addition to these issues, despite offering the intervention during a time when participants already had to be at the hospital, there were still recruitment difficulties. For example, the participant who withdrew from the study after completing several baseline sessions was on an early morning shift (i.e., 5:30 a.m.) and often had to be awakened when the researcher arrived to collect data. This was one reason indicated for withdrawing from the study. The other family that withdrew immediately after recruitment had a history of a lost transplanted kidney due to non-adherence. This is an important consideration as it may be those most in need of intervention are also most likely to have a history, and possibly a long one, of non-adherence to potentially helpful interventions.

An interesting ethical issue, with strong implications for healthcare resource allocation, is whether adherence promoting interventions should be mandatory, as opposed to optional, for patients and families in which nonadherence has been most problematic. Another difficulty in recruitment was that several of the adolescent dialysis patients arrived on their own using
medical transportation. This meant that parents were often unavailable during their child’s
treatment and unable consent for their own or their child’s participation.

Despite mixed results, the current project does provide some support for the feasibility of
the intervention in improving adherence in adolescents with HD-dependent ESRD. The current
data suggests that the intervention may be most successful in families with parents who already
have or are willing to employ high levels of monitoring. Also, the treatment is likely to be most
effective for adolescents who are motivated to improve adherence. Future research should
incorporate measures of family functioning and motivation for change to better assess these
hypotheses.

Given the relatively small population size of pediatric hemodialysis patients, the
continued utilization for small sample methodologies is appropriate for the immediate future of
research in this area. Future research would benefit from using stronger single-subject
methodologies, such as multiple-baseline design, that allow for greater experimental control.
These designs allow for a close examination of individual responses and factors that may be
associated with those differential responses. After a sufficient accumulation of case study and
single subject design studies, researchers could develop collaborative, multi-site investigations
that allow for larger sample sizes and the use of randomized clinical trials to improve adherence
in this population. Pursuit of this plan of research is important because, despite being a
relatively small population, there are great personal and healthcare costs to non-adherence in
this population. Non-adherence can result in not receiving or losing a transplant, with the
possible eventuality of life-long HD and other medical treatments paid for by government
insurance (Medicare).

Future research in this area also should consider a modification of the intervention skills
to be applicable to younger samples. Such interventions could take a treatment approach,
addressing existing adherence difficulties, or a prevention approach, introducing knowledge and
skills to maximize adherence behavior. Such interventions could be especially helpful for pre-adolescent populations in which issues around transitioning of regimen responsibility are beginning to arise. Research suggests that this transition of responsibility may occur by age 12 (Shemesh et al., 2004). In addition, longitudinal studies of adherence behavior are necessary. Specific to ESRD populations, addressing adherence behavior prior to transplant is especially important, and following the patients post-transplant is much needed. Such studies may help identify the most appropriate times to intervene in order to reduce the likelihood of loss of transplanted organs due to non-adherence behavior.

Although this research was conducted within a major pediatric medical center, there were no standing protocols for addressing psychosocial issues in these patients and families. The standard of care is typically to conduct a psychosocial evaluation at the beginning of a dialysis regimen. Also, psychosocial issues that are brought up by the patients and parents are reviewed monthly. However, issues are not assessed regularly and in depth. If patients do not identify areas of concern, they often are not acknowledged by the busy medical team. Given that receiving a transplant is partially determined by the presence of a stable, adherence-promoting family environment, patients and parents may be hesitant to bring up concerns. Further, although adherence is vital to the survival of these patients and their quality of life, scarce attention is typically given to training in those areas. It is possible that the weekly meetings between the trainer and families promoted rapport that may have encouraged discussion of difficult topics affecting these families. The assessments of adherence clearly allowed for detection of difficulties with Jack of which the medical team were generally unaware. Institutionalization of routine adherence assessments in a medical setting could make it easier for problems to be identified and resolved. Clinical researchers and medical departments should consider the development of routine adherence programs that are incorporated into the standard of care for patients with chronic medical conditions.
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APPENDIX

MEASURES TO AID IN TREATMENT IMPLEMENTATION

Below are descriptions of the scales that will be used to help individualize treatment components. These measures will be given at the baseline data collection.

*Illness Management Survey* (IMS; Logan, Zelikovsky, Labay, & Spergel, 2003). The IMS was designed to assess barriers to adherence encompassing four factors: disease/regimen, cognitive problems, lack social support/self-efficacy, denial/distrust, and peer influences. The measure consists of 27 questions each answered using a 5-point Likert-type scale, from 1 = strongly disagree to 5 = strongly agree. It was adapted to allow the patient to rate how much they agree with the barriers separately for the three areas, medication regimen, dietary regimen, and fluid restriction regimen to help individualize the intervention. Additionally, a parent version was created to assess parental perceived barriers. This measure will be completed by adolescents and parents. Internal consistency in the initial report was good with an alpha of .87.

*KIDCOPE* (Spirito, Stark, Williams, 1988). The KIDCOPE assesses the use of ten common positive and negative coping strategies. This 10-item measure will be completed by adolescents to help guide intervention. At each of the assessment periods in which this measure is administered, it will be completed twice by the adolescent, once to assess coping with having renal disease and once to assess coping specifically with having multiple medical recommendations. The adolescent is asked to come up with a problem related to having kidney disease and a problem related to having multiple recommendations. For each problem, they are asked to answer four questions about how the situation affects them on a 4-point Likert-type scale from Not at all to Very much. For each problem, they then identify how often they use the ten coping strategies on a 4-point scale with 0 = not at all and 3 = all the time. They also rate
how much each of the ten strategies helped on a 5-point scale with 0 = not at all and 4 = very much.

*The Brief Illness Perception Questionnaire* (B-IPQ; Broadbent, Petrie, Mein, & Weinman, 2006). The B-IPQ is designed to assess cognitive components related to illness. These include perceived symptomatology, timeline (acute/chronic), timeline cyclical, consequences, personal control, treatment control, illness coherence, and emotional representations. The measure consists of eight questions rated on a 10-point, Likert-type scale with 1 = not at all and 10= extremely. These data highlight problematic and effective illness beliefs. This measure will be completed by both adolescents and parents. The initial study established adequate test-retest reliability and concurrent validity.
Figure 1. Jane’s medication non-adherence as reported by self and mother. B1 through B5 represent the baseline sessions. M1 through M6 represent the modules as they were presented.

Note: * indicates time during week spent hospitalized. M1 = 4 days; M4 = 4 days; M5 = 7 days; M6 = 3 days.
Figure 2. Jane’s differential non-adherence based on her perception of medication as “vital” or “non-vital” for immediate health. Vital medications include an anti-epileptic, medications for hyperthyroidism, and immunosuppressants. Non-vital medications include vitamins and binders. B1 through B5 represent the baseline sessions. M1 through M6 represent the modules as they were presented.
Figure 3. Jane’s medication management responsibility as reported by self and mother. B2 through B5 represent the baseline sessions. M1 through M6 represent the modules as they were presented.
Figure 4. Jane's mother's medication management responsibility as reported by self and Jane. B2 through B5 represent the baseline sessions. M1 through M6 represent the modules as they were presented.
Figure 5. Jack’s medication non-adherence as reported by self and mother. B1 through B6 represent the baseline sessions. M1 through M6 represent the modules as they were presented.
Figure 6. Jack’s medication management responsibility as reported by self and mother. B3 through B6 represent the baseline sessions. M1 through M6b represent the modules as they were presented.
Figure 7. Jack’s mother’s medication management responsibility as reported by self and Jack. B3 through B6 represent the baseline sessions. M1 through M6b represent the modules as they were presented.
Figure 8. Jack’s mean weekly IWG. B1 through B6 represent the baseline sessions. M1 through M6b represent the modules as they were presented.